Applications of Stuttering Treatment Research
Proceedings of the Third Croatian Symposium

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1951–2015

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Preface

Together, some time during 2007, Suzana Jelčić and I had the idea of a Croatian symposium about stuttering to be held in the Adriatic town of Cavtat. The first Symposium occurred during 2010, then a second during 2013, and a third, which is documented in the pages that follow, during October 2016. With planning under way at the time of writing for a fourth Symposium during 2019, it looks as if the event is destined to join the regularly occurring round of gatherings on the conference calendar of our discipline.

The purpose of these Symposia has always been to convey to speech-language pathologists the recent research findings about the disorder of stuttering, and how to apply them when those affected by the disorder, and the parents of children affected by the disorder, seek health care. That purpose was brought most keenly into focus with instructions to the presenters here to outline the importance of their topic, to state recent research findings about their topic, and to state clearly how the research findings can be applied to clinical practice. Their presentations are organised around such headings.

The format for the three days of the Symposium was that after each presentation the delegates—most of whom were speech-language pathologists—broke into groups with a discussion leader to consider the merits of what they had heard and how it might influence what they would do in their clinics. Then in the afternoon the delegates regrouped and, with the guidance of the discussion leaders, engaged the presenters with a moderated interaction about what they had said that morning.

It seems that everyone involved succeeded in achieving what was intended. A post-symposium survey of 56 delegates showed that 82% responded "strongly agree" to the statement "I was satisfied with the format of the Symposium," and 74% agreed with the statement "I will change my clinical practice after attending the Symposium." Delegates were asked to rate the conference with with a scale where "9" indicated "the best conference you have ever attended." We were gratified with a mean score of 7.7, and that 20% of delegates scored "9" and 38% scored "8."

Dave Rowley contributed much to the development of these Symposia during the past years, and we conducted the 2016 event bearing the crushing sadness of his untimely passing during the previous year. We felt him near us as always, and still do.

Mark Onslow

Stuttering, Temperament, and Anxiety in Preschool Children

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Abstract

Certain dimensions of childhood temperament are a risk factor for internalising difficulties such as anxiety. School-age children and adults who stutter with a history of clinic attendance have an elevated risk of anxiety, particularly social anxiety. Concomitant anxiety in adults who stutter increases the risk of stuttering relapse following speech therapy and can significantly impair quality of life. To ensure that best practice is provided, and to maximise stuttering and psychological therapy outcomes, it is critical to understand whether preschool children who stutter are characterised by temperamental traits that may predispose them to anxiety in later life.

Literature regarding temperament and anxiety in stuttering will be reviewed, including hypotheses that seek to describe their association. Although findings from cross-sectional studies remain inconclusive, evidence from prospective longitudinal research indicates that temperament during early childhood is not a precursor of stuttering onset. However, stuttering may be a precursor of anxiety onset in later childhood, adolescence or adulthood. Clinical implications are discussed. The preschool years represent a critical period in terms of eliminating stuttering and the subsequent development of anxiety. Further research is required to understand additional risk factors for the development of anxiety in stuttering.

Clinical importance of the topic

The preschool years

Stuttering typically starts in the preschool years (Yairi, Ambrose, & Cox, 1996). This is also the time when stuttering treatment is likely to be most effective. The negative consequences of stuttering may also commence in early childhood (Weidner, St Louis, Burgess, & LeMasters, 2015). For instance, preschool children who stutter may show a more negative attitude to communication than non-stuttering children (Vanryckeghem, Brutten, & Hernandez, 2005), and there is evidence that they may be interrupted, mocked, walked away from, and ignored by fluent peers (Langevin, Packman, & Onslow, 2009). These negative social experiences continue through childhood and adolescence to include teasing, bullying, social isolation, and rejection. These experiences may also lead to the development of anxiety (Iverach, Menzies, O'Brian, Packman, & Onslow, 2011; Iverach & Rapee, 2014; Smith, Iverach, O'Brian, Kefalianos, & Reilly, 2014).

The preschool years are a critical period of development. In order to work towards preventing anxiety in stuttering, it is necessary to understand factors that may influence the onset of stuttering and anxiety. One such factor is temperament. Temperament refers to our unique nature, or our way of behaving and responding to the world around us. Temperament can typically be seen soon after birth, and is thought to be genetically determined (Goldsmith et al., 1987; Saudino, 2005). For instance, "some children cry easily

and intensely whereas others are more easy going; some are highly active and always on the go where others are more sedentary; some attend and persist in tasks for long periods of time where others' attention wanders quickly" (Saudino, 2005, p.214). Temperament remains relatively stable over time (Goldsmith et al., 1987; Saudino, 2005), but is also influenced by environmental factors such as parenting and peer experiences.

Understanding the relationship between temperament and anxiety in preschool children who stutter may influence the way that speech-language pathologists manage stuttering. Therefore, the purpose of this presentation is to examine the latest research on temperament, anxiety and stuttering in the preschool years in order to inform clinical practice.

Anxiety and stuttering

Anxiety is part of our survival instinct. When we enter a situation that is threatening, we respond in ways that help us to escape or manage potential danger. For instance, a child who sees that the kitchen stove has caught on fire may run and seek parental protection. This anxiety is helpful when it momentarily protects us from harm or increases our performance. However, anxiety becomes a problem when it is excessive, occurs across a range of situations, causes distress, and interferes with daily functioning. For instance, we might see a child react to a harmless situation, such as attending a party, with a level of anxiety that may be more appropriate in situations containing actual threat or danger, such as a stove on fire.

Anxiety is typically composed of cognitive, behavioural, and physiological components. The cognitive component includes negative thoughts and beliefs, and the expectation of threat. For example, a child may think, "These children won't like me" when entering a social situation. The behavioural component involves strategies to reduce threat or anxiety. For instance, a child may attempt to avoid or even escape feared situations, such as parties or preschool drop-offs. The physiological component of anxiety includes somatic symptoms, such as butterflies in the stomach and increased heart rate. For instance, a child may refuse to attend a party because his/her tummy hurts. Anxiety may occur generally across a range of situations, or may occur more specifically in social situations. When anxiety becomes a problem, an anxiety disorder may be diagnosed.

Research evidence has shown that stuttering is frequently accompanied by anxiety. Of particular note, 22–60% of adults seeking treatment for stuttering may be diagnosed with social anxiety disorder (Blumgart, Tran, & Craig, 2010; Iverach et al., 2009; Menzies et al., 2008; Stein, Baird, & Walker, 1996). Recent evidence has also confirmed that roughly one-quarter of school-age children presenting to speech clinics meet criteria for social anxiety disorder (Iverach et al., 2016).

Social anxiety disorder

Social anxiety disorder is a common anxiety disorder. It involves intense anxiety and fear of negative evaluation in social situations (American Psychiatric Association, 2013). Onset typically occurs between the ages of 8–15 years. It has a lifetime prevalence of roughly 8–13% (Kessler et al., 2005; Ruscio et al., 2008). In adulthood, anxiety typically occurs across a range of situations, such as public speaking, meeting new people, giving presentations at work, and socialising at parties. Social anxiety disorder is frequently associated with distress, low self-esteem, lower education, unemployment, financial dependency, and lower

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socioeconomic status (Stein & Kean, 2000). Therefore, it is important to understand factors that may contribute to the _{development} of social anxiety in stuttering.

Research in the field of psychology has shown us that there are several pathways to the development of social anxiety (Kearney, 2005; Kimbrel, 2008; Ollendick & Hirshfeld-Becker, 2002). Risk factors include genetic predispositions, early cognitive biases, negative peer experiences, relationships with parents, and temperament (Higa-McMillan & Ebesutani, 2011; Hofmann & Barlow, 2002; Kearney, 2005; Kimbrel, 2008; Morris, 2001; Rapee & Spence, 2004; Wong & Rapee, 2016). The combination and timing of these risk factors is likely to influence the development of anxiety (Ollendick & Hirshfeld-Becker, 2002), with the biggest impact occurring in childhood and adolescence (Alfano, Beidel, & Turner, 2006; Morris, 2001; Wong & Rapee, 2016). These risk factors may also interact with other factors such as age, gender, and culture, to influence the development and expression of social anxiety (Rapee & Spence, 2004; Spence & Rapee, 2016).

Research has shown that certain temperamental styles may increase the likelihood that a child will fear or avoid certain situations or people (Fox & Pine, 2012; Higa-McMillan & Ebesutani, 2011; Hofmann & Barlow, 2002; Hudson, Dodd, Lyneham, & Bovopoulous, 2011; Kimbrel, 2008; Morris, 2001; Rapee & Spence, 2004). Behavioural inhibition, in particular, is a temperamental style regarded as one of the most significant and well-established risk factors for social anxiety disorder (Fox & Frenkel, 2013; Fox & Pine, 2012; Fox, Henderson, Marshall, Nichols, & Ghera, 2005; Rapee, Schniering, & Hudson, 2009). Behaviourally inhibited children are often cautious in new situations, and usually withdraw from unfamiliar people, situations, or objects. Research has shown that children who are behaviourally inhibited in early childhood are significantly more likely to be diagnosed with social anxiety disorder in adolescence (Chronis-Tuscano et al., 2009; Schwartz, Snidman, & Kagan, 1999).

Temperament and anxiety in stuttering

Considering that certain dimensions of temperament may be associated with anxiety, and given the high rate of social anxiety disorder among children and adults who present at speech clinics for stuttering treatment, there are a few possibilities. Anxiety may be a precursor to stuttering, a consequence of stuttering, or a combination of both. In order to maximise treatment outcomes for preschool children who stutter, we need to understand the following: (1) do preschool children who stutter show temperamental traits that may predispose them to anxiety in later life; (2) are there any other risk factors for the development of anxiety in stuttering, and (3) how do temperament, anxiety and stuttering interact across the lifespan?

Kefalianos and colleagues (Kefalianos, Onslow, Block, Menzies, & Reilly, 2012) have proposed two possible hypotheses regarding temperament in preschool children who stutter. The first is that particular temperament traits, such as behavioural inhibition or a 'difficult' temperament, may increase risk of stuttering onset. According to Kefalianos and colleagues, this hypothesis is consistent with multifactorial theories of stuttering. These theories propose that emotional reactivity and regulation may interact with communication stressors in the environment and functional brain deficits to trigger stuttering. The second hypothesis is that temperament does not cause stuttering, but rather, anxiety is a consequence of stuttering (Kefalianos et al., 2012). Understanding which hypothesis is true will help to inform the timing and nature of early intervention for stuttering.

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Research findings about the topic

For this presentation, we identified research evidence published in refereed journals regarding the relationship between temperament, anxiety, and stuttering, with particular focus on prospective longitudinal research. The purpose of this review was to determine differences in temperament that may be evident prior to stuttering onset, and to identify findings with implications for clinical practice.

A review

Kefalianos and colleagues (2012) reviewed ten published studies which compared temperament among children who stutter and non-stuttering children (Anderson, Pellowski, Conture, & Kelly, 2003; Arnold, Conture, Key, & Walden, 2011; Eggers, De Nil, van den Bergh, 2010; Embrechts, Ebben, Franke, & van de Poel, 2000; Howell et al., 2004; Johnson, Walden, Conture, & Karrass, 2010; Karrass et al., 2006; Lewis & Goldberg, 1997; Reilly et al., 2009; Schwenk, Conture, & Walden, 2007). All but one study evaluated children after the onset of stuttering. In the majority of these studies, temperament was evaluated using parent questionnaires. One study also used direct observation (Schwenk et al., 2007), and another used electroencephalogram (EEG) recordings to measure emotional reactivity. Across these studies, stuttering children were found to demonstrate lower adaptability, lower persistency/attention span, higher activity levels, and more negative mood, when compared to non-stuttering controls.

However, only one study reviewed by Kefalianos and colleagues (2012) investigated temperament prospectively, that is, before stuttering onset. Reilly and colleagues (2009) reported on a community-ascertained cohort of 1,619 2-year-old Australian children recruited at 8 months of age as part of the Early Language in Victoria Study. A comprehensive range of speech, psychological, and demographic data were collected to determine variables predictive of stuttering onset. In order to evaluate temperament, parents completed the approach/withdrawal items of the Short Temperament Scale. Although approach/withdrawal is an established marker of behavioural inhibition and subsequent anxiety, this dimension of temperament was not found to predict stuttering onset in this study. This indicates that preschool children studied prospectively do not demonstrate temperamental precursors of anxiety prior to stuttering onset.

