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Stuttering and its Treatment

eleven lectures

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Jerry Siegel
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PREFACE

These lectures are intended as a reference for students of speech-language pathology who are learning to provide health care for those who stutter. Regardless, they may be of interest to a broader audience within the speech-language pathology discipline. This text is freely available from the website of the Australian Stuttering Research Centre.[†] It is updated regularly to include newly published research findings and to take account of feedback from users. The year and month of the last update appear on the cover and at the top right of alternate pages.

The lectures constitute a personal view about the reference material that students of speech-language pathology need in order to provide adequate health care for stuttering. That personal view includes judgements about the topics and research publications that students need to be aware of, and judgements about those topics that are beyond the scope of introductory material.

Much of this introductory course is straightforward. However, much of it is complex material that, at present, leaves more questions than answers. Even so, all of it, in my view, is applicable to clinical practice directly or indirectly.

The writing of this material would not have been possible without the bristling intellectual climate in which I have thrived for past decades. Many have influenced the present work, but most directly I am indebted to Ann Packman, Sue O'Brian, Ross Menzies, and Robyn Lowe. I am particularly indebted to Robyn Lowe for assistance with the writing and to Damien Liu-Brennan for scientific copy editing. Sabine Van Eerdenbrugh assisted with helpful comments on a recent version. And more thanks are due to my wife Anne Skyvington than to anyone. She supported and somehow managed to tolerate me while I wrote the first version.

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LECTURE ONE: BASIC INFORMATION

LECTURE TWO: MORE BASIC INFORMATION

LECTURE THREE: THE CAUSE OF STUTTERING

LECTURE FOUR: CLINICAL MEASUREMENT OF STUTTERING

LECTURE ONE: BASIC INFORMATION

Terms relating to stuttering

The disorder

Stuttering and stammering

Worldwide, the term *stuttering* is used most commonly to refer to this speech disorder. The term *stammering* is often used in the United Kingdom and Ireland. However, most publications about the disorder use the term stuttering.

Potential confusion

Troubling issues with terminology for stuttering have long been documented.[1] According to the American Speech-Language-Hearing Association, the disorder "is plagued with inconsistent, confusing terminology. This problem has cultural, historical, linguistic, and practical origins" (p. 29).[2] So, the following material is presented with the intention to make clinical terminology for the disorder as clear as possible.

Other terms

These terms are sometimes used to refer to stuttering: *dysfluency*, *disfluency*, and *nonfluency*. However, as will be discussed shortly, there are arguments for not using them.

Those who have the disorder

Direct and person-first terms

Historically, someone who has the disorder was referred to directly as a *stutterer*, and those with the disorder as *stutterers*. Person-first terminology is a different and more recent approach, intended to avoid any negative connotations from labelling someone with a disorder. Instead, the reference shifts to someone who has a disorder. Accordingly, preferred terms in common use are *a person who stutters, someone who stutters,* or *those who stutter*. Terminology about the disorder is in a continuous state of change, most notably of late with a view to avoiding ableist thinking and language.[3,4,5]

Making a choice

When making a choice about how to refer to those who stutter, clinicians may be influenced by the views of clients. Some clients might prefer direct terms, and some might prefer person-first terms. A useful rule of thumb is to err on the side of caution, and to use person-first terms if there is any uncertainty. When writing a formal report about clients, clinicians may prefer person-first terms. Most scientific speech-language pathology journals require the use of person-first terms for stuttering.

Potential limitations of person-first terms

For all their potential benefits, there are potential limitations with person-first terms for stuttering. Two research publications[6,7] raise doubt about whether person-first terminology for stuttering alters negative perceptions about the disorder. It is also the case that person-first terms invoke present tense, and this can cause awkward expression when writing with past tense. For example, "the research participants were people who stutter." At least one publication[8] has declined to use person-first terminology in order to avoid the wording problems that it causes.

Additionally, it might be argued that the semantics of person-first terminology is misleading about the nature of the disorder. The term *people who stutter* might imply that stuttering is something that speakers do when speaking, rather than it being something that happens to them when they speak. The latter is what in fact happens, as will be discussed during Lecture Three. Person-first terminology also suggests that those affected by the disorder, and who suffer negative effects from it, will necessarily stutter in an obvious way to observers. Yet subsequent lectures show this to be not all true.

When people stutter

Stuttering and stuttering behaviour

As well as being a term to refer to the disorder, *stuttering* can be used to refer to someone's speech being affected by it. For example, "she was stuttering a lot yesterday," and "stuttering on the telephone is a problem for him."

Sometimes clinicians use the term *stuttering behaviour* (spelled *behavior* in the United States) in formal contexts such as written reports and conference presentations. In such contexts, the term *behaviour* to describe stuttering is a little different from the everyday use of the term. Researchers sometimes use the term *stuttering behaviour* in scientific publications. For example, "the observers were instructed to push a button for every stuttering behaviour," and "the stuttering behaviours reportedly began suddenly." However, clinicians generally don't use such formal terminology when talking to clients and parents or when writing notes in client files.

It is possible for researchers to use sophisticated instruments to measure stuttering behaviour. For example, kinematic (movement) measures of lip variability during speech have been shown to distinguish children[9] and adults who stutter from those who don't.[10] However, clinicians generally don't use such instruments.

Dysfluency and dysfluent

The terms dysfluency and its adjective *dysfluent* are often used to describe when people stutter. For example, "his speech has been dysfluent for the past week" and "dysfluency in the workplace is a problem for her." But, strictly speaking, there is a problem there. The opposite of those terms, *fluency* and *fluent,* as they are commonly used in English, do not specifically refer to anything about the disorder of stuttering. They refer to a range of things about the flow of speech, not just stuttering,[11] such as rate, prosody, continuity, and smoothness. The term *fluent*, for example, can be used to mean someone speaking a second language proficiently.

Another problem is that the terms *dysfluency* and *dysfluent* are sometimes used to refer to the effects of other speech or language disorders where the flow of speech is disrupted, such as dysarthria and aphasia. So the use of those terms for stuttering may lead to confusion with other disorders.

The 2013 edition of the Diagnostic and Statistical Manual of Mental Disorders,[12] generally known as the DSM-5, introduced the term *childhood-onset fluency disorder,* and presents it interchangeably with the term *stuttering*. Arguably, this is not at all helpful, but so far seems not to have influenced the field of speech-language pathology. To the contrary, some scholars recently recommended "that researchers and clinicians cease referring to stuttering as a *fluency disorder* and simply refer to it as *stuttering*" [authors' italics] (p. 645).[13]

Disfluency and disfluent

The terms *disfluency* and *disfluent* are sometimes used to refer to stuttering. Strictly speaking, this is not correct. The problem is that the prefix *dis-* does not necessarily mean that something is disordered. The prefix *dys-* does mean that, but the prefix *dis-* can mean something more like different, or not usual. So *disfluency* is not an ideal term to use for stuttering. This is particularly the case when the term is used to refer to speech associated with other disorders, a common example being autism.[14,15,16,17] Lecture Four touches on a particularly confusing use of this prefix with the term *stuttering-like disfluencies*.

Nonfluency and nonfluent

The same semantic problem pertains to the terms *nonfluency* and *nonfluent* that are sometimes used for stuttering. These are potentially confusing because, again, the prefix *non-* does not necessarily mean disordered.

When people do not stutter

No stuttering

No stuttering, and variations such as *not stuttering*, are simple and non-confusing ways to describe someone's speech that has no stuttering. For example, "he reported no stuttering all last week" and "I have not heard you stutter for the past 10 minutes."

Stutter-free speech

The expression s*tutter-free speech* is a more formal way of referring to speech that does not contain stuttering. For example, "his speech was stutter-free during a 5-minute telephone call" and "she was stutter-free during a presentation at work." Clinicians sometimes write *stutter-free speech* in formal contexts such as reports about clients, and in professional or scientific publications, but they may be reluctant to use such a formal expression when speaking with clients or their parents. That issue aside, *stutter-free speech* certainly is a non-confusing term.

Fluency and fluent

Fluency and its adjective *fluent* are other ways to refer to speech that does not contain stuttering. For example, "you were fluent just then when we were talking" and "he has been fluent for weeks now." Pedantically speaking, though, there is the issue previously described that such terms are not stuttering specific. However, clinicians use them commonly, and there is rarely any confusion when they are used to mean speech that does not contain stuttering.

Normal disfluency and normally disfluent

As discussed earlier, the terms *disfluency* and *disfluent* are not correct terms for when people stutter, because the prefix *dis-* does not necessarily refer to disordered speech. However, the terms *normal disfluency* and *normally disfluent* can be used correctly to refer to the usual hesitations and repetitions that can be a part of everyday speech. Examples of normal disfluency might be "well, um, … gosh, I don't know," and "er, I think, perhaps, um, I will have to get back to you about that." There is some evidence to suggest that a longer silent interval between one word and the next contributes to listener perception of disfluency.[18]

It is necessary to refer to such normal speech events in a way that distinguishes them from stuttering. That is because those who stutter will have normal disfluencies in addition to stuttering. It is particularly important to get this terminology right when treating young children for stuttering. That is because when stuttering has been successfully treated and stops being a problem—which is what should occur, as discussed during Lecture Seven—it is common for parents to be overly vigilant and mistake normal disfluencies for stuttering.

Here are some examples of clinical file notes that illustrate this point: "His mother was concerned that stuttering had returned, but in the clinic it became obvious that she was concerned about normal disfluency," and "I made it clear to his father that John is normally disfluent sometimes and not to confuse it with the return of stuttering."

Stuttering moments

The idea of stuttering moments

The idea of a *stuttering moment* is a useful concept for clinical practice. The notion is that those affected have speech that appears to be just like anyone else except for short periods—moments—when stuttering occurs. The first documented evidence of this idea appears to have been during the early 20th Century at the University of Iowa.[19] The idea appeared regularly in subsequent research literature, however the first formal statement of it appears to have occurred some 30 years later:

> the stuttering problem might be approached fruitfully by concentrating on *the moment of stuttering*—that is to say, by dealing with the problem of stuttering as a series of stutterings, by regarding it crucially not as a more or less constant condition, but as intermittent responses. (p. 13)[20]

A series of experiments which concluded during the late 1980s[21,22,23,24,25,26] established, overall, that the speech of those who stutter sounds normal apart from stuttering moments. So, in a clinical sense, it is appropriate to think of stuttering as momentary speech disturbances surrounded by otherwise normal sounding speech. In reality, though, it is possible that the speech physiology of those who stutter is unusual whenever they speak, but the only perceptible problems are what observers perceive as stuttering moments.

Stutters, stuttering, stutterings, dysfluencies, disfluencies

The idea of stuttering moments has been popular since its inception, and to this day clinicians use it during clinical practice. In formal reports they may write *moment of stuttering*, *stuttering moments, stuttering* or *stutterings*, but those terms are generally formal for speaking with clients and parents. It is more common for clinicians to refer to *stutters* or a *stutter* during clinical practice. They may also use terms discussed previously—along with their potential limitations—to describe moments of stuttering: dysfluencies, disfluencies, or nonfluencies.

Stuck words, bumpy words

When talking to young children about their stuttering, clinicians need a different kind of language. Popular terms with young children are *bumpy word* or *bumpy words*, and sometimes the terms *stuck word* or *stuck words* are used. The important thing to remember here is the need to communicate effectively with the child about stuttering, so any terms that do that are useful.

The table is a summary of recommended formal and informal stuttering terms.

	RECOMMENDED	*NOT RECOMMENDED*
THE DISORDER	*stuttering*	*dysfluency [2]*
	[1] stammering	*disfluency [2]*
		nonfluency [2]
THOSE WHO HAVE THE DISORDER	*[3] person who stutters*	*stutterer [4]*
	[3] someone who stutters	
	[3] those who stutter	
WHEN PEOPLE STUTTER	*stutters*	*dysfluency [2]*
	stuttering	*dysfluent [2]*
	[5] stuttering behaviour	*disfluency [2]*
		disfluent [2]
		nonfluency [2]
		nonfluent [2]
WHEN PEOPLE DO NOT STUTTER	*no stuttering*	*fluency [2]*
	[5] stutter-free speech	*fluent [2]*
STUTTERING MOMENTS	*stutters*	*dysfluencies [2]*
	stuttering/s	*disfluencies [2]*
	[6] stuck words	*nonfluencies [2]*
	[6] bumpy words	

[1] May be preferable in the United Kingdom and Ireland, [2] May not be clear that you are referring to stuttering, [3] Person-first terms are a conservative option, [4] Many prefer person-first terminology to this, [5] For use in formal contexts, [6] For use with young children

Defining stuttering

There is no single definition of stuttering

Ideally, there would be a single, straightforward definition of stuttering that was accepted by everyone. That ideal definition would contain words to make it clear who does and who does not have the disorder. Unfortunately, though, after a vigorous debate for a decade, beginning during the early 1980s, the search for such a workable and generally agreed stuttering definition ground to a halt without resolution.[27,28,29,30,31,32,33,34,35,36,37,38,39]

Still, that debate was productive because it established three approaches to defining stuttering. An important point here is that none of the three definitions can be considered as completely satisfactory. They all have limitations, but they also all have some some strengths, that make them useful in different professional contexts. During the 20th Century, three leaders in the field contributed to the development of the three definitions described below: Marcel Wingate, Oliver Bloodstein, and William Perkins.

Objective definitions

The World Health Organization definition

The most common definitions of stuttering are known as objective definitions. They are also known as behavioural definitions and symptomatic definitions. In 1977, The World Health Organization offered what seems to be the most popular definition to date:

> Disorders in the rhythm of speech, in which the individual knows precisely what he wishes to say, but at the time is unable to say it because of an involuntary, repetitive prolongation or cessation of a sound. (p. 202)[40]

A more recent World Health Organization definition has so far attracted less attention:

> Speech that is characterized by frequent repetition or prolongation of sounds or syllables or words, or by frequent hesitations or pauses that disrupt the rhythmic flow of speech. It should be classified as a disorder only if its severity is such as to markedly disturb the fluency of speech.[41]

Wingate's definition

Another older and commonly cited objective definition of stuttering is Marcel Wingate's.[42]

> 1. (a) Disruption in the fluency of verbal expression, which is (b) characterized by involuntary, audible or silent, repetitions or prolongations in the utterance of short speech elements, namely: sounds, syllables, and words of one syllable. These disruptions (c) usually occur frequently or are marked in character and (d) are not readily controllable.
>
> 2. Sometimes the disruptions are (e) accompanied by accessory activities involving the speech apparatus, related or unrelated body structures, or stereotyped speech utterances. These activities give the appearance of being speech-related struggle.
>
> 3. Also, there are not infrequently (f) indications or report of the presence of an emotional state, ranging from a general condition of "excitement" or "tension" to more specific emotions of a negative nature such as fear, embarrassment, irritation, or the like. (p. 488)

Limitations of objective definitions

Objective definitions of stuttering can be regarded only as descriptions of stuttering, not definitions of stuttering, because they cannot be used to set apart those who do stutter from those who do not. This is because there are no observable speech events that can be recorded with words and which categorically distinguish between stuttering and normal speech.[43] At some time everyone has normal disfluencies that can be described with the same terms that can be used to describe stuttering moments.

For example, with the World Health Organization definition, it is true that those who stutter will experience “involuntary, repetitive prolongation or cessation of a sound,” but anyone will do things from time to time that can be described that way. The same can be said about much of Wingate’s definition. For example, everyone has “repetitions” occasionally during speech. This definition has also been criticised because it contains "qualifiers and imprecise terms" (p. 17),[44] such as “readily,” “sometimes” and "usually," and because speech dimensions such as “controllable” and “involuntary” are not observable,[45] as should be the case with an objective definition.

The strength of objective definitions

Objective definitions of stuttering are useful ways to describe the disorder. In particular, Wingate’s definition is a comprehensive and compact description of the disorder, and as such it is useful in various professional contexts. For example, clinicians could use it, or variations of it, when describing the disorder to clients, other health professionals, or to the media.

Internal definition

Perkins’ definition

William Perkins’ definition[30,32] of stuttering is a “temporary overt or covert loss of control of the ability to move forward fluently in the execution of linguistically formulated speech" (p. 431).[32] This is referred to as an internal definition because “loss of control” refers to a speaker’s experience. This contrasts it with the objective, observable features of objective definitions.

Limitations of the internal definition

It has been argued that the internal definition is more a statement about the nature of the disorder than a definition.[46] Also, this definition has in common with objective definitions that it fails to distinguish between stuttering and usual speech. Probably, all speakers would report that, at some time, they lose control of their speech. Another issue is that clinicians cannot observe “loss of control” because it is an experience, not a behaviour.

The strength of the internal definition

The internal definition of stuttering certainly is a valid one, because stuttering is fundamentally a personalised experience for those affected. The proponents of this definition even conducted an experiment purporting to verify this.[47] They showed that a speaker could distinguish recordings of real and faked stuttering shortly after producing them, but neither the speaker nor listeners could distinguish them at later times.

Clinicians rely on internal definition of stuttering during routine clinical measurement of stuttering severity. As will be discussed in Lecture Four, it is essential to obtain client reports of how severe their stuttering is. When clients give you that information they are, in effect, drawing on an internal definition of stuttering. If a client says that stuttering is not present, and has not been present for a significant period, that is important clinical information because of its validity. Internal definition of stuttering can be useful during research about the disorder. For example, in one report,[48] children were asked if they thought that they stuttered as part of determining whether they had recovered from the disorder.

Another reason why internal definition of stuttering is valid is that it reflects what clinicians want to achieve for clients with treatment: a change of the experience of the disorder, and a positive shift of how they feel it affects them. The obvious validity of the internal definition was shown with a report[49] of 430 adults who stuttered who were surveyed about how they thought the disorder should be defined. The researchers concluded that:

> To adults who stutter, the term *stuttering* signifies a constellation of experiences beyond the observable speech disfluency behaviors that are typically defined as stuttering by listeners. Participants reported that the moment of stuttering often begins with a sensation of anticipation, feeling stuck, or losing control. (p. 4356)[49]

Perceptual definition

Bloodstein's definition

Oliver Bloodstein's definition[35] is that stuttering is "whatever is perceived as stuttering by a reliable observer who has relatively good agreement with others" (p. 9–10). In other words, a clinician who has consensus with a community of experienced speech-language pathology observers determines whether stuttering is present or whether it is not.

Limitations of perceptual definition

Bloodstein's perceptual stuttering definition is not clear about what constitutes a "reliable" observer and "relatively good agreement with others."[45] Indeed, a stuttering definition that relies on clinical judgement that is consistent with a clinical community raises the question of how junior clinicians might attain such consistent judgements. The answer to that is conceptually simple; senior clinicians can mentor junior clinicians about what are appropriate judgements. However, there are imposing practical aspects of such mentoring. And there is a risk that different clinical communities, such as those in different countries, may develop different perceptions about what stuttering is and what it is not. There is some evidence that this may occur.[50]

Strengths of perceptual definition

An advantage of perceptual definition is that, if the required consensus exists, it is procedurally simple and clinically workable. When parents bring children who have just begun to stutter to the clinic, they are reporting their perception that stuttering is present. As discussed shortly, there is reason to believe that clinicians generally agree with parents in such cases. So, it is arguable that such parents are reliable observers who have "relatively good agreement with others," and so they are using a perceptual definition of stuttering.

Describing stuttering moments

Taxonomies

Wendell Johnson (who arguably was the most influential researcher and scholar in the field) developed the first system for classification of stuttering moments.[51] This taxonomy was developed specifically for stuttering during early childhood and included eight terms: *word repetition*, *sound/syllable repetition, phrase repetition, incomplete phrase, interjection, revision, broken word,* and *prolongation*. There have been several variants of this initial taxonomy.[52,53] The better-known terms that were added to Johnson's original taxonomy are *disrhythmic phonation, block, blockage,* and *tense pause*. All of these taxonomies deal with stuttering during early childhood, with the exception of one.[54] Presumably, this is because of the profound historical influence on the field of two theoretical perspectives about early stuttering, which are reviewed during Lectures Two and Three: the Diagnosogenic Theory and the Continuity Hypothesis.

Unambiguous stuttering moments

For the most part, those who come to a clinic complaining that they or their children stutter will be referring to many unambiguous stuttering moments that occur during each day. The term *unambiguous stuttering moments* refers to moments during speech that, to an observer, are clearly stuttering and not normal disfluency.

This does not mean that a clinician will never be undecided about whether a particular speech event is a stuttering moment or a normal disfluency. To the contrary, this is certain to occur, particularly with young children. So, during clinical practice, this is not normally an issue. An exception would be, after successful treatment of young children, when parents need guidance with being certain of the distinction between a stuttering moment and a normal disfluency.

A taxonomy: The Lidcombe Behavioural Data Language

Overview

The following method[52] to describe unambiguous stuttering moments arguably has some advantages. It was developed for use with stuttering clients of all ages, and it describes speech behaviours only; it contains no reference to anything that cannot be observed. Additionally, it appears that with some clinical experience it can be used reliably.[55] It is known as the Lidcombe Behavioural Data Language.

This taxonomy presents stuttering behaviours in three prime categories: *repeated movements*, *fixed postures*, and *superfluous behaviours*. There is nothing new about these terms. Variations of them have been used for decades: for example, *repetitions, prolongations,* and *accessory features*.[42]

Repeated movements

Commonly, clinicians refer to these as repetitions. There are three different types of *repeated movements*.

The first type of repeated movements is *syllable repetition*.

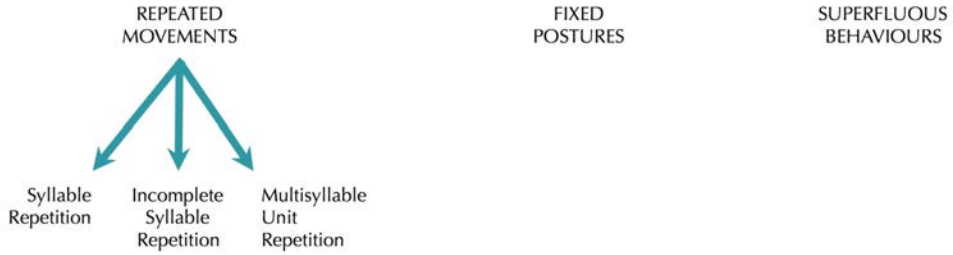
Syllable repetition is straightforward, being a repeated movement of what sounds like an entire syllable. For example, "when-when-when-when," "if-if-if-if-if," and "not-not-not-not."

Not all syllable repetitions are repetitions of entire syllables. Some of them are repetitions of parts of syllables, which are termed *incomplete syllable repetition*, meaning that the speaker did not repeat an entire syllable but part of one.

Some of the distinctions between a syllable repetition and an incomplete syllable repetition are quite obvious. For example, "can-can-can-can" might be heard as a repetition of the entire syllable, with all its phonemes. But with "ca-ca-ca-ca-can" the speaker has produced only the first two phonemes of the syllable before eventually saying the entire syllable. In which case it is an incomplete syllable repetition. Careful listening to stuttering moments may be needed to make this distinction.

Returning to the example of the syllable repetition "not-not-not-not," if "no-no-no-not" was heard it would be an incomplete syllable repetition. Also, a syllable repetition might be "I-I-I-I-I." At first it might seem that this could only be a syllable repetition, but again, careful listening is needed. The word "I" is a diphthong in most spoken English and the speaker might not complete the two vowel-like parts of this, and instead something like "uh-uh-uh-uh-I" might be heard while attempting to say "I." In which case, it would be an incomplete syllable repetition.

Repeated movements can also involve more than one syllable, in which case the term *multisyllable unit repetition* is used.

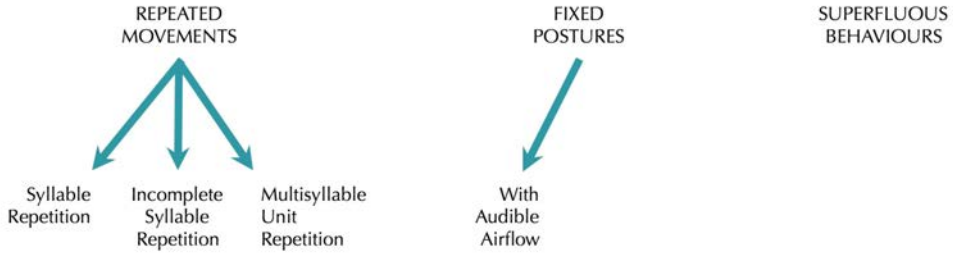


Examples of multisyllable unit repetition would be "I was-I was-I was-I was hoping," "I think that-I think that-I think that-I think that," and "then-I then-I then-I."

Fixed postures

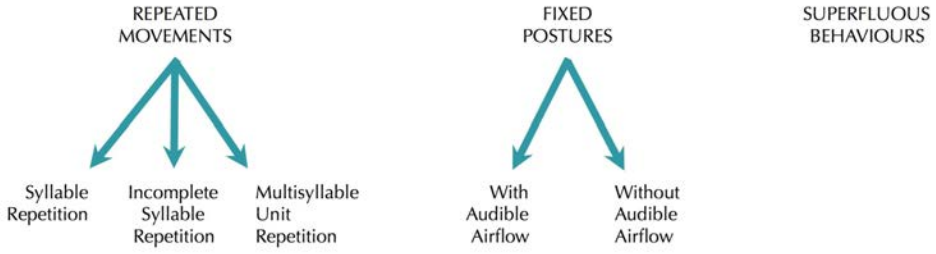
Fixed postures are in a sense the opposite kind of stuttering behaviour to repeated movements because they are not an atypical movement but an absence of typical movement. During fixed postures what normally is seen to move during speech—mostly mouth, jaw, and lips—stops moving. It can stop moving for a period so short that it might be necessary, when learning to describe stuttering moments, to look at a video carefully many times to detect it. It is far more obvious when fixed postures happen for quite a long period of several seconds. In severe cases, fixed postures can stop speech for half a minute, which of course seriously disrupts communication.

The first category of fixed postures is *with audible airflow*.



There are many kinds of airflow that can be audible. These include articulatory and laryngeal fricative noises and, more commonly, phonation. Clinicians often refer to fixed postures with audible airflow as "prolongations," because that is exactly how they sound: as if the speaker is prolonging a sound.

The second category of fixed postures is *without audible airflow*.

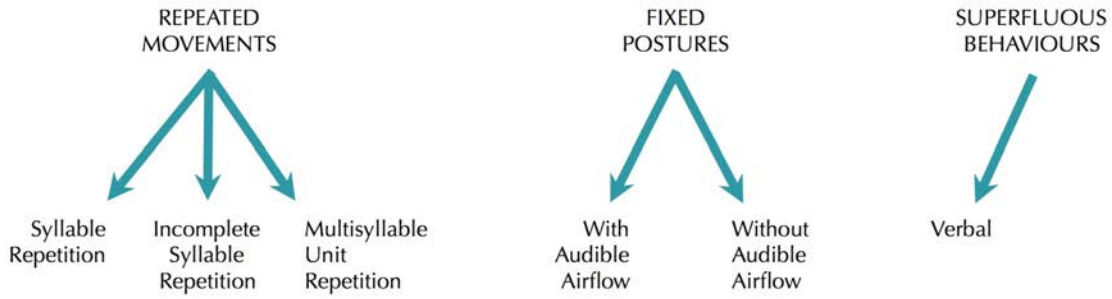


During these no airflow is audible. But it is necessary to listen carefully to be sure that there really is no sound. Sometimes the audible airflow during fixed postures can be barely audible. Clinicians often refer to fixed postures without audible airflow as "blocks," because they give the impression that something is blocking speech.

Superfluous behaviours

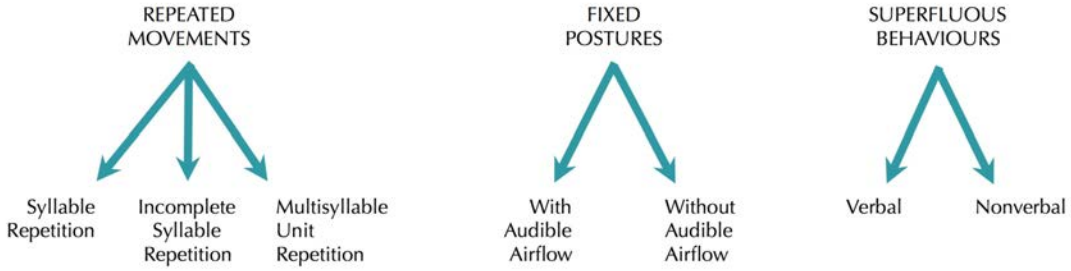
The final category of stuttering moments is *superfluous behaviours*. These are redundant to the intended meaning of the utterance as it normally would be spoken; hence, the term superfluous. These are often the most socially distracting of the observable problem behaviours of stuttering.

The first kind of superfluous behaviours is *verbal*.



It can be a challenge to identify some verbal superfluous behaviours because it is not clear whether they are redundant to the intended utterance. Johnson's taxonomy refers to them as interjections,[51] which is a term that assists with understanding how they can sound. An example would be "oh well-oh well-well-um-um."

The other kind of superfluous behaviours is *nonverbal*.



Nonverbal superfluous behaviours are easy to identify because they are obviously redundant to the intended meaning of the utterance. They include compressed lips, open mouth, breath holding, blinking, nostril dilating, eyebrow raising, grimacing, facial, head, and torso movements, inspiratory airflow, grunts and other inappropriate noises, and aberrant fluctuations in pitch and loudness. Stuttering is an idiosyncratic disorder. It is rare to see two people whose stuttering looks identical, and nonverbal superfluous behaviours are the most idiosyncratic features of the disorder.

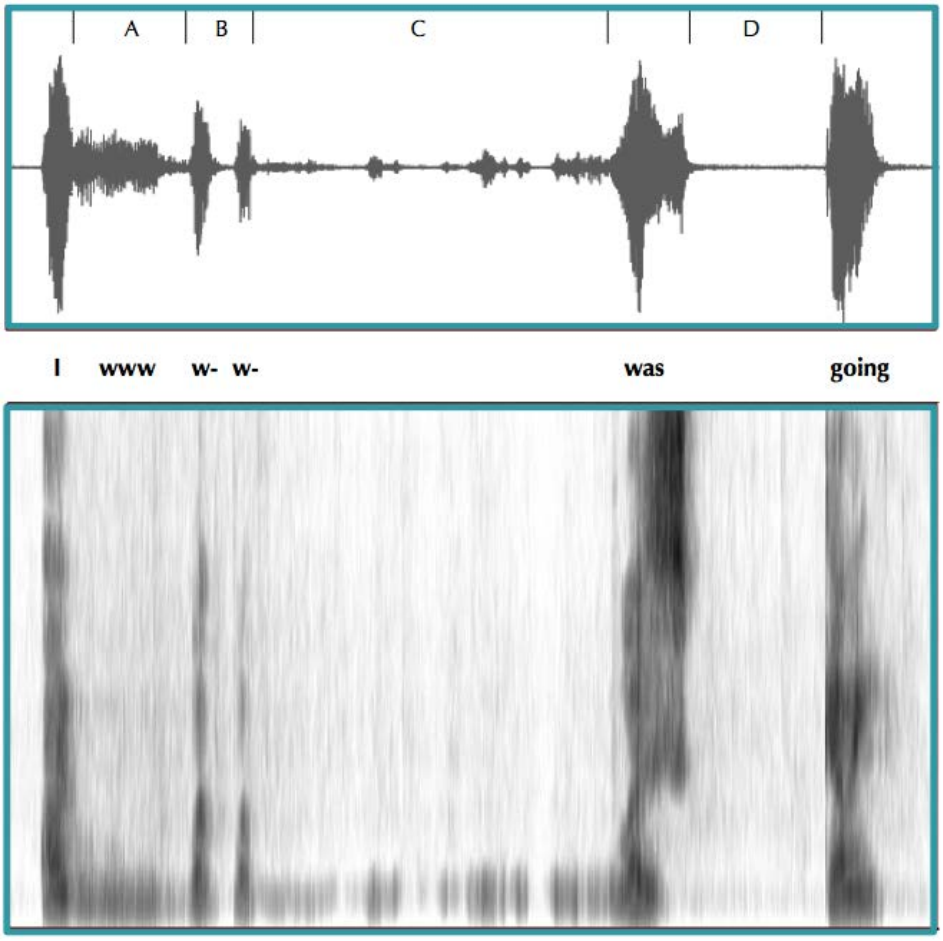
Stuttering behaviours combine in one stuttering moment

The seven stuttering behaviours described with this taxonomy, or with any taxonomy, are not mutually exclusive.[43] One, many, or even all of the seven stuttering behaviours can be present during one

stuttering moment.[56] In fact, it is rare for a stuttering behaviour to have only one of the seven stuttering behaviours by itself. For example, a stuttering moment that is a repeated movement could be a syllable repetition and an incomplete syllable repetition at the same time. Such a stuttering moment might sound like "ca-ca-ca-can-can-can." Or a repeated movement could be a syllable repetition, an incomplete syllable repetition, and a multisyllable unit repetition all at once. That might sound something like "ca-ca-ca-can-can-can-can-I-can I-can I." It also seems that the distribution of stuttering behaviours is the same in Cantonese as it is in English.[57] That finding occurred regardless of the fact that Cantonese differs markedly from English because it is tonal and syllable-timed.

A study of 3,100 stuttering moments from 147 adolescents and adults[58] reported that around half of the stuttering moments contained a repeated movement, around half contained a superfluous behavior, and around two thirds contained a fixed posture. The most commonly occurring combination of stuttering moments was fixed postures and superfluous behaviours, and the least commonly occurring combination contained superfluous behaviours only. For 18% of the stuttering moments, repeated movements, fixed postures, and superfluous behaviors occurred together.

The waveform (top panel) and spectrogram (bottom panel) in the following figure show a stuttering moment on "was" that is a fixed posture with audible airflow (Segments A and C) and incomplete syllable repetitions (Segment B). Subsequently, there is a fixed posture without audible airflow during another stuttering moment prior to the word "going" (Segment D).



Some practical examples of describing stuttering

Communicating with other clinicians

When writing about a client to another clinician who is expert with stuttering, it may be preferable to incorporate formal terminology, such as in this example:

> Most of Mr Williams' stuttered speech contained fixed postures with audible airflow, with his jaw almost shut and airflow comprising alveolar fricatives. Most of these lasted more than 1 second, with several of them lasting more than 10 seconds. During these fixed postures he had extraneous nonverbal behaviours, typically grimacing with his eyes closed, brow furrowed, and head tilted downwards and to the left. None of Mr Williams' stuttering moments involved repeated movements.

Communicating with other professionals

With reports about clients to other professionals, such as a teacher or doctor, more general terminology may be preferable to such discipline-specific terminology:

> Most of Mr Williams' stuttered speech contained speech blocks with audible airflow, with his jaw almost shut. Most of these speech blocks lasted more than 1 second, with several of them lasting more than 10 seconds. During these blocks he had nonverbal behaviours, typically grimacing with his eyes closed, brow furrowed, and head tilted downwards and to the left. None of Mr Williams' stuttering moments involved repeated movements.

The Distribution of stuttering moments

The influence of spoken language

Initial word consonants

Stuttering moments do not occur randomly during speech. Early during the last century, seminal research from the University of Iowa[59] showed that their occurrence follows rules to a considerable extent. Stuttering was shown to occur more commonly on consonants than vowels, with the vast majority of stuttering—more than 90%—occurring at the initial sounds of words. That finding has been replicated[†] many times.[60,61,62,63,64,65,66,67,68] although failure to replicate has occurred.[69] The effect has been show to occur with Japanese children who stutter,[70] with a mean age of 5 years 9 months.

First word of an utterance

In addition to occurring commonly on the first sound in a word, stuttering moments occur commonly on the first word of an utterance,[60,62,63,68,71] although, again, a failure to replicate has occurred.[72] The effect has also been reported for the first word of clauses.[73]

Rare at the end of words

It also appears that sometimes, but rarely, stuttering can occur with repeated movements at the end of words.[74,75,76,77,78,79,80,81] However, queries have been raised that such repeated movements may not be connected with stuttering.[79,80,82] One report[80] found dysfluency types other than repeated movements to occur at the end of words, but reported that they are difficult to identify perceptually.

"Difficult" sounds

Clients commonly report that certain sounds are "difficult" because stuttering is likely to occur with them. The seminal report, referred to previously,[59] showed individuality among those who stutter for sounds that are stuttered more often, and hence, considered to be "difficult." Another report from the

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[†] It is a general rule that research findings are not particularly believable unless they have been reported by researchers who are completely independent of the researchers who found them originally.

same laboratory[83] verified this finding, by showing that there is no general rule about which sounds are difficult for those who stutter.

Content words

Another early report from the University of Iowa[84] indicated that traditional grammar influenced the occurrence of stuttering. Words with heavy semantic content, such as nouns, verbs, and adjectives, are stuttered more often than words with little semantic content, such as articles, conjunctions, and interjections. Or, to say it another way, stuttering is more likely to occur on content words than on function words. This finding has been replicated many times,[61,68,69,71,85,86,87,88,89,90,91] and there is some suggestion that the effect may be language specific, with a report that it does not occur in Arabic.[92] However, it has been reported to occur in Persian.[93] One report[94] suggested that with bilingual speakers the effect might be present in the first language but not the second.

Intriguingly, there are replicated findings that with children these situations are reversed. There are reports of more stuttering occurring on function words than on content words for English,[95,96,97,98] German,[99,100] Spanish,[101] and Spanish-English bilingual[102] children. A report with Korean-speaking and English-speaking 3–7 year-olds[103] found that this effect was reversed for the Korean-speaking children, with them stuttering more on content words than function words. Two reports[88,90] have reported such a finding with studies of adults compared to children. This effect is of interest when attempting to understand the cause of stuttering, as will be discussed in Lecture Three.

Stressed syllables

Some reports have found that stressed syllables are stuttered more often than unstressed syllables,[60,104,105,106,107,108] although others have failed to find such an effect.[66,67,109,110] A report has extended such research to lexical tone with 20 Mandarin speaking Taiwanese children with early stuttering, with a mean age of 4 years 9 months.[111] Results showed that "stuttering-like disfluencies" (see Lecture Four) were around twice as likely to be associated with syllables carrying Tone 3 or Tone 4 compared to syllables carrying Tone 1 or Tone 2. The authors plausibly speculated that results "may be attributed to the increased level of speech motor demand underlying rapid F0 *[fundamental frequency]* change both within and across syllables" (p. 115).[111] This explanation might apply also with findings in English of more stuttering on initial word consonants and stressed syllables. However, a report about stuttering in Cantonese[112] found no differences for stuttering moments across its six tones. A study of Japanese children who stuttered,[70] with a mean age of 5 years 9 months, reported no differences between "heavy and light syllables."

Utterance duration and grammatical complexity

Increased utterance duration[†] is associated with increased syntactic complexity, and has been associated with increased stuttering.[66,113,114,72] These findings have been replicated many times with children,[115,116,117,118,119,120,121,122,123,124,125,126] although it is probably fair to say that the findings are not as marked and consistent as with adults. Consistent with these findings are reports that long words (measured with syllables or letters) are stuttered more often than short words.[127,128] A report[125] linked this effect to a measure of speech motor function (lip aperture variability) with a group of 7–12 year-old children who stuttered and a control group. The children who stuttered showed more lip aperture variability than controls as utterance duration increased. This is consistent with current perspectives of stuttering as a problem with neural processing impairment, as will be discussed during Lecture Three.

Clustering

Another feature of stuttering is clustering, which is the occurrence of a series of stuttering moments at one time during speech. This has been reported several times for early stuttering,[129,130,131,132,133,134] and also with adults.[135,73]

[†] Utterance duration is usually measured with words, syllables, or morphemes.

How predictable are stuttering moments?

An early report[89] indicated that 95% of stuttering moments could be accounted for by "initial sound, grammatical function, sentence position and word length" (p. 183). A later source[127] was consistent with that finding, reporting that 95% of stuttering moments can be accounted for by the word initial phoneme, grammatical class, word length, and word position in the utterance. In other words, most stuttering moments occur according to rules, but it is not possible to fully account for the occurrence of every stuttering moment.

Adaptation, consistency and adjacency

A mysterious effect

After around five readings of the same passage, stuttering decreases on average by half. This so-called adaptation effect was a much researched aspect of the disorder during the last century.

Why the adaptation effect occurs is a mystery. There are data to suggest that it is caused by subtle changes in speech motor function that occur over successive readings,[136] and there are data to suggest the opposite.[137] There is also some evidence that motor learning may explain it.[138] That explanation is bolstered by evidence that the adaptation effect seems to occur with the "stuttering-like disfluencies" (see Lecture Four) of Parkinson disease.[139] The effect might also be explained if anxiety about speaking systematically reduces after several readings.[140,141]

Even more mysterious …

Making the adaptation effect even more puzzling is the consistency effect and the adjacency effect. The consistency effect is that stuttering tends to occur on the same words during repeated readings of a passage, suggesting anxiety about specific words.[142,143] The adjacency effect is when stuttered words are removed from a passage and it is read again, and stuttering tends to occur on words located near the removed ones.[144,145] Both these effects might be explained in terms of anxiety about certain words.

How stuttering affects people

Treatment seeking

Stuttering can have a lifelong impact, and consequently those affected commonly seek treatment. An Australian survey study[146] of 852 adults who stuttered, with a mean age of 49 years, indicated that 72% of them were assessed by a speech-language pathologist, and 73% received treatment from a speech-language pathologist. As with many health issues, early intervention is a desirable option for stuttering. For 135 children in that survey, with a mean age of 11 years, parents reported that 95% received assessment and 92% received treatment from speech-language pathologist.

This section describes many reasons why those who stutter might seek clinical services. The disorder affects speech, quality of life, educational and occupational attainment, and mental health.

Speech impact

Reduced verbal output

A self-evident but much overlooked impact of stuttering is reduced verbal output. This occurs because the speech behaviours described earlier are time consuming. Those who stutter appear not to say as much as their peers within a given time, or take longer to say it, or a combination of both. According to an early study of the matter,[147] when given a spontaneous speaking task, those who stutter say, on average, around one third less than those who do not stutter. A more recent publication[148] replicated that finding; group of control speakers had a mean of 867 words spoken in 5 minutes compared to a mean of 584 words for a stuttering group, which is one third less.

With severe stuttering, speech rate can be below 50 syllables per minute, which is a speech output of less than a quarter of fluent peers. So a person severely affected for a lifetime may say only a quarter of what is possible, or take four times as long as others to say what is intended.

Variable stuttering severity

Stuttering severity is notoriously variable. A survey of 204 adults,[149] mostly from the United States, indicated that 97% of them experience variability of stuttering severity. Findings indicated this to be the most frustrating aspect of the disorder for them.

Stuttering severity is likely to vary with differing audience sizes and types,[150,151,152] generally with more stuttering as audiences become larger. Stuttering severity varies also across different everyday situations.[153] It seems that there will be more stuttering when speaking to people than when speaking alone, as discussed at the end of this lecture. Experiments that have involved repeated measures of participants in the same speaking situation have shown clinically significant stuttering variability in that same situation.[154,155,156] A study of six participants over five clinic visits spanning 2 weeks [157] showed that in two cases stuttering severity was four or five times greater on some visits than others. There is a report[158] that stuttering changes even with different postures, with more stuttering when lying down than when sitting.

Statistical process control charts are a method of studying variation, and this method has been applied to stuttering.[159] The stuttering severity of 10 adults was studied during the course of their speech during a single day. Results showed that all 10 participants showed predictable variation around their mean severity. However, five of the participants had stuttering severity that was unpredictable across the day, and suggestive of an "out of control system," showing severity scores more than three standard deviations from their means during the day.

Word avoidance

It is well known that those who stutter may attempt to limit the impact of their stuttering on daily life by avoiding words.[160,161,162] Scanning ahead for words that are difficult, and avoiding them with circumlocutions, is a common strategy. However, there are some words that cannot be avoided. Examples are name, telephone number, address, and a destination on public transport.

Grammatical constraints

There have been reports that those who stutter have restricted use of grammar.[148,163] The latter of these papers reported that, compared to controls, those who stutter spoke with fewer clauses per utterance and fewer elaborate clause constructions. Also, the stuttering group used less modality than nonstuttering peers. The term modality, in systemic functional linguistics, refers to "linguistic resources to express opinions, attitudes, and politeness, and therefore potentially engage with conversation partners" (p. 481).[148] A particularly noticeable reduction of modality occurred with interpersonal metaphors, indicating that the stuttering participants were less inclined to project opinions with clauses such as "I believe …" and "I think …" These results, with traditional grammar and systemic functional linguistics, were replicated in a more recent report,[164] where the authors concluded that those who stutter have "a reduced openness to interpersonal engagement within communication exchanges" (p. 536).

A follow-up report on the participants in the latter study after speech treatment[165] indicated some improvement in flexible language use related to interpersonal engagement, but not a complete resolution of the issues. A 12-month follow-up[166] showed these treatment gains to be maintained, with evidence of continued improvement. There is also a suggestion from a study[167] of eight 7-year-old children who stuttered and eight control children that such problems may begin early during the course of the disorder. There has since been initial work to develop a questionnaire assessment of these pragmatic language functions.[168]

Quality of life impact

The importance of stuttering and quality of life was highlighted by an issue of the *Journal of Fluency Disorders* being devoted to the topic.[169,170,171,172]

The quality of life impairment that stuttering can cause is well demonstrated in a film depicting the life of King George VI.[173] The United States President Joe Biden stuttered severely as a child and remains affected as an adult.[174] He described the disorder as "the single most defining thing in my life" [175]

Two studies[176,177] used a medically oriented qualify of life instrument to show that stuttering participants had poorer quality of life compared to controls. One study[177] reported that stuttering affects quality of life as adversely as life threatening conditions such as neurotrauma and coronary heart disease. Presumably this is because, in contrast to those diseases for the most part, stuttering is present across the lifespan. A report has linked the quality of life impairment of stuttering to the lack of speech spontaneity it induces.[178] There is some evidence that stuttering restricts participation in some forms of sport and exercise.[179] There is evidence that teenage girls who stutter are affected more by the disorder than teenage boys.[180]

Another study[181] recruited 78 participants, four of whom stuttered, to a "willingness to pay" and "quality adjusted life years" analysis of the disorder. The nonstuttering participants were provided with detailed information about stuttering, and results indicated that participants would pay "with amounts of money equal to two to four times their annual incomes" (p. 309)[181] for a clinical improvement to mild or "cured" stuttering. Additionally, respondents "equated substantial improvements in severe stuttering with a gain of up to 18 additional years of full-health life" (p. 309).[181] These results were consistent with quality of life impairments measured for serious medical illnesses.

Bandura's[182] notion of self-efficacy refers to the extent to which people believe they will be able to achieve things. With reference to those who stutter, a report[183] of 39 adults found a relationship between high levels of self-efficacy and low OASES impact scores[†]. The effect was found to occur independent of stuttering severity. For those who stutter, self-efficacy was found to relate to a feeling of spontaneity while speaking.[178]

Occupational impact

The modern importance of communication and occupation

During the past century there has been systematic change with how much speech is needed for common occupations, and this has implications for those who stutter. In the United States,[184] 80% of occupations relied on manual skills at the start of the 20th Century, with only 20% of occupations relying prominently on communication skills. By the 1950s the proportion of such "white collar" occupations relying on communication skills had increased to 38% and the figure was 62% at the start of this century. For Australia, in 1966, 45% of occupations were "white collar," rising to 69% in 2011.[185]

Stuttering impairs occupational attainment

Considering the importance of communication in occupations, it is not surprising that stuttering has an impact on occupational attainment. A survey that included 713 adults who stuttered[146] reported that 55% indicated that stuttering had been a barrier to finding employment, and 66% indicated it had been a barrier to career progression. Another report showed that 70% of 200 stuttering adults thought the disorder prevented promotion and 20% declined the challenge of a promotion because of it.[186] One report[187] even indicated that 7.5% of participants had employment terminated because of their stuttering. Another report indicated that speech rehabilitation resulted in improved occupational level and promotion prospects.[188]

The disorder appears to affect everyday experiences in the workplace.[189,190,191] Employers have reported that those who stutter are less employable or promotable than others.[192] Members of the public seem to reflect these attitudes.[193,194] A study of questionnaire responses of 20 adults in Jordan[195] showed that the following beliefs were ranked highly from a list of 10: "you face challenges to get a leadership position because of your stuttering" (ranked first); "employers exclude you from getting a job because of your stuttering" (ranked third). These beliefs seem to occur also in Japanese society.[196] A survey study of human resource management students in the United States[197] found that 42 of 43

[†] OASES is a measure of the impact of stuttering, which is discussed in Lecture Four.

potential careers were thought by the students to be less suited to someone who stutters than someone who does not.

A study of a large British birth cohort[198]—participants studied from birth—supported these findings by indicating that those who stutter are more likely to have lower socioeconomic occupation status than those who do not. A large United States cohort[199] observed from childhood produced more definitive findings: those who stuttered earned less annually compared to controls, and those who stuttered were more likely to be unemployed than controls. The study reported a particular disadvantage for women who stuttered compared to men who stuttered.

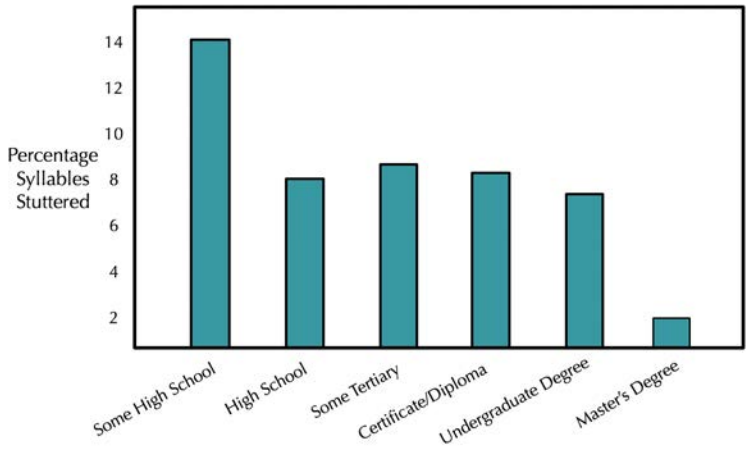
Educational impact

The school years

Some early publications identified education problems for children who stutter during the school years,[200,201] and those results have been replicated in a more recent report.[202] A compelling, large cohort,[203] based on 1988 data from The United States National Health Interview Survey, confirmed those reports. Stuttering school children were significantly more likely to repeat a grade than control children. From six Italian schools, a report[204] studied 52 children who stuttered and 374 controls, aged 8–17 years with a mean age of 11 years. Compared to the controls, there was evidence that the children who stuttered had lower academic outcomes. There is evidence that children experience social isolation in school from fear of speaking in the classroom, and that they habitually avoid it.[205,206,207,208] The extensive clinical and theoretical aspects of this matter are explored in detail during Lectures Nine and Ten.

High school onwards

The birth cohort study mentioned earlier[198] reported that stuttering had no effect on educational outcome. However, there is evidence that the disorder has a negative impact on education attainment. One report showed a negative linear relationship between stuttering severity and education attainment.[209] In other words, there is a tendency for those with more severe stuttering to attain less during education. Those data show that the stuttering of those who do not complete high school may be six times more severe than those who complete a postgraduate qualification. This is shown in the figure below.[210] The vertical axis gives a measure of stuttering severity (see Lecture Four). The horizontal axis shows various levels of education attainment, ranging from partial completion of high school to completing a master's degree at university. The effect in the figure—a significant negative correlation between stuttering severity and educational attainment—was replicated in a survey of 722 adults.[146]



One report211 produced a troubling statistic that the websites of only 13% of 359 public universities provided information about alternative teaching and assessment methods for students who stutter, and

only 51% of the disability liaison officers of those universities responded to an email enquiry about the topic. The authors pointed out that this could disempower potential university students who stutter because they cannot make informed choices about universities with pertinent disability services. Consequently, once at university, they might be unable to optimise their learning environments. A study of questionnaire responses of 20 adults in Jordan212 showed that the following belief was ranked second on a list of 10: "your teachers/professors exclude you from participation in the classroom because of your stuttering." A survey of 246 adults who stuttered and 246 controls[213] in the United States reported that the former group perceived more negative perceptions from their college professors than controls. Additionally, the adults who stuttered felt less comfortable approaching their professors.

Anticipation of stuttering

A common effect

It has been known since the 1930s that those who stutter anticipate its occurrence with some reliability.[214,215,216,217,218,219] This knowledge has been bolstered by reports with adults[220,221,222,223,224] and children[225,226,227] during reading tasks that have established eye gaze patterns consistent with anticipation of difficulty with certain words. All of this knowledge has figured in many of the influential causal theories in the history of thought about the disorder (see Lecture Three): primary and secondary stuttering theory, the Diagnosogenic Theory, approach-avoidance theory, and the Anticipatory Struggle Hypothesis.

The experience of anticipating stuttering

A study of 30 adults[162] reported their experiences of anticipating stuttering, and around half reported "they experience anxiety or uncertainty when they anticipate stuttering" (p. 44). All reported using at least one proactive response to the feeling of anticipating stuttering. For example, "an attempt to hide or escape from an impending moment of stuttering" (p. 42) was reported by 87% of them. Circumlocution, and including something in conversation that was not originally intended, was the most common avoidance response.[†] Consistent with that report, word substitution was independently reported by 82% of another cohort of stuttering participants.[228] Avoiding situations was also a common proactive response to anticipating stuttering.

Self-management strategies, either learned in a clinic or self-generated, were reported by 87% of participants.[228] Those included variants of the speech restructuring technique to be discussed later this lecture, relaxation procedures, and reducing speech rate. Forty per cent of participants reported consciously deciding to not alter speech in any way in response to a feeling of anticipation. The participants indicated that the experience of anticipating stuttering can be helpful to them and also harmful, with 43% reporting that it can be both. However, 37% reported that it is of no help at all and a minority of 13% reported that it is always helpful.

Anticipation of stuttering is connected to what is commonly referred to as *covert stuttering*.[229] This term refers to attempted stuttering concealment using techniques such as word avoidance, circumlocution, and situation avoidance. It seems that it is common for those who stutter to pass through a stage where they initially practice covert stuttering but eventually abandon those efforts.[230,231]

Research has begun[232] to develop an instrument to measure anticipation of stuttering events: the Premonitory Awareness in Stuttering Scale. This 12-item scale was adapted from a similar scale used for tics, and showed that adults who stutter report anticipation of "speech disruptions" more often than control speakers.

[†] Clients commonly report that this can be a functional issue, such as not ordering a particular menu item in a restaurant to avoid stuttering while giving the order to the waiter.

Social anxiety

Situation avoidance

The effects of stuttering on people in the ways just discussed—their speech output, occupational and educational attainment, social stereotypes, and situation avoidance—are probably connected to a common effect of stuttering. That effect is social anxiety, and is considered in detail during Lectures Ten and Eleven. As will be discussed then, a common effect of social anxiety is to avoid speaking situations.

Situations commonly avoided

An early report[233] documented situations that are commonly avoided by those who stutter, using 50 stuttering participants prior to treatment and 100 controls. They indicated their avoidance of 40 standard speaking situations. The following table[‡] presents the top 15 situations that were avoided by the groups, with the most avoided situations at the top of the list. The ranking is ordered according to the stuttering participants.

AVOIDED SITUATION	*STUTTERING*	*CONTROLS*
Asking a question in class	*1*	*2*
Speech to unfamiliar audience	*2*	*1*
Telephoning to make enquiries	*2*	*19*
Short class recitation	*4*	*8*
Reading aloud to friends	*5*	*14*
Introducing one person to another	*6*	*18*
Introducing oneself	*7*	*7*
Telephoning for a meeting or appointment	*8*	*24*
Parlour games requiring speech	*9*	*10*
Telephoning for a taxi	*9*	*26*
Giving your name over the phone	*11*	*21*
Asking for a job	*12*	*6*
Participating in committee meetings	*13*	*12*
Telling a joke to a stranger in a crowd	*14*	*5*
Giving someone a message	*15*	*27*

Most obviously from the table, the telephone was a recurring avoided situation reported by those who stuttered compared to controls. It also seems that those who stutter avoided group speaking situations more often than controls. The report also showed that those who stutter were the most comfortable with people they knew, such as friends and family, and did not commonly avoid those situations.

A more recent publication[234] used another situation checklist[235] and compiled data from 88 adult participants seeking treatment for stuttering and 209 controls. The checklist includes items dealing with emotional responses such as anxiety and worry, and items dealing with the "likelihood of speech

[‡] Adapted and reproduced with permission: Trotter, W & Bergmann, M (1957), Stutterers' and nonstutterers' reactions to speech situations, *Journal of Speech and Hearing Disorders, 22,* 40–45. © 1957 American Speech-Language-Hearing Association.

breakdown." For emotional responses, the three items that best distinguished the stuttering and control participants were "talking on the telephone," "asking a teacher or supervisor a question," and "being rushed." For likelihood of speech breakdown, the three items that best distinguished the two groups were "talking on the telephone," "talking with teachers or supervisors," and "saying a sound or word that previously has been troublesome" (p. 1137).[234]

The telephone`

The problematic nature of talking on the telephone for those who stutter is further shown by that speaking situation being at the top of their hierarchies of feared and avoided situations. One report was a survey of 223 British participants.[236] Those who rated their stuttering to be severe reported making fewer telephone calls per week than those with milder self-ratings of severity. Thirteen per cent of participants reported always using an alternative to the telephone and 55% reported sometimes doing so.

In this report more than a third of those with self-reported severe stuttering said they always used alternatives to the telephone, and more than half reported sometimes having others make calls for them. Sixty per cent agreed with the statement that "it is more difficult to speak to someone on the 'phone than 'face-to-face'" (p. 308–309).[236] Recurring reasons given for this were that nonverbal communication is not possible by telephone, reactions to stuttering are unknown on the telephone, a lack of understanding of stuttering by the conversation partner, and time pressure. Generally, making calls was reported to be more troublesome than answering them. Compared to participants older than 50 years, twice as many participants younger than 30 years reported always using alternatives to telephoning.

Another study was an interview report of 130 stuttering participants.[237] They were asked, "of all your feared talking situations, where would you rank calling on the telephone?" (p. 235). They were also asked about answering the telephone. Overall, 72% ranked making calls among their top three feared situations, and 54% made that rating for answering the telephone. As with the previous report[236] those effects were much more pronounced for severe cases. Participants were given a list of telephone calling options to rate on a fear scale, and the following were the most highly rated: someone from a different culture, the opposite gender, directory assistance, telephone operator, store enquiry, and an older person.

An interesting way to gain insight into these issues has been reported[238] by faking stuttering—often referred to as pseudostuttering—while telephoning a stranger, such as a travel agent or a department store staff member. Twenty-nine graduate speech-language pathology students who did this found the experience rather sobering, with evidence that it may have promoted negative self-perceptions.

Personality

A well-known reference text[239]concluded that "there would seem to be some justification for the inference that stutterers on the average are not quite as well adjusted as are typically fluent speakers" (p. 308). Publications since that review confirm an impression that those who stutter may have unusual personalities compared to those who do not stutter. A report with the Minnesota Multiphasic Personality Inventory[240] showed significant differences between the two groups. Another report[241] compared 93 adults seeking treatment for stuttering with matched controls using a test called the NEO Five Factor Inventory, which assesses five personality domains: Extraversion, Neuroticism, Openness, Agreeableness, and Conscientiousness. Results showed that the stuttering participants were all within the normal range for the five domains but had higher Neuroticism and lower Agreeableness and Conscientiousness scores than controls.

Another report[242] using the same assessment and groups of around the same size of 87, replicated that initial report about Neuroticism but found the opposite for Conscientiousness and Agreeableness, reporting higher scores than controls. Using the NEO Five Factor Inventory again, another report[243] was consistent with the Neuroticism finding by reporting that it correlated with high impact of

stuttering among those who stutter.[†] However, using the same measure with a culturally different population, another study[244] found only higher Agreeableness scores for the stuttering group, but no other differences. Two reports have also linked stuttering to perfectionism.[245,246]

In short, there are inconsistent and slight differences found across studies. Possibly, this is because some personality disorders can be explained simply as the effects of stuttering, and some cannot.[247]

These generally inconclusive findings about stuttering and personality are consistent with the inconclusive results of a study[248] dealing with a related construct: temperament. The researchers concluded that, from among 13 aspects of temperament, there is "a nontrivial tendency for AWS *[adults who stutter]* to experience decreased positive affect compared to AWNS *[adults who do not stutter]*" (p. 2691). The topic of temperament and stuttering during the early years of life is covered in detail in Lecture Ten.

A review of qualitative research about experiences with stuttering

There is a body of literature involving interviews with those who stutter to establish information about their experiences with the disorder. This method generates information in a much different way to most of the studies mentioned so far during this lecture. A synthesis of such qualitative reports since 2000[249] involved 17 studies that met methodological criteria. The authors reported that five themes figured prominently in that literature:

> (1) Avoidance is used to manage stuttering …
> (2) Stuttering unfavourably impacts employment experiences …
> (3) Stuttering shapes self-identity …
> (4) Stuttering leads to negative reactions … both actual and perceived …
> (5) Stuttering impacts relationships adversely. (p. 2237–2239)[249]

Stuttering stereotypes

A significant topic

Many research publications have shown that those who stutter are affected by negative stereotypes about their disorder.[250,251,252,253,254,255,256] This appears to be true across the lifespan from childhood to adulthood.[257,258,259] The topic was considered to be of sufficient importance to warrant a conference about it.[260]

Fictional stereotypes

Stuttering is frequently depicted in movies and television, and more often than not, with a negative stereotype.[261] A review of 29 works of fiction that contained a character who stuttered[262] indicated that "most often, characters who stuttered encountered mean-spirited teasing, name-calling, demeaning remarks or bullying from one or more of the characters" (p. 617). A detailed examination of stuttering portrayed in film[263] typically shows it to be used in a negative fashion, for example, as a comic device or as a sign of weakness.

Responses to stuttering

A report[264] collected listener responses to speech read by participants who stuttered and controls. Without any identifying information about members of the former group, the listeners judged them to be less intelligent, less likeable, and more anxious than the controls. A study of 324 adults from the United States,[265] two thirds of whom were men, involved a list of 15 stigmatising experiences. For most of these experiences, the majority of participants reported having experienced them during their lifetimes. Interestingly, "most participants reported experiencing them never or rarely in the past year" (p. 55). An interview with seven adults who stuttered[266] raised the issue of those who stutter

[†] Neuroticism is a tendency to experience high levels of negative emotion such as anxiety, anger and sadness. Impact of stuttering was measured with the OASES, which is discussed in Lecture Four.

experiencing microaggressions, which are subtle manifestations of negativity about marginalised groups.

During adolescence, it seems that peers find those who stutter "nervous" and less attractive than others.[267,268,269,270,271] One of these findings[271] was that 736 adolescent and young adults stated that images of young people labelled as a person who stutters were less physically attractive than images without that label.

A cross-cultural issue

There is compelling evidence that communities of various cultures hold negative stereotypes about those who stutter. A review of that evidence[272] stated,

> the public view of stuttering is generally unfavorable and … listeners often ascribe negative traits like anxious, shy, nervous, unassertive or introverted to people who stutter. (p. 54–55)

Since this review, publications have reported stuttering stereotypes in the United States,[273] Polish,[274] and Chinese populations.[275] As noted earlier, this stereotyping extends to occupational suitability.[193,194,273] There is evidence that those who stutter may hold self-stigmatising thoughts in response to such community attitudes, and that those self-stigmatising thoughts may contribute to psycho-social harm.[276]

Stereotypes and speech-language pathologists

Added to this literature is a recurring finding over several decades implicating speech-language pathologists in the perpetuation of negative stereotypes about the disorder. Clinicians in the United States,[277,278] the United Kingdom,[279,280,281] Korea[282] and Turkey[283] are reported to have negative and empirically unjustifiable attitudes to stuttering, although the research shows some signs of improvement of this problem over time. One report[284] raised an interesting suggestion that negative attitudes toward stuttering by speech-language pathologists are not necessarily revealed with standard, overt testing methods; there may be implicit attitudes that emerge with another means of testing.

POSHA-S

A long-standing project has used a tool called the *Public Opinion Survey of Human Attributes-Stuttering* (generally referred to as POSHA-S) to explore worldwide attitudes to the disorder. A summary of a series of European reports[285] from 1,111 respondents found some variation, including less positive than average attitudes in Italy and more positive than average attitudes in Norway and Sweden. That international database was used to report that stuttering seems more stigmatised than obesity but less stigmatised than mental illness.[286] Subsequently, reports dealing with attitudes in Australia,[287] Turkey,[288] Portugal[289] and Egypt[290] have been published. A report with the POSHA-S[291] showed that British, Arabic and Chinese students enrolled in a British university had stereotypical stuttering attitudes, and that their attitudes differed based on their home cultures. A report with that database[292] showed that protective services workers—police officers, fire-fighters, security guards—had more negative beliefs about those who stutter than other occupational groups. Using a different survey instrument, another group[293] reported that awareness and accurate knowledge about stuttering was limited in Japan.

Dealing with stigma about stuttering

Another report[294] presented information about the relative merits of different approaches to dealing with stuttering stigma, based on procedures used for dealing with stereotypes about mental illness. The first approach to dealing with stigma about stuttering is to provide public information about what it is like to experience the condition. The second approach is to provide public education based on the traditional "fact and fiction" approach about a disorder. Finally, the protest approach draws attention to the injustice and inappropriateness of stigmatising a condition. The report found that all three approaches had value for reducing stereotypes about stuttering.

Stereotypes and teachers

Some reports suggest that United States teachers of children younger than 12 years,[295,296,297] special educators in schools,[298] and school administrators[299] may well hold negative and unjustifiable stereotypes about the disorder. More recent reports from the United States[300] and India[301] were more favourable, although another report showed that United States teachers did not have more accurate beliefs about stuttering than the general public.[302] A study of New Zealand teachers[303] found they did not have negative attitudes about the disorder, but they did have knowledge gaps about its causality (causality is discussed in Lecture Three). A study of 74 early childhood educators in Greece[304] reported that they had overall positive attitudes toward 2–5 year-olds who stuttered. The report also indicated a relationship between knowledge about stuttering and positive attitudes to it.

A recent report[305] of 262 Kuwait teachers and 209 trainee teachers was rather sobering. The report found, among other things, that 81% of the teachers and trainee teachers believed stuttering to be caused by emotional problems, 76% believed it to be precipitated by "a very frightening event," and 15% believed that "a virus or disease" (p. 60)[305] was responsible. Additionally, 20% of them indicated that "people who stutter should try to hide their stuttering," 72% believed those who stutter to be "nervous or excitable," 82% believed they were "shy or fearful," and 35% indicated that those who stutter "have themselves to blame for their stuttering" (p. 61).[305] A follow-up study[306] showed that such stereotypes could be corrected during teacher training with a 17-minute educational video about stuttering. A report of interviews with 10 Belgium teachers of adolescent students[307] showed that they felt stuttering could become a problem if attention is given to it, and that they tried to minimise any reaction to stuttering and rarely talked about it in class.

Stereotypes and universities

There is some evidence that stereotypes about stuttering extend to university environments. University students have been shown to have negative attitudes to the disorder.[308,309] One report[310] showed that university professors and students scored students who stutter as having more negative personality traits than other students. However, another report[311] of student perception failed to find overriding stereotypes, and another found "neutral to positive perception" (p. 206)[312] of students who stuttered. Speech-language pathology students have been shown to have more positive attitudes toward stuttering than others, with some evidence that there may be differences across countries.[313] A report of students from an Australian university[314] indicated a positive attitude toward stuttering, and suggested a connection between that result and curriculum content. An experimental report of professor evaluations of oral student presentations found a that, in some situations, professors may compensate for students who stutter.[315] This occurred in cases where a student stuttered and also had poor communication skills.

Self-disclosure and stereotypes

Some researchers[316] have made the important point that, in addition to being a disorder that is stigmatised, stuttering is not always readily apparent because it can be so variable from situation to situation (as discussed during Lecture One). Further, it can be actively concealed, such as by means of "covert stuttering" described earlier. With a survey of 505 adults who stuttered, the researchers presented data showing that the extent to which stuttering was concealed adversely impacted quality of life.

The opposite of concealment is to self-disclose a disorder. In the case of stuttering, self-disclosure is when, at the start of a social interaction or speaking to listeners for the first time, speakers declare that they stutter. There is evidence that self-disclosure can positively influence listener reaction to stuttering,[317,318,319,320,321,322] with only one report of no effect.[323] Self-disclosure by a 12-year old and his teacher was shown to be beneficial.[324] A study reported that favourable perceptions about a 12-year old were associated with a written disclosure statement by his mother, rather than by his teacher or by himself.[325] There is some evidence to associate self-disclosure with speaker perceptions of speech spontaneity.[178]

Much of the literature about self-disclosure suggests that the procedure might be used to deal with anxiety about speaking.[317,318,320,323] In which case, from the perspective of clinical psychology, there is a reservation about its use (a potential safety behaviour), which is discussed during Lecture Ten.

Guidelines for interacting with those who stutter

An important topic

According to one publication, there is no shortage of recommendations to the public about how to interact with those who stutter.[326] Yet, as noted in this publication, little of the advice has been generated by those who stutter. Consequently, the authors elicited the views of 148 adults, most of whom had received treatment or support from a self-help group for stuttering. Two thirds were men. From a list of 24 items, the following three actions were rated most highly supportive, in this rank order:[326]

(1) "maintain eye contact"
(2) "wait to let a PWS *[person who stutters]* say what he/she wants to say"
(3) "assuming the listener also stutters, to ask the PWS how they can help the listener with his/her own stuttering problem" (p. 5)

The following three actions were rated as least supportive:

(1) "'faking' stuttering during conversation"
(2) "telling the PWS *[person who stutters]* how he/she should feel about the problem"
(3) "trying to 'help' the PWS by finishing stuttered words" (p. 5)

From written responses to a question about desirable and undesirable responses during communication, the following nine actions were listed as supportive by at least 10 respondents, in this rank order:[326]

(1) "maintaining eye contact"
(2) "being patient, understanding, sensitive, friendly or non-judgemental"
(3) "listening"
(4) "asking about stuttering"
(5) "allowing the PWS *[person who stutters]* to finish his or her words or sentences"
(6) "showing empathy, interest, compassion, or respect"
(7) "treating the PWS normally"
(8) "engaging him or her in conversation"
(9) "helping with the stuttering" (p. 8)

Ten or more respondents considered these two actions not supportive:[326]

(1) "finishing one's words or sentences"
(2) "ridiculing one's stuttering (e.g., making fun or mocking)" (p. 8)

Two caveats about eye contact

The authors presented two caveats about the recurring finding (shown in the list above) that respondents indicated eye contact to be the most supportive action by conversational partners. First, it may cause discomfort during conversation with someone who stutters by creating a feeling of "staring." This is a justifiable concern, considering that extended stuttering moments may elicit unusual eye contact patterns. Second, eye contact is not desirable in some cultures.

Conditions that reduce or eliminate stuttering

The fluency inducing conditions

Changing customary speech

A fascinating feature of the disorder is that speakers can change how they speak in certain ways and this can reduce stuttering or even get rid of it completely while they are using those speech changes.[327] Sometimes the changes can be subtle. For example, it is common to hear of actors who stutter but do not do so when they are on stage. Presumably, part of the explanation for this is the change to customary speech while on stage: louder, slower, perhaps with a different accent, and so on.[†]

The Modified Vocalisation Hypothesis

There are some well-known changes to the customary way of speaking that are not so subtle as when actors speak on stage. The term "fluency inducing conditions" is attributed to Wingate, who proposed a Modified Vocalisation Hypothesis to explain why they reduce stuttering.[328] Wingate proposed that all fluency inducing conditions can be explained because "speaking under all of these conditions emphasizes vocalization and continuity of vocalization" (p. 682).[328] There is much research about these conditions, which is outlined in a reference text (Chapter 11).[239] Some of the common fluency inducing conditions are described below.

Singing

Arguably the most commonly known feature of stuttering is that it goes away during singing. There are some who question whether this always happens (p. 425),[329] but it is generally accepted as a feature of the disorder. Singing has never been directly linked to the development of a treatment method. However, an acoustic analysis of adolescents who stutter and controls during singing[330] showed changes consistent with a popular treatment method to be discussed shortly: *speech restructuring*. In short, singing stabilises and simplifies speech motor activity. (And, as with acting, the words of a song are not spontaneous, but are known in advance.)

Rhythmic speech

Speaking in time to a rhythm has a similar universal effect on stuttering. This has been the source of many therapy techniques during past decades, and even past centuries according to common belief. Writings by Plutarch in 75 BC convey that, Demosthenes, a famous Greek orator who lived during the third century BC, stuttered and consulted the Greek actor Satyrus, who prescribed that Demosthenes should run or walk uphill while speaking, among other treatments.[331] This is commonly interpreted as the first therapeutic use of rhythm to reduce or eliminate stuttering.

Since then there have been many clinical applications of rhythmic speech. Many have not been particularly successful, or, just simply dubious. An example of the latter are the now infamous "stuttering schools" that proliferated in the United States during the first part of the 20th Century.[332] Miniature in-the-ear metronome devices also emerged during the 1970s[333] but never attained any demonstrable success.

There has been a great deal of research about the rhythm effect, dating from the first half of the last century.[334] A recent, sophisticated functional magnetic resonance imaging report[335] provided the first neurophysiological details about how rhythmic speech might control stuttering.

[†] Another part of the explanation is that actors on stage know what they are about to say; there is nothing spontaneous about their speech..

Reduced speech rate

Virtually everyone who stutters will report being told at some time to "slow down." This advice is presumably based on an assumption by casual observers that the problem of stuttering arises from attempting to speak too quickly. Reduced speech rate is a component of many modern treatments.

Of itself, however, the speech rate reduction needed to attain clinically useful stuttering reduction may not be functionally useful. This contention is supported by a report[336] that a 30% reduction of reading rate did not significantly reduce stuttering. For the severest of the participants, the 30% speech rate reduction reduced stuttering severity by 35%, and left the participants with considerable stuttering.

Chorus reading and shadowing

When someone who stutters reads in chorus—that is, at the same time—with someone who does not stutter, stuttering disappears during the reading. Even more curious, if someone who stutters repeats what another person has just said during a spontaneous monologue, but a few words later, the same thing occurs. The former condition is called *chorus reading* and the latter condition is called *shadowing*. There is some evidence that the Modified Vocalisation hypothesis could explain the chorus reading effect.[337] There is also evidence that chorus reading might be explained by a rhythm effect where the speech pattern of the reader who stutters is influenced by the reader who does not stutter.[338] Neither of these speaking conditions has influenced modern treatment practices.

Verbal response contingent stimulation

Response contingent stimulation research with stuttering

During the early 1950s, and ending some decades later, there was a series of laboratory experiments showing, in short, that if those who stutter receive electric shock or a loud noise after stuttering moments, then their stuttering decreased, and in some cases stopped altogether, only to return when the shock or noise ceased. There are at least 50 publications to that effect, dating from the early 1960s.[339,340,341,342,343,344,345] The shock or noise is called *response contingent stimulation of stuttering*. The results of those experiments show that stuttering has operant features.

A disorder with operant features

It is important to state that stuttering has operant features, not that stuttering is an operant. If a behaviour is freely emitted and readily controllable, and changes with response contingent stimulation, then the behaviour is referred to as an operant. However, stuttering is not a freely emitted problem behaviour. As will be discussed during Lecture Three, stuttering is atypical neural processing of speech, which is beyond the control of those affected. A treatment for stuttering that incorporates response contingent stimulation can be referred to as a treatment with operant methods, or an operant treatment.

Verbal response contingent stimulation of stuttering

Laboratory research of shock and loud noise with stuttering stopped during the mid-1970s. However, it did lead to the discovery that response contingent stimulation of stuttering could be verbal, and could functionally control stuttering.[346,347,348] Research showing that this was an option for children[349,350,351] established clinical possibilities that have been fruitful, particularly for treatment of early stuttering, as will be discussed during Lectures Six and Seven. In short, a treatment based on parent verbal response contingent simulation—the *Licombe Program*—has been shown with many clinical trials to be efficacious.

There is some evidence, albeit patchy,[352,353] that the Modified Vocalisation Hypothesis might explain the verbal response contingent stimulation effect. There is also evidence, again not particularly compelling, that an explanation might be that the verbal response contingent stimulation reduces stuttering by inducing simplifications to spoken language.[354,355]

Auditory feedback

Delayed and altered auditory feedback, and speech restructuring

These terms refer to when airborne speech feedback is altered with an electronic device by means of unilateral or bilateral headphones or an in-the-ear device similar to a hearing aid. The first such effect to be discovered was *delayed auditory feedback*,[356,357] often referred to as DAF. Subsequent to a famous report of it being used to reduce stuttering,[358] this discovery prompted much research that continues to the present, and has profoundly influenced treatment practices. Generally, delayed auditory feedback creates a slow and unusual drawling speech pattern that reduces or eliminates stuttering. This is the basis for the most popular of modern treatments for adolescents and adults who stutter. It is rare for delayed auditory feedback devices to be used clinically these days, and clinicians simply teach those who stutter how to use a novel speech pattern to reduce or eliminate stuttering. These treatments, discussed during Lecture Eight, are referred to generically as *speech restructuring*.[359] In short, speech restructuring treatments have been shown with many clinical trials to be efficacious.

For nonstuttering speakers, delayed auditory feedback can induce disfluencies that once were thought to resemble stuttering,[360] and this prompted many theories that stuttering was caused by a problem with speech feedback. However, it is now accepted that these disfluencies are not stuttering. Potentially, delayed auditory feedback devices are problematic, because there have been reports of them[329] inducing transient speech problems (p. 372–373).

Altered auditory feedback devices are a modern development of delayed auditory feedback. In addition to delaying speech feedback, these devices alter pitch upwards or downwards. Such devices are commercially available, but their clinical value appears to be questionable at present, as discussed during Lecture Eight.

Masking

Stuttering is significantly reduced or eliminated when the speaker's voice is not fed back because of noise—commonly white noise—presented through earphones. To return to Demosthenes, there are some sources that suggest Satyrus prescribed that Demosthenes also practise speaking on the seashore above the noise of a roaring ocean. It is tempting to speculate that Satyrus thus discovered and found a clinical application for the masking effect in, addition to the rhythm effect.

Speaking alone

Intriguingly, historical textbooks in the field have claimed anecdotally that a defining feature of the disorder is that stuttering does not occur when speaking alone without a listener.[361,362,363,364] Empirical reports confirm that there will be more stuttering when speaking to people than when speaking alone.[365,366,367] However, only one study has observed participants speaking when they are deceived into believing they are speaking alone.[368] Under those conditions, almost no stuttering occurred. There is much experimentation needed into this effect because of its potential clinical importance. As discussed during Lecture Ten, social anxiety is strongly associated with stuttering. Further laboratory experimentation using control groups is needed to determine whether the speaking alone effect occurs because social anxiety is removed altogether, or whether it occurs for other reasons. As the authors of the latter paper[368] note, other explanations are possible. For example, changes to customary speech could occur linguistically, or with changes to speaking volume or speech rate.

Summary

The disorder of stuttering can be associated with potentially confusing terminology that is best to avoid. It is a clinically useful idea that the disorder involves moments of stuttering that interrupt speech. There is no all-purpose definition of stuttering, but three common definitions can be used in different clinical contexts. The observable behaviours of stuttering are many and complicated, so it is clinically important to have ways to describe them clearly. The distribution of stuttering moments during spoken language is generally influenced by initial word consonants, and those who stutter commonly find that certain sounds are often stuttered. Stuttering affects quality of life across

educational, occupational, and mental health domains. The disorder commonly causes social anxiety, which is connected to situation avoidance. Those who stutter are marginalised in society by negative stereotypes. There are well established guidelines for how to interact with those who stutter. There are many conditions that reduce or eliminate stuttering, and many of those are used in successful treatment methods.

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LECTURE TWO: MORE BASIC INFORMATION

Early and persistent stuttering: A fundamental clinical distinction[†]

Early stuttering

Overview of early stuttering

For the purposes of these lectures, early stuttering is the stage of the disorder during which there is a chance that recovery from its problem behaviours will occur, either because of natural recovery, treatment, or a combination of both. The period of early stuttering begins at onset and extends for some years after that.

Treatment of early stuttering

Treatment for early stuttering is intended to remove the problem behaviours of the disorder.

Tractability of early stuttering

Early stuttering appears to be extremely tractable; in other words, responsive to treatment. Many children recover naturally without any formal treatment, as discussed later during this lecture. It is possible—even likely—that early treatment facilitates natural recovery.[1] As will be discussed during Lecture Nine, it seems that stuttering starts losing clinical tractability at some time during the period when children are 6–11 years old. A review of treatment reports available at the end of the last century[2] concluded this about the need for early intervention: "treatment after more than 15 months have elapsed does not appear to have been as effective … as treatment initiated sooner" (p. 223).

Parent contact with children

Formal education in most English speaking countries begins between the ages of 4 to 6 years. Parents generally have most contact with their children prior to this age. It is true, though, that many children with early stuttering will attend pre-school, kindergartens, or day care centres, for all or part of the week. Parent contact with children during each day is a clinically central feature of early stuttering intervention. Parents do the treatments outlined during Lecture Six when they are with their children.

Early treatment is the best option

As discussed during the previous lecture, educational and occupational limitations are often encountered by those who stutter, and those limitations are likely to be associated with social anxiety. As will be discussed during Lecture Ten, there is good reason to believe that negative peer conditioning during the years of early stuttering is implicated in the origins of social anxiety for those who stutter. Considering this, and considering that the disorder is at its most tractable during that period of life, early treatment is clearly the best option.

Persistent stuttering

Overview of persistent stuttering

For the purposes of these lectures, persistent stuttering is the stage of the disorder during which there is no reasonable chance that recovery will occur, either because of natural recovery, treatment, or a combination of both.

Persistent stuttering lasts a lifetime, and, after a period of early stuttering, may worsen throughout life. This is apparent from comparing data sets that measure stuttering severity during the years of early stuttering and during adulthood.[3] The majority of studies suggest that stuttering, and the various problems associated with it, do not abate with advancing age.[4,5,6,7] Conversely, there have been

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[†] Thanks to Robyn Lowe, Sue O'Brian, Ann Packman, and Ross Menzies for assistance with this material.

suggestions that stuttering decreases in prevalence and severity with older age.[8,9] A cross-sectional survey[10] of 852 adults, with a mean at of 49 years, and 135 children, with a mean age of 11 years, collected self-reported severity ratings using a 9-point scale. Results showed a pattern of increased scores up to 18 years of age, followed by a systematic decrease of around two scale values during subsequent decades of life (Figure 2, page 6). A study of 3,100 stuttering moments from 147 adolescents and adults[11] reported that, during that span of life, stuttering became more behaviourally complex. With advancing age, repeated movements, fixed postures, and superfluous behaviours tended to appear.

Treatment of persistent stuttering

Treatment for persistent stuttering is designed to help clients control stuttering or deal with its effects, or both. Those affected by persistent stuttering may or may not seek such treatment or any form of support to deal with its effects.

Identifying stuttering

Clinical identification of stuttering

Persistent stuttering

Generally, clinicians don't need to diagnose stuttering in a clinic and tell people that they have the disorder. Those who have stuttered for much of their lives will be fully aware of it. So, those with persistent stuttering who present to clinics seeking help will nearly always be correct that they stutter. The only clinical task is to confirm the presence of stuttering rather than some other disorder, as discussed shortly.

Early stuttering

During the 1980s and 1990s many protocols were developed for distinguishing between stuttering and normal disfluency during the early years of life.[12,13,14,15,16,17,18,19,20,21,22] This topic was considered so important that a prominent clinical journal published two reviews in this area during the early 1990s.[23,24] However, during this century there have been no further empirical developments or reviews published about differential diagnosis, which might reflect that such protocols are currently thought to be clinically unnecessary.[25] Some authorities in the field, after an earlier attempt to develop a differential diagnostic protocol,[18] have endorsed such an opinion:

> In our experience, the identification of early stuttering in clinical settings is seldom difficult. We wonder why several authors … have expressed a different opinion, emphasizing the great overlap and possible confusion between early stuttering and normal disfluency, and cautioning clinicians of the difficult task. (p. 214–315) [30]

A report published shortly after this statement confirmed it. Children who stuttered and a control group were studied speaking with parents and clinicians at home and at the clinic, with the conclusion that

> a clinician could, with some degree of confidence, predict whether a diagnosis of stutterer or nonstutterer based on a typical clinician–child conversation in a clinical setting would hold true in other environments. (p. 208)[26]

Consistent findings, attesting to the ease of stuttering identification during its early stages, emerged from a study of nine 3–5 year old Icelandic speaking children.[27] The researchers divided 7-minute speech samples from each of them into 5-second intervals and presented them to English and Icelandic clinicians. Neither group had any difficulty identifying which of the 5-second speech samples contained stuttering. However, an earlier study of children with early stuttering presented different findings:

> situational variability can make it more difficult for clinicians to correctly identify and document a child's need for treatment based upon objective measures of the child's speech fluency collected in a single speaking situation. (p. 199)[28]

Early identification errors

There have been no studies designed specifically to determine how often there is a stuttering identification error when very young children are brought to clinics. However, one discussion of early stuttering identification noted of one specialist clinic that

> 1,140 assessments for stuttering were conducted during the period 1994 to 2000, and a file audit showed that only 10 preschool children referred during that period (0.9%) were not identified to be stuttering. (p. 25)[25]

So, if that information is correct, for every 100 children brought to experienced clinicians for a consultation about stuttering, there would be uncertainty about one of them.

There are sources of anecdotal support from experienced clinicians and researchers about the accuracy of parent stuttering identification.[29,30] For example, a prominent authority stated, "indeed, I can recall only a handful of parental misdiagnoses of early childhood stuttering in more than 35 years of clinical practices in its identification and treatment" (p. 6).[29] Another authority stated that "typically, parents of young children who stutter correctly diagnose the problem, making the professional evaluation a task of describing and quantifying the disorder rather than differentiating it from other disorders" (p. 313).[30] In that context, it is of interest that a survey report of United States paediatricians[31] showed that it will not necessarily prompt a referral to a speech-language pathologist if a parent reports that a child "may be exhibiting signs of stuttering."

On balance, it seems reasonable to state that parents generally know that their children have begun to stutter when they bring them to clinics. Sometimes doctors, or staff at pre-school day care centres or kindergartens, identify children who stutter and prompt parents to bring them to clinics. The clinical task of verifying the presence of stuttering is not challenging.

Stuttering identification across languages

There is evidence that Dutch speakers can recognise the disorder in Brazilian and Portuguese speakers.[32] In another study,[33] English-Spanish bilingual and English-speaking monolingual observers, who were speech-language pathologists, identified more "disfluencies" in Spanish video speech samples than English samples by the same speaker. It is not clear in this report, but the speaker appears to have been a nonstuttering adult. A study of 18 nonstuttering Spanish-English bilingual 5- and 6-year-olds[34] produced a consistent result based on audio recordings of their narratives. More disfluencies were identified that are normally associated with English speaking children of that age.

Another report[35] involved video recordings of readings by two Spanish-English bilingual participants who stuttered and English-speaking speech-language pathologists. For one of the participants, but not the other, the speech-language pathologists noted a higher frequency of stuttering in Spanish than English. However, overall, the authors interpreted findings to "suggest that SLPs *[speech-language pathologists]* can accurately assess and diagnose stuttering in clients from culturally and linguistically diverse backgrounds" (p. 40).[35] A further report[36] involved two Spanish-English bilingual 6-year-olds, one who stuttered and a control who did not. Based on audio recordings, many Spanish-English bilingual speech-language pathologists diagnosed the control child as stuttering. The authors concluded, "it appears that bilingual speakers may be at unique risk for false-positive identification of stuttering" (p. 72).[35] However, although one of those authors has speculated about diagnostic issues with Spanish children with early stuttering,[37] there are no convincing data about the matter as yet.

Screening for early stuttering

The benefits of early stuttering screening

As with any early childhood health problem, there would be benefits of population screening to identify stuttering as soon as possible after onset. In principle, that screening would allow cases to be identified for clinical management at an optimal period during early development of the disorder during the first years of life.

Screening sensitivity and specificity

Screening is not assessment. It is typically a much briefer procedure than assessment, and it is designed to determine who should have an assessment and who should not. *Sensitivity*, also known as the *true positive rate*, is the proportion of cases correctly identified as stuttering at screening, according to the results of a full assessment. *Specificity*, also known as the *true negative rate*, is the proportion of cases correctly identified as not stuttering at screening, according to the results of a full assessment.

Errors are inherent with any screening process, false negative identification being the most serious of them. This occurs when a stuttering child is erroneously identified as not having the disorder at screening; in other words, when a child really is stuttering but is not identified as such. The clinical issue here is that when it finally becomes apparent that an error has occurred, and that the child really is stuttering, the optimal time for stuttering treatment may have already passed.

False positive identification occurs when a child is erroneously identified as having the disorder at screening. In other words, when a child is not really stuttering but is identified to be stuttering. Such an error is unlikely to do any harm because the mistake would become apparent as soon as an attempt at treatment began.

To return to the issue of how accurate parents are at identifying early stuttering when they bring children to clinics, the matter can be restated in the following way if parents are thought of as a screening procedure: there is good reason to believe that parent identification of early stuttering is sensitive, with a high true positive rate; however, the specificity of parent identification of early stuttering is unknown. In other words, the false negative rate is unknown.

There is currently no accepted screening method

Much as it is needed, at present there is no generally accepted way to screen for stuttering during early childhood.[38] Surprisingly, such an important topic has attracted almost no research, with apparently only one preliminary report from more than 20 years ago.[39]

Conclusions

With adults and adolescents there would rarely, if ever, be a need for a clinician to make a diagnosis of stuttering. With early stuttering, there seems to be no justification for any more than a case history and observation of a child's speech to diagnose early stuttering when parents bring children to a clinic. Spanish is one of the most common languages; hence, it is of interest that normal disfluencies may be more prevalent in that language than with English. Although there has been some speculation about the implications of this for early diagnosis, at present there is no empirical reason to believe that diagnosing early stuttering in Spanish is clinically problematic. There is no generally accepted method for screening early stuttering.

Speech and language disorder comorbidity

An ambiguous literature

Some research has reported how many children have stuttering and another speech or language disorder, or have stuttering comorbidities, to use the correct term. There is no doubt that this will occur sometimes.[40,41,42,43] A study of clinicians[44] indicated that 44% of 467 school-age children who stutter reportedly also had a language or phonological disorder. Another study[45] reported that 34% had articulation disorders and 14% had phonological disorders. However, another report[46] found no such difference between stuttering and control children. A recent report[47] studied 58 stuttering children and 40 control children for a 4–5 year period. No systematic differences were reported for phonology across the period of study. A recent review of the literature[48] concluded that research about the topic is ambiguous. Given such ambiguity in the literature, it is not surprising that clinicians are uncertain about concurrent management of children with comorbid stuttering and speech sound disorder.[49]

The same ambiguity pertains to findings about language problems with early stuttering and school-age[†] children. Many reports have found that children who stutter have language less advanced than peers[50,51,52,53,54,55,56,57,58,59] and many have found that not to be the case.[60,61,62,63,64,65,66] The 4–5 year study referred to previously[47] found that the children who stuttered "though within normal limits" were "slightly behind … on broad measures of language development" (p. 23). The ambiguity of this literature is highlighted by two reviews of the available literature at around the same time that came to opposite conclusions. One report[57] was a meta-analysis[†] of 22 studies with a conclusion that stuttering was associated with lower language test scores than control children. However, two more recent reviews of the literature[66,67] reported that available research did not support any such conclusion. At present, little is known about bilingualism and childhood stuttering.[68] A review[69] of an often-cited seminal study from early last century,[70] which purported to implicate bilingualism in stuttering onset, demonstrated that its results were not believable.

To make the literature even more difficult to interpret, some authors[71] pointed out that no study of language and early stuttering had used "conversational language samples collected in a naturalistic, non-contrived play environment with peers" (p. 649). They developed a method to rectify that situation and showed that it was viable with four children with stuttering in pre-school play environments.

Possible bias

An issue here is that children who have comorbid speech and language disorders with stuttering are more likely to be referred to a clinic than children who stutter but have no other speech and language disorders. Therefore, published figures could well be overestimates of stuttering comorbidity for the disorder in general.[43] Also, as considered during Lecture Ten, children who stutter could be socially withdrawn. This could be another source of bias because they may be reluctant to speak during language testing, leading to underestimation of their language skills.[60]

ADHD comorbidity

An even more ambiguous literature

There have been some suggestions during the past two decades of an association between attention deficit hyperactivity disorder (ADHD) and childhood stuttering. In 2003, a tutorial about ADHD and stuttering[72] was based (inexplicably) on two research findings at that time which do not seem at all remarkable. The first[73] was a survey of 241 speech-language pathologists about 3–20 year-olds they were treating for stuttering. Their reports suggested that 3% of their caseloads may have had ADHD, which was well within community prevalence rates. The second paper[74] prompting the 2003 review was a report of 50 children who stuttered whose parents used a survey to establish that 26% of them could be described with the vague term "attending disorder."

Subsequent reports have not done much to clarify this initial confusing picture, because they have not incorporated generally accepted diagnostic procedures for ADHD. Instead, they report about children with and without "ADHD symptoms" based on parent screening methods,[75,76,77] teacher reports,[78] or adult recall of childhood symptoms.[79] A report of 356 adults[80] found that they were likely to indicate experience of ADHD symptoms, but no diagnoses were included in the report. A report of 3–17 year olds from the United States National Health Interview Survey (N=62,450)[81] involved children whose caregivers reported stuttering during the previous 12 months, and reported a doctor or health professional diagnosing ADHD. The boys in the sample had greater odds than girls for a diagnosis of ADHD.

[†] The term school-age refers to children who are at the stage of education commonly referred to as primary school or elementary school, spanning the age range 7–12 years.

[†] Meta-analysis is a systematic review that synthesises evidence from numerous empirical reports.

Another report,[82] used 84 adults with a confirmed ADHD diagnosis and 207 controls, and found stuttering in 18% of the former group. A problem with this report, however, is that methods to identify stuttering were not specified. Regardless, 2% of the control group were reported as stuttering, which seems reasonably accurate, as will be discussed later during this lecture.

On balance, all this research is worth noting for two reasons. First, ADHD involves impaired regulation of attention and behaviour, which is a topic that features in research about early childhood temperament and stuttering, which will be considered during Lecture Ten. Second, the topic is clinically pertinent because, as will be discussed during Lectures Six and Seven, many treatment methods for children require considerable compliance from them, and this might be challenging for children with ADHD. Indeed, one of the reports just mentioned[77] found that children with early stuttering required more time to complete stuttering treatment if they had more "ADHD symptoms."

Legal stuttering identification

Clinicians may be asked to provide a written report to a lawyer, or give verbal evidence in a court. They might be asked to comment on a claim that stuttering began after a physically or psychologically traumatic event. Or, they may be asked to comment on a case of suspected malingering. For example, people who have been heard to not stutter while committing a crime have been known to fake stuttering to give the impression that they could not have been the offender.[83] Publications are available to assist clinicians with preparing such legal assessments.[83,84,85]

Theoretical perspectives about stuttering identification

The Continuity Hypothesis

The previous assertion that stuttering identification is not a clinical challenge is based on the idea that stuttering and normal disfluency are categorical things; they are different and, hence, for the most part easily recognisable. But a different perspective about this emerged in 1970[86] in the form of what is known as the Continuity Hypothesis. In effect, this idea is that stuttering and normal disfluency are not categorical things, but lie on either ends of a continuum. In other words, stuttering is an extreme form of normal disfluency:

> there are few if any aspects of early stuttering which cannot be found occasionally and mildly in the speech of most normal young children. Seen from this point of view, stuttering as a clinical disorder is largely a more extreme degree of certain forms of normal disfluency. (p. 30)[86]

This proposition proved to be rather controversial, with an experiment shortly after purporting to show that it was wrong,[87] and that stuttering and normal disfluency in fact were "two reliable and unambiguous response classes" (p. 691). There was disagreement about the experiment,[88,89] and some years later another experiment came to the opposite conclusion.[90]

The Continuity Hypothesis still sometimes appears today in peer-reviewed clinical journal publications. For example, "parents learn which types of disfluencies are typically associated with childhood stuttering … and which disfluency types are typically associated with normally (dis)fluent speech (p. 121).[91]

The Diagnosogenic Theory

Another historical influence promoted the idea that stuttering and normal disfluency lie on a continuum. This was Wendell Johnson's extremely influential Diagnosogenic Theory—now defunct—that implicated normal disfluency in the cause of stuttering. The theory is considered in detail during Lecture Three, but, in short, it stated that stuttering emerged from parents believing that their children's normal disfluency was stuttering.

Disorders to distinguish from stuttering

There are some disorders that are broadly similar to stuttering, although not in the sense that they resemble each other and require a challenging differential diagnosis process to identify. Identifying them is straightforward with a case history and basic clinical observation. Their case histories and

clinical features are obviously different from the disorder described so far during this lecture, which is sometimes referred to by the term *developmental stuttering* to distinguish it from the disorders that will now be reviewed. The first task for a clinician when meeting new clients is to confirm the diagnosis of developmental stuttering rather than one of the following disorders, and to check that stuttering is not comorbid with one of them.

Acquired stuttering

Acquired stuttering is "a broad term and probably the most common one to denote a fluency disorder of non-developmental origin" (p. 42).[92] This guide to terminology for acquired stuttering[92] suggests three terms for subcategories of acquired stuttering. *Neurogenic stuttering* refers to cases arising from neurological damage such as traumatic brain injury, stroke, and neurodegenerative disease. *Drug-induced stuttering* refers to the effects of medication. A detailed review of this topic is available,[93] indicating that 57% of reported cases are linked to antipsychotic drugs. *Psychogenic stuttering* refers to "a dysfluency that is somehow associated with a psychological problem or an emotional trauma" (p. 42).[92]

Reports about psychogenic stuttering are common and clinically puzzling, and it possible that some or all of them are a combination of neurogenic and psychogenic factors.[94,95,96,97,98,99,100,101,102,103,104,105,106,107] The most recent report about this matter[108] suggests a differential diagnostic procedure for neurogenic and psychogenic stuttering, and it suggests assessment and treatment procedures. The report presents two case histories of mild traumatic brain injury caused during military action.

Neurogenic stuttering seems to be more common than drug-induced stuttering and psychogenic stuttering. Reviews of neurogenic stuttering are available.[109,110,111,112] It appears that those affected by neurogenic stuttering are rarely anxious about it, which is the exact opposite of developmental stuttering, as will be considered during Lecture Ten. Nonverbal superfluous behaviours are common with developmental stuttering, but seem to be rare with neurogenic stuttering. One report,[113] based on five cases of neurogenic stuttering and 35 cases of developmental stuttering, suggests that "phonetic, word class, word length, and word position variables" (p. 1) are more similar than different for the two conditions. Another report,[114] with 3-minute video samples of four cases of neurogenic stuttering and four cases of developmental stuttering, suggests that the difference between the two may not be easy to distinguish. On balance, then, it seems essential during assessments for clinicians to explore the client case history to exclude any chance of neurogenic stuttering being mistaken for developmental stuttering. One report[115] described stuttering onset after concussion in three adolescents and two 2-year-olds who had no family history of stuttering. Contrary to usual cases of developmental stuttering, the three adolescents recovered within 8–10 weeks.

Overall, neurogenic stuttering is a poorly understood condition, but understanding of it has improved with a study of 319 hospital patients with a mean age of 71 years.[116] Of that group, the researchers diagnosed 5.3% with neurogenic stuttering, with 2.5% of the group having the condition for 6 months. There was considerable comorbidity among the 17 patients diagnosed with neurogenic stuttering. Eleven of them also had aphasia, nine had dysarthria, two had apraxia, and five had cognitive problems. Symptoms described as "stuttering" are sometimes reported after concussion. For example, one report[117] stated "difficulty initiating speech, often repeating 'dadadada' before finding her words" (p. 137).

Cluttering

It is possible that someone who has the rare speech disorder *cluttering*[118,119,120] could be mistaken for having stuttering. The features of cluttering[118] are rapid and mostly irregular articulation, disfluencies that are dissimilar to those of stuttering, and impaired intelligibility because of indistinct and abbreviated articulation. Stuttering and cluttering can be comorbid, with a recent report showing seven of 11 participants with cluttering to also have stuttering.[121] So, as well as someone with cluttering being mistakenly identified as stuttering, it is possible for someone to have both disorders and for stuttering to be overlooked. The World Health Organization defines cluttering as follows:[122]

> A rapid rate of speech with breakdown in fluency, but no repetitions or hesitations, of a severity to give rise to diminished speech intelligibility. Speech is erratic and dysrhythmic, with rapid jerky spurts that usually involve faulty phrasing patterns.

Tic syndromes of early childhood

It is possible, but rare, for diagnostic confusion to occur with stuttering and childhood tic disorders, many of which are transient during childhood.[123] Such confusion is most likely to occur when tics have a vocal component. Motor tics (nonverbal tics) will occur when people are not speaking, and that does not happen with the superfluous behaviours of stuttering. The most likely error is for Tourette Syndrome to be mistaken for stuttering. Tourette Syndrome requires one or more vocal tics and two or more motor tics for diagnosis. A report suggested that as many as one fifth of children with Tourette Syndrome may have speech that resembles stuttering.[124]

Neurological disorder

When extremely severe cases of stuttering develop suddenly during early childhood, the disorder may be mistaken for a neurological disease. Doctors have been known to refer cases of severe early stuttering for neurological evaluation. However, speech-language pathologists usually don't make that mistake. On that topic, there is an interesting report of three adult sisters who were diagnosed with late-onset Tay-Sachs disease.[125] The report states that the first of them "developed a stutter at approximately age 10" and the second "developed a stutter at age 8" (p. 289). Videos of the participants accompany the report, and the second participant states during the video that she stuttered as a child. The videos clearly show speech motor problems, but a diagnosis of developmental stuttering is not warranted for any of the participants. Similar diagnostic issues were present in another report involving 453 patients who received deep brain stimulation treatment for Parkinson' disease and who were reported to be stuttering afterward.[126]

An unusual case history

The following case history illustrates a rare instance of when someone presents to a speech clinic with stuttering but obviously it is not straightforward developmental stuttering, and potentially it is comorbid with one or more of the disorders just that were just described.[†]

A 9-year-old boy presented to a clinic with no family history of stuttering or reports of him or his twin brother ever stuttering. He recalled that while camping with family and friends he showered after swimming and then noticed that he was stuttering. Shortly after at assessment the clinician noted syllable repetition, incomplete syllable repetition and nonverbal superfluous behaviours of muscle contractions around his mouth.

Two weeks later the stuttering stopped, and the clinician did not hear from the family again until 18 months later, when his mother reported that the stuttering had returned. The clinician saw the boy again and observed tics as well as stuttering, and so suggested assessment by a paediatric neurologist. The clinician also suggested a psychiatric assessment, and his mother was receptive to that idea, having been concerned about her son's anger and sensitivity.

Stuttering and genetics

Background

It has been known for a long time that genetics is involved with stuttering. For a speech-language pathology readership, comprehensive[127,128] and compact reviews[129] are available, along with an overview of the current status of the field.[130] More technical reviews are available[131,132,133] with more

[†] Thanks to Michelle Taylor for this case history.

focus on the science of genetics, and there is an overview of specific genetic mutations identified to date.[134]

Clients and their parents who don't know it already can be relieved to hear that genetics is involved with the condition, rather than it being a psychological problem, as suggested by common stereotypes discussed earlier. Clinically, it can also be useful to introduce a discussion of what causes stuttering by stating that genetics are involved with it.

The first previously cited review paper[127] outlines the progress in accumulating knowledge about genetics and stuttering in four methodological phases: familial incidence, twin studies, family aggregation, and biological genetics. The subsequent overview follows those headings.

Familial incidence

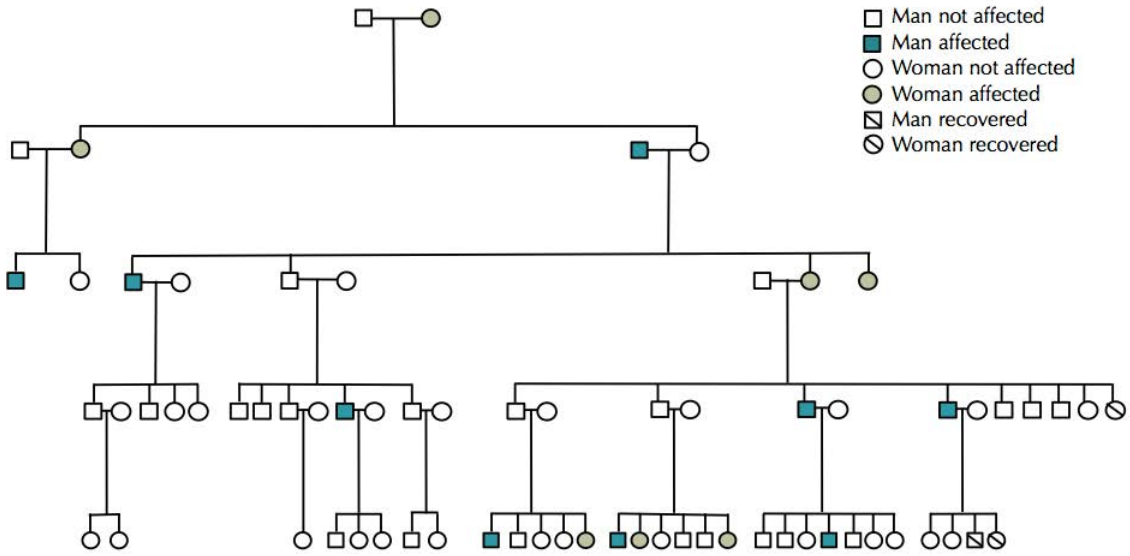
Family history is common

Fundamental evidence for genetic involvement with a disorder is vertical transmission: in other words, a family history. A review of 25 reports dating from 1937 to 2019[128] (Table 2-5, p. 65) shows that, overall, around two-thirds of those who stutter report a family history. So, it is more likely than not that a client will report a family history.

Underestimates are likely

There is good reason to believe that such participant-report data underestimate the true family history rate, with many family members having affected relatives but failing to report it.[135] So, clinically, if an interview suggests that there is no family history, there is some room for doubt.

A famous family



The diagram above[‡] is from a 1940 report of five generations of an Iowa family in the United States.[136] During clinical practice clinicians routinely see many families with stuttering running through them, although not usually as densely as this example. It is probably worth learning to draw family

[‡] Adapted and reproduced with permission: Gray, M (1940), The X family: A clinical and laboratory study of a "stuttering" family, *Journal of Speech Disorders, 5,* 343–348. © 1940 American Speech-Language-Hearing Association.

pedigrees, or learning to use a program that constructs a pedigree, for noting in clinical files. The symbols in the diagram (but not the colour coding) are standard.

More boys and men are affected than girls and women

Proportions of boys at reported onset range from 50%,[251] 61%,[248] and 68%.[254] The ratio of men to women affected by stuttering ranges from 3:1 to 5:1 in various reports,[128] which translates to 75–83% men. There are two features of those figures to note. First, it seems that there are more boys and men stuttering than girls and women. Second, considering all publications about the matter, the reported ratios for young children seem to be more evenly balanced between genders than are ratios for adulthood. So, in short, it seems that fewer girls begin to stutter than boys, and that they are more prone to recovery than boys. There have been independently replicated reports linking the gender imbalance to prenatal testosterone levels,[137,138,139]as occurs with some other disorders.

Studies of twins

Identical and non-identical twins

Studies of twins are another way of establishing a genetic basis to a disorder. Identical twins (monozygotic) are genetically identical people, but non-identical twins (dizygotic) are like any other siblings, except they develop *in utero* at the same time. If both twins have a disorder it is referred to as concordance, and if only one twin has a disorder it is referred to as discordance.

Monozygotic concordance is greater than dizygotic concordance

If the incidence of monozygotic concordance for stuttering is higher than dizygotic concordance, it strongly suggests that a genetic factor is involved with the disorder. In other words, if the incidence of identical twins both stuttering is more than that for non-identical twins, then it suggests genetic involvement. (The assumption underlying this reasoning is that the living environments of both types of twins are the same.)

It seems to have been discovered in the 1930s that, indeed, monozygotic concordance for stuttering is greater than dizygotic concordance,[140,141] with the first attempt to quantify concordance rates some years later.[142] In 1981 a seminal study of 30 twins[143] was published and its findings were subsequently replicated with larger cohorts.[144,145,146,147,148,149]

A large parent self-report study of 10,500 5-year olds[149] found, for reports of "probable stuttering," dizygotic concordance for boys of 36% and 34% for girls, and monozygotic concordance for boys of 53% and 61% for girls. This report estimated "probable stuttering" to be 42% inheritable; in other words, a 42% genetic contribution to the disorder. The highest estimate of genetic contribution to stuttering occurred in a self-report study[148] of 1,896 11-year-old twin pairs, which suggested 80% and 85% genetic contributions to stuttering for boys and girls, respectively.

In short, twins who are genetically identical are more likely to both stutter than are non-identical twins. But monozygotic concordance for stuttering is not 100%. A broad interpretation of these studies is that much—or perhaps the majority—of stuttering can be accounted for by genetic factors. The remainder of cases would be accounted for by genetic or nongenetic factors that are not known about yet.

Family aggregation studies

Clues about genetic models

Statistical analyses of family history data can give some clues about how a disorder is transmitted genetically.[127] However, the retrospective methods of that approach, relying on recall, are a limitation of the method. It appears that two reports during the 1930s[150,151] were the first applications of the family aggregation method with stuttering. Following this, after two and a half decades, was the first comprehensive report.[152] This study involved 213 people with stuttering, or probands to use the proper genetic term, and their families. This study showed an increased stuttering incidence among first-degree relatives of probands—parents, siblings, and children—than for the general population. The

report also found that fathers and brothers of those affected had more than twice the stuttering incidence as mothers and sisters of those affected.

A take-home message

Kenneth Kidd and colleagues at Yale University added to the family aggregation data,[153] and they subsequently published data about 600 probands.[154,155] A take-home message was provided by pooling data for families covered in the first comprehensive report[152] and the Kidd studies:

> For men who ever stuttered, 9% of their daughters and 22% of their sons will be stutterers; while for the fewer women who ever stuttered the risks are higher, as 17% of their daughters and 36% of their sons will be affected. (p. 229)[156]

This information implies that although women are affected by the disorder less often than men, they are more likely to have a child who stutters.

Stuttering and birth rank

Sometimes when there is a family history of stuttering, parents can be concerned that stuttering could be transmitted by non-genetic means, such as with a sibling developing the disorder by "copying" the stuttering of another sibling. Parents who stutter might have a similar concern that somehow their stuttering will encourage their children to begin stuttering.

A report from the Yale group verified that such concerns are not justified[157] by showing no association between birth rank and stuttering among siblings. If stuttering could be transmitted by sibling "copying," children born earlier in the family would have more opportunities to copy stuttering models than children born later in the family, and a different finding would have been expected from the Yale data. Consequently, there is no reason for parents to be concerned that stuttering can be transmitted by "copying."

Biological genetic evidence

Genome-wide association study

Direct genetic evidence is another technique to explore the genetics of stuttering. Until recently, the most up-to-date procedure was with genome wide linkage studies, which trace patterns of stuttering inheritance through generations using genetic markers. The review paper mentioned earlier[129] lists six studies that have provided evidence of multiple chromosome linkage.[158,159,160,161,162,163] These studies make it seem probable that the disorder is polygenic in nature, meaning that many genes are involved in the genotype, and raising the likelihood that other genes are involved and await discovery. However, it has been noted that there is little consistency of results across studies reported to date[127] (see Table 3, p. 42). The genome-wide association study, or whole genome association study, is a technique that involves those affected and not affected with a disorder. To date, this method has been reported in one study about stuttering, which again suggested polygenic inheritance.[164]

Candidate gene analysis

Another source of biological genetic evidence of the disorder is candidate gene analysis, which provides information about contributions of specific genes. Reports using this method have identified mutations in four genes to date (GNPTAB, GNPTG, NAGPA, AP4E1).[165,166,167,168,169,170,171,172] It is possible that a mutation in these four genes could cumulatively account for 20% of stuttering in unrelated cases.[133]

These gene mutations are part of a biological lysosome cell pathway. Lysosomes are small membrane sacs in animal cells that contain many enzymes, and are often described as the cell's recycling bin. They are responsible for many metabolic functions, and mutations of the genes that encode lysosome enzymes cause many lysosomal storage diseases. It seems that mild mutations of the genes are associated with stuttering, and severe mutations are associated with serious disorders including neurological disease involving intellectual disability and white matter pathology.

Two studies have implicated gene mutations with grey matter volume development and energy metabolism in the development of stuttering.[173,174] The latter of those studies[174] were followed up with a magnetic resonance imaging study of 26 children who stuttered and 44 controls, with a mean age 6.5 years. For both groups there was a correlation between glucose uptake and grey matter volume in brain regions involved with speech and language processing. The researchers suggested that these findings could somehow be involved in early stuttering development.

A mouse model of stuttering

Considerable interest has been generated recently with an (arguably adventurous) attempt to establish a mouse model of stuttering.[175] A so called knock-in mouse was developed with a mutation in the GNPTAB gene, which is associated with the lysosome cell pathway. Compared to control mice, the researchers reported that the ultrasonic vocalisations of the mice with the mutated gene were fewer and with longer pauses between. More repetitions of vocalisations were also reported for the experimental mice. The researchers concluded that their results established "the mouse as an attractive model for studying this disorder" (p. 1009).[175] A subsequent publication[176] reported the same effect with two other gene mutations associated with stuttering, and suggested astrocytes—a central nervous system cell—"as a site of the neuropathology, leading to a deficit in interhemispheric connectivity in stuttering" (p. 17515).

Genetic mutations and stuttering treatment

There has been a preliminary attempt to assess whether carrying a mutation in one of the four genes mentioned earlier is associated with the outcome of stuttering treatment.[177] The authors of that work astutely remark that, although they can account for only a minute proportion of stuttering cases, "these four genes are closely related functionally" and that "all are involved in the process of intracellular trafficking." They argue that "deficits in this cellular function are now recognized as causative of a wide range of neurological disorders." They draw on the results of their knock-in mouse study[175] to suggest "that the control of vocalization is an innate, conserved biological process, and that genetic deficits in this control could affect therapies designed to correct such deficits" (p. 12). However, assessing the effects of genetic mutation on stuttering treatment outcome is certain to be a complicated and protracted endeavour.

Conclusions

Around 70% of those who stutter report a family history, so genetic involvement appears not to be always present, suggesting that genetics is not necessary for stuttering. And studies of twins make it obvious that genetics does not always lead to stuttering, so it is not sufficient for stuttering. Even though studies of twins support estimates that stuttering is around 80% inheritable, genetic mutations have been discovered that can explain no more than 20% of stuttering occurrences, with many of those mutations uncovered in stuttering-dense consanguineous families.

It is clear that the genetics of stuttering is complex, with incomplete penetrance within families, and some rare individuals with mutations of a single gene as a major contributor to the disorder. All this makes it virtually certain that stuttering is a polygenic disorder, caused by combined actions of more than one gene, such as with hypertension and coronary heart disease.

Atypical neural processing[†]

Overview

There is extensive evidence from brain research with children and adults who are stuttering that the white fibres that connect areas in the brain that are critical for the complexity of spoken language are not functioning as well as they should. This research literature expands each year, and a 2017 edition

[†] Thanks to Ann Packman for guidance with this material.

of the *Journal of Fluency Disorders* was assigned specifically to the topic. Reviews of neuroimaging studies[178,179,180] cite more than 100 reports up to 2018. This research incorporates evidence of unusual structural and functional non-dominant—right sided—brain activity in speech areas. A review[181] presents 47 papers that implicated functional issues with the supplementary motor area, which is responsible for planning and execution of complex movements. Atypical neural processing associated with stuttering may not be associated specifically with speech functions. One paper reported "wide-spread decreases in connectivity for adults who stutter in regions associated with sensorimotor, cognitive, emotional and memory-related functions." (p. 1)[182]

White and grey matter

The more recent studies suggest that the problem is one of connectivity; transmission of information along the white matter fibres of the brain are atypical in areas involving spoken language. White matter fibres form complex connections between executive areas of the cortex, and are critical for the development of complex neural networks needed for spoken language. Neuroimaging studies of stuttering continue to accumulate rapidly, and overviews and meta-analyses of this body of research are available.[178,183,184,185,186,] Atypical neural processing is not constrained to white brain matter. There are reports that grey matter of the brain is affected in adults[187,188,189,190,191] and children.[192,193,194]

The arcuate fasciculus and the corpus collosum

Reviews of brain imaging research[178,179,180] and subsequent publications,[195,196,197,198,199] show, compared to controls, differences in two prominent brain structures for adults and children who stutter: the corpus collosum and the arcuate fasciculus.

The *corpus callosum* is a large white matter fibre structure connecting the two hemispheres of the brain. The *arcuate fasciculus* is a bilateral bundle of white matter fibres that are fundamental to speech and language production, linking parts of the cortex responsible for expressive and receptive language. These are traditionally known as Broca's area and Wernicke's area, respectively.

An essential context for all this information, however, is that it is not constrained to the disorder of stuttering. For example, atypical neural processing has been reported to be associated with specific language impairment[200] and speech sound disorder.[201]

A current hypothesis

Some researchers[202] have drawn on the results of neuroimaging research and genetic research implicating lysosomal metabolism, as previously discussed, to present a testable hypothesis: the onset and development of early childhood stuttering is linked to abnormal or late myelination[†] of perisylvian fibre tracts. A publication has sustained this hypothesis.[203]

The critical issues

Atypical neural processing is not sufficient for stuttering

It is obvious that atypical neural processing, alone, is not sufficient for stuttering to occur. That is obvious because of the many situations in which those who stutter can speak without stuttering, such as the fluency inducing conditions described during Lecture One. Also, as will be discussed shortly, stuttering does not occur when children first start to speak, but occurs sometime after during early language development. If atypical neural processing is somehow fundamental to the disorder, something additional must occur for stuttering to appear.

Is atypical neural processing necessary for stuttering?

What is yet to be determined is whether atypical neural processing is necessary for stuttering to occur: whether it is always present when stuttering is present. And even if atypical neural processing is

[†] Myelination is an early developmental process that coats each axon of neurones with a fatty substance called myelin. The myelin sheath provides optimal speed and efficiency of nerve cell transmission.

necessary for stuttering, it needs to be determined whether they are part of the cause of stuttering or, as several researchers have suggested, are a consequence of it.[178,195,195,204] The latter explanation is plausible considering the cortical plasticity of the developing brain,[205] and the fact that not only does the brain drive behaviour but behaviour can change the brain.[206] This issue does not pertain specifically to stuttering. It has, for example, been discussed in relation to developmental language disorders.[207]

Some suggestion—but not conclusive evidence—of a causal role has emerged from findings of grey and white matter structural anomalies for participants with stuttering ages 8–13 years compared to control children,[208,209,210,211] and similar results for younger children 3–10 years old.[192,198,212,213,214] This issue has been summarised as follows:

> … studying children who stutter (CWS) is important because the brains of CWS have had far less time to change in response to stuttering; observation of differences in brain activity in CWS is, therefore, more likely to reflect causal mechanisms of the disorder. (p. 3).[178]

Evidence for causality

Lecture Three deals with the cause of stuttering and, in short, there is no convincing evidence yet that atypical neural processing is causal in nature, rather than resulting from years of stuttering.[†] There is only an implication—a substantive one—from their existence in children that they are causally related. Direct evidence would include findings that they are necessary for stuttering to occur, being present in all those who stutter. Researchers have indicated that another source of such evidence would be data from longitudinal studies with age-matched controls.[195] Such evidence has been provided.[202]

Another key source of evidence for causality would be the existence of atypical neural processing prior to stuttering onset in genetically at-risk children.[215] There is some preliminary evidence to that effect, with a study of neonates who were genetically at risk of stuttering, and controls.[199] Those preliminary findings suggested that corpus collosum differences from controls exist shortly after birth. No indications were found for arcuate fasciculus differences. The researchers suggested that the latter finding may have occurred because the fibre tracts in the left hemisphere responsible for speech do not fully myelinate until the age of 2–3 years.[195]

Clinical applications of neuroimaging research

Ideally, the benefits of research about atypical neural processing with stuttering will eventually improve treatment for the disorder.[216] In the interim, the authors of that article note that the clinical implications of this research so far are that those who stutter "will be buoyed to know that the myth of stuttering as a psychological/psychiatric disorder is being debunked by current research illuminating the neurological foundations of stuttered speech" (p. 116). Indeed, this research is eventually destined to alleviate the social marginalisation of those affected by stereotypes, as discussed earlier during this lecture. It should also contribute to alleviating the lasting impact of decades of theorising about stuttering being a psychological problem, as discussed during the next lecture.

Epidemiology of Stuttering

Epidemiology

Epidemiology has come to international prominence with the Covid pandemic. It is the study of health issues and problems in populations, and factors that influence them. There are two types of epidemiological research designs: *observational* and *experimental*. Observational research designs do not attempt any intervention, and common methods of these are cross-sectional studies, cohort

[†] Lecture Three, which deals with causality, raises the prospect that the relation between stuttering causality and brain structure and function may not be a simple, linear cause-effect relationship; there may be some interaction between cause and effect.

studies, or case-control studies. Common experimental studies that test interventions in populations are randomised controlled trials or quasi-experimental designs.

In the case of stuttering, the favoured methods are observational; it is not ethical to do experiments that expose children to things that might cause stuttering. The only published example of such an experiment in stuttering research is infamous,[217,218] with eventual consequences of a public apology from the university concerned and legal compensation to the participants decades later.

The value of stuttering epidemiology

Stuttering epidemiology research can provide useful information for day-to-day clinical practice. Perhaps most importantly, it can establish how prevalent the disorder is and provide information about its natural developmental course through early childhood if it is not treated.

Epidemiological studies can compare children who begin to stutter with those who do not. Such studies can provide clues about what might cause, or somehow be associated with, stuttering onset and development. Such research can also provide clues about how to predict which children will begin to stutter.

Epidemiology and public health

Apart from day to day clinical practice, epidemiological information has a broader impact on stuttering treatment services because it establishes public health information that can change government health care policy. In cases where a disorder occurs frequently, causes significant distress, and can be successfully treated—as is the case with stuttering—information to that effect can prompt governments to provide adequate health care services for it. In cases where adequate health care services are lacking, that situation can be repaired by astute advocacy from clinicians, those who stutter, and the public. There are examples of public advocacy leading to Government enquiries and reports about communication disorders, which have included stuttering.[219,220]

Point prevalence of stuttering

Point prevalence

Point prevalence of a health issue or disorder, often referred to simply as prevalence, is how many people are affected by it at any one time. The most common method of establishing point prevalence for a disorder is to use a cross-sectional design, where a population sample is assessed at one time. Often, prevalence studies involve assessments at different ages.

Two essential caveats

A comprehensive review of stuttering epidemiology research up to 2012[129] details all the caveats that need to be kept in mind about stuttering prevalence. However, there are two central caveats that have the overall effect that estimates of stuttering point prevalence could well underestimate the true value.

Identifying participants

In the case of stuttering, ideally, researchers would assess all participants in a study to determine whether they are diagnosed with stuttering or not. But for practical reasons more than anything else, commonly that does not happen, and most of the available stuttering point prevalence information comes from reports given by relatives, or by self-report.

As discussed during the previous lecture, although self-report about stuttering may be believable for those presenting to clinics for help, the same may not necessarily apply to those who are recruited from the general population to participate in a study. For example, many adults will not necessarily recall having periods of stuttering when they were children, and they may not recall such childhood experiences of their relatives. Yet a common method with cross-sectional study of stuttering populations is to ask those who stutter or their relatives about recall of stuttering within their families. This is known to be a notoriously unreliable procedure, with one report finding that it results in overestimates of stuttering history within families,[221] and another finding the opposite.[222]

A socially avoidant population

Another potential problem is that those who stutter quite often will, to some extent, be socially avoidant because they are socially anxious, as will be discussed during Lecture Ten. So, because a point prevalence study of stuttering requires a one-off social engagement of participants with researchers who are strangers, point prevalence estimates of stuttering could well underestimate the true value.

Telephoning households to find people who stutter[135] seems at first thought to be a way around this problem. However, as also discussed during Lecture One, there is good reason to believe that those who stutter may avoid speaking on the telephone.

Estimates of stuttering point prevalence

A well-known reference text[128] documents 46 international prevalence studies from 1893 to 2019 dealing with children (Tables 2-1 and 2-2, p. 43–45). The mean reported point prevalence in those tables is 1.6%. However, the standard deviation is quite large at 1.9, because the 46 estimated values vary considerably. The lowest reported prevalence figure is 0.3% and the highest is 11.2%. It is of interest that the data for United States children (Table 2–1) have a mean of 1.1% prevalence with standard deviation of 0.6, but the data for other countries (Table 2–2) have a mean of 2.0%, with much more variability, having a standard deviation of 2.3. This suggests that either the point prevalence of stuttering varies internationally, or that the variation is some kind of statistical error arising somehow from different methods used in the two sources.

The latter seems to be the most likely explanation, since there is no sound theoretical reason to suppose that the point prevalence of stuttering would vary so much from country to country. In fact, it has long been accepted that stuttering prevalence is the same for all races and cultures.[223] When a speaker is bilingual, stuttering occurs in both languages.[224] However, there are reports that more stuttering is likely to occur with a second language than a first.[225,226,227] A review[228] highlighted that nearly half the world population is bilingual or multilingual, yet surprisingly little is known about how this relates to stuttering. Those authors conducted a systematic review that found language proficiency and dominance to influence stuttering severity. However, they found little support for the idea that syntactical and phonological differences between languages, and their syllable structure, are involved in determining stuttering severity in bilingualism. An account of the historical origins of stuttering and bilingualism is available.[229] †

The review article mentioned previously[129] notes that one recent study with numerous participants (N=119,367)[230] reported more stuttering among African Americans than other Americans. Why that could be the case is challenging to explain, as is the convincingly reported high prevalence among those with Down Syndrome. A review of the pertinent literature[231] drew attention to reports that 10–45% of those with Down Syndrome stutter. A later report of children with Down Syndrome 3–13 years old estimated a 30% prevalence.[232]

A review article[129] presents a table of prevalence studies conducted this century, along with the conclusion "it is clear that prevalence under age 6 is considerably higher than in later periods in life" (p. 74). A more detailed version of that table is presented below.‡

As the authors of the review article note, their conclusion is consistent with the occurrence of natural recovery after onset, as will be discussed shortly. Their conclusion is also consistent with early stuttering being particularly responsive to treatment compared to treatment in later periods of life, as discussed in subsequent lectures. Also, as the authors note, it may well be the case that early

† Copies of this Bulgarian publication are available from the author on request: john.vanborsel@ugent.be

‡ Adapted and reproduced with permission: Yairi, E, & Ambrose, N (2013), Epidemiology of stuttering: 21st century advances, *Journal of Fluency Disorders, 38, 66–87*. © 2013 Elsevier.

childhood stuttering treatment interacts positively with a trend for natural recovery. Other authors have also offered this suggestion.[233]

PARTICIPANTS	*AGE IN YEARS*	*PREVALENCE*	*MALE/FEMALE RATIO*
1,113[234]	*4–5 [1]*	*2.2*	*0.7:1*
4,983[235]	*4.5*	*5.6*	*[3]*
3,165[236]	*2–5*	*2.6*	*2.6:1*
10,000[237]	*5–13*	*0.3*	*[3]*
21,027[238] *[2]*	*6–10*	*0.8*	*5.1:1*
	11–15	*0.5*	*4.7:1*
	16–20	*0.3*	*1:9:1*
12,131[135]	*2–99*	*0.7*	*2.3*
119,367[230] *[2]*	*3–10*	*2.0*	*2.5 [4]*
	11–17	*1.2*	

[1] Not clear in the report but this probably is the age range

[2] The study reports data separately for the different age groups

[3] Not reported

[4] For the entire sample

A large data set

There are data about stuttering among 3–17 year olds that come from analysis of the extensive United States National Health Interview Surveys (N=119,367),[230] which is the principle source of health information about United States citizens. It includes a range of developmental disabilities: learning disability, autism, ADD/ADHD, cerebral palsy, hearing impairment, visual impairment, intellectual disability, and seizures. Something about this study makes it more believable than other reports of stuttering prevalence. Parents were visited for an interview, and were asked if "a doctor or health professional" (p. 1035)[230] had ever told them that their child had one of those disabilities. This, at least to some extent, gets rid of a common problem with this type of population research: inaccurate self-identification, or inaccurate identification by others such as parents or teachers. In this data set, a "doctor or health professional" reportedly made the diagnosis.

The study[230] indicated a point prevalence of 1.6% for stuttering, which is much higher than the estimate of 1.2% from the cross sectional studies of children discussed earlier. Of all the developmental disabilities in that study, stuttering was the equal ranked third most prevalent.

But still, the results from that study[230] might be an underestimate, regardless of any merit with its methods. One reason is that a minority of parents of young children with communication disorders seem to seek health care advice about such disorders.[239] Therefore, they may not necessarily find themselves in a situation to be told by a doctor or health professional that their child has a disorder. Another reason is that children younger than 3 years were excluded from the data set, yet some information to be discussed shortly shows that many cases of stuttering begin earlier in life than that.

Cumulative incidence of Stuttering

Cumulative incidence

Cumulative incidence, sometimes referred to simply as incidence, is the number of new cases of a disorder during a certain period. It does not include recoveries during that period. So, for example, the cumulative incidence of a disorder up to 12 years of age remains the same regardless of how many recover from the disorder. Sometimes cumulative incidence is discussed without specifying the period, or without reference to recoveries, which can be confusing.

Childhood cumulative incidence

A caveat

The most rigorous way to determine childhood cumulative incidence of a disorder is with prospective epidemiological methods, which study its developmental course with a cohort of children. In the case of a disorder such as stuttering, however, a study of natural development is not without methodological problems. Any such attempt by necessity must alert parents to the first developmental signs of stuttering. Yet, as stuttering develops naturally in the community, not all parents will have such awareness; therefore, such studies have a fundamental validity problem that they are not really studying the natural developmental course of early stuttering.[†] There have been three longitudinal studies published for stuttering, which are discussed next.

The 1,000-family study

A prospective study of a cohort of children published in 1964,[240] known as the *1,000-family study*, is an epidemiological landmark of the field. Children born in Newcastle-Upon-Tyne, England, during May and June of 1947, were assessed regularly for a range of health conditions. The table presents cumulative stuttering incidence at various ages. The following table incorporates information provided in another source[241] (p. 10) about the numbers of children in the cohort at various ages, and the data from Figure 3 (p. 32) of the original publication[240] about the numbers of children who stuttered. As mostly occurs with longitudinal studies, the participant numbers decrease over time, which is known as participant attrition.

AGE	*PARTICIPANTS*	*NUMBER STUTTERING*	*CUMULATIVE INCIDENCE*
Birth	*1,142*		
1 year	*967*	*0*	*0*
5 years	*847*	*30*	*3.5%*
15 years	*763*	*9*	*6.6%*

Albeit a landmark study, and decades ahead of its time, the methods of stuttering identification used for the 1,000-family study are currently recognised by modern standards as a serious weakness that damages the credibility of its results.[129,242] Rather than clinicians, stuttering was identified as present or absent by "health visitors" with a nursing background, who are a feature of the British medical health system.

The Bornholm studies

A more recent large-scale longitudinal study[243] involved all 1,042 children born during 1990 and 1991 on the Danish island of Bornholm. The health services of this island included "a free speech and hearing evaluation" (p. 49)[243] by a clinician. Parents of all the children were recruited just prior to their third birthday, and 1,021 parents agreed to participate in the study and receive the evaluation.

The study did not involve subsequent, identical longitudinal assessments to identify later cases of stuttering onset. Instead, 5 years later, when the children were 8 years old, the researcher inspected the school records of the children for indications of stuttering and interviewed "various community people, such as nurses, social workers, and teachers, who were in position to know about the children" (p. 51).[243] Then, 4 years later, when the children were 12 years old, "all four clinicians who

[†] Thanks to Ross Menzies for this critical point.

cover the island's entire school population were interviewed by the author and were asked to examine their records for any indication of new stuttering cases" (p. 51).[243]

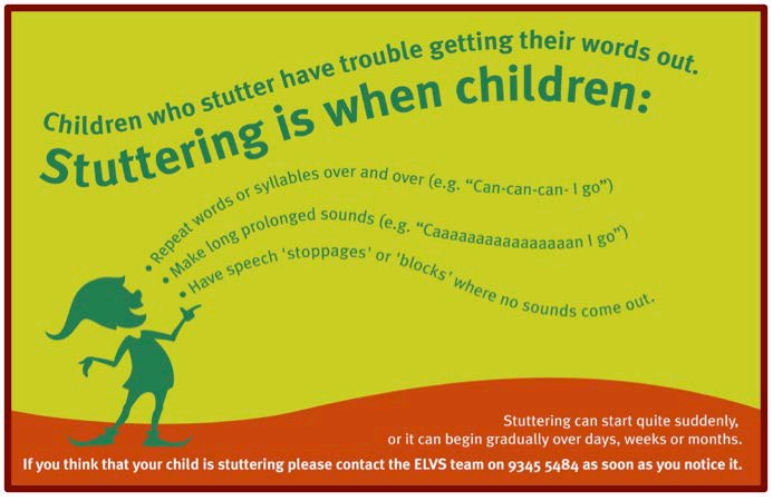
The report indicated a 3-year cumulative incidence of 5.0%; 51 of the 1,021 children were stuttering at 3 years of age. At 8 years of age two additional cases were identified, giving an 8-year cumulative incidence of 5.2%. The 12-year cumulative incidence remained unchanged at 5.2%. Of the children who stuttered, 52% were boys and 48% were girls.

In the review article mentioned previously[129] the authors describe a subsequent Danish publication from Bornholm[244] that is not available in English. They indicate that the original author reported another study of

> 928 children, comprising 92% of the island's newly born children during a different set of two consecutive years … each child was individually evaluated soon after his/her 3rd birthday. The same criteria for stuttering as in the first (2000) study were employed but the procedures were more direct. Specifically, the children's speech samples were audiotaped and evaluated by the examiner to verify the presence of stuttering and to rate its severity … *[the researchers]* identified 176 children who stutter … 101 boys and 75 girls, yielding a 17.7% *[3-year cumulative]* incidence. Whereas one is inclined to doubt such a high figure, we emphasize that, in our judgment, very careful procedures, surpassing those of the first Bornholm study, as well as other many previous studies, were employed, including diagnosis of active stuttering by both parents and two speech-language clinicians, or detailed parent reports of past stuttering … the current first author had the opportunity to observe several identification sessions conducted on Bornholm and can testify to the thoroughness of the procedures. (p. 71)[129]

The Early Language in Victoria Study (ELVS) reports

The children in this report were part of a cohort study of child language development in Melbourne, Australia: the Early Language in Victoria Study (ELVS).[245,246,247] The study was a prospective community cohort design, which means that the children were recruited before stuttering onset and studied longitudinally. There were 1,911 children recruited beginning at 8 months of age, with repeated observations at each subsequent birthday. The ELVS cohort was recruited randomly during 2003 and 2004 from more than 80% of Melbourne parents who visited a maternal and child health nurse when their child was 8 months of age.

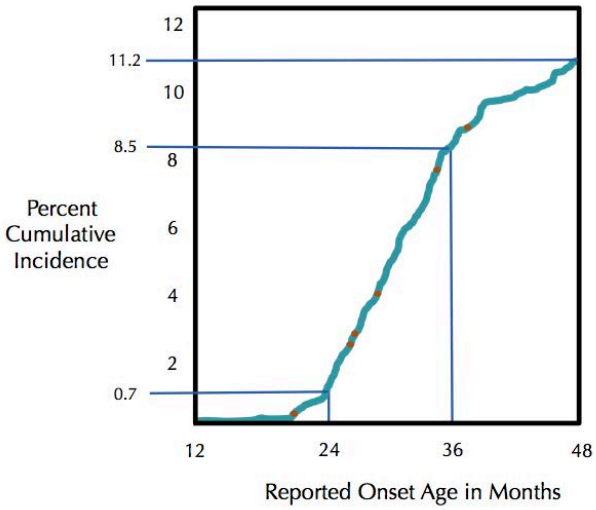


The study of stuttering within the ELVS cohort began when the children were 2 years old, and recruited 1,619 children, which was 85% of the original sample. The parents were sent a fridge magnet (pictured above) describing stuttering and stating "if you think that your child is stuttering please contact the ELVS team … as soon as you notice it" (p. 276).[248] The parents were sent reminder letters about the study every 4 months for 12 months.

When a parent reported stuttering onset, a clinician visited the home, recorded a case history, and made a video recording of the child's speech. By 3 years of age, 158 parents reported stuttering onset. The visiting clinician confirmed the presence of stuttering for 137 children and was unsure about 21 cases. A panel of clinicians reviewed video recordings for these 21 cases and agreed that their stuttering should be considered "'borderline." For the study analyses, these children were classified as nonstuttering.

The first ELVS report[248] showed a 3-year cumulative incidence of 8.5%: 137 of 1,619 children. A subsequent report of the cohort 12 months later,[249] without any participant attrition, gave a 4-year cumulative incidence of 11.2%: 181 of 1,619 children. That represents one in nine children. Of the children who stuttered at that time, 59% were boys and 41% were girls.

The graph[‡] shows the cumulative incidence of parent reported stuttering onset by age, with specific values for 2 years, 3 years and 4 years of age. The graph conveys that the peak period for stuttering onset is between 2 and 4 years. Note that between 3 and 4 years the cumulative incidence plot is still rising but flattening, suggesting that onset rate is slowing but that more cases will appear after 4 years later in childhood.



Conclusions about childhood cumulative incidence

Perhaps the most cautious interpretation of the three prospective studies of the matter to date is that an exact figure for childhood cumulative incidence of stuttering has yet to emerge. The estimate for a 3.5% 5-year cumulative incidence from the 1,000-family study is the least believable among estimates because of the generally acknowledged limitations of that early work by modern methodological standards. It is puzzling that the two Bornholm studies, from the same research team using similar methods, would produce such discrepant 3-year cumulative incidence figures of 5.0% and 17.7%. The ELVS community cohort, with data collection prior to stuttering onset, produced a 3-year cumulative incidence of 8.5% and a 4-year cumulative incidence of 11.2%. On balance, considering that the Bornholm studies began at 3 years and may have missed children stuttering before that, the ELVS reports may be the most believable. But, regardless of what the eventual correct figure proves to be, it is clear at present that stuttering during early childhood is an extremely prevalent disorder.

Lifetime cumulative incidence

The *lifetime cumulative incidence* of stuttering is the risk of being affected at some time during life, including transient periods. In a reference text[128] there is a review of studies where 44,129 people in total were asked whether they had ever stuttered. The authors concluded that

[‡] Adapted and reproduced with permission: *Pediatrics, (132) 460–467*. © 2013 American Academy of Pediatrics.

> … it would seem that a plausible figure for the lifetime incidence of all those who at some time in their lives, either consider themselves or are considered by their parents to be PWS, is at least as high as 8% to 10% … (p. 58)

The authors of a separate review article mentioned earlier[129] agree with that conclusion. In other words, considering any brief periods of stuttering during childhood, and recovery with and without treatment, there is a one in ten chance, and possibly more, that a person will experience the disorder at some time during life.

Stuttering onset

Onset occurs during the early years of life

There is a general consensus that stuttering onset occurs early during the early years of life,[128] and this consensus comes from a review of many studies about parent recall of stuttering onset. There have been some studies of parent interview shortly after onset. A study of 10 children 2–3 years old within 2 months of reported onset had a reported mean onset at 30 months.[250] Another study reported information about 22 children who stuttered, up to their fourth birthday, who had been stuttering for up to 1 year.[251] The children presented with stuttering at a speech clinic. All children reportedly had begun to stutter by 36 months, with mean onset age 28 months.

These results are consistent with the ELVS report of 3-year cumulative incidence,[248] which reported median onset of 30 months. In that study, the lower end of the interquartile range of stuttering onset was 27 months, with the lowest onset reported at 12 months. There were 137 cases reported by parents to have occurred before 3 years of age, with 11 parents reporting stuttering before 2 years of age. The median onset age for the 4-year cumulative incidence ELVS report[249] was 31 months.[†] These results were consistent with a report of 87 children,[254] mean age 39 months, whose parents were interviewed before 1 year post reported onset. The mean reported onset age in that study was 33 months. A report of another cohort of 58 children by the same researchers[252] was a mean onset of 35 months with a range of 19–68 months.

A caveat to keep in mind here is that those reports were not designed to detect cases of stuttering that might occur during the age range 5–11 years. Judging by the 4-year cumulative incidence ELVS graph shown previously, it seems quite possible that such onsets will occur. With the reservation about its methodology in mind, the 1,000-family study reported a rise of cumulative incidence from 3.5% at 5 years to 6.6% at 15 years. However, the first Bornholm study, which was more methodologically believable, reported 3-year cumulative incidence of 5.0%, but a 12-year cumulative incidence of only 5.2%. Regardless of what the eventual correct data will be, it is probably safe to say so far that the bulk of stuttering onset occurs during the early years of life.

Onset can be sudden and severe

An unusual feature of stuttering, unlike other speech problems such as phonological or language disorders, is that it appears after a period of normal speech development. This typically is distressing for parents,[253] particularly when stuttering onset is sudden. Reports show that half of cases develop within 1 week and a third develop during a single day.[251,254] The ELVS community cohort replicated these findings,[248] reporting that 50% of cases developed during 1–3 days and 37% during a single day. It occurs sometimes that parents report a child going to bed speaking normally and at breakfast being severely affected by stuttering. There are reports of stuttering suddenly beginning during the course of an unremarkable day. One of many available case studies describes such sudden onset.[255] Stuttering is not necessarily slow to develop in terms of severity either. Severe stuttering has been reported shortly after onset, including fixed postures and superfluous behaviours.[250,256,,257]

[†] That median onset figure is not reported in the paper.

Repeated movements are prominent at onset

Historically, reports have associated repeated movements with early onset. An influential 1932 account of the early stages of the disorder described them as routine.[258] More modern reports substantiate their prominence at onset.[259,260] One study[250] reported nearly all of 22 parents stating that their children repeated whole words and syllables at onset, and another[248] reported that 71% of parents recalled repeated movements at onset. But of course, if a parent reports repeated movements, it does not mean that various kinds of fixed postures and superfluous behaviours did not also occur during the stuttering moments that had repeated movements.

Is stuttering onset predictable?

Statistically

From a statistical viewpoint, the ELVS prospective cohort study provided a few positive results.[248] At 3 years of age, being a boy, being a twin, having advanced vocabulary, and having a mother with advanced maternal education, were significantly associated with stuttering onset. At 4 years of age the results were the same,[249] with two minor exceptions. Advanced vocabulary did not predict stuttering onset. However, scores for the Communication and Symbolic Behaviour Scales[261] were higher for children who began stuttering, suggesting more advanced communication development. So, the answer to the question is "yes," according to the ELVS data, there are statistical predictors of stuttering onset.

The authors of the ELVS reports argued that the findings about advanced vocabulary and advanced maternal education can be explained, but the finding about twinning is puzzling. Another report from the United States National Health Interview Surveys showed that, along with many developmental disabilities, stuttering was associated with low birth weight.[262] The ELVS cohort had few twins on which to base a claim of a connection between twinning and stuttering onset. However, low birth weight is common with twins, so it might be that the ELVS data are consistent with the United States data.[263] However, a British study[264] of three birth cohorts with more than 56,000 children showed no association between stuttering and birth weight.

Research about statistical prediction of stuttering onset is rare, and there seem to be only two other pertinent reports with reasonable participant numbers. One was a study of 87 children with early stuttering, mean age 39 months.[254] The researchers studied them retrospectively an average of 5.8 months after reported stuttering onset. A positive finding was reported, consistent with the ELVS cohort study, that gender was associated with stuttering onset.[†]

Another report[265] followed 96 children who were genetically at risk to begin stuttering and a control group. Twenty-six of the at-risk group began to stutter. The children who started to stutter had significantly faster articulatory rates than the children who did not; however, that difference was not significant 1 year later. Additionally, no associations were found between the linguistic skills of the children who began to stutter and those who did not. No differences were found for maternal communication style between the two groups.

Clinically

Statistical significance does not necessarily mean clinical significance (as will be discussed in more detail during Lecture Five). The variables that predicted stuttering onset in the first ELVS report at 3 years were able to statistically account for only 3.7% of the cases of stuttering onset in the cohort. The predictors for the 4-year study were only able to explain 3.3% of the variance. So, the short answer to that question is "no," according to the ELVS data, no variables were able to explain a clinically important proportion of stuttering onset.

[†] The report does not provide statistical analysis of gender data. However, they appear to be significant: *Chi Square*=10.35, *p*=.0013.

Hopefully, future research will reverse that situation. It would be extremely useful to predict a clinically important portion of stuttering onsets. Such knowledge would have considerable public health benefits. For example, parents of high-risk children could be told of the risk and be monitored for onset by a clinician so the best early intervention could be provided at the right time. Or, high-risk children could be given preventive treatments before stuttering onset. However, for now, not enough is known to allow any of that.

Natural recovery from early stuttering

What is the natural recovery rate?

The next issue is how many children who begin stuttering early in life will recover naturally without needing treatment. As was the case with cumulative incidence, the most reliable way to estimate natural recovery is with prospective studies involving repeated observations of cohorts during early childhood.

Three essential caveats

Is natural recovery really natural?

A complicating factor here is that there are grounds to believe that many parents do clinically useful things for early stuttering, independently of any clinician input. Indeed, it would be unrealistic to expect parents to do nothing when a child begins to stutter. Several reports have indicated that parents attempt to assist children with their stuttering in various ways.[240,266,267,268] Commonly recurring reports are that parents appear to say "stop and start again" and "slow down" to their children when they stutter. Such verbal responses may constitute the verbal response contingent stimulation of stuttering described in the previous lecture. As such, they may well be clinically useful things for parents to do.

Natural recovery and treatment bias

During such extended studies of children who stutter, it is not ethical for researchers to prevent parents of children with early stuttering from seeking treatment so that they can study the natural course of the disorder. Consequently, it is important to know how much of reported natural recovery is in fact due to treatment that parents sought and received for their children during the period that researchers were studying them. So, interpretation of natural recovery reports needs to be tempered by information about how many of the children received treatment. The review that follows shows—surprisingly—that such information is usually not available.

Has recovery really occurred?

Most of the data about natural recovery are based on self-report, not the judgements of speech-language pathologists about whether recovery occurred or not. As such, there is some room for doubt. This was shown with a study[269] of 15 participants who reported natural recovery without any treatment, and 15 controls. Nine of the self-reported recovery group (60%) stated "that they still had a tendency to stutter" (p. 826). Independent observers watched videos of the participants. For the control group, 84% of the observers judged that the speaker "never stuttered," but for the self-reported recovery group, only 57% of the observers made that judgement. All this raises a possibility that a portion of those who report natural recovery from stuttering may, in fact, still have mild stuttering that is not clinically significant. And perhaps most importantly, there is a tacit assumption by researchers that natural recovery refers to an absence of all the behavioural problems associated with the disorder. However, a study of 254 adults[270] made clear that those who have the disorder do not necessarily see it that way at all. When considering recovery, they focused on the absence of cognitive and emotional effects of the disorder, the capacity to communicate, and having control over the disorder.

The definition of recovery

As discussed shortly, it is fundamental to clinical practice to know how many children recover from stuttering without any treatment. But perhaps the most sobering of caveats about natural recovery was a report[271] illustrating that knowing this depends on how natural recovery is defined. Table 1 in that

report shows a range of natural recovery, in 23 longitudinal studies, from 6.3% to 94.0%, with a median of 66.7%. Obviously, these disparate results are of no functional use of any kind, and to gain insight into them the researchers studied 38 pre-school children on two occasions 7 years apart; first when they were stuttering at 2–5 years old, and again when they were 9–13 years old. Four criteria to determine recovery from stuttering were applied, and the resulting recovery rates, are presented in the table.

CRITERION	*RECOVERY RATE*
(1) Less than 3.0 %SS in video speech samples	*94.7%*
(2) Two expert speech-language pathologists judge no stuttering in speech samples and parents agree that recovery occurred	*71.7%*
(3) Criterion 2 plus child self-report of recovery	*55.3%*
(4) Criterion 3 plus 0 %SS in video speech samples	*13.2%*

In short, determining natural recovery based on stuttering count measures in speech samples is not at all valid and will grossly overestimate recovery rate. Maximum validity is obtained by asking children whether they think they have recovered. The most stringent criterion asks that of children, and also requires zero stuttering in speech samples. Probably the most clinically useful information from this table is that, 7 years after early stuttering during the pre-school years, around half of children will appear to have stopped stuttering according to speech-language pathologists observing speech samples, and according to the reports of parents and the children concerned.

These reports are consistent with others. A study[272] of 15 pre-schoolers followed up 9 years after diagnosis reported 73% recovery based on speech measures alone, but the rate dropped to 60% when the children were asked whether they recovered. The clinical common sense to be obtained from all this was highlighted in a study of 16 children[273] who were studied for a mean of 19 months after onset. The parents reported that four of the children had recovered at that time, but, based on clinician judgements about the presence of stuttering in speech samples, only one child was considered recovered. In other words, relying only on parent report could result in false negative decisions about the presence of stuttering during the pre-school years. As the authors concluded,

> parent report of natural recovery during the pre-school years should not be taken at face value; rather, it should be subject to confirmation by a speech-language pathologist listening to at least one audio or video recording of representative everyday conversational speech of the child concerned. (p. 56)[273]

Prospective reports of natural recovery for older children

The largest cohorts

The earliest prospective report of natural recovery was from the 1,000-family study,[240] which indicated that 34 of the 43 children who stuttered (79%) had recovered by age 16 years. There was no report of treatment history. As noted earlier, though, there are reasons to be wary about the results from this cohort.

The best-known prospective study of natural recovery is the Illinois Early Childhood Stuttering Project,[274] which followed 84 children for 4 years post onset and reported that 74% recovered naturally.[275] Although parents "were informed about availability of clinical services in the area" and that "parents decided if and when their child received treatment for stuttering" (p 1101),[275] the report provides no detailed information about treatment received by the recovered children.

The Illinois group reported results from another cohort of 81 children,[276] of whom 58 were retained in the study for 4–5 years post onset. At that time, 39 were reported as recovered and 19 as persistent, for a recovery rate of 67%. The report contains no mention of treatment history for the children. The first

Bornholm study[243] reported that, of the 51 children who stuttered at 3 years of age, 36 (71%) had recovered at 5 years of age. Again, no information was provided about treatment history of the recovered children.

A report from the ELVS cohort of natural recovery at 7 years[277] presented recovery data for a portion of the children who were recruited at 2 years of age. Of 181 children confirmed to be stuttering at 4 years of age, 103 were studied at 7 years, and 67 had recovered. This provided a recovery rate of 65% at 7 years of age. Within that cohort at 7 years, 39 parents provided reports about stuttering treatment. For the children with persistent stuttering, 17% reported some kind of intervention during early life. Fewer parents of the children who recovered from stuttering—13%—reported that their child received treatment.

A large cohort report from the Twins Early Development Study[144] in the United Kingdom asked parents with a questionnaire at ages 2, 3, 4, and 7 years whether their children were stuttering. With the caveat that parent report has limitations, 1,085 children were reported to be stuttering on at least two of those assessments, 950 (88%) appearing to have recovered naturally. Again, no data about treatment history were reported.

Smaller cohorts

A report of 23 children with early stuttering[278] indicated that 16 of them (70%) had recovered 6 years later. Again, though, no details were provided about treatment history. A study of 22 children[279] with mean age 4.2 years reported that 15 (68%) recovered during a 2-year period. After the first year "parents had the option of continuing only observation and testing or having their chid receive treatment" (p. 112) but no information is provided about how many recovered children received treatment.

The prospective study of 15 children with early stuttering mentioned earlier[272] followed them up from diagnosis to 9 years later, and reported that 11 of them (73%) had recovered. There were two innovative parts to this study. First, complete treatment reports were presented, and all but one of the children had received treatment since their follow-up. This highlighted the impossibility of determining with this type of study what is recovery from natural causes and what is recovery from treatment. Second, the authors asked the children whether they thought they had recovered, and when they did so it appeared that only six of them (60%) might have recovered.

Another report[280] is worth noting, although it was not a prospective study, but a retrospective report of children diagnosed earlier as stuttering. Of the 15 cases aged 2–5 years, 12 (80%) had recovered by 7 years, although no treatment history was reported.

The only discordant prospective finding about natural recovery[281] involved a follow-up of 22 children with early stuttering who were diagnosed in a speech clinic and whose parents declined treatment. Eight of them were younger than 6 years at assessment and all were found to be stuttering 6–8 years later. In light of the issue discussed earlier about the confound of treatment in natural recovery studies, it is intriguing that this is the only report with information about treatment history; none of the eight children received treatment and none recovered.

Conclusions

Taken together, these findings suggest that, after onset during the early years of life, around two-thirds to three-quarters of children will recover naturally at some later time. However, exact figures about how many children recover, and when they recover, have yet to be reliably determined. Overall, the findings about natural recovery are confounded by unknown treatment histories of recovered children. It is also problematic that nothing is known about whether parents of the children in recovery studies made any therapeutic responses themselves to their children's stuttering.

Prospective reports of natural recovery soon after onset

Why is this important?

Lectures Six and Seven show that the merits of early intervention for early stuttering have become apparent during recent decades. Hence, it is now obvious that effective treatment needs to occur at some time shortly after onset. Clinicians cannot wait for many years in the hope that natural recovery will eventually occur. That being the case, information about the rate of recovery during the early years of life is essential to consider during clinical decision making.

Four data sources

Data from the Illinois Project[275] provide information about this matter. The mean age of the 84 children in this study was 40 months at recruitment. Table 4 (p. 1105) of the report shows that five children (8.1%) had recovered at 12–17 months after recruitment.

The ELVS report of 4-year olds[64] indicated that nine of the 142 children (6.3%) recovered within 1 year of onset. However, that result was from a community cohort. It is possible that children from that population who would be taken to clinics would have a higher recovery rate during the first year post onset.

A randomised controlled clinical trial of early stuttering treatment[282] also provided some indication of what the recovery rate shortly after onset might be. The trial had a control group of 25 children who received no treatment for 9 months. Three of these children (12%) appeared to recover during this period. A limitation of this data set is that it was not designed as an epidemiological study and hence did not have enough children for any confident conclusions. However, its strength is that it was an exclusively clinical group of children.

Sixteen children with a mean age of 36 who presented at a clinic[273] were studied for a mean of 19 months. During that period, they received no treatment. Only one of the children (6.3%) was confirmed by parents and the speech-language pathologist to have recovered.

Conclusions

Based on these studies, it seems reasonable to conclude that the natural recovery rate 9–18 months post onset is no more than 10%. In other words, it seems that there is some chance of natural recovery within 1 year post onset, but it is a quite a small chance. Those estimates are based on community and clinical samples.

Is natural recovery clinically predictable?

Reported family history

A review[128] of 21 reports dating from 1937 to 2005 (Table 3-5, p. 95) shows that 88% of families of children with "persistent" stuttering had a reported family history. However, 63% of families with "recovered" children had a reported family history. That might be interpreted to mean that a family history of stuttering can predict to some (unknown) extent whether a child will recover. A study of 1,043 relatives of 66 children who stuttered[283] reported a genetic trend for persistence and recovery from stuttering. The large cohort Twins Early Development Study[144] mentioned earlier replicated that finding.

The Early Language in Victoria Study (ELVS) cohort

The ELVS report on 4-year-olds[249] explored a range of putative predictors that might explain natural recovery within the first year post onset. The most prominent finding was that boys had a greater chance of recovery with an odds ratio of 1.5 (95% *CI*=1.1–2.1, *p*=.02). That means that boys had an estimated 1.5 greater odds of recovering than girls, and the plausible range for the true odds ratio

value, with 95% certainty, was somewhere between 4.7 and 10.9.[†] None of the girls recovered within the first year of onset, and 10% of the boys did. The intriguing ELVS finding associating stuttering onset with twinning,[248] recurred, with a reported odds ratio of 3.3 for twins recovering (95% *CI*=1.4–7.4, *p*=.005). That finding is consistent with the connection of a genetic link to natural recovery. The report also linked maternal education to recovery, including greater odds of recovering with a mother having a degree or other postgraduate qualification. The odds ratio was 1.8 (95% *CI*=1.1–2.9, *p*=.004). Four of the nine children who recovered in this report received professional help, four from a speech-language pathologist.

The ELVS report of natural recovery by 7 years of age[277] included a range of predictors: gender, family history of stuttering, language skills, temperament, child quality of life, and nonverbal cognition. Children who recovered were significantly more likely to have strong language skills than children who did not. Girls with better language skills at 2 years had better odds for recovery than girls who did not. The odds ratio was 7.1, with a wide 95% confidence interval of 1.3–37.9. That means, in short, that the result should be considered with caution because of that wide confidence interval, and because a low odds ratio of 1.3 would not be particularly important clinically. The effect for language skills was not found for boys. No other predictors were found. Of the children who recovered, 13% received some kind of treatment for stuttering, and 17% of the persistent group received treatment.

The Purdue Stuttering Project cohort

Developmental trend of language as a predictor of natural recovery

A report from the Purdue Stuttering Project[284] involved 74 children who stuttered with a mean age of 57 months at the start of the study. Steep growth of syntactic development during yearly clinic visits over 3 years was reported to predict natural recovery at the end of that period. In this report, and with other reports for this cohort, judgments of recovered or persistent stuttering were based on a combination of speech-language pathologist and parent judgment, and speech measures ("Stuttering-Like Disfluencies," to be discussed during Lecture Four).

There was an odds ratio of 11.1 (95% *CI*=1.9–65.4, *p*<.01) in favour of children with steep syntactic development. There are two reservations about interpreting this study, apart from the usual reservations about such a wide confidence interval for the odds ratio. The first is that the children had been stuttering for some years at the time the study began. The second is that the report does not indicate how many of the recovered children received treatment. Regardless, a contribution of this report was the idea that developmental language trends, rather than static measures at one time, may be involved with predicting natural recovery from early childhood stuttering.

These results are consistent with a report from the same cohort[285] for 65-month-old children, 19 of whom recovered and 13 persisted with stuttering. While the children watched video cartoons, EEG data were collected for an event related potential (N400) associated with lexical processing of visual material. Analysis of variance generated evidence that the children who persisted with stuttering had less advanced development of semantic processing. Eight of 19 (42%) of the recovered group had received treatment.

Phonology as a predictor of natural recovery

A smaller study from the Purdue Stuttering Project,[286] reported predictors of natural recovery for 40 children with early stuttering and 25 controls. The children were followed for a mean of 38 months until a median age of 7 years 11 months. Regression analyses showed two statistically significant phonological predictors of recovery: consonant production and nonword repetition abilities. Odds ratios were not reported. No language measure was a successful predictor. The authors reported that 27 of the stuttering group (68%) had received treatment at the time of their first assessment. They reported that they were unable to statistically adjust for this potential confound.

[†] See Lecture Five for more details about odds ratios.

Algorithm as a predictor of natural recovery

With the same cohort, researchers[287] confirmed that the Stuttering-Like Disfluency taxonomy was unable to add anything to the predictive value of existing measures of whether 3- and 4-year-olds will recover. Hence, they reported a study of 4- and 5-year old stuttering children (N=47) to determine any predictors of whether they had recovered when they were 6–9 years old. No information was provided about whether the children received treatment during the period of study. They used a complicated algorithm[18] based on Stuttering-Like Disfluencies to determine whether recovery could be predicted in the 4- and 5-year olds: Weighted Stuttering-Like Disfluencies (to be discussed during Lecture Four). The algorithm is derived from coded transcripts of language samples.

The researchers reported detailed sensitivity and specificity results for the Weighted Stuttering-Like Disfluency score. The sensitivity of the index for the 29 children who recovered—the proportion of correct predictions—was 83.3% at a cut-off score of 4.2 (Table 2). Regression analysis showed an increased odds of 1.2 for stuttering persistence for each 1-unit increase of Weighted Stuttering-Like Disfluency scores. At the cut-off Weighted Stuttering-Like Disfluency Score of 4.2, three of the 18 predictions (17%) were false negative. In other words, 17% of children predicted to recover in fact did not.

Statistical modelling of natural recovery

Another report from Purdue Stuttering Project cohort[288] involved 52 children who stuttered, studied from a mean age of 4 years 6 months for a mean period of 3.2 years. They had been stuttering for a mean of 1.6 years prior to the study. Thirty-one of the children recovered during the period of study. For this report, there was no indication of whether the children received treatment for their stuttering. The report involved detailed statistical modelling of recovery and persistence using several variables: reported age of stuttering onset and stuttering duration, family history of stuttering, family history of recovery from stuttering, several speech and language assessments, and the Weighted Stuttering-Like Disfluencies algorithm.

Results showed that gender, age, age at reported onset, and time since reported onset had no predictive value for persistent stuttering. Successful statistical modelling of persistence was established with four variables: reported family history of stuttering, Weighted Stuttering-Like Disfluencies, the Consonant and Phonological Process Inventory subtests of the Bankson–Bernthal Test of Phonology,[289] and the Nonword Repetition Test.[290] A multivariable model involving all variables was found to have better predictive value than individual variables. For that model, prediction accuracy in the range of 80–100% was reported. The combined error rate—false positives plus false negatives—was around 10% for the model. Removing either one of the phonology assessments (Bankson–Bernthal Test of Phonology or Nonword Repetition Test) resulted in higher error rates.

The Illinois cohort

The two prospective Illinois cohorts have contributed preliminary suggestions about the predictability of natural recovery. The first cohort[291] implicated language and phonological skill, genetics, and certain types of stuttering moments as predictors of natural recovery or persistence. However, these were flagged only as "promising predictors" (p. 51). The second Illinois cohort[252] of 81 children provided similar suggestions, adding increased variability of jaw displacement and negative temperament as predictors of early stuttering persistence. However, those results were presented with the qualification that "results were not definitive" (p. 12).

Another preliminary finding emerged from the first Illinois cohort about a connection between natural recovery and breastfeeding.[292] Forty-seven mothers were studied retrospectively, 30 of whose children recovered naturally and 17 of whose children did not. Data showed a statistically significant effect for boys, with an odds ratio of 0.17, indicating that 1 year of breastfeeding was associated with around one-sixth the odds of persisting with stuttering. However, the report provided no estimate of the confidence interval for the reported odds ratio, making the finding difficult to interpret. Regardless, this finding can be explained in terms of fatty acid nutrition and neural tissue development. However the authors were suitably cautious about the preliminary nature of the finding.

A small cohort

The small-cohort study mentioned earlier[278] for 23 children with early stuttering generated data about predictors of recovery 6 years later.[293] For the 16 (70%) who recovered, analysis of variance was used to provide evidence of an association between lower articulation rate and simpler maternal language and natural recovery 6 years later. No details were provided about treatment history.

Two reviews of natural recovery

A systematic review of the topic[294] incorporated 35 studies with methodologies thought to be acceptable. A fundamental requirement for inclusion was that studies needed to include children who stuttered and control children. Several predictors were established, but none had benefited from independent replication. Four variables were identified as replicated predictors of recovery from early stuttering:

> (1) phonological abilities,
>
> (2) articulatory rate,
>
> (3) change in the pattern of disfluencies …
>
> (4) trend in stuttering severity over one-year post-onset (p. 359) (Numbering in parentheses added.)

The authors concluded that it is too soon to draw any conclusions from this body of research because of inconsistencies in the methods employed. They concluded that "there is a need for systematic and replicated testing of the factors identified before initiating their use for clinical purposes" (p. 359). With regard to an abrupt decline of stuttering severity one year after onset as a predictor, the authors caution that:

> … a significant pitfall of relying on trend in stuttering severity as a predictor is that the factor needs a waiting period of one year to predict the future course of the disorder. In the case of early recovery in stuttering, the recovery period will be within 18 months post-onset. … a year wait period for initiating intervention for stuttering can considerably reduce the outcome of the intervention program. (p. 368)[294]

Another review of the topic[295] published in the same year as the previous one, included children with early stuttering who were younger than six years and who were followed-up for at least 2 years. From 11 cohorts, 41 studies met eligibility and methodological criteria. Results were that some variables significantly distinguished children who did recover from children who did not. Children who did not recover were more likely to:

> (1) … be male;
>
> (2) begin stuttering at a later age;
>
> (3) have known family histories of stuttering …
>
> (4) produce higher stuttering frequencies; and
>
> (5) perform lower on measures of speech sound accuracy, expressive language, and receptive language. (p. 2995) (Numbering in parentheses added.)

The authors noted that effect sizes detected were modest, with the largest being a risk ratio of 1.9 for reported family history. This suggests that children with a family history have nearly twice the risk of persisting than children without a family history. The next largest effect size was gender, with boys having 1.5 times the risk of persisting than girls. The next largest effect size was for reported age of onset, with the persistent group having a mean of 40 months and the recovered group having a mean of 34 months.

In contrast to the previous report,[294] the authors stated that their findings were suitable for application to clinical practice, although they cautioned that the findings cannot be applied to individual clinical children. They argued that the results indicate the need for "comprehensive speech-language evaluations when working with young children who stutter" (p. 310), stating that such evaluations

> … may provide some guidance that is useful to clinicians as they evaluate whether a child is presenting with characteristics associated with higher risk for persistence until empirically supported cutoff scores are available. (p. 310)[295]

Clinical conclusions about the clinical predictability of natural recovery

The short answer to the question of whether natural recovery is predictable is similar to the answer to the previous question of whether stuttering onset is predictable. In a statistical sense the answer clearly is "yes." But, as with stuttering onset, the clinical applicability of that statistical information is concurrently an important matter, and not a straightforward one. Some considerations about it are now presented. The section concludes with an observation that a final process of validation in developing these models has yet to be initiated.

Natural recovery prediction has error rates

In one sense, efforts to predict natural recovery are screening procedures, which were discussed earlier in this lecture. As such, they are associated with error rates. They have a true positive rate (sensitivity) and a true negative rate (specificity), along with false positive and false negative rates. As noted earlier in this lecture in the context of screening, a potentially harmful outcome is false negative. In the case of predicting natural recovery, this is forming a view that a child will recover naturally from early stuttering when the child in fact does not. Publications from the Purdue cohort have noted the clinical seriousness of such an error:[287,288] "In the case of early intervention, failing to identify a TP *[true positive]* may have profound, lifelong ramifications" (p. 2562).[287] This is because of the superior efficacy of early stuttering intervention compared to intervention for persistent stuttering, as noted earlier in this lecture. A delay of early intervention based on a mistaken judgment about natural recovery could be a serious clinical error.

Regrettably then, there seems to be only one publication providing an estimate of false negatives for predicting recovery: 17%. This was for the single predictor of Weighted Stuttering-Like Disfluencies.[287] The most comprehensive statistical modelling of natural recovery to date,[288] from the Purdue cohort, does not supply information about false negatives, only "total number of errors (false positives plus false negatives)" (p. 2929).

How should predictors be used clinically?

Because of the inherent error rates in methods to predict natural recovery, researchers have acknowledged that nothing can be said with certainty about whether an individual child will recover naturally or not. That said, the question remains about what clinical use these clinical predictors might be. The authors of the Purdue reports, while acknowledging the clinical time required to measure children's phonological skills and a Weighted Stuttering-Like Disfluencies score, recommend that those assessments, along with determination of family history, be routinely included in clinical assessment of pre-schoolers who stutter. That view was endorsed with a systematic review.[295]

However, as yet, there is no clear clinical guideline to emerge from this research that makes clear, when such effort is made, what should be done with the results of that clinical assessment. The Purdue researchers have offered passing suggestions that they may be useful for prioritising treatment services[287] and deciding when to monitor a child for recovery rather than providing treatment.[288] The lack of clarity in this matter is arguably underscored by a statement in the latter report obviating the value of recovery prediction in any clinical case of early stuttering involving parent or child concern. (Presumably, that would be the case when most children are brought to a clinic.):

> … regardless of their risk profile, if a child (or their parent) is expressing concern, anxiety, or negative feelings and attitudes toward their communication abilities, that child (and family) would clearly benefit from intervention … (p. 2922)[288]

Natural recovery prediction and the timing of early intervention

As noted in a systematic review,[294] there is a tension between the need for early stuttering intervention and the clinical use of existing reports about predicting natural recovery. This is because reports have

studied children who have been stuttering for several years, and during that period the tractability of early stuttering may decrease. In other words, waiting for some years in the hope that natural recovery will occur is potentially associated with a clinical penalty in the event that natural recovery does not occur. Added to that issue is that a clinically significant period elapses in many cohort studies before the first observation; 1.6 years in one report from the Purdue cohort.[288]

Complicating this issue is that the ELVS cohort, which began when the cohort was 2 years old—much earlier than any other cohort—established recovery predictors that were different to other cohorts. For example, during that early stage of stuttering development, the ELVS cohort reported that family history was not predictive of recovery at all, and that gender was involved. This is the opposite of findings from the Purdue cohort, and could be explained by the younger ELVS cohort.

Finally, it is difficult to apply existing research about predictors of natural recovery to early intervention for the simple reason that the effects of treatment and natural recovery cannot be disentangled in that literature. For example, in the ELVS cohort, parents reported that 15% of the children received some form of treatment, and in the Purdue cohort around two-thirds of children reportedly received treatment.

Another complicating factor affecting clinical judgements about natural recovery and when to begin stuttering treatment is that stuttering is associated with a range of mental health issues, and those problems begin during childhood. The impact of that on when to begin stuttering treatment is an onerous consideration that will be taken up during Lecture Ten.

Validating prognostic models[†]

It is a justifiable viewpoint, expressed in one systematic review,[294] that it is too soon to apply this body of research to clinical practice. It is accepted practice in health care statistics that a prognostic model needs to be validated, in the sense of being shown useful for clients other than those from whom the data were derived. A seminal paper on the topic[296] outlines several reasons why this is necessary. First, mathematical prognostic models are likely to provide overoptimistic estimates of how they will apply to the real world. For example, the model developed from the Purdue cohort[288] relies on professional, community clinicians completing a range of formal assessments. Second, mathematical prognostic models are prone to statistical error. One such source of error is limited sample size, which certainly is a consideration in the literature about estimating natural recovery rates. Finally, the model may not apply to locations other than those in which they were developed. In the case of the Purdue model,[288] there is much work yet to be done to establish whether it applies to other clinical communities worldwide.

Waiting list prioritisation for children who stutter

Considering how common childhood stuttering is, and its potential effects on people throughout the lifespan (see Lecture One), it is not surprising that a recent report[297] found that speech-language pathologists around the world prioritise treatment of childhood stuttering above all other developmental speech and language disorders. The report was a survey of 264 speech-language pathologists from 10 countries: Australia (n=182), United States (n=37), United Kingdom (n=15), Canada (n=9), New Zealand (n=6), Ireland (n=4), Scotland (n=1), South Africa (n=1), China (n=1), and The Netherlands (n=1). The speech-language pathologists worked mostly with children (78%), with 89% working with 3–5 year olds, 83% working with 6–12 year olds, and 43% working with 13–18 year olds. Most of the speech-language pathologists (74%) reported a waiting list in their workplace.

Results indicated that the highest waiting list priority was given to children who stutter above children with other childhood speech and language disorders. The speech-language pathologists indicated stuttering as a priority most frequently (47%) compared to disorders of language (36%), speech (30%),

[†] Thanks to Mark Jones for guidance with this material.

and voice (17%).[†] Reports indicated, compared to other speech and language disorders, that children who stutter most commonly bypassed waiting lists and immediately received assessment and treatment services.

Summary

Early and persistent stuttering are clinically significant developmental stages of the disorder. Diagnosis of stuttering is generally not a clinical challenge, with adults and parents usually being correct with their identifications. However, there are some disorders that potentially could be mistaken for stuttering. There is an ambiguous body of literature about whether speech and language disorders are comorbid with stuttering. Genetics is involved with the disorder, although the complete picture of how is not clear at present. Children and adults who stutter have atypical neural processing. However, it is not yet clear how that relates to the cause of the disorder. Stuttering is a common disorder that is extremely prevalent during early childhood. A comprehensive database shows stuttering to be the equal third ranked of a range of developmental disorders. Its 4-year cumulative incidence could be as high as one in nine children. Onset occurs early during life unexpectedly, unpredictably, and often rapidly. Two-thirds to three-quarters of children will recover at some later time; however, the probability of recovery during the first year after onset is low. Preliminary work has been done to establish workable prognostic models of natural recovery. Speech-language pathologists around the world prioritise treatment of childhood stuttering above all other developmental speech and language disorders.

[†] The highest priority was given to feeding difficulties, at 89%.

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LECTURE THREE: THE CAUSE OF STUTTERING[†]

TWO REASONS CAUSALITY IS CLINICALLY IMPORTANT

Explaining cause to clients and parents

A fundamental way to cope with having a disorder is to understand its cause. So, an obvious way causal theory of stuttering influences daily clinical practice is when clinicians use it to explain what stuttering is and why clients or their children are affected by it. This is particularly important with early stuttering. As discussed during Lecture Two, stuttering onset during early childhood is unpredictable, and its onset can be sudden and severe. This can be distressing for parents, so an explanation of why this happens is fundamental to clinical practice.

Treatment credibility and expectancy

Ideally, there will be a transparent link between what clinicians explain about the nature and cause of stuttering and how they propose to treat it. A treatment that makes sense this way is likely to be more credible to clients and parents than one that makes no theoretical sense. The notion of treatment credibility is "how believable, convincing, and logical a given treatment is" (p. 27).[1] A related notion outlined in that paper is treatment expectancy, which refers to what clients believe can be achieved with a treatment.