Given that all other findings reviewed by Kefalianos and colleagues (2012) were obtained with children who had already started to stutter, Kefalianos and colleagues concluded that it was too early to draw a firm conclusion about the accuracy of either of their proposed hypotheses. Two subsequent literature reviews by Alm (2014) and Conture, Kelly, and Walden (2013) also noted the inconclusive nature of findings regarding temperament and stuttering. Alm (2014), in particular, found that the risk for stuttering was not predicted by an emotionally reactive temperament. However, he did find evidence of heightened inattention and hyperactivity/impulsivity, which may be relevant to a subset of preschool children who stutter. Due to the inconclusive nature of findings overall, both Kefalianos and colleagues (2012) and Conture and colleagues (2013) emphasised the need for longitudinal studies of children attained prior to stuttering onset in order to clearly understand the relationship between temperament and anxiety in stuttering.

Temperament and stuttering longitudinally

In 2014, Kefalianos and colleagues (Kefalianos, Onslow, Ukoumunne, Block, & Reilly, 2014) reported on a prospective, community cohort of 183 preschool children who stutter and

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1,261 control children, recruited in infancy as part of the Early Language in Victoria Study (ELVS; Reilly et al., 2006). This study deserves particular attention because it is the only prospective longitudinal study of temperament in early stuttering, providing unique information about factors involved in the onset of stuttering. Children from ELVS were drawn from a broad sociodemographic range of advantaged and disadvantaged regions. Based on parent responses to the Short Temperament Scale, preschool children who stutter did not show innately different temperament styles to control children at ages 2–4 years, and they did not demonstrate temperamental precursors of anxiety before or shortly after stuttering onset.

In a follow-up study of 173 children who stuttered from the ELVS study, Kefalianos and colleagues (Kefalianos, Onslow, Ukoumunne, Block, & Reilly, 2017) found little evidence of a relationship between stuttering severity or stuttering behaviours and temperament in the preschool years. However, 6 year olds with fixed postures of the speech mechanism with no audible airflow (commonly known as 'blocks') did show precursors of anxiety. This suggests that preliminary symptoms of anxiety may occur as a result of experiencing effortful stuttering behaviours. These findings provide the most compelling evidence about temperament and anxiety in early stuttering, and support the hypothesis that anxiety is a consequence of stuttering rather than a cause. Evidence does not support the hypothesis that temperament may be a precursor of stuttering onset.

In light of these findings, it is necessary to understand the development of anxiety following stuttering onset. In 2014, Smith and colleagues reviewed findings from 13 studies evaluating anxiety in children who stutter, including only one study of preschool children who stutter (van der Merwe, Robb, Lewis, & Ormond, 2011). In that study, van der Merwe and colleagues (2011) did not find evidence of differences in anxiety for preschool children who stutter and non-stuttering controls. This was based on salivary cortisol levels and parent responses to the Preschool Anxiety Scale (PAS; Spence, Rapee, McDonald, & Ingram, 2001). Smith and colleagues concluded that the prevalence and timing of anxiety onset in children who stutter could not be determined.

Clinical application of the research findings

Based on research evidence to date, temperament during early childhood is not a precursor of stuttering onset. However, stuttering may be a precursor of anxiety onset in later childhood, adolescence or adulthood. These findings are important for the clinical management of children who stutter, and suggest several directions for future research. For instance, a range of factors influences the development of social anxiety. More research is needed to understand additional risk factors for the development of anxiety in stuttering (Smith et al., 2014). First, do family, environmental, and cognitive factors predict the onset of anxiety in children who stutter? Second, a prospective longitudinal study of children attained prior to stuttering onset is needed to understand whether differences in temperament and anxiety exist between children who subsequently seek treatment for stuttering and those who never seek treatment. This is important because the inconsistent findings regarding temperament and anxiety to date may be partly explained by the treatment-seeking status of children who stutter. Finally, more research is needed to determine whether markers of temperament or anxiety in children who stutter are reduced following stuttering treatment. The research evidence also has implications for all stages of clinical management of stuttering. This includes explaining the disorder to parents, assessing stuttering, addressing anxiety, and planning treatment.

Explaining stuttering to parents

In the first instance, the speech-language pathologist will typically provide parents with information about stuttering. This is important because a parent's beliefs about stuttering can influence their subsequent reactions to stuttering, and their ideas about how stuttering should be treated (Arnold & Li, 2016; Boyle, 2014). The speech-language pathologist can reassure parents that stuttering is not caused by anxiety. However, the negative consequences of stuttering and risk factors for the development of anxiety can also be discussed.

Assessing stuttering

Management of preschool stuttering should be evidence-based and client-centered. It should also be informed by a detailed assessment of the preschool child who stutters. A key part of the assessment process is the case history. During the case history, the speech-language pathologist interviews the parent, and in some instances the preschool child, about stuttering. The case history can include questions about the physical behaviours of stuttering, and also provides the opportunity to evaluate risk factors for social anxiety. Although most preschool children who stutter will not be socially anxious, some children may demonstrate risk factors for social anxiety (Guttormsen, Kefalianos, & Naess, 2015; Langevin et al., 2009), such as negative peer experiences and apprehension about talking. To gauge this during the case history, the speech-language pathologist may ask parents questions such as: "Is your child afraid of talking?", "Does your child avoid or withdraw from social situations?", and "Does your child complain of tummy aches in social situations?".

A useful measure for this purpose is the Preschool Anxiety Scale Revised (PAS-R; Edwards, Rapee, Kennedy, & Spence, 2010). The PAS-R is a parent-report measure designed to evaluate symptoms of anxiety in preschool children. The PAS-R consists of 30 items, seven of which relate to social anxiety and may be relevant to preschool children who stutter. These items include:

> *Item 2: "Worries that s/he will do something to look stupid in front of other people"*
> *Item 5: "Is scared to ask an adult for help (e.g., a preschool or school teacher)"*
> *Item 9: "Is afraid of meeting or talking to unfamiliar people"*
> *Item 12: "Is afraid of talking in front of the class/preschool group (e.g., show and tell)"*
> *Item 15: "Worries that s/he will do something embarrassing in front of other people"*
> *Item 18: "Is afraid to go up to a group of children to join their activities"*
> *Item 25: "Acts shy and quiet around new people"*

The speech-language pathologist should use clinical judgement when determining how to administer the PAS-R. For example, the seven social anxiety questions may be used to guide the clinical interview. However, if there are concerns that a child's anxiety is not just speech-related, the 30-item PAS-R can be administered. The PAS-R does not prescribe cut-offs in terms of whether a child's scores are in the normal or clinical range, but mean scores can be used to determine whether a child's scores are above or below Australian mean scores. Parent responses may also be used more qualitatively to gain a sense of whether social anxiety is causing significant distress or life interference.

Dealing with pre-schoolers who are anxious

Although most preschool children who stutter will not be anxious, what should clinicians do we do when a preschool child who stutters displays risk factors or symptoms of social anxiety? In such cases, referral to a psychologist is the best option. Social anxiety disorder is

the most treatment-resistant anxiety disorder in children, highlighting the importance of early intervention. In the majority of cases, however, referral to a psychologist will not be necessary. For instance, the preschool child may not be anxious, or may only show minimal indicators of risk for anxiety, such as some negative reactions to stuttering, or some negative thoughts or feelings about stuttering. It is important to recognise that these risk factors may potentially lead to the onset of anxiety in later years. Therefore, speech-language pathologists play an important role in monitoring children who stutter for signs of anxiety.

Speech-language pathologists may not feel adequately trained or equipped to manage the social and emotional experiences of preschool children who stutter. This may serve as a barrier to the translation of research about anxiety and temperament into clinical practice. Speech-language pathologists do in fact possess a range of skills that can support preschool children who stutter and their families. Facilitating problem solving around negative peer experiences, advocating for the child at preschool or childcare, and encouraging resilience by focusing on the child's strengths in non-speech related areas of development, are all within the scope of speech pathology practice.

Treatment planning

Stuttering becomes more difficult to treat as children become older. Intervention during the preschool years is optimal and efficacious (Jones et al., 2005). However, treatment is costly and time-consuming, and many preschoolers will recover naturally. The timing of stuttering intervention in preschool children remains a contentious issue. In a recent publication about the epidemiology of stuttering, Reilly and colleagues (2013) concluded that preschool children who start to stutter do not appear to be negatively affected by their stuttering, and that stuttering treatment could be delayed longer than 12 months. However, it is important to acknowledge that the absence of anxiety does not necessarily mean that anxiety is not developing as a result of the negative consequences of stuttering. A recent longitudinal study by McAllister (2016) showed that the emotional functioning of preschool children who stutter may worsen over time to become cause for clinical concern in later childhood. Therefore, treatment should commence sooner if parents are concerned or if the child is distressed, reluctant to communicate, or showing signs of social or emotional difficulties (Reilly et al., 2013). This ensures that the speech and psychological needs of the child are addressed and not left to escalate. Overall, we recommend that speech-language pathologists should not necessarily wait until social or emotional difficulties are present before commencing treatment. The priority and sequence of treatment can be determined collaboratively between the speech-language pathologist and the child's parents, with input from the child's psychologist if applicable.

Parental anxiety

Although the majority of preschool children who stutter will not be anxious, some parents of preschool children who stutter may experience anxiety in relation to their child's stuttering. This anxiety has the potential to result in parental behaviours that model anxiety to the child. For instance, a parent who becomes anxious when their child has to speak in social situations may reduce speaking opportunities for the child, or allow the child to avoid or escape these situations. As a result, the child may learn that social and speaking situations are threatening, and this may contribute to the development of social fears in the child. When a parent is showing signs of anxiety, the speech-language pathologists can provide ongoing opportunities for the parent to discuss their concerns, offer evidence-based

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information about stuttering, and proceed immediately with stuttering treatment. In some cases, referral to a psychologist may be warranted.

Summary and conclusions

Research evidence to date has shown that temperament is not a precursor of stuttering onset. However, stuttering may be associated with negative peer experiences and negative attitudes to communication that may increase risk for later development of anxiety. The preschool years represent a critical period in terms of eliminating stuttering and the subsequent development of anxiety. It is the responsibility of the speech-language pathologist to complete a comprehensive assessment of stuttering, with consideration of the potential social-emotional implications of stuttering. Based on this assessment, the most appropriate clinical management for each child can be determined in terms of the timing of stuttering intervention and level of psychological support required. With optimal clinical management from a speech-language pathologist, and support from parents, preschool children who stutter have the opportunity to meet their potential and lead fulfilling lives.

Questions from discussion leaders

Do you think that resilience training should routinely be given to young children, in case their stuttering, for whatever reason, is resistant to direct treatment?

Stuttering during childhood and adolescence is often associated with bullying, teasing, academic underperformance, and reduced quality of life. If stuttering is not treated or does not respond to treatment, children who stutter are at risk for developing lifelong social, psychological, educational, occupational, and quality of life impairment. Therefore, children who stutter may benefit from resilience training as a supplement to speech treatment. For instance, the Friends Program incorporates cognitive and behavioural strategies to assist children and adolescents in coping with stress and worry (Stallard et al., 2005). Through the program, protective factors such as self-esteem, self-concept, coping skills, hope, and social support are enhanced. This training has the potential to reduce the negative psychological consequences regularly associated with stuttering, thus improving long-term outcomes for children who stutter.

Do you think anxious parents can be reliable and unbiased informants about the level of their child's own anxiety?

In the field of psychology, a multi-informant approach is often used to evaluate psychological symptoms in children (De Los Reyes & Kazdin, 2005). This involves collecting reports from several informants, such as the child, the child's primary caregiver and/or teacher, and mental health professionals. However, discrepancies between child and parent reports of anxiety symptoms in children are common. This suggests that parents might not always be accurate reporters of their child's anxiety, and children themselves may not always report symptoms accurately. This emphasizes the importance of assessing anxiety symptoms from the perspective of multiple informants in order to obtain a more meaningful and comprehensive picture. Having said this, in the absence of reports from other informants, parents may still provide useful information about their child's anxiety, which may prompt further assessment and management if the child is anxious.

Given that parent anxiety could influence child anxiety, should we assess parent anxiety for all pre-schoolers who stutter and treat it if necessary?

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When considering the negative consequences associated with stuttering, it is not surprising that some parents may feel anxious or concerned about their child's stuttering. This concern may prompt parents to seek speech treatment for their child, which may ultimately allay some of their concerns. However, when these concerns lead to excessive anxiety, this may influence the child's own anxiety about their speech. Therefore, it is helpful for speech-language pathologists to be aware of the potential for anxiety to be present for some parents of children who stutter. In cases where a parent shows signs of anxiety, further assessment or referral may be required. A useful measure for this purpose is the Depression Anxiety Stress Scale, which evaluates the three related negative emotional states of depression, anxiety and tension/stress in adults (www2.psy.unsw.edu.au/dass/).