There is evidence that constructs of treatment credibility and expectancy are related to parent compliance with psychological treatment for children.[1] This issue has been found to be pertinent for one of the childhood treatments discussed later in these lectures (the Lidcombe Program):[2,3] "I didn't think that *[the treatment]* was really going to make such a difference and it did" (p. 76).[3]

CAUSALITY IS NOT A SIMPLE MATTER

At first, it might seem that the notion of what causes a disorder is a simple matter. But this is not always the case. Packman and Attanasio's classic reference text about causal theory and stuttering[7] contains a discussion about what causes a bushfire (Chapter 1, p. 10–11). This gives some insight into the potential complexity of studying causality. In that text, which is well worth reading, the first two chapters provide a brief introduction to the philosophy of science that deals with causality. Those chapters cover concepts such as *necessary and sufficient* conditions for something to occur, fallacies of causal reasoning, the philosopher Thomas Kuhn and scientific revolutions, pseudoscience, and paradigms. Packman and Attanasio extend Gerald Siegel's observations[4] that the complexity of stuttering involves domains such as the perspective of those who seek to understand causality, the level of understanding required, and why a causal explanation might be sought.

This is not to say that causality of disorders is never simple. Packman and Attanasio mention single gene anomalies that cause human problems, such as cystic fibrosis or sickle cell anaemia. All those who have the genetic anomaly have the disorder and nobody has the disorder without the genetic anomaly. Or to say it another way, a single gene is *necessary and sufficient* to have the disorder.

The causality of stuttering, however, is not so simple. The concepts of *necessary* and *sufficient* when considering stuttering causality recur throughout this lecture. If a condition is *necessary* for stuttering, all those who stutter must have it. If a condition is *sufficient* for stuttering, those who stutter may have it, but won't necessarily have it. Unlike cystic fibrosis and sickle cell anaemia, there is no one condition that is *necessary* and *sufficient* for stuttering. Comprehensive causal understanding of stuttering involves conditions that, together, are *necessary* and *sufficient* for it to occur. Naturally,

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[†] Thanks to Ann Packman for guidance with this material.

those necessary and sufficient conditions for stuttering to occur need to be present when stuttering develops during early childhood, and they need to be present whenever a stuttering moment occurs thereafter.

An historic and clinically influential causal theory

The Diagnosogenic Theory

There are many early theories of stuttering that are now of historical rather than scientific interest, and overviews are given in various reference texts.[5,6,7] Examples include pyknolepsy theory, perseverative theory, approach-avoidance theory, the Orton-Travis Theory, two-factor theory, primary and secondary stuttering theory, and psychoanalytic theory. Wendell Johnson's Diagnosogenic Theory is regarded as one of these theories now of historical interest.[7,8] However, there is much about its influence on clinical practice that is instructive. One of Johnson's famous students, Oliver Bloodstein, gave an engaging account of the origins of this theory from the field of general semantics.[9]

The rise

The fundamental premise of the Diagnosogenic Theory was the paradoxical and circular idea that stuttering is caused by its diagnosis. In short, parents caused the development of stuttering by falsely believing that their children had begun to stutter when, in fact, they had normal disfluency. According to the theory, it was when parents became anxious about normal disfluencies and tried to make their children stop doing them, that stuttering subsequently developed. Johnson famously expressed his theory by stating that stuttering begins not in the mouth of the child but in the ear of the parent. His theory was formally proposed in 1942.[10]

Part of the extensive influence of this theory throughout the Western World and beyond can be linked to Johnson's "open letter to the mother of a stuttering child," which was first published in a parenting magazine and later in a prominent journal of the American Speech-Language-Hearing Association.[11] There, the advice offered to parents was:

> Do absolutely nothing at any time, by word or deed or posture or facial expression, that would serve to call Fred's attention to the *interruptions* in his speech. Above all, do nothing that would make him regard them as abnormal or disgraceful. (p. 7)
>
> Do not label Fred a 'stutterer.' If you do, you will have an almost irresistible tendency to treat him as if he were as defective and unfortunate as the label implies. (p. 7–8)[11]

The theory and the clinical advice that followed from the Diagnosogenic Theory attained widespread acceptance, and for decades no clinician, or anyone else it seems, would ever think of directly treating early stuttering by calling attention to it. The situation in the 1970s is portrayed here:

> one of us presented a workshop on speech and language disorders to a group of early childhood teachers … Stuttering was included and the presenter used the word *stuttering* when the topic was introduced … the teacher said that they had been taught that using the "label" *stuttering* would cause a child to become a stutterer … they had also been taught that these children were experiencing "disfluencies", that they were not actually stuttering, and that the problem would worsen and they would become stutterers once they were labelled and treated as such. (p. 49)[7]

The fall

The fate of most theories during the course of advancing knowledge in a field is for them to be wrong: they can't all be right.[12] Part of scientific development is the eventual realisation that a theory is wrong, and this is what happened with the Diagnosogenic Theory. Emerging research evidence during the 1970s strongly suggested that it was wrong. A prime example can be seen in the reports of verbal contingent stimulation of stuttering with early stuttering, described during Lecture One. The most

famous of those was a 1972 publication[13] showing that two young children reduced stuttering when attention was called to it, and a similar finding to the same effect was published some years later.[14] If the Diagnosogenic Theory was correct, then calling attention to children's stuttering would have worsened it, not improved it. During the early 1980s the longitudinal Illinois Early Childhood Stuttering Project[15] (see Lecture Two) began to produce data that challenged the theory: the speech of stuttering pre-schoolers and their normally developing peers was completely different. The first public proclamation that the Diagnosogenic Theory was wrong was published in 1983.[16]

There was much controversy and colour surrounding the theory. As mentioned during Lecture Two, Johnson conducted a dubious experiment during the 1930s that was not published[17] but which, decades later, was found to not support the theory.[18] The experiment later resulted in legal proceedings.

On the colourful side, the theory predicted that if a culture could be found with no word for stuttering or concept of what it was, then there would be no stuttering in that culture. Johnson published a report in 1944[19] stating that the Bannock and Shoshone Indians of Idaho in the United States had neither any word for nor concept of stuttering, and consequently none of them stuttered. Correspondence came to light in 1981[20] that Johnson was informed at the time that he was wrong, and that the tribes in question had 18 ways of referring to stuttering. When prefacing the fourth edition of his landmark text in 1987,[21] Oliver Bloodstein announced that the period since the previous edition had seen some "notable surprises," one being "the discovery that American Indians of the Great Plains do stutter and probably did stutter a generation ago, when they were reported not to."

A sobering reflection

The Diagnosogenic Theory provides a telling illustration not only of how a causal theory can influence clinical practices, but how that influence can go awry. A recent report shows that, decades later, when the theory is obviously wrong and scientifically discredited, some clinicians still believe it to be true: a 2014 study of 37 speech-language pathologists and 70 speech-language pathology students[22] reported that "more than half of the participants indicated that parents are the primary etiological factor in stuttering and the word 'stuttering' should be avoided" (p. 778). Even more alarming is a report that 141 modern speech-language pathologists in the United States were reluctant to use the term "stuttering" or to diagnose the disorder with pre-schoolers.[23] A report of a 2019 European conference about stuttering treatment[24] is also sobering. Delegates from 29 countries at that conference recognised the need for early intervention, and acknowledged the compelling evidence base showing that early intervention is efficacious (as discussed during Lecture Seven). Yet, there was a view that early stuttering intervention would cause children to become anxious, even in the face of research evidence that this does not occur. The conference convenors ventured that the only explanation for this view could be the lingering influence of the Diagnosogenic Theory. Perhaps, then, it is not surprising that parent guilt seems to recur during accounts of early stuttering[25,26] and (as discussed during Lecture Seven) during its treatment.[27,28]

The human sciences: Theories, hypotheses, and models

In their classic reference text,[7] Packman and Attanasio outline a distinction between the physical and the human sciences, pointing out that the study of stuttering is in the latter category. In contrast to the physical domain, human behaviour requires a different research approach because of the intrinsic variability of humans. As stated by Packman and Attanasio, when researching about stuttering causality,

> … the experiment needs to be performed with many human subjects to determine if the finding occurs in a sufficient number of subjects to be considered a meaningful effect.[7] (p. 22)

Packman and Attanasio refer to theories, models, and hypotheses as ways to present causal propositions used in the search for understanding. They state that, in that pursuit, the terms "theories," "hypotheses" and "models" are commonly used interchangeably, even though there is a distinction

between them in the strictest sense.[†] Theories typically have more evidence in support of them than hypotheses; hypotheses are more tentative propositions than are theories. A model is "a physical and/or mathematical and/or conceptual representation of a system of ideas, events or processes."[29] Or, stated more simply, models explain how things work, and are commonly presented using boxes, circles, lines, and arrows. Packman and Attanasio refer to several categories of models. Some are unidirectional, specifying a cause-effect relationship in a linear or interactional fashion, and some are transactional, with a bidirectional link between cause and effect.

Testing causal explanations of stuttering

Packman and Attanasio[7] discuss four ways to evaluate a causal explanation: testability, explanatory power, parsimony, and heuristic value. The first two of these are now overviewed.

Testability

To be of any value, a causal explanation needs to be falsifiable. The prime source of information used to evaluate causal explanations, and potentially falsify them, is experimentation. To cut short a long story, experimentation involves observations in contrived circumstances that make them more powerful. For example, imagine a theory of stuttering that states that the problem is in the larynx with vocal fold function. There has been such a theory that attained notoriety in the past.[30] An experiment could explore that notion, by having those who stutter speak using their vocal folds and without using their vocal folds; during lipped speech, when there is no vocal fold function, and during standard speech, when there is vocal fold function. If the theory is correct and stuttering is a problem with vocal fold function, there should be no stuttering during lipped speech. In fact, such experiments have been done[31,32] and it is obvious that stuttering can occur during lipped speech. Hence, the theory that stuttering is a vocal fold problem is shown to be incorrect; it is falsified.

The influential philosopher Carl Popper is credited with the axiom that experimentation does not prove a theory to be correct: it only fails to disprove a theory. However, experimentation can provide results that may be interpreted as disproof of a theory, as shown in the case above. A theory that constantly resists active attempts to disprove it attains increasing credibility. A theory that resists active disproof for a long period can become known as a law. For example, the theory of gravitation is often referred to as the law of gravitation. (But even gravity, it seems, is not immune to observational challenges.[33])

Explanatory power

The more that a causal explanation can explain about its topic the more credible it is. Stuttering presents so many things that need to be explained, and a causal explanation of it needs to be evaluated in light of how well it covers them. In the case of stuttering, it might be argued that there are two categories of what needs to be explained: (1) incontrovertible or almost incontrovertible causal factors for the disorder, and (2) well-known features of the disorder. Those categories are discussed below.

Incontrovertible or almost incontrovertible causal factors

During every moment of stuttering throughout life, all causal factors for the disorder must be operating.[34] Given that, causal explanation needs to accommodate causal factors that have been established with reasonable certainty. Collectively, causal factors that are currently known need to be incorporated in a causal explanation as *necessary and sufficient conditions for stuttering to occur*. Arguably, there currently are three such causal factors that have been established, as outlined below.

Atypical neural processing. As discussed during Lecture Two, many research findings have connected stuttering and atypical neural processing. As discussed there, definitive research has yet to be done to

[†] For the remainder of this lecture, the term "causal theory" is used to refer generically to theories, hypotheses, and models about the cause of stuttering.

establish a causal nature incontrovertibly; it is not yet clear whether atypical neural processing is *necessary* for stuttering, so that all those who stutter must have it. And as pointed out during Lecture Two, such atypical neural processing is not *sufficient* for stuttering because children speak without stuttering for a period during language development before stuttering begins.

Regardless, the research presented in Lecture Two carries a strong implication that atypical neural processing might be *necessary* for stuttering. So, it arguably is justifiable to list "atypical neural processing" among established causal factors for the disorder, albeit in a tentative fashion.

That said, it bears stating that a causal relation between atypical neural processing and stuttering may not be straightforward. To use the terminology presented by Packman and Attansio,[7] a model of how atypical neural processing is involved in stuttering causality may not be unidirectional; it may be transactional, with a bidirectional link between cause and effect. Organic issues with atypical neural processing may exist prior to stuttering onset, and the speaker's subsequent efforts to compensate for them may change them in some way. Arguably, that possibility is a reality considering recent knowledge that experience is known to drive brain development,[35,36] and the existence of evidence that a change to atypical neural processing occurs after behavioural stuttering treatments.[37,38,39,40,41,42,43]

Genetics. Any causal explanation of stuttering needs to incorporate genetics as a causal factor. As outlined during Lecture Two, genetics is obviously involved causally with stuttering, although details are not fully known at present. Lecture Two makes clear that its causal role is that it is neither *necessary* nor *sufficient* for stuttering; some who stutter appear not to have genetic involvement, and some who have genetic involvement do not stutter.

Onset during language development. Obviously, early developing language is a causal factor for stuttering. However, as discussed during Lecture Two, stuttering does not start when children first start to speak. Children speak without any problems for an early period of their language development, then stuttering begins sometime later during language development. A causal explanation of the disorder needs to cover this. In this case, developing language is *necessary* for stuttering development, but not *sufficient*; all children who stutter have some language development, but most children with language development do not stutter.

Well-known features of the disorder

There are many features of the disorder that are well known. Some of them are self evident, and some of them have emerged from programs of research. Causal explanation of stuttering needs to cover these features. Packman and Attanasio[7] argue that, ideally, the explanation needs to be done parsimoniously: that is, simply. All else being equal, they argue, what "explains more with less" (p. 37) is desirable. Some well-known features of stuttering that require parsimonious causal explanation are presented below.

Behavioural diversity. Stuttering is behaviourally diverse. Why does it have such a range of behavioural manifestations involving different types of repeated movements, fixed postures, and superfluous behaviours? Even more challenging for causal explanation is that everyone who stutters does so in a different way, even though they obviously have the same disorder. They have different types and combinations of the seven stuttering behaviours described during Lecture One.

The influence of spoken language. As outlined during Lecture One, stuttering moments are not random but tend to occur more on consonants than vowels, and mostly on initial sounds of words and on initial words of utterances. Those who stutter often encounter idiosyncratic difficulties with particular sounds and words. Stuttering occurs more commonly on content than function words. And a most obvious but commonly overlooked fact for any causal explanation to accommodate is that stuttering does not occur on every syllable spoken; it presents as an intermittent problem involving stuttering moments. It is even more challenging for causal explanation to accommodate that language is not even necessary for stuttering to occur; stuttering can occur experimentally on non-words, where lexical processing is not necessary.[44]

Epidemiology. As outlined during Lecture Two, stuttering begins during the first years of life, but why not later in life? Why does it sometimes resolve naturally but sometimes becomes a lifelong problem?

Why does it sometimes begin abruptly and sometimes gradually, and why are repeated movements often among the first signs of stuttering?

Conditions that reduce or eliminate stuttering. There are diverse fluency inducing conditions as outlined during Lecture One. Stuttering nearly always vanishes when people sing or speak in rhythm, or when they speak under chorus reading or shadowing conditions. Stuttering decreases with verbal response contingent stimulation, and under conditions of altered auditory feedback and masking. How can such a range of diverse conditions reduce or eliminate stuttering?

Stuttering and wind musical instruments. Playing wind instruments has in common with speech that it involves respiratory activity combined with tongue and lip movements. There are intriguing reports, dating from the early 1950s, that some who stutter appear to do so when playing a musical wind instrument.[45,46,47,48] One of those reports[48] provided acoustic evidence of this occurring. So causal explanation of stuttering must deal with how it is capable of affecting a non-speech activity.

Stuttering and manual tasks. Although there have been findings to the contrary, there are research findings that signs of the disorder are to be found outside the speech mechanism. Examples include delayed manual reaction times for those who stutter[49,50,51] and finger movement tasks.[52,53,54] There have also been recurring reports that those who stutter do not perform as well as controls with bimanual motor sequences. One research group has found this to occur with finger tapping, key pressing, handle turning, and even peddle pushing.[55,56,57,58,59,60,61,62] Such results have been independently replicated.[63,64,65,66,67,68] All this is even more intriguing than findings about playing wind instruments and speaking nonsense words, because manual tasks seem to have nothing at all to do with speech.

The effect has been reported for children with a procedure called the Purdue Pegboard Test,[69] but an attempt to replicate those findings with adults failed.[70] Compared to controls, those who stutter have been shown to have more timing asynchrony when playing piano melodies.[71] There is a body of research dealing with the capacity of those who stutter to synchronise with a metronome beat. Mixed results have occurred in studies of stuttering and control groups. That literature is reviewed in the most recent publication on the topic, which reported a positive finding.[67] Considering that the effect has been found not to be present for children with early stuttering,[72] it is possible that it is a consequence of stuttering rather than part of its cause.

Stuttering severity is variable. As outlined during Lecture One, stuttering severity is notoriously variable. It is likely to vary with differing audience sizes and types, with across different everyday situations, and when talking alone. As such, it is different from many disorders where presenting features remain stable over time. Causal explanation of stuttering needs to account for this puzzling feature of it.

Multifactorial models of stuttering causality

A distinction of terminology: A multifactorial disorder and multifactorial models of causality

As stated earlier, stuttering is a disorder with many factors involved in its cause. Hence, it is appropriate to describe it as multifactorial. That said, as Packman and Attanasio have noted[7] (p. 146), a distinction needs to be made between stuttering being described as multifactorial disorder and the use of the term "multifactorial" when referring to models of stuttering that are described in this section.

Multifactorial Models of causality: The fundamental proposition

In short, multifactorial models state that stuttering is caused by the interaction of many factors to be found in the living environments of early childhood, and within children themselves. There is nothing necessarily pathological or atypical about the factors involved. They just interact uniquely for each child to be responsible for stuttering. To say it precisely, these models specify nothing—no causal factors—as *necessary and sufficient* for stuttering development. As discussed shortly, this feature of multifactorial models has attracted criticism.

As discussed earlier, the Diagnosogenic Theory was clinically influential during the middle of the last century. Multifactorial models have been clinically influential during the latter part of the last century

and during this century. Notably, RESTART-DCM treatment and Palin Parent-Child Interaction Therapy are based on multifactorial models, and are discussed in detail during Lectures Six and Seven.

The Demands and Capacities Model

The best known multifactorial model is the Demands and Capacities Model, which, as its name implies, states that stuttering occurs when the demands for a child's fluency exceed the child's capacity to produce it. The demands on children come from the living environment and include excessive parent language expectations, constant time pressures of living, and excessive parent demands for advanced cognitive performance. Four capacities of children are implicated: speech motor control, language development, social and emotional functioning, and cognitive development. In the words of its proponents, "there is no single etiology, but as many etiologies as there are stories of stuttering development" (p. 24).[73]

The prominence of the Demands and Capacities Model prompted an entire issue of the *Journal of Fluency Disorders* to be devoted to it in 2000. The model has been described at many sources,[73,74,75,76,77,78] with its first appearance in a 1987 textbook:

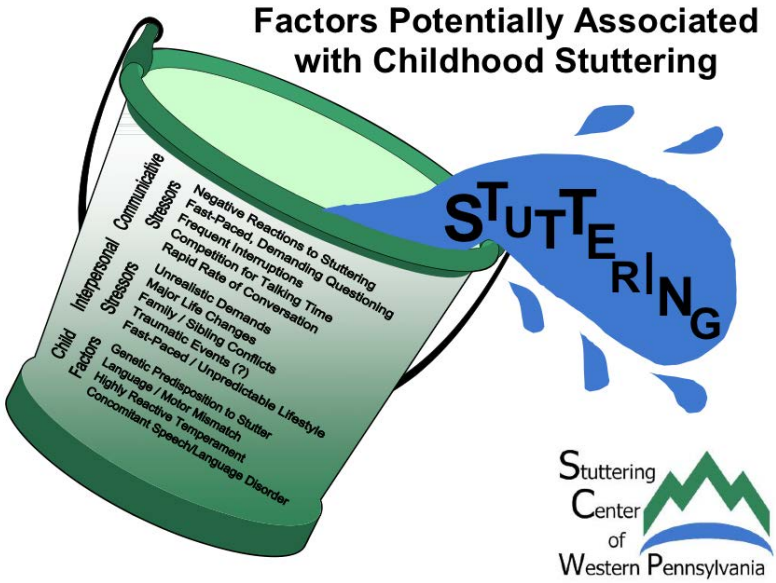
> this growing capacity to talk more easily is paralleled by increasing demands for fluent speech, demands placed on children by the people they communicate with … when the child's capacity of fluency exceeds the demands, the child will talk fluently but when the child lacks the capacity to meet demands for fluency, stuttering will occur. (p. 75)[79]

The model has been depicted graphically as shown in these figures.[‡] The situation on the left shows a scenario where demands exceed capacity, and hence where stuttering occurs. The situation on the right shows a scenario where capacities exceed demands and stuttering does not occur.

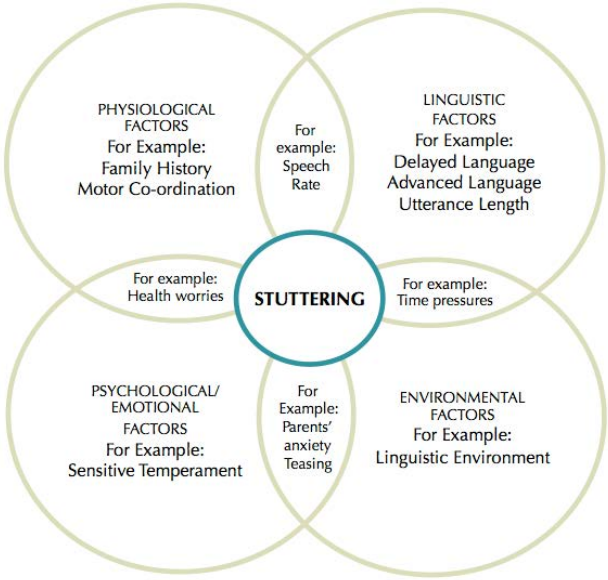
[‡] Reproduced with permission: Guitar, B (2014), *Stuttering: An integrated approach to its nature and treatment* (4th ed.), Baltimore, MD, Lippincott Williams & Wilkins. © 2014 Lippincott Williams & Wilkins

Other multifactorial models

There have been several variants of multifactorial causal models in addition to the Demands and Capacities Model. Two are used internationally as stuttering treatment models,[80,81] particularly in the United States. The following figure[‡] is a graphic from the Stuttering Center of Western Pennsylvania.[81,82] The conceptual similarity between this and the Demands and Capacities Model is apparent, as is the notion that nothing is *necessary or sufficient* for stuttering to occur, as shown by the phrase "factors potentially associated with childhood stuttering."



The Michael Palin Centre in London proffers another variant of multifactorial models,[83] shown in the next figure.[‡] The conceptual similarity with other multifactorial models is apparent. A further variant is known as the Dual Diathesis-Stressor Model,[84] which includes a temperamental proclivity component.



Testability

Although extensively popular theoretically and clinically, the Demands and Capacities Model has been criticised many times,[7,34,85,86,87,88,89] and these criticisms imply criticisms of multifactorial models

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[‡] Reproduced with permission: the *Stuttering Centre of Western Pennsylvania*. © 2004 Stuttering Centre of Western Pennsylvania.

[‡] Adapted and reproduced with permission: *The Michael Palin Centre*. © 2014 The Michael Palin Centre.

in general. These criticisms reiterate the point that multifactorial models are not testable and hence not falsifiable. This is for the simple reason that, as quoted earlier, "there is no single etiology, but as many etiologies as there are stories of stuttering development" (p. 24).[73] It is logically impossible to disprove an indefinite number of causes. As such, multifactorial models are vulnerable to a criticism of being "pseudoscience."

Explanatory power

Causal factors

As noted earlier, multifactorial models specify no causal factors as *necessary and sufficient* for stuttering development. Hence, from that perspective, they have no capacity to explain the causal factors outlined earlier (*atypical neural processing, genetics,* and *onset during language development.)*

Well-known features of the disorder

These models do not rate well in terms of explanatory power for the well-known features of the disorder. An obvious problem for them is explaining the epidemiological fact that most stuttering appears during such a narrow age range in the first years of life. Such models would suggest that it could begin at any time during childhood family life when the factors sufficient for stuttering converge, creating a situation where demands for fluency exceed the child's capacity to produce it. Also, from an epidemiological perspective, it is problematic that the models specify that a cause of stuttering is located in the living environments of early childhood. How could it be, then, that stuttering persists throughout life when that early childhood environment no longer exists? As noted earlier, it is logically essential that "all causal factors must be operating at every moment of stuttering" (p. 226).[34] Additionally, multifactorial models do not explain stuttering variability across time and situations throughout adult life. However, they do explain why stuttering might vary during early childhood in different speaking situations; different situations involve a different mix of demands and capacities.

The future of multifactorial models

Multifactorial models were first proposed two and a half decades ago. From a scientific perspective they have attracted much criticism. Such criticism is justifiable, considering that they are logically impossible to test and that their explanatory power is questionable. Rather than providing theoretical understanding of why stuttering develops during early childhood, they seem only to restate the problem; children begin to stutter because they are unable to do otherwise. Regardless, multifactorial models currently enjoy clinical popularity as a basis for techniques to control of early stuttering, and they have prompted laboratory studies exploring their clinical usefulness[90,91,92,93,94] As will be discussed during Lectures Six and Seven, there have been two clinical trials of such techniques.

The enduring clinical influence of multifactorial models is reflected in a 2022 survey report[95] of 121 Norwegian speech-language pathologists, who were asked: "The first time you meet the parent(s) of a preschool child (0–6 years of age) who is reported to be stuttering, what are the three pieces of advice you give most often?" (p. 941). From a list of 14 options, 78% of the clinicians chose "time to talk," 51% chose "make eye contact," 39% chose "increase pauses," 23% chose "turn-taking," and 16% chose "reduce activity."

Regardless, variants of multifactorial models seem not to have sustained much interest so far in publications this century, although they have been described in a clinical context within two book chapters,[83,96] and they still feature as topics of presentations at international conferences about stuttering. One peer-reviewed scientific journal publication[97] restates an existing model[80] with accompanying explanation of how it is broadly consistent with some aspects of current knowledge about the disorder. Yet the publication seems to add nothing about the explanatory power or clinical insight of the model.

The Interhemispheric Interference Model

The fundamental proposition

It appears that the first formal proposal of the Interhemispheric Interference Model, which implicates the supplementary motor area, occurred in 1987.[98] The model has two parts. The first is that the supplementary motor area of the brain is inefficient, and the second is that the system of hemispheric activation is over-reactive. These two factors are proposed as *necessary* and *sufficient* for the development of stuttering; either factor alone is not necessary. The Interhemispheric Interference Model is an extension of the now defunct Orton-Travis Theory,[99] but departs from it by specifying that those who stutter have normal lateralisation of speech functions. The most recent iteration of the model states, "an anomaly in interhemispheric relations and a deficit in the mechanisms of speech–motor control are each a necessary but not sufficient condition for stuttering" (p. 125–126).[100]

The developer of this model, William Webster, has relied on logic derived from the research findings about bimanual sequence tasks discussed earlier:

> the neural systems underlying such sequential movement control overlap those involved in speech and orofacial movements. Accordingly, anomalies in sequential finger-tapping in stutterers may suggest something about the nature of the "aberrant interhemispheric relations" hypothesized by Orton and Travis. (p. 11)[101]

This prompted the proposal that:

> Although sequential response mechanisms may be lateralized normally in stutterers, the repetitive sequential finger tapping error data suggest that these mechanisms may nonetheless be unusually susceptible to interference. (p. 818)[55]

Testability

In a broad sense, the Interhemispheric Interference Model is supported by many brain imaging findings of unusual hemispheric speech processing with those who stutter. A recent review[178] gave an overview of such literature. A large-scale study of the *planum temporale,*[†] however, was not consistent with the model.[102] It refuted earlier findings[103,104] of differences in symmetry between stuttering and control participants for that anatomical structure. It has been argued[7] that the model is difficult to refute experimentally because neither of its two brain components are operationally defined: the inefficient supplementary motor area and the over-reactive process of hemispheric activation. However, the developers of the model reported that it was verified with an experiment[98] where stuttering and control participants performed a finger-tapping task with a concurrent task using the other hand. The stuttering participants had more interference from the concurrent task than the controls.

Another experiment, though, caused a problem for the model by showing that the same result occurred with a bimanual writing task: writing with both hands concurrently.[105] The results were consistent with a cognitive problem rather than a physical problem with concurrent left and right handed activity. Webster described the problem in a later publication:

> it is unlikely that the interference with sequencing mechanisms in stutterers is strictly an interhemispheric phenomenon as was suggested by the studies of bimanual co-ordination … it unlikely that the origin of that interference is limited to callosal influences. (p. 12)[111†]

[†] The *planum temporale* is an anatomical structure associated with language function, and it is typically asymmetrical between the two hemispheres.

[†] Callosal refers to the corpus callosum, which is a large white matter fibre structure connecting the two hemispheres of the brain.

The Interhemispheric Interference Model received its most sophisticated experimental test using a combination of finger tapping and bimanual crank turning tasks, and two judgement tasks involving the left and right visual fields.[100] The experiment was designed to determine whether the model could explain natural recovery in terms of speech motor control maturation, specifically in the supplementary motor area. Participants were those who had recovered from stuttering, those with persistent stuttering, and controls. Consistent with previous findings, recovered stuttering participants and controls performed better with the bimanual tasks than stuttering participants. However, the stuttering and recovered stuttering participants performed equally poorly on the visual tasks, suggesting that the latter group retained residual interhemispheric problems.

As noted during Lecture Two, a nagging issue with findings about atypical neural processing with stuttering is whether they reflect part of the cause of stuttering or are a consequence of it. One report—although not replicated—has suggested the latter prospect. The report[106] used magnetoencephalography to study lateralisation of brain function for 12 children with early stuttering and 12 controls during picture naming. The children were in the age range of 2–5 years. No differences were reported for the stuttering and control groups. At face value, this falsifies the Interhemispheric Interference Model. If it is correct, for stuttering to occur during early childhood, there should be evidence of unusual lateralisation. The authors concluded that "aberrant lateralization of brain function may be the result of neuroplastic adaptation that occurs as the condition becomes chronic" (p. 1). The conclusion would have been more compelling if, concurrently, the report had demonstrated unusual lateralisation to be present in older children. It is the case, though, that magnetoencephalography has found interhemispheric differences with adults who stutter using other methods.[107,108,109]

Explanatory power

Causal factors

A strength of the Interhemispheric Interference Model is that it seems takes account of current knowledge of causal factors. It overtly specifies *atypical neural processing* as a causal factor. And it is not too much of a stretch to associate atypical neural processing with *genetics*. The model might also explain *onset during language development* in terms of a hemispheric interference problem manifesting at that developmental stage.

Well-known features of the disorder

The Interhemispheric Interference Model certainly can explain the manual sequencing anomalies that have been found in those who stutter. However, its explanatory power is weakened by not only the experiment with bimanual writing tasks described earlier, but by problems with it incorporating other research findings about writing tasks.[110,111] Additionally, explaining the influence of spoken language and stuttering variability seems problematic. That aside, the model does have considerable explanatory power. It is able to explain at least some of the early epidemiology with the natural recovery study.[100] It can explain the fluency enhancing conditions because they could simplify speech motor activity to compensate for a problem with interhemispheric speech processing. Stuttering with wind instruments, nonsense words, and bimanual tasks can be explained by the model, because it does not specify that the brain problems are speech specific.

The future of the Interhemispheric Interference Model

Webster acknowledges in several publications that the broad notion underlying his theory has a long history. It dates back to the early years of the last century to Lee Edward Travis[†] who proposed the Orton-Travis theory of cerebral dominance,[99] the origins of which are apparent in a 1925 report about dyslexia.[112] (Intriguingly, a 2021 report[113] noted commonalities between stuttering and dyslexia, and

[†] Lee Edward Travis is credited as the originator of the speech-language pathology discipline at the University of Iowa, before Wendell Johnson arrived there.

reported that a high prevalence of adults who stuttered met diagnostic criteria for dyslexia.) In 1978 Travis recounted tests of the Orton-Travis theory that were presented in a 1931 textbook,[114] long before the arrival of scientific journals in the discipline:

> When I published the cerebral dominance theory of stuttering in 1931, I presented in its support three laboratory findings: (1) reductions of the patellar tendox reflex latency, (2) reductions in the amplitude of tremors from extended right forefingers, and (3) profound alterations in the alternating phasic movements (opening and closing) of both hands, all during tonic stuttering blocks … (p. 278)[115]

From all accounts, those were ground-breaking research methods for the field.[116] In a 1978 publication[115] Travis outlined how the theory was able to explain a series of research findings in the 1960s and 1970s. In 1986, just before his death, Travis asserted "the stutterer differs significantly from the normal speaker only in his neuro-anatomical organization for speaking" (p. 119).[117]

In short, the idea about interhemispheric interference has been an intrinsic part of thought and research in the discipline about the nature and cause of stuttering. It might be interpreted as an encouraging sign that, for more than 80 years now, the Interhemispheric Interference Model has resisted definitive experimental disproof. And, as discussed during the previous lecture, there is now evidence of atypicality in the corpus callosum—the white matter structure connecting brain hemispheres—among children who stutter. The greatest challenge to date for this line of causal explanation about stuttering is the finding of no lateralisation anomalies with pre-schoolers who stutter.[106] Should that finding be replicated, the Interhemispheric Interference Model would be falsified and its long period of viability would come to an end.

The Covert Repair Hypothesis

The fundamental proposition

Drawing on Levelt's model

The Covert Repair Hypothesis draws on Levelt's well known model of speech production,[118,119] and another model of phonological coding.[120] Levelt's model, in short, comprises three linear processes. The first is the selection of a lexical concept to be spoken. The second is the selection of a word in abstract form (lemma) and its grammatical encoding. Finally, a "mental syllabary" is accessed[121] and the word becomes a set of syllables ready for articulation.

Phonological coding errors

The central proposition with the Covert Repair Hypothesis is that those who stutter have phonological coding errors in the process of preparation for articulation, and that stuttering moments are covert attempts by the speaker to correct these errors before speech execution of the faulty plan.[122,123] Those who stutter have more errors than those who do not, and consequently they need to correct the errors more. These corrections occur before the articulatory sequence occurs, and this leads to repeated movements and fixed postures during speech.

A continuum

The hypothesis does not state that there is anything qualitatively different between those who stutter and those who don't, merely that the former group have slower phonological coding and have more errors in the phonetic plan and, hence, they need to make more corrections. In effect, the hypothesis proposes that stuttering and normal disfluency are on either ends of a continuum. As mentioned during Lecture One, this is known as the Continuity Hypothesis.[124]

Testability

Confirmations

The developers of the Covert Repair Hypothesis, the Dutch researchers Postma and Kolk, have presented support for the hypothesis using research methods involving speech errors of stuttering

participants.[125,126] Other researchers have provided supportive data for the hypothesis,[127] reporting that 5 and 6 year old stuttering children had significantly lower "phonological memory" than control children, as measured with a non-word repetition task. According to a meta-analysis[128] of nine studies dealing with nonword repetition, that effect seems to be robust. Subsequent to the meta-analysis, four observational studies of verbal short-term memory for children with early stuttering and controls reported poorer performance for the former group.[129,130,131,132]

Another report provided further supportive data,[133] reporting that a group of stuttering children with a mean age 5 years 7 months had inferior performance to control children on sound blending and elision tasks.[†] Other researchers have reported that 11 year old children who stutter are slower than controls with a phoneme monitoring task,[134] and less accurate than controls with non-word repetition and phoneme elision tasks.[135] That research group reported that adults who stuttered were slower than controls in repeating non-word phoneme sequences.[136] With adults, researchers[137,138] have reported that a stuttering group showed unusual responses to phoneme monitoring tasks compared to a control group. Researchers have reported[130] that 7–12 year Kannada-speaking stuttering children did not perform as well as controls for nonword repetition and nonword identification tasks.

Another report[130] produced data consistent with the hypothesis using a study of stuttered words in relation to similar sounding words ("phonological neighbours"). The author predicted that, if stuttering involves a phonological coding problem, stuttered words would have fewer similar sounding words nearby than nonstuttered words. The reasoning was that more similar sounding words would facilitate the production of stutter-free phonological encoding, and fewer similar sounding words would increase the chance of a stuttered word. There was some statistical evidence to suggest that was the case.

An eye-tracking study[139] also produced results consistent with the Covert Repair Hypothesis. A group of adults who stuttered and a group of controls read nonwords silently and out loud. During the reading out loud component, the stuttering group had "significantly more fixations and longer dwell times" (p. 475) on the nonwords than controls. During the silent reading component, they had more fixation on the nonwords.

A fundamental problem with such observational studies of stuttering and control groups has been mentioned during Lecture Two; any observed differences may be the result of stuttering rather than being involved in the cause of stuttering. This limitation was displayed with a report purporting to confirm the Covert Repair Hypothesis.[140] The study explored phonological working memory with a stuttering group and a control group of adults, using functional magnetic resonance imaging. Atypical neural processing was identified in brain regions of interest for the stuttering group. However, the authors noted

> … because adults who stutter may develop compensatory strategies for coping with stuttering, the results of our study may be unable to determine the cause and effect relationship between the phonological WM *[working memory]* deficit and stuttering. (p. 10)[140]

However, the authors then argued that similar results for word memory anomalies have been observed with 3–5 year-olds who stutter, "implying the causal role of a deficit in WM *[working memory]* in stuttering" (p. 10).[140]

Reported falsifications

There have been several empirical reports that claim to falsify the Covert Repair Hypothesis. One finding[141] with nine boys who stuttered with normal phonology, and nine boys who stuttered with disordered phonology, was that neither group showed more self-repair behaviours than the other. Another report[142] with 12 stuttering boys of mean age 55 months tested the prediction of the hypothesis that higher articulatory rate would cause more stuttering, but it was not found to be so.

[†] Elision is removing a phoneme from one word to create a new word.

Another report[143] of 32 adult stuttering participants and 32 controls found that the former group made more errors with tongue twisters, which was consistent the hypothesis. However, the number of errors made did not correlate with any stuttering severity scores, which would not be predicted by the hypothesis.

Another challenge to the hypothesis occurred[144] with a study of 12 stuttering and 12 control children, ages 7–12, reciting a list of nonsense words. No significant differences in errors could be found. Another report[145] was of a man who stuttered but only produced phonological errors on stuttering moments that were "part-word repetitions." The authors claimed this to be a logical challenge to the hypothesis. Two publications produced lip electromyographic data with an experimental reaction time paradigm, and argued that the results were inconsistent with a motor planning problem.[146,147] A report[148] compared speed of phonological encoding with stuttering adults and controls and concluded that the data did not support the Covert Repair Hypothesis. Another study of stuttering and control children[149] found no significant differences between the groups for a nonword repetition task. A report[150] by the same group with nonword repetition found "limited support" (p. 1) for problems with encoding speech sounds.

Explanatory power

Causal factors

Although the Covert Repair Hypothesis does not overtly incorporate atypical neural processing within its causal explanation, it is an intuitive prospect that it might be responsible for its proposed mechanism. And it is also intuitive to link atypical neural processing to *genetics*. The hypothesis can explain *onset during language development*; phonological complexity gradually increases with language development to a point where the problem emerges as a quantitative distinction from normal language development that is recognisable as stuttering.

Well-known features of the disorder

The Covert Repair Hypothesis has certainly prompted interest in the current literature, with many examples of researchers using it to broadly frame or interpret their research. The hypothesis indeed does have strong explanatory power, as argued by its developers.[122] It explains the fluency enhancing conditions that might involve speech rate reduction; speech rate reduction would reduce the inherent problem for those who stutter. The hypothesis can explain the repeated movements and fixed postures of stuttering quite well. For example, if an error is detected at the last sound of a syllable, then repetition of the initial sound and vowel that precedes it will occur until the correct sound is ready (for example, do-do-do-dog). The developers presented detailed explanation of various stuttering types according to the theory[123] (Tables 1 and 2). However, the theory, like others, is silent about superfluous behaviours.

The hypothesis does, however, have some serious shortcomings with explanatory power. Its developers acknowledge[122] that the hypothesis does not explain the occurrence of natural recovery. Also, it does not explain why stuttering varies within people and across time and situations.

There are several research findings that are inconsistent with the hypothesis. It has been pointed out,[141] for example, that it is at odds with a finding[151] that speech rate and response latencies—the time taken for a verbal response during conversation—did not differ between children with early stuttering and controls. The hypothesis would predict the opposite.

The hypothesis suggests that the problem of stuttering is constrained to phonology. As such, it is challenged by the findings that those who stutter perform less well with activities that are phonology independent: playing wind instruments and bimanual tasks. The theory has shortcomings with explaining how aspects of spoken language can influence the occurrence of stuttering moments.

The future of the Covert Repair Hypothesis

The Covert Repair Hypothesis is eminently testable and has generated much research. However, so many failures to verify the theory, from different researchers using different methods, casts some doubt

on its longevity. A review of the pertinent literature in one report[148] attributed such results to the range of stuttering severity and types of stuttering involved, and the different research methods used. Another viewpoint would be that if the covert repair hypothesis is true, then phonological encoding problems with those who stutter should occur predictably and should be detected easily with all samples of stuttering participants across a range of research methods. Another reviewer of the theory concluded "the covert repair of errors of phonological encoding cannot account for all instances of disfluency associated with stuttering" (p. 25).[152]

If the Covert Repair Hypothesis should, during the next few years, head towards being one of those theories of historical rather than current interest to the disciple, then that will be worthwhile progress. Expedient abandonment of a theory based on empirical research is welcome progress for a discipline, and clinicians who are ultimately guided by theory about stuttering would be well informed by such a development.

The EXPLAN Theory

The fundamental proposition

Also drawing on Levelt's model

The EXPLAN theory has in common with the Covert Repair Hypothesis that it draws on Levelt's speech processing model to specify a cause of stuttering.

A delayed motor plan

The theory differs from the Covert Repair Hypothesis in proposing that the motor plan is delayed, rather than being incorrect. The theory seems to be foreshadowed in reports from the late 1990s.[153,154,155] The impetus for its development seems to be that stuttering generally tends to occur more often on content than function words (see Lecture One). The authors proposed a hypothesis that the "stuttering of function words is caused by unavailability of instructions for the following content word" (p. 1020).[153] They propose also that, compared to function words,

> the speech plan of a content word is unavailable because planning of such words is relatively slow because of their more complex semantic content, their phonetic composition, and their greater length when compared to function words. (p. 1028)[153]

It appears that the first formal statements of the theory occurred some years later,[156,157] introducing the term EXPLAN theory to capture the fact that it deals with speech planning and execution.

Probably the most comprehensive, formal outline of the theory was presented in 2004.[158] The theory deals with the planning of speech as the linguistic aspect of the process, and execution of speech as the motor component. Stuttering occurs when the motor plan is late in presentation for speech execution. According to the theory, this occurs because planning of the linguistic segments of content words is slow; they are more difficult to plan than function words. The theory suggests that

> whole-word repetitions (and also pauses) are ways of stalling motorically (repeatedly executing a previously generated program) on material prior to other material that is difficult to plan … whereas prolongations part-word repetitions and word breaks reflect planning problems (the repetition, prolongation and hesitation within words signify that the plan was not right or was only partly prepared). Prolongations, part-word repetitions and word breaks are referred to as advancing to indicate the speaker has moved forward prematurely in the speech stream and to contrast with what happens in stalling. (p. 56)[159]

So, stuttering occurs when the speaker either uses whole-word repetitions to delay the execution of a motor plan for a content word that is not yet ready. Or, the speaker abandons that delaying strategy and instead attempts to progress to speak the incompletely prepared word, hoping that the plan for it will arrive in time. This causes other, more complicated speech perturbations.

A continuum

As with the Covert Repair Hypothesis, the EXPLAN Theory incorporates the Continuity Hypothesis, linking the normal disfluency of early childhood to stuttering development. This argument is stated clearly in one publication, and draws on the notion that "young speakers, whether they are diagnosed as stutterers or not, would exhibit similar nonfluencies" (p. 346).[155] It is the shift during adulthood from disfluencies on function words to disfluencies on content words, for which there is a delayed motor plan, that is responsible for persistent stuttering. Early onset stuttering is the simple repetition of function words, to delay things, because the content word is not ready. Persistent stuttering in adulthood is when the speaker essentially abandons the delaying tactic with function words, and attempts to move forward with the content words that are not fully planned, resulting in different, more complicated stuttering moments.

Testability

It might be arguable that research thought to confirm and falsify the Covert Repair Hypothesis, as discussed earlier, could have the same impact on the EXPLAN theory. Regardless, the EXPLAN development team reported "very few dysfluencies" (p. 345)[155] for children or adults on function words that occurred after content words, which was interpreted as supporting the theory. That paper also argued it was consistent with the theory that for all age groups "dysfluency … occurred predominantly on either the function word preceding the content word or on the content word itself, but not both" (p. 345).[155]

Recently an independent group directly tested the EXPLAN theory[160] with an argument that during early stuttering it predicts that the phonetic complexity of the second word of an utterance will predict whether stuttering occurs on the first word of an utterance. The authors found that for fourteen 3-year-olds this was not the case. Another independent group[161] conducted two experiments with adults using a "semantic blocking" research protocol, and reported results consistent with the theory.

Explanatory power

Causal factors

The explanatory power of the EXPLAN Theory for established causal factors is roughly equivalent to the Covert Repair Hypothesis, as described earlier. *Atypical neural processing* might be responsible for its mechanism, with a link to *genetics*. It offers a mechanistic explanation of *onset during language development*.

Well-known features of the disorder

The leader of the development team for this theory points out its strengths in explanatory power.[158] It offers an explanation for the influence of spoken language on stuttering. It explains the intermittent nature of stuttering; why it does not occur on every syllable. The reason is that delays in motor planning occur according to the difficulty of what is being planned. It also explains the prominence of stuttering on function words during early stuttering and the switch to prominence of content words during persistent stuttering. and it does credibly explain different types of stuttering moments. As with the Covert Repair Hypothesis, it does explain the fluency inducing conditions in terms of reduced speech rate. The theory, like others, is silent about superfluous behaviours.

Being a theory that deals with specific interruption to the process of speech production, it shares a number of shortcomings with the Covert Repair Hypothesis. It does not explain why natural recovery occurs. Nor does it explain why stuttering onset can be sudden or gradual. Being a speech process theory, it also shares with the Covert Repair Hypothesis that it cannot explain findings about playing wind instruments and bimanual tasks. Further, it seems to be a fatal problem for the theory that stuttering can occur experimentally on non-words, where lexical processing is not necessary. It shares with the Covert Repair Hypothesis a shortcoming with explaining why stuttering varies within people and across time and situations.

The future of the EXPLAN Theory

It is difficult to offer any projection about the future of the EXPLAN Theory because formal statements of it only emerged this century, barely a decade ago. The theory is testable and perhaps it will resist disproof, with experimentation during coming years by researchers independent of its development. At present, though, its weakness seems to be its limited explanatory power for the well-known features of the disorder.

The Packman & Attanasio 3-Factor Model of Stuttering Moments

Background

Known generally as the P&A Model, this is a model in the true sense of the term, as outlined earlier; it explains how things work. It does not propose to explain why the disorder develops, but proposes to explain the factors that, together, are *necessary* and *sufficient* for the occurrence of individual stuttering moments. As such, it incorporates the logic that "all causal factors must be operating at every moment of stuttering" (p. 226).[34]

Packman and Attanasio developed the P&A Model,[7,34,162] advancing earlier thinking outlined in previous publications.[163,164] They acknowledge that it incorporates components developed from earlier work by Zimmermann and Wingate. They credit Zimmerman and colleagues with the notion that the speech motor systems of those who stutter may be unusually susceptible to variability.[165,166,167] They credit Wingate with the notion that prosody, of which syllabic stress is a part, is somehow disturbed with stuttering.[168,169,170] Wingate recognised that the effect of rhythmic stimulation involves changes to stress, and, specifically, that rhythmic speech reduces stress contrasts.

The fundamental proposition

There are three factors in the P&A Model, as follows:

Atypical neural processing

The model assumes some kind of a central nervous system issue that gives some children an inherently unstable speech motor system. The first published account of the P&A Model[34] suggested atypical white matter connectivity as the likely neural processing problem, as other researchers had done.[171] This problem manifests as a deficit in neural processing that makes speech prone to perturbation. In the model, the neural processing impairment is a *necessary* but not *sufficient* condition for stuttering to occur. In other words, everyone who stutters must have it, but it is possible to have it and not stutter.

Triggers for stuttering moments

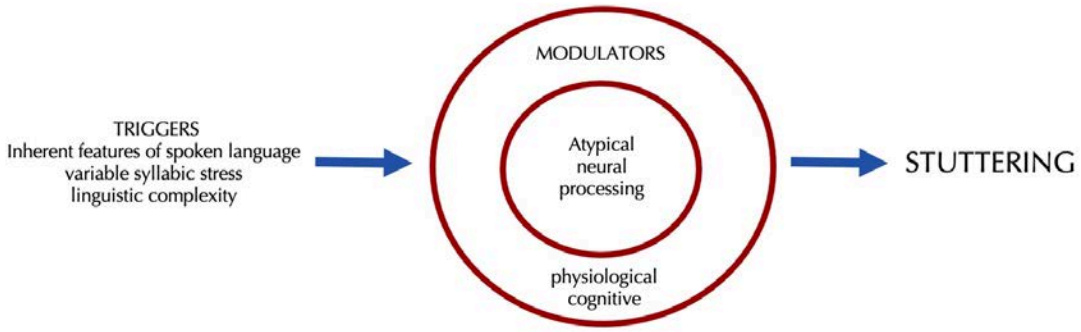
The underlying neural processing impairment and triggers for stuttering moments are, together, *necessary* and *sufficient* for stuttering moments to occur. Moments of stuttering are triggered by certain features of spoken language. These are the variables that increase the motor task demands placed on the already unstable speech system: the varying of stress or emphasis from each syllable to the next, and linguistic complexity. This pushes those who stutter beyond what their unstable speech system can deal with, thereby triggering stuttering moments.

This idea draws on the watershed time during early language development when children begin to produce linguistic stress contrasts. So, for example, they will say "*dad*-da," emphasising the first syllable, instead of "dad-da." That may not seem much of a difference, but it is a leap of speech motor control. Subsequent to initial triggering of stuttering moments by early attempts to produce linguistic stress contrasts, maladaptive responses by children to struggle with the problem cause continued stuttering development.

Modulators

According to the P&A Model, the threshold above which moments of stuttering are triggered is modulated, differently for each individual, according to the level of physical arousal at the time. Anxiety is a prominent cause of physiological arousal, which may lower the threshold for stuttering

moments to occur and may be associated with more of them occurring. Cognitive factors might also lower the threshold for stuttering moments to occur. The model draws on evidence that stuttering increases with the physiological arousal presumably associated with increased audience size.[172,173,174] The model also draws on evidence that stuttering increases when a competing linguistic activity diverts attention away from speaking.[175,176] The P&A Model is illustrated in the figure.[‡]



Testability

To date, there has been no critical test of the P&A Model. One such test would be to scan the brains of genetically at-risk infants prior to stuttering onset, and to demonstrate the presence of a central nervous system anomaly in all those who subsequently began to stutter. Because the model posits that atypical neural processing is a necessary condition for stuttering, no child who develops stuttering should be without such an anomaly prior to stuttering onset. Although there has been a preliminary scanning study of neonates at risk of stuttering and controls,[177] it does not provide such conclusive evidence. The developers of the theory also venture that their proposition "would be falsified if stuttering were shown to occur during nonsyllabized vocalization, for example during the production of extended vowels" (p. 359).[164] Also, the model would be challenged if stuttering did not decrease under experimental conditions that reduced linguistic stress contrasts. Such an experiment should be possible because acoustic correlates of linguistic stress are well known, including syllable intensity, fundamental frequency and duration.

The P&A Model suggests, for those who stutter, a systematic and measurable relation between physiological arousal and stuttering rate. The model would be refuted if there was shown experimentally, in groups of those who stutter, to be no correlation between physiological arousal and the occurrence of stuttering moments. Existing data touching on this matter are not so favourable for the P&A Model. Experiments about this matter suggest that there may not be such a systematic relation. Such a relation has not been found with stuttering contingent electric shock,[178] with challenging interviews compared to supportive ones,[179] with feared sounds,[180] or with standard reading and conversation tasks.[181] Additionally, there seems to be no relation between self-reported anxiety and either subjective or objective measures of stuttering severity.[182] There is, however, acoustic evidence[183] that emotional arousal causes more second formant frequently fluctuation with those who stutter compared to controls.

Explanatory power

Causal factors

The P&A model draws on existing knowledge about the disorder. It overtly posits *atypical neural processing* within its causal explanation, and it is intuitive to link it to *genetics*. The P&A Model provides a mechanism to explain *onset during language development* during a period of development when linguistic stress contrasts emerge.

[‡] Adapted and reproduced with permission: Packman, A (2012), Theory and therapy in therapy: A complex relationship, *Journal of Fluency Disorders, 37, 225–233*. © 2012 Elsevier.

Well-known features of the disorder

The P&A Model was designed specifically to explain research findings about stuttering, so not surprisingly, it does this well. Its original development was intended to explain the findings that vowel duration variability decreases with treatments that incorporate a fluency enhancing condition.[184,185] Indeed, the model can explain those treatments in terms of reduced vowel duration variability, which compensates for speech motor system instability by reducing linguistic stress during speech. A recent, functional magnetic resonance imaging report[186] provided the first neurophysiological details about the mechanism by which rhythmic speech might control stuttering.

The model can also explain the prominence of repeated movements at stuttering onset. They are the child's response to the problem by attempting to stabilise the speech system by minimising linguistic stress contrasts. Subsequent development of idiosyncratic fixed postures and superfluous behaviours are a less adaptive response to the problem. If myelination is involved in the neural processing impairment (see Lecture Two), the P&A Model can explain natural recovery for some children and a lifetime of stuttering for others. The posited underlying problem with neural processing can explain differences in stuttering severity across situations and within individuals. The severity of the underlying neural processing impairment would influence the baseline severity of stuttering across individuals, and the modulating effects of physiological and cognitive factors would explain idiosyncratic stuttering differences between speaking situations and times.

The model explains much of the influence of spoken language on stuttering, because stuttering on initial sounds and initial words of utterances is associated with linguistic stress. Stuttering is more likely to occur on linguistically complex utterances than simple utterances. The model is consistent with evidence that linguistically complex utterances contribute to instability of speech movements.[187]

Another issue with the explanatory power of the model is shared by all others reviewed during this lecture: the developers have not offered an explanation why around one third of children begin to stutter suddenly, during the course of a single day, or how verbal response contingent stimulation can control stuttering so well during early childhood.

Epilogue

The start of this lecture included reference to defunct causal theories of stuttering that were developed during the early decades of the 20th century. Many theories developed during the last decades of the 20th century have attracted little attention during this century, according to whether they have featured in recent peer-reviewed journals or published conference proceedings. Arguably, such theories are potentially destined to join those with an historical place in the discipline. These include the Sensory-Motor Modelling Theory,[188] the Neuroscience Model,[189] the Anticipatory Struggle Hypothesis,[190] the Two-Factor Theory,[191] the Neuropsycholinguistic Theory,[192] and the Suprasegmental Sentence Plan Alignment Model.[193] Those theories have been reviewed in several reference texts.[5,6,7] Should any of them generate future interest they will feature in subsequent iterations of these lectures. In the event that a new causal explanation is proposed and generates interest during this century, it will also be included in these lectures. As an example, one proposition this century[194] might well generate interest because it comprehensively explains how speech treatments, and the various speaking conditions outlined during Lecture One, can reduce or eliminate stuttering. In any event, the coming and going of so many causal theories about stuttering during the past century reflects its status as a puzzling disorder, arguably among the most puzzling that has affected humans.

Summary

The cause of the disorder is a necessary topic for discussion with clients and parents. Causal explanation also guides treatment development in the long term. The viability of a causal explanation includes its testability and explanatory power. Five causal explanations of stuttering have attracted interest during this century: multifactorial models, the Interhemispheric Interference Model, the Covert Repair Hypothesis, the EXPLAN Theory, and the P&A Model. On balance, no causal explanation has

yet resisted experimental disproof sufficient times, or over a sufficient period, to warrant any confidence. At present, though, it seems reasonable to say that stuttering appears to be somehow associated with an issue of neural processing, although details are far from clear. This broad concept, in various iterations, has resisted disproof since the early years of the last century.

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LECTURE FOUR: CLINICAL MEASUREMENT OF STUTTERING[†]

Six reasons for clinical measurement

Assessment

Clinical measurement provides a formal way to document the nature and severity of the impact of stuttering for clients when they first come to a clinic. Such impact may be behavioural or non-behavioural. Much of the non-behavioural impact of stuttering will be related to anxiety, which is outlined during Lecture Ten. Measurement of anxiety is described during Lecture Eleven. The behavioural impact of stuttering will be related to stuttering moments and how often they occur, as described during Lecture One.

This does not mean that clinical measurement is necessary to detect the impact that stuttering has for clients. To the contrary, with clinical experience, that impact will be obvious. However, clinical measurement provides numbers that quantify the impact of stuttering. For many reasons, it is a useful thing to record these numbers during client assessment. There are reference texts[1,2] that provide an overview of most formal clinical assessments for stuttering adults and children. Many of these assessments are not discussed here because they are used mostly in research contexts, not clinical contexts.

Communicating with clients

Clinical measures establish a common language between clinicians and clients, or parents, that can be used to communicate easily about everyday stuttering severity. For example, if a parent stated of a child "he was a 5 all yesterday," the clinician would immediately have a clear picture of the child's stuttering severity during that day. Such communication between clinician and client is essential in order to assess whether treatments are working as planned.

Stating treatment goals

When clinicians give stuttering treatments, they need a clear idea of what they are intending to achieve. Clinical stuttering measures can be used to convey to clients, or their parents, what those intended achievements are. For example, a clinician may say to a client: "That test shows that the impact of your stuttering is much less than six months ago." The formal description of this process uses terms such as *setting of treatment targets*, *treatment target criteria*, or *treatment goals*. Some treatments have standard, built-in treatment target criteria that may not be advisable to change, and for other treatments it is usual for the clinician and client to determine the treatment goals together.

Using measurement to document treatment goals and whether they have been met is part of treatment accountability. Stuttering treatment has to be funded, either through government or private sources. Those who provide that funding—government health care providers or the clients—need to know the outcome of their investment. Clinical measurement is an ideal way to provide that accountability by documenting client health improvements and how many hours of funded treatment were required to attain those improvements.

Assessing progress toward treatment goals

Clinical measurement does not stop after assessment. It is necessary to determine if a treatment is working as planned and that satisfactory progress is being made towards treatment goals. If progress

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[†] Thanks to Sue O'Brian for guidance with this material. This lecture deals with clinical measurement of stuttering severity, impact of stuttering, and speech satisfaction for those who stutter. Measures of anxiety are discussed during Lecture Eleven (Treatment of Social Anxiety and Stuttering).

towards those goals is not satisfactory, clinical measures may assist with documenting and exploring why that is the case, so that the problem can be fixed.

Managing maintenance of treatment gains

Health care resources for stuttering are valuable. Consequently, those resources are used inefficiently if clients do not maintain their treatment gains, and if they return to the clinic for more treatment, perhaps several times, after their original treatment. As will be discussed in Lecture Ten, such post-treatment relapse is a serious problem with adult clients. Post-treatment relapse is not so much of a problem with young children who stutter who successfully complete treatment, but it does occur.[3]

Clinical measurement can be used to monitor clients' post-treatment progress to detect any signs of impending relapse and to provide a clinical response if it begins to occur. The period after treatment that is designed to prevent relapse is referred to as *maintenance*. It is an indispensable part of any stuttering treatment.

Keeping track of daily stuttering severity changes

As noted in Lecture Three, stuttering severity is notoriously variable. To reiterate, stuttering severity is likely to vary with how many people are being spoken to at one time, with usually more stuttering when there are larger audiences. Stuttering will typically change severity across everyday situations, with lower severity typical of familiar conversation partners, and more severe stuttering likely when speaking with formal acquaintances and figures of authority. It is essential for clinicians to use clinical measurement to know about and keep track of such day-to-day variations during clinical management. For example, a clinician might ask a client to use a technique to control stuttering in a daily situation in which severe stuttering typically occurs. The clinician may ask the client to measure stuttering severity in that situation each day to explore whether systematic improvement is occurring.

Percentage syllables stuttered (%SS)

Overview

The views of 12 scholars in the field about stuttering assessment[4] included that a core component is "speech fluency and stuttering behaviors" (p. 2379), but there was no universally agreed method for such assessment. Percentage syllables stuttered, commonly abbreviated to %SS, is one such assessment. Compared to severity rating scales, which are considered later in the lecture, %SS is not a straightforward procedure; it requires equipment, and it is more logistically and arithmetically complicated, and clinically challenging. As such, clinicians may choose only to understand %SS for the purpose of reading clinical research literature, in which it features prominently. They may prefer not to use it in the clinic.

Percentage of syllables stuttered is a measure of the percentage of spoken syllables that are stuttered. It is sometimes referred to as a stutter-count measure because it is based on a count of unambiguous stuttering moments. To reiterate from Lecture One, unambiguous stuttering moments refers to moments during speech that are clearly stuttering and not normal disfluency.

Percentage syllables stuttered is based on syllables spoken, the syllable being a fundamental unit of speech production.[5] The average number of syllables in each word spoken increases from childhood to adulthood as language complexity develops. During adulthood the ratio is around 1.5 syllables per word spoken, but during the early years of life the ratio is much lower at 1.15, according to one source.[6]

When measuring %SS, syllables are thought of as being stuttered or not stuttered. For example, if someone speaks 900 syllables and 98 of them are unambiguous stuttering moments, that is 10.1 %SS. If someone speaks 1,435 syllables and 75 of them are unambiguous stuttering moments, that is 5.2 %SS. Percentage syllables stuttered is usually written to one decimal place.

How to measure percentage syllables stuttered (%SS)

A study explored the relative merits of standard and "challenge" phone calls to assess %SS.[7] The latter calls involved occasionally (but courteously) interrupting, disagreeing with, and talking over participants. Results showed little difference between the two approaches in terms of statistical analysis. However, the challenge phone calls elevated participant anxiety slightly, and for some participants this resulted in clinically significant increases of stuttering severity. Hence, the researchers argued that, in clinical practice, challenge phone calls might be a more valid speech assessment than standard phone calls.

There is evidence that, for adults, a %SS score during a 10-minute everyday conversation is representative of stuttering severity during that entire day.[8] That finding has been replicated with adolescents,[9] with the caveat that the finding pertains to group data only, but not to individuals. The former of those studies[8] is useful when interpreting %SS scores clinically. The speech of 10 adult participants was studied continuously during a 12-hour day, during which time their mean number of syllables spoken was 33,617, with a range of 17,274–50,463, and a standard deviation of 9,027 syllables. So that means, for example, if an adult stutters at 10 %SS for a 12-hour day, there could be somewhere between 1,700 to 5,000 stuttering moments during that day. Such data are currently not available for children.

When calculating %SS, a syllable is counted as stuttered only once, regardless of how many stuttering behaviours are associated with it. For example, consider "yesterday I-I-I-I, you see I, you see I, well, um I-I-I-I was here." That is counted as six syllables—"yesterday I was here—with one of them stuttered. The fact that there were repeated movements and superfluous verbal behaviours with saying "I," and two attempts to say it, does not change the fact that, for the purposes of calculating %SS, it was just one stuttered syllable.

Less commonly, percentage words stuttered is used, which is a similar measure but calculated by counting every word spoken, not every syllable spoken. That measure is now used rarely for research and clinical practice.

It is sometimes claimed that %SS is an objective measure, but, pedantically speaking, that is not correct. When measuring stuttering with %SS an observer needs to make a perceptual judgement about whether a syllable is stuttered or not; there is no objective truth to it. Another pedantic point is that the terms "stuttering frequency" and "stuttering rate" are often used interchangeably when referring to %SS. In a strict sense, however, percentage is a measure of proportion, so those terms are not correct. But they are commonly used, nonetheless. It is more correct to refer to %SS as a measure of stuttering severity.

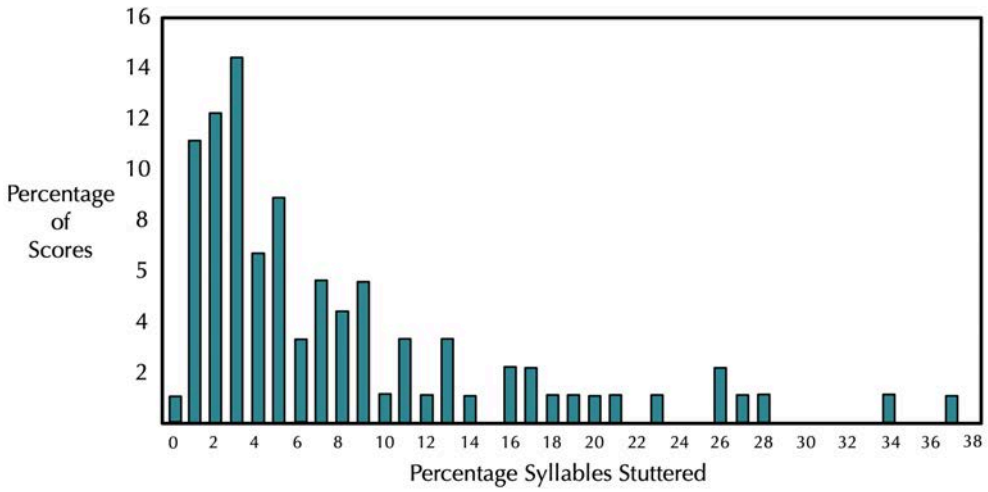
Percentage syllables stuttered scores are not normally distributed

With a clinical measure it is convenient if the population values are normally distributed, so the mean score falls in the middle of a normal distribution, with half the scores above the mean and half below the mean. It is known, then, that around two-thirds—68.2% to be exact—of cases are within one standard deviation either side of the mean. This helps to interpret extreme scores in terms of how far from the mean they are.

The distribution of %SS scores is not normal.[10] There are more mild cases than severe cases.[11,12,13] Information from the latter report[13] about adults is reproduced in the diagram below.[‡] The correct way of describing this distribution is to say that it is skewed to the right, and resembles a negative binomial distribution. The situation seems fairly much the same with early stuttering,[14] although %SS scores generally seem to be lower at that time of life.[11] In the figure below there are few people with scores greater than 20 %SS, but many with scores below 10 %SS. The median score is 4.8 %SS. The 60th

[‡] Adapted and reproduced with permission: O'Brian, S et al (2004), Measurement of stuttering in adults: Comparison of stuttering-rate and severity-scaling methods, *Journal of Speech, Language, and Hearing Research, 47*, 1081–1087. © 2004 American Speech-Language-Hearing Association.

percentile is 6.5 %SS, meaning that 60% of the scores—at least from this data set—are below 6.5 %SS.[†] For research that involves %SS, the implications of skewed scores are a little complex because there are mathematical issues with how they should be analysed and interpreted. Those statistical issues are discussed in detail elsewhere.[15]



Equipment for percentage syllables stuttered measurement

During clinical practice, %SS is typically measured during a conversation with the client/child, or while observing a conversation between a child and parent. A two-button counting device is used for measuring %SS, such as the one in the following photograph. One button is pushed for every syllable spoken without stuttering and the other button is pushed for every syllable spoken with unambiguous stuttering. The device automatically calculates %SS. Commercially available devices or smartphone applications can be used, or software is available for laptops.[16,17]

Considerable training is required to learn to use such equipment, but such training is readily available.[16,17] A disadvantage of using a smartphone application for measuring %SS is that the buttons are not mechanical. This may make it difficult to maintain eye contact with the client while measuring %SS online, as the clinician is doing in the photograph.

[†] The %SS scores in the figure are rounded to the nearest whole value. That is why there is one case of a zero score; the actual score was 0.3 %SS.

Limitations of percentage syllables stuttered

Validity

Counting the number of stuttering moments is not necessarily a valid reflection of how severe stuttering appears to an observer. This is because some stuttering behaviours may seem to be more severe than others. For example, fixed postures may appear to be more severe than repeated movements. Regardless, as discussed below, generally there is a strong and clinically significant correlation between %SS and perceptual measures of stuttering severity. But, of course, that will not necessarily be the case for any individual client.

Not viable for self-assessment

It is useful if clients and parents can self-administer a stuttering severity measure during everyday life. But considering the equipment and training needed for measuring %SS, this is not a clinical option. Consequently, clinical use of %SS is normally constrained to measurement in the clinic by the clinician. An option to obtain %SS measurements of clients during everyday speech is to have them make audio recordings—or even video recordings—of themselves, or have parents make recordings of their children, to bring to the clinic. Such recordings can be made easily with phones. Although it is time consuming to measure %SS from such recordings, in some clinical contexts the effort would be justified.

Reliability

Reliability is a general term for how well a measure gives the same score when used several times.[18] *Relative reliability* refers to how well a measure rank orders *groups of participants*. It is most commonly measured with a correlation coefficient or an intraclass correlation index. *Absolute reliability* (sometimes known as *agreement*) refers to the closeness of individual scores to each other and to a hypothetical "true score." It can be measured with percentage agreement, standard error of measurement, or limits of agreement.

Percentage syllables stuttered is a notoriously unreliable measure. The first report about this was in 1940,[19] showing poor absolute reliability: 20 clinicians ranged from 37–136 counts of stuttering moments from the same speech sample. During subsequent decades consistent evidence of such reliability problems emerged.[20] One paper[21] lists 32 research reports that contain measures of reliability. A recurring problem was absolute reliability: if one observer gives a certain %SS score, there is no guarantee that another observer will give the same score or a similar score.[22,23,24,25]

In the most notorious of these studies,[22] researchers gave the same 10 audio recorded samples, eight of which contained stuttering, to 26 clinicians in four countries and asked for their %SS measures; for which, the clinicians gave entirely different scores. Of particular concern was that scores for some samples in the low range had considerable variation: 0–4.2 %SS, 0.6–3.5 %, 0–2.1 %SS, 0–4.8 %SS, and 0–2.1 %SS. Such results suggest that some clinicians would consider some samples to contain no stuttering at all, while other clinicians would consider that the same samples had clinically significant stuttering that would require treatment. A more recent study[26] showed that students and generalist clinicians recorded less than half the number of stuttering moments as experienced clinicians.

In response to these reliability problems, a time-interval stuttering-count procedure[27] was adapted for stuttering.[28,29,30] With this method, the observer notes whether short periods of speech, such as 10 seconds, are stutter-free or whether they contain one or more stuttering moments. However, a subsequent review[31] showed that this method did not solve the reliability problems with stuttering-count measures.

It also appears that %SS reliability problems cannot be solved by listening to speech samples twice and counting syllables the first time and stuttering moments the second time.[32,33] The latter of these studies[33] also reported that it did not help to slow down speech samples while counting syllables and stuttering moments. A recent report about training procedures for stuttering counts[34] was more encouraging; however, the training by no means solved the reliability problems with stuttering counts.

There is evidence[35] that inexperienced observers are more reliable when they indicate whether each utterance contains one or more stuttering moments, compared to when they judge whether each syllable contains a stuttering moment. The same report found that reliability increased when observers had access to transcripts of the speech concerned. The observers were also able to complete the assessment task more rapidly when they had access to transcripts.

There is evidence[36] that %SS training in English does not necessarily generalise to using %SS with another language. Twenty-five English speaking clinicians, who did not speak Spanish, were trained to use %SS. They did not attain relative and absolute reliability when measuring %SS for Spanish speakers.

Severity rating (SR) scales

Overview

Differing numbers of scale divisions

Severity rating (SR) scales are a different type of clinical measure to %SS. Severity ratings are perceptual measures, where an observer listens to a sample of stuttered speech and uses the SR scale to record an overall judgement of severity. Or, a client can self-assign a SR score.

Severity rating scales have been around for years in various forms. They vary according to how many scale divisions there are, but the number is arbitrary. There is no real reason to think that a certain number of divisions is better than any other. Seven-point scales, 9-point scales, 10-point scales, and 11-point scales are commonly used.[†] Often, but not always, some or all of the scale divisions have labels telling the user what they represent.

Commonly used clinical severity rating scales

And example of a severity scale used for research is in the Illinois Early Childhood Stuttering Project (see Lecture Two). One version of the scale is 0 = *normal disfluency*, 1 = *very mild stuttering*, and 7 = *very severe*,[37] and another version is 0 = *normal speech*, 1 = *very mild stuttering*, and 7 = *very severe stuttering*.[38] A scale commonly used during treatment of early stuttering (see Lecture Six) is 0 = *no stuttering*, 1 = *extremely mild stuttering*, and 9 = *extremely severe stuttering*. A scale commonly used for older children, adolescents and adults (see Lectures Eight and Nine) is 0 = *no stuttering*, 1 = *extremely mild stuttering*, and 8 = *extremely severe stuttering*. Scores are commonly written in clinical files as SR 1, SR 2, SR 3, and so on.

A behavioural measure

Severity ratings have in common with %SS that they are intended as behavioural measures of stuttering severity. However, when clients score their own SRs, their scores may be inclined, to some extent, to reflect their nonbehavioural experiences with the disorder, notably speech anxiety. So, it is important to instruct clients not to allow such factors to influence their SR scores; their speech related anxiety can be measured using procedures outlined during Lecture Eleven.

Presenting severity rating scales to clients

It is useful to present SR scales visually to clients and parents, as well as describing them, and for them to have a copy in some form for their use outside the clinic. Here is how a SR scale might look when presented to clients:

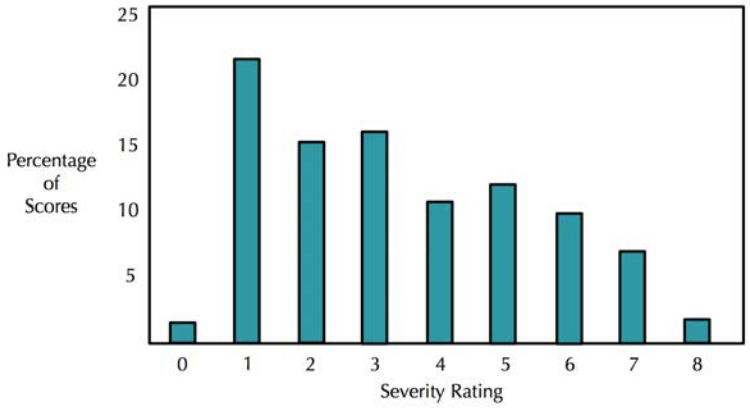
[†] An example of an 11-point scale is 0–10.

Equal interval ordinal scales

These SR scales are called equal interval ordinal scales. "Ordinal" means a sequence of numbers, and "equal interval" means it is intended that each scale division represents the same severity increment. Whether, in practice, such scales really are equal interval scales, or whether people tend to bunch up scores somewhere on them, is a complicated matter of psychophysics. That topic has been covered with specific reference to ordinal scaling of stuttering measurements.[39]

Severity rating scores are not normally distributed

As is the case for %SS scores, SR scores are not normally distributed, although their distribution is more normal-looking than for %SS.[10] The graph below[‡] shows clinician SRs on a 9-point scale using data from a report mentioned earlier with 90 adult stuttering participants.[13] The scale is 0 = *no stuttering*, 1 = *extremely mild stuttering*, and 8 = *extremely severe stuttering*. It shows mean SRs rounded to the nearest whole scale value. Cases that scored up to a mean SR 0.4 were rounded down to SR 0. Although these scores are not plainly normally distributed, they are certainly more normal-looking than the previous graph of %SS.[†] The median score is SR 3.0.



The data in that report[13] are clinician severity ratings. However, it appears that self-report severity rating data from adults provide a much more normal-looking distribution.[10,40] The latter report[t40] was an online survey of 505 adults who stuttered, using a similar scale to describe stuttering in everyday situations, and the distribution was nearly perfectly normal-looking.

Reliability of severity ratings

Logically, there is more chance of attaining adequate reliability with SR than with %SS, simply because SR scales have much fewer potential scores. By 2011, there were 11 research reports about SR reliability for stuttering as indicated in a publication (see Table 1, p. 1287).[21] Those reports show that while SR scales are not altogether free of problems, they are probably more reliable than %SS scores. There is also evidence that shows SR scores to be more reliable than %SS in a clinical context

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[‡] Adapted and reproduced with permission: O'Brian, S. et al. (2004), Measurement of stuttering in adults: comparison of stuttering-rate and severity-scaling methods. *Journal of Speech, Language, and Hearing Research, 47,* 1081–1087. © 2004 American Speech-Language-Hearing Association.

[†] The paper used a 1–9 scale, but the graph converts the data to a 0–8 scale.

where there is a need to detect changes over time with individual clients.[21]

For children, there is some evidence that, with little training, clinicians and lay listeners agree when using both SR and %SS scales,[41] and that parents of children who stutter have close rating agreement with clinicians.[42,43] One report[44] used a 10-point SR scale with 3–6 year olds speaking seven languages and clinicians who spoke those languages: Danish, English, French, German, Greek, Italian, and Persian. Results showed that neither language nor clinical experience influenced the clinician ratings. However, one report[45] showed that clinicians do not use the scale reliably when adult clients speak an unfamiliar language of Mandarin. A study of 25 English speaking clinicians,[36] who did not speak Spanish, showed adequate relative reliability with a 9-point SR scale when it was used for Spanish speech samples. However, there was a problem with absolute reliability among the clinicians, leading the authors to caution about the clinical generalisability of the scale into another language.

Ideally, clients and their clinical peers would all give the same SR for the same speech sample. In practice, though, experienced clinicians generally accept one-unit margins as acceptable limits, such as SR 6–7, SR 4–5 and SR 7–8.

A severity rating scoring guide[†]

After listening to a client for whatever period seems reasonable to be a valid speech sample, these four questions can be used to guide the assigning of a SR.

Were there any unambiguous stuttering moments?

If not, then the score is SR 0, which means *no stuttering*. If there were some ambiguous stuttering moments that could have been normal disfluency or might have been stuttering, then SR 1 would be appropriate, meaning *extremely mild stuttering*. Also, SR 1 would be appropriate if there was one unambiguous stuttering moment that was brief but not particularly bothersome: perhaps a syllable that was quickly repeated two or three times without a fixed posture or superfluous behaviour. Possibly, SR 2 might be appropriate in that situation, particularly if there was more than one such brief stuttering moment, indicating a little more severity than *extremely mild stuttering*.

Would a casual observer notice the stuttering?

A rule of thumb is that a casual listener would not normally notice SR 0–1: someone without a speech-language pathology background who would not make a sophisticated judgement about speech. That would be someone from the public who the client might encounter during everyday life, such as an accountant, bus driver, shopkeeper, waiter, lawyer, and so on. If it seems that such a casual observer would notice the stuttering, the SR would be 2 or more.

How much does it affect communication?

As discussed during Lecture One, stuttering is time consuming and on average those who stutter can say one third less than those who do not stutter, and with severe stuttering speech output might be less than a quarter than that of peers. So, a prime consideration when assigning a SR score is the extent to which stuttering affects communication. Reduced speech output will be one part of that consideration, as will how socially distracting stuttering may be because of superfluous behaviours. In cases where superfluous behaviours are particularly socially distracting and time consuming, a clinician may feel that communication is particularly affected.

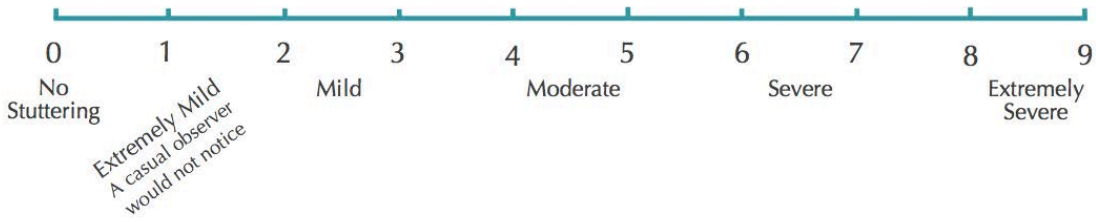
Was it mild, moderate, or severe?

It is useful to think of four categories to describe how stuttering affects communication: *mild*, *moderate*, *severe*, or *extremely severe*. *Mild* would be SR 1–3, *moderate* would be SR 4–5, *severe* would be SR 6–7, and *extremely severe* would be SR 8 (or 8–9 in the case of a 10-point scale). The categories *extremely mild*, *mild*, *moderate*, *severe*, and *extremely severe* are useful, incidentally, as

[†] Thanks to Sue O'Brian for this material.

informal descriptions of client stuttering severity. A report might read, for example, "I assessed this 37-year old man today who presented with extremely mild stuttering."

The diagram is a summary of the guide for a final decision about a SR score.



The clinical population as reference

Severity rating scales are of most use clinically if they are assigned with reference to the clinical population of those who stutter. In other words, SR 5 means a client is similar to others who stutter and come to clinics with that stuttering severity. At present, the only way to establish such reference points is with clinical experience and mentoring from a senior colleague. There are no generally available training methods to show inexperienced clinicians what a group of experienced clinicians believe are representative SR scores for the clinical population.

Clinical knowledge about the severity of the general population can be used to guide clients when using the SR scale. For example, if a client says that speech during a conversation in the clinic was SR 7, the clinician might say, "that was more like a 5." After watching a parent and child talking for a while, a clinician could ask "what SR would you give his speech just then?" The parent might say "4," and the clinician might say "yes, I agree," or the parent might say "3," and the clinician might say, "I would have given that a 4." Most clients and parents quickly learn to match the SR that the clinician would give.

Advantages of severity ratings

Simple

In contrast to %SS, a compelling advantage of SRs is that they are simple and require no equipment. Additionally, it seems that extensive training is not needed to learn to use them,[41,42,43] so they are particularly suitable for clients and parents, who can use the measures with themselves or their children. Further, they can be used easily with other languages without the need for detailed translation.[46]

This means that clinicians can have direct access to information about how severe stuttering has been during a certain period. For example, a clinician might say to a client "how was your stuttering last week" and receive a reply "1." In which case, the clinician knows that the client's stuttering was extremely mild during the previous week. Another example question would be "how has your stuttering been during phone calls to that customer?" The simplicity of the SR scale allows it to be used with considerable clinical flexibility. For example, a clinician may ask a client or parent to record a "typical SR" and a "worst SR" during a defined period during a day, or for an entire day.

Valid

It seems that SRs are more valid than %SS because they take account of all behavioural features of stuttering rather than just a count of stuttering moments. There is some evidence that clinician SRs take account of some relevant information about severity that %SS does not, which is discussed shortly.

Covert

Severity ratings can be done covertly. Clinicians can assess clients' stuttering severity in the clinic without them being aware it is occurring. This prevents the so-called Hawthorne Effect with stuttering

assessment, where behaviour can change when it is overtly assessed. In the case of children, parents are able to use SRs to covertly measure their children's stuttering at any time of the day when they are together.

The relation between %SS and SR

A strong relationship

There is a strong correlation of .91 between these two scoring methods when used by clinician observers for the same speech samples.[13] The effect has been shown in Kannada.[47] This means that the two measures can be used interchangeably with some confidence, but with two reservations. In the study mentioned,[13] what prevented a higher correlation were several cases where the %SS score did not correspond at all with the SR score. This occurred several times when samples of stuttering had high proportions of repeated movements or low proportions of fixed postures.

The second reservation about the matter is a report[48] that, to be reliable, %SS scores depend more on audiovisual samples than SR scores. Percentage syllables stuttered scores were 18% higher when scored using audiovisual samples than audio only samples, but this did not occur for SR scores. This is not an issue when talking face to face with clients and measuring stuttering severity, but it does suggest that SR is a preferable measure when clients bring audio recordings of their speech to the clinic.

For clinical research purposes, it seems that, at least for early stuttering, %SS and SR do equally well for documenting the results of stuttering treatment during clinical trials, and so the simplicity of SRs makes them a better option in that context.[10,49] (Clinical measurement during clinical trials is discussed during the next lecture.)

There is some evidence that, considering behavioural complexity, clinician severity ratings are a more valid measure than %SS and client self-rated stuttering severity.[50] As outlined during Lecture One, a stuttering moment can involve repeated movements, fixed postures, or superfluous behaviours. The report[50] found that clinician-rated stuttering severity had a significant relationship to the complexity of stuttering moments in terms of how many repeated movements, fixed postures, and superfluous behaviours are present. No such relationship was found for %SS or client-rated stuttering severity. When explaining the latter finding, the authors suggested that, when judging severity, those who stutter focus on the experience of stuttering rather than its behavioral manifestations.

Repeated movements and fixed postures

Repeated movements are generally not as socially distracting as fixed postures, and they quite often consume less time. So, if a %SS score for a sample is quite high because of many stuttering moments with repeated movements, it will not necessarily mean that the SR score for that sample will be high also. Observers may not think that all these stuttering moments with repeated movements are particularly severe stuttering.

Conversely, consider a speech sample that has a quite low %SS score because there are only a few stuttering moments, but those few stuttering moments are fixed postures and they are particularly socially distracting and time consuming. Such a sample might score a low %SS but a higher SR because the distracting and time-consuming nature of those fixed postures leads an observer to believe that stuttering is quite severe.

Percentile ranks for %SS and SR

The table[‡] shows the comparative percentile ranks for the two measures for an adult caseload.[13] Clinicians gave both measures based on 3-minute video speech samples. The table shows, for example, that the 50th percentile for SR is 3.0 and for %SS is 4.8. In other words, for that data set, half of a clinical caseload will be below those values and half will be above.[†]

PERCENTILE	SR	%SS
10	1.2	1.2
20	1.5	2.0
30	1.9	2.9
40	2.6	3.6
50	3.0	4.8
60	3.9	6.5
70	4.7	8.6
80	5.6	12.6
90	6.8	19.5
100	8.0	36.9

The relation between %SS and SR during treatment

Appendix One to this lecture is an example of a clinical file showing the use of %SS and SR measures during treatment of a child for 12 weeks. The clinical file illustrates the association between the two measures. The clinician has made a %SS measure in the clinic each week, as well as recording parent SRs for each day of the week prior to each clinic visit.

The child scored SR 0 consistently for the last few weeks of the file record, with only the occasional SR 1, and the clinician %SS scores were virtually zero for that period. This is an example of a successful treatment of childhood stuttering. The parent SR scores are indicating 0 = *no stuttering* most of the time, with the occasional SR 1 = *extremely mild stuttering*, which a casual observer would probably not notice. The clinician %SS scores verify that result.

Syllables per minute (SPM)

Sometimes a clinical measure of speech rate, most commonly syllables per minute, is associated with %SS. Devices that measure %SS typically have a timer that allows syllables per minute measures to be generated. Because stuttering moments consume time, if stuttering decreases after treatment, then speech rate would be expected to increase. It is necessary to use this clinical measure during a treatment that incorporates speech rate targets, which some modern treatments for adolescents and adults do. Progress has been made toward development of a smartphone application for monitoring and feedback of client speech rate.[51]

Speech naturalness (NAT) measurement

Why measure speech naturalness?

The speech restructuring treatments that figure prominently in these lectures are clinically useful for reducing stuttering but may not produce speech that sounds completely natural. This has been known to be clinically problematic for a long time.[52] Speech restructuring treatments involve a trade-off between speech that has no stuttering moments, or a few stuttering moments, and speech that sounds natural. So, a measure of speech naturalness is useful during such treatments to measure how natural clients sound and to guide them in attaining speech that sounds as natural as possible, while providing the desired stuttering reduction.

[‡] Adapted and reproduced with permission: O'Brian, S et al (2004), Measurement of stuttering in adults: comparison of stuttering-rate and severity-scaling methods, *Journal of Speech, Language, and Hearing Research, 47*, 1081–1087. © 2004 American Speech-Language-Hearing Association.

[†] The paper used a 1–9 scale, but the table converts the data to a 0–8 scale.

A scale of speech naturalness

A 9-point scale was developed during the 1980s and 1990s and is now used widely by researchers, and sometimes clinicians, to record speech naturalness.[53,54,55,56] For research purposes, it has been shown mathematically that:

> For posttreatment data, the average of three independent raters, and for pretreatment data, the average of five independent raters should give a result within one scale point of the hypothetical true score for the speaker in at least 80% of samples. (p. 718)[57]

There is evidence[58] that speech pathology listeners and general community listeners give different scores to clients who are using a speech restructuring technique to control their stuttering. The community listeners gave scores 1.6 scale values higher—more unnatural—than the speech pathology listeners. Also, among the community listeners, men gave scores 1.3 higher—more unnatural—than women.

Stuttering-Like Disfluencies

Stuttering-Like Disfluencies is a measure that is used for research publications from the Illinois Early Childhood Stuttering Project (see Lecture Two) and has also been used by some other researchers as a measure of stuttering severity:[59]

> The three disfluency types most typical of stuttering in young children (part-word repetition, monosyllabic word repetition, disrhythmic phonation) were combined to form a global category that we labelled Stuttering-Like Disfluencies … (p. 38)

The language of the measure—"stuttering-like"—is ambiguous, and consequently it has been criticised several times because it is not clear to what extent it relates to stuttering or normal disfluency.[60,61,62,63] Part of the issue is that the taxonomy specifies that children who do not stutter have fewer than 3.0 Stuttering-Like Disfluencies per 100 syllables, which implies that nonstuttering children show speech behaviours that can be referred to as "stuttering-like." The potential problem arising from this paradoxical terminology is illustrated in a publication[64] that used the measure and had the following wording in its title: "… frequency of stuttering in young children who do and do not stutter" (p. 2133).

There is a complicated algorithm based on Stuttering-Like Disfluencies: Weighted Stuttering-Like Disfluencies.[65] The algorithm is designed to predict natural recovery from early stuttering. It is derived from coded transcripts of language samples:[66]

> The weighted SLD is computed by summing PW *[part-word]* and SS *[single-syllable]* repetitions per 100 syllables of speech and then multiplying this value by the mean number of PW and SS RUs *[repetition units]* combined. This value is added to twice the sum of blocks and prolongations (collectively called as DPs) *[disrhythmic phonations]* (p. 2559, italics added)[67]

The Stuttering Severity Instrument (SSI-4)

The Stuttering Severity Instrument examiner's manual[68] is now in its fourth edition, often abbreviated to SSI-4. The SSI-4 is a more detailed measure of stuttering severity than either %SS or SR. It involves a composite single-number index that contains information about %SS, the duration of the three longest stuttering moments, verbal and nonverbal superfluous behaviours, and speech naturalness. The speech naturalness scale is the one described above. The superfluous behaviours, referred to as "physical concomitants," are scored on a 6-point scale where 0 = *none* and 5 = *severe and painful looking.* The SSI-4 can be scored manually or with a computerised version. For comparison purposes, there are normative data for 72 young children, 139 school-age children and 60 adults. In order to use this measure, the forms and manual need to be purchased from the publisher. The test has been translated to Persian.[69]

The SSI-4 is designed for research and clinical applications. It is reported often in stuttering research reports, although not as commonly as %SS. It takes considerable time to complete because client speech needs to be transcribed and analysed. Its time requirements are not an issue for research applications, but may be an issue for clinical applications where a stuttering severity measure is required at each weekly appointment.

There have been several reports questioning the reliability of this measurement instrument, which have been reviewed in a more recent report that again questioned its reliability.[70] Another report[71] shows that the SSI-3 (the previous version to SSI-4) provides no additional information than can be obtained from a SR scale. Considering this, and considering that it involves expense to purchase and clinical time to complete, the SSI-4 may not be a useful routine measure for generalist clinicians. However, clinicians who specialise in stuttering treatment may wish to purchase it and commit the time needed to complete the assessment before and after treatment, and perhaps on one or two occasions during treatment.

The Speech Efficiency Score (SES)

A group of researchers has begun developing this measure as an alternative to stutter-count measures.[72] The SES is derived from waveform analysis of speech, and calculates "the portion of the time during which the speaker produces speech fluency out of the overall speech time" (p. 62).[72] Encouraging results were presented for 15-second audio speech samples, showing that the SES is a viable alternative to SR and %SS. The intention of this research is to eventually to develop "algorithms for automated segmentation and calculation of the SES" (p. 67).

The Overall Assessment of the Speaker's Experience of Stuttering (OASES)

The impact of stuttering can be measured with the Overall Assessment of the Speaker's Experience of Stuttering,[73] commonly known as the OASES. It is designed to reflect the World Health Organization's International Classification of Functioning, Disability, and Health.[74] Each of the OASES questions requires a response on a 5-point scale, with higher scores reflecting more adverse impact. There are OASES Australian normative data to supplement North American normative data.[75]

The OASES is a questionnaire with four categories of questions about the impact of stuttering: general information, reactions to stuttering, communication in daily situations, and quality of life. The OASES score is the total of the four sections. It takes around 20 minutes for the client to complete. The scale was developed in 2006[76] and is starting to appear regularly in publications. To use this measure, the forms and manual need to be purchased from the publisher.

The OASES can be a useful part of a clinician's assessment tools for documenting impact of stuttering before and after treatment. For this purpose, there are three versions for different ages. The OASES-S is for school-age children 7–12 years, the OASES-T is for adolescents 13–17 years, and the OASES-A is for adults 18 years and older. The OASES-A has been shown to have acceptable reliability and validity,[75,76] and preliminary results for OASES-S and OASES-T are reported in the treatment manual.[73] The OASES-S has been translated into Dutch (OASES-S-D)[77] and Portuguese (OASES-S-PT),[78] and the OASES-A has been translated into Japanese (OASES-A-J)[79] and Hebrew,[80] with data showing it to be reliable and valid in those languages. All OASES versions have been translated to Swedish,[81] and been shown to be reliable and valid. The OASES-A has been shown to capture dimensions involving spontaneity while speaking[82] and feelings of everyday satisfaction with communication.[83] OASES scores have been shown not to relate to objective measures of stuttering severity such as %SS nor the SSI-IV.[83,84]

A systematic review dealing with measures of the psychological impact of stuttering on school-age children[85] (see Lecture Eleven) included the OASES-S. It was one of two tests with some support for its measurement properties. The authors of the review planned only to include measures of psychological impact that had their developmental data reported in peer-reviewed journals. Consequently, they

waved that criterion, and included the OASES-S in the report, even though the only supportive data for it are reported in the commercially available test manual. This is a caveat for the use of the test: its supportive data are not peer reviewed and are only available for scrutiny when the user has purchased the manual.

The Wright and Ayre Stuttering Self-Rating Profile (WASSP)

The Wright and Ayre Stuttering Self-Rating Profile (WASSP) is another stuttering impact measure that was also designed to reflect the World Health Organization's classification system.[86,87,88] The WASSP is not as empirically developed as the OASES, and as yet there are no normative data. It appears to have been designed both as a clinical and research measure, with intended clinical application for assessment and demonstrating post-treatment client changes. The developers indicate that its contents can be used to plan treatment. Each of its 24 items is scored with a 7-point scale from *none* to *very severe*. Those items measure the domains of "stuttering behaviours (8 items), thoughts about stuttering (3 items), feelings about stuttering (5 items), avoidance due to stuttering (4 items), and disadvantage due to stuttering (4 items)" (p. 84).[88]

The WASSP developers report that the test has adequate reliability and validity.[88] It is briefer to administer than the OASES, with 10-minutes completion time reported. Purchase from the publisher is required for its use. It appears that the test is used often in the United Kingdom where it was developed, but availability elsewhere currently seems to be limited.[88] A Turkish version[89] has been shown to have acceptable reliability and validity.

Simple speech satisfaction scales

Often, clinical reports measure client speech satisfaction with a simple scale, such as a 9-point scale where 1 = *extremely satisfied* and 9 = *extremely unsatisfied*. Versions have been reported where 1 = *extremely happy* and 9 = *extremely unhappy*. Parents can use such scales to measure satisfaction with their children's speech. In addition to having the advantages of simplicity and validity of client self-rating scales, a speech satisfaction scale can be clinically useful because it is an overarching measure that (presumably) takes overall account of any behavioural and nonbehavioural features of the disorder that impact on the client.

A more complicated version has been suggested [90] where clients make a judgment about their speech using a 10-point scale where 1 = *very bad* and 10 = *excellent,* with intermediate points on the scale labelled as *bad, very strongly insufficient, strongly insufficient, insufficient, sufficient, more than sufficient, good, and very good*.

A simple, overarching scale[83] is the Satisfaction with Communication in Everyday Speaking Situations (SCESS). It involves the question "considering all the issues associated with your stuttering, how satisfied are you with your communication in everyday speaking situations at the present time?" The scale is 1 = *extremely satisfied* and 9 = *extremely dissatisfied*. The SCESS scale relates well to the OASES, self-reported stuttering severity, but not to %SS.[83]

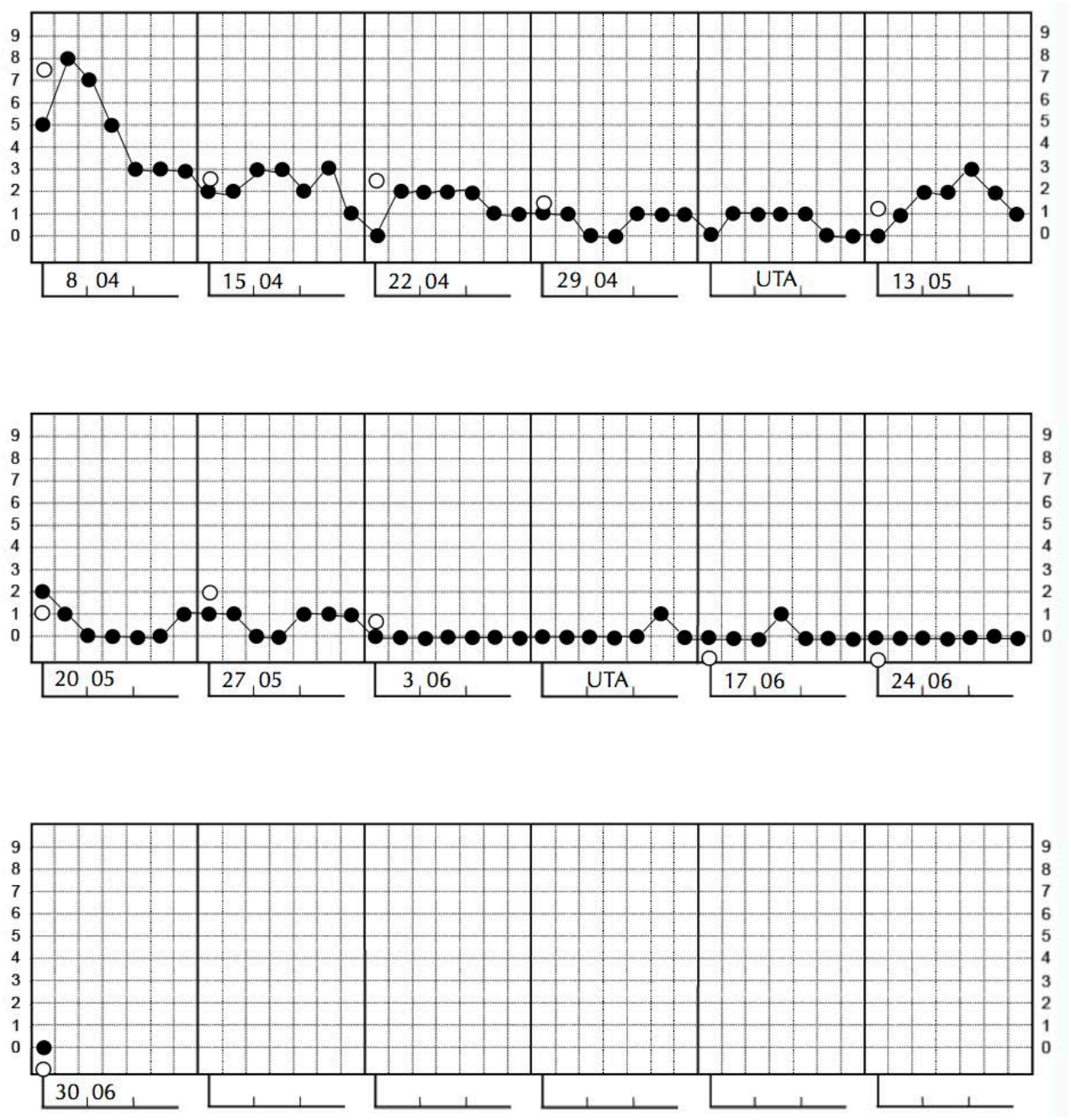
Summary

Clinical measurement is essential to assess clients and communicate with them about their stuttering. It is also essential to state treatment goals, to assess progress towards them, and to manage the maintenance of those treatment goals. Stuttering severity can be measured most conveniently with %SS and SR. Severity rating measures have clinical advantages related to their simplicity and validity, and their covert use with clients when needed. Generally, %SS and SR measures seem to measure the same dimensions of stuttering severity, with some important caveats. Speech naturalness is a useful measure for treatments that involve a trade-off between stuttering control and natural sounding speech. There are options available for measuring the impact of stuttering for clients before and after treatment. A glossary of clinical measurement procedures is presented in Appendix Two.

Appendix One

%SS and SR measures during clinical management of a child with early stuttering

Closed circles are parent SR scores for each day. Open circles are clinician %SS scores during each clinic appointment. The numbers under the chart are the dates of the first clinic appointment of the week. UTA = unable to attend.



Appendix Two

Speech and impact measures for stuttering

SPEECH		
Percentage syllables stuttered	*%SS*	*A stutter-count measure of the proportion of spoken syllables that contains an unambiguous stuttering moment.*
Severity rating	*SR*	*A perceptual measure of stuttering severity using an ordinal scale.*
The Stuttering Severity Instrument	*SSI-4*	*A more detailed and time consuming measure of stuttering severity than either %SS or SR.*
Syllables per minute	*SPM*	*A measure of speech rate.*
Speech naturalness	*NAT*	*A perceptual measure of how natural speech sounds using an ordinal scale.*
IMPACT		
Overall Assessment of the Speaker's Experience of Stuttering	*OASES*	*A stuttering impact measure for adults, adolescents and school-age children involving domains of general information, reactions to stuttering, communication in daily situations, and quality of life.*
Wright and Ayre Stuttering Self-Rating Profile	*WASSP*	*A stuttering impact measure for adults, adolescents and school-age children involving domains of stuttering behaviours, thoughts about stuttering, feelings about stuttering, avoidance due to stuttering, and disadvantages due to stuttering.*

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LECTURE FIVE: EVIDENCE-BASED PRACTICE WITH STUTTERING

What is evidence-based practice?

Evidence-based practice, or evidence-based medicine as it is sometimes known, is a health care philosophy that incorporates evidence from systematic research. Its philosophy applies not only to provision of health care to individuals who seek it, but also to government health care policy and administration.[1] Evidence-based practice originated with clinical medicine, but has attained widespread, international acceptance in many health care domains, including speech-language pathology. The best-known definition is:

> Evidence-based medicine is the conscientious, explicit and judicious use of current best evidence in making decisions about the care of individual patients. (p. 71)[2]

Another more recent definition explicitly mentions the mathematics involved with generating research evidence. Much research that is clinically useful—but not all of it—involves numbers of some kind derived mathematically:

> Evidence based medicine is the use of mathematical estimates of the risk of benefits and harm, derived from high-quality research on population samples, to inform clinical decision-making in the diagnosis, investigation or management of individual patients. (p. 1)[3]

A comprehensive video is available,[4] containing an interview with a speech-language pathologist, which overviews how evidence-based practice applies to health care generally.

Speech-language pathology and evidence-based practice

Evidence-based practice has influenced the discipline of speech-language pathology. The American Speech-Language-Hearing Association is by far the largest professional speech-language pathology association in the world, and arguably the most influential. In 2005 it proclaimed:

> It is the position of the American Speech-Language-Hearing Association that audiologists and speech-language pathologists incorporate the principles of evidence-based practice in clinical decision making to provide high quality clinical care. The term evidence-based practice refers to an approach in which current, high-quality research evidence is integrated with practitioner expertise and client preferences and values into the process of making clinical decisions.[5]

What evidence-based practice is not

Not a rulebook

Evidence-based practice is not a rulebook about how to provide treatment. Rather, it is a philosophy to guide treatment decisions:

> Evidence based medicine is not "cookbook" medicine. Because it requires a bottom up approach that integrates the best external evidence with individual clinical expertise and patients' choice, it cannot result in slavish, cookbook approaches to individual patient care. External clinical evidence can inform, but can never replace, individual clinical expertise, and it is this expertise that decides whether the external evidence applies to the individual patient at all and, if so, how it should be integrated into a clinical decision. (p. 72)[2]

Not a source of all clinical knowledge

To fully understand how evidence-based practice influences clinical practices with stuttering, it is essential to know the limits of science. In short, systematic research is an indispensable source of

knowledge to guide clinical practice, but, as stated in the previous quote, it is by no means the only source of knowledge that a clinician draws on. The abstract of Gerald Siegel's seminal paper on the topic summarises the limits of science:

> Science is a powerful tool when it addresses the kinds of questions it was designed to answer, but there are also important questions in communication disorders that fall outside the limits of science. Three such areas are discussed: Questions concerning social and personal values, questions that call for logical rather than scientific endeavors, and questions that should not be posed because we already know the answers and would not be influenced by contrary findings. (p. 306)[6]

Some examples of domains of clinical practice that fall outside the limits of science are empathy and emotional support for clients, listening skills, and hope and belief that intervention will help them. These are sources of knowledge cited by Siegel that relate to social and personal values and logic, and which research would not change.

Not a replacement for common sense

A paper published in the British Medical Journal[7] makes a point about common sense during clinical practice very well. The authors report no evidence that parachutes improve health outcomes when jumping from aircraft, and therefore recommend that common sense might be applied to the matter of determining the health care value of parachutes.[†] Their point, simply, is that the quality of client care will be compromised if common sense is deleted from clinical reasoning. An example with stuttering treatment would be a client with intellectual disability. Common sense indicates that the results of treatment research might not apply to such a client as they would other clients.

How to do evidence-based practice

There are many expositions about how to do evidence-based practice, but the following is a simplified summary based on the steps of the process as described at a classic source.[8] Evidence-based practice has been applied to health rehabilitation generally[9] and specifically to speech-language pathology[10] and stuttering treatment.[11] An issue of the *Journal of Fluency Disorders* was devoted to the topic of evidence-based practice and stuttering.[12,13,14,15,16]

Step One: Find out what the client needs

In order to be fully informed, clinical judgements need to be "moderated by patient circumstances and preferences" (p. 737).[17] In other words, in addition to research evidence, clinicians need to incorporate what they establish about clients' needs and their circumstances. An example of the importance of client circumstances in clinical reasoning would be a case where parents of a stuttering child are separated and share custody. In that case, evidence-based practice decisions may be different to when parents are living together and one parent spends the day with the child during the week. Another example would be an adult who seeks control of stuttering during everyday conversations. Some clients will wish to control stuttering in certain situations only, and some clients will wish to control stuttering during the entire speaking day.

What might clients need?

At the most basic level, the clinician needs to determine why clients have come to the clinic. This idea of complaint-centred treatment is certainly not new for stuttering.[18,19] Broadly speaking, the issues that cause someone who stutters to present to a clinic will relate to either behavioural or non-behavioural matters. Either there will be some need to control stuttering, or a need to deal with a non-behavioural feature of the disorder, or a combination the two. Most likely, non-behavioural issues will involve speech-related social anxiety in some way, as will be discussed during Lectures Ten and Eleven.

[†] There is no mandate that scientific journals are humourless.

Information pertinent to this matter was presented for the clinical experiences of 71 adult members of the National Stuttering Association, which is a United States self-help group for stuttering.[20] Behavioural treatments to deal with stuttering were the most commonly reported intervention received, but 49% of respondents reported receiving treatment that "involved reducing the fear of stuttering or of speaking situations" (p. 120).[20] Fifty-three per cent reported a combination of behavioural and non-behavioural approaches, and 25% reported non-behavioural treatment "with little emphasis on speech" (p. 120).[20] Thirty-three per cent "were disappointed because treatment did not address their feelings about their speech" (p. 122). Not surprisingly, the 9% who stated that "their therapist did not seek information from them in the decision-making process" (p. 122) reported dissatisfaction with the treatment process.

A survey of 24 children and adolescents who stuttered and their parents[21] was conducted after treatment. Responses were roughly evenly divided among the clients and parents about a preference "to speak without stuttering" or not. An e-Delphi Survey report[22] of 35 adult who stuttered and 13 speech-language pathologists, who were expert in stuttering treatment, established views about core components for adult stuttering treatment. The statements where a consensus was achieved included "management of communication-related anxiety" (p. 121) and "working on speech directly to reduce the amount of stuttering" (p. 122).

With early stuttering it is usual that the prominent need will be for behavioural stuttering control. With older clients, the situation may not be as straightforward, and it might take some time to establish client need. As will be discussed during Lecture Ten, from the school-age years through adolescence to adulthood, it seems that the likelihood of social anxiety becoming a clinically pertinent issue increases.

Step Two: Find the relevant evidence

The next step is to know or find the best evidence about how to provide what clients need. There are three broad categories of such evidence that inform stuttering treatment: basic research, treatment process research, and treatment outcome research.

Basic Research

Basic research deals with the nature and the cause of the disorder. An example is research showing the possibility that a child with early stuttering will have another speech or language disorder. This information will influence your assessment procedures.

Lecture Two covered epidemiological research about the nature of stuttering that clinicians may take account of when planning a treatment. For example, information about the chance of natural recovery from stuttering during the first year after onset will be a consideration in deciding when to begin early intervention, as will be discussed during Lecture Ten.

Another example of basic research that informs evidence-based practice is from Lecture Three, which presented information about the cause of stuttering. Research was presented that tests the veracity of various current theories. In deciding whether to intervene with early stuttering using a treatment based on the Demands and Capacities Model, a clinician may wish to form a view about whether that theory is substantiated by basic research.

Treatment process research

Treatment process research deals with how treatment functions, as well as factors that can affect how it functions. An example from the previous lecture is that speech restructuring treatments involve a trade-off between stuttering control and natural sounding speech. This information guides clinicians when deciding whether to recommend such a treatment for a client.

Another example of treatment process research that informs evidence-based practice is that percentage syllables stuttered (%SS) has been shown to be a notoriously unreliable measure, as outlined during the previous lecture. This research may influence a clinician's treatment process in various ways. For example, it might prompt a clinician to constantly calibrate %SS scores against a community reference, such as the training sources mentioned during Lecture Four.[23,24]

Treatment outcome research

For stuttering, and arguably for most health care domains, clinical trials are "the most fundamental, clinically interpretable, and useful output unit of stuttering treatment research" (p. 402).[25] They provide creditable research that conveys how useful treatments might be for improving the health of clients. The publication of a compelling clinical trial can change evidence-based clinical reasoning by increasing confidence in a particular treatment. Such a trial may introduce a completely new procedure to consider for clients. Or a clinical trial may show a treatment to be of limited or of no value. Clinical trials are discussed in detail shortly.

Step Three: Do the treatment and evaluate its effects

The final step in the evidence-based reasoning process is to administer the treatment and evaluate its effects. This can be done with regular application of the simple clinical measures outlined during the previous lecture, such as %SS and SR. The graph in Appendix One of that lecture is an example of clinical measurement used to establish that a child responded favourably to treatment and continued to do so. If these clinical measures had not shown that a change was apparent after several weeks of treatment, the clinician would have needed to problem-solve and make changes to the treatment process.

Scientific standards for clinical evidence

Peer-reviewed scientific journals

Clinician consumers of research need to determine whether research is sufficiently creditable to warrant consideration. A defensible rule of thumb here is whether the research has been published in a peer-reviewed journal that is listed in a creditable data base such as PubMed or the Web of Science. Publication sources apart from peer-reviewed journals are sometimes referred to as the "grey literature," and include student theses, books, book chapters, and internal institutional reports.

That being said, the standards of scientific journals, and the rigour of their peer review, are not at all uniform. For example, some peer-reviewed journals do not meet standards for inclusion set by prestigious databases such as the Web of Science.[26] This has prompted a strong caution that "some … published articles belong in the bin, and should certainly not be used to inform practice" (p. 31).[3] In any event, there is cause to regard with serious reservation any research that has not been reviewed and endorsed by peers within the scientific community and cleared for publication by the editor of a respected journal.

Hierarchies of evidence

Subsequent to a decision about whether research is sufficiently credible to warrant consideration, clinician consumers of research then need to make a further judgement about the standard of the research. To inform that decision, there are some generally accepted overarching standards for health care research, presented as hierarchies of evidence. Prominent examples are the University of Oxford Centre for Evidence-Based Medicine,[27] the National Health and Medical Research Council of the Australian Government,[28] and the Cochrane Consumer Network.[29]

A common theme appearing in all of those is that at the top of the hierarchy—the most convincing evidence for health care—is a systematic review that synthesises evidence from numerous randomised controlled trials using meta-analysis. Scientific journals and textbooks regularly publish systematic reviews, and The Cochrane Collaboration[30] is a well-established and trusted online source of systematic reviews.

Apart from a systematic review, those hierarchical classifications[27,28,29] then specify that a minimum of one randomised controlled trial is the next best level of evidence. They specify methodological variants of randomised designs as less compelling, such as pseudo-randomisation and cluster randomisation. Non-randomised designs are relegated to lower levels. These include case control and cohort studies, followed by case studies of groups and individuals and time series studies of

individuals. The Oxford Centre for Evidence-Based Medicine[27] specifically places "expert opinion" as the least admissible source of evidence for the value of a treatment. In the context of stuttering treatment, reliance on expert opinion rather than on scientific research to guide treatment has been described as assertion-based practice.[31] Experts, many of them charismatic, commonly proclaim the merits of certain treatments when there is no creditable research evidence to support their claims. The media often endorses such claims because of their charismatic nature.

Detailed methodological critique

When a clinician decides that a published research paper is worth considering, it is then necessary to make a value judgement about its methodological credentials. This judgement needs to be informed by detailed and rigorous scientific knowledge. For example, a clinician may devalue the importance of a report on the grounds that the authors used a misleading statistical analysis procedure. There are many sources of guidance for how such detailed critiques of scientific publications might be conducted.[3,32] Such texts often contain checklists for evaluating scientific publications, which include detailed items such as "if the statistical tests in the paper are obscure, why have the authors chosen to use them?" and "were outliers analysed with both common sense and appropriate statistical adjustments?" (p. 223).[3]

There is a website available that provides methodological critiques of treatment reports in speech-language pathology,[33] and gives each report a quality rating. The site was modelled on similar sites for clinical psychology and physiotherapy. It includes critiques of many papers dealing with stuttering.

For most common health problems, there are specifically designed standards for health care research. In the case of stuttering treatment research, a detailed, 136-item checklist has been proposed[34] which clinicians can use to critically appraise stuttering treatment research evidence. The authors reported that inexperienced judges are able to use it reliably. The checklist was based on research standards that have been historically endorsed by leading scholars and researchers in the field.

This proposed checklist[34] has some controversial features. Its authors acknowledge that the randomised controlled trial is the gold standard for health research; however, they argue that it should not necessarily be placed at the top of the hierarchy of evidence for use by clinicians for evidence-based practice with stuttering because "the vast majority of stuttering treatment research uses other designs" (p. 127).[34] Another controversial feature of this checklist is that it does not necessarily require data to be collected by the standard "blinded" method, where the observer who collects the data does not know anything about the research or whether speech samples are pre-treatment or post-treatment. As a precursor to their checklist, the authors argue that stuttering treatment research ideally should have the following five fundamental methodological credentials:

(1) a randomised design or a single-subject time series experimental design
(2) data collected by a blinded observer or an unblinded observer who has agreement with a second blinded observer
(3) treatment outcome measures at pre-treatment, during treatment, and at post-treatment
(4) outcome measures are collected in the clinic and outside the clinic
(5) when a report shows reduced stuttering, speech rate and speech naturalness are shown to be normal.

The single-subject time series experimental design referred to in the first point involves many types of research designs,[35] which are sometimes called N=1 or N-of-1 trials.[36,37] They are recommended for use in situations which include rare disorders where it is difficult to obtain sufficient participant numbers for traditional clinical trials. However, this is not the situation with stuttering, which is a common disorder, as discussed during Lecture Two. A suggested summary of current views about N-of-1 trials in speech-language pathology[38] is that, to be compelling, they should incorporate "replication in at least five studies showing similar treatment effects with at least 20 patients and involvement of at least three research teams in at least three institutions" (p. 244).

Clinical trials of stuttering treatment

What is a clinical trial?

Because of the prominence of the clinical trial in health research, it is useful for clinicians to have some criteria for determining what is and what is not a clinical trial. There are many definitions available. The World Health Organization defines a clinical trial as

> … any research study that prospectively assigns human participants or groups of humans to one or more health-related interventions to evaluate the effects on health outcomes.[39]

The National Institutes of Health, which funds health research in the United States, defines a clinical trial this way:

> A prospective biomedical or behavioral research study of human subjects that is designed to answer specific questions about biomedical or behavioral interventions (drugs, treatments, devices, or new ways of using known drugs, treatments, or devices). Clinical trials are used to determine whether new biomedical or behavioural interventions are safe, efficacious, and effective.[40]

The major Australian government health funding bodies and Australian Universities involved with health research define it this way:

> A clinical trial is a form of human research designed to find out the effects of an intervention, including a treatment or diagnostic procedure. A clinical trial can involve testing a drug, a surgical procedure, other therapeutic procedures and devices, a preventive procedure, or a diagnostic device or procedure. (p. 33)[41]

Definition of a clinical trial for these lectures

For the purposes of these lectures, the following definition of a clinical trial is used because it is designed specifically with reference to stuttering treatment, and it incorporates reasonable consensus from within the speech-language pathology discipline:

> A clinical trial of a stuttering treatment is (a) a prospective attempt to determine the outcome or outcomes of (b) at least one entire treatment with (c) at least one pre-treatment and one follow-up outcome of at least 3 months in the case of a reported positive outcome, and (d) where outcomes involve speech observations that are independent of treatment and derived from recordings of conversational speech beyond the clinic. (p. 404)[25]

This definition contains an essential component of speech measurement outside the clinical setting. At present the gold standard for doing so involves the %SS measure, as outlined during the previous lecture. However, two recent reports introduced some potential flexibility here with findings that, for early and persistent stuttering, %SS and SR scores beyond the clinic do equally well for documenting the results of stuttering treatment during clinical trials.[42,43] The caution here, though, is that these findings have yet to be replicated.

Although this definition of a clinical trial is used throughout these lectures, it is an arbitrary perspective on the matter. The many reviews of the evidence for stuttering treatment efficacy present differing views about what should be regarded as a clinical trial of stuttering treatment. For example, this report[44] presents a far more liberal view of the matter than that just described, including reports that contain "any outcome relating to a positive effect on … communication or … social and emotional wellbeing" (p. 678)[44] without regard to follow-up or whether speech measures were collected beyond the clinic. At the other extreme is the 136-item checklist mentioned previously.[34] A middle ground position is a 29-item checklist presented by other authors.[45]

That being said, some details about the presently used working definition[25] are as follows.

Clinical trial standards

Prospective methods

Using the definition just outlined,[25] or in fact any definition, a retrospective study would not be regarded as a clinical trial of a stuttering treatment. Examples of retrospective studies would be file audits of previously treated clients,[46] and clinical follow-up of such cases.[47] That is not to say that retrospective reports of stuttering treatment outcome are not useful publications to consider during evidence-based practice. To the contrary, they are useful demonstrations of the potential efficacy of a treatment and may be important preliminary precursors to a clinical trial, and may in some circumstances be considered during evidence-based clinical reasoning.

Study of complete treatments

Many reports of stuttering treatment do not report about the entire treatment. Such reports are more appropriately termed clinical experiments than clinical trials. Again, that is not to say that clinical experiments are of no value during evidence-based practice. For example, a clinical experiment is capable of producing evidence that a treatment can stop the developmental course of stuttering.[48] However, clinical experiments do not contribute information about the outcome of the entire treatment.

Beyond-clinic speech measures

It is a generally accepted rule about clinical behaviour change that it should be measured beyond the clinic to be sure that it has really occurred.[49] This is because of what is known as discriminated learning, which refers to the learning of behaviour change that occurs in the clinical setting where it is taught, but not necessarily outside the clinic. Regardless, it is common sense that clinical trials need to show stuttering reductions outside the clinic, because treatment needs to improve speech during everyday life. Authorities in the field of stuttering agree about this matter.[50,51,52,53,54,55] The most common speech measure for clinical trials of stuttering treatment is blinded %SS scores.

Follow-up period

The clinical trial definition presented earlier specifies that speech measures are collected after a follow-up period of at least 3 months. That is a liberal requirement. Normally, for a treatment to be regarded as useful, researchers would need to demonstrate that clinically significant treatment effects remain in place for a year or more after treatment. Often, clinical trials of stuttering treatment do report data with such follow-up periods.

Phases of clinical trial development

Phases I to IV

Clinical trials normally proceed with four developmental stages, from Phase I to Phase IV. The components of each stage, particularly the number of participants involved, differs from discipline to discipline. For example, there are normally more participants in clinical trials of drugs than in clinical trials of stuttering treatments. There is a short and readable introduction to clinical trials that was written specifically for a speech-language pathology audience.[56] It is a general rule that clinicians can have more confidence in the results of clinical trials when they are at a more advanced phase of development. Given equivalent methodological rigour, a Phase III clinical trial is far more compelling than a Phase II clinical trial.

The CONSORT Statement

Standards for an acceptable clinical trial are specified in the Consolidated Standards of Reporting Trials (CONSORT) Statement.[57] The group who drives and maintains the CONSORT Statement comprises scientists and medical journal editors. Many prestigious medical journals will not accept a clinical trial unless it conforms to the CONSORT Statement, and there is an increasing trend for

authors of clinical trials, including trials of stuttering treatment, to indicate that their trial design conforms to those guidelines.

Phase I clinical trials

An early stage of treatment development

Phase I clinical trials are normally the first stage in a sequence of treatment development. For stuttering research, they normally involve only a few participants. Their purpose is to develop preliminary evidence that justifies continued development of the new treatment. The kinds of information sought with Phase I trials are whether clients will comply with the treatment, whether it is safe, and whether there is any suggestion that there might be a treatment effect.

Non-randomised

Phase I trials are not randomised, meaning that there is only one group that receives the treatment. Measures are made pre-treatment and then post-treatment. Because of the few participants that are involved in Phase I clinical trials, and because they are non-randomised, they are the least compelling of clinical trial evidence.

Phase II clinical trials

A "green light" for further trials

The next stage of clinical trial development is the Phase II trial. These normally have more participants than Phase I trials and are designed to collect more convincing evidence of any potential treatment effect. A Phase II trial can give a "green light" for the conduct of a Phase III Clinical Trial. During Phase II trials, the safety and viability of the new treatment continue to be monitored, the treatment is adjusted according to need, and the final treatment protocol is developed.

Can be randomised

Phase II trials are normally non-randomised, but they can be randomised.[58,59] Randomisation means that there are two groups, often a control group who receives no treatment and an experimental group who receives the treatment being developed. A variation is for two or more treatments to be compared against each other, possibly with that comparison involving a no-treatment control group. Randomisation of Phase II trials is a method with controversial features (Chapter 13).[60]

The importance of randomisation

There is a well-known effect where non-randomised trials overestimate the true effect size.[61] In other words, they suggest that the treatment is better than it really is. The most common sources of bias in non-randomised trials are placebo effects and regression to the mean. Regression to the mean is where those who stutter seek clinical help when their stuttering is at its worst, only to improve subsequently because of natural variation. For clinical trials involving early stuttering, there is another ever-present source of bias: children in such trials might recover from stuttering not because of the treatment but because of natural recovery (see Lecture Two).

Randomisation ensures that the trial is as free of bias as a trial can be. However, clinical trials can never be completely free of bias. This is because participants in a clinical trial need to volunteer in order to be involved in the research, and such volunteers may be unlike those who generally present to clinics for speech treatment.

An advantage of a randomised Phase II trial is that it enables a mathematical calculation of what the true effect size is, in ways to be discussed shortly. When beginning a Phase III trial, it is necessary to have some idea of the effect size because it is used mathematically to determine how many participants are needed for such a trial.

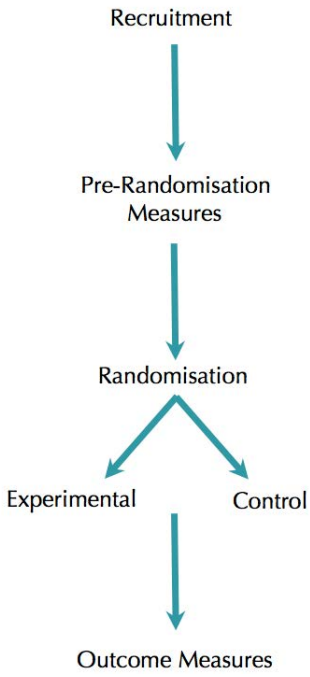
Phase III clinical trials

The "gold standard"

Phase III clinical trials are often referred to as the gold standard of clinical trial research. They are expensive and logistically taxing for researchers to undertake. A comprehensive guide to Phase III clinical trials of stuttering, written specifically for a speech-language pathology audience, is available.[62] Salient points from this guide are summarised in the flow chart above.

Recruitment

First, participants are recruited to the trial and give their consent to be randomised to one of the groups. For most trials of stuttering treatment, participants are recruited from clients presenting to a clinic, although newspaper and other types of advertising can publicise the trial. After recruitment, the pre-randomisation measure or measures are collected. These are referred to as pre-randomisation measures, not pre-treatment measures, because in a randomised controlled trial involving a no-treatment group, half the participants in fact receive no treatment.



Randomisation

Next, an independent person randomises the participants. Ideally, the independent person is a biostatistician, or a researcher who implements a randomisation method that has been prescribed by a biostatistician. There are several different ways of randomising participants to trials, according to features of the trial design and how many participants there are.

Treatment arms

As with Phase II trials, there can be three or even more groups, or *arms* to use the correct term. Treatments can be compared to each other or to a control arm. A trial with a no-treatment control arm and an experimental treatment arm compares the experimental treatment with no treatment. A trial can compare two treatments that are completely different, or it can compare variations of the same treatment. An example of the latter would be a clinical trial comparing a treatment given to participants in a clinic to a treatment given to participants by video telehealth. With clinical trials that compare two treatments there also can be a no-treatment control arm.

Primary outcomes

In a clinical trial, outcome measures are fundamental to how the outcome of the trial is judged. The CONSORT Statement strongly suggests that a randomised trial should have no more than one primary outcome. The reason given for this is that, from mathematical and logical viewpoints, more than two outcomes makes it difficult to interpret the results of the trial.

Secondary outcomes

Although ideally there should be one primary outcome for a randomised trial, there can be several secondary outcomes. Secondary outcomes are measures that are used to complement the primary outcome as measures of interest. For example, a randomised trial of a treatment to control stuttering may have %SS as the primary outcome, and secondary outcomes might be clinician and client SR scores, along with NAT and SPM scores (see Lecture Four).

Effect sizes

The most trustworthy estimate of effect size is obtained from a randomised trial where the response of one or more treatment groups is compared with the response of a no-treatment control group. Effect sizes can be estimated mathematically. A basic method for estimating effect size is Cohen's *d*.[63] This is

the difference between the mean primary outcome of the experimental and control group divided by the average standard deviation of the two groups. This gives a measure of effect size in standard deviations. By convention, a Cohen effect size of 0.2 is regarded as small, 0.5 medium, and 0.8 and greater as large. Cohen effect sizes can be larger than 1.0. There are several variations of the method for calculating Cohen's *d*.

Another way to measure effect size is the *odds ratio*, and this has been reported in several trials of stuttering treatment. This is a measure of the odds having a certain health outcome. For example, an odds ratio of 6.5 in a randomised controlled trial might mean that the group who received the treatment had 6.5 times greater odds of attaining below 1.0 %SS at post-treatment than the group who received no treatment. An odds ratio of 4 is generally considered to be favourable. There are related indices of effect size that have not yet appeared in the stuttering treatment literature: absolute risk reduction, relative risk, relative risk reduction and "number needed to treat."

Three ways effects can be significant

Measures of effect size are normally accompanied by a measure of statistical significance. For example, a report might indicate that a treatment group had better odds of attaining below 1.0 %SS than the control group, and report that *OR*=7.5, 95% *CI*=4.7–10.9, $p<.0001$. The way to read this is that the odds ratio was 7.5 with a 95% confidence interval of 4.7–10.9. In other words, the plausible range for the true odds ratio value, with 95% certainty, was somewhere between 4.7 and 10.9. If the confidence interval contains zero, there is no evidence of a difference between the groups.

However, this is not the entire story about the significance of effect sizes for stuttering treatment, or any treatment.[64,65] A difference may be statistically significant but of no practical significance. For example, a group may have a mean pre-treatment score of 12.7 %SS and a post-treatment score of 10.3 %SS. This could well be a statistically significant difference; however, such a small change is unlikely to be of any clinical significance. The term *personal significance*[65] takes account of the extent to which—regardless of numbers—a treatment remedies the life problems and consequent presenting clinical complaints that it causes.

In clinical psychology, the *reliable change index* is commonly used to define what is considered to be a clinically significant change,[66] using a statistical method to determine whether a pre-treatment to post-treatment change is statistically believable. The procedure requires information about the standard error, which is the standard deviation of a sampling mean. Such data are available for stuttering[67] and, hence, it is possible to use this procedure for %SS data before and after treatment, and this method has been demonstrated with stuttering treatment.[68] The reliable change index also allows assessment of whether treatment moves the client from a dysfunctional range to a functional range of performance on a clinical measure. The authors of the previous paper[68] argue— contentiously—that this can be done for %SS scores by using 0.5 %SS as the cut-off score for normal speech after treatment.

Drop-outs

With randomised controlled trials, researchers are required by the CONSORT statement to report drop-outs. Knowing how many participants dropped out of a trial, and from what arm, influences the confidence that can be placed in the results. If, for example, one quarter of all participants in the experimental arm dropped out of treatment, that would need to be taken into account when evaluating the results of the trial.

One approach to dealing with clinical trial drop-outs is by *intention to treat analysis*.[69] This means analysing trial outcomes of participants according to the treatment group into which they were randomised, regardless of whether they completed treatment or what treatment they completed.[†] One way to do this is by applying *last observation carried forward*. This means that if a participant drops

[†] Sometimes participants "drop-in," which means they receive the experimental treatment even though they were not randomised to receive it.

out after collection of pre-randomisation speech measures, for example, then those pre-randomisation speech measures are included as post-randomisation measures. This provides a conservative rather than a liberal estimate of effect size.

Phase IV clinical trials

Once Phase III clinical trials have established the value of a particular treatment, Phase IV clinical trials are used to determine how well they work among the community of clinicians who need to use them during everyday professional practice. Phase IV of clinical trials development is often referred to as translational research.

When discussing the merits of treatments, the terms *efficacy* and *effectiveness* are often used interchangeably. However, strictly speaking, they mean different things. Efficacy refers to a demonstration of the effects of a treatment under the specialised conditions of a clinical trial as conducted by professional researchers. Usually, clinical trials are conducted in dedicated research facilities, with specially trained clinicians, and explicit attempts are made to ensure that the treatment is being done correctly.

The strictly correct use of the term effectiveness refers to whether a treatment is useful when used by a community of professional clinicians who operate in the "the real world" of treatment, as demonstrated by Phase IV clinical trials.

Finding stuttering research to inform evidence-based practice

Finding clinical trials as they are published

Thousands of research papers have been published in scientific journals about stuttering, and hundreds are added every few years. Clearly, it is challenging for clinicians to keep up with such a burgeoning body of literature. However, based on the previous argument about the fundamental importance of clinical trials to everyday clinical practice with stuttering, it seems reasonable that finding and reading clinical trials from within the emerging literature should be a priority. There are several databases of scientific research that can assist clinicians to do this.

Step One: Set up regular database email alerts

Clinicians who have access to a library with research databases can arrange for those databases to send regular email alerts about publications about stuttering (and, of course, any other professionally pertinent research topics). For clinicians without institutional access to databases, there are freely available databases that can send regular email alerts.[70,71]

The most useful search string should include "stutter* or stammer*" in the title and abstract fields.[†] Adding the term "trial" or "clinical trial" to the search string will not necessarily be useful because clinical trials may not use the term "clinical trial" in either the title or the abstract.

Arguably, it is best to have email alerts sent once per week, for two reasons. First, it spreads out the workload of sorting through publications to find which are clinical trials (see below). Second, if a convincing clinical trial is published, then it is best to know about it as soon as possible because it could influence your clinical practices.

Step Two: Scan publication titles

When an email notification indicates that there has been a publication dealing with stuttering, the titles can be scanned for any publications that are obviously not clinical trials. For example, titles such as "eye gaze patterns during social interactions with stuttered speech," and "interhemispheric signal processing with stuttering" can be excluded immediately from results of a search for clinical trials.

[†] If a database will not accept the "*" truncation character, "stutter," "stuttering," "stammer," and "stammering," will need to be entered separately.

Step Three: Read abstracts of possible trials

If a title that comes up in a search that looks as if it might be a clinical trial, the abstract of the report can be inspected to see if it may indeed be a clinical trial. Examples of titles that suggest clinical trials are "Intensive stuttering modification therapy: multidimensional assessment of treatment outcomes"[72] and "Evaluation of a stuttering treatment based on reduction of short phonation intervals."[50] The latter report contained the following,

> All speakers achieved stutter-free and natural-sounding speech during within- and beyond-clinic speaking tasks at the completion of Maintenance. All were tested 12 months after completion of Maintenance. (p. 1229)[50]

This strongly suggests that the report might qualify as a clinical trial that needs to be read carefully. On the other hand, the abstract of another paper contains

> The study involved assessment of the children's speech fluency and a client satisfaction questionnaire that sought parents' opinions about which aspects of the treatment were beneficial. … Responses to the questionnaire indicated that treatment helped families learn about stuttering and about strategies that facilitate children's fluency. Evaluation of the children's fluency revealed that all participants achieved improved fluency at the conclusion of treatment and at long-term follow-up. (p. 118)[46]

This gives no indication of whether this could be considered a clinical trial, according to the present definition. Consequently, it needs to be read in detail to make a final determination. As it turns out, this is not a clinical trial according to the present definition,[25] because it was based on retrospective file audit data rather than prospective beyond-clinic speech measures.

Step Four: Read the clinical trials

When it is clear that the report is a clinical trial of a stuttering treatment, the paper can be read in detail. During this reading, clinicians can form a view of how, if at all, the trial could influence clinical practices. Part of this view should be the phase of clinical trial development. To reiterate, a Phase I nonrandomised clinical trial with few participants will be less compelling than a Phase III randomised trial with many participants.

The burden of work for all of this, so far, is reasonable. Based on current publication rates, it is extremely unlikely that in the near future more than 10 clinical trials would be published in any one year. If 1 hour is devoted to reading each clinical trial, the burden of work over an entire year keeping up with clinical trials of stuttering treatment would be 10 hours at most.

Step Five: Read as many other stuttering treatment reports as possible

Steps One to Three will identify abstracts of stuttering treatment reports that are not clinical trials but are basic research or treatment outcome research that can usefully inform evidence-based treatment practices. Reading all these increases the workload for a clinician who treats stuttering. However, it has been argued that:

> even the most complex stuttering treatment journal article can be assessed … in less than approximately an hour. Multiplied across many articles, this is an important time commitment, but it is not unreasonable, especially considering the alternative of continuing to spend time providing ineffective or otherwise less than ideal treatment. (p. 134)[34]

SUMMARY

Evidence-based treatment practices are an ethical requirement of the speech-language pathology discipline. Evidence-based practice incorporates judgements about the best research evidence to inform clinical management decisions with clients. Clinician judgements about the quality of research evidence can be informed by publications in peer-reviewed scientific journals, accepted hierarchies of evidence strength, and detailed methodological critiques of research publications. Clinical trials are

the fundamental output of clinical research that informs treatment practices. Clinical trials evolve treatment development in four stages that move from preliminary evidence of treatment effects to evidence of population effectiveness. Reviewing the scientific literature requires a time commitment by clinicians, but that time commitment is worthwhile in terms of its rewards.

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LECTURE SIX: EVIDENCE-BASED TREATMENTS FOR EARLY STUTTERING

Early intervention with telehealth

Telehealth

Current early stuttering interventions were developed for the traditional format of weekly clinic visits. The term *telehealth* refers to treating clients when they are not in the clinic. There are reviews available of telehealth in speech-language pathology with specific reference to stuttering treatment.[1,2] Professional speech-language pathology associations have also reviewed telehealth service provision and associated professional issues with it.[3,4,5]

During the Covid pandemic, 106 United States and Canadian speech-language pathologists[6] were surveyed about delivering the Lidcombe Program (a treatment to be discussed shortly) with telehealth. A majority of 94% indicated that they would include telehealth in future Lidcombe Program delivery. They reported the treatment to be easily adaptable to telehealth, with benefits including "time efficiency, flexibility of scheduling, and improved clinical processes" (p. 1). Reported challenges involved technology, establishing a clinical relationship, and identifying mild stuttering.

Telehealth seems viable for presenting the Lidcombe Program in a school setting. According to interviews of five school-based Australian clinicians,[7] the key issues were (1) understanding and managing the required technology, (2) logistics of the procedure in a school setting, (3) support from colleagues and the school, and (4) establishing family engagement with the telehealth model.

Video telehealth is currently accessible using laptops, tablets, and smartphones. The number of households with Internet connections is increasing rapidly, and internet transmission rates are constantly improving.

Advantages of telehealth for early stuttering intervention

Limited infrastructure needed

The traditional clinical infrastructure for pre-school children and parents is not needed for telehealth. All that is needed is a workspace, an Internet connection, and a laptop, tablet, or phone. Software that the clinician can use to video record entire treatment sessions is either free of charge or inexpensive.

Benefits of home clinician contact

Parents do not need to prepare their child for travel to the clinic, and either arrange childcare for siblings or bring them along to the clinic. There is evidence that parents can find this to be a significant burden added to doing the treatment.[8] A significant clinical advantage of telehealth is that the clinician sees the parents doing treatment with their children in their own homes. Not only from the perspective of common sense, but also in terms of generalisation theory,[9] this makes clinical sense.

Families isolated from in-clinic services

In large and sparsely populated countries, there will be many families who are isolated from in-clinic treatment services. Telehealth is useful for the many families who are in such a situation. In Australia, for example, one third of families live rurally, and, apart from geographical isolation, such isolation presents many barriers to treatment access.[10]

Facilitates specialisation

Telehealth facilitates clinical specialisation. It enables a specialist clinician to treat children who are located anywhere in the country. Additionally, with flexible working hours, a clinician can treat children anywhere in the world. Telehealth can therefore facilitate a clinician becoming known nationally and internationally as a specialist in stuttering.

Telehealth and stepped care

The four advantages of telehealth early stuttering intervention suggest that it may have a place within a stepped care model of healthcare delivery. That model contains two fundamentals.[11] First, it provides the simplest and most cost efficient method of health care that is efficacious. Second, it is self-correcting so that clients progressively escalate to more resource intensive, and more costly, models of health care if they are shown to need it. So, if families do not respond to telehealth early stuttering intervention, they might then go to a clinic each week. Or an intervening step might be that telehealth Lidcombe Program intervention is supplemented by occasional clinic visits. Treatment can begin at any step, not necessarily the first. Work has begun to develop a standalone Internet Lidcombe Program treatment that does not require a clinician,[12] suggesting the possibility of such treatment as the first intervention in stepped care. The stepped care intervention model has been shown efficacious with management of several disorders,[13,14,15,16,17] but there seems to have been only one description of the stepped care concept applied to stuttering.[18]

Three early stuttering treatments supported by clinical trials

Three early stuttering interventions that have clinical trial evidence to support them, using the definition of a clinical trial presented in Lecture Five[19] —either Phase I, Phase II or Phase III evidence—are reviewed here. These treatments are the Lidcombe Program, the Westmead Program, and two treatments that are conceptually similar to each other: Palin Parent-Child Interaction Therapy and RESTART-DCM treatment. The relative strengths and limitations of the three treatments are discussed, expanding on a previous discussion of the matter.[20] In the next lecture, the clinical research that supports the three treatments will be presented. Of all treatments considered, the Lidcombe Program has the most extensive evidence base of clinical research, so it is considered with the most detail.

The Lidcombe Program

Background

Basic research that led to its development

Lecture One described an extensive body of basic stuttering research from the 1950s and 1960s showing that response contingent stimulation can reduce stuttering to a clinically useful extent. During the 1970s, laboratory experiments showed that verbal response contingent stimulation could be used with children and could obtain similarly useful stuttering reductions.

The most famous of these experiments was the so-called puppet study of 1972,[21] which adapted a technique developed a decade earlier.[22] The researchers set up an illuminated puppet that conversed with children who stuttered. Under the experimental conditions the light was turned off during moments of stuttering, effectively making the puppet disappear. The researchers showed that this ingenious application of verbal response contingent stimulation successfully controlled the early stuttering of two children. That control generalised beyond the laboratory and was maintained for around 1 year. The Lidcombe Program involves the operant method (see Lecture One) of parents providing verbal response contingent stimulation to children who stutter. An early description of this type of approach appeared in 1971.[23]

Clinical resource materials

The Lidcombe Program Treatment Guide[24] is a clinical reference for the treatment which is available at the website of the Australian Stuttering Research Centre. At the website there is a clinical severity rating (SR) chart for parents and clinicians (*Child Stuttering Severity Chart eForm*).[25] It is reproduced in Appendix One of this lecture. The website also contains a pamphlet for parents about the treatment, written in several languages. Information about the Lidcombe Program has been made available beyond the speech-language pathology discipline to general and paediatric medical practitioners with overviews in medical journals.[26,27] Clinical checklists are available for clinicians to use to ensure they are doing the treatment correctly. One of these was validated by users,[28] and another is presented with case studies of its use.[29] One of those checklists[28] contains 63 items, and is reproduced at the end of the Lidcombe Program Treatment Guide.

The Lidcombe Program Trainers Consortium[30] has members in 12 countries and provides postgraduate training for the treatment. This training involves two days of instruction and demonstration, often with subsequent clinical follow-up. The developers of the Lidcombe recommend that clinicians do not attempt it without Consortium training. The Lidcombe Program is endorsed by the professional associations of several countries.[31,32,33]

Overview

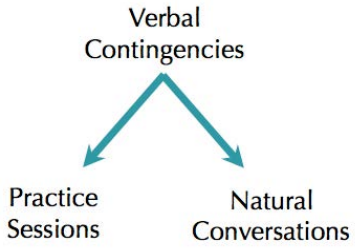
A behavioural treatment

The Lidcombe Program is a behavioural treatment, designed to deal with children's stuttered speech. It uses operant methods, even though, as discussed during Lecture One, stuttering is not freely emitted problem behaviour and in no proper sense is it an operant.

The Lidcombe Program is unlike the other two treatments considered during this lecture. It does not require children to change their customary speech pattern in any way, and it does not require any change to the customary living environments of children to remove features of those environments thought to cause or sustain stuttering.

Parents give verbal response contingent stimulation

Parents do the Lidcombe Program with training and supervision by a clinician. It involves parents giving their children verbal response contingent stimulation—verbal contingencies—for not stuttering and for stuttering. They do this during practice sessions with their children, designed specifically for this purpose, and during naturally occurring conversations with their children. On most occasions it is the parents who give the treatment to their children, but sometimes it may be caregivers.



Clinical measurement

Regular measurement of children's stuttering severity occurs during the Lidcombe Program.

Parents have an appointment each week

During each weekly appointment the clinician teaches parents how to do the treatment and ensures that it is being done properly. The treatment guide[24] specifies what occurs during each clinic appointment, and in what order.

Treatment goals during Stage 1 and Stage 2

Lidcombe Program treatment goals are no stuttering or nearly no stuttering for a long time. The goal of Stage 1 is no stuttering or nearly no stuttering, and the goal of Stage 2 is for that to be sustained for a long time. Stage 2 of the treatment is sometimes referred to as maintenance.

The severity rating (SR) scale[†]

Treatment goals specified with SR scores

Parents give their children a SR for each day and clinicians give a SR during each clinic appointment. They use the SR scale described during Lecture Four 0 = *no stuttering*, 1 = *extremely mild stuttering*, 9 = *extremely severe stuttering*. Lidcombe Program treatment goals are specified with SR scores.

> To progress to Stage 2, the following two criteria need to be met for three consecutive clinic appointments that are 1 week apart: (1) parent SRs of 0–1 during the week preceding the clinic appointment, with at least four of those seven SRs being 0 and (2) clinician SRs of 0–1 during the clinic appointment. A minimal requirement during Stage 2 is for parents to record SRs only during the week preceding the clinic appointment. However, the clinician may direct parents to record SRs more often during Stage 2. (p. 10)[24]

A flexible measurement

Parents give a SR to their children's speech for each day. This measurement procedure has some flexibility, as outlined in the treatment guide:

> Variations of the SR procedure can be added to the treatment process if the clinician thinks it would be useful, commonly one SR for the morning and one for the afternoon. Clinicians may wish parents to use supplementary SRs for a particular speaking situation that occurs each day, such as at dinner and bath time, and shopping. These are recorded in addition to the daily SRs. Other options are for parents to record a highest and lowest SR for each day. (p. 2)[24]

SR scores to determine if treatment is working as planned

Severity rating scores are used to check that children's clinical progress is satisfactory. If progress is not satisfactory then SR scores will alert the clinician and the problem can be solved. Such problem solving is a routine part of Lidcombe Program administration.

Accurate parent severity ratings are essential

It is essential for parents to use the SR scale accurately, or the treatment cannot work properly. If parents underestimate their children's stuttering severity with the scale, it can result in them being admitted to Stage 2 prematurely, before they have, in reality, attained the SR treatment goals just outlined. For example, parents might give an average SR of 0.3 for the week before a clinic appointment when the appropriate average SR is 2.3. The opposite situation, where parent SRs are too high, would waste clinical resources by causing the child to take longer for treatment than necessary.

Parent severity rating training

It is a simple matter to prevent such problems. To quote from the treatment guide,

> During the first clinic appointment, after the clinician has explained the SR scale, the parent or the clinician, or both, converse with the child for a few minutes until the child displays a reasonably representative amount of stuttering. After a few minutes the clinician asks the parent to assign a SR to the speech sample. The clinician indicates whether that is an appropriate score and, if necessary, suggests a different score. All subsequent clinic appointments begin with the parent conversing with the child, the parent assigning a SR score, and the

[†] Prior to 2015 the Lidcombe Program used a 1–10 SR scale, and publications before then contain that version of it.

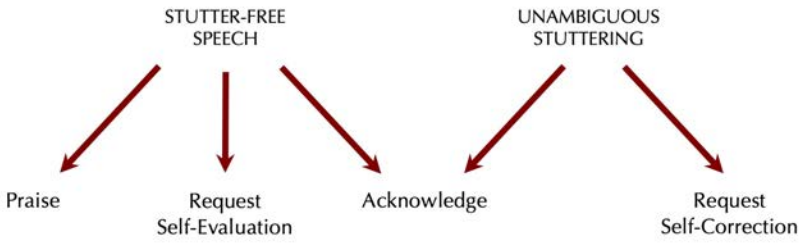
clinician either confirming that the score is appropriate or providing corrective feedback. (p. 3)[24]

Percentage syllables stuttered (%SS)

There has been a recent recommendation, with theoretical and empirical justification, for why %SS is no longer an essential part of the Lidcombe Program.[34] However, for reasons outlined in that publication, some clinicians prefer to use %SS during each clinic session when using the treatment.

Parent verbal contingencies

There are five verbal contingencies in the Lidcombe Program that the treatment guide specifies as essential. Three of these are verbal contingencies for stutter-free speech, and two are for unambiguous stuttering moments. An overview of the essential Lidcombe Program verbal contingencies is shown in the figure below.



Verbal contingencies for stutter-free speech

Verbal contingencies for stutter-free speech are central to the Lidcombe Program because, above all else, children must enjoy the treatment for it to work properly. Parent verbal contingencies for stutter-free speech are inherently positive and supportive, which is essential for clinical dealings with children who have early stuttering.

The first parent verbal contingency for stutter-free speech is *praise*. The clinician teaches parents to occasionally praise their children for not stuttering. Parents can be taught to say things like "That was lovely talking without bumps," or "Good talking, no stuck words," or something similar. It is essential for parents to do this in their own way. Every parent has a different style with a child, and different children need to be praised in different ways. It is also essential that parents are genuine with their praise and also that they don't do it excessively.

The second parent verbal contingency for stutter-free speech is *request self-evaluation*. This verbal contingency can be used when a child does not stutter for a certain period. That period can be as brief as a single utterance or as long as several hours. When no stuttering occurs for such a period, the parent can ask the child to self-evaluate stuttering during that period. The parent could say something along the lines of "Were there any bumps there?" and the expected response from the child would be "No." Or, a parent could say "Did you say all that smoothly?" and the expected response would be "Yes."

The third verbal contingency for stutter-free speech is *acknowledge*. The difference between this and the previous two verbal contingencies is that the conversation is not paused at all. This is most important; the child's everyday communication cannot be constantly disrupted each day by parent verbal contingencies. Also, acknowledging stutter free speech is different from praising stutter-free speech because it is a matter-of-fact statement rather than a positive comment. Examples would include: "That was smooth" and "No bumpy words."

Verbal contingencies for unambiguous stuttering

These need to be introduced carefully because some children can be initially apprehensive about them. Also, verbal contingencies for stuttering are more likely to make children react negatively to the treatment than are contingencies for stutter-free speech. They are used less frequently than verbal contingencies for stutter-free speech. In other words, most of the verbal contingencies that children

receive during the Lidcombe Program are for stutter-free speech. As is the case with verbal contingencies for stutter-free speech, every parent has a different style with a child, and different children will need to receive verbal contingencies for stuttering in different ways.

The first verbal contingency for unambiguous stuttering is *acknowledge*. As with the verbal contingency to acknowledge stutter-free speech, this verbal contingency needs to be not at all disruptive. The parent just notes that stuttering has occurred and moves on, saying something like "That was bumpy" or "That was a stuck word."

The second verbal contingency for unambiguous stuttering is *request self-correction*. The parent asks the child to repeat the utterance without the stuttering moment. Mostly the child can do that, but if the child fails to do so, it is usually best for the parent to let it go. Examples of request self-correction would be to say "Can you try that again" or "See if you can say that without the bump." Request for self-correction occurs occasionally, not on the majority of or on most stuttering moments. The exception to this rule is when child has only a few stuttering moments each day, which occurs towards the end of treatment.

Optional parent verbal contingencies

The Lidcombe Program Treatment Guide specifies two additional verbal contingencies that parents can use but which are optional. The first of these is *praise for spontaneous self-evaluation of stutter-free speech.* Older children with early stuttering receiving the Lidcombe Program, in particular, will sometimes spontaneously self-evaluate their speech as stutter free, saying something like "I didn't do any bumps." In such a case, a parent may respond with something like "Good boy, you're listening for your smooth talking."

The parent needs to be sure that the praise is for self-evaluation of stutter-free speech, not praise for stutter-free speech. Parents need to understand the difference between the two. For example, "Good boy, you're listening for your smooth talking" is praise is for self-evaluation of stutter-free speech, and "Good boy, that was smooth talking" is praise for stutter-free speech.

It is generally thought not to be a good idea to praise spontaneous self-evaluation of stuttered speech, such as "I just did a bump." The reason for this is that it might confuse a child if the parent's praise follows a moment of stuttering. If a child does spontaneously self-evaluate stuttering, parents can note that it occurred and tell the clinician at the next clinic appointment. Naturally, this is a desirable situation and a sign that the Lidcombe Program treatment process is working well.

The second optional verbal contingency is *praise for spontaneous self-correction*. When children correct a stuttered utterance without being asked by a parent to do so, parents can offer praise. Again, older pre-school children are the most likely to do this. The verbal contingencies that parents might use here include "Good girl, you fixed that bumpy word all by yourself," and "You fixed that stuck word, good boy."

Some essential things about parent verbal contingencies

They are for unambiguous stuttering moments

Lidcombe Program verbal contingencies for stuttering are for unambiguous stuttering moments. If parents have any doubt about a moment of stuttering, it is not a problem, and they can choose to not apply a verbal contingency. All children with clinical levels of stuttering will have many unambiguous stuttering moments each day, and parents will have plenty of them to work with. This normally only becomes a clinical issue at the end of Stage 1 when children have SR 0 or SR 1—*no stuttering* or *extremely mild stuttering*—during most days.

Teach verbal contingencies for stutter-free speech first

Clinicians don't teach parents how to do the verbal contingencies all at once. Normally, they first teach parents to do verbal contingencies for stutter-free speech so that children can become comfortable with the treatment. Then, they implement the parent verbal contingencies for stuttered speech with children when they are sure they are ready for it. It makes clinical sense to introduce

verbal contingencies for stutter-free speech before verbal contingencies for stuttering, because it is an inherently positive approach.

Be sure parents are doing them correctly

The clinician needs to be sure that parents are doing verbal contingencies correctly, according to instructions. The way to do this is, at each clinic appointment, to have parents demonstrate exactly how they have been doing the verbal contingencies with the child during the previous week, and to give them feedback. This can be an imposing clinical task for junior clinicians. It involves watching parents give verbal contingencies, making constructive comments, and then demonstrating improvements with the child. However, it is essential to do this during clinic appointments. Otherwise, the treatment process will not work properly if parents continue to do verbal contingencies incorrectly.

Verbal contingencies must be a positive child experience

The Lidcombe Program treatment process will not work properly if verbal contingencies amount to a negative experience for the child. Verbal contingencies cannot be constant, intensive, or invasive. It is an essential clinical skill to identify when this is occurring during treatment, or even better, to identify when it might occur and prevent it. For some parents, it is necessary to introduce the treatment slowly and carefully so they can be sure that the child is receiving supportive and enjoyable verbal contingencies. Otherwise, during clinic appointments, it will be obvious that the child is not happy with the treatment, and clinical outcomes will be predictable.

Verbal contingencies during practice sessions

Practice sessions

The clinician teaches the parent to present verbal contingencies during practice sessions for 10–15 minutes usually once per day, sometimes twice per day. Fewer or more each day can be recommended by the clinician as judged advisable. The parent typically sits with the child at a table, with suitable activities such as books and games. Such structure is not essential, however, and treatment during practice sessions can be done in many situations. But in many cases, perhaps most, the formality is useful.

Their purpose

Apart from parent training, the point of verbal contingencies during practice sessions is to accustom children to what the treatment procedures will be, and to focus their attention on the treatment target of "no stuttering." Overall, verbal contingencies during practice sessions establish a positive experience of the Lidcombe Program for the child.

Manipulating syntactic complexity and utterance duration

Lecture One presented research evidence that stuttering increases with increasing syntactic complexity and utterance duration,[†] and that these findings have been replicated with children. Clinicians can use this information to teach parents to manipulate these variables when giving verbal contingencies during practice sessions to minimise the occurrence of stuttered utterances, if needed. With such

[†] Utterance duration is usually measured with words, syllables, or morphemes.

manipulation, treatment during practice sessions can involve giving the child a chance to respond to a range of utterance durations: from one- and two-word responses to several utterances. It will depend on the child's stuttering severity at the time of the activity. This parent manipulation of syntactic complexity and utterance duration is not a static procedure; parents change their utterance duration and language complexity as needed, according to the children's stuttering severity during the practice sessions.

Verbal contingencies during natural conversations

Natural conversations

When the clinician forms a view that it is appropriate, parents begin to judiciously introduce verbal contingencies during natural conversations. The natural conversations are everyday speaking situations with children: at mealtimes, in the bath, on the way to pre-school, in the park with the family, whilst shopping, and so on. Eventually, verbal contingencies during natural conversations will replace verbal contingencies during practice sessions, and the latter will not occur at all.

Their purpose

The fundamental clinical premise of the Lidcombe Program, based on laboratory research, is that parent verbal contingencies are the active treatment agent. So, when the clinician feels it to be appropriate, it is logical for parent verbal contingencies to occur during natural conversations with children.

Stage 2

The purpose of Stage 2

There are three purposes of Stage 2. The first is to systematically hand over complete responsibility for management of children's stuttering to their parents. Second, Stage 2 is designed to detect any signs of impending relapse. As mentioned during Lecture Four, relapse after speech treatment for stuttering is common with adults. Although not so common with early stuttering, it does occur after the Lidcombe Program. In fact, half the children in one report[35] showed some transient signs of stuttering a mean of 5 years after their treatment began. So, the third purpose of Stage 2 is, after having detected any such signs, to prevent relapse from occurring.

Treatment goals for Stage 2

To progress to Stage 2, children need to meet the criteria mentioned earlier for two consecutive fortnightly appointments:

> (1) parent SRs of 0–1 during the week preceding the clinic appointment with at least four of those seven SRs being 0, (2) clinician SRs of 0–1 during the clinic appointment. (p. 10)[24]

Performance contingent maintenance

The idea of a performance contingent maintenance schedule was introduced to stuttering treatment, and its potential benefits were shown, in 1980.[36] It amounts to the parent and child returning to the clinic and having to sustain treatment targets for increasingly longer intervals; two appointments 2 weeks apart, then two appointments 4 weeks apart, followed by the same thing at 8 and 16 weeks between appointments. If the child does not meet the Lidcombe Program treatment criteria at any appointment, the parent and child return to the start of the sequence. Stage 2 normally takes a year or more. The importance of following this procedure was shown in a report that half of children during Stage 2 fail to meet treatment criteria at least once during Stage 2.[37]

A common Stage 2 problem

When children attain the Lidcombe Program treatment criteria and there is no stuttering or nearly no stuttering, parents or clinicians, or both, can become complacent and not follow through with the prescribed Stage 2 maintenance program. This causes a serious risk that relapse will occur. The researchers who published a long-term clinical follow-up of the treatment[35] suggested that clinicians encourage parents to watch carefully for any signs of post-treatment stuttering during Stage 2. It is essential that verbal contingencies for stutter-free speech continue to occur during Stage 2, and that any unambiguous stuttering moments receive verbal contingencies from parents.

The Lidcombe Program problem solving

Problem solving is a routine part of the Lidcombe Program. A study of common problems arose from 60 consultations with expert clinicians about cases where children were not improving.[38] Appendix Two at the end of this lecture shows the most common problems that needed to be solved. A more detailed and recent publication[39] presented 124 clinical challenges that occur during the Lidcombe Program treatment process, and presented strategies to deal with each of those challenges.

Clinical strengths and limitations of the Lidcombe Program

Strengths

Replicability

The basis of the treatment process is replicable, with measurement and verbal contingencies clearly described in the Lidcombe Program Treatment Guide.[24] The guide specifies what occurs during each clinic appointment, and in what order. The benefit of a replicable treatment is that any properly trained clinician can be confident of doing it exactly the way it was demonstrated to be efficacious in clinical trials.

Conceptual simplicity

The essence of the treatment is that parents present five verbal contingencies to their children during practice sessions and natural conversations, and measure their stuttering daily with a simple severity rating scale. Although the treatment is simple in concept, in practice it can be challenging to adapt it in a different way for every family, and to be sure that parents are doing the treatment correctly. These two features of the treatment—adapting it for each family and being sure that parents are doing it correctly—are essential for it to be successful.

Limitations

It is not ideal for immediate early intervention

The Lidcombe Program requires compliance from children. They need to participate in daily practice sessions and cooperate with the parent verbal contingency procedures. As discussed in the next lecture, it is common for clinicians to delay treatment for a period after onset. As noted in Lecture Two, many children begin stuttering prior to 30 months of age. In the event that a clinician decides to begin treatment immediately with a child who has begun to stutter at that age, the Lidcombe Program may not be ideal. In fact, in the next lecture, treatment process research is discussed which shows that treatment times are longer for younger compared to older children who receive the Lidcombe Program.

Safety issues

There is a safety issue with the Lidcombe Program concerning its use of parent verbal contingencies. Research has revealed the possibility that occasionally a child could react negatively to verbal contingencies.[40] It is possible, therefore, that without proper clinician management to prevent such an event, a parent could give verbal contingencies in a negative and punitive manner that might be detrimental to a child's well-being. This does not occur often, but it can occur.

Treatments based on Multifactorial Models: I. Palin Parent-Child Interaction Therapy[†]

Background

A treatment based on a multifactorial model

This treatment was developed at the Michael Palin Centre for Stammering Children in London. The treatment is one of many based on the multifactorial models described in Lecture Three. To reiterate briefly, these models state that what triggers stuttering and sustains it is subsequently found in predisposing motor, physiological, language, and developmental child variables and the way they interact with their living environments. None of these variables is *necessary or sufficient* for stuttering; they interact uniquely with the stuttering of each pre-school child. Palin Parent-Child Interaction Therapy is based on a theoretical position that is broadly consistent with this thinking.

The factors specifically mentioned by the developers of this treatment include:

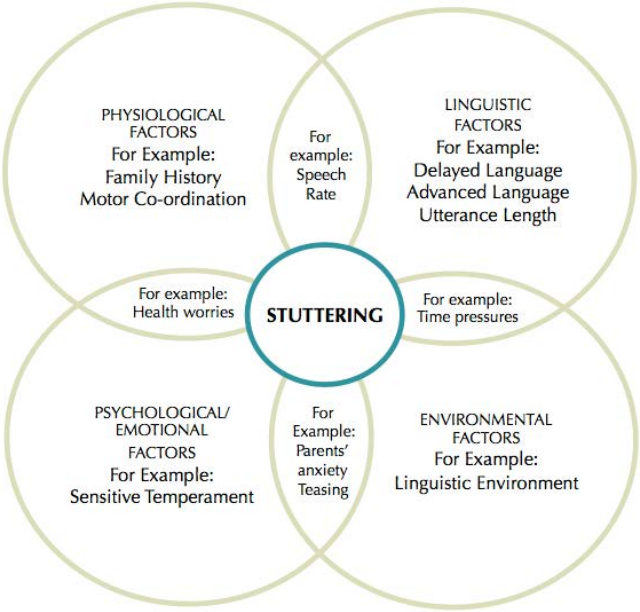
(1) Psychological aspects such as child temperament and parent anxiety

(2) Physiological factors such as gender, genetic history, and motor skill

(3) Language development

(4) Aspects of the living environment such as pace of life, communication and interaction style, parent language complexity, and rapid parent speech rate compared to that of the child.

Here again is the figure[‡] giving an overview the multifactorial model on which this treatment is based, which was presented during Lecture Three



Other treatment influences

One of the developers of the treatment has noted that[41]

> There is certainly an emphasis in identifying the individual child's strengths and needs, based on a belief that stuttering is multifactorial, heterogeneous and that the inherent vulnerability to stuttering is influenced by internal and external factors. The therapy itself is influenced by many approaches, including family systems theories, cognitive behaviour therapy, behaviour therapy, and solution focused brief therapy. (p. 3)

Clinical resource materials

Two journal reports contain overviews of the treatment.[42,43] A slightly longer overview appears in a book chapter,[44] and there is a comprehensive manual available.[45] Additionally, the Michael Palin Centre conducts a 3-day training for the procedure.[46] A web page at the Michael Palin Centre website contains general advice, in text and video media, to parents of children with early stuttering.[47] The advice to parents at that location includes the following:

[†] Thanks to Alison Nicholas at the Michael Palin Centre, London, for assistance with preparing this description of the treatment.

[‡] Adapted and reproduced with permission: the *Michael Palin Centre*, © 2014 Michael Palin Centre.

> "Having a short (5 minutes) one-to-one time with your child on a regular basis, when you are both calm and not in a rush and you are not likely to be interrupted
>
> Thinking about your child's general well-being, his sleeping and eating habits, his health and his pace of life
>
> Looking at your family's conversations - are you letting each other finish what you want to say? Is anybody hogging all the talking time? Do you interrupt each other when trying to speak?
>
> Building your child's confidence by focusing on what he is doing well and praising him for this
>
> Thinking about your child's language and whether he is trying to use sophisticated words and sentences to express himself. What kind of language are people using when they talk to him?"

Overview

Pre-treatment assessment

Palin Parent-Child Interaction Therapy begins with a detailed assessment that takes account of general speech and language development in addition to stuttering. The assessment is also designed to establish the extent to which children are aware of stuttering and how it may be affecting them socially and emotionally. This is needed because a key feature of the multifactorial model on which this treatment is based is that the putative factors responsible for stuttering combine uniquely for each child. In other words, the triggers for stuttering and what sustains it are different for every case of stuttering.

Diverse treatment goals

A fundamental difference between this treatment and the Lidcombe Program is that Palin Parent-Child Interaction Therapy is not designed to achieve "no stuttering" or "nearly no stuttering." Additionally, it has diverse goals. As stated by one of the developers of this treatment, "our aim is not zero stuttering during intervention. We seek to establish a decreasing trend in stuttering, reduced parental anxiety, and increased parental confidence in managing the stuttering*" (*p. 4*).*[41]

And, stated at another source:

> The main focus of Palin PCI is the child, his or her profile of skills, and facilitating further development of the natural occurring fluency within the environment. It also aims to build on parents' or caregivers' knowledge and confidence in what helps and enhances existing behaviours that support fluency. (p. 69)[44]

And at another source:[43]

> Palin PCI is explicit about the need to help parents address issues such as managing anxiety about stuttering, helping children manage emotions, confidence building, and other behaviour management such as setting boundaries and routines with, for example, sleeping, eating and turn taking. (p. 63)

Individual treatment design for families

With Palin Parent-Child Interaction Therapy, the clinician forms a judgement about which of the factors, mentioned earlier, will be targeted in a treatment program: psychological, physiological, language, or living environment. The clinician has 40 "interaction strategies" available, classified within 12 categories, as outlined in Chapter Six of the treatment manual:[45]

> (1) Following the child's lead in play
>
> (2) Letting the child solve problems for himself
>
> (3) Using more comments than questions during conversation

(4) Complexity of questions at child's level
(5) Using language which is appropriate for the child's level
(6) Using language which is semantically contingent on the child's focus of attention
(7) Using repetition, expansion and rephrasing of the child's utterance
(8) Giving the child time to initiate, respond and finish his talking
(9) Matching the parent's rate to the child's rate
(10) Using pausing before and between utterances
(11) Use of eye contact, position, touch, humour and/or surprise
(12) Using praise and reinforcement. (p. 91–125)

Additionally, there are 19 "family strategies" outlined in Chapter Seven of the treatment manual:[45]

(1) Managing two languages
(2) Openness about stammering
(3) Building confidence
(4) Giving children feedback
(5) Sincerity
(6) Consistency
(7) The language of praise
(8) Reactions to praise
(9) Helping parents to build up their child's confidence
(10) Turn-taking
(11) Dealing with feelings
(12) Difficulties with separation
(13) High standards
(14) Helping parents to manage their child who has very high standards
(15) Sleep
(16) Behaviour management
(17) Routines
(18) Pace of life
(19) Emerging issues. (p. 127–168)

The treatment manual[45] outlines "child strategies" in Chapter Eight, stating that "our research has shown that most children achieve fluency with the interaction and family strategies … However some children's fluency continues to be a cause for concern and we introduce direct fluency therapy at this stage" (p. 169). These strategies incorporate speech restructuring treatment components:

(1) Rate reduction
(2) Pausing to think
(3) Easy onset
(4) Being more concise
(5) Eye contact/focus of attention. (p. 169)[45]

The treatment process

"Special time"

Palin Parent-Child Interaction Therapy incorporates "special time," which is a 5-minute period that each parent spends individually with the child three to five times per week. The purpose of special time is to provide a comfortable environment in which parents can practice the targeted interaction changes. It is expected that the changes to parent interactive style will generalise to beyond these talking times, but nonetheless they continue to occur throughout the treatment.

Clinic visits

The treatment involves six weekly 1-hour clinic visits. The format of each clinic visit is consistent, with the exception that during the first clinic visit the results of the assessment are conveyed to parents and the routine for special time is established for the family. During special time parents keep a diary about the activity conducted and the targets that they implement, and this diary is presented to the clinician for discussion at the start of each session.

Both parents are required to attend each clinic session, during which a version of special time is conducted in the clinic and is video recorded. The clinician is nondirective during the treatment, and parents are encouraged to select their own treatment targets based on their observations of the within-clinic video. Parents are encouraged to identify the interaction styles that they are already using to support the child's fluency and they then select an interaction style that they would like to perform more often, and the introduction of any new targets is discussed with the clinician.

The consolidation period

After the six weekly clinic visits, there is a 6-week "consolidation" period, which occurs entirely at home. The purpose of this is for parents to consolidate the skills they have learned and generalise them to the home environment. There is no mention in any of the documentation about the treatment stating that targeted family interaction changes should generalise to beyond the home environment. However, the clinician may involve nursery or school staff as appropriate.

During the consolidation period, parents send their special time diaries to the clinician each week, and they receive written feedback from the clinician. An example of such a diary is presented in the treatment manual.[45] Subsequent to the 6-week consolidation period, review clinic visits are scheduled at 3 weeks, 3 months, 6 months, and 1 year.

Standard treatment period

Unlike the Lidcombe Program, Palin Parent-Child Interaction Therapy has a specified number of six clinic visits. It does not involve speech criteria that are used to establish an end to the treatment, although %SS measures are collected at assessment, pre-treatment, and post-treatment, along with parent rating scales. The clinician considers these measures when making a decision about the end of treatment.

Treatment flexibility

Although the treatment prescribes that there are six initial weekly clinic visits, there is some flexibility in allowing more if judged necessary. The developers state[42] that the duration of six clinic visits was selected because, at the time the treatment was originally developed, that number of clinic visits was a standard British National Health Service allowance to clinicians for treatment of children. The treatment manual states:

> In the first instance, we will book six sessions and then you will practise at home for six weeks. You will then come back in for a review session, when we can decide if he needs any more therapy. For many children, all we need to do at that stage is see them from time to time to keep an eye on things. (p. 84)[45]

Treatments based on multifactorial models: II. RESTART-DCM Treatment

Background

RESTART-DCM[†] treatment has much in common with Palin Parent-Child Interaction Therapy, conceptually and procedurally. RESTART-DCM also has popularity in common with that treatment,

[†] Rotterdam Evaluation Study of Stuttering Therapy-Demands and Capacities Model

being widespread in the Netherlands since the 1980s and "taught to Dutch students of speech therapy for the past 25 years" (p. 2).[48]

Overview

In common with Palin Parent-Child Interaction Therapy, the treatment is a hybrid, involving strategies derived directly from multifactorial models of stuttering causality and speech restructuring techniques. As stated in the treatment manual:

> The RESTART-DCM approach is never limited to simply providing advice to the parents. Depending on what is found to be necessary, the speech (stuttering) therapy provided will focus on behaviour changes, coping with emotions and skills training. If lowering the demands and promoting the capacities should fail to resolve the stuttering problem to a satisfactory extent, speech fluency may be worked on directly by *modelling* slower, more relaxed, smoother speech (p. 4)[31]

Additionally, all children are given an oral motor assessment.[49] If that assessment "should reveal that the oral motor skills are insufficient, reinforcement of the motor skills is a relevant therapy goal" (p. 12), and the child is treated with a method involving speech motor drills[50] in addition to the basic procedures.

Assessment

The RESTART-DCM treatment manual[31] outlines five standard tests of language, articulation and oral motor function. Additionally, two 10–15 minute video recordings are made of the child and parents playing together in their customary fashion. This interaction is then scored using the form in Appendix One of the treatment manual, which follows these categories (the italics indicate any "unfavourable behaviour" [p. 19] that is noted on the video):

> (1) Questions parent to child: *many; open; a commanding tone, or with little time for the child to answer*
>
> (2) Turn-taking behaviour: *talking simultaneously, interrupting; interaction times are too short*
>
> (3) Parent response to stuttering: *negative verbal reaction to the stuttering; negative non-verbal reaction to the stuttering*
>
> (4) Parent(s) linguistic behaviour: *introduce a new topic; correct child's verbal behaviour; make utterances that increase time pressure*
>
> (5) Articulation and/or speech rate
>
> (6) Other parental behaviour: *give negative attention; show directive action* (p. 19)[31]

In relation to categories (2) and (5), it appears that the treatment is not, strictly speaking, a clinical application of a multifactorial model of stuttering because the treatment is not different for every child: all parents speak with reduced speech rate to their children and with increased interturn speaker latencies:

> A rule of thumb to establish the right speed is to speak just as slowly as the child does when speaking fluently, unless the child has a relatively rapid rate of speech, i.e., > 3.5 syll /sec. In that case, the parent must learn to speak at slow-to-normal speed … (p. 8)[31]

Parents will rarely speak with a lower articulation rate than their children, so parents routinely need to reduce their rate during the treatment.[51] The same applies to parent interturn speaker latencies,[51] because parents need to have latencies of 1–2 seconds during conversation with their children with "definitely no overlapping speech" (p. 8), which rarely occurs naturally.

The treatment process

According to the treatment RESTART-DCM manual,[31] parents and children attend the clinic for an hour each week initially, but there is flexibility about the duration. Generally, after four visits, parents are invited to attend a clinic session during which the child is not present.

During the first clinic visit the clinician explains causal factors for stuttering and discusses stuttering with parents, with the discussion supplemented by written material presented from Appendix Two of the treatment manual. During the first session parents are instructed to provide "parent-child special times" for 15 minutes per day at least 5 days a week. As treatment progresses, these times are used to practise skills to lower demands and reinforce capacities according to clinician guidance. Parents keep a log of these special times.

Typical treatment sessions involve the clinician observing the parent playing and talking with the child, discussing progress during the previous week, and having the parent demonstrate treatment procedures that were used during the previous week. The clinician then outlines changes to clinical procedures for use during the coming week, demonstrates them to the parent, and has the parent attempt the procedures.

The following components of the treatment are outlined in the manual:

(1) Reducing motoric demands
(2) Reducing linguistic demands
(3) Reducing emotional demands
(4) Reducing cognitive demands
(5) Reinforcement of the speech motor capacity
(6) Reinforcement of linguistic capacity
(7) Reinforcement of the emotional capacity
(8) Reinforcement of the cognitive capacities
(9) Direct therapy with children aimed at more fluent speech. (p. 7–16)[31]

When the following program criteria are met, a 24-month maintenance phase begins, comprising three 30-minute clinic visits each month followed by one visit every 3 months for 21 months. The program criteria are

> The child has normal-fluent speech (very young children for approximately six weeks and older children [aged 4 1/2 -6] approximately 3-4 months) or exhibits only incidental disfluencies that are minimally abnormal (occasional repetitions with usually one iteration).
>
> The parents implement a fluency enhancing environment or the speech therapist/fluency expert judges that the parents can maintain the rest of the modification on their own.
>
> The child's speech is acceptable to the parents, the child and the speech therapist/fluency expert.
>
> The parents know what to do if a relapse should occur. (p. 16)[31]

Clinical strengths and limitations of treatments based on multifactorial models

Strengths

They can be used for immediate early intervention

With these multifactorial model treatments, the child does not have to do anything at all. It is only the parents who do the therapy. It is a completely passive treatment from the perspective of children. As such, these treatments are often described as indirect. Because of this they are suitable for children who stutter at any age.