Do you think all these mental health problems will be obviated if stuttering is controlled effectively during the pre-school years?

A growing body of evidence has confirmed the prevalence of social anxiety disorder among adults seeking treatment for stuttering. Most recently, evidence has emerged that school-age children who stutter may demonstrate a significantly higher rate of social anxiety disorder than non-stuttering control children. This suggests that anxiety may develop earlier than previously thought. The preschool years present a critical period of development, with effective speech treatment potentially protecting children who stutter from developing the chronic psychological and social problems reported by adults who stutter. However, it is not yet known whether controlling stuttering in the preschool years will obviate mental health problems later in life. In order to work towards preventing anxiety in stuttering, additional research is needed to determine risk and protective factors associated with anxiety in children who stutter.

Is it possible that direct treatment such as the Lidcombe Program might increase a child's anxiety about stuttering?

There is no evidence of any adverse psychological effects before, during, or after Lidcombe Program treatment (Woods, Shearsby, Onslow, & Burnham, 2002). There is, however, much evidence for negative social and emotional consequences of stuttering beginning during the preschool years if stuttering persists after onset. Naturally, as with all childhood speech pathology treatments, the clinician is proactive in preventing adverse reactions from children and will respond appropriately in the unlikely event they occur.

We would like to follow up your response to the last question, where you say "There is no evidence of any adverse psychological effects before, during, or after Lidcombe Program treatment (Woods, Shearsby, Onslow, & Burnham, 2002)." However, that surely is an incautious statement, since the Woods et al. involved only eight children, specifically selected because they were treatment successes, and assessed with only two psychological measures. What is your response to that?

That is a valuable point. On those grounds, we would like to revise our original response as follows:

One small study investigated the psychological impact of the Lidcombe Program with eight children who successfully completed treatment (Woods, Shearsby, Onslow, & Burnham, 2002). That study reported no evidence of any adverse psychological effects before, during, or after treatment (Woods, Shearsby, Onslow, & Burnham, 2002). More research is needed to explore this issue.

There is, however, much evidence for negative social and emotional consequences of stuttering beginning during the preschool years if stuttering persists after onset. These negative consequences are likely to intensify with age (Guttormsen, Kefalianos, & Næss, 2015). Naturally, as with all childhood speech pathology treatments, the clinician is proactive in preventing adverse reactions from children and will respond appropriately in the unlikely event they occur.

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The Lidcombe Program for Early Stuttering: Are We Really Sure?

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Abstract

The importance of early intervention in management of stuttering is widely acknowledged. It is vital therefore that the most effective form of treatment is offered during the limited time frame when early stuttering is most tractable. The Lidcombe Program has empirical evidence from Phase I to Phase III clinical trials, providing the most comprehensive evidence base of treatments for early stuttering. There is evidence of the efficacy and safety of the treatment, and effectiveness studies indicate that community clinicians with appropriate training are able to match outcomes of clinical trials across a broad range of settings. Research data provide benchmarks such as predicted treatment sessions to Stage 1 and expected change in stuttering severity early in treatment. A randomised trial conducted in the Netherlands indicates insufficient evidence of a clinically significant difference between outcomes for the Lidcombe Program and an indirect Demands and Capacities model of treatment. Whilst quantitative data are useful, they do not illustrate the complexity of the Lidcombe Program. Qualitative information about parent experiences of the program raises considerations for treatment and shows that progress is not always straightforward. Successful outcomes for the Lidcombe Program are dependent on a balance between treatment fidelity and its individualisation.

Clinical importance of the topic

The importance of early intervention

The importance of early intervention in the management of stuttering is widely acknowledged (Gottwald & Starkweather, 1999; Yaruss, Coleman & Hammer, 2006). Clinicians assess the risk for persistent stuttering and decide if treatment is indicated. If treatment is required, the most effective form of intervention needs to be offered during the limited time frame when early stuttering is most tractable. Clinicians aim to integrate individual client variables and clinical expertise with the best available external evidence based on systematic, scientific research. It is important therefore to understand the research upon which the Lidcombe Program is based and how it informs clinical decision making.

A conceptually simple treatment

The Lidcombe Program targets children's speaking and so is different from multifactorial approaches where stuttering is considered a complex disorder influenced by child, family and environmental factors, and where no stuttering is not the main treatment goal (Rustin, Botterill, & Kelman, 1996). In contrast, the Lidcombe Program is conceptually simple, requiring parents to present a maximum of five different verbal contingencies to their children and to measure their children's stuttering with a severity rating scale each day. Children are not required to change their speech pattern nor do parents modify the child's communication environment.

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Not always straightforward

When children progress through the treatment in the expected way, it is straightforward. However, there are child and parent variables to consider during the process of Lidcombe Program treatment that have not been accounted for in quantitative research studies. These include linguistic and cultural background, parent perception of stuttering, parents' perception of themselves as the therapist, parenting styles, the nature of the parent-child relationship, and parent education and literacy. Random group assignment in Lidcombe Program randomised clinical trials aims to ensure that groups are equivalent across all clinically pertinent variables. However, random assignment cannot entirely resolve the problem, it only minimises it to some extent (Krause & Howard, 2003).

Research findings about the Topic

Overview of research findings

The Lidcombe Program has research findings from case note audits, experimental studies, and randomised clinical trials. There is clinical trial evidence for the efficacy of the treatment with a standard treatment format (Harris, Onslow, Packman, Harrison & Menzies, 2002; Jones et al., 2005; Lattermann, Euler & Neumann, 2008; Lewis, Packman, Onslow, Simpson, & Jones, 2008), and studies to show the effectiveness of the Lidcombe Program within a community setting (O'Brian et al., 2013), in a group treatment format (Arnott et al., 2014) and in a telepractice format (Bridgman, Onslow, O'Brian, Jones & Block, 2016). Meta-analysis of randomised controlled evidence for 134 children (Onslow Jones, Menzies, O'Brian, & Packman, 2012) showed that the pre-randomisation percentage syllables stuttered (%SS) for children in the Lidcombe Program group and the no treatment control group were about the same. Children in the control group showed some improvement due to natural recovery. However, the Lidcombe Program children did better, showing 7.5 times greater odds of having minimal stuttering than the children who did not receive the Lidcombe Program.

Stuttering is a relapse prone disorder and so it is important to investigate whether treatment gains are maintained in the longer-term. Jones et al. (2008) followed up the children in the Jones et al. (2005) trial for 3–7 years post-treatment, at which time three of the 29 children treated had relapsed appreciably. A Swedish case study report of the Lidcombe Program (Femrell, Avall & Lindstrom, 2012) showed that the six children who completed treatment maintained fluency 21 months after completing Stage One. A data based study in the United States (Guitar et al., 2015) followed up children 1 to 5 years after treatment. This showed that the mean pre-treatment score of 12.6 %SS reduced by 96% to a mean post-treatment score of 0.5 %SS.

Concern that a direct behavioural approach could be harmful for children's psychological development (Cook & Rustin, 1997) was allayed by a study (Woods, Shearsby, Onslow, & Burnham, 2002) of child psychological measures and child-mother attachment post-treatment. Additionally, no evidence has been found that the Lidcombe Program suppresses child language development, nor leads to changes in child or parent speech rate or pragmatics (Bonelli, Dixon, Bernstein Ratner, & Onslow, 2000). Similarly, Lidcombe Program treatment does not seem to be associated with a decrease in the length of children's utterances or sentence complexity (Lattermann, Shenker, & Thordardottir, 2005).

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Treatment dosage

Zeng, Law and Lindsay (2012) highlight the insufficient attention paid to "dosage" within the field of speech and language therapy research and recommend exploration of dosage variables. A strength of the Lidcombe Program evidence base is the inclusion of data relating to number and frequency of treatment sessions. The Lidcombe Program treatment guide (Packman et al., 2015) prescribes weekly visits of 45–60 minutes duration in Stage 1 and, when necessary, that parents administer daily 10–15 minute verbal contingency practice sessions each day in addition to verbal contingencies during everyday, natural conversations. Around a 30% reduction of severity ratings are expected within the first four weeks of clinic visits (Onslow, Harrison, Jones & Packman, 2002). File audit data show the median number of clinic visits required to reach Stage 2 is 16, with a range 11–23, and that children with more severe stuttering require more clinic visits than children with milder stuttering (Kingston, Huber, Onslow, Jones & Packman, 2003; Koushik, Hewat, Shenker, Jones & Onslow, 2011). These data are useful in treatment, caseload management, and allocating clinical resources. Evidence specifying the required dose of parent verbal contingencies for stutter-free speech and for unambiguous stuttering during natural conversations would assist clinicians. When children are referred to clinics shortly after stuttering onset, caseload management is facilitated by the knowledge that initial "watchful waiting," to detect signs of natural recovery, will not negatively impact treatment outcomes (Jones, Onslow, Harrison & Packman, 2000; Kingston et al., 2003). Benchmarking data enable clinicians to predict likely treatment time and to identify slower than expected progress, however, we also need to know possible reasons for this.

Language factors

Many children living in socio-economically disadvantaged communities present with delayed language development (Locke, Ginsborg & Peers, 2002). A study by Rousseau, Packman, Onslow, Harrison and Jones (2007) aimed to determine whether pre-treatment phonological and language development predict Lidcombe Program treatment time. The findings showed no relationship between children's phonological development and time taken for Stage 1. The children with a higher mean length of utterance progressed through Stage 1 more quickly than those with shorter mean length of utterance. Unexpectedly, children with higher receptive language scores took longer in treatment, suggesting other unmeasured variables exert an influence. Interestingly, children with more severe pre-treatment stuttering had poorer comprehension and general language function. Rousseau et al. indicated that further investigation is required before confident conclusions can be made from such research.

The necessary exclusion criteria applied in many clinical trials relating to family linguistic background means that their findings may not include the population of bilingual children who stutter. Given that much of the world's population is bilingual, this calls into question the generalisation of Lidcombe Program research evidence to many clinical settings. Research specifically examining the effectiveness of the treatment with bilinguals would explore a different aspect of language competence and provide vital evidence for working with multi-lingual populations. Shenker and Roberts (2006) studied the long-term outcome of the Lidcombe Program for 14 bilingual children, 2–7 years post-treatment. Results were found to be similar to studies for unilingual children (Lincoln and Onslow, 1997; Miller and Guitar, 2009). Eleven of 14 children had maintained a level below 1.0 %SS and the other three children had very low levels of stuttering. Another useful finding was that for matched non-stuttering peers, levels of "stuttering-like disfluencies" present in English/French bilinguals were similar to the 8% found in another study (Roberts & Hérbert, 2001). These

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disfluencies have the potential to confuse parents who are learning to distinguish between stutter-free speech and unambiguous stuttering and may impact on time required for parent training.

Vong, Wilson and Lincoln (2016) investigated Lidcombe Program outcomes with three preschool children in Malaysia. Two bilingual participants achieved near-zero levels of stuttering 12 months post-treatment in both languages, though treatment targeted only one. Vong et al., suggested that the longer treatment duration required for these children resulted from a mismatch between the Lidcombe Program and the culture of Malaysia. Praise and acknowledgement were problematic because such feedback is used infrequently in Malaysian culture. The caregivers required 5–8 treatment sessions before reliably demonstrating correct delivery of these contingencies for stutter-free speech.

The Lidcombe Program compared to RESTART-DCM

The relative value of direct and indirect treatments for stuttering in children and adults has been debated but rarely compared in research. A Netherlands study (de Sonneville-Koedoot, Stolk, Rietveld & Franken, 2015) aimed to compare the effectiveness of direct and indirect treatment for early stuttering. This was a randomised controlled trial with 99 children in the Lidcombe Program arm and 100 in the RESTART-DCM arm. The RESTART-DCM treatment was based on the Demands and Capacities Model, which aims to decrease environmental demands on speech and increase the child's capacities for fluent speaking (Starkweather & Gottwald, 1990). The primary outcome measure was the frequency measure of %SS at 18 months after the start of treatment. Children with less than 1.5 %SS were regarded as "non-stuttering." The paper reported that the Lidcombe Program reduced stuttering more quickly during the first three months of treatment but there was no statistically significant difference shown between the groups at 18 months.