Limitations

Potentially complex treatments

Palin Parent-Child Interaction Therapy and RESTART-DCM Treatment appear to be the most complicated and logistically challenging treatment for clinicians from among the three discussed

during this lecture. The treatment manuals show that more than 60 therapy strategies are involved with each of the treatments. Data about the matter are limited, but one of the clinical trials of Palin Parent-Child Interaction therapy[42] suggested that, in practice, the treatment might be simpler than it appears at face value. In that trial, from four to six therapy strategies were chosen for each of the six families in the trial. And, as discussed earlier, it seems that there are consistent elements in RESTART-DCM treatment that are used for every child who is treated: reduced parent speech rate and interturn speaker latency.

Issues with the underpinning theoretical model

A treatment based on a theoretical model of the nature of stuttering might be questionable if the model itself is questionable. As outlined during Lecture Three, there are grounds to argue that multifactorial models of early stuttering are indeed questionable, and consequently they have received considerable criticism.

It seems fair also to state that these treatments are not straightforward applications of multifactorial models because Palin Parent-Child Interaction Therapy and RESTART-DCM treatment both involve the clinical option of a variant of speech restructuring if needed. Also, the latter treatment involves a speech motor training program, in the event that a child fails an oral motor assessment.

The Westmead Program

Background

An old technique

This treatment is currently in early developmental stages at the Australian Stuttering Research Centre, Sydney, Australia. It uses the well-known rhythm effect, or what is often called syllable-timed speech. As described during Lecture One, this is a fluency inducing condition that seems to have been used to treat stuttering centuries ago. It appears that the earliest documented modern use of this as a stuttering treatment occurred during the 1930s.[52,53] To summarise, when adults who stutter speak while they are saying each syllable to a rhythmic beat, either aided by a metronome or not, they stop stuttering. That is, until they stop speaking rhythmically, at which time stuttering resumes.

Early application to early stuttering

During the early 1980s some researchers looked for clinically useful effects when children spoke in rhythm.[54] The children in this report began speaking during each session with syllable-timed speech at 80–120 beats per minute, saying two-syllable words until they reached a target speech rate, which was from 104–112 beats per minute. Then, during each session, the children spoke in a sequence from three single-syllable phrases, to four-six syllable phrases, then conversational speech. During the last three sessions the rhythmic speech was phased out. The treatment was done solely within the clinic, with three visits per week for 5 weeks. The researchers concluded that the treatment was worthy of further investigation, but no subsequent reports were published.

An intriguing experiment

An experiment[55] involving 9–11 year old boys showed that instructions were not necessary for them to decrease stuttering in the presence of a metronome. The researcher played a metronome in the background with a group of 20 children. Half of them were instructed to talk to the beat of the metronome and the other half received no instruction. Predictably, the children who were instructed to talk rhythmically did not stutter. But surprisingly, the study showed that the children who received no instruction also showed a significant treatment effect. In other words, the children showed a treatment effect from rhythmic stimulation without being instructed to speak that way. That was certainly most suggestive of clinical value for syllable-timed speech with children.

Clinical resource materials

The Westmead Program Treatment Guide[56] is a clinical reference for the treatment which is available at the website of the Australian Stuttering Research Centre. Also at the website of the Australian Stuttering Research Centre[25] there is a clinical severity rating (SR) chart for parents and clinicians (*Child Stuttering Severity Chart eForm*).

The treatment process

Overview

The Westmead Program directs parents to encourage children to use syllable-timed speech during everyday conversations. The aim is to achieve a normal speech rate and speech that does not sound unnatural in any way. For four to six times each day, for 5–10 minute intervals, the parent and child practise syllable-timed speech, and parents occasionally praise their child for using this speech pattern. Parents also prompt their children to occasionally use syllable-timed speech between these practice sessions. There are no set rules for how often these daily therapy activities should happen; the clinician makes a judgement for each child and family.

Parents have a clinic appointment each week

As with all evidence-based early stuttering treatments, parents and children have a clinic appointment each week. During each weekly appointment, the clinician teaches parents how to do the treatment and ensures that it is being done properly.

Treatment goals during Stage 1 and Stage 2

As with the Lidcombe Program, Westmead Program treatment criteria are "no stuttering" or "nearly no stuttering" for a long time. The goal of Stage 1 is to achieve no stuttering or nearly no stuttering, and the goal of Stage 2 is for that to be sustained for a long time. As with the Lidcombe Program, Stage 2 of the treatment is sometimes referred to as maintenance. The treatment criterion measure is the SR scale, which is used with the Lidcombe Program. Stage 1 concludes when these treatment criteria are met:

> To progress to Stage 2, the following criteria need to be met for two consecutive fortnightly consults: (1) clinician SR of 0 or 1 during the consultation, and (2) daily parent typical SRs of 0-1 during the week preceding the consultation, with at least four of those seven SRs being 0. A minimum requirement during Stage 2 is for parents to document SRs during the week preceding the consultation. However, the clinician may request parents to document SRs more often. (p. 6)[56]

Stage 1

Stage 1 of the treatment has two components, Stage 1A and Stage 1B. During Stage 1A, the parent and child attend the clinic for 30–60 minute sessions so they can both learn to do the syllable-timed speech pattern. During this period the parent and child establish a routine where syllable-timed speech is practiced each day. The clinician teaches the parent, where necessary, to modify utterance duration and grammatical complexity to make syllable-timed speech easier to learn. Generally, children learn to do the speech pattern quickly and are able to do it during conversation during the first few sessions. At this time the clinician directs the parent to have the child attempt it during conversations between practice sessions.

Stage 1B begins when the parent and child are practising and using syllable-timed speech during the day correctly. As with the Lidcombe Program, it is critical to be sure that parents are doing what the clinician intends. Fortnightly appointments begin during Stage 1B.

Stage 2

When children attain the treatment criteria, Stage 2 begins, and the family has clinic appointments less frequently during a period of 1 year. During Stage 2, parents are instructed to gradually stop doing the practice sessions each day. In the event that, during a Stage 2 clinic appointment, the child does not meet treatment criteria, the clinician has the option of either stopping progress through Stage 2, while the problem is resolved, or to return the child to Stage 1 to re-establish treatment gains.

Clinical strengths and limitations of the Westmead Program

Strengths

A simple procedure

Of all the treatments discussed, the Westmead Program is the simplest. Speaking with syllable-timed speech seems to be easy for children to learn. So much so, in fact, that as soon as the parent and child learn to do the procedure, clinic appointments begin to occur fortnightly.

It may be useable for immediate early intervention

Rhythmic stimulation is quite a simple procedure, so it may be more useable with younger children than is the case for the Lidcombe Program.

Treatment credibility and expectancy

There is a strong theoretical basis to the Westmead Program, not in the sense of stuttering causality, but in terms of the mechanism that might explain it. Apart from the fact that syllable-timed speech seems to be the oldest stuttering treatment method on record, the P&A Model described during Lecture Three provides a credible explanation for how it might work; syllable-timed speech removes the stress contrasts that trigger stuttering moments.

Limitations

A repetitive and drill-like procedure

This aspect of the treatment could prove to be troublesome as it develops with further clinical trials. Even though parents rapidly learn to do the treatment with their children, it may prove to be quite wearying for them to sustain for long periods in order to obtain durable stuttering control.

Summary

The early years of stuttering are a time when it is at its most tractable and when parents have optimal contact with their children during daily life. Therefore, early stuttering intervention is a desirable clinical option. There are three treatment types for children with early stuttering for which there is clinical trial evidence: the Lidcombe Program, treatments based on Multifactorial Models, and the Westmead Program. The three treatments differ in clinical processes, and each has distinctive strengths and limitations.

Appendix One

Child Stuttering Severity eForm[25]

Child Stuttering Severity Chart

Name:

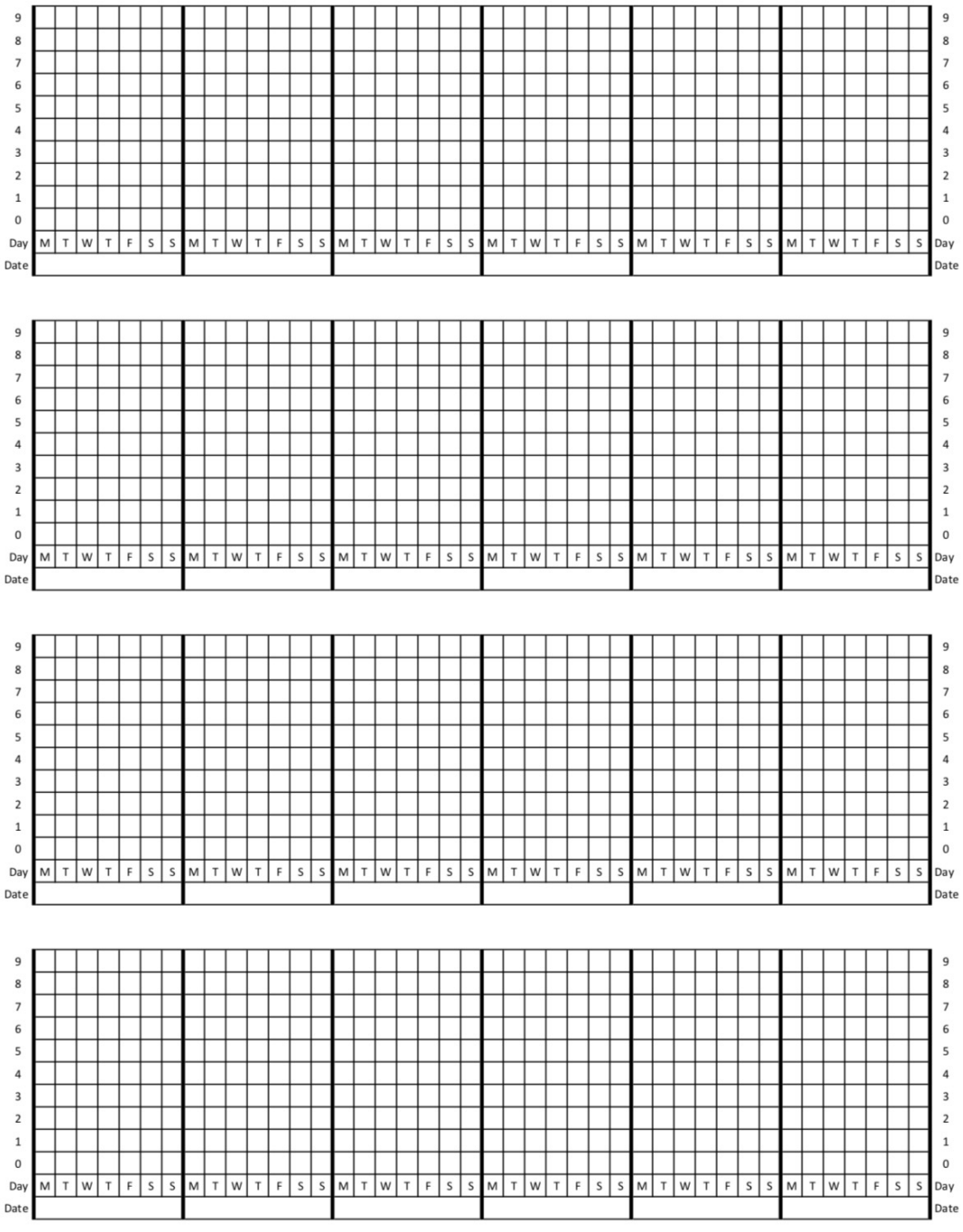
Other Information:

Stuttering Severity (x)

0 = No stuttering

1 = Extremely mild stuttering

9 = Extremely severe stuttering



Appendix Two

Common Lidcombe Program problems[38]

Speech measures	*SRs not collected or collected infrequently.* *Parents unreliable using SRs.*	*14%*[†]
Training parents	*Parents presenting verbal contingencies incorrectly.* *Family members presenting contingencies without training.*	*14%*
Practice sessions and natural conversations	*Treatment during practice sessions used for too long into treatment.* *Treatment during natural conversations introduced too early during treatment.*	*13%*
Verbal contingences for stutter-free speech	*Parents not presenting enough of them.* *Used during practice sessions but not during natural conversations.*	*9%*
Verbal contingences for unambiguous stuttering moments	*Parents using them excessively.* *Parents presenting them in a manner that children don't like.* *Parents presenting them inaccurately.*	*8%*
Low rate of verbal contingencies	*Verbal contingencies given infrequently during practice sessions and conversations.* *No verbal contingencies given at all.*	*8%*
Child has other speech or language problems	*Clinician concurrently many treatment goals for different disorders.*	*8%*
Stage 2	*Entry to Stage 2 without attaining treatment criteria* *Stopping verbal contingencies during Stage 2.*	*5%*
Missing components of Stage 1	*Weekly 45–60 minute clinic appointments.* *Parent training with verbal contingencies.* *Consistent application of treatment.*	*5%*
Stuttering severity fluctuates	*Clinicians unaware this is common.* *Problems measuring treatment progress.* *Clinicians not aware it could be caused by treatment or natural variability.*	*5%*
Stuttering twins being treated	*Clinician uncertainty about treating concurrently or separately.* *Expectations about treatment times.*	*2%*
Sensitive child	*Managing dislike of verbal contingencies.*	*2%*
Parent scepticism	*Managing parent confusion about treatment and doubt about its benefits.*	*2%*
Child unaware of stuttering	*Clinicians uncertain about whether to make child aware of stuttering before treatment.*	*2%*
Problematic parent-child relationship	*Parent focused negatively on stuttering rather than constructive treatment.*	*1%*

[†] Percentage of consultations for which the problem occurred

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LECTURE SEVEN: THE EARLY STUTTERING INTERVENTION EVIDENCE BASE

Clinical trials of one treatment

The Lidcombe Program

In-clinic Phase I-III trials

The Lidcombe Program was developed for the traditional format of weekly clinic visits. For this format, the first Phase I non-randomised clinical trial with Australian children was published in 1990.[1] Subsequently, there has been a Phase I nonrandomised trial with Malaysian children[2] and one with Kuwaiti children.[3] There have been three non-randomised Phase II trials with Australian children,[4,5,6] and one Phase III randomised controlled trial with New Zealand children.[7] One report[8] involved 3–7 years follow-ups of the children treated in that trial. One of the Phase II trials[6] began as a randomised controlled trial, but the researchers could not retain the control group, so it finished up as a Phase II trial with just the children who completed the Lidcombe Program.

The Phase III randomised controlled trial[7] recruited 54 New Zealand children, 12 of them girls, and randomised 29 of the children to a Lidcombe Program arm and 25 to a no-treatment control arm. Two children dropped out of each arm. The primary outcome was percentage syllables stuttered (%SS), measured in three everyday childhood speaking situations at pre-randomisation, and again at 3, 6, and 9 months post-randomisation. At 9 months post-randomisation, %SS for the Lidcombe Program arm was 1.4 and 3.9 for the control arm. This result was statistically and clinically significant.

A Phase III randomised controlled trial[9] compared a standard treatment arm to a group treatment arm involving three families per group. The latter treatment arm involved a "rolling group" model, where a new family entered the group each time a family left the group. Fifty-four children were randomised, and clinical outcomes for the standard and group treatment arms were consistent with outcomes from other clinical trials. However, the children in the group arm required around half the number of clinical hours than the children in the standard arm. Therefore the group Lidcombe Program treatment model was clinically efficient, although the treating clinicians in the trial "found group treatment to be more taxing but clinically gratifying" (p. 1606).[9]

A Phase II randomised trial[10] with three arms compared the traditional weekly visit treatment format with two alternatives: two clinic visits each week and one clinic visit each two weeks (fortnightly). Thirty-one children were randomised to one of the three service delivery models. The conclusions that can be drawn from this study are limited because of its preliminary nature, having low participant numbers and high dropout rates; six, seven and eight children remained in the three treatment arms at 9 months post-randomisation. However, with this in mind, results showed no evidence of any difference for %SS scores at that assessment. Generally, it seemed that two clinic visits each week was not a feasible model for practical reasons. Despite the varying number of clinic visits per week, the median number of weeks to complete Stage 1 was similar for the groups. Of particular interest was the similar number of weeks needed for the groups, with a clinic visit each week and one visit fortnightly: 23 and 24 weeks, respectively. The authors concluded that, considering the health economics of the matter, further clinical trialling of the matter is warranted.

Telehealth Phase I-II trials

Telehealth is considered in detail during Lecture Six: technology used to treat clients when they are not in the clinic. There have been three low-tech telehealth trials of the Lidcombe Program with Australian children using the telephone: two Phase I trials[11,12] and one randomised Phase II trial.[13] A Phase I trial of video telehealth has also been published.[14] A Phase III randomised controlled telehealth trial has been published as well, with an in-clinic and a telehealth arm.[15] The design is known as a parallel, open plan, non-inferiority randomized controlled trial. Results showed no reason to believe that the telehealth Lidcombe Program was less efficacious in terms of stuttering severity outcomes, or cost, than the clinic presentation. In fact, the telehealth arm of the trial had 17% shorter treatment consultations than the clinic arm. There was no reason to believe that parents and children in either arm of the trial had a different relationship with the treating clinicians. For both treatments, there was a general association between stuttering severity and parent satisfaction with fluency.[16] A clinical guide to conducting the Lidcombe Program by telehealth is available.[17]

It is not clear at present where telehealth Lidcombe Program developments will lead. It could turn out that this treatment method will be suitable for the majority of families. On the other hand, this may not be so and the final place for telehealth Lidcombe Program intervention may be as part of a stepped care public health approach to early stuttering, as described during the previous lecture.

The Lidcombe Program in different cultures

A systematic review[18] identified eight data-based studies of the treatment in non-English speaking countries. The languages involved were Arabic (Kuwait), Baluchi and Persian (Iran), Dutch (The Netherlands), Swedish (Sweden), China and Malaysia (Mandarin), Bulgaria (Bulgarian). The review concluded that the treatment is efficacious in different cultures and languages, although it can take longer to complete than in English. The review concluded that it

> … can meet the needs of bilingual children and families and seems to be deliverable in a multilingual context even when the service deliverer and the child do not speak the same language. (p. 12)

The treatment focusses on being a positive experience for children, and, as such, praise and acknowledgment for stutter-free speech is usually a clinically essential parent verbal contingency. However, when formulating a Phase I trial of the Lidcombe Program in Malaysia,[2] the researchers pointed out that the treatment was developed for Western cultures. Based on a study of Malaysian parents and children with early stuttering,[19] they concluded that "praise and acknowledgment of desirable behaviours … appear to be used only infrequently in Malaysian cultures, and that when they occur, may not be varied in expressions" (p. 30).[2]

Four Malaysian children with early stuttering were studied, one of whom was treated in Mandarin Chinese and the others in English. Based on beyond-clinic recordings 12 months after Stage 1, one child had %SS scores of zero, and another child had scores below 1.5 %SS. The third child had scores around 3.0 %SS and the fourth child did not reach Stage 2. The numbers of clinic visits to reach Stage 2 were 21, 31, and 57, which were longer than usual treatment times for the Lidcombe Program (to be reviewed shortly). The researchers reported that this seemed to have been caused by additional time required to teach the parents verbal contingencies, particularly praise for stutter-free speech. The researchers suggested approaches to the cultural issues about praise with the Lidcombe Program, such as variation of tonal and facial expression.

Similar themes emerged during a Phase I trial with Six Kuwaiti children with early stuttering.[3] Four of the children completed Stage 1 and, based on beyond-clinic recordings, were stuttering below 1.0 %SS during Stage 2. The authors reported that praising the children did not come naturally to the parents, and more time was spent training them to use verbal contingencies than is typical for Western parents. Additionally,

> Cultural factors were evident in the current study, such as the inability for women from traditional Bedouin families to drive to sessions on their own and

> relying on their husbands and other family members for transport. Other reasons for missing sessions included religious holidays … (p. 230)

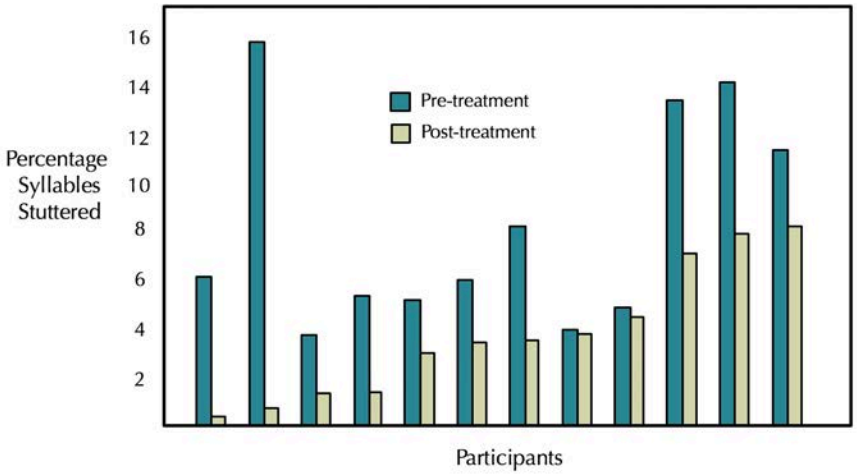
The Lidcombe Program with co-occurring speech sound disorder

A Phase I trial[20] with five boys, ages 3–4 years, involved treatment for stuttering with the Lidcombe Program concurrently with treatment for speech sound disorder. The children were assessed at pre-treatment, at entry to Stage 2 of the Lidcombe Program, and at 9 and 12 months after the start of treatment. The primary stuttering outcome measure was %SS based on two 10-minute conversation samples in everyday situations. Four of the children completed Stage 1 in 14–22 clinic visits, which is consistent with clinical benchmarks (to be discussed shortly). One child did not complete the treatment. Pre-treatment stuttering for the four children was in the range 2–15 %SS, and at 12 months post-treatment they were all below 1.0 %SS. At 12 months post-treatment, all children had shown clinically significant improvement with speech sound disorder to within developmental expectations. The authors concluded that "young children with co-occurring stuttering and speech sound disorder may be treated concurrently using direct treatment approaches" (p. 251).[19]

Palin Parent-Child Interaction Therapy

In-clinic Phase I trials

The developers of this treatment have reported two Phase I clinical trials of it using in-clinic service delivery, with a total of 12 children.[21,22] The first trial[21] recruited nine families, of whom three dropped out, and the latter trial[22] recruited six children who were retained in the trial. The results across the two non-randomised trials are presented in the figure below for the 12 children. For some of the children in the figure, follow-up data are for 6 months post-treatment,[22] and for some the follow-up data are 12 months[21] post-treatment.



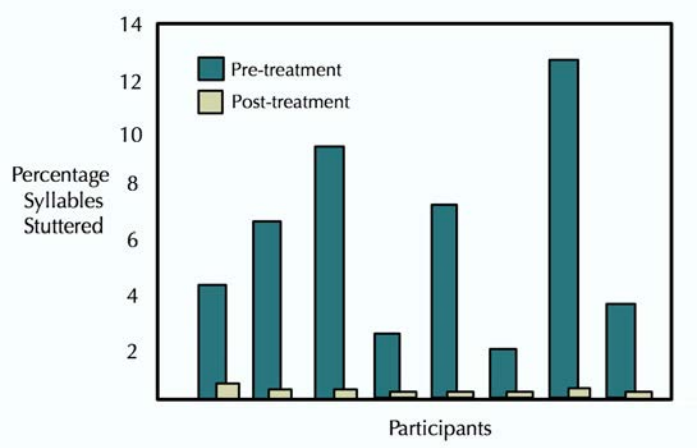
The first four children, on the left of the graph, achieved stuttering reductions to around 1.0 %SS or lower, but this was not the case for the other eight children. Two children showed almost no stuttering reduction. Overall, the pre-treatment to post-treatment reductions of %SS were 64% for the 12 children. Considering that non-randomised designs overestimate effect sizes, this result could arguably be a reflection of natural recovery. These data need to be interpreted keeping in mind that no stuttering or nearly no stuttering is not a goal of Palin Parent-Child Interaction Therapy.

The Westmead Program

In-clinic Phase I-II trials

Subsequent to Phase I trials,[23,24] a Phase II trial[25] recruited 17 children. Only eight of these children completed the treatment. With that caveat in mind, along with the caveat about interpreting non-randomised evidence, the corresponding figure here suggests that the treatment may have some merit.

For the eight children who completed the trial, the mean post-treatment score at entry to Stage 2 was 0.2 %SS.



Clinical trials comparing two or more treatments

Lidcombe Program compared to RESTART-DCM treatment

Method

This is the largest randomised controlled trial reported for any stuttering treatment,[26] randomising 99 children to a Lidcombe Program arm and 100 children to a RESTART-DCM arm. The trial was in The Netherlands, with the treatments presented in Dutch. To be eligible for the trial, children were required to have been stuttering for at least 6 months and to be stuttering more severely than 3.0 %SS. The children randomised to the Lidcombe Program arm had a mean age of 51 months, and children randomised to the RESTART-DCM arm had a mean age of 52 months. Children in the former group were treated with a version of the Lidcombe Program treatment guide available at the time,[27] and children in the latter group were treated with the RESTART-DCM manual mentioned during the previous lecture.[28] Children were followed up for 18 months after the start of treatment. A detailed methodological evaluation of the RESTART trial has been published.[29]

Treating clinicians

Twenty-four clinicians at 20 clinics throughout the Netherlands treated the children. All clinicians received training from the Lidcombe Program Trainers Consortium[30] and "DCM based treatment training is included in the regular clinical education in the Netherlands" (p. 3).[26] Clinicians had a mean of 3.7 years of experience with the Lidcombe Program and a mean of 15 years of experience with the RESTART-DCM treatment. The researchers reported various strategies designed to maximise treatment fidelity, including 3-monthly clinician meetings and clinician treatment logs. The experiences of the treating clinicians are described in a separate publication.[31]

Primary outcome

The primary outcome was "the percentage of nonstuttering children at 18 months, operationalized as ≤1.5% syllables stuttered" (p. 4).[26] This measure was derived from %SS, at 18 months after the start of treatment, measured from three 10–15 minute audio recordings of the children during a period of 2 weeks. Two of these recordings were during conversations with parents and others at home, and one was with a non-family member outside the home. At 18 months after the start of treatment, 28% of the children in the Lidcombe Program arm had not completed Stage 2 of their treatment, and 35% of the children in the RESTART-DCM arm had not attained final treatment targets.[32] There were 21 drop-outs (11%): nine in the RESTART-DCM arm and 12 in the Lidcombe Program arm. It is not stated in the report, but, for the 72 children who completed Stage 1 in the Lidcombe Program arm, the mean number of treatment sessions was 16.4 and the median number was 14.5.[33]

Secondary outcomes

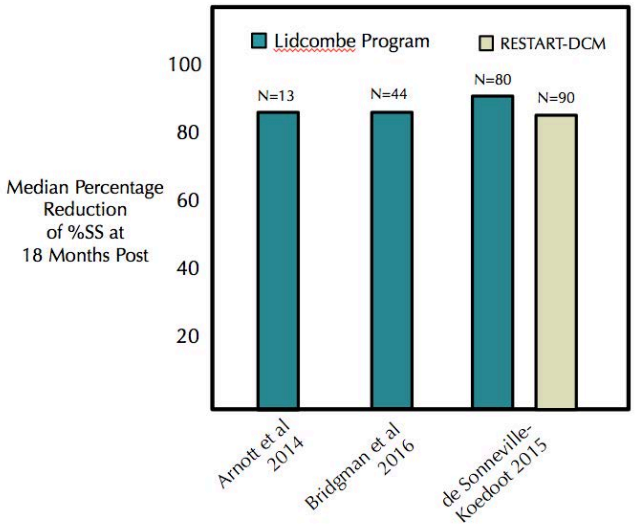
A range of secondary outcomes was reported, including %SS and parent and clinician severity rating with an 8-point scale. These were reported for pre-treatment, and 3, 6, 12, and 18 months after the start of treatment. It is not made fully clear in the report, but the %SS measures at pre-treatment, and 3, 6, and 12 months after the start of treatment were based on the same methods as the %SS measures at 18 months after the start of treatment: three audio recordings of the children beyond the clinic.[34] Additional secondary outcomes were a health-related quality of life measure (EQ-5D),[35] a measure of child attitude to communication (KiddyCat)[36] (see Lectures Ten and Eleven), and three measures of child emotional and behavioural problems derived from the Child Behavior Checklist.[37]

Primary outcome results

At 18 months after the start of treatment, 86 children remained in the Lidcombe Program arm, and 91 remained in the RESTART-DCM arm. There were 76.5% of "non-stuttering" children in the Lidcombe Program group and 71.4% of "non-stuttering" children in the RESTART-DCM group. These differences were not significant. Results remained nonsignificant when the cut-off %SS scores for "non-stuttering" were changed to 1.0 %SS and 2.0 %SS.

In terms of the 95% confidence intervals that were reported (see Lecture Five), it is appropriate to interpret these results as showing no evidence of a difference between the treatments, rather than evidence that the treatments are equivalent. For %SS scores 18 months after the start of treatment, the mean difference between treatments was 0.3 %SS, with a 95% confidence interval for the difference of -0.4–0.9 %SS. That confidence interval contains zero (see Lecture Five). Additionally, for the percentage of "non-stuttering" children at 18 months after the start of treatment, the 95% confidence intervals were 66–84% for the Lidcombe Program and 61–80% for RESTART-DCM. Arguably, the range of these confidence intervals includes differences that are clinically significant.

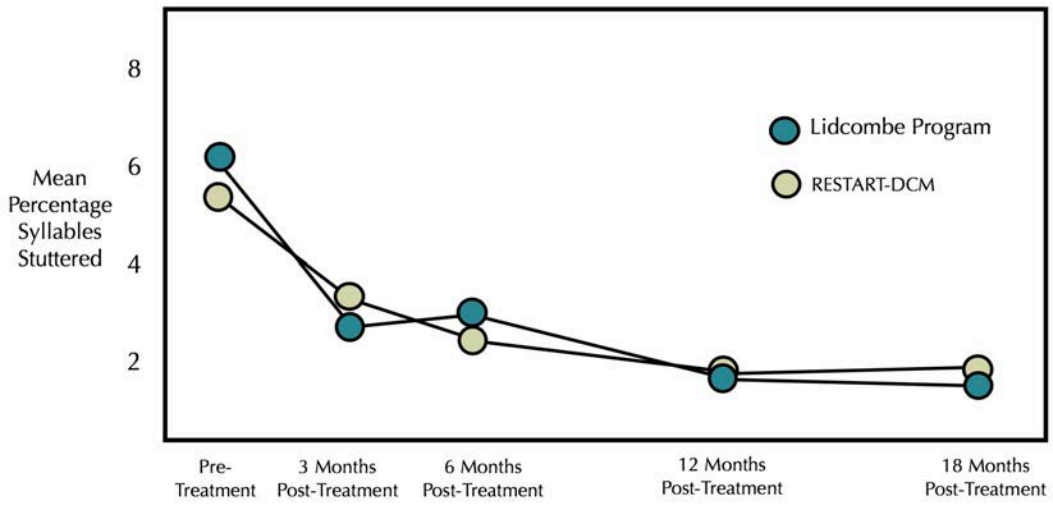
It would be useful to have some way of comparing the outcomes of the RESTART trial with the outcomes of the dedicated Lidcombe Program clinical trials discussed earlier. Any such comparison needs to be guarded because the trials concerned were conducted in different countries, at different times, and with different research protocols. There is also the problem of possible—even likely—differences of %SS scores by clinicians in different countries, as discussed during Lecture Four. Arguably, a measure of reduction of %SS scores from pre-treatment to 18 months post-treatment would go some way to offsetting any such reliability problems. So, the figure presents median percentage reductions for the RESTART trial and the data from the two standard treatment arms from Lidcombe Program randomised trials at 18 months post-randomisation.[9,15] With the caveat that such a comparison needs to be guarded, the data in the figure suggest no evidence that outcomes in terms of stuttering reductions are different across the three trials.



Secondary outcome results

The paper reported that "most outcome measures were slightly in favour of the direct approach (LP), but the few significant interaction terms were deemed negligible due to their small effect sizes" (p. 11).[26] For the entire 18-month pre-treatment to post-treatment period there was a statistically significant effect favouring the Lidcombe Program for %SS and parent severity rating (see Table 2, p.

8–10), but with small effect sizes. In other words, reported effects were statistically but not clinically significant (see Lecture Five). There were no significant changes pre-to post-treatment for the quality of life measures. With the Lidcombe Program group, there were significant post-treatment improvements for the three measures of emotional and behavioural problems, but these were attributable to a pre-treatment difference between the groups. For the attitude to communication scores, there was an almost statistically significant (unadjusted p=.06) post-treatment improvement for both groups. The figure[‡] presents the %SS measures for the trial at pre-treatment, and 3, 6, 12, and 18 months after the start of treatment.



An economic evaluation of the two treatments

An economic evaluation of the RESTART clinical trial[38] reported that, at 18 months after the start of treatment, health outcomes were slightly better for the Lidcombe Program than for the RESTART-DCM treatment. One measure attained statistical significance with a small effect size (Cohen's d=0.17): quality adjusted life years. The authors concluded that "cost-effectiveness and cost-utility ratios were in favour of the LP. The LP is considered a good alternative to RESTART-DCM treatment in Dutch primary care" (p. 106).[38]

A critique

The RESTART trial attracted a negative critique[39] pointing out that it was without a control group that received no treatment, hence that it presented "no value for clinical management because the treatments investigated were not shown to be more effective than no treatment" (p. 65). The treatment was also criticised because of the paradoxical criterion of ≤1.5 %SS as non-stuttering.[†] The authors responded[40] by conceding that the RESTART trial could not determine whether either treatment was better than natural recovery, but pointed out that the goal of the study was not to do that. In relation to

[‡] Adapted and reproduced with permission: De Sonneville-Koedoot, C., Stolk, E., Rietveld, T., & Franken, M-C. (2015). Direct versus indirect treatment for preschool children who stutter: The RESTART randomized trial. *PLoS One, 10,* e0133758. © 2015 de Sonneville-Koedoot et al..

[†] That criticism about the primary outcome seems reasonable. For example, at a childhood speech rate of 200 syllables per minute, ≤1.5 %SS represents up to 180 stuttering events for every hour of speech. Clearly, that cannot be described as "non-stuttering." It is arguable that a simpler and more interpretable primary outcome for the trial would have been the gold standard of post-treatment %SS scores compared across treatments groups, as is routinely used in clinical trials.

the issue of the criterion for non-stuttering, the authors reiterated that the results of the trial were identical when different criteria of ≤1.0 and ≤2.0 %SS for non-stuttering were applied.

Lidcombe Program compared with two Westmead Program versions

Background

A clinical trial[41] was conducted prompted, by the limitations of the Lidcombe Program that were discussed during Lecture Six, and prompted by the potential advantages to the Westmead Program as outlined in that lecture. Additionally, the authors argued that the Westmead Program is potentially useful because it does not require the dedicated practice sessions required with the Lidcombe Program, and that non-randomised clinical trial data suggest that it may require fewer treatment hours. Two versions of the Westmead Program were devised for the trial, one incorporating the verbal contingencies of the Lidcombe Program. The rational for this was a high dropout rate in a previous trial and a suggestion "that families tended to withdraw from treatment at the point when low-level stuttering severity had been attained but not stabilized" (p. 507).[41] The report raised the idea that "for such cases, the final stages of clinical progress need to be hastened with the addition of contingencies for stuttered and stutter-free speech" (p. 507).

Method

The trial was a three-armed randomised controlled trial with the Lidcombe Program as the control group and the two Westmead Program versions as the experimental groups, one with and one without verbal contingencies. There were blinded outcome assessments at 9 months post-randomisation. There were 91 children recruited, 33 to the Lidcombe Program arm and 28 and 30 to the two Westmead Program arms. This was the first clinical trial of either treatment without a lower age restriction; children of any age who were stuttering were eligible to be participants.

Treating clinicians

The treatments were conducted in Melbourne and Sydney, Australia, in two community and two university research clinics. All clinicians had received Lidcombe Program training from the Lidcombe Program Trainers Consortium and received Westmead Program training from its developers.

Primary and secondary outcomes

The primary outcome was %SS measured at 9 months post-randomisation from two 10-minute recordings of the children. One of those involved the children speaking to a non-family member outside the home, and the other with an adult family member at home. The secondary outcome was the number of clinic visits required to complete Stage 1.

Results

No evidence was found of any difference in %SS scores between the groups at 9 months post-randomisation. There was evidence to support earlier trials that treatment times were shorter for the Westmead Program, with a median of 30 clinic visits to complete Stage 1 of the Lidcombe Program and only 18 and 16 visits for the Westmead groups.

Limitations

A major limitation of this study is that, although the authors attempted to fix the Westmead drop-out problem with an adapted Westmead Program, they failed to do so. The drop-out rates for both Westmead arms were 43%, and there was also a substantial Lidcombe Program drop-out rate of 27%. This weakens the confidence that can be placed in the results, even though a statistical technique called multiple imputation was used to adjust for the problem.

The authors argued that some novel aspects of the trial may have accounted for dropouts. There were no speech and language exclusion criteria, and 13 of the children were younger than 3 years, which has never occurred previously in a clinical trial of early stuttering intervention. It was also the first trial of either treatment involving community clinics. Regardless, the authors concluded that "parents and childen may simply find the treatment boring" (p. 13).[41] The authors also stated that they are

attempting to deal with this problem by developing a standalone internet version of the Westmead Program.

Clinical translation

The Lidcombe Program

One translational study (see Lecture Five),[42] an in-clinic Phase IV trial, has explored whether the results of Lidcombe Program clinical trials can be achieved in clinical communities. The study involved 31 Australian community clinicians who treated 57 children with early stuttering. Outcome measures were %SS during everyday childhood conversations at 9 months after the start of treatment. Statistical regression modelling was used to determine whether any variables could predict that outcome: (1) pre-treatment stuttering severity, (2) speech or language disorders in addition to stuttering, (3) whether the clinicians had received training from the Lidcombe Program Trainers Consortium,[30] (4) the duration of weekly clinic visits, and (5) the mean period between clinic visits. At 9 months post-treatment, 12 children (21%) had withdrawn from their treatment, 47 (65%) had competed Stage 1, and eight (14%) were still in Stage 1.

The mean 9 months post-treatment stuttering severity for all the children was 1.7 %SS. However, Consortium training was a significant predictor of outcome. Children treated by Consortium trained clinicians attained a mean of 1.1 %SS at 9 months post-treatment and those treated by clinicians without such training scored a mean of 2.4 %SS, which is more than double. No other predictors of outcome were found. The authors concluded that, for clinicians with Consortium training, Lidcombe Program community outcomes are able to match those attained in clinical trials.

A study involving six community clinicians[43] evaluated the cost-effective rolling-group Lidcombe Program model used in a previous randomised trial.[9] Participants were 19 children with early stuttering, mean age 49.1 months, treated by six generalist clinicians in four Australian rural towns. Within clinic measures of %SS were collected at pre-treatment and at 6 and 9 months after the start of treatment. Percentage syllables stuttered scores were 7.4 at pre-treatment and 1.4 and 1.3 at the 6 and 9 months post assessments, respectively. Those results were obtained in benchmark treatment times for the Lidcombe Program (to be discussed shortly). The report was supplemented with a resource involving perspectives from participating clinicians about the rolling-group treatment model.[44]

Randomised clinical experiments

According to the operational definition of a clinical trial presented during Lecture Five, a clinical trial involves evaluation of an entire treatment. The rationale given for such a criterion was that clinicians need information about the efficacy of an entire treatment in order to determine whether they might wish to use it. However, several reports have been published which have all the features of a randomised controlled trial according to this definition, with the exception that they are evaluations of parts of a treatment. These reports might be termed randomised clinical experiments.

The Lidcombe Program

Sixteen weeks of Lidcombe Program compared to no treatment

An experiment[45] with German children is sometimes cited as an independent replication of the Phase III Lidcombe Program trial,[7] and its title states that it is a clinical trial. However, the report involved only 16 weeks of treatment. Forty-six pre-school children, four of them girls, were randomised to receive either 16 weeks of the Lidcombe Program treatment or 16 weeks of no treatment. One child dropped out of the treatment group. After 16 weeks of treatment, the children in the Lidcombe Program had 1.6 %SS in everyday speaking situations and the control children 6.9 %SS. This result was statistically and clinically significant.

Twelve weeks of Lidcombe Program compared to no treatment

With a similar design to the German study, an Australian experiment[46] randomised 29 pre-school children, four of them girls, to receive either 12 weeks of the Lidcombe Program treatment or 12 weeks of no treatment. Six children dropped out, leaving 10 in the treatment group and 13 in the control group. At 12 weeks post-randomisation, the children in the Lidcombe Program group had a mean 3.5 %SS during everyday conversations and the children in the control group had a mean 5.8 %SS. This result was statistically and clinically significant.

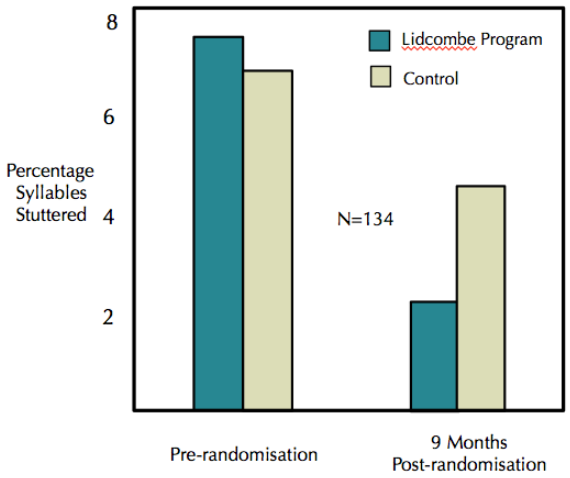
Twelve weeks of Lidcombe Program compared to RESTART-DCM

As a preliminary study to the randomised trial discussed earlier,[26] a Dutch study[47] randomised pre-school children to a Lidcombe Program group and a RESTART-DCM group. Thirty children were randomised. Seven dropped out, leaving 11 children in the Lidcombe Program group and 12 in the RESTART-DCM group. Based on beyond-clinic recordings, results after 12 weeks of treatment were 3.7 %SS for the Lidcombe Program group and 3.1 %SS for RESTART-DCM. This result was clinically and statistically nonsignificant.

Interpreting their findings cautiously, and foreshadowing their later randomised trial, the authors concluded that "randomized controlled trials of LP versus DCM treatments are feasible" (p. 197).[47] They also correctly pointed out that further study of the matter is necessary with control groups in order to obtain fully interpretable results.

"Meta-analysis" of the Lidcombe Program

The ultimate endpoint of clinical trials research is a systematic review of meta-analysis for many randomised controlled trials (see Lecture Five). For clinical trials of stuttering, that is not yet possible. However, the next best thing is an analysis of randomised clinical evidence for the Lidcombe Program that involves a no-treatment control group. That randomised clinical evidence includes randomised controlled trials,[7,13] and two randomised clinical experiments.[45,46] The mean post-randomisation period for those reports is 6.3 months.



The results of this analysis[48] are shown in the accompanying figure. In total, it involved 134 children. At pre-randomisation the stuttering severity of the Lidcombe Program and control groups were about the same. There was some predictable improvement with the control children because of natural recovery. However, at a mean post-randomisation period of 6.3 months, the Lidcombe Program children did better than the control children.

The Lidcombe Program odds ratio[†] was 7.5 for attaining below 1.0 %SS at 6.3 months post-randomisation. That means that, at 6.3 months post-randomisation, children who received the Lidcombe Program had 7.5 times greater odds of having "no stuttering" or "almost no stuttering" than children who did not receive the Lidcombe Program treatment. The 95% confidence interval was 2.7–20.9, meaning that there was an estimated 95% chance of the true odds ratio being between those two values.

[†] See Lecture Five for discussion of odds ratios.

It is necessary when interpreting this meta-analysis result to keep in mind that, with a mean post-randomisation period of 6.3 months, not all of the 134 children involved received the full treatment. So the odds ratio for those children may have been greater had they received the full treatment. Therefore, it would be justifiable to conclude that the odds ratio is at least 7.5.

Data-based case studies

For the present purposes, data-based case studies are reports published in peer-reviewed journals that are either retrospective—involving previously treated children—or reports that do not involve speech measures beyond the clinic, or which do not incorporate a clinically meaningful follow-up period. However, their conclusions about treatment outcome focus on speech measurement.

The Lidcombe Program

Ten Swedish children were enrolled in a case study report of the Lidcombe Program.[49] Six of them completed the treatment and reduced stuttering, according to measures in the clinic, from a pre-treatment mean of 6.7 %SS to a post-treatment mean of 0.1 %SS at the end of Stage 2. The post-treatment assessment was "21 months or more after achieving fluency" (p. 251),[49] which presumably means 21 months after completing Stage 1. Several other case studies have been reported for the Lidcombe Program with British,[50] Canadian,[51] and United States children.[52] The latter was a data-based follow up of 15 children 1–5 years after treatment. Based on video recordings of the children speaking in their homes, mean pre-treatment scores were 12.6 %SS and mean post-treatment scores were 0.5 %SS, which was a 96% reduction. Several individual case studies of the Lidcombe Program have been reported with French Canadian children with early stuttering.[53,54,55] Those cases were consistent with a report of Malaysian children treated with the Lidcombe Program[19] that treatment in one language generalises to another language. A report with a Belgian 3 year 4 month old child[56] showed treatment success by telehealth.

Palin Parent-Child Interaction Therapy

A retrospective file audit[57] of 55 children treated at the Michael Palin Centre in London involved 38 boys and 17 girls with a mean age 53 months at the start of the study. Percentage syllables stuttered was measured before treatment and 3, 6, and 12 months after the start of treatment. Measures were based on clinic video recordings "while the child described a series of "What's wrong?" pictures with the SLPs *[speech-language pathologists]* in the clinic" (p. 1214).[57] The duration of the recordings was not specified. Additionally, children were measured with the KiddyCAT, which is a parent report measure of child attitude to communication (see Lectures Ten and Eleven).

From before treatment and 12 months after the start of treatment, %SS scores were, respectively, 6.7 and 2.3, which was a 66% reduction. This is consistent with the 64% reported for the 12 children in two clinical trials of the procedure,[21,22] discussed earlier. KiddyCAT scores reduced from 4.6 to 2.0, showing an improvement of the children's attitude to communication.

A family-focussed treatment approach based on a multifactorial model

There has been a case study report[58] of a treatment that is broadly similar to the treatments discussed previously based on multifactorial models: Palin Parent-Child Interaction Therapy and RESTART-DCM Treatment. This treatment, developed at the Stuttering Center of Western Pennsylvania at the University of Pittsburgh, in part draws specifically on the Demands and Capacities Model. A treatment manual is available.[59]

Similar to the Palin Parent-Child Interaction Therapy treatment process, this family-focussed treatment approach "typically consists of six to eight sessions, 45 min in length, scheduled once per week or every other week" (p. 120).[58] It is also consistent with Palin Parent-Child Interaction Therapy that the treatment goal does not overtly specify no stuttering or nearly no stuttering as a treatment goal. Instead, the treatment

> is designed to help young children who stutter (between the ages of 2 and 6) improve their speech fluency while simultaneously ensuring the development of healthy communication attitudes and effective communication skills. (p. 119)[58]

Also consistent with Palin Parent-Child Interaction Therapy, and also RESTART-DCM Treatment, is that "direct fluency shaping and stuttering modification" (p. 119)[58] procedures are implemented if needed. Another similarity is that the clinician works with parents to construct individual treatment plans according to need. The multifactorial "bucket analogy" (see the diagram in Lecture Three) is presented to parents during this process:

> For example, if parents report that their schedule at home is busy and that they often feel rushed, and if they believe that this contributes to time pressures that affect the child's speech, then the parents and clinician may brainstorm ways of reducing these time pressures. The parents may then work to set aside a set period of time each day so the child can interact with the parents with less time pressure, or they may consider different scheduling options for the child's activities in order to allow for more one-on-one time. (p. 121)[58]

The specific "parent communication modifications" considered are as follows:

> (a) use and modeling of an easier, more relaxed manner of speaking … (b) use of increased pause time between speaker turns so as to reduce time pressures the child may feel when communicating; (c) reduction of demands to speak and increased time pressures often associated with "rapid-fire" questioning, if present; and (d) reflecting, rephrasing, and expanding on children's utterances to provide a positive communication model. (p. 123)[58]

The results of the report were fairly consistent with the two clinical trials of Palin Parent-Child Interaction Therapy,[21,22] indicating that six of 17 children "continued to stutter following completion of the parent-focused treatment" (p. 128).[58] Mean scores for "stuttered types of disfluencies per 100 words" (p. 126) were 16.4 before and 3.2 after the intervention. Parents scored "how often the child was able to speak *without* (authors' italics) stuttering" (p. 126–127) with a 5-point scale: 5 = *always*, 4 = *almost always*, 3 = *sometimes*, 2 = *rarely*, and 1 = *never*. The post-treatment mean for three beyond-clinic speaking situations was around 2.8.

A case study of several treatments

A report with six Iranian children with early stuttering[60] involved the Lidcombe Program, Palin Parent-Child Interaction Therapy, and a hybrid treatment that combined the two treatments. For several reasons, this report does not provide useful information for clinicians. No reason was given for combining the two treatments. Also, there are transparent problems in the report with treatment fidelity. For example, components of the Lidcombe Program are described which do not exist, such as "following children's model in play," "reducing parent's speech speed," and "observing turn taking in families" (p. 33).[60] Finally, all children received a 12-week combination of all three treatments, making the results uninterpretable.

Therapist drift

The Lidcombe Program

Therapist drift

Departure from manualised procedures, or *therapist drift*[61] to use the correct term, is thought to be clinically undesirable. It may be a justifiable assumption that adherence to the Lidcombe Program Treatment Guide will produce optimal treatment results. However, currently there is little empirical support for that contention, but there are three reports that therapist drift with the Lidcombe Program does occur. These reports are now discussed.

Therapist drift in a translational study

During the translational study outlined previously,[42] the 31 community clinicians generally adhered to the Lidcombe Program Treatment Guide. However, around half of them varied from the prescribed 45–60 minute treatment sessions and used 30-minute sessions instead. Also, fortnightly clinic visits often occurred instead of the prescribed weekly visits, sometimes because of clinician scheduling and sometimes because of client failures to attend their clinic sessions. The mean number of days between clinic visits was 15.4, rather than the ideal seven days specified in the treatment guide. However, there was no evidence that these fidelity problems affected outcomes. The result that fortnightly clinic visits did not affect outcomes was consistent with another file audit of 134 North American pre-schoolers treated with the Lidcombe Program.[62]

The translational study also showed that around half of the 31 clinicians did not use the recommended procedure of having parents demonstrate verbal contingencies in the clinic each week. It was not possible to determine statistically whether this treatment fidelity problem affected outcome. Regardless, the authors concluded:

> The fact that only half the SLPs *[speech-language pathologists]* in the present study routinely requested the parent to demonstrate treatment in the clinic, however, is concerning. Without observing parents demonstrating treatment, it is not possible for SLPs *[speech-language pathologists]* to confirm that parents are implementing the treatment safely and correctly, and that the child is responding positively. However, the study design did not allow exploration about why SLPs *[speech-language pathologists]* in the present study largely chose not to adhere to this important treatment procedure. (p. 601–602)[42]

Therapist drift in two observation studies

This important treatment fidelity issue—that parents may not be doing verbal contingencies the way the clinician has instructed them during the Lidcombe Program—was explored with three children with early stuttering during their treatment.[63] It was encouraging that the three parents presented more verbal contingencies for stutter-free speech than stuttered speech during practice sessions, as specified in the treatment guide. This was also the case with verbal contingencies during natural conversations, although more contingencies for stuttering occurred in such situations.

The report had some sobering features, however. One parent continued with the treatment during practice sessions despite the child saying he did not like the activity. Another parent focused on the rules of the game being played rather than the child's speech during such treatment. All parents were observed to give incorrect verbal contingencies, such as praising stuttered speech, during treatment in practice sessions.

To assist clinicians with ensuring Lidcombe Program treatment fidelity, the report concerned[63] presents an empirically developed checklist of procedures for treatment during practice sessions, which focusses on the important issues of parent verbal contingencies. The paper contains two case histories of the checklist being used.

A larger study[64] involved 40 parent-child pairs during Stage 1 of the Lidcombe Program. The parents recorded the practice sessions they did each day, and kept a diary of their use of verbal contingencies during natural conversations. There were some positive results. The mean duration of practice sessions was 12.7 minutes, and the median number of practice session per day was once per day. Those findings were consistent with the Lidcombe Program Treatment Guide. Most verbal contingencies during practice sessions were for stutter-free speech, as specified in the Treatment Guide; 91% were for stutter-free speech and 6.8% were for unambiguous stuttering, with only 2.7% of verbal contingencies being incorrectly applied.

However, according to parent diaries, the number of verbal contingencies during natural conversations was lower than expected: an average of 8.5 contingences per day for stutter-free speech and 1.7 contingencies per day for stuttered speech. Also, an unexpected and puzzling association was found between the number of verbal contingences for stuttering during natural conversations and the

number of clinic visits to complete Stage 1. It was expected that more verbal contingencies would be associated with fewer clinic visits, but the opposite trend was reported.

Therapist drift in a survey study

A survey of 277 Australian speech pathologists[65] reported that around half of them said they departed from the procedures specified in the Lidcombe Program Treatment Guide. A common reason given for such departures was that the Australian public health providers sometimes do not allow the full treatment to be given, allocating only treatment "blocks" of time to any one child, with the blocks of time not long enough for the treatment. (This is reminiscent of how British health care managers allocated treatment with Palin Parent-Child Interaction Therapy, as discussed during the previous lecture.)

Other problems documented in the survey were workplace service restrictions, including one report of children waiting up to 12 months for treatment. Other identified problems included allocating the requisite time in school settings that provided treatment services. For the study sample, 23% of clinicians were located in schools. This is a particular problem in the United States where a public law states that all children who are disadvantaged because of disability must receive prompt remediation.[66] Consequently, United States clinicians with many children on their caseloads are by law not permitted to have a waiting list; all affected children must be treated promptly. In many such cases, children would not be able to receive a complete Lidcombe Program treatment as specified in the treatment guide.

Mechanisms of Action

Multifactorial treatments and the Westmead Program

As discussed during the previous lecture, treatments based on multifactorial models and the Westmead Program have transparent, putative underlying "mechanisms of action"[67] for any treatment effects. Although there are no data at present to substantiate such notions, the former treatments might operate because multifactorial models are correct, and the Westmead Program might operate because of the acoustic effects of syllable-timed speech.

The Lidcombe Program

However, there is no obvious mechanism of action to explain how the treatment effects of the Lidcombe Program might occur. Several studies have attempted to find such a mechanism, and these are now discussed.

Child and parent language

One suggestion[68] has been that children, or parents, might simplify their language production after the treatment. An initial report[68] with nine parent-child pairs found that to not be the case, with a range of parent and child language measures not changing from pre-treatment to post-treatment. Measures included speech rate, inter-speaker turn latency, mean length of utterance, developmental sentence scoring, number of different words, requests for clarification and requests for information. No differences were found for the pre-treatment to post-treatment period. In fact, maternal speech rate increased after the treatment and parents decreased their rate of questioning. There was a slight suggestion, however, that the children did not meet full developmental language expectancies during the pre-treatment to post-treatment period.

Subsequently, the result of no pre-treatment to post-treatment language change was replicated with four children.[51] At post-treatment, the children increased their mean length of utterance, percentage of complex sentences, and number of different words. This result was replicated with another eight children,[69] showing no change of mean length of utterance, type-token ratio, and a phonological measure of percentage consonants correct.

A longer term study[70] was conducted on 11 children with early stuttering prior to Lidcombe Program treatment, at 9 months post-treatment and 18 months post-treatment. Measures were mean length of

utterance, number of different words, and subordination index. During the period of study, there was no evidence associating Lidcombe Program treatment with restricted language development. The children showed no differences from the developmental trajectories of normative data for the three language measures.

Acoustics

To date, there has been one attempt to find an acoustic explanation for the apparent efficacy of the Lidcombe Program;[69] perhaps children use a slightly different speech pattern post-treatment that controls stuttering. However, this report found no changes for vowel duration, intervocalic interval, voice onset time, or articulation rate.

The contribution of verbal contingencies

The Lidcombe Program is based on laboratory studies showing that stuttering has operant-like properties because it responds to contingent stimulation (see Lecture One). The construction of the treatment around five parent verbal contingencies carries the assumption that those contingencies are essential to the reported Lidcombe Program treatment effects.

An experiment[71] was designed to explore this assumption by randomising 34 parent-child pairs to two groups. The first group received the standard Lidcombe Program, and the second group received the Lidcombe Program without the verbal contingency *request self-correction*. The researchers measured the number of weeks and the number of clinic visits for the children to attain a 50% reduction of stuttering severity. They reported no significant differences between the groups. This result challenged the contribution of the verbal contingency *request self-correction* to the efficacy of the treatment, and it suggested the need for further research about the matter. This study supplements the study of parent treatment fidelity with verbal contingencies discussed earlier.[64]

The issue was further explored with a randomised controlled noninferiority trial.[72] The control arm was standard Lidcombe Program treatment, and in the experimental arm all the verbal contingencies were removed. Participants were 74 children aged between 3 years 0 months and 5 years 10 months, 37 of whom were randomised to each group. At 18 months follow-up, 31 children remained in the control group and 26 remained in the experimental group. The authors concluded that:

> Findings of noninferiority were inconclusive for the primary outcome of stuttering severity, based on a margin of 1.0 percentage syllables stuttered. ... The inconclusive finding of noninferiority means it is possible that verbal contingencies make some contribution to the Lidcombe Program treatment effect. (p. 3419).

Together, the two studies raise an issue in need of resolution: how do verbal contingencies contribute to Lidcombe Program treatment effects?

Speculation about cortical plasticity

It is reasonable speculation, regardless, that the treatment somehow rectifies problems with neural processing that are associated with stuttering (see Lecture Three). Perhaps an efficacious treatment such as the Lidcombe Program induces children to "adopt a compensatory neural growth pattern that successfully makes up for the deficient brain regions" (p. 77).[73] Another author has suggested a similar mechanism in terms of the malleable nature of the developing brain.[74] A report of dyslexic school-age children was consistent with this possibility, showing changes of grey matter volume after 8 weeks of therapy.[75]

This idea incorporates the well-known notion of "cortical plasticity." Not only does the brain drive behaviour, but behaviour drives the brain. A review of the topic[76] presents converging lines of evidence that "suggest an active role for dynamic myelination in adult brain plasticity and indicate myelin plasticity may be an additional route by which experience can shape brain structure and function" (p. 86). For example, learning to juggle can change grey matter structure in areas that support visual learning.[77] Another study showed changes in the occipito-temporal cortex after only seven days of juggling learning.[78]

If such mechanisms of action are involved with the Lidcombe Program, they could be verified experimentally, such as with brain scanning of experimental and control children before, during, and after a treatment period.

Treatments based on multifactorial models

A review of the topic[79] and a subsequent empirical investigation[80] found no reason to implicate unusual parent language behaviour with early stuttering, or any reason to believe that changing parent language behaviour may be clinically useful with early stuttering.

As noted in the previous lecture, during RESTART-DCM treatment all parents reduce speech rate and increase their interturn speaker latency. With Palin Parent-Child Interaction Therapy those changes seem to occur often during treatment. Therefore, experimental evidence to verify the capacity of those variables to control early stuttering is of interest. A recent review[81] of five laboratory experiments of parent reduced speech rate for children with early stuttering[82,83,84,85,86] concluded that stuttering reductions of around 50% were observed overall under such conditions. However, effects were not observed for every child studied. One of those experimental reports[83] concluded that study of the effects of extended, everyday parent use of such techniques is warranted.

An experimental study of a 5 year 9 month old child who stuttered[87] involved increased interturn speaker latency for 15 sessions during a 7-week period at the family dinner table. The parents and the child's 10-year-old brother participated, with the children using wooden blocks to signal the need to have a conversational turn. Results suggested that the procedure was responsible for a 40–50% reduction of stuttering during the experimental conditions compared to baseline. A laboratory experiment[88] with three boys who stuttered involved three experimental sessions of "no interruption" for two of them during conversation with a researcher. For one boy, aged 6 years 2 months, a stuttering reduction of around 50% was observed. No effect was observed for the other boy, age 5 years 6 months. A study of 27 children with early stuttering,[89] with a mean age of 4 years 0 months, showed that parents could be taught to slow their speech rate and increase interturn speaker latency. In the clinic, "stuttering-like disfluencies" (see Lecture Four) of the children decreased by 36%.

Treatment safety

The Lidcombe Program

As noted in the previous lecture, a potential limitation of the Lidcombe Program is that it is possible for a parent to misuse the treatment and give verbal contingencies in a punitive and excessive manner. Indeed, during the early development of the Lidcombe Program, concerns were raised that the treatment might send an overall negative message to children that would affect their self esteem and establish unhelpful cognitions.[90,91,92]

In response to those concerns, the Lidcombe Program developers verified for eight children with early stuttering that the treatment is psychologically safe.[93] Measures with the Child Behavior Checklist[94] showed no behavioural indications of any changes with the children pre-treatment to post-treatment that might suggest anxiety, aggression, withdrawal, or depression being associated with the treatment. Additionally, the Attachment Q-Set, which measures the strength of bond between parent and child, showed that there were no changes after treatment. In fact, if anything, attachment appeared to improve. These results were confirmed by the randomised trial discussed earlier,[26] which reported some suggestion of post-treatment improvement for Child Behavior Checklist and KiddyCat scores after Lidcombe Program treatment.

Naturally, though, findings about the safety of the Lidcombe Program pertain to clinically appropriate management of any threats to safety. If there is any chance that a parent will use verbal contingencies in a punitive manner, the clinician needs to deal with that situation without delay.

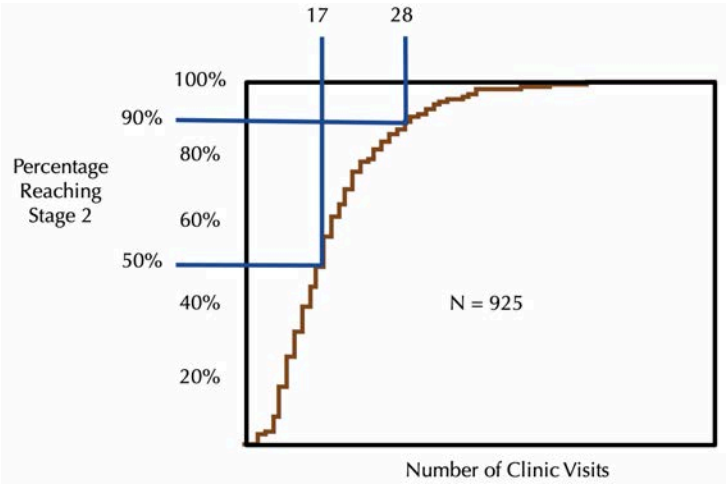
Treatments based on multifactorial models†

As with the Lidcombe Program, there is a basic issue about the safety of these treatments that needs to be dealt with. Treatments based on multifactorial models change features of everyday childhood life that appear essential to healthy development. Active participation in conversation[95,96] and sustained interaction with adults[97,98] is known to be fundamental to early linguistic development. A review of three decades of literature[99] presented four critical aspects of healthy oral language childhood development: "family dynamics, … interaction with parents, immediate social environment, and encouragement given to the child in the first years of life" (p. 350). As yet, there has been no research directed at the effects of changing these features of early childhood life during treatments based on multifactorial models, but obviously it is required.

How long does treatment take?*

The Lidcombe Program

There is a sufficiently comprehensive data set that gives an indication of how many Stage 1 clinic appointments may be required with the Lidcombe Program; in other words, how many clinic appointments are required to attain "no stuttering" or "nearly no stuttering." More than a thousand children have been participants in Lidcombe Program clinical research, and the following figure† contains information about treatment time for children, based on six file audits,[52,62,100,101,102,103] seven clinical trials,[4,9,10,15,26,41,72] one prospective follow-up,[52] one translational study,[42] and one prospective observation study.[64] Each of those studies, which involved a total of 925 children, reported a median number of clinic appointments to attain Stage 2 criteria. The average reported median clinic visits across those studies was 17.# The range of median clinic appointments reported in those studies to attain Stage 2 is 11–30.



† Thanks to Juliet Imeson for guidance with material in this section.

* Thanks to Michelle Donaghy for guidance with material in this section.

† Adapted and reproduced with permission: Jones, M et al (2000), Treating stuttering in young children: Predicting treatment time in the Lidcombe Program, *Journal of Speech, Language, and Hearing Research, 43*, 1440–1450. © 2000 American Speech-Language-Hearing Association.

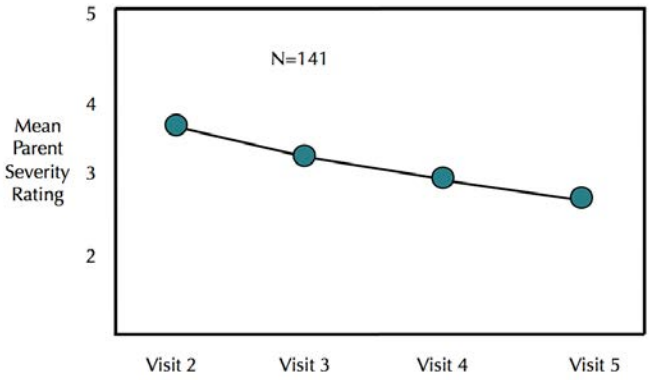
Two of these publications contain around 40% of the cohort (N=316) who were treated during 1989–2001. At that time, the Lidcombe Program criteria for progression to Stage 2 needed to be attained for 1 week only, not the three consecutive weeks specified in the current treatment guide. These reports contain a median of 11 clinic visits, compared to 16 for the remaining studies. Consequently, the treatment times in these reports were adjusted by adding five more clinic visits to their median values, bringing them into line with the subsequent studies.

The graph of these data in the figure above is known as a recovery plot, and it shows the proportion of children to attain Stage 2 and the number of clinic appointments to do so. With the median number of clinic appointments being 17, half of those 925 children required fewer than 17 clinic appointments for Stage 1, and half required more than 17 clinic appointments. The estimated 90th percentile for the recovery plot is 28 clinic appointments. In other words, 90% of cases will have attained Stage 2 by 28 clinic appointments.

An important note here is that these data describe trends in large groups of clinical children, as do the results of the clinical trials described earlier. As such, care is needed in evidence-based reasoning to form a judgement about the extent to which these data apply to any individual clinical child. One consideration will be comorbid diagnoses and case features. Two of the prior reports,[100,101] for example, describe nine children—3% of the caseload—who dropped out of Lidcombe Program treatment because of comorbid speech and language problems, challenging behaviours, and complex family problems. Indeed, little is currently known about treating children with speech disorders that are comorbid with stuttering.[104]

Additionally, the treatment time data just described, pertain to a population of clinicians, and care is needed about applying them to an individual clinician. Treatment times for individual clinicians will vary according to the nature of their caseloads and their clinical experience and training.

The next figure‡ is the mean parent SR for a report of 141 cases,[102] showing an average reduction of around one third during the first five clinic appointments (4 weeks of treatment).



How does a treatment delay affect the treatment process?

The Lidcombe Program

The next figure,* below, shows the results for 316 children in the studies mentioned earlier.[100,101] The recovery plot on the left shows the children who had been stuttering for more than 12 months, and the recovery plot on the right shows the children who had been stuttering for less than 12 months. The children who had been stuttering for less than 12 months have the same shape of recovery plot as the others, however it is moved to the right to a statistically significant extent. This means that both groups

‡ Adapted and reproduced with permission: Onslow, M et al (2002), Beyond-clinic speech measures during the Lidcombe Program of early stuttering intervention, *ACQuiring Knowledge in Speech, Language and Hearing, 4*, 82–85. © 2002 Speech Pathology Australia.

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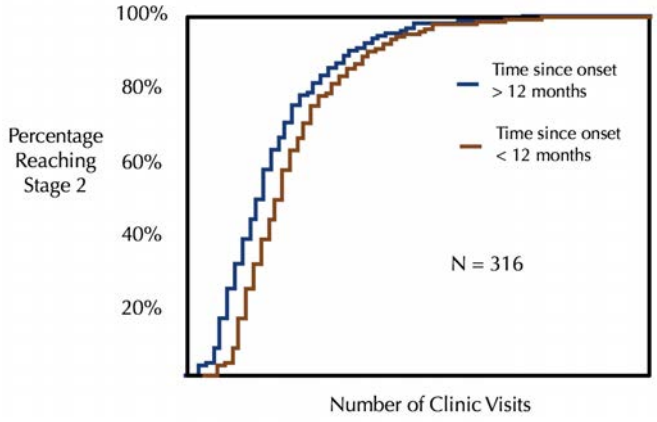
of children, overall, responded in the same way to the treatment, but the children who had been stuttering for less than 12 months required a few more clinic appointments to reach Stage 2.



So, in terms of important clinical significance, it appears that delaying the Lidcombe Program for up to a year after stuttering onset is unlikely to jeopardise a child's responsiveness in terms of time taken to reach Stage 2.

This research result is intuitive, considering, as discussed during the previous lecture, that the Lidcombe Program places a cognitive load on children to understand the treatment process. It appears from these results that children who have been stuttering a little longer, and consequently are a little older and more cognitively developed, are a little more responsive in terms of time taken to reach Stage 2. In short, there are empirical and logical grounds to suggest that the Lidcombe Program is not optimally suitable for very young children who have begun to stutter.

Do case variables affect the treatment process?

The Lidcombe Program

Pre-treatment stuttering severity

Six studies[4,42,52,62,100,101] have used a statistical technique called logistical regression to predict treatment time with the Lidcombe Program. These analyses show that pre-treatment stuttering severity accounts for around 20% of the number of Stage 1 clinic visits required. This finding is intuitive; if there is more stuttering it takes longer to control it. The prospective observation study of parents doing the Lidcombe Program discussed earlier[64] reported the same effect with a strong association. The study of the Lidcombe Program in a student clinic mentioned earlier[103] also reported that effect. In a study of a clinical caseload,[105] the 10 children who took the longest to complete Stage 1 had more severe stuttering than 10 children who completed Stage 1 in the shortest time.

Another clinically useful perspective on this matter was presented in the Lidcombe Program translation report with community clinicians.[42] There was a 17% increase of Stage 1 clinic visits for every one SR scale value pre-treatment. So that could make quite a difference for a child with SR 4 compared to a child with SR 8. Such difference could prompt a clinical decision to begin intervention earlier with a more severe child so that treatment is completed before the school years.

Phonological and language development

There is logistical regression evidence in one clinical trial[4] that phonological development does not predict treatment time. However, this report found that, together with pre-treatment stuttering severity, receptive language scores and language development (Mean Length of Utterance), predicted 34% of the variance of clinic visits needed for Stage 1.

The result is a little difficult to understand, because better language development was associated with shorter treatment time, but higher receptive language scores were associated with longer treatment time. The former result seems intuitive, but it is not at all clear why better receptive language would be associated with longer treatment time. Without replication of the result, it is possible that the latter, unintuitive result is a Type II statistical error, where a finding is reported when in fact it is not true.

Siblings treated previously

The study of the clinical caseload mentioned previously[105] reported a novel, but intuitive finding. One variable that characterised the children in the short treatment time group was that a sibling was more likely to have been treated with the Lidcombe Program previously.

Predicting treatment outcome

All reports discussed so far have dealt with predicting treatment *time* for the Lidcombe Program, but there is one report that predicted treatment *outcome*.[106] For a cohort of 277 children who received Lidcombe Program treatment, 32 variables were used to predict short-term and medium-term treatment outcome. Outcomes at 6–9 months and 12–18 months after the start of treatment were measured with parent report of stuttering severity. The study also explored predictors of whether parents would drop out of treatment. The 32 predictor variables spanned domains of demographics, stuttering severity, child speech and language, and child and parent psychometrics.

Results were that better language skills and "easy" childhood temperament (see Lecture Ten) were statistically associated with better treatment outcomes. However, those results, albeit intuitively correct and statistically significant, were not clinically significant. Those variables only accounted for a minute portion of the variance of treatment outcome. No predictors of treatment dropout were found except—intriguingly—that parents who failed a personality screening relating to their impulsivity were 3.5 times more likely to drop out of treatment than parents who did not. The authors cautioned that the latter finding requires replication with a full personality assessment before it can be given credence.

Parent experiences

The Lidcombe Program

The importance of this topic

There have been three studies of how parents experience the Lidcombe Program.[107,108,109] These reports provide useful information to forewarn clinicians about both positive and negative features of the treatment that parents might encounter. In particular, clinicians can be forewarned about potential adverse parent experiences. This material is supplemented with an interview report[110] of a parent and child 7 years after completing the Lidcombe Program.

One report[107] surveyed 35 parents whose children had recently completed Stage 1 of the treatment. Results showed the Lidcombe Program to be a generally positive experience for parents. Some reported lacking confidence to do it, but valuing clinician support with overcoming that feeling. Some parents reported a sense of empowerment with doing the treatment. Some parents reported difficulty doing the treatment, mainly with finding the time each day for it. Most parents reported a positive response from children to the treatment, but some children did not like being interrupted by parent comments. There were no parent reports of adverse reactions, such as reduced talkativeness, and "in fact, many parents reported their child was more confident and more talkative as a result of the treatment" (p. 422).

Another report[108] found three typical paths (outlined below) through the treatment based on interviews with 14 parents of children who were being treated. Six of the parents were interviewed on two occasions. All but one interview was with mothers.

Path One: Straightforward parent experiences

The first path reported was a straightforward one, with parents enthusiastic and innovative. They easily incorporated the treatment into their lifestyles in an enjoyable manner and were able to do some independent problem solving instead of relying overly on the clinician. These families attained a quick and steady therapeutic response, with the child assuming some responsibility for the treatment.

Path Two: Straightforward parent experiences then problems

The second treatment path reported was a straightforward one initially, with problems subsequently encountered. Parent guilt about not being able to commit properly to the treatment, and guilt about stuttering itself, began to emerge from them. These parents became needy of support, the visits to the clinic became a burden for them, and they found the treatment difficult to sustain. The children of these parents began to be unresponsive, and even irritated, by parent verbal contingencies.

Path Three: Problems from the outset

The third treatment path through the Lidcombe Program involved encountering problems from the outset. These cases were in the minority though. Such parents had trouble doing the verbal contingencies, and were not particularly adept at leading the child to do the treatment. These parents would doubt their capacity to do the treatment, and they focussed on problems they were having rather than how to solve them. These parents, more than the others, talked about "their anxieties, feelings of inadequacy, guilt and distress" (p. 24).[108] The beliefs of such parents about stuttering and parenting were not a good fit with the Lidcombe Program. It seems clear that if the Lidcombe Program continues with sporadic or little progress, such parents can become distressed.

Treatment implementation problems

Another report[109] involved 16 mothers who were each interviewed nine times during the course of treatment during a 6-month period. The key findings of the report dealt with treatment implementation, perception of the treatment, and parent emotions.

The first finding dealt with obstacles implementing the treatment. Those obstacles can be summarised as

(1) Problems finding time to do the treatment

(2) Forgetting to do the treatment

(3) Problems managing siblings throughout the treatment.

Fourteen of the 16 mothers had more than one child and so issue (3) was prominent. Despite these implementation problems, a number of benefits were reported, including an increase of quality time with the children, along with an improved bond between them, and increased knowledge about stuttering. Improved parenting skills were also a feature of the treatment, which is not surprising because it is a behavioural treatment that places some demands on children.

Parent perceptions

Another emerging theme was about treatment credibility and expectancy. As discussed during Lecture Three, these are important issues related to treatment compliance. Mothers' perception of the treatment could be too ambitious, expecting it all to be over in a few weeks. And there was also an issue of just not expecting it to work:

> I wouldn't hesitate for anyone to try it because I didn't think it was going to work. I didn't think that saying smooth and bumpy talking was really going to make such a difference and it did. (p. 76)[109]

Parents could also be taken by surprise by not expecting to have to do the treatment themselves and anticipating that the clinician would do the job for them. Parents offered suggestions about how the treatment could improve and proffered the need for "bigger picture" information about the treatment, more treatment documentation, and a support group.

Another aspect of this theme was the children's reaction to the treatment. It was commonly reported that they were becoming aware of when they were stuttering and, therefore, self-evaluated stuttering. Eventually, they became more self-confident and lost some of their shyness. Direct signs of children enjoying the treatment were reminding parents to do treatment, and clear signs of enjoying praise for stutter-free speech. There were, however, two emerging topics suggesting negative reactions to the treatment. These were negative reaction to verbal contingencies and suggestions that the child had done something bad by stuttering. Interestingly, some children seemed to react negatively to the word "smooth" and reacted better when parents substituted something different like "great talking."

Parent emotions

Parents were reported to experience nine emotions during treatment.[108] The five most common were judged as those emotions that appeared most strongly and were most often mentioned, and how many mothers reported them. The first was "empowerment and responsibility," with parents realising it was up to them alone to incorporate the treatment into their lives. The accompanying responsibility could lead to anxiety and pressure to perform well with the treatment. Indeed, "anxiety" was a strong theme emotion. Parents could be concerned about doing the treatment properly and by a fear that their children were being teased and bullied at school, and worried that their children would still be stuttering when they went to school.

"Parent guilt" occurred for many reasons, such as a belief it was they who caused the stuttering, guilt that they were not doing the treatment correctly, and guilt about not finding time to do it during the day. Eight mothers reported distress related to stuttering severity and the experience of having to watch the child stutter during treatment. The final parent emotion was referred to as a "cycle of confidence." Mothers' confidence followed the ups and downs of their children's stuttering and their success in implementing the treatment. This cycle of confidence was reported throughout the entire 6 months of the study.

The early stuttering intervention evidence base: Summary and conclusions

The Lidcombe Program

Independent reviews consistently report that the evidence base for the Lidcombe Program is the most comprehensive available among early stuttering treatments.[111,112,113,114,115,116,117,118,119,120] This evidence base includes nonrandomised clinical trials, nine randomised trials, randomised clinical experiments, case studies, treatment process reports, and qualitative studies of parent experiences. A Cochrane Review[119] noted that the treatment is unique with its demonstration of superiority over no-treatment controls. Research publications continue to emerge, as described earlier during this lecture. A survey that included 124 Australian children who stuttered, with a mean age of 11 years,[121] reported that 50% of them had received the Lidcombe Program.

Regardless, for the evidence-based practitioner, this empirical base is far from ideal according to accepted standards of health care research. The Cochrane Review[119] concluded that results should be interpreted cautiously because the "certainty of the evidence" is "very low and moderate" (p. 2). Additionally, the review cautioned that the randomised trials concerned had a "high risk of overall bias" (p. 2).

Treatment fidelity, or implementation fidelity refers to whether a treatment is administered as intended, and is an important consideration with treatment translation in general,[122,123] and the issue is thought to be important with stuttering treatment,[124,125] and particularly so with randomised trials comparing stuttering treatments for early stuttering.[126] A review of implementation fidelity[127] for behavioural, parent-implemented treatments for pre-schoolers included a majority of studies of the Lidcombe Program. No study reported ideal components of implementation fidelity, leading the authors to conclude that "rigorous measurement and reporting of [fidelity implementation] in future intervention studies is required in order to better inform evidence-based practices for interventions with [children who stutter]" (p. 1)

That said, a strength of the Lidcombe Program evidence base is that it involves direct assessment of treatment effect size from randomised control trials and randomised controlled experiments that compared the treatment with a no-treatment control group. Another strength of the evidence base is that it contains replicated findings that are independent of the original Lidcombe Program developers, most notably the randomised trial comparing the Lidcombe Program with RESTART-DCM.[26] That being said, there has been no replication of the randomised controlled trial of the Lidcombe Program[7] showing an effect greater than natural recovery in a no-treatment control group.

For more than two decades, research has sought to establish the mechanism or mechanisms that underlie the Lidcombe Program treatment effects. This research continues today, but at present, the mechanism, or mechanisms, underlying the Lidcombe Program remain unclear. Their discovery may eventually lead to the development of a treatment with much different characteristics to the current structure of the Lidcombe Program.

Treatments based on multifactorial models

A strong feature of the evidence base for this style of treatment is that it includes the largest randomised clinical trial of a stuttering treatment reported to date,[26] although positive clinical trial results for this style of treatment await replication. Replication is a substantive issue here, considering that the non-randomised Phase I trials of Palin Parent-Child Interaction therapy did not produce convincing evidence of a treatment effect. Those trials showed a quarter of the children with post-treatment stuttering severity below 1.5 %SS, compared with three-quarters of the children in the randomised trial. Replication of the results of clinical trials of treatments based on multifactorial models is confounded by the fact that two of its prominent treatment variations—RESTART-DCM and Palin Parent-Child Interaction Therapy—have clinically dissimilar treatment goals. It has been argued that a limitation of the evidence base for treatments based on multifactorial models is that, in contrast to the Lidcombe Program, it contains no direct estimate of effect size for the treatment derived from comparison with a no-treatment control group.[39] But as noted earlier, the RESTART trial provides no evidence that effect sizes, in terms of percentage stuttering reduction, are different from those obtained with the Lidcombe Program.

The Westmead Program

This treatment is at the early stages of its development. Data available are nonrandomised Phase I and Phase II trials, and one randomised controlled trial comparing it to the Lidcombe Program. However, the latter trial was severely compromised by high drop-out rates. In order to be as compelling as the evidence that is available for the Lidcombe Program and RESTART-DCM Treatment, evidence from further randomised trials will be required.

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LECTURE EIGHT: EVIDENCE-BASED SPEECH RESTRUCTURING TREATMENTS FOR PERSISTENT STUTTERING

Speech restructuring treatment

Background

Overview of speech restructuring

Speech restructuring refers to the use of a novel speech pattern to reduce stuttering or eliminate stuttering while sounding as natural as possible.[1] During speech restructuring, clients learn to speak initially with a slow, drawling speech pattern that is stutter-free. The speech pattern is then shaped toward stutter-free speech that is as natural sounding as possible. Surveys of those who have sought treatment for stuttering[2,3,4] confirm that this approach is desirable for a substantive portion of them.

Terminology

There are many variants of this clinical technique currently in use, referred to with many different terms: *prolonged speech*, *smooth speech*, *easy speech*, *fluency shaping*, and *precision fluency shaping*. They include target speech behaviours taught to clients such as *reduced speech rate*, *extended vowel production*, *light articulatory contacts*, *gradual onset of vocalisation* (also known as *gentle onsets*), and *continuous breath flow during speech*.

The mechanism of speech restructuring

All these speech pattern techniques sound quite similar, and it is quite likely that they all work in essentially the same way to control stuttering. The target speech behaviours of the treatment have been associated with post-treatment acoustic changes such as reduced articulation rate, reduced duration of phonation intervals, reduced variability of vowel duration, increased voice onset time, vowel duration, and intervocalic interval.[5,6,7,8,9,10] However, no specific underlying acoustic mechanism has been found that might explain how the treatment functions. As discussed during Lecture Three, it is a theoretically tenable idea that stuttering moments reflect an inherently unstable speech motor system. If this is so, it is plausible that these speech patterns somehow, in a manner currently unknown, offset that problem by stabilising the speech motor system.

A variant: Stuttering modification

The term *stuttering modification* (sometimes *stutter more fluently*) is related to speech restructuring. Although it appears to be used less commonly these days, it refers to an alternative approach to controlling stuttering with a novel speech pattern. Charles Van Riper was an extremely influential clinician, largely because he developed the "stutter more fluently" technique.[11] The technique is not intended to impose an overarching speech pattern to control stuttering. Instead, the technique provides a way for clients to stop or reduce struggle with individual stuttering moments. Terms for the components of this technique are *cancellations*, *preparatory sets*, and *pull-outs*. These techniques and the history of their development are described in a reference text (p. 429–431).[12] However, as noted there,

> "stuttering modification" … is a popular therapy in many places, although large-scale data verifying its effectiveness are still relatively sparse more than half a century after initial reports of its use. (p. 431)

Since this statement, there have been two reports of treatments that have incorporated Van Riper's techniques,[13,14] although neither report conformed to the discipline standards for a clinical trial of stuttering treatment provided in Lecture Five. The first report had all the features of a clinical trial except that speech measures were made in the clinic at 6 months post-treatment, and the second report was a file audit.

Another variant: Voluntary stuttering

This is another commonly recommended procedure that is related to stuttering modification, which, again, has limited research about its clinical value. It is also referred to as *negative practice*, *pseudostuttering,* and *bouncing*. Although it is a stuttering modification behaviour, it is designed "to reduce fear, anxiety, and/or negative emotions associated with stuttering" (p. 290).[15] As such, it is classifiable within anxiety management strategies known as *behavioural experiments*, which will be discussed during Lecture Eleven. The report just mentioned[15] surveyed 206 participants who had knowledge of the technique, and reported that around half reported that it assisted with their fear of stuttering, and around a third reported that it made them feel more confident with their speech. It was clinically important, though, that the report noted that around two-thirds of the clients reported discomfort using the technique and that "when they first used voluntary stuttering it was too emotionally difficult for them to use in everyday situations" (p. 295).[15]

A brief history of speech restructuring

A long time ago …

The history of speech restructuring has been described in detail.[16] Lecture One described how Satyrus seems to have used rhythmic speech during the third century BC to help Demosthenes with stuttering. This appears to be the first recorded use of a novel speech pattern to control the disorder. It is also generally believed that Satyrus recommended that Demosthenes speak above the roar of an ocean. It is completely possible that this was another use of speech restructuring to assist with stuttering; speaking with increased volume may have induced a novel speech pattern.

Twenty centuries later …

This brief historical overview jumps some 20 centuries to 1724, when an American minister of religion, Cotton Mather, published the following in a medical treatise, which describes a technique that has obvious conceptual similarity to modern speech restructuring:

> While you go to snatch at Words, and are too quick at bringing of them out, you'l be stop'd a thousand Times in a Day. But first use yourself to a very deliberate Way of Speaking: a Drawling that shall be little short of Singing. Even this drawling will be better than Stammering; especially if what you speak, be well worth our waiting for.[17] (p. 460)

It is clear that the use of speech restructuring variants occurred many times during subsequent centuries, and a full historical account is given in a 1984 text.[18] However, the present brief account jumps to 1951, when the effects of delayed auditory feedback—described during Lecture One—were reported. The oddities of speech under the influence of delayed auditory feedback were referred to initially as "artificial stutter."[19] This began a long period of research about the effects of delayed auditory feedback on stuttering, which eventually did not lead to any real understanding about the nature and cause of the disorder.

The 1960s onwards …

Although the discovery of the effects of delayed auditory feedback did not yield any theoretical insights, its clinical impact was extensive. During the early 1960s Israel Goldiamond discovered that those who stutter could overcome the effects of delayed auditory feedback by using a slow and drawling speech pattern. As it happened, that speech pattern could keep stuttering in check. He coined the term *prolonged speech* for this speech pattern, published a report about its clinical value in 1965,[20] and a revolution began with treatment for persistent stuttering. Goldiamond's technique was to establish a novel and slow speech pattern to control stuttering, using a delayed auditory feedback device. The next steps of treatment were that the delay was systematically reduced and speech rate was systematically increased.