The results of this large and interesting trial are somewhat difficult to interpret. First, children were classified as "non-stuttering" post-treatment at 1.5 %SS so some were obviously stuttering. Second, the trial did not include an untreated control group and some of the reduction of stuttering is likely due to natural recovery across both groups. In the absence of a non-treatment control, the effects of either treatment over and above natural recovery were not demonstrated.

What is the active treatment agent?

There is a debate in treatment research concerning whether or not we need to know which aspects of a treatment are active components and, if these can be identified, whether or not we need to understand how they work in order to implement them effectively with less straightforward clients (Campbell et al., 2000; Hayhow, 2011).

Verbal response contingent stimulation is considered to be the active treatment agent of the Lidcombe Program, although there may be other mechanisms operating in treatment that assist recovery (O'Brian & Onslow, 2011). A study by Donaghy et al. (2015) investigated the role of the verbal contingency "request self-correction of stuttering," which is one of the two contingencies in the treatment for unambiguous stuttering moments. This experiment included 38 children who had been stuttering for at least 6 months and presented with more than 3.0 %SS. The control group comprised parent-child dyads receiving the standard Lidcombe Program as outlined in the treatment guide (Packman et al., 2015). The experimental group received the standard Lidcombe Program treatment without the instruction to use the verbal contingency "request self-correction of stuttering." The Primary

outcome measure of number of clinic visits to 50% reduction of stuttering severity showed little difference between the two groups. This result present some issues. Might that particular verbal contingency become more powerful after the initial decline in %SS, or is it redundant to Lidcombe Program treatment effects? If it is redundant to treatment effects, is the same true of the other verbal contingencies of the treatment? If so, what are the treatment agents responsible for observed treatment effects? For example, are home practice sessions, where conversations are structured to achieve low levels of stuttering, an important treatment agent?

An observation study of three children by Carr Swift et al. (2011) documented parent use of verbal contingencies. Results showed that parents, as instructed, presented more contingencies for stutter-free speech than stuttered speech during practice sessions. However, one parent did not maintain the recommended ratio when delivering treatment during natural conversations and, contrary to clinician instruction, introduced verbal contingencies during natural conversations in addition to practice sessions from the start of treatment. Additional concerning observations were a parent who continued with a practice session even though the child disliked the activity, and another parent who focussed on the rules of the game rather than the child's speech. All parents were observed to give incorrect verbal contingencies in practice sessions, such as offering praise for stutter free speech in response to stuttering.

Adverse parent experiences

Given the evidence that parents may not implement treatment as we expect, qualitative research data about adverse parent experiences of the Lidcombe Program enable clinicians to be forewarned of potential difficulties. Research on mothers' experiences of the Lidcombe Program (Goodhue, Onslow, Quine, O'Brian, & Hearne, 2010) showed that some felt anxious and under pressure to perform well with treatment. Some reported feeling empowered as well burdened by the responsibility of implementing home treatment. Some mothers experienced a cycle of confidence where their confidence in carrying out the treatment was related to the child's progress. In a similar study, Hayhow (2009) reported that the majority of cases found treatment straightforward, three started well but then progress stalled while two mothers found treatment problematic from the outset.

Clinical application of the research findings

Treatment fidelity

Positive outcomes for the Lidcombe Program require a balance between adherence to essential treatment procedures and individualisation for each child and family. For research findings to be useful in clinical practice, they need to extend our understanding of factors that potentially limit treatment integrity and those that influence its individualisation.

It is important to consider whether stuttering treatment is being implemented as intended (Thomas & Howell, 2001) since lack of fidelity to the treatment procedures upon which research findings are based could reduce treatment effects. For the Lidcombe Program, treatment fidelity means that clinicians need to understand the active components and to train parents to apply these correctly. It is possible that when adapting treatment, clinicians move away from the procedures outlined in the treatment guide and inadvertently omit active treatment components. Evidence that clinicians who receive training from the Lidcombe Program Trainers Consortium have more favourable clinical outcomes than those

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who do not (O'Brian et al., 2013), suggests that economising on training could lead to unacceptable costs for clients and continued costs for services.

Therapist drift

Psychologist Glenn Waller (2009) raises issues about evidence-based treatment and therapist drift, which he describes as a common occurrence. He suggests clinicians can drift away from implementing the full range of tasks required for effective behavioural change and become the unintentional agent of treatment failure. Clinician belief about what should be useful during treatment can affect judgement, prompting believe that therapy is working even if there is evidence to the contrary. It could be speculated that the strong evidence base for the Lidcombe Program might lead clinicians to persist with treatment in the belief that it should be effective, even if clinical evidence from an individual client suggests the contrary. This could discourage clinicians from the proper clinical response if a continuous reduction of stuttering severity does not occur during the early stages of treatment; issues affecting progress need to be identified and addressed.

Parents do not always implement treatment in the way they have been trained, raising safety concerns. However, research data have not revealed the frequency of errors in the use of parent verbal contingencies or whether in fact errors decreased over time as parents became more skilled. Clinicians and researchers do need to be cautious when making assumptions about how treatment procedures are actually implemented in the home and find a means of verifying that their instructions are being followed. Ordinarily, parents demonstrate home therapy in clinic and are guided and encouraged by the therapist to develop their skills. Implementation at home may be more challenging than in the supportive environment of the clinic. There are reported difficulties for some parents in leading sessions, in managing siblings and in developing the required problem-solving skills for adapting treatment.

It is a concern that some clinicians studied were not checking, within clinic, parents' ability to use contingencies correctly and safely (O'Brian et al. 2013). This lack of scrutiny makes it impossible to know whether parents can independently structure practice sessions and use verbal contingencies appropriately. Research has indicated that some parents feel anxious about conducting therapy and so it is possible that clinicians omit aspects of treatment to minimise the experience of negative emotion for the parent or themselves. In these instances, more clinician feedback and support may be required for parents to enable them to engage fully in sessions and gain confidence in demonstrating treatment in clinic.

Caseload issues with therapist drift

There is evidence that therapist drift may be context driven where externally imposed targets such as caseload size may lead clinicians to reduce session duration or frequency (O'Brian et al. 2013; Rousseau, Packman, Onslow, Robinson, & Harrison, 2002). Interestingly, there was no evidence that this affected outcomes. To date, research has not demonstrated at what point a reduction in session duration or frequency impacts on treatment effectiveness. However, clinicians need to consider, before starting treatment, whether they have capacity to offer the regularity and duration of sessions that the Lidcombe Program requires. This is particularly important where there are linguistic and cultural variables that can impact on treatment time. It is useful to be forewarned about potential slower response to treatment when working with language delayed children.

Research alerts us to the possibility of mismatch between treatment procedures and cultural practices when implementing the Lidcombe Program. We need to be prepared to spend time discussing these differences with parents, negotiating how treatment proceeds, and allowing for additional training as required. Although findings indicate that bilingual children achieve similar outcomes to monolingual children, research has not provided evidence specifically about outcomes where treatment has to be conducted with an interpreter. It is likely that treatment delivered in this way will require longer sessions and more visits in Stage 1; also the potential for loss of treatment fidelity is greater. In these circumstances, matching research benchmarks for treatment duration is unrealistic, hence it is important for clinicians to gather data to establish local norms in order to monitor clinical effectiveness and anticipate resource requirements.

Is therapist drift always undesirable?

A positive aspect of therapist drift from treatment procedures is that it can lead to future developments and improvement. Any departure from the Lidcombe Program treatment guide (Packman et al., 2015) must be intentional, clinically reasoned, and its outcome documented so that it can add to practice based evidence. An interesting issue relating to Lidcombe Program treatment is that of structure. Treatment is implemented in practice sessions and natural conversations. No attempt should be made to modify conversation structure within natural conversations. However, some parents report that increasing structure in natural conversations, at times of increased stuttering, helps their child to attain stutter-free speech. This is therefore an example of a departure from treatment protocol with apparently positive outcomes for some children.

Research into mothers' experiences of the Lidcombe Program showed that although many children enjoyed treatment, some reacted negatively to contingencies for stutter-free speech. Hayhow (2011) refers to cognitive evaluation theory (Ryan & Deci, 2000) as a possible explanation for these contingencies not working well. The theory suggests that praise may have an intrinsic motivation for the child, or extrinsic motivation, which is associated with trying to please others. Praise is more likely to have positive effects on progress if it holds intrinsic meaning for the child and supports development of autonomy. Praise for stutter-free speech might encourage children to feel they are gaining control over their speaking. However, if over-used, children can perceive praise to be controlling. The meaning attached to praise changes as children develop with age, and eventually may serve to remind them of their speech difficulty and so cease to be rewarding in the way it is for a younger child. Treatment can be individualised by using verbal contingencies that are more acceptable to the child and parent provided that they remain specific and speech related.

Summary and conclusions

In conclusion, clinicians have a professional duty to ensure practices are evidence based and that the treatments offered are effective for communities served. So, how confident can clinicians be that the Lidcombe Program meets these requirements?

We can be certain that research about the treatment has been rigorous, using a systematic framework of clinical evaluation. The evidence base is strong, showing the Lidcombe Program to have a clinically important treatment effect. Although the complexity of its implementation in some cases should not be underestimated, children from a diverse range of backgrounds benefit from this approach. A real strength of the treatment is that it has been devised by the partnership of researchers and clinicians working together to produce

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research evidence that applies directly to clinical practice. Clinicians need to ensure they are aware of research findings as they emerge and respond by updating their practices accordingly. Provided treatment is implemented correctly, there is little doubt that the Lidcombe Program is a useful choice for many preschool children who stutter.

That being said, discussions with follow-up days after Lidcombe Program Trainers Consortium workshops reveal that some clinicians find that the Lidcombe Program is not equally effective for the entire range of families on their caseloads. They report that they cannot always match the treatment benchmarks provided in research data. Typically, these are clinicians working within inner-city settings where caseloads are linguistically, culturally and socio-economically diverse. The issues faced by these clinicians about the implementation of the Lidcombe Program are not fully answered by the available empirical evidence.

Questions from discussion leaders

Do you think it really matters if clinicians drift from the treatment guide? For example, if they choose to never use one or two of the specified verbal contingencies?

We have some evidence to indicate that drift from the treatment guide such as a reduction in clinic visit frequency or duration may not be detrimental to outcomes. In addition, it has been shown that leaving out the verbal contingency "request self-correction of stuttering" may not affect progress. However, we need to be cautious about moving away from the treatment guide when we do not know enough about the role of individual verbal contingencies in treatment and the consequences of omitting them. It would be particularly concerning if the exclusion of some contingencies risks leading parents towards using a ratio of more contingencies for stuttering than for stutter free speech when that is not in the best interests of the child's treatment.

What is your view about the management sequence for the Lidcombe Program with preschoolers with one or more speech and language disorders comorbid with stuttering?

In my view, there is no one approach appropriate for all children. For cases where the speech or language disorders are relatively mild, I would usually want to work on fluency first to ensure treatment can occur during the time period in which it will be most effective. Some improvement in children's speech and language skills often occurs alongside Stage 1 of Lidcombe Program treatment.

When children present with severely impaired language or speech skills then stuttering management needs to be planned on an individual basis. This involves taking into account which disorder is having the greatest impact on the child's communication abilities and which is most likely to respond initially to treatment.

(*Editors' note:* Subsequent to the conference, Unicomb, Hewat, Spencer, and Harrison, [2017] published a report of successful concurrent Lidcombe Program treatment and treatment for speech sound disorder.)   .

Given the obvious cultural issues associated with the Lidcombe Program, how do you promote it to clinicians from other cultures?

There are a number of reasons why Lidcombe Program could be a good treatment choice for clinicians from a range of cultures. Research has demonstrated that it translates to

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bilingual populations of children and different community settings and so we know that satisfactory outcomes can be achieved for children from different backgrounds. Practice sessions can be based around daily activities and so be made culturally appropriate. Mismatches between the requirements of the Lidcombe Program and the typical ways in which parents interact or play with their child can pose a challenge in therapy. Provided the clinician is able to understand the parents' perspective and has awareness of cultural practices, then these can be incorporated as part of the individualisation of treatment.

On balance, considering your reservations about the treatment, what is your overall assessment of the value of the Lidcombe Program for treating preschool children who stutter?

I regard the Lidcombe Program as an effective treatment for preschool children that works well in most cases. The approach has great value as a direct, behavioural treatment and has been shown to lead to long-term improvements in children's fluency. I do not have reservations about the treatment per se but rather some concerns about the initial decision making process when there is a complex range of factors that could impact on parent training. Before starting treatment, we need to be sure that parents will be able to implement the Lidcombe Program accurately and safely and that we have the skills and resources to support them.

What do you think the RESTART clinical trial conveys to evidence based clinicians who often use the Lidcombe Program to treat early stuttering?

As evidence based clinicians we need to be prepared to change our practices in response to new research findings. The results of the RESTART trial suggest that similar outcomes could be achieved for the Lidcombe Program and RESTART-DCM treatment. However, the absence of a no-treatment control group in that trial means that the findings are difficult to interpret and so do not really give us a clear message about how these treatments compare. I hope that further trials will be conducted in the future so that we are able to understand more fully the relative merits of direct and indirect approaches.

To follow up your response to the last question, is there any hypothetical case of a pre-schooler who stutters for whom you would consider RESTART-DCM treatment? If so, can you outline such a case?

I consider it important to be skilled in a range of therapy approaches for early stuttering, because one treatment is never going to be the right choice for every case. So, whilst the Lidcombe Program is the treatment I most frequently use, there are certain children and families for whom this is not the most appropriate option. For example, I may select an indirect treatment such as RESTART-DCM when it appears that parental management of stuttering is placing an unhelpful level of demand on the child's speech. This might include comments about speech, even criticism for stuttering or unrealistic expectations for language production. I would be cautious about using a direct treatment in this situation as it may result in further pressure being placed on the child's speaking. As a starting point, I would want to support parents to work through their concerns and anxieties, with a view to modifying their approach and reducing demands on the child.

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Multifactorial Treatments with Young Children who Stutter

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Abstract

The focus of this presentation is to explain the rationale for multifactorial approaches and the current status of research into interventions, with a particular emphasis on Palin Parent-Child Interaction Therapy (Kelman & Nicholas, 2008). In line with the structure of the conference, this presentation seeks to report on the clinical importance of the topic, the research findings about the topic and clinical applications of the topic. Multifactorial models are a method of organising and integrating the literature in order to understand the complexity and variability of stuttering in young children. The models inform the assessment and therapy process and emphasise the importance of taking into consideration the physiological, linguistic, emotional and environmental variables which may influence stuttering and the impact that it has on the child and the family. A combination of indirect and direct strategies is included, although typically the indirect strategies are implemented in the first instance. While the principles of intervention are the same between individuals, the importance of modifying the content of therapy is emphasised across all these approaches. Palin Parent-Child Interaction Therapy is one such program. A summary of the program is described in depth, along with the evidence base to support it.

Clinical importance of the topic

The need for intervention with preschool children who stutter is clear. Not all young children will continue to stutter into adulthood (Yairi & Ambrose, 2013) and not all will suffer significant consequences in the short or long term. But for some, stuttering is associated with negative stereotyping, peer rejection, reduced academic achievement, behavioural, emotional, and social difficulties (Davis, Howell, & Cooke, 2002; Iverach et al., 2016; McAllister, 2016; St. Louis & Tellis, 2015). Even if the disorder does not persist, the experience at the time can result in increased anxiety, upset, frustration and worry for the child and the parents (Plexico & Burrus, 2012; Vanryckeghem, Brutten, & Hernandez, 2005). The aim of this paper is to outline the relevance and contribution of a multifactorial approach to assessment and intervention with young children who stutter. While a range of approaches will be referred to, Palin Parent-Child Interaction Therapy (Kelman & Nicholas, 2008) will be used as the main example.

Multifactorial Models

When describing the Demands and Capacities model, Starkweather (1997) was explicit that the model says little about the cause of stuttering. As with most other multifactorial models, the aim is to provide a framework to organise the literature and to inform the assessment process and content of therapy. Using one of these models to understand the factors that may be significant in relation to an individual child's stuttering, the clinician can identify the child's strengths and needs, as well as the priorities for intervention. The therapy program may then be developed in order to devise an individualised therapy program. Multifactorial models help clinicians and clients to understand the complexity, heterogeneity and variability of the disorder, as well as the individual's responses and reactions to stuttering and to therapy.

It is generally accepted that stuttering is a neurophysiological problem, a motor speech disorder explained by structural and functional differences in the brain (Chang, 2014). A child is born with a predisposition to stutter, but according to what we know from the field of epigenetics and from genetic research about stuttering, whether a child stutters or not is not entirely explained by whether it was inherited from a parent. We know, for instance, that stuttering is more common in families where it already exists and that when one twin stutters, the chance that the second twin will stutter is higher in monozygotic twins compared to dizygotic twins (Kraft & Yairi, 2012). However, monozygotic concordance is not 100%, leading to the conclusion that it is the addition of extrinsic and intrinsic variables that determine the extent to which the genetic predisposition finds expression (Starkweather, 2002). It is the clinician's role to explore the factors that are influential, both in terms of those that are unhelpful, but also those that are helpful. It is not presence of any one factor or combination of factors that is so important as the interaction between them (Smith & Kelly, 1997).

There are a number of multifactorial models that have been proposed. Examples are: (1) The Component model (Riley & Riley, 1979), which organises variables into physical, temperamental, and social attributes; (2) The Demands and Capacities model (Starkweather & Gottwald, 1990) and (3) the Three Factor model (Wall & Myers, 1995), which consider psycholinguistic, psychosocial, and physiological factors, (4) the Dynamic Multifactorial model (Smith & Kelly, 1997) and the CALMS model (Healey, Trautman, & Susca, 2004), which represent cognitive, affective, linguistic, motor, and social factors; and (5) the Palin Multifactorial model (Rustin, Botterill, & Kelman, 1996), which considers the relevance of physiological, linguistic, environmental, and psychological factors.

Across the models, the individual factors are grouped together in a similar fashion. In all, stuttering is viewed as a dynamic disorder with the factors and the relationship between the factors changing over time. The authors of the Demands and Capacities model discuss these factors in terms of whether they are intrinsic or extrinsic to the child. The factors are discussed in the context of the child's capacity for fluent speech and the relationship, and potential mismatch, between this capacity and the intrinsic and extrinsic demands for fluent speech. The demands or capacities may be within normal limits, but it is the mismatch between the demands and capacities that is critical.

The Palin Multifactorial model

The Palin Multifactorial model (Rustin, Botteril, & Kelman, 1996) is used to understand the complexity of the disorder and inform assessment and therapy at The Michael Palin Centre in London. There are four broad categories of factors that interact with each other: physiological, linguistic, environmental, and psychological.

Physiological factors. There is now a substantial body of evidence that genetics have a role in stuttering onset (Kraft & Yairi, 2011) and that stuttering is a neurodevelopmental disorder which might be explained by structural and functional differences in the brain (Chang, 2014). There is evidence emerging that there may be further differences between those children whose stuttering resolves and those stuttering persists (Chang, Zhu, Choo, & Angstadt, 2015). The consequence of these structural and functional differences manifest themselves as less stable motor patterns of speech production (Ambrose, Yairi, Loucks, Seery, & Throneburg, 2015; Smith, Goffman, Sasisekaran, & Weber-Fox, 2012). In terms of whether the stuttering will resolve or persist, there is evidence that period of time since onset and/or the pattern of dysfluency over time may be predictive of chronicity, and girls are more likely

to resolve the disorder naturally than boys (Yairi & Ambrose, 2005). Anecdotally, parents at the Michael Palin Centre report other physiological factors that have an impact on the child's fluency on a daily basis, such as tiredness or sickness. Attempts at a faster rate of speech are also often reported to have a negative impact on fluency.

Linguistic factors. There have been a number of studies that have explored the linguistic abilities of children who stutter compared with non-stuttering controls. The results are equivocal, with studies finding reduced skills for children who stutter (Hakim & Ratner, 2004), mismatches in linguistic skills (Coulter, Anderson, & Conture, 2009), and evidence that children who stutter may also have advanced language skills (Reilly et al., 2013; Watkins, Yairi, & Ambrose, 1999). In a review of the literature, Ntourou, Conture, and Lipsey (2011) concluded that the evidence across studies indicates that children who stutter have language skills that are subtly below controls. While often still within normal limits, these can be significant in relation to stuttering, particularly in combination with environmental factors. There is little evidence regarding the relationship between language and chronicity, but there have been many studies that have explored the relationship between language and the moment of stuttering. With preschool children, stuttering is more likely to occur at the start of utterances (Richels, Buhr, Conture, & Ntourou, 2010) and clauses (Bernstein Ratner, 1997), on function words (Howell, Au-Yeung, & Sackin, 1999; Richels, Buhr, Conture, & Ntourou, 2010), and on stressed syllables (Natke, Sandrieser, van Ark, Pietrowsky, & Kalveram, 2004). Stuttering also is more frequent as length and complexity of utterance increases (Gaines, Runyan, & Meyers, 1991; Ratner & Sih, 1987).

As with language abilities, the research into the phonological skills of children who stutter has resulted in inconsistent or non-replicated findings. Certainly, there are children who stutter who have comorbid delayed or disordered phonology, although the extent of the co-existence is unclear. Reports of up to 40% comorbidity (Yaruss, LaSalle, & Conture, 1998) have been argued to be an over-estimation (Nippold, 2000) and Ambrose et al. (2015) concluded that it is only the acquisition of late phonemes that lags in children who stutter and is associated with chronicity.

Psychological factors. One of the intrinsic variables that has received much attention in recent years is temperament. While some studies have found no temperamental differences between young children who stutter and their fluent peers (Reilly et al., 2009), other studies suggest that there are a significant group of children who stutter who are more reactive and less able to regulate their emotions (Karrass et al., 2006) and less able to adapt to new situations (Anderson, Pellowski, Conture, & Kelly, 2003). We know that emotional arousal can have an impact on stuttering, with emotions such as excitement, frustration, and anxiety reported by many to be less helpful emotional states. People who are sensitive will reach a threshold to arousal and reactivity much sooner than those with typical sensitivity thresholds.

In addition to the potential for intrinsic temperament and emotional state to influence stuttering behaviour, we are also aware that the stuttering affects the emotional state in the moment and over time. There is evidence that some children will experience anxiety about their speech even from an early age and that this tends to increase over time (Vanryckeghem & Brutten, 2007; Vanryckeghem, Brutten, & Hernandez, 2005). How we feel affects what we do and children's emotional reactions and behavioural responses to stuttering, such as loss of eye contact or word and situation avoidance, can have consequences for their ability to communicate and participate (Langevin, Packman, & Onslow, 2009).

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But it is not just the child who is affected by the stuttering. Parents feel anxious and upset, they are worried about their child's reaction to the stuttering, and fear that their children will suffer socially and will not reach their potential (Plexico & Burrus, 2012). This can have an impact on their confidence as parents, their interaction style and their management of their children in general (Biggart, Cook, & Fry, 2006; Kloth, Kraaimaat, Janssen, & Brutten, 2000; Plexico & Burrus, 2012).

Environmental factors. According to Kraft et al. (2014), environmental factors refer to the combination of biology, living conditions, culture, and experiences shared with parents and other members of the same household. There is no suggestion that a child's environment causes stuttering, or that children who stutter are exposed to more a demanding environment that explains the onset of the stuttering (Kloth, Janssen, Kraaimaat, & Brutten, 1995). However, there is some evidence that when children begin to stutter, adults—not just the child's own parents—change their interaction style in response to the stuttering (Kloth et al., 2000; Meyers & Freeman, 1985a, 1985b). These changes are not necessarily outside the normal range, but these or other environmental factors may interact with physiological, psychological, and linguistic factors, so that the child who stutters may find it more difficult to be fluent in what are typical or normal interactions (Miles & Ratner, 2001). There is evidence that parents can make changes to their own behaviours which can facilitate fluency, such as reducing their rate of speech (Guitar, Schaefer, Donahue-Kilburg, & Bond, 1992; Stephenson-Opsal & Ratner, 1988), increasing response time latency (Newman & Smit, 1989), and developing structured turn taking (Winslow & Guitar, 1994). But these are not necessary or beneficial for all children (Zebrowski, Weiss, Savelkoul, & Hammer, 1996).

Therapies based on multifactorial models

There are a number of therapy programmes that have been developed from the theories of stuttering as a multifactorial disorder. In each of the following programs, therapy encourages parents to make changes to their interaction with the aim of creating a more fluency and communication enhancing environment. Each of the methods emphasises the need to consider the individual child's strengths and needs and to develop an individualised program accordingly. The programs are guided by principles of intervention rather than being a set of standardised procedures. While often described as indirect, all of the programs describe direct speech work as an option to be considered as part of a child's therapy program.