In the reference text mentioned earlier[18] there is a detailed historical account of this style of treatment spreading through the Western World, and probably beyond. The use of delayed auditory feedback was soon found to be unnecessary and was replaced with recorded models of the requisite speech

pattern and clinical instruction. There have been more clinical trials of speech restructuring treatment, with more participants, and with more independent replications, than for any other stuttering treatment. By the end of the 1970s there was sufficient research for a meta-analysis of the efficacy of speech restructuring treatments, and a conclusion that the method was more efficacious than any other.[21] Things have not changed since, with it being the most promising way for adults to control their stuttering, should they wish to do so. A survey that included 625 Australian adults who stuttered[22] reported that around a quarter of them had received a speech restructuring treatment.

Programmed instruction

A technique for behavioural control

More often than not, speech restructuring treatments reported worldwide incorporate programmed instruction, which is a technique invented by the behaviourist B. F. Skinner. Its principles and their application to speech-language pathology have been outlined in detail.[23] It is a technique for learning behavioural control—of stuttering in this instance—with small increments arranged in a hierarchy of what is presumed to be easier to more difficult. Clients learn to master the hierarchical increments in small steps within a pre-determined sequence.

Performance continent progression

Some clients will take longer than others for any given programmed instruction sequence, and one reason is that progress through the incremental steps is performance contingent. In other words, there is a criterion, or several criteria, for completing each of the incremental steps. For example, one of the criteria for completing an incremental step might be no stuttering. In which case, in the event of a stuttering moment occurring while a client is attempting to complete a step in the programmed instruction hierarchy, the client has to return to the start of the sequence.

A fundamental assumption

Programmed speech restructuring contains a clinical assumption that each step in the hierarchy is more difficult than the previous one for the client to achieve. Therefore, it is also assumed that moving through the hierarchy is a productive way to learn, and that success at one step depends on success at the previous step. Those assumptions may well be true, but they are not substantiated by any research about stuttering treatments.

Models of programmed instruction

Most speech restructuring clinical trials have involved programmed instruction, in a set sequence. Commonly, intensive treatment formats are used. These raise health economics issues about efficient use of clinical resources. For example, there has been an observation[24] that the results of a 5-day, non-residential intensive treatment[25] appear similar to the results of a 3-week residential treatment.[26]

In fact, intensive speech restructuring treatment does not seem essential to a positive treatment outcome. A clinical trial[27] suggested that 10 one-hour sessions of individual treatment produced equivalent results to a treatment version involving an intensive treatment day. And there has been a clinical trial[28] showing that 16 two-hour sessions over four consecutive days produced equivalent results to two 2-hour sessions for 8 weeks.

With many programmed instruction stuttering treatments, the hierarchical sequence involves speech rate increments, commonly syllables per minute (SPM). The target speech rate increases with a sequence of six steps from extremely slow, such as 50 SPM, to a target speech rate somewhere near typical rates, or appreciably slower, such as 200 SPM. It is not realistic to require clients to speak with their speech rate exactly at specified SPM values, so they are given some leeway, such as plus or minus 20 SPM. There are several speaking tasks within each step, usually from five to 10. The speaking tasks might involve monologues, or conversation with a clinician.

Instatement and transfer

The part of treatment just described is often referred to as *instatement*, and sometimes as *establishment*. Subsequently, in such treatments the client traditionally enters a *transfer* phase. During the transfer phase procedures are introduced that are designed to generalise the newly learned speech skills to everyday speaking situations. The transfer phase can involve a whole new programmed instruction sequence involving speaking tasks that are arranged hierarchically, from easy to difficult.

The clinical trial evidence for speech restructuring treatment

There have been more positive clinical trials of speech restructuring for persistent stuttering—more than 30— than for any other treatment. These trials are by many independent researchers in different countries, dating from 1973. A systematic review conducted in 2019[29] confirmed that this treatment for persistent stuttering has stronger evidence than any other.

Effect size

As discussed during Lecture Five, effect size for a treatment can be assessed by comparing a treatment arm in a clinical trial with a control arm that receives no treatment. However, neither of the randomised trials of speech restructuring[27,30] involved a no-treatment control arm, so the effect size for the treatment cannot be estimated.

Another problem with knowing what the effect size might be for speech restructuring treatment is the high drop-out rates in clinical trials. In one trial[31] 20 of 32 dropped out, and in another trial[26] 13 of 30 dropped out. Equally troubling is that some trials with substantial participant numbers did not report whether there were any drop-outs: one trial with 36 participants,[32] one with 39 participants,[33] and another with 44 participants.[34] In fact, only one trial[25] with substantial numbers reported few drop-outs: two of 80 participants. So the usual bias where non-randomised trials overestimate effect size[35] would be enhanced by these drop-out rates.[†] Only two clinical trials of speech restructuring treatment have used the technique of intention to treat analysis, mentioned during Lecture Five, to compensate for this problem.[27,30]

In light of all this, perhaps a reasonably conservative statement is that some of those who wish to attain clinically significant reduction of their stuttering will be able to succeed in doing so. The proportion of those who will succeed is unknown. The overall tenor of clinical trials—biased as they are—conveys that significant stuttering can be reduced to below 4 %SS or even below 1 %SS. There is some evidence that those stuttering severity reductions are not associated with changes to the behavioural complexity of stuttering.[36] As outlined during Lecture One, a stuttering moment can involve repeated movements, fixed postures, or superfluous behaviours. The report[36] indicated that, after speech restructuring treatment, there was no change in the overall behavioural complexity of stuttering moments. In other words, while the amount of stuttering reduced, the types of stuttering behaviours remained constant.

Again with the reservation that the clinical trials concerned may be biased, it appears that clinically significant stuttering reductions may be obtained for periods of 1–2 years, which is the general follow-up period in clinical trials. The longest follow-up periods for clinical trials that showed sustained stuttering reductions were 9–12 years (N=12),[37] this being a long-term follow-up of an earlier trial,[31] and 10 years (N=17).[38] Both results were for multi-week, intensive residential treatments.

As mentioned several times throughout these lectures, post-treatment relapse is a recurring problem with speech restructuring treatment.[39,40] Relapse rates were specifically reported in some of the clinical trials that have been published for adults. For example, one trial[32] reported 30–60% relapse at 12–18

[†] This reasoning assumes drop-outs are connected to undesirable features of the treatment. However, it is possible that the reverse is true. Participants could drop-out from clinical trials because the treatment is so rapidly efficacious that they do not bother to continue their participation in the trial.

months, depending on how relapse was defined. Another trial[26] reported 24% relapse in terms of more than 6.0 %SS at 12 months post-treatment.

Speech naturalness

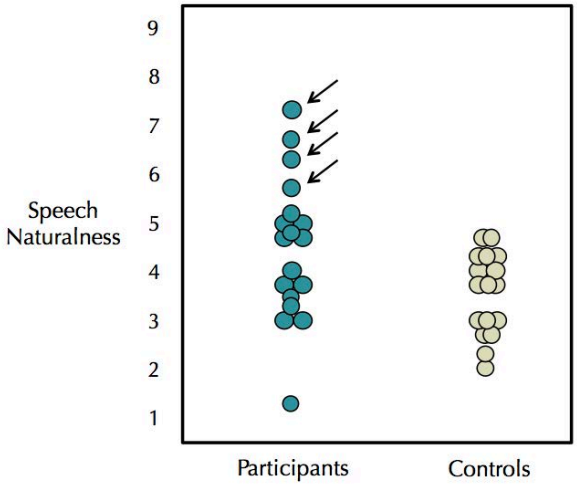
How speech sounds

As discussed during Lecture Four, gains from speech restructuring treatment are typically achieved at the cost of speech that does not sound perfectly natural. This problem has been known for decades.[39] There is an extensive body of literature dealing with the problem of post-treatment speech naturalness with speech restructuring treatment; 30 pertinent publications are documented in a reference text[12] (p. 341–342). There is no research evidence to support the contention, but it seems likely that unnatural sounding post-treatment speech contributes to the relapse problem.[41] A theoretical mechanism for this would be that unnatural sounding speech prompts listeners to evaluate such speech negatively, causing anxiety for the speaker. Such anxiety is known to have a detrimental effect on maintenance of speech restructuring treatment benefits, as will be described during Lecture Ten.

Following is a recent statement of the problem of speech naturalness and speech restructuring treatment:[42]

> Communication effectiveness can be diminished if gains in fluency are achieved … through the use of speaking techniques that are so burdensome and unnatural that the individual has difficulty using them on a consistent basis. (p. 290)

Consequently, since the early 1990s, journal editors generally do not accept speech restructuring clinical trials for publication without speech naturalness assessment of some kind. The accompanying figure,[‡] from a clinical trial,[43] illustrates the issue. Listeners assigned speech naturalness (NAT) scores to post-treatment speech samples of 18 participants who received speech restructuring treatment, and matched controls. As a group, the treated participants scored a little less than one NAT scale value higher than controls: means of 4.5 and 3.6 respectively. The figure shows that, with the exception of four participants marked with arrows, the group who received treatment attained NAT scores around the range of controls.



How speech feels

An important clinical issue with speech naturalness is that clinicians cannot assume that speech feels as natural to clients as it sounds. In fact, there is research to show that how natural speech sounds and how natural speech feels may be different things altogether.[44,45] The latter of these reports involved interviews of clients after speech restructuring treatment. Results showed one thing that drove clients to receive treatment was that they felt different because of their stuttering. However, the treatment by no means took away that feeling, but worsened it: feeling different from their normal way of speaking. This is important information for clinicians. A treatment that controls stuttering but either sounds or feels unnatural may not be particularly useful to a client.

[‡] Adapted and reproduced with permission: O'Brian, S et al (2003), The Camperdown Program: Outcomes of a new prolonged-speech treatment model, *Journal of Speech, Language, and Hearing Research, 46,* 933–946. © 2003 American Speech-Language-Hearing Association.

Speech restructuring I: The Camperdown Program

Background

Overview

This treatment is an example of a non-programmed speech restructuring model for adults. In summary, the treatment incorporates a video demonstration of the *Camperdown Program Training Model*. The clinician guides the client in using the speech pattern in the training model to develop an individualised *fluency technique* to reduce or eliminate stuttering during everyday speech and to sound as natural as possible.

Nonprogrammed instruction

The development of the Camperdown Program as a nonprogrammed treatment was prompted by a laboratory experiment[46] with three adults who had never experienced speech restructuring treatment. They learned a speech restructuring pattern and were then able to use it to control stuttering and sound reasonably natural simply by being instructed to do so. Programmed instruction was not necessary for them to attain this laboratory result.

No speech targets

As described earlier, speech restructuring clinical procedures typically involve teaching clients target speech behaviours: extended vowel production, light articulatory contacts, gradual onset of vocalisation, and continuous breath flow during speech. However, those speech targets have long been recognised as a threat to treatment replicability.[47] The term "treatment replicability" refers to whether clinicians can do a treatment in the same way that the clinicians did the treatment in published clinical trials.

The replicability problem with such speech targets was illustrated in a report[48] where seven clinicians, being experienced with one particular speech restructuring treatment, were shown video recordings of clients demonstrating the target speech pattern at various stages of that treatment. The clinicians did not agree at all about whether the clients were using the program target behaviours correctly or incorrectly.

This result prompted the Camperdown Program to be developed without using any speech targets during the treatment process. Instead, the clinician shows the Camperdown Program Training Model of the required speech pattern to clients and asks them to imitate it and to control stuttering while doing so. The clinician gives feedback about how closely the client imitates the Training Model, using whatever instruction is appropriate.

No transfer phase

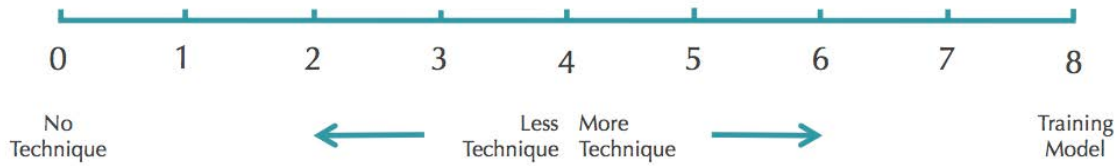
Another report[49] that influenced the Camperdown Program development showed that a traditional speech restructuring treatment involving a transfer phase remained efficacious when its transfer phase of treatment was replaced with speech practice. In response to this publication, Camperdown Program development proceeded without a formal transfer phase.

Clinical resource materials

The Camperdown Program Treatment Guide is available from the Australian Stuttering Research Centre website.[50] At that website there are downloadable clinical forms: (*Camperdown Program Situations Measurement Chart eForm, Camperdown Program Fluency Cycles Chart eForm, Camperdown Program Daily Measurement Chart eForm.*) Additionally, at this website there are downloadable video demonstrations of the Camperdown Program Training Model at around 70 SPM. The website also contains other downloadable clinical materials for use during the treatment process. The description of the Camperdown Program in the following sections draws freely from the treatment guide. A separate publication[51] outlines the use of technology, such as the Scenari-Aid website,[52] during the treatment process.

Stage I: Teaching treatment components

Stage I typically involves weekly clinic appointments of around 1 hour duration. One purpose of Stage I is for clients to learn imitation of the Camperdown Program Training Model using one of the downloadable video examples. Another purpose of Stage I is for clients to learn how to use the Stuttering Severity Scale and the Fluency Technique Scale. The Stuttering Severity Scale has nine points, where 0 = *no stuttering*, 1 = *extremely mild stuttering*, and 8 = *extremely severe stuttering*. The Fluency Technique Scale was based on research that developed the scale of speech naturalness described during Lecture Four. It has nine points, where 0 = *no technique* and 8 = *very obvious technique*. The use of these scales is described in detail in the Camperdown Program Treatment Guide. The Fluency Technique Scale appears below.



Clients are required to imitate the Training Model using fluency technique 7–8 and to speak using it spontaneously without stuttering, with a stuttering severity score of 0. Clients imitate the video Training Model and the clinician gives feedback about the attempts, without reference to any speech targets. The clinician may direct clients' attention to certain parts of the Training Model and encourage them to listen again and try to copy that section more closely.

Clients learn to use the Stuttering Severity Scale by giving a score to their speech, based on recordings of themselves and during real time, for short periods of 1–5 minutes. Clients compare their stuttering severity scores with those given by the clinician. Clients are required to measure their severity to within one scale value of the clinician's score.

Stage II: Establishing stutter-free speech

During Stage II, clients:

(1) Consolidate their learning of the Training Model from Stage I

(2) Work with the clinician to develop an individualised fluency technique that sounds natural and useable for stuttering control

(3) Continue self-evaluation of their stuttering severity and fluency technique

(4) Establish problem-solving skills for Stage III, which involves generalisation of stutter-free speech to everyday speaking situations.

Stage II can be done individually with weekly clinic appointments of around an hour duration, or in an intensive group format. Typically, clinicians find the former to be a more practical option.

Fluency cycles: Overview

During Stage II, clients rotate through a series of fluency cycles. They use massed practice for clients to establish their own fluency technique that works to control their stuttering and, eventually, sounds as natural as possible. One cycle consists of three parts: Fluency Technique Practice, Experimentation, and Planning. Each part takes 3–4 minutes. Clients complete as many of these cycles as needed to achieve program criteria and progress to Stage III.

Fluency cycles: A. *Fluency Technique Practice*

The aim of Fluency Technique Practice is for clients to consolidate their learning of the Training Model that occurred during Stage I. Repeating this consolidation process during a series of "fluency cycles" ensures that the basic skill of controlling stuttering is continually reinforced. Clients do not

make any attempt to sound natural. Clients practise fluency technique 7–8 of the Training Model. Throughout Fluency Technique Practice, the aim is for clients' speech to continue to sound like the model and to remain stutter-free. After practicing, clients record a stuttering severity score and a fluency technique score. The clinician gives feedback in the same manner as in Stage I.

Fluency cycles: B. Experimentation

The Experimentation part follows the Fluency Technique Practice part of a fluency cycle. The aim here is for clients to develop an individualised fluency technique and begin to sound more natural while maintaining low levels of stuttering. Clients experiment with using and evaluating as many different features of the Training Model as might be needed to control their stuttering. It is intended that each client will ultimately develop an individualised fluency technique to control stuttering.

The Experimentation part consists of three steps: Goal setting, Evaluation after speaking, and Evaluation after listening to the speech recording.

Goal setting. Clients set stuttering severity and fluency technique goals to achieve. The primary goal is to remain stutter-free, hence the stuttering severity score goal is always 0. However, clients decide the fluency technique goal, differently each time if necessary. Clients determine the fluency technique goal by:

(1) Reviewing evaluations of previous cycles in terms of their success with controlling stuttering

(2) Evaluating how much fluency technique will be needed to control stuttering.

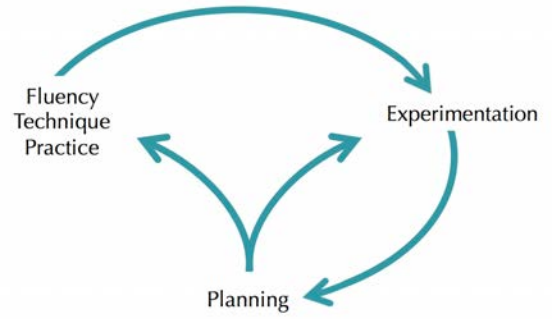
It is important for clients to do this goal setting with limited guidance from the clinician. Being able to plan a stuttering control strategy based on evaluation of previous performance is critical to the success of the Camperdown Program. Clients then speak for 3–4 minutes attempting to meet the set goals. Clients make an audio recording of this speaking task.

Live evaluation. The second step is client evaluation of the speaking task without listening to the audio recording. Clients record a stuttering severity score and fluency technique score for the speaking task. The clinician does not discuss these scores with the client at this stage. This process is designed to simulate everyday situations where clients need to evaluate their speech, and make decisions about stuttering control without clinician assistance.

Recording evaluation. The third step is an evaluation of the audio recording. Clients listen to the audio speech recording to confirm or correct the evaluations made during the evaluation after speaking step. This is done in consultation with the clinician and agreement about the evaluation needs to be reached by both.

Fluency cycles: C. Planning

Planning is the third part of a fluency cycle. The aim is for clients, initially with guidance from the clinician and ultimately alone, to use self-evaluations from previous fluency cycles to plan a strategy and set stuttering severity and fluency technique goals for the next cycle. During the planning part of a fluency cycle there are two ways for clients to proceed, as the accompanying diagram shows.



If a client attains a stuttering severity score of 2 or more during the previous Experimentation part of a fluency cycle, the client returns to the Fluency Technique Practice part to start the next cycle.

If a client attains a stuttering severity score of 0–1 during the previous Experimentation part of a fluency cycle, the client chooses to begin the next cycle either at Fluency Technique Practice or Experimentation. Clients who choose the latter may choose to speak with a more natural sounding fluency technique goal than the previous time.

Regardless of how clients proceed at the Planning part of a fluency cycle, they begin at least every third cycle with the Fluency Technique Practice part. The point of this is to constantly consolidate their basic fluency technique skill.

Stage III: Generalisation

During Stage III clients attend the clinic each week to:

(1) Consolidate use of their fluency technique to control stuttering
(2) Compare their speech measures during the appointment with the clinician's
(3) Review their fluency technique practice routine with the clinician and revise it as needed
(4) Present recordings of their speech and speech measures in daily situations and discuss them with the clinician
(5) Use those recordings and speech measures as a focus of discussion with the clinician to deal with any emerging problems
(6) Devise a hierarchy of difficult speaking situations to assist generalisation
(7) Modify their measurement procedures for the coming week if needed.

Clients progress to Stage IV when stuttering and fluency technique goals are met for three consecutive weekly consultations

Stage IV: Maintenance of treatment gains

The goals of Stage IV are for clients to:

(1) Maintain target stutter-free speech during the clinic appointment
(2) Present target stuttering severity and fluency technique scores for typical speaking situations
(3) Present audio recordings of themselves to confirm these scores
(4) Discuss with the clinician how they have dealt with any problems that sustain treatment benefits.

Clients attend 1-hour clinic appointments that become less frequent according to progress. Discharge occurs when the client and clinician are satisfied that the client has developed self-management skills that are sufficient to sustain treatment gains. Commonly, clients have access to local self-help group meetings, which may be helpful during Stage IV.

Clinical trial evidence for the Camperdown Program

Phase I and Phase II clinical trials

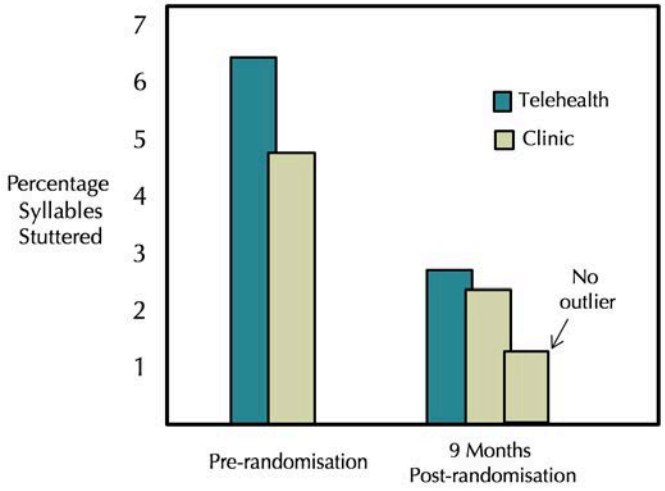
Using the Lecture Five definition of a clinical trial, there have been several supportive Phase I and Phase II trials of the Camperdown Program speech restructuring treatment model. Three trials involved a standard clinic treatment format,[53,43,54] (the latter with adolescents) and one reported results at a university student clinic.[55] Another was a low-tech telephone telehealth trial with adults,[49] and another was a video telehealth trial with three adolescents.[56]

One report involved an experimental version of a standalone Internet presentation of the treatment that did not require a clinician.[57] The results of this standalone Internet Phase I trial with two adults was encouraging. A subsequent trial of the standalone Internet version recruited 20 adults.[58] As with all clinician-free Internet treatment programs, compliance was an issue. Five participants completed the treatment and five completed more than half of it. Four of the five who completed the treatment reduced their stuttering severity by more than half, and two of those who completed more than half the treatment reduced their stuttering severity by an equivalent amount. Results were confirmed by participant reports of stuttering severity. These results suggest that standalone Internet Camperdown Program treatment may be a useful component of the stepped care approach to stuttering described during the previous lecture.

A Phase III clinical trial of telehealth

As mentioned during Lecture Six, there are some compelling advantages for telehealth treatment services with early stuttering. In the case of adults, there is the advantage for the many young adults who wish to reduce their stuttering for employment reasons, and are reluctant to take time off work for treatment. In such situations, telehealth treatment can minimise work disruption by reducing travel time to the clinic. Additionally, treatment within the client home maximises cultural and community support during treatment.[59] A study of telehealth assessments with 14 adults who stuttered[60] gave seven of them an in-clinic assessment and seven of them a video telehealth assessment. The 70-minute assessments were done by speech-language pathology students, and involved interview and formal testing procedures. Results showed that the two assessment methods were comparable in terms of time required, assessment results, and the client experiences of assessment.

A randomised Phase III trial[27] compared the standard clinic Camperdown Program presentation with an experimental, low-tech telehealth version presented by telephone. For the telehealth adaptation, "home practice replaced the face-to-face programme group intensive day" (p. 110).[27] The trial used a non-inferiority design, which establishes whether an experimental treatment variation is not inferior to the original. Twenty adult participants were recruited to each arm of the trial.



Results are presented in the figure above.‡ The telehealth group had more severe stuttering than the group that received the standard, in-clinic treatment. This can occur with small participant numbers, even though participants are randomised to each group. Three participants dropped out (8%) and their data were analysed by intention to treat with last observation carried forward (see Lecture Five). After treatment there was no difference in outcomes between the two groups, and in fact it is arguable that the telehealth group did better, considering that their stuttering was more severe pre-randomisation.

There is another reason to think that the telehealth group did better than the standard, in-clinic group. This is because of what is known as an outlier in that group. One of the in-clinic participants did not respond to the treatment at all. When the authors (controversially) removed that participant from the analysis, the results for the telehealth group looked even better.

The telehealth group required a mean of 10 hours 17 minutes for treatment, and the standard group required 12 hours 54 minutes. When these values were statistically adjusted for differences of pre-treatment variables such as stuttering severity, prior treatment and family history, the telehealth group

‡ Adapted and reproduced with permission: Carey, B et al (2010), Randomized controlled non-inferiority trial of a telehealth treatment for chronic stuttering: The Camperdown Program. *International Journal of Language and Communication Disorders, 45,* 108–120. © 2010 Taylor & Francis.

used 221 minutes less contact time—3.7 hours—than the standard group. That result was clinically significant and also statistically significant.

Speech naturalness assessment used a control group as a reference. There was a statistically significant result where both treatment groups had a mean speech naturalness score one scale value less natural than control speakers. In other words, to some extent, control of stuttering was attained at a cost of speech that sounded unnatural to some extent. These results were similar to those for a Phase I trial of the Camperdown Program.[43]

No replications

On balance, this body of clinical trial evidence might be interpreted as a sound data base attesting to the efficacy of the Camperdown Program model. However, there has been no independent replication of any of these results, and all the trials cited previously were from the same research group, albeit from researchers located in three different cities. As such, the results require cautious interpretation.

Speech restructuring II: The Comprehensive Stuttering Program

Overview

This evidence-based treatment model is outlined as a contrast to the Camperdown Program. It is a 3-week residential treatment that incorporates speech restructuring targets, programmed instruction, and a transfer phase. The Comprehensive Stuttering Program was developed during the 1980s[61] and is conducted at the Institute for Stuttering Treatment and Research[62] in Edmonton, Canada. It appears that a downloadable treatment manual is not available. However, a description of the treatment as it is currently conducted is available.[63]

Although the 3-week residential intensive treatment model can be adapted as needed, "it is the preferred format for the majority of clients" (p. 214).[63] This intensive format involves 90 hours of therapy with 6 hours per day. The Comprehensive Stuttering Program has three standard, formal phases: acquisition (instatement), transfer, and maintenance. The transfer phase involves a series of beyond-clinic speaking tasks, including speaking to strangers, telephoning businesses, shopping assignments, and group presentations.

The programmed instruction sequence begins at 40 SPM and with an eventual target of 190 SPM +/- 40 SPM. The speech pattern is taught using the following speech targets: "prolongation," "easy breathing," "gentle starts," "smooth blending," and "light touches" (p. 217).[63]

These speech targets are taught with constant attention to attaining natural sounding speech, and clients learn to use a 10-point speech naturalness scale, which is used during the treatment process. The treatment incorporates Van Riper's "stuttering modification" techniques ("stutter more fluently") that were described earlier.

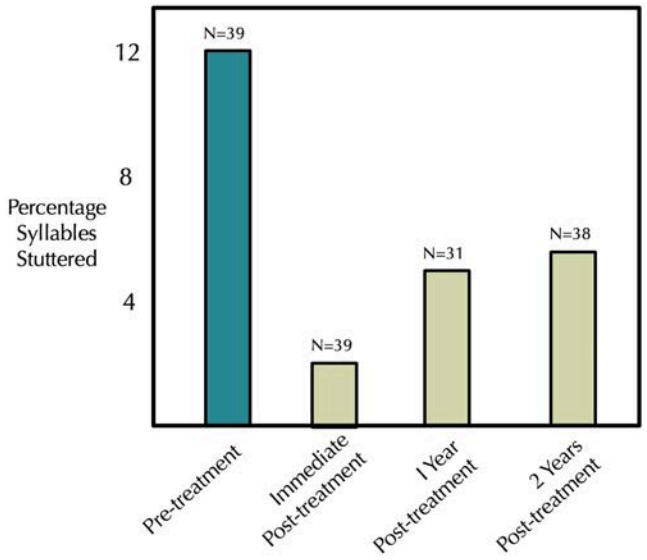
The Comprehensive Stuttering Program involves a substantive nonbehavioural component with cognitive behaviour therapy for each client (a psychological intervention: see Lecture Eleven). The Comprehensive Stuttering Program does not include any standard clinical psychology measures, and it appears that speech-language pathology staff, rather than clinical psychologists, administer the cognitive behaviour therapy component.

Clinical trial evidence for the Comprehensive Stuttering Program

As with the Camperdown Program, there has yet to be an independent replication of clinical trial results for this treatment. Phase I and Phase II clinical trials were published in the 1990s,[26,64] the former involving 42 participants, 17 of whom were adults and 25 were adolescent. Subsequently, a Phase II trial was reported[33] involving 39 participants, 14 of who were Canadian and 25 were Dutch. Although the trial purports to be for adults, the age range of the Dutch group was 17–53 years, and the range for the Canadian group was 15–42 years. So, a small, unknown number of participants were adolescents.

Speech measures were made pre-treatment, immediately post-treatment, and at 1 and 2 years post-treatment. At 2 years post-treatment there was only one drop-out from the study. Data were not

available for eight participants at 1 year post-treatment. Results are presented in the figure, with the Canadian and Dutch participants pooled.



Speech naturalness data were presented in this clinical trial, but are difficult to interpret. The Dutch participants' speech naturalness was measured with a procedure not common in stuttering clinical research: a 7-point bipolar naturalness scale.

The standard speech naturalness (NAT) measure was used for the Canadian participants, however participant speech samples were presented to listeners along with "140 other speech samples" (p. 238).[33] No indication was given of the nature of those speech samples and how they may have influenced NAT scores for the trial participants. With this reservation, the mean NAT score at 2 years post-treatment was 2.9, which certainly suggests natural sounding speech. However, mean speech rate data presented in the report (Table 1, p. 241)[33] are 135 SPM at 2 years post-treatment for the Dutch participants and 153 SPM for the Canadians. The target SPM range for the Comprehensive Stuttering Program treatment process is 190 SPM +/- 40 SPM, with "most clients … speaking at a rate of 150 to 190 SPM, which is on the lower end of the normal range" (p. 219). This might be interpreted to suggest that Table 1 (p. 241) of the report shows unusually slow speech and, hence, unnatural sounding speech.

In another report with one of the longest follow-up periods on record,[38] the same research group reported that at 10 years post-treatment 17 participants, when telephoned unexpectedly, showed that they had retained similar treatment effects to the participants who were studied at 2 years post-treatment.[33] No speech naturalness data were presented to bolster confidence in this result. However, self-report data indicated that

> at 10 years follow-up the majority of participants who responded reported that (1) they were generally satisfied with their current speech, (2) they had the ability to use techniques to control speech most of the time or more often, (3) their confidence in their ability to speak improved and (4) they had to pay attention to speech most of the time or almost always to be fluent. (p. 120)[38]

Speech restructuring III: Intensive smooth speech

A Phase II trial

Smooth speech treatment is a variant of speech restructuring that involves

> instruction on respiratory control, where easy, relaxed diaphragmatic breathing during speech is demonstrated. … another fundamental characteristic of smooth speech is the use of gentle onsets and offsets. This is achieved by starting the phrase with exaggerated airflow and using soft articulatory contacts. In addition, a phrase/pause speech pattern is taught. (p. 812)[65]

The treatment incorporates programmed instruction with speech rate increments from 50 syllables per minute (SPM) and targeting a final rate of 160–200 SPM. There are formal transfer, generalisation and maintenance phases. The maintenance phase lasts 12 months.

A Phase II trial[65] had three treatment arms and a control group, although it was not randomised. Two of the arms were smooth speech, both given intensively, one with parents present and the other without parents present. The trial was for children 9–14 years old.

For the smooth speech arm without parents present, participants received 5 hours of training prior to the treatment day to ensure that the requisite speech pattern had been learned. Then groups of 3–5 participants received intensive treatment for a week of 7-hour days. For the smooth speech arm with parents present, 2–5 parent-child pairs participated in 7-hour day groups, which were held once a week for 4 weeks. Parents were involved with the treatment process and did treatment at home on non-clinic days.

Data based on recordings beyond the clinic were not available for the standard clinician group, but were available for the clinician-parents group. For this group, 25 children were recruited, with a mean age of 10.5 years, and beyond-clinic pre-treatment stuttering severity of 10.9 %SS. Twenty-seven children were recruited for the standard clinician group, with a mean age 10.6 years. There were 20 children in the control group with a mean age of 10.9 years and pre-treatment stuttering severity of 8.8 %SS.

Results

It is not clear from the report whether there were any drop-outs. At 1 month post-treatment, the mean stuttering severity in the clinician-parents group was 2.9 %SS. Control group data were not available at 12 months post-treatment. Speech rate increased for the clinician-parents group from 131 SPM pre-treatment to 174 SPM at 12 months post-treatment. To further assess speech naturalness, the researchers argued that a 9-point scale was

> too difficult for the parent and child to make a distinct and easy judgment so it was adapted to a 5-point Likert scale, with 1 representing "poor," 2 "fair," 3 "moderate," 4 "good," and 5 "very good" speech naturalness. (p. 815)[65]

Unlike the original 9-point scale described during Lecture Four, this scale was accompanied by a detailed description of how to use it. The mean speech naturalness score for the children was 2.6 at pre-treatment and 3.5 at 12 months post-treatment, suggesting that the children may have attained reasonably natural sounding speech. This result, however, is difficult to interpret because no control group was available at 12 months post-treatment, and no methodological details are presented for how the speech naturalness scores were obtained.

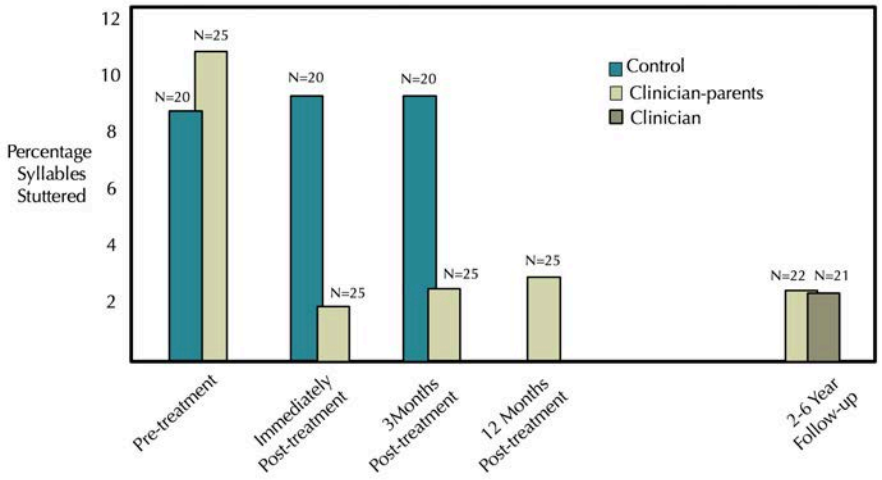
Anxiety reduction

The trial used the State-Trait Anxiety Inventory for Children[66] as a secondary outcome. Anxiety for the treatment groups showed significant change from pre-treatment to 12 months post-treatment. However, it appears that these gains were not clinically significant, because the children were in the normal range of scores pre-treatment.

Follow-up

A separate report for the trial was a 2–6 year follow-up[67] of 22 children in the clinician-parents group and 21 children in the clinician group. Treatment gains seemed to be retained in terms of %SS scores, with even faster speech rates than the original report, and better mean speech naturalness scores of 4.5. Results of the initial trial and follow-up are shown in the figure,[‡] with mean %SS scores for beyond-clinic assessments.

[‡] Adapted and reproduced with permission: Craig, A et al (1996), A controlled clinical trial for stuttering in persons aged 9 to 14 years, *Journal of Speech and Hearing Research, 39,* 808–826. © 1998 American Speech-Language-Hearing Association.



Speech restructuring IV: data-based reports

Two data-based case studies

It is of interest that a data-based case study[68] of a 1-week intensive speech restructuring treatment with fifteen 6–8 year olds showed clinically significant stuttering reductions in the clinic at 18 months post-treatment, and evidence of reasonably natural sounding speech. However, speech measures were not reported beyond the clinic. Another data-based case study without beyond-clinic data[69] contained six children ages 5–8 years. This was a 1-week intensive speech restructuring treatment, which used training with delayed auditory feedback. However, that report presented modest post-treatment stuttering reductions.

The Kassel report

A more comprehensive data-based report with children involved the Institute of the Kassel Stuttering Therapy in Germany.[70] The title of this paper refers to it as a "therapeutic trial," but that label may be questionable because of methodological issues discussed shortly. In broader terms, its status as a clinical trial may be questioned because it does not inform evidence-based practices in the sense of being replicable by clinicians in other settings. The report is not accompanied by a treatment manual or access to the requisite software. In the event that a clinician wishes a client to receive the treatment, the only option at present is to make a referral to this commercial program. The treatment is fully funded by the German health insurance system. Regardless, the substantive group of children and parents in this report makes it worthy of critiquing.

The treatment

The treatment is designed for children 7–9 years old. The format is intensive residential, with six to eight children and parents receiving 8 hours of treatment per day for 6 consecutive days, followed by three "weekend in-patient refresher courses" (p. 3) during a 6-month maintenance period. Presumably, those weekends also involve 8 hours of treatment per day. The staffing of the weekend courses is not specified, but the intensive phase involves "three therapists and perhaps one or two interns" (p. 30). The content of the refresher courses were not specified. However, the content of the 6-day intensive component is described as follows:

> In addition to specific speech trainings and exercises at the computer, speech games, activity games (preferably those which require verbal interactions), painting, short lectures on stuttering and speech physiology, the program includes exchanges of experience and leisure activities. (p. 3)[70]

Subsequent to clinicians describing the technique to the children, computer software "trains soft syllable onsets at the beginning of an utterance" (p. 3). Parents are required to "use soft syllable onset

all or most of the time, the child only in separate explicitly designated situations" (p. 3). Subsequent to the week of intensive training, the child uses the speech technique in structured situations at home, and then is encouraged to use it during naturally occurring speech situations.

Participants

The report involved 119 children, 108 boys and 11 girls, with a mean age of 8.0 years. The age range is not presented, but the authors state that around a third were younger than 7 years, with an unspecified number younger than 6 years and an unspecified number older than 9 years.

Outcomes

Percentage syllables stuttered was measured in the clinic by the treating clinicians at pre-treatment, after the intensive phase, and at the end of the 6-month maintenance phase. Measures were made in the clinic with brief "speech samples of at least 500 syllables (occasionally less, especially before treatment)" (p. 5). These methods do not justify classifying the report as a clinical trial based on the guidelines presented in Lecture Five. From pre-treatment to the end of 6-months maintenance, mean %SS scores changed from 9.4 to 5.6, which is a 40% reduction.

At 18 months and 3 years after the intensive phase, the mean %SS scores were 4.7 and 3.9. However, these data cannot be compared with the data collected earlier in the study because the methods of collecting the data changed. Although no methodological details were presented, it seems that clinicians telephoned the families at home for these later assessments.

From pre-treatment to 3 years after the intensive phase, OASES-S scores reduced from 2.4 to 2.0, which represents a change from "moderate" to "mild/moderate" impact of stuttering. However, as the authors point out, that result is difficult to interpret because the self-report OASES-S was designed for 7–12 year olds, but a third of the children were younger than 7 years.

Some caveats

This report presents some challenges when interpreting its results. It presents a unique health economics scenario for the discipline, because the treatment is funded entirely by the German health insurance system. In particular, the computer software for families is funded by insurance companies conditional on them conforming to requirements of the 6-months maintenance period. This makes it difficult to determine how the treatment might apply to other health care settings.

As the authors point out, speaking to clinicians during speech assessments probably biased the data in favour of a treatment effect because of discriminated learning. This is why the usual method to measure %SS with children is for parents to record their children conversing in everyday situations. In that context, it is difficult to interpret the clinical meaning of a 40% reduction of measured stuttering severity.

In fact, the report provides no information about the extent to which the children generalised the use of "soft syllable onsets" and how their speech sounded to listeners when using them. The authors did conduct a speech naturalness evaluation of pre-treatment and post-treatment speech samples using a standard speech naturalness scale (see Lecture Four: 1 = highly natural, 9 = highly unnatural). The authors concluded that, overall, "the speech of the treated children did not become as natural as the speech of non-stuttering children" (p. 9).

Ultimately, judgments about the merits of the Kassel program need to be considered in the context of the resources it involves. The treatment is designed to alleviate stuttering with "soft syllable onsets," but there seem to be no treatment progression criteria based on success in attaining that goal. Consequently, all children receive an entire treatment package involving them and a parent for 12 eight-hour days: six days during the intensive phase and three weekends during maintenance. Based on 6–8 children in an intensive group, with 3–5 clinical staff, this would involve 50–60 clinician hours per child treated. The value of using so many family and health care resources needs to be evaluated in light of the evidence presented for their benefits. Its value also needs to be evaluated in the context of clinical trials of treatments that have involved 6–9 year olds and documented outcomes in a fraction

of the clinical time required than for the Kassel treatment. Those treatments are presented during Lecture Nine.

Speech restructuring V: Video self-modelling as a supplement

The procedure

Self-modelling is positive behaviour change based on people observing themselves being free of a problem behaviour. It has been shown useful for managing various problem behaviours,[71] and in theory can be used as an additive to improve the effects of any stuttering treatment. Clinicians can make a video recording of clients speaking without stuttering, using the speech restructuring technique. Then, simply, clients are instructed to watch the videos regularly.

Basic research

The mechanisms by which self-modelling might work are unclear, but there has been a suggestion that it relates to self-efficacy and self-belief.[72,73] With stuttering, two single-subject laboratory experiments[74,75] showed promise for children and adolescents, with encouraging results continuing at 2–4 years follow-up.[76] Subsequently, a laboratory study[77] was conducted with three adults using an experimental single-subject design. The researchers made self-modelling videos of the participants by having them repeat any stuttered utterances until they were stutter-free, and then editing the stuttering moments from the videos. During the experimental condition participants were instructed to speak the way they did on the videos. One participant showed clinically significant stuttering reductions under that condition.

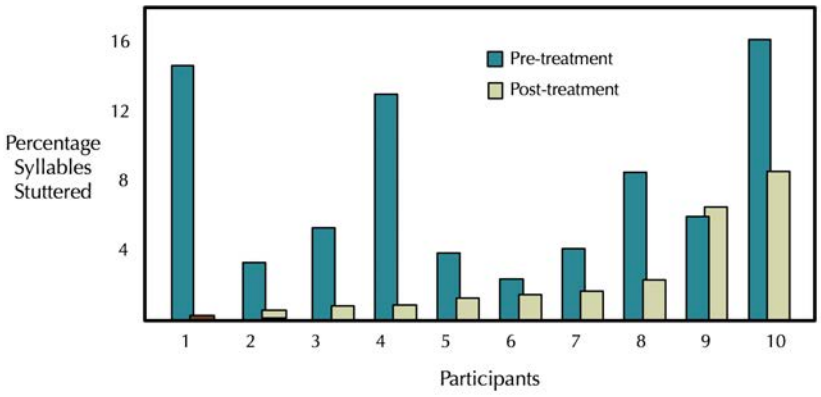
A data-based case study of video self-modelling: Relapse management

Design

A case study report[78] explored the potential value of video self-modelling with the common relapse problem after speech restructuring stuttering treatment. The study recruited 12 adults who had received speech restructuring treatment but had relapsed. The researchers gave them a 1-hour clinical session during which the participants re-established speech that was stutter-free and as natural sounding as possible. For each of the participants, three 5-minute videos were constructed that contained no stuttering. The mean NAT score for them was 3.8. The participants were asked to watch the videos once per day for 1 month. Twelve participants were recruited and two (17%) dropped out.

Results

Results are presented in the accompanying figure,[‡] which shows pre-treatment and post-treatment stuttering severities immediately after the 1 month of watching the videos. Participants 1–8 restored their stuttering severities levels to those that are associated with successful speech restructuring treatment



[‡] Adapted and reproduced with permission: Cream, A et al (2009), Self-modelling as a relapse intervention following speech-restructuring treatment for stuttering, *International Journal of Language and Communication Disorders, 44,* 587–599. © 2009 Taylor & Francis.

outcomes. Participant 10 reduced severe stuttering by around half and one participant did not respond at all. The mean post-treatment NAT score was 3.9, which was comparable to the result for another speech restructuring treatment by the same research group,[43] suggesting that the regaining of stuttering control required some compromise of speech naturalness.

Clinical applications

This finding suggests that with just an hour of clinical time spent, clinicians can successfully manage clients who have relapsed after speech restructuring treatment. Additionally, the technique could be used with the intention to prevent the occurrence of relapse. This is potentially far more efficient than the common "booster" or "refresher" sessions that are reported in descriptions of speech restructuring treatments,[63,70,79,80,81] where clients return to the clinic to receive a substantial portion of their treatment to restore their speech benefits, or to provide resistance to relapse occurring.

A Phase III trial of video self-modelling: Speech restructuring supplement

Design

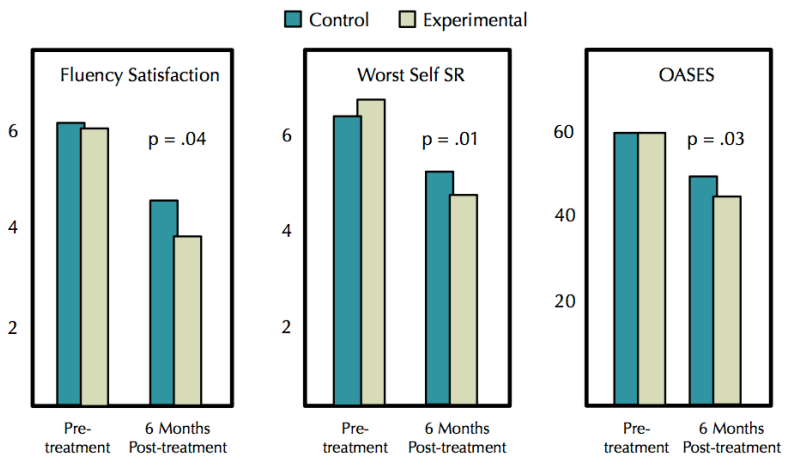
The Phase I trial results of video self-modelling led those researchers to explore whether building video self-modelling into a speech restructuring treatment process might improve outcomes. So, 89 participants, 64 adults and 25 adolescents, were recruited into a Phase III trial.[30] One arm involved standard speech restructuring and the experimental arm involved standard speech restructuring plus video self-modelling. The participants were treated at different clinics around Australia, using different speech restructuring treatment models.

Towards the end of the treatment, the researchers made 4.5-minute self-modelling video recordings of all clients. Clients in the experimental group were given their self-modelling video and instructed to watch them for 5 minutes each day for a month, with the following instruction: "Try to talk without stuttering using your speech technique as you see yourself doing on the video" (p. 890–891).**Error! Bookmark not defined.** Five participants (6%) dropped out, and their results were analysed with intention to treat analysis by last observation carried forward.

The primary outcome was %SS measured from unscheduled telephone calls to participants from strangers. There were several secondary outcomes: Subjective Units of Distress[82] scores for anxiety after the telephone calls, self-rated SR scores for eight nominated speaking situations, avoidance of those eight speaking situations, satisfaction with fluency, and impact of stuttering measured with the Overall Assessment of the Speaker's Experience of Stuttering (OASES) [83] (see Lecture Four).

Results

Three significant results were found: fluency satisfaction, improvement in the speaking situation self-rated most severely, and the OASES. The OASES scores for the experimental group dropped from moderate impact at pre-treatment to mild-moderate impact at post-treatment. Results are presented in the figure below. The paper does not present data separately for adult and adolescent participants, although the authors state that there were no differences in outcomes between the two age groups. So, the figure, although it represents adults and adolescent results combined, is likely to be reasonably representative of the adult group results.



There can be some flexibility with incorporating the results of the two trials of video self-modelling into clinical practice. For example, a clinician may require clients to watch self-modelling videos daily for a month after treatment and then systematically withdraw how often they watch them until they can manage to sustain their treatment benefits without any watching. Then, in the event of impending relapse, a client could return to daily watching. Only in the event that a return to daily watching failed to prevent signs of impending relapse would a client need to come to the clinic for further consultation.

A clinical experiment

Participants in this experiment[84] were three adults who had received an intensive, residential speech restructuring treatment.[63,85] They were studied with a multiple baseline across participants experiment, which is one of many experimental designs used with individuals. Prior to the experiment, their stuttering severities were 6.4 %SS, 7.7 %SS and 16.2 %SS. Each of them was given two 4-minute self-modelling videos, which they were instructed to watch at least twice per week for 5 weeks. Stuttering severity during the 5-week experimental period was measured each week with three recordings, one of which was independent of the clinic. The researchers reported that two of the participants reduced their stuttering severity. One participant reduced %SS scores by around one fifth, and another by around a third. However, the data are difficult to interpret because the reported %SS scores were an amalgam of within- and beyond-clinic measures. All three participants reported that the self-modelling procedure benefited them: "the most robust improvements in the self-report data were reduced avoidance behaviors followed by reduced expectancy to stutter" (p. 39).[84]

SUMMARY

Adults

According to clinical trials, speech restructuring is the most efficacious speech treatment for persistent stuttering during adulthood. For some adults, despite its many limitations, the treatment is clearly capable of controlling stuttering to a clinically significant extent and for a clinically significant period. Yet currently, without randomised controlled evidence, nothing is known about effect sizes that might be expected. There is convincing evidence that video self-modelling is a useful adjunct to speech restructuring treatment.

Adolescents

For adolescents, speech restructuring has the strongest supportive clinical trial evidence of all available treatments for that age group, with independent replications. There is evidence that video self-modelling can be useful with this age group. Arguably, considering the potential benefits against the little effort required to implement a video-self modelling procedure, it should be a routine adjunct

during and after a speech restructuring treatment process with adolescents. This is discussed in more detail at the end of the next lecture. It is also necessary to note that the major drawback of this style of treatment—speech that sounds and feels somewhat unnatural—may be a particular clinical issue with adolescents.

School-age children

For school-age children, the evidence for speech restructuring is less compelling than it is for adolescents, with only one substantive clinical trial reported,[65] which is without independent replication. This clinical trial contains participants 9 years and older. Consequently, there is no clinical trial evidence that younger children in the age range 6–8 years would benefit from the treatment. It is of interest, however, that there is a data-based case study of intensive speech restructuring with 6–8 year olds.[68] As with adolescents, speech that sounds and feels unnatural is likely to be a particular clinical issue with this age group.

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LECTURE NINE: OTHER EVIDENCE-BASED SPEECH TREATMENTS FOR PERSISTENT STUTTERING

Syllable-timed speech

Method

This treatment[1] is an adaptation of the Westmead Program for early stuttering discussed during Lectures Six and Seven. Two early reports suggested that the procedure may be useful with older children.[2,3] Another report, mentioned during Lecture Six,[4] exposed a group of 9–11 year olds to a metronome beat and their stuttering decreased without any instructions.

The treatment has two stages. During Stage I children use syllable-timed speech to establish a low level of stuttering, and the purpose of Stage 2 is to maintain those treatment benefits for a clinically significant period. During Stage 1 the children come to the clinic with a parent for 30–45 minute appointments, during which they learn and practise syllable-timed speech. The child is taught to use the speech pattern to sound as natural as possible throughout these clinic appointments. During the appointments parents are instructed to praise their children for using the speech pattern, and to remind them to use it if they lapse to a customary speech mode.

When the child is able to sustain the use of syllable-timed speech during a conversation, parents are instructed to practise the technique with the child for 5–10 minutes, 4–6 times per day. When this is achieved parents are instructed to prompt their children to use the speech pattern at random times each day, at which time clinic appointments occur fortnightly.

The treatment incorporates the SR scale, and the child is admitted to Stage 2 if %SS in the clinic is less than 1.5 and SRs for each day are 0–1 for two consecutive fortnightly clinic appointments. During Stage 2 children are required to maintain these speech targets for 10–12 months with a performance contingent maintenance program.

A Phase I trial

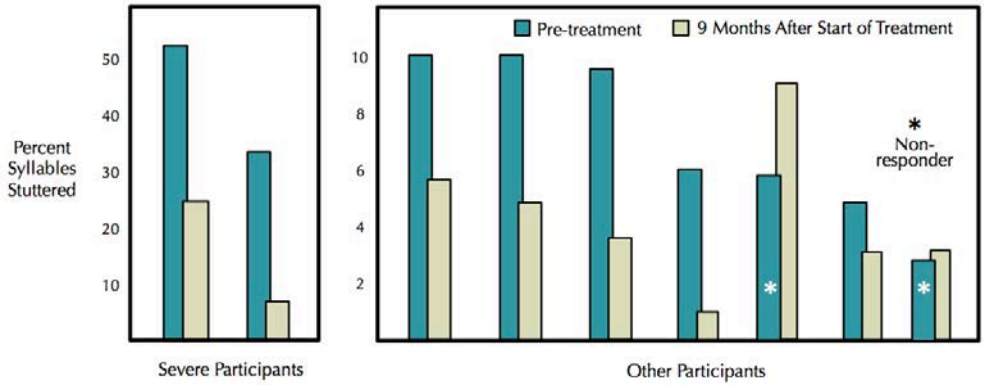
Ten children were recruited to a Phase I trial,[1] ages 6–11 years. One child dropped out from the treatment. Outcomes were measured at pre-treatment and at follow-up 9 months after the start of treatment.

Results

Stuttering severity

Two children did not respond to the treatment. Of the seven children who responded to the treatment, five reduced their stuttering by more than 50% at 9 months follow-up, and two children showed large reductions of 81% and 87% at 9 months follow-up. Mean reduction for the nine children was 54%. Results are shown in the figure.[‡] Two severe children are on the left and the remaining seven are on the right.

[‡] Adapted and reproduced with permission: Andrews, C et al (2012), Syllable-timed speech treatment for school-age children who stutter: A Phase I trial, *Language, Speech, and Hearing Services in Schools, 43*, 359–369. © 2012 American Speech-Language-Hearing Association.



Speech naturalness

To assess speech naturalness, the researchers obtained 15-second stutter-free speech segments from the children at follow-up, and played them to 10 unsophisticated listeners. The listeners were instructed to write down words or sentences to describe the children's speech. On only one occasion listeners gave any suggestion of a speech pattern change that might be associated with syllable-timed speech, with a report that one child sounded "mildly monotonal" (p. 365).[1] However, that child was one of those who did not improve.

Speech satisfaction and stuttering impact

For the seven children who completed the treatment, their mean speech satisfaction dropped from 6.0 pre-treatment to 2.7 at follow-up. Their impact scores on the OASES scale for this age group dropped from a mean of 54 to 40, showing a change from moderate to mild-moderate impact.

Verbal response contingent treatment I: Self-imposed time-out

The procedure

Lecture One reviewed the extensive laboratory evidence of the controlling properties of response contingent stimulation, and Lecture Six outlined the Lidcombe Program model of verbal response contingent stimulation for treating early stuttering. A model of verbal response contingent stimulation that is suitable for adults, which has supportive clinical trial evidence, is known as *self-imposed time-out*. This means that when a stuttering moment occurs, the client stops speaking for a few moments, then resumes speaking. The duration of that period of self-imposed time-out seems not to matter.[5] All that seems to matter is that it is contingent on stuttering.[6] Research participants who use this technique generally choose quite a brief time-out period.[7]

Clinical advantages

The advantage of the self-imposed time-out technique is that it does not use an overt speech pattern. Additionally, there is reason to believe that it might invoke existing speech skills, such as those learned with speech restructuring.[8,9] And in fact, three of four participants in a laboratory report[8] who had severe stuttering, and who attained more than a 60% stuttering reduction with self-imposed time-out, had received previous speech restructuring treatment.

Clinical trial evidence for self-imposed time-out

Phase I trials

The first clinical trials of verbal response contingent stimulation with adults were Phase I trials[10,11] according to the definition in Lecture Five. These reports demonstrate potential value of the technique with single-subject experimental designs, each report using one participant.

A "regulated breathing" technique, to be reviewed shortly, appears to focus mostly on self-imposed time out, although it includes several other components. Drawing on a clinical experiment with 21

school-age children and adolescents who stuttered,[12] a single subject experiment was performed with a 9-year old and a 14-year old boy.[13] (A variation of this technique has been reported in single-subject experiments with five children aged 5–11 years[14] and with eight 6–10 year-olds.[15] However, neither of these reports contained beyond-clinic data. The variations included "exhale slightly before beginning to speak on a natural exhalation of air [p. 296].[14])

A randomised Phase II trial

A treatment described as habit reversal[16] was originally developed to treat "nervous habits and tics" and was later applied to adult stuttering clients in a randomised Phase II clinical trial,[17] described as regulated breathing. The treatment is multidimensional, but appears to focus mostly on self-imposed time out:

> In order to regulate breathing, the client was instructed to stop speaking when a stuttering episode occurred and to take a deep breath by exhaling and then inhaling. (p. 41)[17]

Forty adults were randomised to five treatment arms, four being a variant of the regulated breathing procedure, and a fifth being a self-monitoring placebo. It is difficult to interpret a five-arm trial with only eight participants per group, but modest stuttering reductions of around 50% were reported.

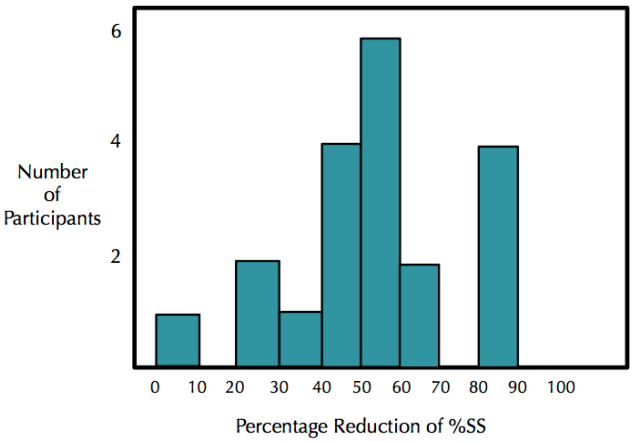
A nonrandomised Phase II trial

The last trial of self-imposed time out to date[18] was a non-randomised Phase II trial that recruited 30 participants, 26 of whom were adults and four were adolescents. The trial involved instatement, generalisation and maintenance phases of treatment. Twenty-two participants completed the first two phases, and 18 of these completed maintenance and remained in the trial at 6 months post-treatment. Therefore, the trial was affected by a high drop-out rate of 40%.

The instatement phase began with the clinician first imposing time-out, and then the client learning to self-impose it. Subsequently, participants learned to use self-imposed time out during everyday speaking situations and to self-evaluate their speaking performance. A subsequent maintenance phase assisted participants to sustain their treatment gains.

The mean reduction of %SS scores for the 22 participants completing the instatement and generalisation phases was 54% from pre-treatment to 1 week post completion of those phases. That result was sustained for the 18 participants for whom data were available at 6 months post completion of the instatement and generalisation phases.

As shown in the accompanying figure,[‡] more than half of the participants reduced their stuttering by more than 50%, and four of them reduced their stuttering by 80–90%. The report indicated that there was no difference in responsiveness between the adults and the four adolescents in the trial. The figure shows the percentage reductions for 22 participants at the conclusion of the instatement phase.



Speech naturalness for participants was assessed with the NAT scale, with the result that "around half of the participants achieved post-treatment speech naturalness scores

[‡] Adapted and reproduced with permission: Hewat, S et al (2001), Control of chronic stuttering with self-imposed time-out: preliminary outcome data, *Asia Pacific Journal of Speech, Language and Hearing, 6,* 97–102. © 2001 Taylor & Francis.

within or near the range of control subjects" (p. 38).[18]

Clinical applications

The verbal response contingent treatment model seems to have some treatment effects with persistent stuttering, but it seems not to have the strong effects that appear to occur for early stuttering. It is fairly clear that the clinical trial evidence for them is, on balance, neither as comprehensive nor compelling as it is for speech restructuring. However, it is arguable that the evidence is strong enough to suggest that they will have clinical value for some adults and adolescents.

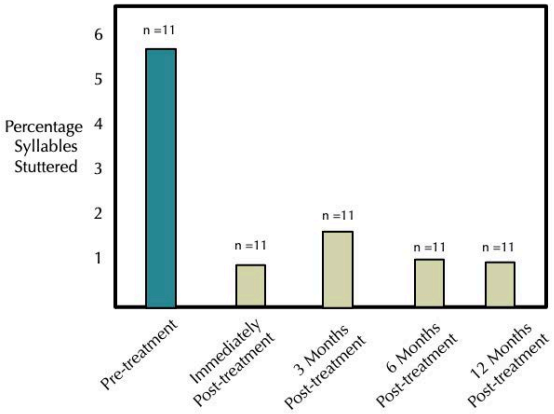
The most obvious disadvantage of the treatment is its limited clinical trial evidence base. And, although self-imposed time-out is clearly less effortful for clients than speech restructuring, it is not clear exactly how much effort clients must use to sustain benefits during everyday speech. It is unknown at present, for example, what proportion of stuttering moments must receive time-out for a clinical effect and whether it needs to be sustained to retain the effect. The Phase II trial merely states "the majority of participants reported that they were using time-out only 'sometimes' 6 months after therapy" (p. 40).[18]

Based on available clinical trial evidence, a verbal response contingent treatment model might not routinely be the first treatment of choice for adults and adolescents. There are, however, some situations where a clinician might consider them the first treatment of choice. Examples include when a client is unable to learn a speech restructuring pattern, or cannot sound natural enough, or has a long history of unsuccessful treatment with speech restructuring, or simply does not like the speech restructuring technique.

Verbal response contingent treatment II: The Lidcombe Program

A Phase II trial

Although the Lidcombe Program was developed for children younger than 6 years, there has been one Phase II clinical trial of the treatment for children older than that.[19] Fifteen children were recruited to the trial and four dropped out. The remaining children had a mean age of 8.3 years with a range of 6.10 years to 12.4 years.



Results

The trial showed a stuttering reduction of 89% from a mean pre-treatment stuttering severity of 5.3 %SS. A mean of 15 hours of treatment was required, with a median of 12 hours, to reach Stage 2. The data are presented in the figure. The report contained no speech naturalness assessments, perhaps with the assumption that speech naturalness would not be an issue with a verbal response contingent stimulation treatment.

These results are consistent with a follow-up of eleven 6–10 year old children who were treated with the Lidcombe Program.[20] The children were telephoned three times during one week at a mean of 70 weeks post entry to Stage 2, with a range of 9–187 weeks. At that follow-up their mean %SS score was 1.9.

Verbal response contingent treatment III: GILCU

Method

Another model of verbal response contingent stimulation is Gradual Increase in Length and Complexity of Utterance, commonly referred to as GILCU. This well known treatment program has been detailed in two editions of a text.[21,22] The fundamentals of the procedure were first described in

1965[23] and variants of it have been described elsewhere.[24,25,26] The developer of the GILCU model spent many decades training clinicians in the United States to use it.[27] It has also been used in the United Kingdom,[28] Germany,[29] and Hong Kong.[30] GILCU has been discussed in several publications.[27,31,32] The treatment is designed for children.

The GILCU program begins with a programmed instruction Establishment phase containing many steps, beginning with the child being required to speak a word without stuttering and moving up through a series of steps up to 5 minutes of reading, monologue, and then conversational speech. It qualifies as a model of verbal response contingent stimulation because the contingency for stutter-free speech is praise, sometimes paired with redeemable tokens. The prescribed verbal contingencies for stuttering are (surprisingly) "stop" and "speak fluently" (p. 64).[25]

Parents are trained to identify stuttering and to implement home practice. Branching steps are included to provide additional remedial training when a child doesn't succeed at any step. The procedure contains transfer and maintenance phases, during which verbal contingencies are systematically withdrawn.

A Phase I trial

There has been only one report by the developers of the treatment that qualifies as a Phase I clinical trial according to the Lecture Five definition. The trial[33] recruited four children, one of whom dropped out. It is not clear from the report whether any of the children were adolescents. Probably, though, most were in the school-age range because the four children had a mean age of 11.3 years.

Results

Participants had a mean pre-treatment stuttering rate of 5.9 stuttered words per minute, and at 9 months follow-up the three that remained had a mean score of 1.0 stuttered words per minute, for an 83% stuttering reduction. For the Establishment phase, a mean of 9.6 hours of treatment was required, 6.4 hours for the Transfer phase, and 2.0 hours for the Maintenance phase. The report contained no speech naturalness assessments, perhaps with the assumption that speech naturalness would not be an issue with a GILCU procedure.

Concluding comments

A total of 208 participants of all ages have been reported in GILCU data-based clinical studies.[32] Yet only three of those participants[33] met a reasonable clinical trial requirement of beyond-clinic speech measurement with a follow-up period. It is therefore probably a reasonable observation that the GILCU clinical trial evidence base is not well advanced, even though "the programme has been in use for 40 years" (p. 228).[32] Another GILCU report[25] appears to be a clinical trial with a 14-month follow-up of 6 children. However, clinicians collected those data in the clinic. Beyond-clinic measures were obtained during the study, but the latest were at the end of the Transfer phase, without a follow-up period.

Hybrid treatments I: The Oakville Program

Method

The researchers who published the Phase I trial of syllable-timed speech for children[1] sought to improve the modest, and inconsistent, result in that trial of mean stuttering reduction of around 50%. They proposed that a way to do that might be to add parent verbal contingencies to the treatment.[34]

The hybrid treatment is essentially the same as the syllable-timed speech treatment,[1] except that during Stage 1, after syllable-timed speech practice is introduced at home, parent verbal contingencies for stuttering and stutter-free speech are introduced.

A Phase II trial

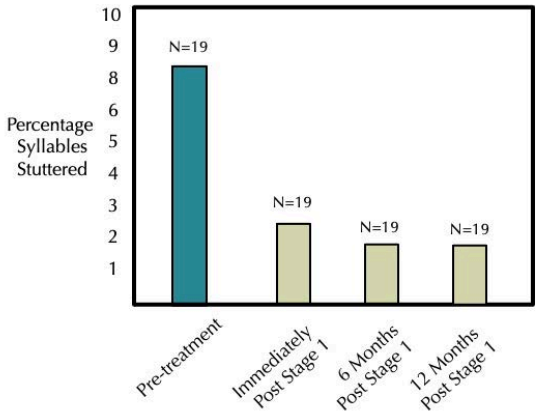
Twenty-two children, aged 6–11 years, were recruited to the trial,[34] 16 of whom were boys and six were girls. Fourteen of the children had previous treatment, and nine had comorbid disorders,

predominantly with speech and language. Outcomes were measured pre-treatment and 6 and 12 months after completion of Stage 1.

Results

Stuttering severity

Three children withdrew from the trial before completing Stage 1, leaving 19. Results are presented in the figure.[‡] The mean reduction of %SS scores at 12 months post Stage 1 completion was 77%. The mean %SS score at 12 months post Stage 1 was 1.9, with a range of 0.2–5.6 (SD=1.6). At each assessment the children reported their typical stuttering severity in each of the eight situations used in the Phase I trial.[1] The group mean pre-treatment typical stuttering severity (not presented in the figure) was 5.4 and 1.9 (SD=1.2) at 12 months post Stage 1.



Impact of stuttering

At each assessment the children completed the Assessment of the Child's Experience of Stuttering, which is an earlier version of the OASES-S (see Lecture Four). The mean pre-treatment score was 50.6 and 33.9 at 12 months post Stage 1. This change represented an improvement from moderate to mild-moderate impact.

Post-treatment speech

To determine if listeners could detect any signs of speech rhythmicity post-treatment, 10 seconds of stutter-free speech pre-treatment and 10 seconds post-treatment was selected for each child. A group of listeners was asked to score each sample with a five-point scale where 0 = *not at all rhythmic speech*, and 4 = *extremely rhythmic speech*. Results showed no sign that the children were speaking with any detectable rhythm post-treatment.

A treatment process study

With a separate study,[35] the same research group reported speech process data for four children, ages 8–11 years, who received the Westmead Program. As expected, the syllable-timed speech treatment reduced variability of vowel duration post-treatment. However, there was no compromise post-treatment in terms of speech rate, measured with articulation rate, and language use, measured with utterance length and complexity. For one child, perceptual judgments by observers suggested some effects of rhythmicity post-treatment.

Hybrid treatments II: DELPHIN Speech Treatment

Method

The methods of this German treatment for children and adolescents are outlined in the one available English report about it.[36] Although it is not fully clear, the treatment seems to incorporate elements of speech restructuring:

> The first goal is the acquisition of costo-abdominal breathing ... Then the patient is taught in one-on-one-sessions what Schültz calls the "deblocking impulse," a

[‡] Adapted and reproduced with permission: Andrews, C et al (2016), Phase II trial development of a syllable-timed speech treatment for school-age children who stutter. *Journal of Fluency Disorders, 48,* 44–55. © 2009 Elsevier.

> kind of sigh, a letting go. The deblocking impulse is formed without pressure. It leads to a relaxation of the region around the larynx and impedes a cramping of the false and true vocal folds. This deblocking impulse should be used in every syllable with the main accent … Approximately, 5 days later, the second step follows. Now the patient learns what Schültz calls "nasal swinging accent": a gentle voice onset with nasal character in words beginning with a vowel. The nasal character facilitates a soft voice onset. (p. 159)[36]

There is no treatment protocol available to provide more precise details about this treatment, but it incorporates progressive muscular relaxation, and the unusual inclusion of

> … European drum sessions to foster sense of community and to improve concentration and coordination skills. In contrast to African drum playing, where the right hand is mainly used, here the player alternates from one hand to the other after each beat; both halves of the brain are activated and are said to be better connected. During the drum sessions, the speech technique is used, and for some patients the movement of the hands facilitates the speech technique. (p. 160)

A Phase II trial

Fifty-six participants were recruited with a median age 13.0 years, 42 of whom were boys. Inexplicably, a small but unknown number of adults were included, with an age range of 8–36 years for participants. Participants were treated in groups of 7–10. The treatment duration details are not clear apart from the fact that it was resource intensive: "during the intensive therapy, a patient attends 140 sessions (mostly group sessions)," and "during the 2 years after the intensive therapy, the patient has maximally 2 weeks and four single days for the stabilisation phase, usually 50–80 logopedic sessions" (p. 160).[36] Various speech measures were collected at pre-treatment and at various times up to 12 months post-treatment, and one of those measures involved a telephone call to participants outside the clinic. The Strengths and Difficulties Questionnaire[37] was used to assess emotional and behavioural problems pre-treatment and post-treatment.

Results

Stuttering severity

It is difficult to interpret the results of %SS scores for pre-treatment and post-treatment telephone conversations, because they were based on short samples of 250 syllables. The pre-treatment mean was 12.7 %SS and the post-treatment mean was 4.3 %SS.

Speech naturalness

A five-point scale was used to evaluate whether the parameters "prosody," "breathing," and "tonus during phonation" were "normal" or "markedly deviant." Although improvements were reported, it is not possible to determine the extent to which participants sounded natural at post-treatment.

Strengths and Difficulties Questionnaire

Improvements were reported from pre-treatment to post-treatment. However, it is not possible to attribute those improvements to the speech treatment component or to non-speech treatment components (progressive muscular relaxation and group based activities such as "drum sessions").

MACHINE AIDED TREATMENTS

Background

Potential benefits

For obvious reasons, a machine that could do stuttering treatment automatically without a clinician, or that could produce clinically significant stuttering reductions when worn, would be well worth having, because the clinician would have to do little, if anything. And if the machine was inexpensive and portable, clients could do their treatments when and where they chose. And if the machine reduced

stuttering when it was worn, it would simply obviate the whole problem of persistent stuttering for the wearer during that period.

A questionable history

For these reasons, credible attempts to develop machine driven treatments are welcome. The history of clinical stuttering research contains many attempts to establish machine driven treatment,[38] and unfortunately, on many occasions these attempts have been accompanied by commercialism without sufficient evidence for clinical efficacy. The more memorable devices that could be worn by those who stutter are the electronic metronome and the Edinburgh Masker.

Both these devices relied on two of the fluency enhancing conditions mentioned during Lecture One. The electric metronome was a device resembling a monaural hearing aid that produced a metronome beat to the ear. The Edinburgh Masker presented a masking signal during speech. This device involved a throat microphone held to the outside of the larynx with a strap, a headset, and a masking unit carried on a belt or in a pocket. In the long run, though, the value of these devices was never determined by a clinical trial, and they appear to be no longer clinically available and not used much in clinical practice, if at all.

Altered auditory feedback

A topic of current interest

The fluency enhancing condition called *altered auditory feedback* (see Lecture One) is delayed auditory feedback plus an alteration in pitch upwards or downwards. There has been much basic research about altered auditory feedback that has established a place for it as one of the fluency enhancing conditions. Around the same amount of basic research has been done about it as for time-out, although the effects of altered auditory feedback are not as reliable across participants as is the case for time-out. A recent laboratory study[39] reviews much of that literature.

SpeechEasy

Altered auditory feedback has emerged as a current controversial topic in stuttering research with the commercialisation and advertising of a device known as SpeechEasy.[40] It resembles a monaural hearing aid and presents altered auditory feedback to the wearer. One source of controversy about it emerged in a publication[41] in which the device was cited as an example of pseudoscience. Predictably, a vigorous exchange followed in the literature.[42,43] During the course of that exchange the developers of the device disclosed a financial interest in it.[42]

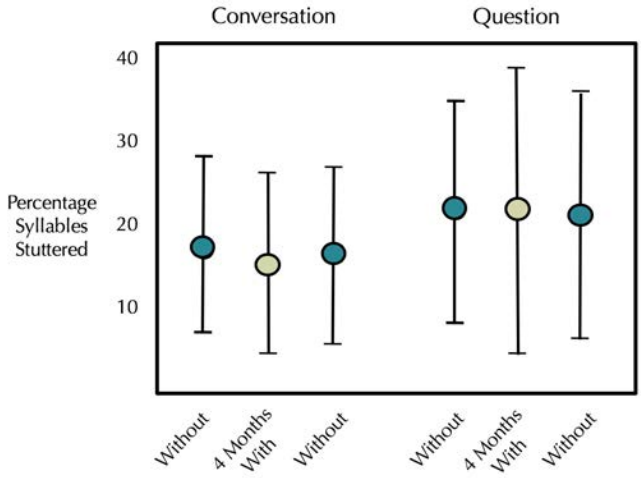
A Phase I trial of SpeechEasy

The clinical trial evidence for altered auditory feedback is not particularly encouraging. The only clinical trial that has been published is a Phase I trial of the SpeechEasy device by a group that is independent of its developers.[44]

Eleven adult participants were recruited and fitted with the device, wearing it for 4 months with a guideline to wear it for at least 5 hours per day. A little publicised feature of the device was documented in this trial. Namely, that it is not intended as a standalone device, but is intended to be combined with features of speech restructuring. Participants were instructed to use

> several active techniques to alter one's speech pattern, such as easy vocal onsets, prolongations, continuous phonation, starter sounds … and fillers. Participants were told that these active strategies could be introduced at their discretion to help initiate voicing and/or enhance responsiveness … (p. 520)[44]

Participant speech was measured beyond the clinic on three occasions while conversing and asking questions of a stranger: pre-treatment, during the 4 months of wearing the device, and during a withdrawal phase without the device. Results are summarised in the figure,[‡] with the means showing no effect from wearing the device. The error bars are the 95% confidence intervals.



Differing interpretations of the trial

The researchers who did the trial did not recommend moving on to a Phase II trial. The developers of the device protested vigorously,[45] claiming that the clinical trial was methodological flawed. The researchers who conducted the trial retorted by saying there was nothing wrong with their methods.[46]

A subsequent report

The developers of the SpeechEasy device then published a report[47] described as a "randomized clinical trial" with 18 adult Brazilian Portuguese participants diagnosed with stuttering. One group of 11 participants received SpeechEasy treatment for 6 months "with no training to use any fluency enhancing techniques" (p. 772). Another group of seven participants received treatment involving

> both fluency shaping and stuttering modification techniques—negative practice, smooth speech, resisting time pressure and use of voluntary disfluencies. In addition to the practice of these speech motor skills, the treatment programme emphasized self-observation and included systematic cognitive and attitudinal intervention. (p. 772)[47]

Pre-treatment and 6 months post-treatment assessments occurred, but the results of this report are uninterpretable because both groups showed only a 40% reduction of stuttering severity, and measures were based solely on 200 syllables of "monologue speech, conversational speech and oral reading" (p. 772) from within the clinic.

Modifying phonation intervals

The opposite of speech restructuring?

This device uses an ingenious idea that seems to be the opposite of speech restructuring. Rather than adding speech pattern features to control stuttering, it takes some away. What it takes away are short phonation intervals. It does this by means of a throat microphone held in place with a band, a signal processing box, and a display on a laptop computer. The laptop display indicates the proportions of phonation intervals within certain duration ranges, and gives the speaker feedback on attempts to reduce the number of phonation intervals within certain ranges. The potential of the device is supported by several laboratory research reports.[48,49,50] It is of interest that a report[51] showed four of seven clients who received a standard speech restructuring treatment reduced frequencies of short phonation intervals post-treatment. So, regardless of how successful modification of phonation intervals may prove to be as a treatment, it seems that modification of phonation intervals is not necessary for stuttering reduction.

[‡] Adapted and reproduced with permission: Pollard, R, et al (2009), Effects of the SpeechEasy on objective and perceived aspects of stuttering: A 6-month, Phase I clinical trial in naturalistic environments, *Journal of Speech, Language, and Hearing Research, 52,* 516–533. © 2009 American Speech-Language-Hearing Association.

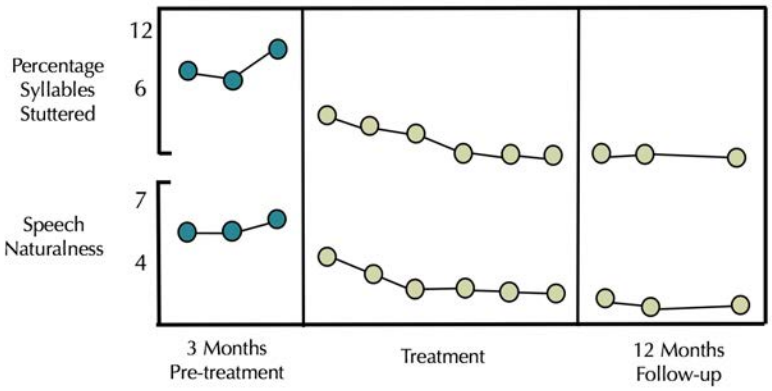
The treatment phases incorporated programmed instruction requiring the client to speak without short phonation intervals. The machine fails participants at any step for not meeting phonation interval criteria. The modifying phonation intervals program contains instatement, generalisation, and maintenance phases. The instatement phase is intensive, requiring six 3-hour treatment days per week, for a period of from 3–12 weeks, depending on the rate of client progress.

Two data-based case studies

There have been no clinical trials published for modifying phonation intervals treatment. However, after the initial laboratory research findings, two data based case studies were published.[52,53] They are data based case studies rather than clinical trials because outcome measures within and beyond the clinic were a part of the treatment: 3-minute conversations for which the number of syllables was not reported. In other words, as part of their treatment, clients were trained to speak without stuttering in situations that were used to evaluate treatment outcome.

The first report[52] was five adult participants studied with a single subject experiment. The following figure shows mean %SS and NAT scores for the five participants during a 3-month pre-treatment period. It also shows scores for the treatment period, which involved 2 weeks of instatement and 8 weeks of generalisation. Subsequently, data are presented for a 12-month follow-up period. The figure shows one of several treatment data sets presented by the authors, based on telephone conversations within the clinic, without any feedback from the machine. Treatment speech measures beyond the clinic yielded similar results.

The figure[‡] shows that %SS and NAT scores were quite high during the pre-treatment period. The high NAT scores would be expected with the presence of clinically significant stuttering. The %SS scores reduced to clinically significant levels with almost no stuttering during treatment, and NAT scores stabilised at what appears to be within the normal range. These results were sustained during a 12-month follow-up period.



The second data-based case study[53] compared 10 participants who received an intensive speech restructuring treatment with 17 participants who received the modifying phonation intervals treatment. Surprisingly, five of the participants in this second report were participants in the first report.[52] Consequently, it is not possible to ascertain mean outcomes for the 12 novel participants in the second report. Additionally, it is not clear whether the MPI software and hardware were functionally similar in the two reports, or whether the five participants in the original report 14 years earlier used a different apparatus.

Regardless, the authors showed that there was no significant difference between the speech restructuring group and the modifying phonation intervals group at 12 months follow-up. It seems likely that the outcomes for the 12 novel participants in the second report attained similar outcomes to the five participants in the initial report, with the exception that they did not sound as natural, according to scores with the 9-point NAT scale.

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‡ Adapted and reproduced with permission: Ingham, R J et al (2001), Evaluation of a stuttering treatment based on reduction of short phonation intervals, *Journal of Speech Language and Hearing Research, 44,* 1229–1244. © 2001 American Speech-Language-Hearing Association.

Awaiting a clinical trial

As yet, no clinical trial of the treatment has been published. The treatment is provided as a clinical service by certified United States clinicians who are trained at the University of California, Santa Barbara. The modification of phonation intervals software and hardware is available only for purchase by certified clinicians.[54]

Noninvasive brain stimulation

Noninvasive brain stimulation is a term describing clinical methods "to modulate the excitability of the brain via transcranial stimulation" (p. 173).[55] The two methods are transcranial magnetic stimulation and transcranial direct current stimulation.

Transcranial direct current stimulation "involves application of a weak electrical current across the head via electrodes placed on the scalp, modulating the resting membrane potential of neurons in the underlying cortex" (p. 2).[56] The technique has been used with mixed results for a range of conditions, particularly psychiatric illnesses, and for cognitive enhancement with healthy participants. This treatment development was spurred by applications of the technique to rehabilitation of limb and speech motor function with stroke patients. There have been feasibility studies with stuttering.[57,58,59] There is a detailed review of eight studies of the technique applied to stuttering reduction in adults.[60]

There has been a case study report of the treatment as an adjunct to speech treatment.[61] A randomised clinical experiment was reported[56] with the technique as such an adjunct. Thirty participants were randomised to receive either five consecutive days of 20-minute speech treatments with transcranial direct current stimulation, or five consecutive days without that adjunct. Participants who did not receive the transcranial direct current stimulation received a sham treatment involving a dosage that was presumed to be ineffective. The 20-minute speech treatment sessions comprised a sequence designed as a hierarchy of increasing difficulty: chorus reading with a clinician, chorus reading with recorded speech, syllable-timed speech with monologue, and syllable-timed speech in conversation.

The primary outcome was change in "percentage of disfluent syllables" from pre-treatment to 1 week post-treatment, and from pre-treatment to 6 weeks post-treatment. At each assessment, the measure was generated from clinic speech samples of reading and conversation. The first 2 minutes of each sample was analysed. Thirty participants received treatment after randomisation. The control group showed no change. Overall, for the experimental group there were changes from pre-treatment percentage disfluent syllables scores: 27% reduction at 1 week post-treatment and 22% reduction at 6 weeks post-treatment. However, at 6 weeks post-treatment, although gains remained for the reading task, the percentage of disfluent syllables score for conversation had returned to pre-treatment levels. Perhaps controversially, the authors concluded that "transcranial direct current stimulation combined with behavioural fluency intervention can improve fluency in adults who stutter" (p. 1161).[56]

Electromyographic (EMG) biofeedback

Method

In short, an electromyography (EMG) machine uses surface electrodes to detect muscle action potential as muscles contract, and displays it visually. Clients attempt to change muscle action potential using such feedback. The procedure is used to treat a number of tension-related disorders and found its way into basic stuttering research with several promising laboratory reports.[62,63,64,65]

The trial of speech restructuring with 9–14 year olds described earlier, with and without parents present, involved an experimental arm with EMG biofeedback:

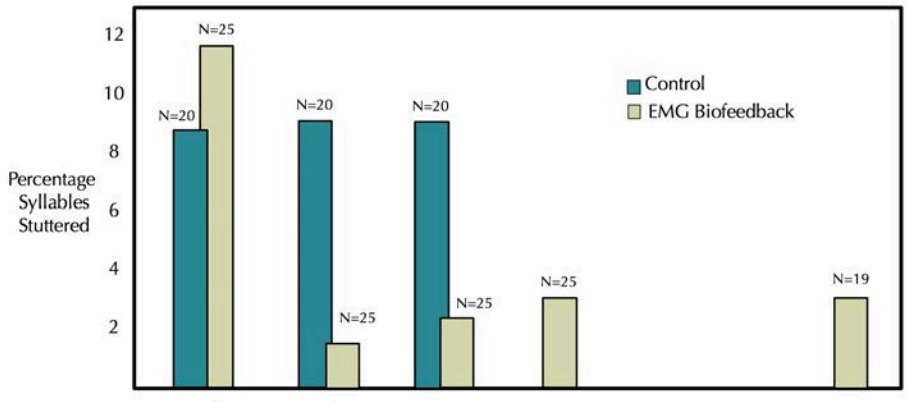
> All children had their own computer so that speech muscle activity was monitored simultaneously … if the child's muscle tension was too high a high-pitched sound would occur, indicating that the child should relax the speech muscles. Initially, the children were taught to raise and lower their muscle tension without speaking (around 2 hours). Once the child was able to distinguish differing levels of muscle tension … he or she was required to

perform the same task without observing the screen or being able to hear the sound.[65]

Subsequently, the children followed a programmed instruction sequence lasting two days, starting with words and finishing with conversational speech. The programmed instruction included with and without EMG biofeedback during speaking. On the third day the children attempted speaking without the EMG biofeedback and were able to enter a transfer phase when they could speak without stuttering and without biofeedback.

A Phase II trial

The arm with EMG biofeedback in this trial[65] involved five intensive 6.5-hour days. As with the other arms of the trial, it is not clear whether there were any drop-outs. Results in the following figure,[‡] including follow-up,[67] show that the 5-day intensive treatment with the feedback produced results equivalent to those produced with intensive speech restructuring. At pre-treatment, mean stuttering severity was 11.5 %SS and at 12 months post-treatment was 2.9 %SS, showing a 75% stuttering reduction. The mean clinician pre-treatment speech naturalness score with the 5-point scale described previously was 2.6, and at post-treatment was 4.6, again suggesting that speech was reasonably natural sounding.



Two failures to replicate

The methods of the original trial[65] were replicated in another non-randomised trial[66] using identical equipment and following the biofeedback treatment manual.[67] However, results failed to replicate the original result. Participants were 10–16 years old, which was a similar age range to the original study. Eleven of 12 participants recruited finished the study. Results showed only a modest mean stuttering reduction of 37% in conversations outside the clinic after the treatment.

Another non-randomised trial[68] again failed to replicate the positive findings of the original trial.[65] Three adolescents, one aged 13 years and two aged 15 years, were treated with a direct replication of the original study, again using identical equipment, and following the biofeedback treatment manual. Treatment effects were modest again, with %SS reductions for the three participants while talking on the telephone outside the clinic as follows, from pre-treatment to post-treatment: 12.8 to 9.1, 3.7 to 3.0, and 32.3 to 29.8.

[‡] Adapted and reproduced with permission: Craig, A et al (1996), A controlled clinical trial for stuttering in persons aged 9 to 14 years, *Journal of Speech and Hearing Research, 39,* 808–826. © 1998 American Speech-Language-Hearing Association.

Pharmacological treatments

A long search

Since the 1960s, there has been a search for a pharmacological stuttering treatment, and a large number of clinical trials have been conducted for various compounds. These include anticonvulsant agents for treating epilepsy, antidepressants, antipsychotic agents, cardiovascular agents, and dopamine antagonists.

Two reviews

A 2006 review of this topic[69] identified 31 reports that met what the authors determined to be minimum methodological requirements. The authors identified trials using the above mentioned pharmacological compounds and concluded that

> The results of this systematic review of pharmacological treatments for stuttering are straightforward and are overwhelmingly negative. Of 31 studies reviewed, only 11 met three or more of five basic methodological criteria. Of those 11 plus 4 other relatively well-designed articles, only 1 provided data to show that stuttering was reduced to below 5%, the lenient outcome criterion selected for this review … One other provided data to show that stuttering was reduced by at least half … (p. 348)[69]

The authors concluded their review with a statement that there is no evidence that anyone who stutters has ever benefited from drug treatment and that it is unlikely that anyone ever will. Some other authorities in the field agreed with them.[70]

A more recent review[71] of drug trials for children and adolescents, extending back to the 1960s, included two older papers and one more recent paper that were not covered by the previous review. Seven papers were identified that met prescribed methodological criteria. Only one paper was found that the authors felt was methodologically strong enough to constitute reasonable evidence, and it found that a cardiovascular agent had no effect on stuttering. The authors concluded with the suggestion, that, surprisingly, never seems to have been thought of before: that drug treatment for stuttering might be improved if combined with standard speech-language pathology interventions.

A subsequent report[72] did evaluate the effects of olanzapine and haloperidol when added to a speech-language pathology treatment. However, the treatment was far from "standard," comprising "mixed treatment sessions that included 'air flow technique' and 'break Valsalva maneuver (sic.)' as well as 'desensitization' from Van Riper's protocol" (p. S271). This, plus the absence of contemporary speech measures, and side effects "such as mild drowsiness, dry mouth, and lethargy" make the results of the trial not compelling.

The search for a drug treatment continues, with a constant stream of preliminary reports. For example, there has been a preliminary study of ecopipam,[73] a dopamine receptor antagonist, with 10 adults who were studied during 8 weeks of using the drug. Stuttering severity reductions were measured from 900–2,300 syllables of within-clinic speech. Some stuttering reductions, and reductions of OASES scores, did occur, but arguably were not sufficiently large enough to be clinically significant. An intriguing report contained a suggestion that green tea improved the speech and psychological status of adolescents who stuttered compared to controls.[74]

In short, at present there is no reason to consider drug treatment for stuttering. Regular reports continue to emerge about the effects of various drugs on stuttering, but they continue to not conform to accepted standards for a clinical trial in this field.

Group-based residential support

A report documented the benefits of a 2-week residential Camp SAY for childen who stuttered aged 8–18 years.[75] The camp is not formal therapy focused on stuttering. Its purpose is to promote "social interaction with people who stutter as a means of achieving positive change for the participants" with activities including "theater, swimming, arts and crafts, horseback riding, and sports" (p. 20). Campers have an opportunity to meet with a speech-language

pathologist once or twice per week for additional support. OASES scores were collected on the first day of camp, the last day of camp, and at 6 months follow-up.

There were small but statistically significant changes of OASES mean Impact Scores, which were sustained at follow-up. The changes may not have been clinically significant, with all means remaining within the mild-moderate impact score range. Regardless, as discussed during the next lecture, speech-related social anxiety is likely to be a clinically significant issue for this age group, and the effects of this style of intervention on that aspect of childhood stuttering require future documentation.

Some clinical notes about persistent stuttering

Adolescents as a unique clinical group

A life transition

Clients in the age range 13–17 years are a unique group. The adolescent—teenage—years are a transition from childhood to adulthood and are accompanied by changes not experienced at any other time of life. Clinical challenges associated with adolescence arise from changes during this period that span physical, cognitive, emotional and social domains. These changes can impact the client and the family,[76,77] and can be a significant consideration when planning and implementing treatment.

Adolescents can be thought of clinically as neither children nor adults.[78] Many adolescents will experience stress at some period during this time of life, which needs to be taken account of clinically, but many will not.[79,80] Other common features of adolescence that may need to be accounted for clinically are the emergence of a sense of independence and autonomy,[76,81,82] developing importance of the peer group,[79,83] and a decline of motivation.[84,85,86,87,88] The emerging importance of the peer group is associated with a need to fit in to the norm,[77] which can cause problems if an adolescent stutters.

Parents during treatment

A report of how 13–17 year old adolescents interacted with their parents about stuttering[89] is useful information for clinicians who treat this age group. An important finding was that some of these adolescents made the decision to attend a speech clinic themselves, and for others it was the parents who motivated them to do so. Most of the adolescents reported that they found parent assistance with their stuttering to be helpful, but it was clear that unsupportive parent input might occur. They commonly reported that helpful parent input

> involved the use of speech skills, where parents would remind participants to use certain speech strategies, provide advice, or sometimes practice speech skills with participants. Also, participants reported parents providing assistance in the form of listening and not interrupting them when talking. (p. 50)[89]

However, the minority who reported unsupportive parent input found it to be “frustrating and perceived in an unsupportive manner” (p. 50).

Telehealth and adolescents

For adolescents, there are particular advantages to telehealth. Telehealth treatment, which appears viable for adolescents,[56] can give them the independence that is important to them. If they wish, their parents need not be involved in treatment at all. Parents, too, can benefit from telehealth treatment of their adolescent children. For example, they do not need to take them to and from the clinic. In particular, video telehealth presents treatment within an adolescent-friendly medium that facilitates client empowerment and self-management. Finally, the Internet for social purposes, using laptop, tablet and smartphone devices, are now part of the lives of most adolescents who have access to such technology.[90] Treatment methods for this age group can readily include technology[91] such as the Scenari-Aid website.[92]

A preliminary trial considered during the previous lecture,[93] which showed positive treatment outcomes, revealed some useful information for this treatment format with adolescents. Naturally, with

so few participants conclusions must be guarded, but there was some suggestion that the video telehealth format was a better prospect for these adolescents than those in the other Phase I trial[54] who were treated in a clinic.

At another source describing this telehealth trial,[94] one adolescent participant commented that for treatment sessions she did not need to "race home from school and get ready and go somewhere" and found treatment "comfortable ... because I was in my own house and in a more familiar place." Another said treatment was easier because he could "just hop up on my computer instead of going to the clinic." His father said, “he was just so relaxed. I think this is a big call but he is probably more relaxed with [his SLP] than he is at the school he goes to, and with his teachers. This is a big winner."

School-age children and clinical tractability

The evidence for changing tractability

Clinical trials of early intervention compared with clinical trials of adult intervention indicate that stuttering is at its most tractable shortly after onset and at its least tractable during adulthood. Effect sizes are larger for early stuttering than for adults, novel speech patterns are not required as part of efficacious clinical management, and there are far fewer signs of relapse than with adults. The adolescent clinical trials considered during this lecture provide no reason to believe that adolescent clients are more clinically tractable than adults. Hence, it seems possible that a change in tractability might occur during the primary school years.

A Phase III clinical trial of the Lidcombe Program for children with early stuttering[95] shows no sign that %SS outcomes are affected by age. Table 5 (p. 662) of this report shows that outcomes were the same for the 28 children in the trial younger than 4 years as for the 19 children older than 4 years. So, there was no sign of changing treatment responsiveness during the period of early stuttering.

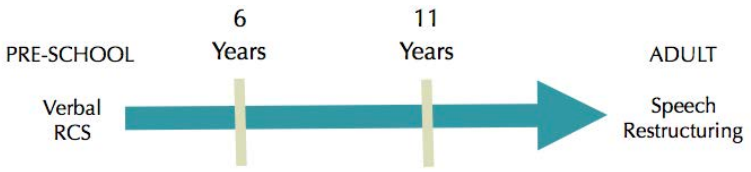
However, it seems to be a different story with the retrospective follow-up of children previously treated with the Lidcombe Program when they were 6–10 years old.[20] One of the children in the report, Participant 4, did not to respond to the treatment, apparently because of compliance issues. For the remaining children, aged 6–10 years, in Table 2 (p. 284), analysis shows a significant, moderate negative correlation between age in months when treatment began and the percentage reduction of %SS scores at follow-up (r = -.72, p = .012).[†] In other words, there is evidence that increasing age during the school years is associated with decreasing treatment effect sizes at follow-up. For the 11 children in this report, around half of the treatment effect at follow-up can be accounted for by age at the time of treatment (r^2 =.52).

So, to summarise, verbal response contingent stimulation is suitable and efficacious for children with early stuttering, but speech restructuring is usually suitable for adults, and there is reason to believe that responsiveness to verbal response contingent stimulation decreases with age during the school years.

A treatment selection model

The information above can be incorporated into a treatment selection model for school-age children. The model would, with increasing age, have verbal response contingent stimulation becoming progressively less suitable during that time of life, and speech restructuring becoming progressively more suitable. The model is presented in the figure.

[†] This analysis is not reported in the paper.



The model suggests verbal response contingent stimulation as a first intervention of choice for school-age children, because it is a simple treatment and does not require a novel speech pattern. The last treatment of choice for school-age children would be the more complicated speech restructuring technique, with its associated disadvantages, supplemented with video self-modelling.

Considering the clinical importance of the school-age time of life for stuttering, with its apparent changing clinical tractability, it is lamentable that clinical research to guide clinicians with this age group is so sparse. Reviewers of interventions for this age group[96,97,98,99] have not been able to identify a consistently efficacious treatment for them. The gravity of this situation prompted the editor of an international speech-language pathology journal dealing with school-age children to issue a call to rectify the situation urgently,[100] and more than 100 researchers and clinicians endorsed that call.[101]

Adaptation of the Lidcombe Program for school-age children

The model of treatment selection for school-age children in the diagram above may prompt clinicians in some circumstances to use the Lidcombe Program with that age group, considering that there is encouraging clinical trial evidence of its value for them.

However, some adaptations to the clinical process are necessary when using the Lidcombe Program with school-age children. The language used to present verbal contingencies will be different, as perhaps will the activities for presenting verbal contingencies during practice sessions. It is also realistic to expect children to participate more actively in the treatment, such as engaging with parents and the clinician in scoring and recording SR scores, and using self-imposed contingencies such as spontaneous self-correction.

One group of Australian, American, and Canadians who use the treatment with school-age children comment as follows:

> Although school-age children's interests are often captured by the latest fad toys, we still find that more traditional games, toys, books, magazines, comics and catalogs continue to be useful as stimulus materials. (p. 153)[102]

The authors of that report recommend that parent SR scores are used during the treatment, and that if the child contributes SRs they are used only as a supplementary source of useful information during treatment. They also recommend that during clinic appointments there is open discussion with the child about the types, frequency, and wording of verbal contingencies that will be used. They remark that school-age children generally prefer that verbal contingencies not be used in the presence of their peers. They also state that, because reading aloud in class is a routine part of school life, it is often useful to include reading aloud without stuttering as a therapy task. Additionally, they say token rewards are more likely to be useful with this age group, although tick charts may be sufficiently motivating in many cases.

A common source of trouble with adapting the Lidcombe Program to this age group is the limited contact parents often have with children. The authors of the article[102] suggest that in some cases, when it is appropriate, an older sibling, grandparent, or relative may be able to contribute to the treatment, providing that such a person attends all clinic appointments.

Teachers and school-age children who stutter

The classroom

Lectures Ten and Eleven deal with the common association between stuttering and social anxiety. Not surprisingly, then, there are several sources of anecdotal and research evidence that implicate fear of

speaking in the classroom as a potential issue for stuttering primary school children.[103,104,105,106] A common clinical picture of a school-age stuttering child is one who is quiet and withdrawn, reluctant to participate in classroom activities, and is constantly anxious about being called on to speak in class. As expressed in an early report:

> In school, he generally sits in the rear of the class, rarely initiates discussion or answers questions spontaneously, and he avoids most situations which might provoke the slightest fear of stuttering. Even though he may be intellectually superior to most of his classmates, he minimizes his own potentialities, capacities, and gifts by remaining silent and not risking the possibility of a stuttering effect. (p. 141)[107]

The importance of teachers

That early report[107] noted the importance of early school experiences because they often represent the first excursion into the world by children who stutter without daily parent contact. Accordingly, teachers can be critical personnel in the lives of children who stutter, particularly during the school years. If a stuttering child is anxious in the classroom and feels that it is a dangerous and threatening place, a teacher can make the classroom feel much safer. Participants who were interviewed as young adults made it clear that the importance of the classroom experience extends into adolescence.[108] An adaptation of the OASES (see Lecture Four) for carers (OASES-C) reported that teachers of children 2–6 years old associated stuttering with a mild-to-moderate impact.[109]

How might teachers help

A useful teacher approach to fear of classroom speaking seems to have been originally suggested in 1940.[110] This paper contained sensible advice to teachers of not suggesting to children any techniques for controlling stuttering. Instead, the teacher can confidentially discuss with a child how help might be offered in the classroom, and together they can formulate a strategy for handling the matter. The interview study mentioned previously[108] indicated that such a constructive, individualised approach rarely occurred for the participants. A report of New Zealand teachers suggested that they have limited knowledge of strategies to assist students who stutter.[111]

It might be expected that anxiety about reading aloud in class can worsen for children who stutter when the class takes turns to speak. Apprehension about speaking, and quite often physiological signs of anxiety, can build steadily. Such anxiety about an impending classroom speech has featured in interviews of adults recounting school experiences, with this one being particularly informative:

> If I thought there was a teacher that would randomly pick kids to read or would go down the row and everybody gets a turn, I'd have my mother talk to them and once again explain my situation, so that I did not have to read in class because any time they started that my ears would get hot, I'd start getting nervous, I couldn't sit still, I just started to sweat, and the only thing I could think about was counting down the time until I had to read. (p. 77)

> I used to go upstairs to the second floor bathroom and just keep flushing the toilet so that nobody could hear me getting sick, and then that kind of physical behavior lasted with me a long, long time because as a young adult and as an adult whenever I had a speaking situation coming up I would get physically ill. (p. 78)[105]

An account by United States President Joe Biden is similar.[112] A common sense approach here would be for the teacher to ask the child for a preference about speaking order. For example, if the child wishes to speak toward the start of the order and has a name towards the end of the alphabet, the sequence of children speaking could occur in reverse. Or, the teacher could call on children randomly, with the exception of the child concerned, who is called on at an agreed time. Or, if the child is sitting towards the back or the front of the class, the speaking could be done in order of seating position.

A comprehensive review[113] examined the empirical evidence for common recommendations about how teachers can assist with school-age students who stutter. That review pointed out that many

recommendations have been made to call on students to speak early during the class to reduce anxiety. The review points out that there is no direct empirical evidence about the merits of that approach. However, some laboratory experiments of wait-time to speaking with stuttering participants suggest that it is justified. Based on these experiments, the review presents the caveat that the student "should not be the initial speaker, or should not read in the earliest position" (p. 9).[113]

The review describes two additional approaches to children who stutter in the classroom: anti-bullying interventions and giving a presentation about stuttering. A report[114] showed that an anti-bullying school program, involving 4 hours of teaching with manuals and videos, could positively influence peer attitudes and bullying for school-age students who stutter. Another report[115] suggested that a 45-minute presentation about stuttering improved attitudes about stuttering, although participants were adolescent students. A subsequent report found that improvements were retained 7 years later.[116] Positive results were reported with a 9-year-old boy who included a classroom presentation about stuttering in his treatment,[117] and by speech-language pathologists who gave a classroom presentation as part of treatment for a 10-year-old girl.[118]

Some modern resources

Some modern suggestions about how teachers might help children who stutter in the classroom are available.[119,120,121,122] A video production by The Michael Palin Centre in London promotes teacher awareness of stuttering,[123] and is available at their website.[119] This video is a useful resource for clinicians who have contact with teachers of children who stutter. Clinicians may also direct parents to it so they can show it to the teacher of their stuttering school-age child. An overview of how speech-language pathologists might assist school age clients with bullying includes teacher involvement.[124]

Summary

Adults

There is clinical trial evidence that self-imposed time-out may be a treatment option in some cases of adult persistent stuttering. There is clinical trial evidence that the SpeechEasy device has no clinical value. However, there is promising preliminary evidence for another machine-based treatment: modification of phonation intervals. There is no reason to consider pharmacological treatment for stuttering control with any client (of any age).

Adolescents

EMG treatment cannot be considered without further clinical trials. There is almost no evidence that any of the verbal response contingent stimulation treatment models are suitable for this age group. There is no evidence for syllable-timed speech treatment with adolescents.

School-age children

A prominent finding for stuttering treatment in school-age children is a positive non-randomised Phase II clinical trial of the Lidcombe Program model of verbal response contingent stimulation.[19] One caveat to that, however, is that there is no independent replication of the finding at present. Another caveat is that the Lidcombe Program treatment model was developed for young children, and so is unlikely to be completely suitable for older children. The obvious clinical advantage of a verbal response contingent stimulation treatment model for school-age children is that it does not require a speech pattern. Consequently, post-treatment speech naturalness probably will not be a clinical issue.

There is a Phase II non-randomised trial of syllable-timed speech for school-age children.[34] On balance, it seems that the results of the trial are not as promising as for the Lidcombe Program with that age group. In contrast to the Lidcombe Program trial for school-age children,[19] the overall effect size appears to be more modest, and two children did not respond to the treatment at all. On the positive side, syllable-timed speech is by far the most procedurally simple of treatments for school-age children. Further, any treatment effects associated with syllable-timed speech for this age group seem

not to be associated with unnatural sounding speech. Clearly, further clinical trials are needed to establish information for clinicians about the relative merits for school-age children of syllable-timed speech and the Lidcombe Program.

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LECTURE TEN: STUTTERING, SOCIAL ANXIETY, AND MENTAL HEALTH[†]

Background

A changing view about stuttering and anxiety

Past decades

Research perspectives about stuttering and anxiety have changed during recent decades. A review of the topic at the close of the 20th century[1] showed, that during the mid 1980s, prominent scholars of the day agreed that there was little convincing evidence of a relationship between stuttering and anxiety.[2,3,4]

However, a follow-up review of the area 10 years later[5] showed that things had changed. Continued research, with methodological improvements, allowed the authors of the review to conclude that there is compelling evidence of a relationship between stuttering and anxiety. The authors of the review drew attention to mounting evidence of clinically significant anxiety levels associated with stuttering, and evidence that adults who stutter may—but not necessarily—experience psychological problems related to anxiety. They reported progress in clinical management of anxiety with those who stutter, and new ways to measure it clinically.

More recent views

Another decade later, in 2021, there was evidence of research in the field detecting anxiety early in the lives of those who stutter. A systematic review and meta-analysis of the topic[6] concluded that "the summary effect size indicates that children and adolescents who stutter present with increased anxiety symptoms … compared with non-stuttering peers" (p. 1).

Accordingly, research about stuttering and anxiety reviewed in this section has profoundly influenced modern clinical practices. In 2021, the views of 12 scholars in the field[7] included the need for core clinical assessment covering domains related to speech anxiety: "(d) reactions to stuttering by the speaker; (e) reactions to stuttering by people in the speaker's environment; and (f) adverse impact caused by stuttering" (p. 2379).

Anxiety

Expecting harm

Generally speaking, expectancy of harm drives anxiety. Examples of exceptions to this generalisation include innate infant anxiety states, such as fear of separation and fear of strangers. It appears that anxiety involved with stuttering focuses on anticipation of harm in social situations, where speech is required. Examples of such harm would be social rejection, being laughed at, or being ignored. In fact, one report showed expectation of such social harm to specifically be the issue; those who stutter appeared to be troubled only by such expectations, not expectations of any physical harm.[8]

Three components

Anxiety is commonly described as a complex psychological event composed of verbal-cognitive, behavioural, and physiological components.[1,9,10,11] Clinical psychologists group the following cluster of emotions as related to anxiety: scared, shy, panicky, and insecure.

[†] Thanks to Ross Menzies and Lisa Iverach for guidance with this material.

Verbal-cognitive

Anxiety commonly includes thoughts and expectancies about negative, harmful events. The prevailing psychological perspective is that, with the exception of some evolutionary anxiety responses, such as fear of heights, water, and spiders, emotions come from thoughts that emerge in response to events.

The kind of harm that might be expected by those who stutter in social situations is fear of negative evaluation. They may perceive social situations and performance based situations, such as addressing a group of people, as threatening and dangerous. They may have a fear that listeners in such situations will form some kind of a negative view about them, and that negative view will cause them harm in some way, such as humiliating or demeaning them.

Naturally, most people will experience social anxiety in some situations, particularly when speaking to a group of people. However, at some point, anxiety about such situations becomes clinically problematic because it interferes with usual enjoyment of life activities. The destructive and unhelpful thoughts that can drive the social anxiety of those who stutter are well known and discussed shortly. Examples include thoughts that "people will wonder what's wrong with me if I stutter" and "people will think I'm strange."

Behavioural

People who are socially anxious may avoid social situations to some extent, or to use the common expression, they may be "socially avoidant." They may also avoid situations that focus on speech performance, such as talking to a group of people. Or, in extreme instances, they may show escape behaviours once they are in such situations by fleeing from them.

Physiological

Those who stutter and are anxious about social situations may endure them with considerable distress, which could, but not necessarily, be manifested with physiological symptoms. They may experience symptoms such as sweating, blushing, increased heart rate, heart palpitations, hyperventilation, dry mouth, shortness of breath, nausea, headache, shaking, and muscular tension. Those physiological symptoms can prompt cognitive symptoms such as mental blocking, difficulty concentrating, and feeling flushed. During research, physiological anxiety responses can be measured with salivary cortisol, skin conductance, blood pressure, respiration rate, and heart rate.

Loosely connected components

Clinical psychologists think of the above three anxiety components—verbal-cognitive, behavioural, and physiological—as being loosely related to each other.[10] They do not systematically increase or decrease together in a lawful way. From one anxiety-provoking situation to another, one of them may increase while another may decrease or not change. They will not necessarily all be present when someone is anxious. This feature of anxiety is well known in clinical psychology and has been shown to be the case with those who stutter.[12,13] The latter of those reports involved participants speaking to a virtual audience and a virtual empty room. Although a measure of the verbal-cognitive anxiety component (Subjective Units of Distress; see Lecture Eleven) was elevated in the former condition, stuttering severity, heart rate, and skin conductance showed no changes.

One study of 13 adults who stuttered and 15 controls[14] showed elevated skin conductance and lower respiratory sinus arrhythmia in the stuttering group. Respiratory sinus arrhythmia is heart rate fluctuations linked to breathing that occur naturally; during inspiration, heart rate increases and during expiration heart rate decreases. Lower respiratory sinus arrhythmia is associated with social anxiety. One of two self-reports of anxiety in the study showed an association with respiratory sinus arrhythmia.

In particular, clients who stutter may be distressed by destructive thoughts and beliefs about negative social evaluation, but will not necessarily have any signs or history of situation avoidance, escape, or physiological arousal. It is quite possible for someone who stutters to always enter feared situations, and experience distress when doing so, but without any physiological signs. Probably, with most

clients who stutter, the prominent sign of anxiety is what they will tell clinicians: thoughts about harmful social evaluation.

State and trait anxiety

Anxiety linked to personality, or temperament, is referred to as *trait anxiety*. In contrast, the term *state anxiety* can be used to refer to immediate emotional responses to everyday experiences. Naturally, there is a link between the two, with those having trait anxiety likely to experience more state anxiety than others.

Stuttering, anxiety, and anxiety disorders

Stuttering and anxiety

There is an extensive research literature, with much recent growth, showing that those who stutter are more socially anxious than controls. Indirectly, this is verified by research, overviewed in Lecture Three, that more stuttering occurs with larger and more threatening audiences. Key findings from the last century were that those who stutter have high levels of anxiety according to standard clinical psychology measures,[15,16] and that stuttering and control subjects can be accurately distinguished using such measures without any speech data.[17,18] The former report developed the Speech Situation Checklist[17] for the identification of speech-related anxiety with participants who stuttered and participants who did not. The checklist refers to situations such as "talking to a stranger," "being interviewed for a job" and "introducing yourself," and asked participants to rate how each situation caused "fear, tension, anxiety, or other unpleasant feelings" (p. 354).[17] For 21 selected situations, the reported strength of such emotions was able to predict with 93% accuracy whether participants stuttered or not. This result was replicated in the later study[18] comparing treatment-seeking adults with controls. Results showed that 10 of the Speech Situation Checklist responses were 97% accurate for distinguishing between the groups. A survey that included 621 Australian adults who stuttered[19] asked about "anxiety level due to stuttering." Around a quarter indicated that they were "a little anxious," around a quarter indicated that they were "fairly anxious," and around a quarter indicated "very anxious." There were 12% who indicated "extremely anxious," and 10% who indicated "not anxious" (p. 6).

Research findings this century continue to confirm that, as a group, adults who stutter are socially anxious compared with those who do not stutter.[11,20,21,22,23,24,25,26] A meta-analysis[27] confirmed this for trait anxiety and social anxiety. Studies were included in the analysis if they used proven psychometric measures of trait and social anxiety, and if they compared a stuttering group of participants with a control group. Eleven studies dealt with trait anxiety and eight dealt with social anxiety. Findings confirmed that those who stutter differ as a group from those who do not stutter, with an effect size (see Lecture Five) of d=0.57 for trait anxiety and a much greater effect size of d=0.82 for social anxiety. The latter effect size showed those who stutter to be nearly a standard deviation above controls for social anxiety measures. In short, any individual who presents to a clinic with stuttering will likely—but by no means certainly— have a history of social anxiety. There is some evidence that anxiety-related problems with stuttering may affect racial and ethnic groups differentially.[28] The United States National Health Interview Surveys for 2010–2015, based on 875 parent reports for 4–17 year-olds, suggest that Hispanic and African-American children were less likely than white children to show early signs of such problems.

Salivary cortisol is a physiological marker of anxiety which has been associated with the disorder several times.[29,30] A study of 19 adult men who stuttered and 19 matched controls[31] reported higher levels in the former group. Dry mouth (xerostomia) can be a symptom of anxiety, and the stuttering participants showed a higher score than controls on a short self-report measure known as the Xerostomia Inventory. Consistent with this result, a physiological measure of unstimulated saliva flow rate was lower for the stuttering participants than the controls. Physiological makers of skin conductance and heart rate, together with self-report psychological measures, have found to be elevated with stuttering participants compared with controls.[32]

Stuttering and social anxiety disorder

What is social anxiety disorder?

Social anxiety disorder, once known as *social phobia*, is described in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5)[33] published by the American Psychiatric Association. Social anxiety disorder involves a pervasive fear of humiliation and embarrassment in social and performance situations. Those affected have an intense fear of negative evaluation and judgement by others.[34,35,36,37] Social anxiety disorder can have adverse effects on the lives of those affected, causing social avoidance and generally restricting the usual enjoyment of interactions with others. For example, it produces fear and avoidance of activities such as disagreeing with others in a social situation, expressing a controversial viewpoint, and in any way being the centre of attention.[38] Social and performance situations are commonly avoided, or endured with extreme distress, often accompanied by the physiological symptoms described earlier. An important feature of social anxiety disorder is that the expectation of humiliation and embarrassment in social situations is unrealistic and irrational in light of the actual threat.

Stuttering and social anxiety disorder

There is evidence that those who stutter and seek clinical help are likely to have social anxiety disorder. The population prevalence of the disorder is 8–13%.[39,40] However, case reports of social anxiety disorder are common for those who stutter,[41,42] with the condition reported for 40%,[43] 44%[44] and 60%[45] of cases in speech clinics. The latter of these reports indicated that such cases have 34-fold increased odds of meeting criteria for social anxiety disorder diagnosis compared with age and gender matched community controls. These reports are consistent with studies reporting that, in general, adults who stutter have anxiety scores higher than controls but slightly lower than those with psychiatric conditions.[16,23,46] However, a report of older stuttering participants after a lifetime with the disorder[47] did show anxiety scores in the range associated with social anxiety disorder. A recent review of social anxiety disorder and stuttering is available.[39] A report[48] compared the demographics of clients presenting to speech clinics for stuttering treatment with and without social anxiety disorder. Apart from the group with social anxiety disorder being significantly younger, no demographic differences were found.

As outlined during Lecture One, a stuttering moment can involve repeated movements, fixed postures, or superfluous behaviours. It is intuitive to predict that the presence of social anxiety in those who stutter might be connected to stuttering that is more behaviourally complex, containing stuttering behaviours that are potentially more socially distracting, with more fixed postures and superfluous behaviours than repeated movements. However, there is evidence that is not the case. A study of 3,100 stuttering moments from 147 adolescents and adults[49] showed no relation between the behavioural complexity of stuttering and the presence of mental health disorders, anxiety, or depression. Based on that result, the researchers indicated that, clinically, there is no reason to expect that the presence of complex stuttering behaviours suggests concomitant mental health issues.

The reality of peer responses to stuttering

There is no doubt that many, if not most, adults with clinically significant stuttering have experienced negative peer social reaction at some stage because of their stuttering. However, for a social anxiety disorder diagnosis, the expectation of social humiliation and embarrassment must be unrealistic and irrational in relation to the actual threat. Arguably then, the prevalence of social anxiety disorder and stuttering is consistent with the results of a study involving 324 adults from the United States[50] that was mentioned during Lecture Two. The study involved a list of 15 stigmatising experiences, and "most participants reported experiencing them never or rarely in the past year" (p. 55).[50] The list included experiences such as "people have been unkind to me," "people have avoided looking at me," and "people made fun of me or picked on me" (p. 55). Yet, the majority of participants agreed that "because of my stuttering, I have worried about other people's attitudes toward me," "I am fearful that others will reject me if they hear me stutter," and "because of my stuttering, I have felt embarrassed in

social situations" (p. 56). Not surprisingly, then, a report[51] showed that adults who stutter perceive their communication competence to be below than that of controls.

It is of interest, then, that there is no research that directly measures exactly how often those who stutter encounter negative social peer reactions during everyday life. One report from 1954[52] showed that many store clerks, when spoken to by someone who was stuttering, reported that they experienced embarrassment, pity, and sympathy. There were some early reports of physiological listener responses to stuttering, and a more recent report[53] verifies such negative reactions, and provides evidence also of listener skin conductance and heart rate changes when confronted by severe stuttering. There are data[54,55,56] that show listeners looking away from videos of people stuttering more often than for control speech. A report[57] suggested that, overall, listeners do not respond differently in terms of turn-taking behaviours when talking with those who stutter and those who do not. Findings showed, though, that during stuttered utterances, conversational partners tended to interrupt and complete the utterance for the speaker. Also, for those with moderate compared with mild stuttering, listeners used more reinforcers such as "um-hum" and "right." However, all this does not quantify how often such peer reactions occur during everyday life. Nor does it indicate whether someone who stutters would necessarily be aware of such reactions.

Stuttering and other anxiety related disorders

DSM-5 disorders

Many DSM-5 psychological disorders involve anxiety. Some examples are generalised anxiety disorder, mood disorders, depression, and personality disorders. There is some evidence that who stutter are at risk of having these disorders also.

There are reports[58,59] that show stuttering clients who seek treatment when compared to controls have 4.5 times more chance of having generalised anxiety disorder, 2.1 times more chance of having any mood disorder, 1.9 times more chance of experiencing a major depression, and 3.0 times the chance of having any of the personality disorders, three of which are anxiety related: obsessive compulsive disorder, dependent personality disorder, and avoidant personality disorder. However, another report about stuttering and personality disorders,[60] which used a different assessment to one of the report just mentioned,[58] failed to find any evidence of personality disorders among adults seeking treatment for stuttering.

The Symptom Checklist-Revised

A study of 200 stuttering and control adults[24] with the Symptom Checklist-Revised (SCL-90-R)[61] found significantly elevated scores for the former group, with many symptoms that may involve anxiety: somatisation, obsessive compulsiveness, interpersonal sensitivity, depressive and anxious mood, hostility, phobic anxiety, paranoid ideation, and psychoticism. A subsequent report from that cohort[62] reported the relationship between the Global Severity Index, which is an overall measure of mood state from the Symptom Checklist–Revised, and a measure of self efficacy, which is the expectation of being able to accomplish tasks. Findings were that, for a 5-month period, positive self-efficacy was associated with better mood. Another report[63] found that 30–40% of 129 adults who stutter experienced negative mood, and it was noted that the rate was similar to social anxiety disorder.

Substance abuse

There is a strong association between anxiety and substance abuse. However, this seems not to be the case with those who stutter and seek treatment.[59] That finding was replicated with a community sample within a British birth cohort study[64] where participants who reported no stuttering at 16 years (N= 10,491) were compared with those who did report stuttering (N=188). There was no evidence of an association of stuttering with alcohol or smoking.

Sleep patterns

A report from a United States health data base[65] studied sleep patterns in adults who stuttered. The authors' motivation for the study was a suggestion about irregular biological rhythms in the literature

about temperament and early stuttering,[66,67] and two reports[68,69] from the United States National Health Interview Survey linking insomnia with childhood stuttering. The report used the National Longitudinal Survey of Adolescent Health (N=13,564). Part of the survey collected data about insomnia and hours of sleep. Participants who indicated "yes" to the question "do you have a problem with stuttering or stammering?" (p. 4) (n=261) reported an average of 20 minutes less sleep per night than controls. Additionally, those participants with self-reported stuttering were twice as likely to have insomnia than controls. The authors interpreted their findings in terms of a link between sleep problems and anxiety, and hence between sleep problems and stuttering.

Depressive symptoms and suicidal ideation

Another report used the National Longitudinal Survey of Adolescent Health (N=13,564) to explore depressive symptoms and ideation.[70] Respondents were asked questions about depressive symptoms and whether they had "seriously considered committing suicide in the last 12 months" (p. 4). Results showed that, compared to controls, adolescents and young men and women who stuttered reported higher levels of depressive symptoms. For the females, there were signs of depression increasing with age. The males who stuttered (but not the females) were more likely than controls to report suicide ideation. Another paper reported that teenage girls who stutter experience more negative impact on their lives than teenage boys.[71]

The Clark And Wells model of social anxiety disorder

The puzzle

Those who have phobias such as heights, water, and spiders, generally manage to avoid what they fear and never learn that their assessment of the threat is unrealistic. For example, those who are flight phobic may never fly. Consequently, they may never learn that what they fear—that the plane will crash—does not happen to them. However, for social anxiety disorder, it is a different matter. For those affected, even if they are socially avoidant in general, it is virtually impossible to avoid social encounters entirely. Yet they still don't learn that their constant and pervasive fear of social humiliation and embarrassment is unrealistic. In other words, they persist with that belief even though experience provides constant evidence to disconfirm it.

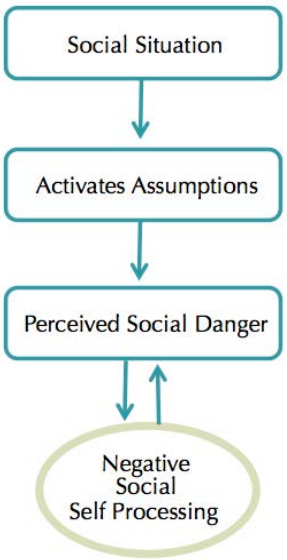
The Clark and Wells model

A major contribution of this model[34] is that it explains that puzzle about social anxiety disorder; why social anxiety persists in the face of constant experiences that should disconfirm the belief that social situations are harmful. There are many models to explain this,[72,73,74,75,76,77] and one has been devised specifically for stuttering.[78] However, the Clark and Wells model is the most influential, having been confirmed several times with tests of hypotheses derived from it. Several cognitive models of social anxiety have been developed and validated, but the Clark and Wells model dominates clinical programs for social phobia internationally. Psychologists commonly incorporate the model of social anxiety disorder within their social anxiety management procedures, with efficacious results.[79,80,81,82] Components of the model feature in a treatment for the social anxiety of those who stutter that will be discussed during the next lecture. An overview of the model has been presented in an easily understandable way,[83] and this overview is essential reading for speech-language pathologists who commonly encounter stuttering clients with social anxiety. The following description of the model draws heavily from that source.

Three assumptions of the model

The model rests on three assumptions about those affected. The first involves the existence of excessively high standards of social performance, such as believing you should always be entertaining and intelligent, and must never make a social slip-up: "I must not show any signs of weakness" and "I

should always have something interesting to say" (p. 406).[83] The second assumption involves beliefs about performing in a certain way in social situations: "If I am quiet, people will think I am boring," "If people get to know me, they won't like me" (p. 406). The third assumption of the Clark and Wells model is unconditional negative self beliefs, such as "I'm boring," "I'm stupid," and "I'm different from everyone else" (p. 407). Such assumptions cause a perception of an impending social situation to be threatening.



Negative self-processing in social situations

An impending social situation activates the assumptions just described, as shown in the accompanying figure,[‡] which signal that the impending social situation is threatening, making it a source of perceived social danger. Then, during the social encounter, negative self-processing prevents disconfirmation of the social event as dangerous.

Observer perspective

A prominent aspect of that negative self-processing is that attention shifts from the actual situation that is occurring towards an image or impression of what people think is occurring: how they think they appear to others. One way this is expressed is that the person affected sees an *observer perspective* of the situation rather than a *field perspective*. When most people are asked to recall a social encounter, they will relate to their own field of vision—their field perspective—which is of course what is appropriate for them to recall.

However, those affected with social anxiety disorder, and other anxiety disorders also, are known to report an observer perspective.[84,85,86,87] They will report how they looked to others in the social situation, which is usually uncomfortable and awkward, from the perspective of an observer. It is obvious that something is amiss with this situation, because it is impossible to see an observer perspective of yourself; it is only possible to imagine one. This prevents disconfirmation of perceived danger from the actual social situation: "what they see in the image is not what the observer would see but rather their fears visualized" (p. 408).[83]

Findings about observer perspective have been reported with interviews of adult stuttering and control participants.[88] The stuttering group reported more recollection of intrusive and recurrent mental imagery than controls. The stuttering group were distinctive for their recurring imagery themes of shame, sadness, helplessness and frustration.

Another report[89] of 30 participants seeking treatment for stuttering and 30 controls involved recall of a situation in which they felt anxious. The stuttering group was significantly more likely than the control group to recall impressions and images that were from the observer perspective. Further, the stuttering group was more likely to recall images that were negative, distorted and from that observer perspective. The authors concluded that the results could be caused by the same maladaptive social processing that occurs with anxiety disorders. This is an example of observer perspective recall from a stuttering participant:

> It was kind of over the shoulder, over my right shoulder. You could just make out the side of my head and the top of my shoulder and the person I was talking to was sort of in the full frame. And um occasionally when I was having trouble saying something I would kind of turn my face to the right so you could get a profile look of my face and how I'm struggling to complete the sentence. (p. 5)[89]

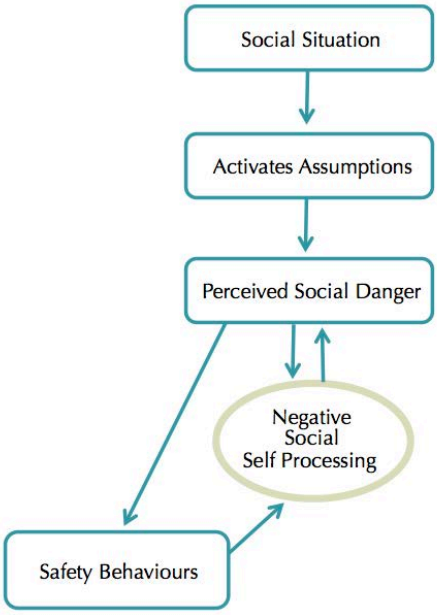
[‡] Adapted and reproduced with permission: Clark, D M (2001), A cognitive perspective on social phobia, in W R Crozier & L E Alden (Eds), *International handbook of social anxiety: Concepts, research and interventions relating to the self and shyness* (p. 405–430), Hoboken, NJ: Wiley. © 2001 John Wiley & Sons, Ltd.

Bias toward negative social stimuli

The other aspect of negative self-processing is that those with social anxiety are disinclined to recognise positive social input that would disconfirm the threatening nature of social encounters. For example, they avoid positive faces in favour of negative faces,[90] detect negative social information more accurately than positive social information,[91] have slow recognition of positive social stimuli,[92] and pay excessive attention to emotional social stimuli.[93,94]

There has been a finding to this effect with stuttering participants,[95] who looked at positive faces less often than controls when speaking to a group. They also looked less often and for a shorter time at all audience members when compared with controls. The results of Stroop task[†] studies[96,97] are broadly consistent with that finding. The latter of these two studies reported that, compared with controls, participants in the stuttering group showed bias toward socially threatening words such as "inept," "foolish," "failure," and "inadequate." Another method that has shown negative attentional bias with socially anxious participants is the dot-probe task, which is sometimes referred to as the probe detection task. A study of 43 adolescents who stuttered and 43 controls[98] with a dot-probe task found that the stuttering group had attentional bias to threatening faces. However, one report has failed to show such an an effect for stuttering participants, at least for those who were not socially anxious.[99] Another report[100] with 48 adults who stuttered and 42 controls failed to find any negative bias in responses to written descriptions of various social situations.

So, if someone is socially anxious and perceives social situations as dangerous, a destructive cycle of negative processing of thoughts can begin during the social encounter, and over subsequent encounters, that makes the perceived threat worse. Overall, this intensifies the negative self-processing. That destructive cycle is shown with the bidirectional arrows in the figure.[‡]



Safety behaviours

Attempting to prevent feared outcome

Safety behaviours are used by those who are socially anxious as an attempt to prevent a perceived threat or negative event from occurring. Those with social anxiety disorder use them commonly in social situations. Examples of safety behaviours commonly used by those with social anxiety disorder are to reduce the chance of social penalty by avoiding eye contact, allowing a conversational partner to do most of the talking, and keeping answers short. A list of common safety behaviours is presented in the Subtle Avoidance Frequency Questionnaire.[101]

Safety behaviours prevent fear extinction

There is evidence[102,103,104,105,106] that safety behaviours in fact maintain anxiety by preventing learning that fears are unfounded, and that situations are not as dangerous as they are perceived to be. Or, to use the correct jargon, safety behaviours prevent *fear extinction*. So, avoiding eye contact stops someone learning that people are giving accepting looks, and gives the mistaken impression that

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[†] With Stroop tasks, participants name the colours of the text in which different words are printed. The task can be used to assess reaction time interference.

[‡] Adapted and reproduced with permission: Clark, D M (2001), A cognitive perspective on social phobia, in W R Crozier & L E Alden (Eds), *International handbook of social anxiety: Concepts, research and interventions relating to the self and shyness* (p. 405–430), Hoboken, NJ: Wiley. © 2001 John Wiley & Sons, Ltd.

avoiding eye contact provides protection from harm, when in fact there was no potential harm. Likewise, allowing a partner to do most of the talking prevents learning that nothing socially negative happens from talking.

Safety behaviours can cause the feared outcome

In addition to preventing fear extinction, safety behaviours can have the effect of causing feared social outcomes to occur, rather than providing protection from them. For example, speaking little, keeping answers short, and avoiding eye contact can make someone appear to be uninterested in engaging with others. This can lead to the feared negative outcome. Accordingly, with the Clark and Wells model of social anxiety disorder, the use of safety behaviours in response to the original perceived social danger can feed into a cycle of worsening perceived social danger, as shown in the figure above.

Eliminating safety behaviours during anxiety treatment

Safety behaviours are routinely targeted for elimination during anxiety treatment.[34,83,107] A review[108] of studies of anxiety related conditions such as social anxiety disorder and obsessive-compulsive disorder showed that removal of safety behaviours promoted reduced perception of threat in the long term.

Safety behaviours during stuttering treatment

It appears that health professionals can unwittingly promote safety behaviours through problem solving advice for their clients.[109] There is evidence that this occurs with speech-language pathologists who treat stuttering clients.[110] That report surveyed 160 speech-language pathologists and reported evidence that they may recommend what might, in effect, be safety behaviours to adult clients in order to manage anxiety. The survey results generated a list of 34 potential safety behaviours, and factor analysis revealed five categories of them. They are presented in the table below.

SAFETY BEHAVIOUR CATEGORY	*EXAMPLES*
General Safety Behaviours	*Avoid topics that make you anxious*
	Ask many questions
	Point rather than speaking
	Allow your partner to talk for you
	Talk little
Practice and Rehearsal	*Rehearse mentally before speaking*
	Practice the speech restructuring technique just prior to speaking
	Rehearse answering the phone mentally before answering
General Avoidance	*Avoid unnecessary talking on a bad day*
	Keep answers short
	If anxious avoid difficult words
Choose Safe and Easy people	*Immediately before an important speaking situation*
	In socially threatening situations
	Say "relax" to yourself when anxious

Control Related

Speak slowly when anxious

Try to take deep breaths

The most commonly recommended of the *General Safety Behaviours* was avoiding anxiety provoking topics. Clinical psychologists recognise this strategy as a common safety behaviour that is intended to protect against social threat. Clinical psychologists also recognise silent rehearsal before speaking as a safety behaviour, and the majority of the participants reported recommending it to clients as a *Practice and Rehearsal* safety behaviour. More than half of the speech-language pathologists reported giving advice listed under *General Avoidance* once or more.

The authors of the study concluded that further research is needed to determine how often clients follow such advice. Additionally, they stated a need to determine whether any, or all, of such recommendations are in effect safety behaviours in the event that clients do use them. In other words, there is a need to determine whether they are adaptive and helpful, or in fact prevent fear extinction.

A subsequent report[111] of 133 clients who sought anxiety treatment for stuttering indicated that 132 of them reported using one or more of 27 safety behaviours. Most commonly reported were:

> "try to avoid difficult words," "rehearse sentences mentally before saying them," "keep your answers short," "choose safe or easy people to talk to in socially threatening situations," and "try to avoid difficult syllables." (p. 1249)

There was evidence that the reported use of many of the safety behaviours correlated with scores for measures known to be associated with stuttering-related anxiety: Fear of Negative Evaluation and Unhelpful Thoughts and Beliefs About Stuttering (see Lecture Eleven). This suggests a connection between anxiety and use of safety behaviours by those who seek clinical help for stuttering. That being said, in clinical psychology it is not currently clear which client behaviours should be considered safety behaviours and which behaviours should be considered healthy adaptive behaviours in a situation that causes anxiety.[102,109]

Safety behaviours and speech restructuring

A review of this topic[112] drew attention to how clinical use of speech restructuring could place clients in a situation where they attain control of stuttering at the expense of perpetuating speech-related anxiety. In other words, there is a potential conflict between speech restructuring and management of social anxiety:

> For those clients who wish to control their stuttering and where speech restructuring is deemed the most suitable approach, it is possible that speech restructuring may (a) induce or increase self-focused attention, (b) promote the use of safety behaviors, and (c) become a safety behavior itself. (p. 59)[112]

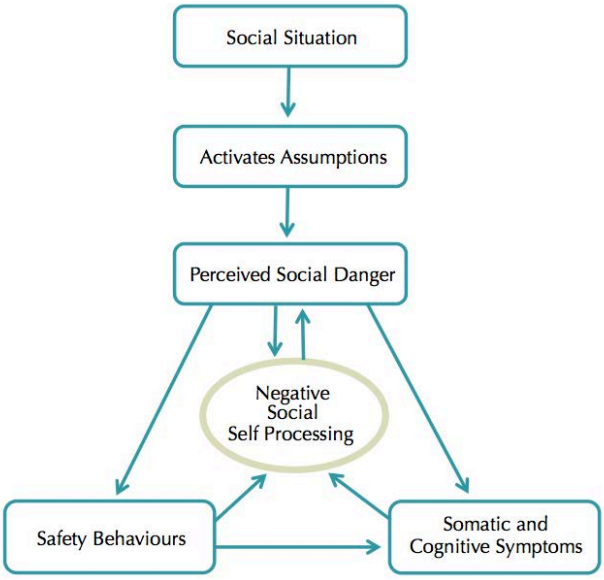
The authors of that article present a detailed approach to dealing with this clinical challenge.

Self-disclosure and safety behaviours

In this context, it is worth considering the commonly recommended technique of self-disclosure with clients who stutter. The technique was overviewed during Lecture One. As noted during that lecture, recommendations for its use connect it with speaker anxiety. In which case, it may be a safety behaviour. There is a need to determine whether the technique, in fact, prevents fear extinction and has the effect of exacerbating a feared outcome.

Somatic and cognitive anxiety symptoms

The other part of social anxiety disorder is that anxiety is not necessarily confined to unhelpful thoughts, as discussed previously. As soon as anxiety is present somatic symptoms could occur, as shown in the figure:[‡] sweating, blushing, increased heart rate, heart palpitations, hyperventilation, shortness of breath, nausea, headache, shaking, feeling flushed, and muscular tension. These physiological symptoms may prompt cognitive symptoms such as mental blocking and difficulty concentrating. The person then becomes self focussed, and attends to these physiological symptoms as well as cognitive appraisals such as negative thoughts. This process acts to confirm negative thoughts and beliefs that the situation is dangerous. Such responses can feed into a destructive cycle associated with social encounters to make the whole experience extremely distressing, not only for psychological reasons but for physical reasons.



Before the feared situation

The Clark and Wells model deals not only with what occurs during the social situation but what happens before and after it. It is typical for those with social anxiety disorder to ruminate in advance of the situation about all the past failures and negative social experiences that they have had. This can even occur so vividly that the start of the negative self-processing that occurs within a situation may even occur before it happens. This recollection of past failure, which is not at all based on reality, can lead to so much expectation of a repeat episode that the person may at this point choose to avoid the situation rather than enduring the distress of being in it. Again, all this provides further failure to disconfirm irrational beliefs about social dangers.

After the feared situation

After the event, those with social anxiety disorder can conduct a "post mortem of the event" (p. 411).[83] Rumination may continue in a destructive fashion, even though the anxiety and distress associated with the event might have subsided. In fact, such rumination might reaffirm a belief that the event was negative, and it is added to a list of past failures. Post-mortem rumination about innocuous events can be interpreted as a reflection of poor self-worth, such this example:

> a patient at a dinner buffet mentioned how much he liked a bread and butter pudding. Later in the evening, he heard his hostess say she disliked bread and butter pudding. Afterwards, he thought his comment revealed he was unsophisticated and worthless. (p. 411)[83]

[‡] Adapted and reproduced with permission: Clark, D M (2001), A cognitive perspective on social phobia, in W R Crozier & L E Alden (Eds), *International handbook of social anxiety: Concepts, research and interventions relating to the self and shyness* (p. 405–430), Hoboken, NJ: Wiley. © 2001 John Wiley & Sons, Ltd.

Anxiety impairs speech treatment

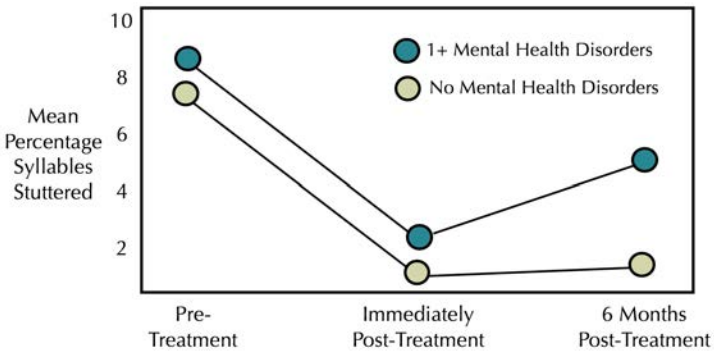
The problem of post-treatment relapse

For decades it has been known that after speech restructuring treatment to reduce or eliminate stuttering only around one third of clients are able to sustain their treatment benefits.[113,114] In other words, the relapse rate after speech treatment is around two-thirds:

> One-third of the clients achieved and maintained satisfactory fluency … one-third of the clients achieved satisfactory fluency during treatment but experienced significant regression over time … almost one-third of all clients studied either failed to complete a treatment program or were unavailable for subsequent follow-up assessment. (p. 16)[113]

Anxiety and post-treatment relapse

No plausible explanation for this relapse rate was forthcoming until a publication linked it to anxiety, showing that the one-third of clients with self-reported relapse had elevated anxiety scores on the Spielberger Trait Anxiety Scale.[114] This was verified with a study of 64 adults who received a speech restructuring treatment in clinical trials.[115] Around two-thirds of them were diagnosed with having one or more mental health disorders, the majority of which involved anxiety. Everything looked fine for the one-third of the group who had no such mental health disorders, as shown in the figure.[‡] Immediately after treatment the stuttering severity of the group reduced and remained that way 6 months later. This is a classic example of a successful short-term speech treatment outcome.



But consider the two thirds of the group who had one or more mental health disorders. This is a much different result. In the first instance, these clients had more severe stuttering than those who had no mental health disorders. They also were a less clinically responsive group. They reduced their stuttering by around two thirds, but the group with no mental health disorders reduced their stuttering by nearly 90%. However, the important finding is that at 6 months post-treatment the group with no mental health disorders showed little sign of relapse. But this is not at all the case for the group with one or more mental health disorders, who started to relapse. This is a classic example of a poor short-term treatment outcome because of relapse.

The origins of social anxiety with stuttering: The early years

Obviously, the social anxiety problems that commonly trouble adults who stutter begin at some time earlier in life. It is important, then, when young children present to clinics with stuttering, to identify any signs that might signal the potential for future development of social anxiety problems.

[‡] Adapted and reproduced with permission: Iverach, L et al (2009), The relationship between mental health disorders and treatment outcome among adults who stutter. *Journal of Fluency Disorders, 34,* 29–43. © 2009 Elsevier.

Direct evidence: Psychometrics

The ELVS Cohort

The report for the ELVS cohort (see Lecture Two) at 4 years of age[116] presented data from the Pediatric Quality of Life Inventory Parent-Proxy Report (PedsQL),[117] which is a medically oriented quality of life scale completed by parents. It showed that, at 4 years of age, no differences for "psychosocial health related quality of life" (p. 464)[116] were associated with stuttering. There are qualifications about interpretation of those data because the PedsQL scale is not a standard measure of childhood anxiety used in clinical psychology literature. However, some of the scale items clearly do pertain to anxiety, such as "I feel afraid," "I feel sad," and "I worry about what will happen to me" (p. 139).[117]

The ELVS report[116] also featured the Strengths and Difficulties Questionnaire, which is an assessment used commonly in child mental health research.[118,119,120] It measures emotional and behavioural problems with five scales: "emotional symptoms, conduct problems, hyperactivity-inattention, peer problems, and prosocial behavior" (p. 1337).[119] There are 25 test items with five items for each of those scales. To each of 25 statements about the child, parents indicate either *"not true," "somewhat true,"* or *"certainly true."* A "total difficulties score" is obtained from the sum of all scales except the prosocial behavior scale. An "internalizing scale" is obtained from the sum of the emotional symptoms and the peer problems scales, and an "externalizing scale" is obtained from the sum of the conduct problems and the hyperactivity-inattention scales.

At 4 years of age in the ELVS report, no differences were reported for Strengths and Difficulties Questionnaire measures for the control children and the children who had begun to stutter.

The Millenium Cohort

However, another report[121] provided opposite results for the Strengths and Difficulties Questionnaire using the Millenium Cohort.[122] The Millenium Cohort comprises some 19,000 children who were born in the United Kingdom during 2000 and 2001. One of the many questions asked of parents when their children were 3, 5 and 11 years old was whether the child had "stuttering or stammering" during the previous 12 months. At these ages parents were also asked to complete the Strengths and Difficulties Questionnaire, along with many other assessments. Data were available for 3-year-olds (n=173), 5-year-olds (n=194), and 11-year-olds (n=194) reported to be stuttering, and were compared with control children. The Total Difficulties scores showed statistically and clinically significant differences from controls at all ages. The report concluded that "cohort members who were reported to stutter were more likely than those with typically developing speech to experience behavioural, emotional and social difficulties" (p. 27) and that "early social, emotional and behavioural difficulties may be apparent in children who stutter as young as 3 years old" (p. 30).[121]

The United States National Health Interview Survey

The results from the Millenium Cohort were replicated in a study[123] that used data from the United States National Health Interview Survey for 2010–2015. Among many other health related questions, parents were asked whether their children had "stuttering or stammering" during the previous 12 months. A short, six-item version of the Strengths and Difficulties Questionnaire was used in the National Health Interview Survey.[124] The first five items "related to being well-behaved, experiencing worry, being unhappy or depressed, social behaviors, and attention to tasks," and required parents to respond *"not true," "somewhat true,"* or *"certainly true"* ("Data Description," para. 2).[124] The sixth item dealt with "functional impairment due to difficulties with emotions, concentration, behavior, or being able to get along with other people." For this item, parents responded "no," "yes," "minor difficulties," "yes, definite difficulties," or "yes, severe difficulties" ("Data Description," para. 2). Comparisons were made between children in the sample 4–5 years old whose parents reported stuttering (n=144) and whose parents did not report stuttering (n=7,171). Significant differences were found for all domains, with odds ratios around 2–3.

The Generation R Study cohort

A report[125] used the Generation R Study, which is a prospective Netherlands community cohort study from foetal life onwards. When the children were 9 years old parents where asked by survey "does your child currently stutter?" and "has your child ever stuttered in the past?" (p. 4566). Children were classified as "stuttering history" if one answer was "yes" and "stuttering persistence" if both answers were "yes." The sample obtained was 3,421 children, of which 118 were classified as "stuttering history" and 27 were classified as "stuttering persistence." To screen behaviour-related mental health problems in the children, the Child Behavior Checklist was administered at ages 1.5 years, 4.0 years, 5.0 years and 9.0 years.[126,127] The children's temperaments were assessed at 0.5 years and 6.0 years with the Revised Infant Behavior Questionnaire[128] and the Child Behavior Questionnaire.[129]

The study was built around two Hypotheses: Hypothesis 1 was that behaviour problems and temperament during the pre-school years (1.5 years and 3.0 years) predict stuttering history and stuttering persistence. Hypothesis 2 was that stuttering history and stuttering persistence predict behaviour problems and temperament. These hypotheses are bidirectional: Hypothesis 1 suggests causal involvement of behaviour problems and temperament in stuttering, and Hypothesis 2 suggests them to be an effect of stuttering. The authors concluded "we found hardly any evidence for our first hypothesis" (p. 4572). The only significant result from six Child Behavior Checklist and six Infant Behavior Questionnaire subtests was a temperament score of "recovery from distress." There was support for Hypothesis 2: stuttering persistence during the pre-school years was associated with Child Behavior Checklist scores later in life, and with stuttering history associated with negative affectivity temperament scores at 6 years.

Clinical cohorts

A report of 427 children[130] included the Child Behaviour Checklist, the Short Temperament Scale for Toddlers,[131] and the Short Temperament Scale for Children.[132] The children were treated for early stuttering beginning when they were younger than 6 years. For that cohort of children who presented clinically, the authors reported "nothing unusual about behavioural and emotional functioning, or the temperaments" (p. 1) of the children. The report used the Depression Anxiety and Stress Scales[133] with parents and reported that their scores were within normal limits. However, the Recent Life Changes Questionnaire[134] suggested that "a third of parents were experiencing moderate to high life stressors at the time of seeking treatment" (p. 1). This prompting the authors to speculate that "life stresses were instrumental in prompting parents to bring their stuttering pre-schoolers to clinics, possibly with the onset of their children's stuttering exacerbating such stresses" (p. 15).

Another report[135] was consistent with the ELVS report, showing no health related quality of life issues with a group of 197 children, ages 3–6 years, who were participants in a clinical trial of early stuttering treatment (see Lecture Seven). Compared to normative data, there were no systematic problems for the children according to four medically oriented quality of life instruments.

Small studies

A report[136] incorporated data for eight children who showed pre-treatment scores in the normal range for the Child Behaviour Checklist. Another report[137] presented data for seven stuttering and seven control children and showed that they did not differ on the Preschool Anxiety Scale,[138] which is a more direct parent report measure of anxiety. This report also showed no differences for salivary cortisol. Those two reports, however, do not contain enough participants to be particularly convincing.

The Pictorial Scale of Perceived Competence and Social Acceptance for Young Children[139] is a direct assessment based on child responses. It was given to 28 children with stuttering, mean age 4 years 9 months, and a control group of children.[140] There were no significant differences between the two groups for test scores; however stuttering severity was a significant predictor of social acceptance, accounting for 20% of the variance in test results.

A significant result was found in a study of Turkish children who stuttered and controls.[141] The report included 15 children who stuttered and 15 controls aged 3–6 years. Significant differences were found

for the Child Behaviour Checklist between the groups for all seven scales of the test, notably "Anxious Depressed" and "Emotionally Reactive."

What does it all mean?

Small studies that are well controlled mostly show no differences between stuttering and control groups for psychometric measures of anxiety. However, two large cohorts—the Millenium Cohort and National Health Interview Survey—found differences. The authors of both those reports acknowledge the limitations of the parent-report method for identifying stuttering compared direct diagnostic methods such as in the ELVS cohort and other studies. Parent *"yes/no"* reports of stuttering in a questionnaire are not particularly compelling. That being said, significant, replicated, large-cohort findings between stuttering and nonstuttering childen for the Strengths and Difficulties Questionnaire cannot be disregarded. Such findings so early during the developmental course of stuttering need to be considered when forming a clinical view about when to begin early intervention for the disorder, as will be discussed shortly.

Regardless, there is no consistent pattern of evidence that early stuttering is associated with psychometric measures of anxiety; there are only suggestions of some association. Considering the strong association between persistent stuttering and anxiety measures, as discussed earlier during this lecture, it seems compelling to conclude—as did one study of the matter[125]—that anxiety-related mental health issues are a consequence of early stuttering rather than being involved in its causality.

Direct evidence: Early childhood temperament

Temperament

It is estimated that 20–60% of adult personality traits can be accounted for by temperament. Temperament is a stable, innate, and constitutional tendency to react to the environment or interact with it in a certain fashion. According to a popular definition,[142] "temperament describes our early emotional, motor, and attentional equipment, along with the regulating capacities that allow us to control our reactions and put them to good use" (p. 7). Temperament is influenced by biological features that include genetics. Childhood temperament is generally accepted as a risk factor for anxiety later in life.

The most commonly used classification of childhood temperament was developed by Thomas and Chess,[143] and involves nine parent-reported dimensions. One of those is "approach/withdrawal," which refers to how children respond to new situations: whether they readily engage in them or retreat from them. This dimension of temperament—behavioural inhibition or shyness—is regarded as a risk factor for anxiety disorders later in life.

Temperament and early stuttering: Some fundamental caveats

There is a rapidly accumulating, diverse body of literature dealing with the association between early childhood temperament and stuttering. At present, this body of literature is inconclusive, and is being interpreted differently by many researchers. A reason for its inconclusive nature could be that it is constrained by inherent limitations. So, a discussion of those limitations is useful before considering that literature.

A fundamental (and often overlooked) caution about this body of literature is that any findings of association between temperament and early stuttering may pertain to speech and language disorders generally rather than to stuttering specifically.[144,145,146] This was illustrated with a report[147] associating negative emotionality with poorer receptive vocabulary for pre-schoolers who stutter; a finding that has been reported also for nonstuttering children.

That aside, given that temperament is a stable, innate construct present at birth, there is a potentially misleading implication in reports of temperament measured after the onset of stuttering. As outlined during this lecture, it is clear that young children may encounter all kinds of adverse psychological events after stuttering onset. Therefore, it might be more appropriate to think of "temperament" measures after stuttering onset as personality measures, which reflect the interaction between

temperament and environmental experiences.[†] Indeed, inspection of the commonly used measures of early childhood temperament suggests the likelihood that, in part, they reflect experiences of stuttering.

In short, any association between early stuttering and measures of temperament may reflect the effects of stuttering rather than its causes.[150,148] This clearly is a possibility because most children involved with research about stuttering and temperament have been stuttering for some years. Clearly, a link between temperament and early stuttering is not firm evidence of causality, but it does raise the possibility of a causal connection.[149]

Another reservation about this literature is that it is observational, not experimental. In other words, it is based on simply observing the temperaments of children who stutter and children who do not stutter. As such, it can only establish an association between stuttering and temperament. It cannot definitely establish anything about the causality of stuttering.

And finally, many measures of temperament for children with early stuttering are based on parent report, which may be influenced by the presence of the disorder. In other words, when parents report about their children's temperament characteristics, it is not clear to what extent their reports are confounded by the fact that the child stutters.

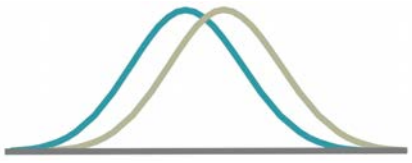
Three reviews of early childhood temperament and stuttering

A full review of the topic is beyond the scope of this lecture, but the pertinent research is cited in three current reviews.[146,149,150] One of them[146] cites evidence that, for all children, a so-called "difficult" temperament heightens the risk of anxiety disorders later in life. "Difficult" temperament includes "nervous, high strung or tense," "appears fearful or anxious," "appears worried," "not as happy as other children," and "has difficulty having fun" (p. 153).[146] The review concluded:

> Using the guideline that independent replication of findings makes them trustworthy, there is an inevitable conclusion to this review. For stuttering children during the preschool years, there may be some association between temperament and stuttering … The guarded nature of this statement arises because of the modest scope of the research on which it is based, amounting to 10 publications, and because of some inconsistencies with results … (p. 158)[146]

Another review[150] was published the following year, and incorporated six new studies. Those authors also were tentative in their conclusion that "childhood stuttering may be associated with constitutionally based temperamental/emotional processes, many of which are believed to be open to environmental influences" (p. 128).[150] In another publication,[151] that research group summarised their view of the consistent findings to have emerged from a generally inconsistent literature about stuttering and early temperament. Compared with controls, they stated that children who stutter are (1) less adaptable, (2) have poorer attention and attention regulation, and (3) have a negative mood.

A review of the topic[148] focused on the effect sizes reported in the literature, recording all of them in tables. A central argument in the review is not only that inconsistent effects are reported but that maximum effect sizes reported are moderate. The review author makes the point that the two distributions for such an effect size look something like the figure on the right. There is extreme overlap between the two groups and no real separation between them. All of this led the reviewer to conclude:



> Children who develop stuttering (as a group) are *not [author's italics]* characterized by temperamental traits such as shyness, social anxiety, or general anxiety … A subgroup of CWS *[children who stutter]* tends to show somewhat elevated traits of inattention and hyperactivity-impulsivity. (p. 18)[148]

[†] Thanks to Ross Menzies for this content.

Parent measures of temperament

The prospective ELVS cohort[177] was used to determine the presence of temperament anxiety markers prior to and after the development of stuttering. This report involved 183 children with stuttering and 1,261 nonstuttering children, aged 2–4 years. At the children's second, third, and fourth birthdays, parents completed the Short Temperament Scale,[152] which is based on the Thomas and Chess temperament classification.

No differences were found at any age for the "approach" and "easy difficult" scales, which are thought to be anxiety precursors. At 3 years of age significant differences were found for the "reactivity" and "persistence" scales, indicating that at that age children who stuttered "were less reactive to environmental stimuli and had a reduced ability to attend to a task until completion" (p. 1314).[177] However, there was no evidence for a continued difference with "persistence" at 4 years, and "reactivity" was not measured at that age. The authors concluded that there were

> no signs of temperament precursors of anxiety before stuttering onset or shortly after. Results suggest, at most, that temperament is influenced somehow during the period after stuttering onset, but with a waning developmental influence subsequently. (p. 1314)[177]

A study of 123 children with ages 9–14 years[153] explored how temperament affects the impact of stuttering. Correlations were established between OASES scores and the Early Adolescent Temperament Questionnaire-Revised,[154] which is a self- and parent-report measure. Moderate correlations were found between "Surgency" and "Negative affect" temperament scores and OASES Overall Impact scores, which prompted a conclusion that

> … on the one hand, more extravert and less fearful/shy children experience a lower overall impact of their stuttering; on the other, children with higher levels of irritability and frustration experience a higher overall impact of their stuttering. (p. 427)[153]

However, the authors noted that correlation does not establish causality, and that the study did not exclude the possibility that temperament scores simply reflect experiences with stuttering.[153] Evidence to support that contention was reviewed earlier from the Generation R Study cohort.[125]

That conclusion from the Generation R cohort was consistent with the conclusion from the review noted earlier,[177] that temperament is most influential on early stuttering but less so later in childhood. The authors of the study of 123 children[153] noted that a correlation between stuttering severity and the temperament dimension of "effortful control" had been reported for early stuttering.[155,156] However, their study found no correlation between stuttering severity and temperament scores for 9–14 year-olds, and such a correlation seems absent also with persistent stuttering in adulthood.[157]

An innovative feature of the study of 123 children[153] was that, using the Early Adolescent Temperament Questionnaire, parent measures of temperament were supplemented with child measures. This is of interest because of the caveat mentioned earlier that parents' reports of temperament might be confounded by their children's stuttering. Moderate correlations were found between parent and child temperament scores, leading the authors to dismiss that prospect. However, because moderate correlations were found, another possibility is that parent reports about their children's temperaments might be influenced considerably by variables other than temperament, which might include the children's stuttering.

Using the same cohort of children, another report by the authors with 132 children who stuttered, ages 9–14 years,[158] reported data from the Early Adolescent Temperament Questionnaire-Revised, and the Revised Children's Anxiety and Depression Scale.[159] The latter scale measures anxiety and depression symptoms, with a parent- and a child-reported version. They reported significant, low to moderate correlations between child- and parent-rated versions of the temperament scale (surgency and negative affect) and the depression scale. However, the children's scores on the anxiety and depression scale were not considered to be clinically significant.

The second Illinois cohort[160] (see Lecture Two) studied 58 children with early stuttering and 40 control children. According to one subtest of the Children's Behavior Questionnaire-Short Form,[161] there was a significant difference between the groups during the 4–5 years of the study for Negative Affectivity. The children in the stuttering group who did not recover had greater negative affect than the control group and the children who recovered.

If there is a relationship between early stuttering and temperament, there might be a correlation between stuttering severity and temperament measures. Ten studies have reported such a correlation or association,[155,156,162,163,164,165,166,167,168,169] and six have failed to show a correlation.[153,158170,171,67,172] One of these reports[169] with 47 young children showed small but significant correlations between "percentage of stuttered disfluencies" during short narratives in a laboratory and "surgency," which is an index of emotional reactivity involving high levels of positive affect, derived from the Children's Behavior Questionnaire-Short Form. However, only around 10% of the variance was explained. The report failed to find any similar correlations for skin conductance.

Another study[156] followed up on an earlier finding[155] with 98 children from a university clinic, having a mean age of 6 years 7 months. The authors sought to replicate the earlier finding of an association between the temperament dimension of "effortful control" and parent measures of stuttering severity. The effortful control temperament dimension, measured with the Children's Behavior Questionnaire-Short Form,[173] describes the capacity to regulate focus and attention. A regression model found a statistically significant relationship between parent measures of stuttering severity and effortful control, with 13.6% of the variance explained.

Clinical applications

There have been two suggestions[151,155] that an association between temperament and stuttering severity would contribute to sustaining early stuttering. This idea, that early childhood temperament somehow contributes to the development of early stuttering, has been echoed by other researchers.[174] Some have gone as far as suggesting the following about their findings:

> Holistic treatment for stuttering in children should include the behavioral components of enhanced self-regulation of EC *[effortful control]* (i.e., attention, inhibitory control, and perceptual sensitivity) alongside traditional approaches to augment the effectiveness of each. (p. 12)[156]

Indeed, one of the authors took up this notion with a clinical trial, which will be discussed at the end of this lecture.

Some authors have gone as far as suggesting (cautiously) that there may be clinical applications of findings associating temperament and early stuttering.[150] In essence, they suggest that childhood temperament may be a consideration in determining whether a child should have a "direct" treatment such as the Lidcombe Program or an "indirect" treatment based on multifactorial models (see Lectures Six and Seven). In support of their speculation, they presented preliminary results suggesting that temperament predicts outcome of an "indirect" therapy.[175]

The topic of clinical applicability of findings about temperament and early stuttering has been debated.[176] The arguments presented there in favour of their clinical applicability might be overviewed as:

> Stuttering is a complex, multifaceted developmental disorder with numerous research findings that highlight a wide range of individual differences and interrelations between and among multiple variables/factors .... Consequently, there is a need to obtain and integrate parent- and self-report, behavioral, and physiological data when studying, evaluating, diagnosing, and developing treatment plans for children who stutter … Further, substantial longitudinal evidence from other fields (e.g., psychology and child development) clearly demonstrates the interrelations between developmental challenges, parent-child attachment, emerging self-regulation processes, and socialization – with particular attention to communication on long-term (i.e., across the life span) academic, social, psychological, and vocational outcomes … Given its

> heterogeneity, early attention to the child in context, as a whole, is similarly critical for understanding the onset, development, and exacerbation and/or amelioration of stuttering … (p. 4)

And the arguments presented there against the clinical applicability of research about temperament and early stuttering might be overviewed as:[176]

> … that constitutes the following logical fallacy: stuttering is a multifactorial disorder, therefore a multifactorial assessment and treatment approach necessarily follows. … with the current state of evidence about temperament and early stuttering, it is not pertinent to the day to day business of providing health care to children who stutter. … none of the data reported so far describe how pre-treatment temperament affects treatment outcome. Nor do any available data describe how clinical responses associated with a certain type of child temperament can affect treatment outcome. … the critical clinical issue is a link between such temperament features and early stuttering development. One way to establish such a link is to discover features of temperament that distinguish stuttering from control participants prior to stuttering onset. At present, only one cohort attempted to do that, and failed, albeit with only one temperament measure and without any independent replication.[177] Emerging findings to the contrary would be a clinical game-changer. (p. 5–6)

Perhaps one way of summarising this clinical controversy is to present the two extreme views that have been presented by researchers and scholars. One extreme is that some children will have an unusual temperament, and some children will begin to stutter. Therefore, some children who stutter will have an unusual temperament, and how the treatment of those children needs to accommodate their temperament will be clinically obvious. The other extreme view is that temperament is involved with stuttering development in a clinically crucial way, or perhaps even a casual way, and hence temperament requires comprehensive assessment pre-treatment in order for satisfactory treatment to occur.

Direct evidence: Autonomic nervous system studies

The physiology of anxiety and early stuttering—one of the three loosely related components of anxiety—has been studied by comparing the autonomic nervous systems of children with early stuttering and controls.[151,168] One report[151] studied 20 children with early stuttering and 20 controls. The study dealt with the temperament feature of emotional regulation, using indices of sympathetic and parasympathetic nervous system activity.[†] Skin conductance was used to measure sympathetic activity, and respiratory sinus arrhythmia was used as a parasympathetic measure. Respiratory sinus arrhythmia is heart rate fluctuations linked to breathing that occur naturally; during inspiration heart rate increases and during expiration heart rate decreases. Lower respiratory sinus arrhythmia is associated with social anxiety in adults.

The children watched a neutral screen during a baseline condition.[151] Then they watched short videos that successfully elicited positive and negative emotions. Then, the children told a story about the videos. Some important significant differences between the two groups were reported. The children who stuttered showed lower (parasympathetic) respiratory sinus arrhythmia during the baseline, which theoretically means they had increased vulnerability to a sympathetic response. Additionally, the children who stuttered showed more (sympathetic) skin conductance increase during positive emotions while watching and talking about videos. Interestingly, a similar effect size was reported (Table 3) which was consistent with the observations in a previous review.[150]

The authors concluded[151] that their results suggested autonomic nervous system involvement with early stuttering. They speculated about the nature of that involvement: that such activity may divert the necessary attentional resources from speech and communication. Naturally, the presence of unusual

[†] The sympathetic nervous system controls responses to perceived threat, and the parasympathetic nervous system controls homeostasis at rest.

temperament and early stuttering raises the caveat discussed earlier of whether such unusual temperament is involved in the cause of stuttering, or whether it is merely an effect of stuttering. The authors acknowledged this issue but argued that it is unlikely that their results were caused exclusively by the experience of stuttering.

In their conclusions to another study of respiratory sinus arrhythmia and early stuttering,[168] those authors again presented this caveat but argued a view that on balance, "cognitive, emotional, and related processes appear to play a meaningful role in the onset and/or developmental trajectory of childhood stuttering" (p. 2146). The same research group reported a skin conductance study of nine young children with persisting stuttering and 23 who they considered recovered around 2 years later.[178] During a stressful picture naming task, data showed a significant 14% difference between the groups, but no differences from controls. Again, the authors concluded that "that emotion should be considered in any comprehensive account of childhood stuttering," regardless of "the directionality of effect" (p. 149).[178]

A report by the same group[179] involved 18 children who stuttered and 18 controls, mean age 4 years 5 months. The groups did not differ for baseline skin conductance, heart rate, or respiratory sinus arrythmia. However, when presented with faces of negative valence, compared to controls the stuttering children showed an emotionally reactive effect in terms of higher heart rate and greater decrease of respiratory sinus arrythmia. The authors concluded that "emotional reactivity and regulation has clinical significance for preschool-age stuttering and should be considered during assessment and treatment of stuttering in children" (p. 14), but no elaboration of that statement was provided.

A study[180] of 47 children who stuttered and 25 control children, with a mean age of 4 years 8 months, failed to replicate the findings discussed above. The children engaged in speech and nonspeech tasks, while measures were made of skin conductance and blood pulse volume. The researchers reported that "overall, the results of our study do not support the hypothesis that atypically high levels of sympathetic arousal are associated with speech production in preschool children who are stuttering" p. 11).[180] Additionally, no differences were found between the two groups of children for the parent temperament measure the Children's Behavior Questionnaire–Short Form, and the KiddyCat measure of children's attitude to communication.

Another report[181] did not support the broad notion that there is autonomic system involvement with early stuttering. Thirty two children with early stuttering (mean age 3 years 11 months) and 16 controls (mean age 4 years 1 month) engaged in picture description and a more challenging nonword repetition task. Electrodermal activity was the same for both groups with the picture description task, but was elevated for the nonword repetition task. For the group of children who stuttered, KiddyCat scores were significantly higher, as were the Fear Scale and the Sadness Scale of the Children's Behavior Questionnaire–Short Form. The researchers concluded that

> Our findings suggest that age-appropriate social communication tasks are not inherently more stressful for preschool-age CWS and are not associated with state- related stress or anxiety that is often reported for adults who stutter. However, speaking tasks that place a higher demand on children's cognitive–linguistic system may be more taxing and challenging to preschool CWS than CWNS, leading to a higher level of arousal. (p. 4030)[181]

Direct evidence: Executive function

Overview of executive function

The term *executive function* refers broadly to cognitive activity that exerts control over day-to-day activity, including thoughts, emotions, and behaviour. Executive function develops early in life. From a theoretical perspective, there is overlap between executive function and temperament. Additionally, there is empirical support for the notion that the expression of temperament may be influenced by executive function.[182] A review of executive function is available.[183] There is another comprehensive overview of the topic and meta-analyses of research results pertinent to early stuttering.[174] That

overview outlines three components involved with the concept of executive function: "inhibition," "working memory," and "cognitive flexibility" (p. 1). The authors of the review point out that early development of executive function is underpinned by "a developing attentional system" (p. 1). The organisation of the present discussion of executive function and early stuttering draws from this overview and its meta-analyses of children who stutter and control children. Those meta-analyses involved 29 studies, 48% of which were children with early stuttering, 21% of which were school-age children, and 31% contained children from both age groups.

Executive function: Attentional control

Attentional control is also known as *attentional shifting*. Broadly speaking, this is the capacity to concentrate on one thing. Several aspects of this capacity can be directly measured for children using behavioural tasks and with parent report tools such as the Children's Behavior Questionnaire.[215] Meta-analysis[174] of seven studies showed significant results for the domain of "attentional focus/persistence" (p. 15) when measured with parent report. However, another domain of attentional control—distractibility—showed no significant effect size with parent measures for three studies. Six studies of behavioural attentional control measures were analysed, with no significant effect size. Since that meta-analysis, a report has been published[184] for 15 children who stuttered and 18 controls, ages 8–11 years, using an experimental attention-shifting paradigm. The children who stuttered had more difficulty than controls with task-shifting that required cognitive flexibility.

In this context, it is of interest that a meta-analysis involving 21 studies of attentional ability with adults who stutter[185] showed overall inferior performance to control participants. The author concluded that "stuttering most likely co-occurs with a problematic attentional ability only in a subgroup of PWS [people who stutter] (p. 8)." The author noted that the "subgroup" finding was consistent with that finding for the review of early stuttering and temperament discussed earlier.[150]

In a review of the topic[183] the authors speculate about mechanisms by which "young CWS *[children who stutter]* would have weaknesses in executive function in the first place" (p. 6). One potential explanation is as follows:

> from a resource allocation standpoint, as CWS struggle to plan or execute speech/language and/or attempt to manage their fluency breaks, they may overutilize limited executive function resources, including aspects of attention, to compensate for fluency processes that do not come as automatically for them … (p. 6)[183]

Their second explanation is based on research findings suggesting that early stuttering is associated with inferior language development (see Lecture Two):

> concomitant weaknesses in language processing result in limitations in executive function, which subsequently lead to deficits in other domain-specific processes. (p. 6)[183]

Executive function: Response inhibition

Response inhibition is also known as *inhibitory control*. Response inhibition refers generally to "the ability to resist, subdue, or withhold one's thoughts, behavior, and/or emotional response" (p. 15).[174] With a meta-analysis,[174] five studies of parent report measures of response inhibition showed a significant effect size, and three behavioural measures showed no significant effect size.

With adults, Stroop tasks can be used to measure response inhibition by presenting names of colours printed in a different colour to the name and asking participants to name the colour of the printing. Several methods are available to measure dimensions of response inhibition for children of different ages. One method for young children, which involves no verbal stimulus, is the "baa-meow task."[186] Children are played audio recordings of sheep and cat noises and asked to push a button that is the opposite of the animal they hear.

Another method to measure response inhibition in children is the peg tapping task. Children are asked to tap once when the experimenter taps twice, and vice versa. Thirty children who stuttered and 30

controls, aged 3–6 years, completed that task.[187] Significant group differences were detected, leading the authors to conclude that children who stutter "not only have difficulties with inhibition and impulsivity in the verbal domain, but also the nonverbal domain, suggesting a domain-general weakness" (p. 14).

Executive function: Verbal short term memory

With children in the meta-analysIs,[174] verbal short term memory was measured with nonword repetition tasks of various kinds, and with a forward span task. The latter task "measures the extent to which a person can recall a list of presented digits, letters, or words in the order in which they were presented" (p. 2). Nine studies showed a significant effect size for nonword repetition tasks, and seven studies showed a significant effect for forward span tasks. Subsequent to the meta-analysis, another observational study of verbal short-term memory for children with early stuttering and controls reported poorer performance for the former group.[188]

The authors of the meta analysis concluded that, for verbal short-term memory, there were "robust differences" (p. 15)[174] between the children who stuttered and the control children. Regardless, they tempered this conclusion by stating that those differences were subclinical, meaning that they did not require clinical attention. They applied this same conclusion to their findings for attentional control and response inhibition. This view, that these differences are clinically unimportant, contrasts with a view mentioned earlier.[150]

Executive function: Summary

The term *executive function* refers broadly to cognitive activity that exerts control over day-to-day activity, including thoughts, emotions, and behaviour. There is some theoretical overlap between executive function and temperament. Mixed research findings have been published for two components of executive function: attentional control and response inhibition. This makes it difficult to form any view at presence about the relevance of these constructs to early stuttering. More compelling research findings have occurred for verbal short-term memory. However, in the case of those findings, their importance is difficult to assess because of their sub-clinical nature.

Indirect evidence

Parent-reported child awareness and negative peer reactions to early stuttering

The potential anxiety issues reported for adults who stutter can be attributed to negative social conditioning during peer interactions early in life.[34,36] Consequently, it is important to recognise the body of research suggesting that young children are likely to be aware of their stuttering and that it is capable of causing them distress. This body of research contains evidence of reactions such as talking less and situation avoidance, which are interpretable as early anxiety avoidance behaviours.

An early report of children assessed by one researcher during a 6-year period documented this effect.[189] There were 104 children in the age range 2–4 years, and by 3 years of age around half the children were reported to show awareness of their stuttering and negative reactions to it:

> Of the nine two-year-old subjects, four were said to have reacted to repetitions or other types of blockage by exclaiming, 'I can't talk,' by crying, or by looking down and blushing. In one case, seen three weeks after reported onset, the child was said to have become 'so annoyed' by his repetitions that he hit himself on the mouth and stopped talking for three days. At age three about half the children are said to have exhibited these or other evidences of reaction to stuttering blocks at one time or another. Reactions of this kind appear to be common up to about age six. At these age levels children frequently say, 'I can't talk,' 'Why can't I talk?' or 'Help me talk.' Other verbal reactions reported are 'My goodness,' 'I'm doing it again,' or 'I'll tell you later. (p. 233)[189]

In another early report,[190] five of 22 parents of children who began stuttering prior to 3 years indicated they thought their children were aware of stuttering, with four of them indicating their children were "aware and bothered" (p. 176) by it.

Later reports have been consistent with these findings. One showed that 57% of 1,122 parents of 2-year-olds said that their children showed signs of awareness of stuttering, and the figure had increased to 90% for 7 year-olds.[191] Around two-thirds of children who had been stuttering for less than 1 month reportedly showed awareness of it. Using a Japanese version of the survey in the first study, that result was replicated with 57 Japanese children of ages 3–7 years.[192] Seventy percent of parents reported child awareness of stuttering at 4 years, and 90% at 7 years. Another report of 77 parents of children, mean age 53 months (range 34–73 months),[193] showed that 90% of them reported some kind of negative impact from stuttering. The most common reported reactions were "frustration associated with their stuttering, withdrawal, reduced or changed verbal output, making comments about their inability to talk" (p. 407).[193] Twenty-five per cent of the parents linked stuttering with talking less, and 43% said stuttering negatively affected the children's mood. Twenty-seven per cent reported that peers teased their children about their stuttering, which is a higher rate of teasing than the usual 6–22% range reported for children who do not stutter.[194,195] Thirty-eight Norwegian parents of 2–5 year-olds used an adaptation of the OASES to report the impact of stuttering on their children.[196] Results indicated that most parents thought that stuttering adversely affected their children.

Attitude to Communication

The KiddyCat[197] is designed as a measure of attitude to communication. Children are asked 12 *yes/no* questions, such as "is it hard for you to say your name," "do your words come out easily," and "do people like how you talk?" (p. 229).[198] Consequently, it might be interpreted as having some relation to social anxiety.

Three studies from the United States have shown KiddyCat score differences between children with early stuttering and controls: with 52 stuttering and 62 control children,[198] 45 stuttering and 63 control children,[199] and with 46 stuttering and 66 control children.[200] The same result has been reported for 58 stuttering and 70 control Polish children,[201] for 49 stuttering and 74 control Slovenian children,[202] and 55 stuttering and 53 control Turkish children.[203] The KiddyCat was used in a study mentioned previously[137] with small participant numbers, and it showed no differences between stuttering and nonstuttering children. A report of 59 children with a mean age of 4.8 years (range 2–6) years[204] indicated that neither[205] stuttering severity nor time since reported stuttering onset predicted KiddyCat scores.

Evidence of negative peer responses

There is direct video evidence of negative peer reactions to stuttering in pre-school playgrounds.[206] Four children with early stuttering were video recorded during four 20-minute playground periods. During these 80-minute samples of conversation, negative peer responses to stuttering were reported for three of the four children. The percentages of negative peer responses to stuttered utterances that had communicative intent for the three children were 2.8%, 12.5%, and 28.6%. For the latter child, more than a quarter of stuttered utterances with communicative intent received negative peer responses. Those peer responses included interrupting, mocking, walking away, and ignoring what the child was saying. One child was even assaulted because his stuttering prevented him from resolving a conflict about a toy.