The original Demands-Capacities approach (Gottwald, 2010; Starkweather, Gottwald, & Halfond, 1990), and the version of the approach known as RESTART-DCM (Franken & Putker-de Bruijn, 2007) aim to achieve a balance between the demands to communicate and the child's motor, linguistic, socio-emotional, and/or cognitive capacities. While direct fluency therapy may be introduced early in Gottwald's (2010) Demands-Capacities approach, with RESTART-DCM, the internal and external demands on the child's communication are addressed in the first instance and explicit training on the child's capacities may be added after the demands have been addressed. Both Family Focused Therapy (Yaruss, Coleman, & Hammer, 2006) and Palin Parent-Child Interaction Therapy (Kelman & Nicholas, 2008) have been influenced by the Demands and Capacities model, but have other influences and differing methods. This is not an exhaustive list of multifactorial programs, but these programmes are all readily available for implementation.

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Palin Parent-Child Interaction Therapy

Palin Parent-Child Interaction Therapy (Kelman & Nicholas, 2008) begins with a comprehensive assessment process, which includes a detailed case history taken from the parents. Both parents are involved from the beginning and throughout the therapy. A child assessment includes formal assessment of the child's receptive and expressive language skills, evaluation of stuttering frequency and severity, and delivery of a child interview to explore the child's awareness of the stuttering and the degree to which it is a problem. Using the information and results from the assessment, the therapist is able to complete a Summary Chart to highlight the factors that are relevant for the child, thinking about the physiological, linguistic, psychological and environmental strengths and needs. The management section of the Summary Chart helps the therapist to consider the interaction, family and direct strategies that might be relevant for the child. A short video of each parent playing with the child is made to help identify the strategies that the parent is using that are helpful for the child and potential adaptations that might be helpful in light of the child's identified needs.

Therapy takes place over six once-weekly sessions that last for approximately 1 hour. During these first sessions, the indirect components of the program—the interaction and family strategies—are introduced. Interaction strategies are selected by parents through video observation and might include strategies such as slowing their own rate of speech or increasing pausing. Family strategies might include being open about stuttering, or confidence building. The parents practise the interaction targets during a 5-minute play time at home, which each of them has with the child between three and five times a week. They incorporate family strategies into the family life and routines in the home. Following these sessions, there is a 6-week break from clinic therapy, during which the parents continue to implement the strategies at home and feeds back regularly to the therapist.

At the end of this consolidation period, progress is reviewed at a clinic session and further management decided as appropriate. This may include further interaction or family strategies or direct child strategies might be introduced, such as slowing the pace of talking or rate of speech through techniques such as "bus talking." Most children do not require the direct strategies because most achieve fluency through the indirect components. However, the following may indicate that direct strategies should be introduced: If the child is stuttering more or no less than the start of therapy, reacting negatively, avoiding speaking or changing words, expressing frustration or being upset, trying to find strategies to manage the stuttering. Further, if parental concern is not reducing and they are not noticing signs of improvement beyond the clinic, direct strategies may be indicated. The child's age, level of attention, cognitive skills, metalinguistic skills, and sensitivity to the stuttering will influence how this is managed.

The child's progress is monitored for up to a year post therapy, with regular assessment of stuttering severity and the child's attitude to communication (Vanryckeghem & Brutten, 2007). The following are also assessed during this period: The parents' perspective of the impact of stuttering on the child, the severity and impact on the parents, and the parents' knowledge of stuttering and how to manage it using the Palin Parent Rating Scales (Millard & Davis, 2016)

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Research findings about the topic

There are a number of models used to describe and interpret levels of research evidence. The Oxford Centre for Evidence Based Medicine developed a Levels of Evidence Table (OCEBM Levels of Evidence Working Group, 2011) in response to criticisms of other tables and to encourage greater flexibility in interpretation of research evidence. The aim is to consider research designs in relation to the research questions they may be used to address. In response to the question "does this intervention help?" the Oxford Centre table records "systematic reviews of randomized trials or n-of-1 trials" trials as the highest level of evidence, followed by individual randomised trials, and then nonrandomised controlled cohort studies. However, it is not the design alone that contributes to the level of the evidence that emerges, but also the robustness of the design and the validity and reliability of the results. On this basis, with a randomised controlled trial for the RESTART-DCM treatment, and replicated n-of-1 studies for Palin Parent Child Interaction Therapy, these approaches have the highest levels of evidence of the multifactorial treatments available.

The RESTART trial

The RESTART trial (de Sonneville-Koedoot, Stolk, Rietveld, & Franken, 2015) is a multicentre randomised controlled trial with an 18-month follow-up. Children aged between 3 and 6 years were randomly allocated to one of two treatment groups using a minimisation process to take account of factors thought to affect outcome: gender, stuttering severity, time since onset, family history, therapy during the previous year, and clinician. One treatment was RESTART-DCM (Franken & Putker-de Bruijn, 2007) and the other was the Lidcombe Program (Packman et al., 2015).

Based on 176 children there was no significant difference between the two groups 18 months after the start of treatment with respect to stuttering severity (de Sonneville-Koedoot et al., 2015), however severity did differ between the groups at different time points. For both treatments, most improvement occurred during the first 3 months after the start of treatment, after which stuttering frequency plateaued. Children who received the Lidcombe Program had higher stuttering frequency at baseline and lower at 3 months after the start of treatment. There was a significant difference in parent ratings of stuttering severity over time. Small effect sizes meant that differences in clinician severity ratings at 18 months after the start of treatment were deemed to be negligible. There were no differences in quality of life, communication attitude or behaviour, and there were no significant differences between the number of sessions or the number of treatment hours between the two groups (de Sonneville-Koedoot, Bouwmans, Franken, & Stolk, 2015; de Sonneville-Koedoot, Stolk, Raat, Bouwmans-Frijters, & Franken, 2014; de Sonneville-Koedoot, Stolk, Rietveld, & Franken, 2015). The researchers concluded that the outcomes for both treatments were comparable and proposed that the common factors across the programs may be of greater importance than any differences.

Palin Parent-Child Interaction Therapy

In single subject experimental designs, multiple measures are collected over a baseline period so that the individual can act as their own control and change can be measured and interpreted in comparison to the baseline data. When properly designed and analysed, it is possible to attribute change to the therapy, rather than any other developmental factor. The internal validity is extremely high. While the findings cannot be generalised beyond the individual studied, the external validity is increased by replication. What cannot be

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extrapolated from these studies are population effects (Kazdin, 1982; McReynolds & Thompson, 1986; Zhan & Ottenbacher, 2001).

There are two papers reporting single subject data for Palin PCI, replicated across a total of 12 children (Millard, Edwards, & Cook, 2009; Millard, Nicholas, & Cook, 2008). In these studies, all participants had been stuttering for a minimum of 12 months, and therefore were at greater risk of persistence. Multiple weekly measures of stuttering frequency were taken from speech samples of the child playing at home with each parent. These provided naturalistic beyond clinic speech samples to determine whether there was a trend in the data prior to the start of therapy, to explore whether each child was resolving the stuttering before therapy began.

These measures continued to be made on a weekly basis during the 6-week period of clinic therapy and for the next 6 weeks while the parents continued to implement the therapy at home. In the final phases, Millard et al. (2009) collected data for 6 weeks before the 6-month follow-up and in the 2008 (Millard et al., 2008) study, speech samples were collected once a month for 1 year post therapy. In both studies the data were analysed using Cumulative Sum analysis (Montgomery, 2007). This analysis detects any systematic changes in the data that are over and above what would be expected given the range of variability in the baseline phase. In addition to stuttering frequency, parents' perceptions of change were also explored using an early version of the Palin Parent Rating Scales (Millard et al., 2009), to reflect the broader aims of the therapy program. The studies concluded that the therapy was effective in reducing stuttering frequency and the impact of the stuttering on the child and parents. Parents also reported feeling more knowledgeable and confident in managing the stuttering.

Clinical application of the research findings

There are a number of clinical applications of the evidence pertinent to the Palin Multifactorial Model:

Physiological factors

Given the increasing evidence that indicates that children are born with the predisposition to stutter, parents can be reassured that the stuttering is not caused by something they have done or haven't done. This is supported by the common sense observation that there are many children who are exposed to tragic or dramatic live events or circumstances who do not stutter. Guilt is a common emotion that parents experience in relation to a child's stuttering (Plexico & Burrus, 2012) and one that should be addressed as part of the therapy process.

Evidence that children who stutter have a less robust or efficient speech motor system suggests that they will require additional time to function optimally. Further, attempting to ensure that the child's physiological well-being is maximised should also be helpful. For example, ensuring that the child is having enough sleep can be beneficial. During times of sickness or stress the child may stutter more. Helping parents to recognise this and may enable them to alleviate anxiety during such times.

Linguistic factors

It is clear that the child's language skills should be assessed, for several reasons: there may be underlying language needs, the stuttering may mask language problems, the stuttering

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may be a manifestation of language formulation difficulties, and use of more complex language may influence stuttering frequency. Therapy to support the child's linguistic development may be required to support communication and fluency. It will also be important to consider the relationship between the child's use of language and stuttering, as well as the development of language over time and the linguistic environment of the child.

Psychological factors

There is no evidence that there is a particular temperament that is characteristic of children who stutter, but it is important to take into account the child's temperament when understanding the impact that the stuttering has or may have in the future. A child who rates highly on the temperament dimension of "withdrawal" may be more likely to avoid speaking situations. A child with high "attention" levels may pay greater attention to disruptions to fluency. A child with high "reactivity" will reach the threshold of arousal sooner than others and so may experience stress in situations that others would not react to. The less effortful control children exhibit, the more stress they experience. Reactivity and self-regulation are important factors in development of self-monitoring and awareness in young children who stutter and perhaps they are less able to manipulate their attentional, behavioural, and emotional responses, and hence may be less able to effectively manage their stuttered speech or reduce its severity. While temperament is predominantly static, regulation develops over time. Parents can be supported to find ways to help regulate attention and emotions for the child in the first instance, and to develop the child's ability to self-regulate over time. Resilience is the ability to adapt to stressors and to "bounce back" from difficult situations and experiences. Clearly for children who stutter and their parents, resilience is a characteristic to be encouraged. Resilience can be taught, underlying skills such as flexible thinking and problem solving can be introduced, and confidence developed in both child and parents.

Parents experience a range of emotions and fears for their children in the short and long term. Acknowledging and normalising these worries and emotions, providing information, helping them to identify the bases of their worries and to explore those, can be extremely helpful in realigning parents' attention and ensuring a realistic balance between fears and likely reality. Having coping skills and strategies to draw on should their worst fears be realised will help to reduce the intensity of the worry. Children exist within a family system and will be influenced by the attitudes and behaviours of those around them. Helping parents to feel more confident and positive about the future, even if the stuttering should continue, will be important. Encouraging open discussion about stuttering helps avoid the "conspiracy of silence," encourages desensitisation in both the child and parents, and ensures that the channels for communication are open should they be required in the future. In preschool children, the use of praise, empowering parents to support the child, encouraging joint problem solving, and taking a strengths focused approach to intervention, will help promote a positive and constructive approach to management.

Environmental factors

Parents are able to introduce routines and structure that support physiological factors such as eating and sleeping routines. They can support the child's linguistic development and encourage the long term coping skills such as resilience, flexible thinking and problem solving skills. They are also able to make some changes to their own interaction, which, in addition to addressing the physiological, linguistic and emotional factors, also help to create a more communication and fluency enhancing environment. These might include targets

such as slowing their rate of speech, increasing pausing and following the child's lead in play.

Summary and conclusions

Multifactorial models provide a framework for understanding the complexity and interactions of variables related to stuttering. When therapy is informed by an understanding of stuttering as a multifactorial disorder, it is clear that therapy needs to be individualised and flexible to meet the needs of the child and parents. To do this, clinicians need to understand the principles of the programmes rather than follow step by step instructions. These approaches are evidence based, and given that the aims extend beyond the reduction of stuttering frequency, the concept of successful therapy needs to be expanded to include a reduction of the behavioural and affective impact on both child and parents.

It is clear from all research into therapy with this age group that children's response to therapy is not uniform. The Palin Parent-Child Interaction Therapy and RESTART-DCM research demonstrates that the findings from longitudinal studies of risk for persistence in non-clinical populations of children who stutter may not be directly generalised to the population of children who present to clinics. It would seem that there must be some other factors or combination of factors that influence outcome, other than gender, family history or time since onset. Both the Palin Parent-Child Interaction Therapy and the RESTART-DCM studies have shown that the most therapeutic effect is evident in the first 3 months of therapy, so if there is no progress during this time then the therapy should be reconsidered.

Finally, in terms of future directions for research, it would seem that the similar outcomes observed across the therapy programs in the RESTART study suggest that it may be the factors that are common to the approaches that are critical for outcome, rather than the differences. Exploring what these might be could enhance the effectiveness of the therapy as well as streamline the methods. We also need to better understand which children respond well to therapy, which variables may predict outcome, and thereby find additional methods to support those children whose needs will not be met by current approaches.

Questions from discussion leaders

How many children require only the indirect treatment components of Palin Parent-Child Interaction Therapy and not the direct component?

We haven't studied this systematically, but the data from our single subject papers would suggest two thirds. We have unpublished data that we are currently analysing for 27 children who we have monitored for 1 year from the start of therapy. Of those, six received direct components within that time. So an approximation of two thirds would seem to be reasonable.

Why have you used cumulative sum analysis in clinical research? Isn't the major issue pre-to-post reductions of stuttering?

We do want to know whether there is a reduction in stuttering. However, there are two serious problems with using pre-post frequency measures with this age group. The first is the issue of spontaneous recovery. While we don't know non-treatment recovery rates for clinical populations, it is likely that a proportion of these children will resolve the problem without therapy. So when exploring treatment effect, we want to know if the stuttering is

improving before the therapy begins, and if any observed change is beyond that. The second issue is that stuttering can be highly variable in this age group, so single measures may not reflect true differences, merely a higher or lower score which is within the typical range for the child. Cusum analyses explore whether there is a systematic change—a trend in the data—compared with the baseline. If the cusum limits are crossed then you can assume that the increase or decrease is greater than you would expect for that child, given the variability in the baseline phase. Thus, our results indicate that Palin Parent-Child Interaction Therapy promotes a systematic decrease in stuttering frequency over time that cannot be explained by the natural variability in the child's stuttering.

What are your criteria for adding a direct approach during therapy with child who stutters?

This would be a decision made jointly with the parents and based on ongoing assessment of the child's strengths and needs in the context of the child's environment. Given our data, we would expect the parents to be reporting improved fluency and a reduction in their concern by the time of the first review, 3 months after the start of therapy. If this is not the case, then we would discuss the introduction of more direct strategies with the child. It is important to note that "openness" is encouraged even within the indirect components. If the child is becoming more concerned, the impact of the stuttering is increasing, or if the child is developing unhelpful strategies such as avoidance or concomitant movements, then we would consider the introduction of the direct strategies in addition to the indirect.

Which of the two treatments, Palin Parent-Child Interaction Therapy and RESTART-DCM, do you think has the more compelling evidence for treatment efficacy?

Both approaches have evidence that they are efficacious and this is important because it gives us options for our choice of intervention. The researchers arrive at this conclusion through differing methodologies, but the important question is whether changes in the dependent variable can be attributed to the independent variable. The Palin Parent-Child Interaction Therapy research demonstrates that stuttering and its impact is reduced directly by the therapy. RESTART-DCM research shows that stuttering frequency reduces over time for the majority of children who receive the program, at a rate that is similar to the Lidcombe Program. Further research is needed for both, and you could argue that each program would benefit from answering the question explored by the other and applying the alternative methodology. Palin Parent-Child Interaction Therapy needs to be informed by population effects and RESTART-DCM needs to be informed by no-treatment controls to account for natural recovery and variability and to increase confidence that the results are a direct result of the intervention.

You seemed to imply that n-of-1 research with single cases is as influential as clinical trials with 200 participants such as the RESTART trial. Is that what you intended to convey?

In terms of answering the question "does this therapy work?" single subject trials provide strong evidence about whether the dependent variable is changed by the introduction of the dependent variable. These are different to case study reports which are non-experimental. Participants in the n-of-1 trial act as their own controls and so the internal validity is extremely high and we can be confident that change is the direct result of therapy. The external validity— generalisation of the findings to other children—is limited with one trial, but is increased through replication of cases, of which there are 11 for Palin Parent-Child Interaction Therapy. In answer to the question "are they as influential?" the answer would be no. This is not because of the strength of the findings, but more because the

methods and data are not understood and there is a strong bias towards randomised controlled trials. The most important question that we should be asking is whether the methods and the evidence are robust enough to answer the question "does this therapy work?". This is true for both n-of-1 trials and randomised controlled trials.

We would like to pursue your response to the last question a little further, where you assert that n-of-1 trials and randomised controlled trials are both robust enough to answer the question "does this therapy work?". So if that is correct, why would health care researchers go to the effort of randomised controlled trials that involve many participants? Why wouldn't they simply do n-of-1 trials?

Randomised controlled trials have higher external validity, so the findings may be generalised to a similar population and context beyond the study. They are designed to explore population effects so it is possible with a randomised controlled trial to determine the proportion of children who are likely to respond to the intervention and to what degree. So while both methods answer the question "does this therapy work?", n-of-1 trials are not designed to answer these additional questions about the likely impact on large numbers beyond the study. We need different methodologies to explore different questions and we can combine methods to give us richer data and greater knowledge.

Question from editors

You state that

> *… the Oxford Centre table records "systematic reviews of randomized trials or n-of-1 trials" trials as the highest level of evidence, followed by individual randomised trials, and then nonrandomised controlled cohort studies. However, it is not the design alone that contributes to the level of the evidence that emerges, but also the robustness of the design and the validity and reliability of the results. On this basis, with a randomised controlled trial for the RESTART-DCM treatment, and replicated n-of-1 studies for Palin Parent Child Interaction Therapy, these approaches have the highest levels of evidence of the multifactorial treatments available.*

However, there are many common meanings for n-of-1 trials, but the Oxford Centre defines it as

> *n-of-1 trial: A variation of a randomized controlled trial in which a sequence of alternative treatment regimens is randomly allocated to a patient. The outcomes of regimens are compared, with the aim of deciding on the optimum regimen for the patient. (Centre for Evidence Based Medicine, 2017)*

As such, some clarification is needed, because the trials of Palin Parent-Child Interaction Therapy to which you refer contain only one treatment.

The problem with this definition is that it is not only difficult to find on the website, but this is all that exists, in contrast to other research designs which are explained and discussed in much greater detail. As with the randomised control trial, in an n-of-1 trial the comparison "treatment regime" may consist of an alternative treatment, placebo, or a no treatment condition. In its simplest form, examining the individual's response to intervention can be explored by exposing the participant to one condition followed by the other (AB or BA) (Kravitz et al., 2015, p. 5). But it is for the researcher to strengthen this basic design to increase the validity of the findings and the reader to consider whether the design, methods and data answer the question "does this intervention help?" In their guidelines about how to

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use the Oxford Centre Levels of Evidence table, the authors are clear that the table is "not intended to provide you with a definitive judgment about the quality of evidence. There will inevitably be cases where 'lower level' evidence—say from an observational study with a dramatic effect—will provide stronger evidence than a 'higher level' study" (Howick et al., 2011, p. 2). This is only a tool to assist, "no evidence ranking system or decision tool can be used without a healthy dose of judgment and thought" (Howick et al., p. 1). So I would always encourage people to go back to the source, appraise original research documents themselves, and make their own judgments about whether the authors' conclusions are reasonable, all things considered.

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Treatment of School Age Children who Stutter

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Abstract

The primary purpose of this presentation is to review the available empirical evidence about the important topic of speech treatments for school age children who stutter. For 6–12 year olds, there is nonrandomised clinical trial evidence for three treatment types: the Lidcombe Program, speech restructuring, and syllable- timed speech. A recent trial has been published for a hybrid treatment involving verbal contingencies and syllable-timed speech. For all those treatments, there is an adjunct video-self modelling procedure for which there is supportive evidence. All these sources of clinical evidence are considered in making a recommendation about a clinical approach with this age group of clients who stutter.

Clinical importance of the topic

The topic of clinical management of children aged 6–12 years who stutter is clinically important for three reasons.

Mental health

The first reason clinical management of this age group is important is that if they continue to stutter they are at risk of developing mental health problems that will affect their quality of life for a lifetime. One of the most common of such problems that affect older clients is social anxiety disorder (Blumgart, Tran, & Craig, 2010; Iverach et al., 2009; Stein, Baird, & Walker, 1996), which typically begins right in the middle of this age range. Added to that are the issues that school age children are known to be targeted for bullying and rejection by peers (Davis, Howell, & Cooke, 2002; Langevin, Kleitman, Packman, & Onslow, 2009; Mooney & Smith, 1995), and that the negative effects on mental health of bullying during childhood are well known (Giora, Gega, Landau, & Marks, 2005; Gladstone, Parker, & Malhi, 2006; McCabe, Miller, Laugesen, Antony, & Young, 2010). That is the last thing a school age child who stutters needs. There is also some evidence from a large cohort from the United States National Health Interview Survey showing that stuttering school children are significantly more likely to repeat a grade than control children (Boyle, Decoufle, & Yeargin-Allsopp, 1994). Such educational problems obviously can have lifelong consequences.

The urgency of intervention during the school years

The second reason that this topic is clinically important is the sheer urgency of it. It is widely accepted that the most important goal for preschool children who stutter is that they should stop stuttering as soon as possible. We don't think there has been any argument offered during this symposium that would contradict this proposition, and it is arguable that this is the case also for school age children. They are not yet at the age when we should consider any alternatives as a prime clinical focus, such as counselling them about dealing with the fact that they stutter. The prime clinical focus should be to urgently find a way to reduce their stuttering as much as possible and as quickly as possible. That is not to say that we should not do other things clinically, such as counselling and finding ways to help them deal

with the inevitable problems that school age children have at school, such as bullying, as Jane Harley will discuss this afternoon. But this age group should stop stuttering as much as possible and as quickly as possible. So, for that reason, our focus here is on methods to control the stuttering of school age children, even though that is not the entire clinical story.

The challenge of evidence based reasoning with school age children

Finally, the topic of school age children is critically important because of the challenge that it presents for evidence based clinical reasoning. It is concerning that there is no randomised controlled trial evidence available for this age group. But the argument here is that this is not the end of the story. Why? Because this age group is unique, being located developmentally in the middle of two age ranges for which there is indeed compelling randomised controlled trial evidence. There is replicated randomised controlled trial evidence for the Lidcombe Program with preschool age children up to 6 years (Arnott et al., 2014; Bridgman, Onslow, O'Brian, Jones, & Block, 2016; De Sonneville-Koedoot, Stolk, Rietveld, & Franken, 2015; Jones et al., 2005), and hopefully before long there will be such replicated evidence to support multifactorial treatments for preschoolers. For adults, there is clinical trial evidence for several treatments, particularly for speech restructuring (for a review, see Onslow, 2017, Lecture Eight). So, during the evidence based clinical reasoning cycle with school age children who stutter, we can be informed not only by clinical trials directly with children 6–12 years old, but also by clinical trials for preschoolers and for adults.

So, for the following discussion, client need has been taken out of the evidence based reasoning cycle, and the focus is on clinical evidence, with the assumption that clinicians have benchmark clinical skills in doing treatments that are supported by such evidence. Subsequently the discussion will turn to that other, much overlooked, component of evidence based clinical practice, which is common sense. In all of this talk about scientific evidence we don't want to lose sight of that.

Research findings about the topic

Clinical trials

For this discussion, the clinical trial is considered to be the fundamental unit of clinical research that is informative for clinicians (Onslow, Jones, O'Brian, Menzies, & Packman, 2008). What, then, is a clinical trial of a treatment designed to control stuttering? It depends on individual perspectives, but Onslow et al. (2008) define it as a prospective attempt to determine the efficacy of an entire treatment, with speech measures gathered outside the clinic and a reasonable follow-up period. That is not to say that other research such as file audits or case studies have no value. To the contrary, they are indispensable developments leading up to clinical trials.

Onslow et al. (2008) suggest that 3 months is a reasonable follow-up period, but here the suggestion is that 12 months post-treatment might be such a reasonable follow-up. Also, here we give consideration only to clinical trials with at least 10 children who completed the treatment and who were in the trial at follow-up. That seems to be a reasonable proposition. Clinical trials involve a sample group from the population of children who stutter, with the intention of generalising to some extent from that sample in the clinical trial to the clinical population of children who present to clinics. Any fewer than 10 children just does not feel all that much like such a group, so that is our cut-off point.

The Lidcombe Program

So onward to the first set of research findings. That is fairly simple. There is compelling evidence that the Lidcombe Program is efficacious with under 6-year-olds, and that based on that information there is some likelihood that the Lidcombe Program will be efficacious with a slightly older child in the range 6–12 years. Of course, the Lidcombe Program will need to be adapted for use with older children. Compared to preschoolers, parents will use different language to present verbal contingencies to school age children, and they will use different activities during the practice sessions of the Lidcombe Program.