Those results are alarming because, if they in any way reflect what generally happens to young children who stutter, there is good reason to believe that they may be exposed to the kind of negative social conditioning that could be the origins of anxiety later in life.

Peer awareness

Indirect evidence that children with early stuttering may receive negative social conditioning comes from an ingenious research paradigm involving stuttering and nonstuttering puppets.[207] On three occasions over a 2-year period, 20 stuttering and 20 nonstuttering pre-school children were asked to "point to the puppet that talks the way you do" (p. 233–234). Results showed that the children were generally able to identify with the stuttering and control puppets, and their reliability for so doing increased during the period of study.

A preliminary report has begun for a method to explore pre-schoolers' attitudes to stuttering using avatars,[208] by adapting the well-known *Public Opinion Survey on Human Attributes—Stuttering* (POSHA-S) that has featured in much of the research about stuttering stereotypes discussed during Lecture One. Preliminary results with the POSHA-S/Child for 51 children 3–7 years old indicated the potential for negative attitudes toward the disorder at that time of life. A subsequent report[209] verified this finding. Children 4–10 years old generally had more negative attitudes than adults but showed systematic improvement during that period. That improvement trend was replicated with children and parents from Bosnia and Herzegovina.[210] The POSHA-S/Child was used to assess attitudes to stuttering of 37 pre-schoolers before and after an intervention designed to improve it.[211] The intervention comprised two 30-minute classroom lessons with puppet videos, group discussion, and schoolbook activities. Results showed a post-intervention improvement of attitudes. The result was replicated with Polish children.[212]

A clinical trial of resilience training for stuttering with pre-school children

Background

The authors of this clinical trial[213] based their design on material discussed earlier in this lecture. They cited "strong evidence of the significant differences in self-regulation, emotional reactivity, and resilience" (p. 71) between pre-schoolers who stutter and those who do not. They designed their trial for many purposes, including to determine whether speech treatment improves after intervention and the effects of resilience training on pre-schoolers who stutter and their parents. However, a key issue explored with the trial was raised earlier by those involved with research about early stuttering and temperament: whether a resilience component added to speech treatment for children with early stuttering improves speech outcomes.

Design

The randomised controlled trial was used to compare a group of pre-schoolers who received speech treatment for stuttering compared with a group who received that speech treatment and "an additional resilience component" (p 21).[213] Participants were 28 child-parent pairs with children being a mean age of 4.4 years. Assessments occurred pre-treatment and immediately after 12 weeks of treatment.

Outcomes

Four standard outcome measures were used, but none were specified as the primary outcome. Percentage of syllables stuttered was obtained from a within-clinic sample of 350–450 syllables during a play task. The Parenting and Family Adjustment Scales[214] measure various domains of family functioning and parenting, and the parenting practices domain was used. The Strengths and Difficulties Questionnaire[118] measures emotional and behavioural problems. Finally, the authors used an adapted version of the effortful control subscale of the Children's Behavior Questionnaire.[215]

The treatments

The speech treatment was "a combination of direct and indirect stuttering therapy" including "response contingent principles," "'Demands and Capacities Model' treatment" (p. 74.)[213] (see Lecture Three) and Palin Parent-Child Interaction therapy (see Lecture Six). The additional component in the experimental treatment arm was the Curtin Early Childhood Stuttering Resilience Program.[†]

[†] The authors note a study limitation that this intervention has not been standardised and there is no evidence of its efficacy. It has not been reported previously and no references to it, or to a treatment manual, are cited in the report. The paper incorporates a similarly unreported and and unreferenced pilot assessment connected to the intervention: the Curtin Early Childhood Stuttering Resilience Scale. Again, the authors acknowledge a study limitation that the scale has not been assessed for reliability and validity. Consequently, results from the scale are not discussed here.

Results

Pre-treatment mean %SS was 10.5 and post-treatment was 2.1. There was no evidence that the resilience treatment improved these outcomes. There was no evidence of any improvement post-treatment for the temperament dimension of effortful control. There was evidence of reduced behavioural and emotional problems in both groups, but no evidence that the child resilience training added to that effect. For both groups, there was evidence of improved parenting practices at post-treatment. However, there was evidence that the experimental group that received parent training had more improvement than the group that did not.

Limitations

The authors conclude that "this study provides evidence for the overall effectiveness of early intervention in stuttering therapy" (p. 79). Yet the extent of the contribution is limited by the evaluation of the treatment, which was based on 350–450 syllables of within-clinic speech measured immediately post-treatment without a follow-up period. According to some criteria, the report would not be regarded as a clinical trial (see Lecture Five). The authors also conclude that the experimental treatment was "successful in positively shifting parenting practices, and developing improved self-regulation and resilience" (p. 79). It might be arguable that a more important contribution of the trial is the finding that such changes had no impact on speech treatment outcomes. It is also difficult to interpret the results of the study because there was no indication of whether pre-treatment scores for the Parenting and Family Adjustment Scales, the Strengths and Difficulties Questionnaire, and the Children's Behavior Questionnaire were within or beyond the normal clinical range.

Conclusions

It seems clear that children are likely to be aware of stuttering shortly after onset and that it may well cause them distress. Additionally, early stuttering may be associated with negative peer social conditioning, which is potentially associated with anxiety development later in life. Direct test evidence of psychological problems with pre-school children who stutter so far contains conflicting reports. One of four small studies reported a difference between stuttering pre-schoolers and controls. The ELVS cohort showed no differences from controls for the Strengths and Difficulties Questionnaire during the pre-school years, but the Millennium cohort and the United States National Health Interview Survey showed that there were differences. Such findings so early during the developmental course of stuttering cannot be disregarded.

The ELVS cohort, although yet to be replicated and with limited methods to measure temperament, shows that children who begin to stutter show no signs of temperament markers of anxiety prior to or during early stuttering development. On balance, there is no consistent pattern of evidence that pre-school children who begin to stutter have temperamental markers of anxiety. Hence, findings of an association between early stuttering and unusual temperament are easily interpreted as an "epiphenomenon" reflecting the effects of the disorder, or simply as a benign co-occurrence with the disorder. As an example of the latter possibility, it would not be surprising if children with an anxiety-prone temperament became more anxious in response to early stuttering than children who did not have such a temperament.

On balance, it seems that anxiety, autonomic nervous system involvement, temperament, and executive function are associated somehow with early stuttering. Yet, to date, that body of literature has yet to yield an overarching and generally endorsed explanation of this material that is clinically useful. On the face of it, there are two broad possibilities that might emerge during coming years: Either the findings are an epiphenomenon or they reflect something about the nature and cause of stuttering. In either case, how the topic should influence clinical practices is far from clear at present.

The origins of social anxiety with stuttering: The school-age years and adolescence

Direct evidence

General Anxiety Scale for Children

The 1,000-family study[216] (see Lecture Two) presented extensive early data about anxiety with a group of 80 school-age children who stuttered and 80 controls ages 9–11 years, with mean age 10.5 years. The children received an extensive psychiatric evaluation, including the General Anxiety Scale for Children.[217] A limitation of those data, however, was that speech-language pathologists rather than psychologists or psychiatrists collected them.

State-Trait Anxiety Inventory

It was 30 years until further data about this matter emerged.[218] In the context of a clinical trial, the State-Trait Anxiety Inventory for Children[219] showed no significant pre-treatment differences between 77 stuttering and 20 control children, ages 9–14 years, with mean age 10.9 years. Neither group was unusual according to test norms. A subsequent report[220] replicated this finding with the same stuttering participants and an enlarged group of 106 control children 9–14 years old with mean age 11 years.

Another report[221] used the State and Trait Anxiety Inventory for Children with three groups who were slightly older: 18 who stuttered and were seeking treatment (10–16 years, mean 12.6 years), 17 who once stuttered but recovered (10–16 years, mean 12.7 years), and 19 controls (10–15 years, mean 12.9 years). There was no difference between the groups for trait anxiety, but the stuttering group scored higher for state anxiety in three of four hypothetical situations that were used in the test.

Another report[222] used the State Trait Anxiety Inventory[223] (for adults) with an older group: 19 stuttering adolescents who were seeking treatment and 18 controls between 11–18 years, with a mean age of 14.4 years. Results showed significantly higher state and trait anxiety scores for the stuttering group.

Fear of Negative Evaluation Scale

The report just mentioned[222] used the long version of the Fear of Negative Evaluation scale[224] with adolescents (mean age 14.4 years), and reported significantly higher scores for the stuttering group. However, a caveat to this finding is that the Fear of Negative Evaluation scale was developed for adults, not adolescents.

Revised Children's Anxiety and Depression Scale

A report of 132 children who stuttered, ages 9–14 years,[158] who presented at a speech clinic, were given the Revised Children's Anxiety and Depression Scale.[159] The scale has a child- and a parent-reported version, and measures anxiety and depression symptoms in 8- to 18-year-olds. It has five subscales related to anxiety. All the children's scores were below clinical thresholds.

Revised Children's Manifest Anxiety Scale

Revised Children's Manifest Anxiety Scale[225] data were reported with 18 stuttering and 18 control children in the age range 11–12 years.[226] Although some significant results were reported for the total anxiety and subscale scores, these results could not be interpreted as clinically significant because the children who stuttered were not beyond the normal range of scores. Another report[227] using the Revised Children's Manifest Anxiety Scale involved 36 stuttering adolescents who had received treatment and 36 controls aged 12–18 years, with a mean age of 14.3 years. As was the case with school-age children, the stuttering adolescents scored significantly higher; however, both groups were in the normal range for anxiety.

A report[228] of 23 stuttering school-age boys and girls, ages 6–11 years, and 50 adolescent boys and girls, ages 13–18 years, produced consistent results. The total anxiety score and subscale scores were within normal limits. However, the Revised Children's Manifest Anxiety Scale contains a lie scale, which is designed to detect deceptively positive responses where respondents present themselves in a favourable light. There was evidence that the boys had high lie scale scores, suggesting they may have

been concealing their true levels of anxiety. The researchers speculated that this might explain the many equivocal results about anxiety levels in these age groups.

Personal Report of Communication Apprehension

The Personal Report of Communication Apprehension[229] deals with fear of speaking, with subscales for public speaking, meetings, groups and conversations. A study[230] recruited 39 stuttering adolescents from speech clinics along with 39 controls, ages 13–18 years with a mean of 14.6 years. The adolescents with stuttering showed significantly higher apprehension scores than controls. This result was replicated with a study[231] of 36 adolescents seeking stuttering treatment, ages 11–18 years, mean age 14.2 years. There were no controls, but results for the adolescents were consistent with the first study.[230]

Strengths and Difficulties Questionnaire

The Strengths and Difficulties Questionnaire mentioned earlier[118] was used in a study[232] with 10 boys who stuttered and 10 controls, ages, 8–14 years. The authors reported a significant difference between the groups. The previously mentioned report from the Millenium Cohort[121] used the Strengths and Difficulties Questionnaire to show that 11-year-olds (N=194) reported to be stuttering by their parents differed from matched controls. The questionnaire was used in the United States National Health Interview Survey report mentioned earlier,[121] and significant differences were found for some survey domains with children ages 6–10 years and 11–17 years. Another report, with 35 boys who stuttered and 35 controls, ages 14–17,[233] found a significant difference for the survey domain dealing with peer relationships.

Child Behaviour Checklist

A study of Turkish children who stuttered and controls[141] included children in the ages 7–11 years and 12–18 years, with 15 stuttering children and 15 controls in both age groups. The Child Behaviour Checklist[234] was used and significant differences were found between the groups for nearly all eight scales of the test, notably "Anxious/Depressed."

A report of Kannada-speaking children

Kannada is spoken by around 70 million people in the south of the Indian subcontinent. A report[235] presented data from the Speech Situation Checklist-Emotional Reaction[236] for 100 children who stuttered and 275 controls, aged between 7–14 years. The test assesses anxiety associated with various speaking situations, and was translated into Kannada for the study. The children who stuttered had double the scores attained by the control children. Additionally, there was a developmental trend, with the older children who stuttered in the cohort having much higher scores than the younger children.

Multiple Anxiety Assessments

A report[237] presented a range of assessments for 37 adolescents who were seeking treatment for stuttering, ages 12–17 years and mean age 14.2 years. They were given a computerised, self-administered version of the Diagnostic Interview Schedule for Children.[238] Fourteen of them (38%) met diagnostic criteria for at least one DSM-IV[239] mental health disorder,[†] which is around twice the anticipated rate for adolescents, placing it in the 17–21% range.[240,241,242] Ten of these 14 diagnoses were a mental health disorder involving anxiety: social anxiety disorder, agoraphobia, obsessive-compulsive disorder, separation anxiety disorder, and specific phobia.

For three psychological test scores, the stuttering participant scores were in the normal range: Revised Children's Manifest Anxiety Scale,[225] Child Behavior Checklist,[234] and the Children's Depression Inventory.[243] However, there was a consistent trend for the older adolescents (15–17 years) to have more severe scores than the younger adolescents (12–14 years).

[†] The DSM-IV is the previous edition of the DSM-5, which was mentioned previously.

A study of 75 stuttering 7–12 year-olds and 150 matched nonstuttering controls[244] included the Youth Online Diagnostic Assessment.[245] Results indicated that, compared with controls, the stuttering group had four-fold increased odds for prevalence of any anxiety disorder. For social anxiety disorder there was a six-fold increased odds, with girls much more at risk than boys and with 24% of the children who stuttered diagnosed with the condition.

The Spence Children's Anxiety Scale Child Report[246] and Parent Report[247] showed scores within the normal range, but significantly higher values for the stuttering group. Mean scores for the Strengths and Difficulties Questionnaire Parent Report[248] were within normal limits, however the mean Total Difficulties score and Internalising and Externalising scores were significantly higher for the children who stuttered.

The largest cohort to date[249] involved 102 adolescents, ages 11–17 years, who were seeking treatment for stuttering. Psychological test scores were reported for the Revised Children's Manifest Anxiety Scale,[225] the Children's Depression Inventory,[243] the Youth Self Report and Child Behavior Checklist,[234] and the Assessment of the Child's Experience of Stuttering, which is an earlier version of the OASES-S (see Lecture Four). Stuttering severity measures were reported and showed that the relationship between them and psychological measures was not straightforward. Scores for depression and anxiety were within normal limits, but higher self-reported stuttering severity was associated with higher anxiety and internalising (emotional) problems. The boys showed externalising problems (rule-breaking and aggression) in the clinical range.

A report[250] from the ELVS cohort (see Lecture Two) when the children were 11 years old compared those who were stuttering with those who were not, using the Spence Children's Anxiety Scale Parent and Self Report,[246,247] the Strengths and Difficulties Questionnare,[118] and the School-Aged Temperament Inventory.[251] In contrast to the study of other Australian children reported around the same time,[249] no differences were found. A likely cause of that discrepancy is that one study involved children from a clinic and the ELVS report did not.

Physiological evidence

A study[252] of nine children who stuttered, ages 6–11 years (mean age 9.3 years), without a control group, compared salivary cortisol with normative data. Measures were made four times per day for three consecutive days. No evidence of abnormal levels was found; however, the authors noted that future research might take account of the many potentially confounding methodological issues with making such measures during childhood.

Anticipation of stuttering is common among those affected and is linked to anxiety (see Lecture One). It is possible—even likely—that unusual eye gaze patterns during reading are a physiological marker showing anticipation of certain words, and there is evidence that adults who stutter show such unusual eye gaze patterns during reading (see Lecture One). Three reports have shown that school-age children have such eye gaze patterns that are potentially consistent with anticipation of difficulty with certain words.[253,254,255]

Indirect evidence

Communication attitude

Negative communication attitude has been documented not only for pre-schoolers as outlined previously, but also for older children and adolescents. A detailed review of the topic is available.[256]

The Communication Attitude Test, often referred to as CAT, is the original scale developed for primary school-age children from which the KiddyCAT—described earlier—was derived. For the same reasons that the KiddyCAT is of interest in the context of potential anxiety development for pre-schoolers, the Communication Attitude Test is of interest for primary school children who stutter; it appears to focus on social anxiety.

There is a substantial body of research showing that children from different cultures and languages who stutter have higher scores than controls for this test. The Communication Attitude Test was used to assess 70 stuttering and 271 control 7–14 year old Belgian children.[257] From age 7 years the

children who stuttered had more negative attitudes to communication than peers. Additionally, their communication attitude worsened with time, whilst it became healthier with the controls. Other reports with 143[258] and 110[259] Belgian school-age children have replicated these findings.

These Communication Attitude Test differences have been replicated with Italian 7–14 year olds[260] and Croatian 7–13 year olds.[261] Eighty stuttering and 80 control Japanese school-age children, ages 5–12, also showed differences,[262] and there was evidence that the scores of the stuttering group continued to worsen across the ages studied, while the control group seemed to stabilise. Similar results have been found with Swedish[263] and Slovian[264] children.

Perceived communication competence

For those affected by stuttering during adolescence, the dimension of perceived communication competence might also intuitively suggest an indirect relation to social anxiety. There have been several reports using the Self Perceived Communication Competence scales.[265] The study that reported about communication apprehension[230] also reported data from the Self Perceived Communication Competence scales for the 39 stuttering adolescents from speech clinics and 39 controls (ages 13–18 years with a mean of 14.6 years). The stuttering group had significantly poorer perceived communication competence than the control group. The same research team replicated these significant results[266] with adolescents 13–18 years: a group of 53 receiving stuttering treatment (mean age 15.2 years) and 53 controls (mean age 14.8 years). The previously mentioned study of 36 adolescents[231] found similar results with the Self Perceived Communication Competence scales. Consistent results were found with interviews comparing stuttering and control children ages 5–10 years.[267]

Psychological distress

A birth cohort study[268] identified 217 adolescents who were stuttering at 16 years, according to their parents, 137 of whom completed the Rutter Malaise Inventory.[269] This is not a specific measure of social anxiety but "a 24-item self-completion scale which measures emotional distress such as depression and anxiety and related somatic symptoms such as headaches and tiredness" (p. 459).[268] The clinical history of the participants was not reported. Results showed that the adolescents with stuttering were more likely than the controls from the cohort to experience psychological distress, but not at levels that would put them at risk for clinically significant mental health disorders. However, one study has reported positive levels of self esteem for 48 adolescents who stuttered,[270] and another[271] has reported the opposite effect for 54 participants and controls.

Peer awareness

The earliest report that school-age children are aware of stuttering in peers was in 1958 with 120 children 5–8 years old.[272] They were presented with recordings of a story with and without stuttering, and the older children showed a preference for the latter and sometimes used the term "stuttering." An independent group replicated these results with a similar method two decades later.[273] In this study, 30 nonstuttering children, mean age 6.7 years, preferred to hear a story that was told without stuttering rather than a story told with stuttering. A second group of nonstuttering children, mean age 8.9 years, not only preferred the nonstuttered story, but volunteered the label "stuttering" when describing the other story.

The puppet research method described earlier for pre-schoolers was used with 79 nonstuttering children ages 3–7,[274] and reported consistent results. The children were able to identify with the nonstuttering puppet, again with that capacity increasing with age. It was telling that, from 4 years of age, the children began to offer negative evaluations of the stuttering puppet.

A study with 75 children, mean age 9 years 10 months, used the semantic differential bi-polar adjective pair method.[275] Half the children watched a video of an adult stuttering and the other half watched the same adult not stuttering. For 12 personality attributes, the children assigned significantly more negative scores for the video with stuttering. A study of 64 school children aged 10–14 years, mean 12.7 years, involved videos of a peer speaking at four different stuttering severities.[276] The

children rated the videos for a range of attributes dealing with "themes of peer friendship, listener comfort, and allowing a peer who stutters to take a speaking role in a group project" (p. 208).[276] There was a significant relationship between stuttering severity and the negativity of the peer responses. A similar study with 88 children ages 8–12 years reported the same relation between negative perceptions and stuttering severity.[277] A study of 62 Italian school children who stuttered and 474 controls, with mean age 11 years, reported that the children who stuttered were more rejected by and less popular with peers.[278] However, a report[279] of a small sample 22 stuttering school children, with a mean age of 14 years, in Flanders (Belgium) showed no evidence of peer rejection.

Bullying

Generally, being bullied during the school years is strongly associated with anxiety later in life,[280,281,282,283,284,285] and one report shows this effect concurrently for a control group and a group of adults with stuttering.[286] Hence the association of bullying with stuttering school-age children is of interest in the present context. One report[226] showed stuttering school-age children to have a 63% risk of being bullied compared to 22% for controls. Another report[287] of 28 children who stuttered, ages 7–15 years, found that 59% of them reported being bullied, and 38% reported it to have occurred on most days or every day. And another report[231] of 36 adolescents indicated that 63% reported being bullied less than once per week, but 37% reported it occurring at least once per week. With a study of 53 stuttering adolescents and 53 controls,[266] aged 13–18 years, with a mean age of 15.2 years, more adolescents reported being bullied: 43% compared to 11% for controls. The study discussed earlier,[244] with 75 stuttering and 150 control children, indicated higher scores for the children who stuttered on the Culture Bullying scale of the Personal Experiences Checklist Child Report.[288] The following two items were responsible for the significant result: "other kids make fun of my language" and "other kids tease me about my voice." A report of 54 stuttering adolescents and 54 controls[271] indicated significantly more bullying for the former group using the The Bully–Victimization Scale.[289]

Another report[290] concerned 403 nonstuttering children, mean age 11 years 9 months, who each had a stuttering classmate. The children were asked to categorise their stuttering peers. One of the categories was "bully victim," and 38% of children who stuttered were placed in that category compared with only 11% of nonstuttering children. Consistent with that finding, classmates thought stuttering peers to be less popular, without leadership potential, and more likely to be rejected than others. A study of 97 school children, aged 8–13 years[291] with a mean age of 10 years, used the Peer Attitudes Toward Children Who Stutter scale. The study reported that children who had contact with a stuttering child had significantly more negative attitudes to children who stuttered. The same research group replicated that finding with 760 children ages 6–12 years.[292]

Retrospective reports of bullying by adults have produced results consistent with the above findings. In one report,[293] pertaining mostly to the school years, 83% of 276 stuttering adults reported being bullied at school, with 18% reporting it occurred every day and 41% reporting a few times per week. Almost all respondents reported negative short-term effects of being bullied, and 46% reported long-term effects. These results were replicated with a survey of 324 adult respondents,[294] 82% of whom reporting being bullied at least once per week. Responses suggested that 84% of respondents had difficulty establishing friendships later in life because of the bullying. One report of 332 adults surveyed[295] reported that 56% said they were "affected a lot" by stuttering during the school years. There is evidence that such lasting effects are associated with cyberbullying.[296]

Impact of stuttering

These findings about bullying and mental health with stuttering school-age children are consistent with a report using the OASES impact measure for 50 stuttering 8–11 year-olds, and an adapted measure for controls.[297] The children who stuttered had significantly lower impact scores than peers. This was reflected in overall concern about their speech, increased behavioural and cognitive responses to their stuttering, and compromises to their communication in daily situations.

Conclusions

In contrast to pre-school children who stutter, there is much more direct evidence that the anxiety related mental health issues that affect adults who stutter begin during the school-age and adolescent years. There are signs that such problems worsen during this period, with findings of problem anxiety measures more typical of older participants in studies. Two reports[237,244] have found evidence during the primary school years and adolescence of the diagnosable anxiety related mental health disorders that trouble adults. The latter of these reports found evidence that 24% of school-age children presenting at speech clinics for stuttering treatment were diagnosed with social anxiety disorder. The earlier of these reports contained evidence of worsening anxiety test scores during adolescence. Evidence of bullying during the school years, and negative classroom experiences, are consistent with these findings. The most prominent anxiety disorder with adults who stutter—social anxiety disorder—is typically diagnosed during early adolescence,[298,299] with median onset at 13 years.[40] So it is not surprising that it is present for many school-age children who stutter, warranting referral to a clinical psychologist.

These findings about the early psychological effects of stuttering are consistent with a body of evidence that children who have speech and language disorders are generally at risk of developing mental health problems, many of them involving anxiety.[300,301,302] The latter of these reports was of 258 five-year-olds who were diagnosed with a speech or language disorder (only five were diagnosed with stuttering).[303] Controls had a 21% rate of psychiatric disorder, and the language-impaired group had twice that rate at 40%. At a 14-year follow-up the rate of psychiatric disorder had not changed. The authors concluded that "young adults with a history of early childhood language impairment have one of the highest rates of psychiatric disorder in the community" (p. 80).[302] That could certainly be said of stuttering.

Stuttering, mental health, and the timing of early intervention

Early intervention is by far the best clinical option for the disorder, as outlined during Lectures Six and Seven. Considering epidemiological data and evidence of the potential quality of life impairment from persistent stuttering, and the mental health evidence presented during this lecture, the following policy statement about the timing of early intervention seems justified.

Stuttering typically starts during the pre-school years and is a significant risk factor for mental health problems later in life, particularly social anxiety disorder. Such problems have been reported from 7 years of age, and are associated with long-term impairment of educational and occupational attainment. The origins of those mental health problems have been reported during the pre-school years for children who stutter: negative peer reactions, teasing, stigmatisation, social distress, and signs of emotional and behavioural problems. Although three-quarters of children may eventually recover naturally from stuttering, recovery rate during the first 18 months is estimated to be only 6–8%, as discussed during Lecture Two. However, it is not possible to predict whether an individual child will recover naturally. Consequently, after diagnosis, stuttering should be treated with an appropriate evidence-based treatment as soon as possible.

That position does not appear to be universally endorsed. A report of a 2019 European conference about stuttering treatment,[304] with delegates from 29 countries, raised the prosect of "active monitoring" after stuttering onset, rather than beginning an evidence-based treatment immediately. The conference convenors concluded as follows:

> However, one issue remained unresolved among us: whether to intervene immediately after onset or to delay intervention. And in the event that a decision is made to delay intervention, it seems that some clarity is needed about what "active monitoring" means and how it fits into that management plan. Is it pre-treatment counselling, is it a treatment intended to reduce stuttering, or is it a procedure to determine whether natural recovery occurs? (p. 9)[304]

That report was driven by professional researchers, but another report[305] that included 126 Norwegian speech-language pathologists, appeared to provide a somewhat different perspective. When surveyed about this matter with pre-school children, two-thirds of the speech-language pathologists "reported that they never recommend a 'wait and see' approach when they are initially contacted by a parent reporting that their child is stuttering" (p. 929). If this represents a differing view between professional researchers and professional clinicians, it is a potentially sobering situation.

Summary

Adults with stuttering who present at speech clinics, more often than not, will have clinically significant anxiety that requires intervention. Many such cases will require referral to a clinical psychologist. Such adult clients may warrant a DSM-5 diagnosis, notably social anxiety disorder. If an adult does have clinically significant anxiety, it reduces the chance of effective speech treatment. Clinicians need to be mindful of the possibility that techniques for stuttering control may be safety behaviours that sustain speech-related anxiety. Primary school age and adolescent clients seeking stuttering treatment are more likely than younger clients to experience clinically significant anxiety. There is evidence that the psychological problems associated with stuttering begin early during life. Consequently, after diagnosis, early stuttering should be treated with an appropriate evidence-based method as soon as possible.

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LECTURE ELEVEN: TREATMENT OF SOCIAL ANXIETY AND STUTTERING[†]

Speech-language pathologists and anxiety treatment

As noted during the previous lecture, some clients presenting at speech clinics with stuttering may have clinically significant anxiety that requires intervention. This has prompted statements that any such client with significant anxiety, regardless of whether it amounts to a diagnosable psychological problem, requires clinical management.[1,2]

That being said, not all clinicians will have the necessary training or experience to manage the social anxiety of stuttering clients. Treatment of anxiety is fundamentally in the professional domain of clinical psychologists and psychiatrists. Speech-language pathology professional preparation programs around the world vary in the extent to which they incorporate anxiety management training. However, alone, they are not a qualification to diagnose and manage anxiety disorders.

The authors of a tutorial about anxiety management procedures for stuttering clients[1] state that standard anxiety management procedures are not particularly complicated; however, they caution that they

> should only be used by SLPs *[speech-language pathologists]* who have had appropriate experience and/or training during their professional preparation and/or at some later stage, and that their use should be in accordance with the code of ethics of the individual SLP's *[speech-language pathologist's]* professional body. (p. 195–196)[1]

A report of a 2019 European conference about stuttering treatment,[3] with delegates from 29 countries, led the convenors to conclude that "there was unanimous agreement that SLPs are ideal personnel to provide basic CBT services for persistent stuttering" (p. 10). However, they cautioned that

> … SLPs [speech-language pathologists] of today have a frontier to negotiate. They cannot function without being informed by the field of clinical psychology and, it seems, without ready access to a clinical psychologist when assessing and treating children, adolescents, and adults with persistent stuttering who experience anxiety. (p. 10)[3]

Anxiety measurement for speech-language pathologists

Background

It is important to stress that detecting an anxiety problem and giving it a DSM-5 diagnosis is not simply a matter of administering formal assessments. Clinical psychologists and psychiatrists typically diagnose an anxiety disorder after a period during which they formally test, interview, and generally become familiar with a client. Such an assessment process would cover domains in addition to anxiety that are related to it, such as depression and stress. As noted during the previous lecture, adults seeking treatment for stuttering are often affected by social anxiety disorder. An overview of clinical measures for that specific disorder that clinical psychologists can use is available.[4] The following measures for social anxiety are suitable for administration by speech-language pathologists; they require no formal psychology qualifications to administer. However, they are not diagnostic tools for mental health disorders.

A specific caveat is needed for speech-language pathologists about measuring the anxiety of children, because it is a lot different to measuring the anxiety of adults. One complicating factor is the possible limitations of child report about anxiety. Because of this, it is generally agreed that parent reports are

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[†] Thanks to Ross Menzies and Lisa Iverach for guidance with this material.

essential input for assessing anxiety with children. Clinical psychologists administer tests, observe children, and interview them and their parents—and sometimes teachers—to form a diagnosis. It would be prudent for speech-language pathologists to screen children for anxiety to determine whether referral to a clinical psychologist is necessary.

The Unhelpful Thoughts and Beliefs About Stuttering (UTBAS) scales

Overview

The UTBAS scales provide a stuttering-specific measure of the unhelpful thoughts and beliefs that may drive social anxiety for those who stutter. The scale can be downloaded from the website of the Australian Stuttering Research Centre, along with translations into several languages.[5] A Japanese[6] and a Turkish[7] version of the scale have been validated. The UTBAS scales relate well to the OASES (see Lecture Four).[8]

Clinical psychologists and speech-language pathologists developed the scale[9,10] by producing a list of 66 commonly occurring unhelpful thoughts about stuttering expressed by those who stutter. To complete the scale, the client indicates how frequently each thought occurs using a scale of 1 to 5, where 1 = *never or not at all*, 2 = *rarely or a little*, 3 = *sometimes or somewhat*, 4 = *often or a lot*, 5 = *always or totally*. The numerical scores are added to obtain a total score between 66 and 330.

To supplement this basic scale, there are two other scales that measure how much clients believe each thought, and how anxious each thought makes them feel. The three UTBAS scales are referred to as UTBAS I, II, and III. It is an option to give all three scales and combine the scores for a total UTBAS score between 198 and 990.

Interpreting UTBAS scores

The table below shows means and standard deviations for UTBAS I, II and III, and the total UTBAS score for 140 adult stuttering participants.[10] Of these participants, 24% met 12-month criteria for a DSM-IV social anxiety disorder diagnosis.

UTBAS I		*UTBAS II*		*UTBAS III*		*TOTAL*	
MEAN	*SD*	*MEAN*	*SD*	*MEAN*	*SD*	*MEAN*	*SD*
165	*(52)*	*145*	*(53)*	*159*	*(62)*	*469*	*(160)*

A guide for clinical interview

Arguably, the UTBAS is of most value as a guide for questions during an interview to establish whether a client's speech anxiety might be clinically troublesome. For example, clinicians could adapt certain scale items to ask a client during an interview, "Have you ever thought that people would doubt your ability because you stutter?" (Item 1), "Do you ever have the feeling that people are focusing on every word you say?" (Item 8), "Do you ever think that your stuttering will prevent you from being successful?" (Item 15), or "Have you ever thought that most people view those who stutter as less capable?" (Item 34).

Based on the responses obtained, and based also on general questioning to determine whether the client might be anxious about speech, clinicians may wish to use the UTBAS and other anxiety measures to provide a quantitative indication of the client's anxiety.

Age range

The UTBAS scales were developed for adults but can be adapted for use with adolescents, with some minor wording changes to suit that age group. "I'm of no use in the workplace" is replaced with "I'm of no use in the classroom," and "I can't speak to people I find sexually attractive" is replaced with "I can't speak to people I find attractive." With adolescents, perhaps 16 and 17 year-olds, clinicians

might be reasonably comfortable using the UTBAS means and standard deviations for clients. This may not be so advisable with younger adolescents, and the scale is probably of limited use with school-age children.

The UTBAS-6

There is a six-item screening version of the UTBAS, known as the UTBAS-6,[11] which is presented in the Appendix to this lecture, as well as being downloadable from the website of the Australian Stuttering Research Centre.[5] This scale is recommended for routine clinical assessment of anxiety by generalist speech-language pathologists; the full UTBAS scales are more suitable for in-depth assessment by speech-language pathologists who specialise in stuttering and anxiety.

The six items are able to accurately reproduce the total score for each of the three subscales. The researchers who developed the scale recommend that when the total UTBAS-6 score falls in or above the fifth decile, the client should be referred for psychological assessment. However, this does not mean that a score below the fifth decile excludes a clinically significant anxiety problem. The researchers indicate that "the decision about referral to a psychologist will be based on a combination of UTBAS-6 scores, any other clinical measures, and clinical judgement" (p. 970).[11]

These are the test items of the UTBAS-6, covering negative thoughts in the domains of fear of negative evaluation (1–2), avoidance (3), self-doubt and lack of confidence (4), and hopelessness (5–6):

(1) People will think I'm strange.

(2) People will think I'm incompetent because I stutter.

(3) I don't want to go—people won't like me.

(4) I'll never finish explaining my point—they'll misunderstand me.

(5) What's the point of even trying to speak—it never comes out right.

(6) I'll never be successful because of my stutter.

The figure below shows an example of an UTBAS-6 form completed by a client with clinically significant anxiety.

Brief Version of the Unhelpful Thoughts and Beliefs About Stuttering Scales

	Using the following scale, please read each item below and circle the number which most accurately describes you in terms of: (1) how FREQUENTLY you have these thoughts, (2) how much you BELIEVE these thoughts, (3) how ANXIOUS these thoughts makes you feel.															
	1=never or not at all 2=rarely or a little 3=sometimes or somewhat 4=often or a lot 5=always or totally	How FREQUENTLY I have these thoughts					How much I BELIEVE these thoughts					How ANXIOUS these thoughts make me feel				
1	I'll never be successful because of my stutter	1	2	(3)	4	5	1	2	3	(4)	5	1	(2)	3	4	5
2	People will think I'm incompetent because I stutter	1	(2)	3	4	5	1	2	(3)	4	5	1	2	(3)	4	5
3	People will think I'm strange	1	2	3	(4)	5	1	2	3	(4)	5	1	2	3	(4)	5
4	I don't want to go – people won't like me	1	(2)	3	4	5	1	2	(3)	4	5	1	2	(3)	4	5
5	What's the point of even trying to speak – it never comes out right	1	2	(3)	4	5	1	(2)	3	4	5	1	(2)	3	4	5
6	I'll never finish explaining my point – they'll misunderstand me	1	(2)	3	4	5	1	2	(3)	4	5	1	2	(3)	4	5

The Fear of Negative Evaluation (FNE) scale

The original 30-item FNE scale

The FNE scale was originally published in 1969[12] as a 30-item self-report questionnaire where respondents indicate true or false to statements referring to the expectation and fear of negative evaluation from others. For responses that suggest social anxiety, one point is scored, and for responses that suggest no social anxiety, no point is scored. There are several reports published with FNE data for stuttering participants.[10,13.14,15,16,17]

The Brief FNE scale

Subsequent to the popularity of the 30-item version of the scale, several publications focused on abbreviating the measure to either a 12-item version or an 8-item version.[18,19,20,21,22,23] A general conclusion from this research is that the 8-item version is useable because it has similar properties to the original 30-item scale. The 8-item version is generally referred to as the BFNE-S.

Each of the eight items is scored on a scale of 0–4, where 0 = *not at all characteristic of me*, 1 = *a little characteristic of me*, 2 = *somewhat characteristic of me*, 3 = *very characteristic of me*, 4 = *entirely characteristic of me*. Numerical scores of each item are then summed to give a total score. The range of scores will therefore be 0–32.

These are the test items of the BFNE-S:

(1) I worry about what other people will think of me even when I know it doesn't make any difference.

(2) I am frequently afraid of other people noticing my shortcomings.

(3) I am afraid that others will not approve of me.

(4) I am afraid that other people will find fault with me.

(5) When I am talking to someone, I worry about what they may be thinking about me.

(6) I am usually worried about what kind of impression I make.

(7) Sometimes I think I am too concerned with what other people think of me.

(8) I often worry that I will say or do wrong things.

Age range

The 30-item FNE and the Brief FNE scales were developed for adults, and have not been adapted for younger clients. So the advice for using them with adolescents is essentially similar to that for the UTBAS. For older adolescents, age 16 or 17 years, it may be reasonable to use the norms that are available for adults, and the test items can be useful to guide a clinical interview about anxiety. However, it would be incautious to apply the available norms to younger adolescents or school-age children.

Interpreting Brief FNE scores

Sensitivity is the true positive rate and *specificity* is the true negative rate (see Lecture Two). One report[20] shows Brief FNE sensitivity and specificity values for identifying people with social anxiety disorder (Table 3, p. 826). Based on achieving an ideal trade-off between sensitivity and specificity, the table suggests a cut-off score of 25 for potentially clinically significant anxiety. This score provides sensitivity around 65% and specificity around 80%. In other words, a score of 25 gives a 65% chance of indicating a problem when there is one and a 20% chance of indicating a problem when there is not one.

The report[20] is probably worth reading prior to using the Brief FNE during clinical practice to screen for clinically significant anxiety. The authors point out that the user can consult Table 3 (p. 826)[20] to form a cut-off score according to individual need. So, for example, if a clinician wanted to identify as many clients as possible with clinically significant anxiety, and were not particularly concerned about making a mistake, a cut-off score of 15 might be used. That would give sensitivity of around 90% but specificity—an error rate—of around 40%. Screening always involves such a trade-off between sensitivity and specificity.

Subjective Units of Distress Scale (SUDS)

Previous measures discussed have dealt with *trait anxiety*, which is anxiety linked to temperament. However, the SUDS measures state anxiety, which is an immediate emotional responses to everyday experiences. The SUDS is usually attributed to the psychologist Wolpe during the 1960s.[24] Clinical psychologists today commonly use this scale to evaluate the distress experienced at a particular time

or during a particular situation, or to predict the level of distress for any coming situation. As such, it can be useful for state anxiety assessment during stuttering treatment for adults and adolescents.

Ratings can be made on an 11-point scale from 0–10 or a 101-point scale from 0–100. In either case, 0 = *no anxiety and* 10 or 100 = *extreme anxiety*. The SUDS is quick to administer, and can be used by clients for self-assessment during everyday speaking situations. The scale has been shown to be valid.[25]

For clinical purposes it would be appropriate for a speech-language pathologist to use the SUDS during treatment to determine the level of client anxiety experienced during speaking situations, or when thinking about those situations. This could provide information about whether stuttering reductions in those situations are associated with anxiety reductions, or whether anxiety treatment is needed in addition to speech treatment. The 11-point version of the scale is presented in the diagram.

Communication Attitude Tests: CAT, KiddyCat, BigCat,

There is a long history in the field about measurement of a dimension referred to as communication attitude. The idea of measuring such a construct can be linked, according to one account,[26] to the Anticipatory Struggle Hypothesis, which is an outdated causal perspective about the disorder, mentioned during Lecture Three. The Anticipatory Struggle Hypothesis is summarised in a prominent textbook[27] (p. 149–151). It posits that stuttering is driven and sustained by an early developing belief that speech is difficult. Two seminal developments in measuring such a construct in adults occurred during the mid 20th Century.[28,29] At present, there are three commercially available tests to measure the construct of communication attitude, spanning the ages of pre-school, school age and adults.

The Communication Attitude Test (CAT)

The Communication Attitude Test,[30] often referred to as CAT, is designed specifically for children who stutter who are 6–15 years old. It has been available for some time.[26] It requires children to respond *true* or *false* to 35 questions that include items which suggest that it focusses primarily on social anxiety: "people worry about the way I talk," "my classmates don't think I talk funny," "my parents like the way I talk," "I don't mind asking the teacher a question in class," "some kids make fun of the way I talk," and "I talk well with most everyone" (p. 73).[31] The Communication Attitude Test has been shown, in many cultures and languages, to distinguish between children who stutter and controls, as reviewed during Lecture Ten.

A systematic review dealing with measures of the psychological impact of stuttering on school-age children[32] included the Communication Attitude Test. The authors reported that there were few well-developed measures for this age group, and noted the problematic nature of this situation; psychological issues have been documented to emerge for stuttering children during this time of life (see Lecture Ten). Compared to other tests of psychological impact of stuttering, the Communication Attitude test had the most comprehensive support for its measurement properties.[†] The authors noted that there was no available evidence for a change of the test scores after clinical intervention.

The authors of the review[32] found that few measures of psychological impact of stuttering for school-age children had their developmental data published in peer-reviewed journals. Consequently, they

[†] The authors also cited the OASES for school-age children (see Lecture Four) as having empirical support of its development.

included the Communication Attitude test in the report, even though the only supportive data for it are reported in the commercially available test manual. This is a caveat for the use of the test: its supportive data are not peer reviewed and are only available for scrutiny when the user has purchased the manual.

The KiddyCat

This is a version of the Communication Attitude Test for children younger than 6 years.[33] As noted in Lecture Ten, its items suggest that it has some relation to social anxiety: Children are asked 12 *yes/no* questions, such as "is it hard for you to say your name," "do your words come out easily," and "do people like how you talk?" (p. 229).[34] As noted in Lecture 10, there are several reports dealing with pre-schoolers who stutter and control children.

The BigCat

This is an adult version of the Communication Attitude Test,[35] comprising 34 items, to each of which respondents indicate whether it is "mostly true" or "mostly false." The reliability and validity of the test, including its capacity to distinguish between adults who stutter and controls, has been demonstrated in participants from the United States,[36,37] Poland,[38] Iran,[39] and Kannada-speaking India.[40] As is the case for the Communication Attitude Test and the KiddyCat, the BigCat appears to focus on speech-related anxiety: ''My speech is as good as that of most people,'' ''I will usually have some trouble with my speech,'' ''my speech does not affect the way I interact with people,'' and ''I am self conscious about the way I speak'' (p. 202).

The Spence Children's Anxiety Scale

Overview

The Spence Children's Anxiety Scale has been shown to be reliable and valid, and it has comprehensive normative data available. The scale is well established, extensively used, accessible from a website without charge,[41] and available in many languages.

Child and parent version (8–15 years)

There are child and parent response versions for this age group,[42] the child version containing 45 items and the parent version containing 38 items. Examples of items are "my child worries about things," "my child is afraid of the dark," "my child complains of feeling afraid," and "my child worries about being away from us/me." Responses to items are scored using a four point scale, where 0 = *never*, 1 = *sometimes*, 2 = *often*, 3 = *always*. Individual item scores are used to calculate sub-scale scores for various domains of anxiety: generalized anxiety, panic/agoraphobia, social phobia, separation anxiety, obsessive compulsive disorder, and physical injury fears.

T-scores are available for the raw test scores. T-scores are rescaled so that the distribution has a mean of 50 and a standard deviation of 10. This enables comparison of results across the six subscales. A score of less than 10 above the mean is not considered to be concerning. The scale is not intended as a standalone diagnostic instrument:

> The SCAS is not intended as a diagnostic instrument when used in isolation. Rather it is designed to provide an indication of the nature and extent of anxiety symptoms to assist in the diagnostic process. It is recommended that clinicians use the scale in partnership with a structured clinical interview.[42]

However, speech-language pathologists could use the parent report scale for screening purposes to determine any need for a clinical psychology referral. In which case, it is not advisable for a speech-language pathologist to use the subscales for screening purposes, only the total score.

Pre-school version (3–5 years)

The scales have a version for 3–5 year-olds[43] comprising 28 parent report items that provide an overall anxiety measure. Parents follow instructions, which can be downloaded from the website.[41] Examples of test items are "has difficulty stopping himself/herself from worrying," "is scared of heights (high

places)," and "is afraid of crowed or closed-in spaces." Parents respond to each item using a 5-point scale to indicate the extent to which each of the 28 statements pertain to their children, where 0 = *not true at all*, 1 = *seldom true*, 2 = *sometimes true*, 3 = *quite often true*, and 4 = *very often true*. Individual item scores are used to calculate sub-scale scores for specific aspects of child anxiety: generalized anxiety, social anxiety, obsessive compulsive disorder, physical injury fears, and separation anxiety. A total numerical score is obtained from those responses. T-scores are available for the subscales and the total score. As is the case with the school-age version, it is advisable for speech-language pathologists to use only the total scores for screening.

The Preschool Anxiety Scale Revised

The Preschool Anxiety Scale Revised[44,45] is a parent-report anxiety measure of anxiety for children younger than 6 years. It consists of 28 items. For each item, parents select the response that best describes the child, using a 5-point scale, where 0 = *not at all true*, 1 = *seldom true*, 2 = *sometimes true*, 3 = *quite often true*, and 4 = *very often true*. A total score is obtained by adding scores for all items, with a maximum total score of 112. Individual item scores are used to calculate sub-scale scores for four categories: social anxiety ("worries that s/he will do something to look stupid in front of other people"), generalized anxiety ("has difficulty stopping him/herself from worrying"), separation anxiety ("would be upset at sleeping away from home"), and specific fears ("is afraid of insect and/or spiders").

The Preschool Anxiety Scale Revised is publicly available in English and seven other languages. Normative data are provided[44] for 764 mothers and 418 fathers of Australian 3–5 year old children. Cut-off scores are not provided for determining whether a child is in the clinical range. However, Table 3 (p. 405)[44] provides mean total and subscale scores based on mother and father report. Elevated scores compared to those means can be used to assist clinical judgement about the need for referral to a clinical psychologist.

Evidence-based anxiety treatment for stuttering

Cognitive Behaviour Therapy (CBT)

Cognitive Behaviour Therapy, known generally as CBT, is the flagship clinical psychology intervention for a range of psychological problems involving negative emotions such as anxiety, depression, and anger. It has been shown efficacious with a range of DSM-5 disorders. A search of the Web of Science database[46] shows thousands of publications dealing with the method. A tutorial[1] about CBT for anxiety with stuttering gives an overview of the four standard components of the treatment, with specific reference to stuttering: exposure, behavioural experiments, cognitive restructuring, and attentional training. An overview of Cognitive Behaviour Therapy is given on the Australian Association for Cognitive and Behaviour Therapy website.[47]

There is evidence from the 1970s that anxiety treatments such as desensitisation and meditation may benefit those who stutter,[48,49] and it is common to incorporate CBT, or components of it, within speech restructuring treatments.[50,51,52,53,54,55] There has been a report about the effects of exposure therapy on stuttering[56] using six participants in a multiple baseline design experiment. The exposure task was 10 sessions of speaking to an audience. Before the experiment, all six participants met criteria for diagnosis of social anxiety disorder, and after the experiment only one retained that diagnosis. However, the results are difficult to interpret because the participants also received varying sessions of progressive muscular relaxation, ranging from none to four sessions.

There have been only two conference reports about the value of an entire CBT treatment,[57,58] and only three published clinical trials that conform to the discipline standards for a clinical trial of stuttering treatment as outlined during Lecture Five.

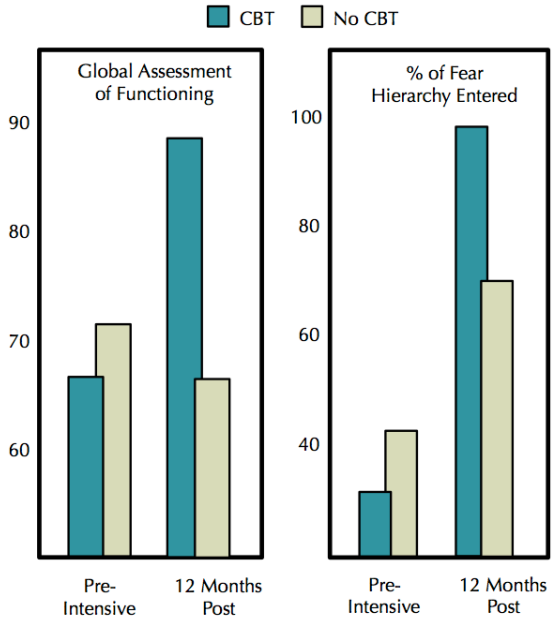
CBT for stuttering: A clinical randomised controlled trial

Design

The first of those trials[59] involved 32 stuttering participants, 60% of whom were diagnosed with social anxiety disorder. They were recruited to a randomised controlled trial of CBT adapted for the needs of stuttering clients. Participants were randomly allocated to receive a CBT package followed by an intensive speech restructuring treatment, or an intensive speech restructuring treatment alone. A clinical psychologist gave the CBT treatments, which were a standard 15 hours, in weekly sessions for 10 weeks. Seven sessions were 1 hour, one was 2 hours, and two were 3 hours. There were seven participant drop-outs (22%).

Results

The trial clearly showed that the addition of CBT to speech restructuring treatment did not reduce %SS scores at all. However, the trial showed no social anxiety disorder diagnoses and an overall general improvement of psychological functioning after CBT. Immediately after CBT, statistically and clinically significant improvements were reported for Global Assessment of Functioning. The DSM-IV index is a score out of 100 indicating general mental health and wellbeing, quality of functioning during daily life free of psychiatric difficulties, and engagement with the world. A clinical psychologist or a psychiatrist gives the score after a full diagnostic interview. Similar statistically and clinically significant results were found immediately after CBT for the 30-item FNE Scale, UTBAS scores, and the Social Phobia and Anxiety Inventory.[60]



At 12 months post-treatment, a statistically significant result for Global Assessment of Functioning scores remained. There was little change for participants in the control arm, but the participants in the CBT arm were in the normal range of psychological functioning post-treatment. Additionally, participants assembled a hierarchy of their least feared to their most feared speaking situation. At 12 months post-treatment, participants in the CBT arm were able, on average, to enter almost 100% of their fear hierarchies. The control arm showed improvement of this measure after speech treatment, but not to the same extent as the participants in the CBT arm. The difference between the groups was significant at 12 months post-treatment. These results are presented in the figure.

iGlebe: A standalone Internet CBT for stuttering

Clinical issues driving the development

Despite its promise, the clinical trial just described raises various clinical issues that researchers have noted.[61] Most obviously, a clinical psychologist gave the treatment. As discussed earlier, it may be appropriate for speech-language pathologists to provide CBT to clients who stutter if they have the appropriate professional preparation and training. However, this raises the matter of whether, on the whole, such speech-language pathology interventions would be as effective as those provided by clinical psychologists. It would be difficult to argue that this would be the case.

Another issue is the limited viability for every speech-language pathologist who manages stuttering caseloads to attain appropriate professional preparation for managing anxiety. Ideally, that preparation would involve formal postgraduate CBT qualifications, which is not a foreseeable prospect for all

speech-language pathologists who routinely manage stuttering caseloads. Nor is it a foreseeable prospect that speech-language pathology professional preparation programs worldwide will universally provide training to benchmark clinical psychology standards for CBT treatment. Nor is it viable that, in every case, clients who require stuttering and anxiety management would receive these services concurrently from a speech-language pathologist and a clinical psychologist.

Standalone Internet CBT as a solution

A potential solution to those problems is a standalone Internet-based CBT treatment for stuttering clients;[61] in other words, an Internet treatment program that does not require clients to have personal contact with a clinician. This is a common approach to mental health problems in clinical psychology.[62]

There are good reasons to foreshadow that a solution to this problem can be an interactive, Internet-driven CBT treatment that requires no clinician. Speech-language pathologists would be able to integrate CBT treatment with speech restructuring treatment without needing psychological training or access to a clinical psychologist, and they would be able to do so in a cost neutral manner.

A review[63] argued that Internet-based CBT treatment could fully replace a human therapist if treatments were customised to the individual user. Instead of a standard approach to treatment, the authors argued that an Internet-based treatment could begin with comprehensive assessment of individual client anxiety features, such as unhelpful thoughts and beliefs, as occurs with a standard clinic assessment. The authors also argued that human therapist simulation would require corrective feedback for incorrect client responses during the learning process of CBT, tracking of client access to the program with encouragement for compliance, and reminders for failures to log on.

Additionally, treatment "dose" was raised as an issue with standalone Internet treatments; users require "a large number of opportunities to engage in cognitive and behavioural tasks relevant to their problems" (p. 251).[63] Finally, the authors argued that the design of a successful Internet-based CBT treatment would need to incorporate features that simulate contact with a human therapist as much as possible.

Program design

Background

Incorporating those guidelines, the authors designed a standalone Internet CBT treatment: iGlebe.[†64] The program design is based on the original clinic treatment[59] and incorporates components of the Clark and Wells social anxiety model described during the previous lecture.[65,66] The program incorporates the faces and voices of a male and female clinical psychologist who communicate to the user throughout the treatment. The treatment is designed around an on-line pre-treatment assessment, conducted within the iGlebe program, which includes the 30-item FNE, UTBAS, and the Depression Anxiety Stress Scales.[67] A Stuttering Specific Avoidance Scale was developed specifically for the program. It presents the user with 55 common daily life situations, and requires each of the situations to be scored on a five-point scale where 1 = *never avoid* and 5 = *always avoid*. When users have completed the treatment they repeat these online assessments.

Section One

Section One introduces the user to the voices and images of the two clinical psychologists who present iGlebe. The program explains to the user the cognitive model of emotion, involving the

[†] In early publications the program was referred to as CBTPsych.

relationship between events, thoughts and emotions; events prompt thoughts, which prompt emotions. Examples are provided of how people are able to control their thoughts, and hence control emotions. Users are shown how both negative and positive thoughts and emotions may emerge from the same event. The example provided is of missing a bus because it departs ahead of schedule. The figure below shows how this event may promote thoughts that lead either to positive or negative emotions.

The iGlebe clinical psychologist then presents a "thinking exercise" where the user is presented with a life situation that does not involve speech. Following this, users are required to write different thoughts about the situation that could lead to the different emotions of anxiety, anger, sadness and happiness. The program then compares the user responses with the responses prepared by the clinical psychologist.

Next, the program presents a scenario where a woman asks a sales assistant for something, stuttering while doing so, and the sales assistant asks her to repeat the request. The user is required to repeat the thinking exercise and responses are again compared to the iGlebe responses. Of the given responses, one that would lead to anxiety is "the sales assistant thinks I am stupid," one that would lead to anger is "she has no right to treat me this way," one that would lead to sadness is "I'm hopeless, I can't do anything right," and a response that would lead to happiness is "I only had to repeat once what I was asking for." Similar thinking exercises recur throughout Section One.

iGlebe then shifts from how thoughts in response to events cause emotions, to the idea of causal thoughts: an idea or belief that of itself would cause an emotion. The program then links in to the three assumptions that underlie the Clarke and Wells model of social anxiety: excessively high standards of social performance, beliefs about performing in a certain way in social situations, and unconditional negative self beliefs. iGlebe presents examples of such causal thoughts.

The program continues with an explanation of common *cognitive errors*, otherwise known as *cognitive distortions*, such as those outlined in the following table. These are typically incorporated within CBT.[68] iGlebe presents numerous examples of these cognitive errors, such as "I am going to make a fool of myself at the party" and "I have to look fantastic all of the time" and the user is required to identify which of the cognitive errors they are.

MIND READING	*Assuming people are thinking negative thoughts about you when there is no real evidence that they are.*
FORTUNE TELLING	*Arbitrarily predicting that things will turn out badly.*
EMOTIONAL REASONING	*The way you feel about yourself is reality: "I feel stupid so I must be stupid."*
MENTAL FILTERING	*Dwelling on the negatives and discounting the positives.*
"SHOULD" THINKING	*Developing negative emotions about yourself and others based on internalised rules about the behaviours of others: "That shopkeeper should not have been rude."*
OVERGENERALISATION	*Interpreting negative events as part of a never ending pattern of defeat.*
ALL OR NOTHING THINKING	*Thinking in black and white categories with nothing between: "My spouse and I disagree on some things so we have a poor relationship."*

DISCOUNTING THE POSITIVES	*Belief that positive achievements don't count in evaluating yourself.*
LABELLING	*Thinking "I am a loser" instead of "I made a mistake."*
PERSONALISATION	*Blaming yourself for something that was not entirely your fault.*
BLAME	*Overlooking how your attitude and behaviour may contribute to problems.*

Section Two

This section uses the online UTBAS user scores from the pre-treatment assessment to create an individualised profile of unhelpful thoughts and beliefs about stuttering. It is a standard CBT technique to challenge such unhelpful thoughts that may cause negative emotions, which in this case is anxiety. These standard cognitive challenges are: (p. 264–265)[64]

(1) What evidence do you have for the thought?
(2) What evidence do you have against the thought?
(3) What would you tell a friend, to help, if he/she had the thought?
(4) Think of your calmest, most rational and supportive friend or family member. How would he/she react to the causal thought? What would he/she say?
(5) Are you worrying about an outcome you can't control? Is there any point to this type of worry?
(6) What does the thought do for you? How does it make you feel? Is it helpful or just distressing?
(7) What good things would you gain if you gave up the thought? How would your life be different if you didn't believe the thought?
(8) If the causal thought was true, what is the worst outcome? Is it as bad as you think?

There are 66 UTBAS items. For all of these items, iGlebe has cognitive restructuring sample answers for each of the eight probe questions in the above table, totalling 528 sample answers. In order to ensure an adequate dose of cognitive restructuring, iGlebe requires users to write at least 40 different restructurings of their unhelpful thoughts and beliefs. For each cognitive challenge that the user writes, iGlebe provides a predetermined challenge from its database for comparison. For example, in response to the unhelpful thought "It's impossible to be successful if you stutter," the following was a user's response to the probe question "what would you tell a friend, to help, if he/she had the thought?" (p. 264):

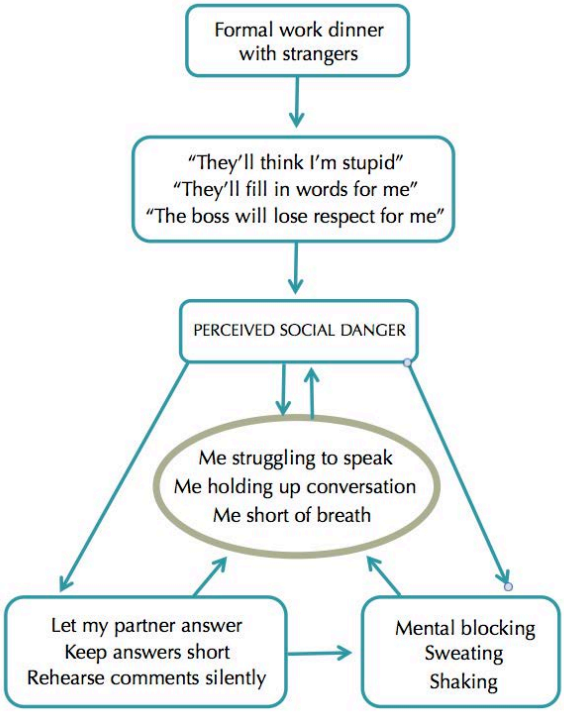
> It is absolutely wrong. You can be successful in many things despite you *[sic]* stutter, look at your past performance, you succeed sometime.[64]

The iGlebe sample answer was:

> Don't be silly! Lots of people who stutter are successful. This thought is so self-defeating. You need to beat it![64]

Section Three

This section is an extensive psychoeducation package based on the Clark and Wells model. Situation avoidance and safety behaviours are explained in detail, with examples. Users are then guided to build an individualised model of their anxiety, incorporating information from their pre-treatment assessments of unhelpful thoughts and beliefs and avoided situations. The guide includes avoided situations, thoughts that drive anxiety and avoidance, safety behaviours, mental self-images, and physical anxiety symptoms. When the user has constructed the model, it is used later in the program to establish behavioural experiments and to target unhelpful imagery for correction. The accompanying diagram gives an example of an individual formulation that might occur for a user.[‡] It relates to the social event of a formal work dinner with strangers.



The impending social event activates the assumptions outlined earlier: excessively high standards of social performance, beliefs about performing in a certain way in social situations, and unconditional negative self beliefs. These assumptions activate causal thoughts for anxiety such as "they'll think I'm stupid," "they will humiliate me by filling in words when I have a speech block," and "the boss will lose respect for me when I make a fool of myself in front of his colleagues." Such thoughts lead to a perception that the impending social event is dangerous. Safety behaviours are planned for during the dinner to avoid the feared outcomes: letting a partner do most of the talking, keeping any answers to questions short, and silently rehearsing every utterance before saying it.

During the dinner, negative self focus includes images of struggling to speak, holding up the conversation with stuttering, and being short of breath. These are compounded by a destructive cycle where safety behaviours have the reverse effect to what is intended by making the speaker appear odd, unfriendly, distant, or aloof, which worsens feelings of self consciousness, thereby feeding into a cycle involving negative self processing and anxiety symptoms of mental blocking, sweating, and shaking during the dinner.

Then, after the dinner there is rumination about how humiliating the whole event was and how awkward it would be speaking to the boss at work the next day. All that can be recalled is struggling to speak, holding up everyone's conversation, and gasping for breath while trying to speak. The dinner is added to a mental list of previous failures and confirms the expectation that such events in the future will be similar.

Section Four

This section presents behavioural experiments about feared situations. One of the iGlebe clinical psychologists says this to the users:

[‡] Adapted and reproduced with permission: Clark, D M (2001), A cognitive perspective on social phobia, In W R Crozier & L E Alden (Eds.), *International handbook of social anxiety: Concepts, research and interventions relating to the self and shyness*, (p. 405–430), Hoboken, NJ: Wiley. © 2001 John Wiley & Sons, Ltd.

> "This is a particularly important component of the treatment package, because it introduces you to behavioural experiments. In previous sections of the program, you've learned that social anxiety is driven by negative thoughts and maintained by safety behaviours and avoidance. Behavioural experiments are designed to test out your negative thoughts in situations where we will ask you to drop your avoidance and your safety behaviours. Behavioural experiments are a way to test out your thoughts or your predications about situations and they're fairly straightforward really to understand. You make a prediction about what will happen in a particular social situation. You enter the social situation and engage in a real way and you discover whether your prediction comes true or not."

The success of the technique relies on the fact that those who are socially anxious typically overestimate the likelihood and seriousness of a predicted negative outcome.

For behavioural experiments, iGlebe uses the avoided situations that each user recorded with the Stuttering Specific Avoidance Scale at the pre-treatment online assessment. Around 10 behavioural experiments are designed for each user. A list of 21 common predictions for those who stutter is presented, from which the user selects three. Examples of such predictions are "people will walk away," "they will not talk to me," and "I will forget what I am going to say." For each prediction the user uses a 100-point scale to indicate the perceived probability of its occurrence. iGlebe leads users to compare the actual outcome with the predicted outcome. Users are instructed to repeat behavioural experiments until they are no longer anxious in the avoided situations.

iGlebe has the capacity to create 3,620 different behavioural experiments for users based on their pre-treatment assessment data. Users are instructed to carefully avoid using their typical safety behaviours, as identified in Section Three, during behavioural experiments. The following is an example of a behavioural experiment provided by iGlebe and how it turned out.

A man has avoided going into banks because of a fear that he would not be able to make his needs known to the teller and that the teller would be condescending to him. His task was to go into a bank and make a deposit into his account. When he entered the bank he was anxious, and more so when he approached the teller. The teller greeted him in a friendly manner and asked how she could help. He stuttered a few times, but nonetheless was able to communicate his request. The teller made the deposit and courteously wished him a good day, without any sign of condescension. So, the outcome was different from the prediction. Even if the teller did notice the man's stuttering, it did not interfere with him achieving the purpose of going to the bank and it did not prompt any condescending behaviour.

Section Five

This section continues material from Section Two, which challenges fear of negative evaluation. Users are guided, by means of a sample essay, to write about "why it doesn't matter what other people think of me." One of the iGlebe clinical psychologists says the following as part of the preparation for this exercise:

> "When you care about the opinions that others hold of you, you're giving them tremendous power over your emotional life. You're saying in a sense that you can only be happy if they're happy with you, you can only feel good if they feel good about you. Giving somebody that much power over your sense of self worth doesn't make any sense if you really think about it."

The second part of this section targets unhelpful "should" cognitions, and the problems they cause. "Should" cognitions refers to internalised rules people have about the behaviours of others and the tendency to become angry if those rules are broken. The iGlebe psychologist informs users of the problems with becoming angry over something you think should not have happened but is now in the past and cannot be changed. Also, the user is informed that people cannot be prevented from behaving in ways you think they should not. Additionally, there are many different perspectives about how people "should" behave, and it is irrational to think that yours is the correct one.

iGlebe provides the example of a man stuttering severely with a shop assistant who said "hurry up, there is a long cue." This made him angry and affected his mood for the rest of the day, and could well have made him anxious about future dealings with shop assistants. He kept thinking "she shouldn't have been so rude." But there is no point in that thinking because it can't change what happened. A better outcome for him would have been just to accept that the shop assistant was tired, rude and insensitive, and too young to know any better, and for him to move on and not waste mental energy and emotions on someone he found to be so distasteful. The iGlebe psychologist points out that there are many different perspectives about this scenario and the man's perspective is just one of them. Other perspectives are that the shop assistant was being considerate to other shoppers, that she had the right to do her job as she sees fit, and that she had the right to free speech. There is nothing innately correct about the man's view that "she shouldn't have been so rude."

iGlebe targets maladaptive "should" thinking by requiring users to select three of 17 "should" cognitions commonly associated with anxiety. For each of the three selected "should" statements, the program guides the user to explore the advantages and disadvantages of each. Users are then required to choose three of the "should" cognitions and to construct their own narrative for each of them. To assist the user, iGlebe provides 34 different sample responses for each of the common "should" cognitions.

Section Six

This section is designed to repair the imagery that leads to a negative self-focus during social situations and to establish a different perception of social encounters. There are four projected benefits from obtaining a healthy control of attention. First, it is intended that gaining control of imagery about social encounters will prevent the post-event rumination that can distress those with social anxiety and perpetuate the anxiety. Second, the problem of distorted observer perspective is targeted so that users obtain a correct picture of how people really respond to them in social situations. Third, attention will focus away from any negative events in a social encounter, which are likely to be minor, towards neutral or positive aspects of the encounter. Finally, it is projected that users will break their cycle of failing to disconfirm negative expectations during social encounters and will be able to find evidence that disconfirms those beliefs.

The first step is skills-based attentional training,[69] which first trains the user to control where attention rests at any moment, using the attentional training technique. The user downloads an audio file from iGlebe, in which the psychologist's voice provides training in shifting attention rapidly from one focus to another. When the user has practised the attentional training technique daily for some weeks and mastered it, the iGlebe clinical psychologist introduces the situational attention refocusing technique:

> "Situational attentional refocusing builds on your ability to place your attention where you wish. There is considerable research … to suggest that anxious individuals place too much of their attention on negative aspects of social settings. I'm sure you've experienced this. Where attention seems caught by one negative person or one negative aspect of the environment, one person who you think is being critical of you. You don't seem to be able to focus on anything else. Well we want you with your new attentional skills to enter social spaces in an unbiased way, moving your attention through the positive aspects of the situation."

iGlebe then presents users with a list of their commonly avoided everyday situations, scored in the online pre-treatment assessment with the Stuttering Specific Avoidance Scale. The user is then required to choose three of these situations and practise the situational attentional refocusing technique for each of them and to record what occurred during each practice. The user is urged to continually practise this technique.

The final part of Section Six deals with problems of mental imagery that involve the biased observer perspective, which often affects those with social anxiety, as discussed during Lecture Ten. The iGlebe clinical psychologist says to the user:

> "Research has consistently shown that people with social anxiety lay down distorted images into their memory. Now this is very important because anxious individuals are basing a lot of their fear on going into social situations on their past memories on how they performed … the memories that socially anxious individuals have include images of what they actually looked like in the events in which they were anxious … they remember seeing themself performing in the social event as if they had been an observer to the event. Now … obviously these negative memories must be false; no one sees themselves when they speak."

In this part, iGlebe presents an example of a social encounter during which stuttering occurs. The images of this encounter recur for the person two years later. The woman recalls seeing herself looking anxious and tense during the event and those present evaluating her quizzically and apprehensively, and generally having a negative view of her, as shown in the image below on the left.

However, this recall of the situation cannot be correct and what actually occurred—the field perspective—was more like the image on the right. Yet, years later, she still ruminates about how badly she thought the social encounter went and the thought of it all makes her ruminate about how badly future social encounters will turn out how and makes her anxious about them before they even occur. The iGlebe program invites users to test whether this is an issue for them. The iGlebe clinical psychologist says to the user:

> "Do you think you show this "observer" or "external camera" bias? You can test this out by simply closing your eyes and remembering images from past anxious speaking situations. Do you see yourself in the image, or are you simply seeing the faces of those around you? Remember, if your memory is displaying images of your own face, it is playing tricks on you! Such images simply cannot be accurate! Unless you are telling us that you were standing in front of a large mirror in these social events, you simply could not see yourself doing anything!"

The final part of Section Six deals with the "re-scripting" technique[70] for faulty images of past events such as in the previous example. Users download an audio file from iGlebe, in which a psychologist's voice provides guidance to mentally go over a past event and re-script it so that it is different from the troublesome version; the mental image becomes one where the social event is going well, people are smiling and enjoying your company, and you are not stuttering. Users are instructed to repeat this exercise several times for each false and biased memory of a social event.

Section Seven

This final section deals with relapse prevention. It emphasises that minor setbacks are inevitable and should not be interpreted as relapse. The clinical psychologist guides users to recognise when they are vulnerable to anxiety setbacks, such as at times of stress or fatigue. The critical point is made that

falling back into avoidance patterns never helps with anxiety setbacks; avoidance only perpetuates and worsens anxiety.

Phase I clinical trials of iGlebe

Background

The developers reported preliminary data for two adult participants.[64] Participant 1 completed the treatment in 2 months with 11 log-ins, and Participant 2 completed the treatment in 3.5 months with 34 log-ins. Results suggested that treatment effects with this intervention may be similar to those attained with the clinic version that was presented by a clinical psychologist.[59] At post-treatment, both participants no longer had social anxiety disorder diagnoses and various anxiety measures showed improvements.

Method

Subsequently, the prototype of the website was refined, and a Phase I clinical trial was reported[61] with 19 adult participants recruited from a speech-language pathology clinic waiting list. Five of these (26%) did not begin the treatment, leaving 14 participants, who were permitted 5 months to complete the treatment. Their average age was 42 years (range 33–77 years). Several psychometric assessments were collected pre-treatment and at 5 months after the start of the treatment, independently of the measures collected by iGlebe.

The presence or absence of social anxiety disorder was assessed with a standard, self-administered computer assessment.[71] Additionally, participants completed the 30-item FNE scale, the UTBAS scale, the Beck Depression Inventory,[72] the State-Trait Anxiety Inventory,[73] the Social Phobia Anxiety Inventory,[60] and the Endler Multidimensional Anxiety Scales-Trait.[74] Impact of stuttering was measured with the OASES, and stuttering severity was measured with %SS from two unscheduled 10-minute telephone calls from strangers.

Results

Users had a mean of 15 log-ins with a mean log-in time of 7 hours and mean period between log-ins of 7 days. However, as outlined earlier, much of the iGlebe clinical procedures occur during everyday situations when users are not logged in. Eight of the 14 participants completed all seven iGlebe sections during the 5-month access period. Users did not have any contact with a clinical psychologist or a speech-language pathologist during the trial.

At pre-treatment, seven participants met diagnostic criteria for social anxiety disorder, and at post-treatment only two retained that diagnosis. However, these two participants did not complete the entire iGlebe treatment, one progressing only to Section Two and the other to Section Four. Two of the four psychometric measures showed statistically significant improvement at post-treatment: the FNE scale and the Social Phobia Anxiety Inventory. UTBAS scores OASES total scores, and OASES quality of life subscale scores showed significant improvement. Percentage syllables stuttered scores showed no change from pre-treatment to post-treatment.

A Phase II clinical trial of iGlebe

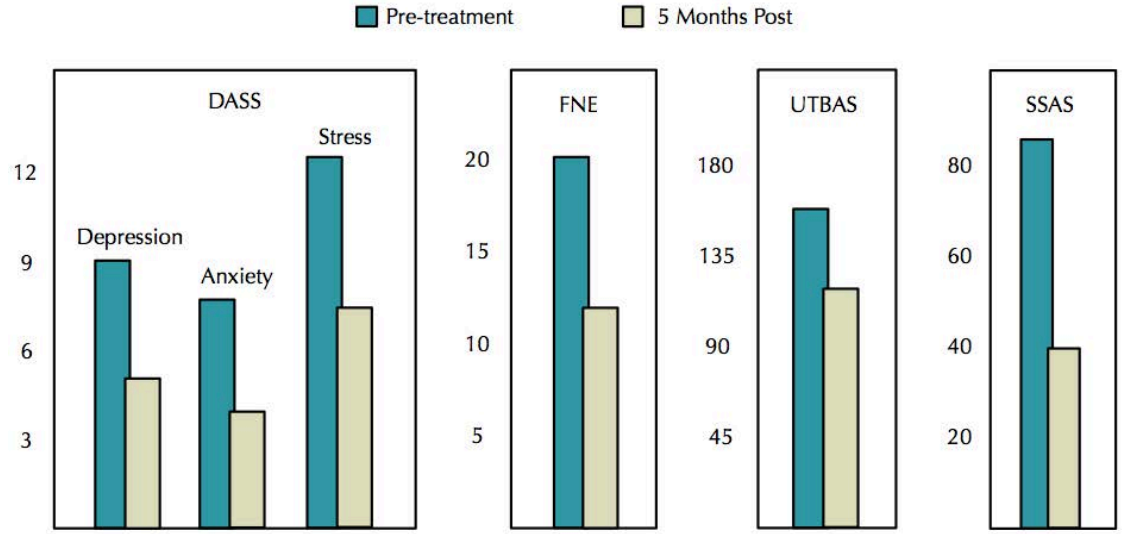
Background

In formulating a Phase II trial of this standalone Internet treatment,[75] the developers noted that the Phase I trials involved pre-treatment and post-treatment assessments at speech clinics. In effect, then, the trials were not standalone in the strictest sense, because such clinic contact may have been somehow associated with participant compliance. Hence the Phase II trial involved no direct participant contact of any kind from researchers or clinicians.

Method

This was an international non-randomised trial with 267 participants who reported a history of stuttering and were given 5 months access to iGlebe. Participants were recruited from 23 countries, with the majority from Australia, The United Kingdom, Canada, the United States, New Zealand, and

South Africa. Participants completed pre-treatment and post-treatment assessments from within the program: The Depression Anxiety and Stress Scale, the Fear of Negative Evaluation Scale, the Unhelpful Thoughts and Beliefs about Stuttering Scale, and the Stuttering Specific Avoidance Scale.



Results

Of the 267 participants recruited, 30 did not log on, 185 did not complete Section 7 within 5 months, and three completed all sections but did not complete post-treatment assessments. Hence, the completion rate for the trial was 18.4% (49 of 267 participants) and the completion rate for iGlebe was 19.5% (52 of 267 participants). This completion rate was far superior to existing standalone Internet treatments for depression and anxiety, which attain below 7%[76] and around 1%.[77]

Without any contact from a researcher or a clinician, statistically significant pre-treatment to 5 months post-treatment reductions were reported for all measures. These results were similar to the Phase I trials of iGlebe and trials of in-clinic CBT for stuttering with a clinical psychologist. Post-treatment scores for the Depression Anxiety and Stress Scale were within normal community values. Results are shown in the figure above.[‡]

A Phase II clinical trial of iBroadway with adolescents

Background[78]

The authors of this report argued that anxiety associated with stuttering is likely to be developing during early childhood. Hence, they adapted the iGlebe program for adolescents: iBroadway.

Method

Participants were 21 adolescents, ages 12–17, years who were seeking treatment for anxiety about their stuttering. Outcomes were the Diagnostic Interview Schedule for Children,[79] the Revised Children's Manifest Anxiety Scale,[80] the Children's Depression Inventory,[81] the Subjective Units of Distress Scale (discussed earlier in this lecture), the UTBAS scale[9,10] (modified for adolescents), the OASES (see Lecture Four), parent-reported speech satisfaction, parent reported typical and worst

[‡] Adapted and reproduced with permission: Menzies, R et al (2016), International Phase II clinical trial of CBTPsych: A standalone Internet social anxiety treatment for adults who stutter. *Journal of Fluency Disorders, 48,* 35–43.

stuttering severity, self-reported speech satisfaction and avoidance of speaking situations, and self-reported typical and worst stuttering severity. Post-treatment measures were collected 5 months after participants first accessed the program.

Results

Eleven of the 21 participants completed the iBroadway modules within the allocated 5 months, which was a favourable compliance rate for internet CBT treatment. For participants who completed the modules, there was a significant post-treatment reduction of mental health diagnoses with the Diagnostic Interview Schedule for Children. There were significant post-treatment decreases for the Subjective Units of Distress Scale, the UTBAS scale, and parent reported speech satisfaction. The authors are planning continued development of the iBroadway program and further clinical trials.

A randomised trial of iGlebe compared to in-clinic CBT

Background

This trial[82] was designed to evaluate the relative effects of iGlebe and CBT treatment by a clinical psychologist.

Method

The design was a two-arm noninferiority randomised controlled trial; a method designed to determine whether one treatment is not inferior in effects compared to another. Assessments occurred at pre-randomisation and at 6 and 12 months post-randomisation. Participants were 50 adults with stuttering who were seeking anxiety treatment, 23 of whom were randomised to receive iGlebe treatment, and 27 of whom were randomised to receive in-clinic CBT by a senior clinical psychologist. The iGlebe treatment involved 5 months access to the program, and the in-clinic treatment involved from four to 11 one-hour weekly treatment sessions.

The primary outcomes were number of mood and anxiety disorders determined with a self-administered computer interview,[83] and the Brief FNE scale. Secondary speech outcomes were %SS based on two 10-minute, unscheduled telephone calls where participants conversed with a stranger, and a self-reported typical SR score for the previous week. Other secondary outcomes were the OASES and the Social Phobia Anxiety Inventory.[84]

Results

Of the 23 participants randomised to receive iGlebe treatment, 18 were available for assessment at 12 months post-randomisation. Of the 27 participants randomised to receive in-clinic treatment, 24 were available for assessment at 12 months post-randomisation. Missing data at 12 months post-randomisation were accounted for by last observation carried forward (see Lecture Five). All psychological outcomes showed a consistent medium effect size with no evidence of inferiority of the iGlebe treatment on any outcome variable.

Improved self-reported SR scores at 12 months post-randomisation provided the first suggestion in the literature that CBT can improve stuttering severity. However, this result was not at all definitive, because it was not reflected in %SS scores. As the authors stated, this could have been because the %SS measure was not valid, or because "CBT treatment produced a favourable change in the way participants perceived their stuttering severity, which prompted a posttreatment lowering of their perceived severity" (p. 10). As the authors also noted, the clinical importance of this issue warrants further research.

A randomised trial of iGlebe supplementing speech treatment

Background

The authors designed this trial[85] to determine whether iGlebe added to the benefits of speech restructuring treatment.

Method

The design was a two-arm randomised experimental clinical trial. Assessments occurred at 6 and 12 months post-randomisation. Participants were 32 adults recruited from a stuttering treatment waiting list. Participants in both arms received Stages 1 and 2 of the Camperdown Program speech restructuring treatment in a 3-day version (see Lecture Eight). The treatment was presented without any components dealing with anxiety. In other words, the speech treatment was exclusively focused on stuttering control. The treatment included a 1-hour follow-up session each month for 5 months. Participants in the experimental arm were given access to iGlebe for 5 months after the 3-day component of the Camperdown Program treatment.

The primary outcome was %SS based on two 10-minute, unscheduled telephone calls where participants conversed with a stranger. Secondary speech outcomes were determined from typical and worst self-reported SR in eight standard speaking situations. Other secondary outcomes were avoidance of speaking situations, the OASES, UTBAS, the Brief FNE scale, the Social Phobia Anxiety Inventory,[84] and number of mental health diagnoses measured with a self-administered computer interview.[83]

Results

Approximately a third of participants completed the program, and the authors noted that this was a better compliance rate than a previous trial.[75] At 12 months post-randomisation the groups that had access to iGlebe had significantly better results than the group that did not. This improvement occurred for typical and self-reported SR and OASES. The result was found to be robust using a statistical technique called multiple imputation to adjust for missing data due to the 18% compliance rate.

It is difficult to interpret this trial of iGlebe because, for some unknown reason, the sample was unusual for treatment-seeking adults, with only five participants having a mental health diagnosis. Improved self-reported SR scores were consistent with the finding of the randomised trial discussed previously.[82] This second trial of IGlebe provided another suggestion in the literature that CBT can improve stuttering severity. However, this result was not at all definitive, for the same reasons that were discussed earlier with the other trial. Clearly, this issue requires detailed exploration with a different research method in order to clarify it.

Acceptance and commitment therapy

Subsequent to the developments of Behaviour Therapy and Cognitive Behaviour Therapy, there has been a so-called "third wave" or "third generation" of cognitive behavioural therapies.[86] One of these is Acceptance and Commitment Therapy, commonly known as ACT. A user guide to ACT is available.[87] This treatment differs from CBT because it focuses on "awareness, acceptance, and understanding the context of thoughts rather than challenging and changing their content" (p. 123).[88] Acceptance and commitment therapy, along with several other third wave therapies, has in common with CBT that it incorporates mindfulness training,[†] although with a greater emphasis. A definition of mindfulness is "the awareness that emerges through paying attention on purpose, in the present moment, and nonjudgmentally to the unfolding of experience moment by moment" (p. 145).[89] The overall purpose of ACT is

> to undermine the grip of the literal verbal content of cognition that occasions avoidance behavior and to construct an alternative context where behavior in alignment with one's value is more likely to occur. (p. 651)[86]

A recent review of the efficacy of ACT was[90] a meta-analysis of 18 randomised controlled trials (N=917). The authors concluded that the treatment was promising, being superior to control conditions. However, there was no evidence of it being superior to established treatments. Another

[†] The attentional training in Section Six of iGlebe described earlier is a mindfulness procedure.

review around the same time,[91] in the context of a general review of third wave therapies, was more guarded, reviewing 13 randomised controlled trials and drawing attention to methodological problems with them, and noting a moderate effect size for ACT. A more recent review by the same author[92] was a meta-analysis of 60 randomised controlled trials, and which reported no methodological improvements in trial quality and a reduction to a small effect size.

A preliminary case study explored ACT with eight adults,[93] followed by a speech treatment program, but reported no striking effects. There has been a description of how ACT may pertain to those who stutter,[94] and a subsequent description of an ACT package tailored specifically for stuttering,[95] and a data-based report about a similar program.[96] In the latter report, 20 participants received eight 2-hour group therapy sessions, with 10 participants per group. The report is difficult to interpret because participants received a combined package of speech treatment and ACT. As such, any psychological improvement could have occurred because of the speech treatment rather than the ACT treatment. The speech treatment was described as "fluency shaping activities, speech rate control, speech naturalness and self-administered timeout for stuttered moments" (p. 291).[96] Results at 3 months follow-up showed statistically significant improvements for stuttering severity during speech at the clinic while talking to a clinician, and improvements of OASES scores. Improvements were also shown in psychometric measures reflecting the success of the ACT therapy process. However, replications of the effects of ACT on stuttering will need to occur before it can be compared with what is known about the effects of CBT.

Another pertinent report[97] is difficult to interpret. Ten stuttering participants were randomly allocated to receive CBT or CBT plus mindfulness training. The report presented data for the entire 10 participants, showing improvements across a range of psychological measures. However, no data were presented to suggest that the addition of mindfulness training improved the effects of CBT.

Inquiry Based Stress Reduction

A randomized controlled trial[98] explored an intervention known as Inquiry Based Stress Reduction. This is a meditation technique that identifies thoughts associated with stress. Then, in a meditative process, those thoughts are cognitively challenged and "reframed" into opposite thoughts. The intention is to "to experience situations that were previously perceived as stressful with peace of mind and connectedness" (p. 4).[98]

Participants in the trial were 65 adults who stuttered, recruited from the Israeli Stuttering Association, social networks, and advertising. Twenty-eight were randomised to the treatment group and 28 to a no-treatment control group. The treatment was provided to the experimental group for 12 weekly, group sessions of 3.5 hours, with 14 participants in each group. Assessments occurred pre-treatment, at the end of treatment, and at 1-month follow-up. Assessments were the OASES-A, the State-Trait Anxiety Inventory,[99] the Psychological Flexibility Questionnaire ,[100] and the Satisfaction-with-Life Scale.[101]

Results showed statistically significant improvements in all measures at 1 month follow-up for the experimental group compared to the control group. Significant improvements occurred for the four OASES subscales as well as the Total Score, which was 3.1 pre-treatment and 2.3 at 1 month follow-up. This represents a change from moderate-severe to moderate impairment.

Although the follow-up period was short at 1 month, these results are promising and require replication. They appear to have potential economic benefit, considering that groups of 14 participants received 14 hours of treatment, which amounts to 3 hours of treatment each.

Summary

Some clients who present at speech clinics with clinically significant anxiety will require intervention for it. This presents a challenge for speech-language pathologists, for whom anxiety management is typically not a primary professional domain. However, there are anxiety measurement procedures suitable for speech-language pathologists, who may wish to provide anxiety treatment with appropriate experience and professional preparation. There is evidence that cognitive behaviour

therapy is efficacious for treating the social anxiety of those who stutter. Additionally, there is evidence that a standalone Internet social anxiety treatment is efficacious. Hence, speech-language pathologists might recommend it for their clients; it has no cost and requires no clinical psychology expertise. This could prove to be a significant advance for speech-language pathologists who do not have professional qualifications for anxiety management.

Appendix

The UTBAS-6 scale[5]

UTBAS-6

Brief Version of the Unhelpful Thoughts and Beliefs About Stuttering Scales

Using the following scale, please read each item below and circle the number which most accurately describes you in terms of:

(1) how FREQUENTLY you have these thoughts, (2) how much you BELIEVE these thoughts, (3) how ANXIOUS these thoughts makes you feel.

	1=never or not at all 2=rarely or a little 3=sometimes or somewhat 4=often or a lot 5=always or totally	How FREQUENTLY I have these thoughts					How much I BELIEVE these thoughts					How ANXIOUS these thoughts make me feel				
1	I'll never be successful because of my stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
2	People will think I'm incompetent because I stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
3	People will think I'm strange	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
4	I don't want to go – people won't like me	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
5	What's the point of even trying to speak – it never comes out right	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
6	I'll never finish explaining my point – they'll misunderstand me	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5

Note: Items 1-6 of the UTBAS-6 are taken from the original UTBAS scales (items 15, 19, 23, 35, 50, and 60, respectively)

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