There will be other differences to preschool treatment with the Lidcombe Program with this older age group. For example, parents have restricted access to children of this age, so children will be expected to be more proactive in self-administering the treatment than is the case with preschoolers. Also, school age children can be expected to make an important contribution to the Lidcombe Program treatment process by contributing self-reported severity ratings. So, the clinician does not have to rely exclusively on parent severity ratings.

A nonrandomised clinical trial has been published with the Lidcombe Program for 11 children in the school age range (Lincoln, Onslow, Lewis, & Wilson, 1996). During the 12 months post-treatment period, there seems to be a treatment effect. We say "seems to be a treatment effect" because you can never be really certain with nonrandomised trials. They tend to overestimate effect sizes. But it seems fair to say that at 12 months post-treatment the children seemed to have greatly reduced stuttering in the range of 1–2 percent syllables stuttered (%SS). What is immediately apparent is that these treatment effects are not as good as with preschool children treated with the Lidcombe Program, for whom we are accustomed to seeing almost zero %SS at follow-up. This is a recurring theme throughout all the clinical trial data presented in this presentation, highlighting the earlier point about the sheer clinical urgency of treating this age group of stuttering children.

Speech restructuring

Let's go to the other side of our age group, to adults, to look for management clues for school age children. There is compelling evidence that speech restructuring treatment is efficacious when adults wish to control their stuttering. And again there is direct evidence with school age children that this treatment may have value. A nonrandomised clinical trial of speech restructuring involved twenty-five 9–14 year olds who participated in a parent-conducted speech restructuring program compared to a control group (Craig et al., 1996). We have the same picture that indeed there are gains to be had from this clinical method at 12 months post-treatment, although it seems that the treatment effect is not quite as good as with the Lidcombe Program, with stuttering rates more than 2.0 %SS.

The other consideration with this treatment is that post-treatment speech does not sound completely natural. There was evidence in the Craig et al. (1996) trial that the children did not sound all that natural at the end of it. We need to keep these considerations in mind with this client age group. We need to be careful because school age children who stutter are prone to social anxiety, peer rejection, and bullying.

A hybrid treatment

The next treatment up for consideration is a hybrid treatment, meaning a treatment made up of two treatment styles. Over the years we have heard a lot about this style of thing—holistic, flexible, multifaceted approaches to treatment. It sounds like a good thing on the face of it, and logically it might be a good thing. If there are two styles of treatment known

to be efficacious and associated with a certain treatment effect, then there might be clinical benefits from combining them. Perhaps if, for example, two treatments associated with an 80% reduction of stuttering are combined to one treatment then the effect size would hypothetically increase to more than 80%.

Andrews et al. (2016) published a nonrandomised clinical trial with 19 children followed up for 12 months post-treatment. The treatment was a combination of syllable-timed speech and the verbal response contingent stimulation procedure that parents use during the Lidcombe Program. Syllable-timed speech is of course nothing new. According to ancient texts about the matter, the Greek actor Satyrus was the first speech therapist, who helped Demosthenes, who stuttered, to attain his greatness as an orator by treating him with syllable-timed speech. A clinical trial was published in 2012 (Andrews et al., 2012) with school age children that did not quite make the cut for presenting today, because it had only nine children with 9 months follow-up. But it was sufficiently encouraging to warrant continued development by combining it with verbal response contingent stimulation.

How does the hybrid treatment work? The syllable-timed speech component of the treatment works simply with parents practising syllable-timed speech during a conversation with their children for 5–10 minutes 4–6 times per day. The Andrews et al. (2016) clinical trial involved 19 school age children for whom there were outcome data at 12 months post-treatment. It is possible that combining syllable-timed speech and verbal response contingent stimulation might achieve something more than with just verbal response contingent stimulation, but the result of this trial suggested that proposition to be obviously wrong.

But there is something of note about this hybrid treatment. To assess how the children sounded after treatment, the researchers presented pre- and post-treatment recordings of the children to untrained listeners. They asked them to assess rhythmicity on a five-point scale where 0 equalled not at all rhythmic and 4 equalled extremely rhythmic speech. Surprisingly, the children sounded less rhythmic after treatment according to the numbers and a *t*-test of the numbers.

Other options

So, for this age group, the evidence based options we have are the Lidcombe Program, a speech restructuring program, and a hybrid syllable-timed speech plus contingencies treatment. After that, evidence based options are few. There have been variants of standard treatment proposed during past decades for this age group, but they have not been trialled clinically to the standards defined for inclusion in this presentation, even though those standards are quite liberal. Examples include self-imposed time-out (de Kinkelder & Boelens, 1998), Gradual Increase of Length and Complexity of Utterance (Ryan & Van Kirk Ryan, 1983), and syllable-timed speech alone (Andrews et al., 2012). The numbers in these studies were quite small, significantly less than the 10 used as a benchmark for inclusion in the present discussion. Also, there have been laboratory reports of self-modelling with this age group, which show promising results (Bray & Kehle, 1996; 1998; 2001).

The Holy Grail of stuttering treatment is a device that can do treatment for us. There was a brief hope a while back that it may have been within our grasp with an encouraging clinical trial of EMG biofeedback with school age children (Craig et al., 1996). However, there were two subsequent failures to replicate that result (Block, Onslow, Roberts, & White, 2004; Huber, O'Brian, Onslow, Packman, & Simpson (2003), so that device cannot be

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recommended. So far there has been no attempt to do a clinical trial of any altered auditory feedback device such SpeechEasy with school age children, which is not surprising considering the negative results from the only trial of that device conducted so far with adults (Pollard, Ellis, Finan, & Ramig, 2009).

Clinical application of the research findings

How you apply all this with school-age children in the clinic is reasonably self-evident. Although the clinical trial evidence is weak, three treatments comprising non-randomised trials can be recommended: Lidcombe Program, a hybrid syllable-timed speech and Lidcombe Program Treatment, and speech restructuring.

What is a realistic speech treatment outcome?

That being the case, it is clear that by the time children have been stuttering into the school years, the treatment effects are not as compelling as with treatment during the preschool years. It seems inescapable to conclude that residual stuttering in the range of 1–3 %SS should be expected at the best with school-age children. Still, that treatment effect is well worth having and it remains to be seen with future randomised controlled trials with this age group whether or not that is an accurate estimate of potential outcomes. Also, it remains to be seen whether that stuttering reduction is sufficient to prevent the onset of the mental health and associated quality of life problems caused by this disorder.

A suggested sequence

But for now, and with regard to the clinical trial evidence of stuttering school age children, what we have in effect is a set of three treatments that present a clinical sequence from simplest to most complex. Start by considering the simplest treatment first, the Lidcombe Program. Second, consider the hybrid syllable-timed speech and Lidcombe Program treatment. As a last option, consider the most complex treatment, speech restructuring.

What is the justification for that recommendation? First, the Lidcombe Program is the simplest treatment and there is no way that it will cause children to sound unnatural. Then, the hybrid syllable-timed speech and Lidcombe Program treatment seems to be the next simplest treatment, and preliminary indications suggest that it might work without promoting unnatural sounding speech. And speech restructuring treatment definitely will make your clients sound unnatural. This is not at all desirable and should be avoided if possible with this client age group who are notoriously prone to social anxiety, peer rejection, and bullying.

Some clinical common sense

So now, as foreshadowed, some clinical common sense when applying the research described to your caseload. First, it is perfectly true that the clinical trial evidence for this age group needs to be further investigated to include the gold standard randomised controlled trials. But the clinical trial evidence for the Lidcombe Program with children younger than 6 is compelling, and does comprise randomised controlled trials. Therefore, for any individual school-age child who stutters who is at the young end of the school age range, make it routine to attempt the Lidcombe Program as a first treatment of choice. How young? That is a question that only clinical common sense can answer. Should that recommendation be overturned with the inevitable cases that have had the Lidcombe Program and experienced relapse? That is more than common sense; it is a matter of sophisticated clinical judgement.

Self-modelling

The other common sense application of scientific evidence to school age children concerns self-modelling. In short, self-modelling is positive behaviour change based on people observing themselves being free of a problem behaviour, in this case speaking without stuttering.

There has been evidence that indicates self-modelling can aid treatment. There has been a randomised controlled trial of this procedure with adolescents and adults as a supplement to speech restructuring treatment (Cream, O'Brian, Onslow, Packman, & Menzies, 2009). That trial showed improvements in self-reported stuttering severity and quality of life when it was added to standard treatment. And a nonrandomised trial showed that it was particularly valuable as a response to post-treatment relapse (Cream et al., 2009). With 10 participants who had relapsed after successful treatment, there was an impressive recovery of speech skills simply by having the clients watch tapes of themselves speaking without stuttering. So, even though there has not been a clinical trial specifically with school age children, we think it is a justifiable recommendation that with every one of your school age clients you make a self-modelling tape for them to view regularly during treatment and to keep for use in the event of post-treatment relapse. Why wouldn't you?

Summary and conclusions

To conclude, we suggest with school age children who stutter to consider the simplest treatment first and to consider the most complex treatment last. We suggest that all treatments with this age group are supplemented by self-modelling. And we have a final common sense suggestion about what you should do in the clinic with the empirical information presented here. There is nothing in the data that say that this is true; it is purely clinical common sense. With a school age child anywhere in the range from 6–12 years, what you need to do in deciding about treatments is to give more preference to the simpler treatments for the children with milder stuttering, and give more preference to the more complex treatments for children who are more severe.

Questions from discussion leaders

Recent evidence challenges the clinical importance of parent verbal contingencies during childhood stuttering treatment, so could you justify their inclusion in a syllable-timed speech treatment?

It is misleading to say that the clinical importance of parent verbal contingencies during childhood stuttering treatment has been challenged. It is more accurate to say that the contribution of verbal contingencies to obtaining Lidcombe Program treatment effects has been challenged. At present the mechanisms underlying the Lidcombe Program of early stuttering intervention are unknown, but it is far too premature to conclude that verbal contingencies have no clinical value for children of any age. The collective weight of laboratory studies indicates to the contrary. In fact, comparison of the two syllable-timed speech trials we mentioned during our presentation (Andrews et al., 2012; 2016) suggests that verbal contingencies could contribute clinically. The former trial contained no verbal contingencies and the latter did, and the latter trial reported a far better clinical result.

Why have there been so few clinical trials for school age children who stutter?

It is difficult to know the answer to that question but we would venture that it relates to the substance of our presentation. Simply, the treatment choice for this age group is more challenging than with other age groups, therefore doing clinical trials with them is more challenging than with other age groups.

Knowing how crucial this treatment is at this age, how do you proceed if a school age child does not wish to engage?

That question has no answer because, obviously, no clinician would attempt a treatment that a child does not wish to engage with. If you are asking what should be done in the event that assessment shows there is no treatment a child will engage with, the answer is equally obvious. If you are asking about clinical strategies for when a child's commitment to treatment wanes, the answer lies in the basic methods of speech-language pathology.

What do you think are the most important research directions now for this age group of children who stutter?

Those research directions that will generate data that guide clinicians in making decisions about which of the treatment options we discussed—or any others—are suitable for individual children in the 6–12 years age range. At present, clinicians have almost no empirical guidance about clinical choices for any of the different ages and clinical presentations of children in that age range.

Do you think that if stuttering is controlled during the school years that psychological problems will not develop, or will stop developing if they have begun to do so?

The answer to that question is informed by data that negative social conditioning experiences can occur with stuttering during the pre-school years (Langevin, Packman, & Onslow, 2009), the onset of psychological problems with school age children who stutter may be as early as 3 years (McAlliser, 2016), and that mental health disorders are commonly comorbid with stuttering during the school years (Iverach et al., 2016). There is no guarantee that school age children who stutter will be immune from anxiety related psychological problems. If noxious conditioning experiences occur early in life the scene could be set for later problems associated with negative social conditioning. Even if a school age child shows no sign of stuttering related mental health problems, a process may have started that will manifest later in life.

To follow up on your response to the last question, what would you do with a school age child if you were concerned about the development of anxiety or other mental health issues? In particular can you comment on this scenario for a child who may be reticent to engage in speech treatment?

That would be one case where consultation with a clinical psychologist would be the wisest and safest first course of action before considering any speech treatment.

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Treatment of Adolescents who Stutter

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Abstract

Treatment of stuttering in adolescents has long been considered to be highly challenging, especially because of high dropout and relapse rates. A few studies have demonstrated that treatment in peer groups can be moderately effective. The use of online treatment with webcam has also shown promise. Studies of treatment for clinical problems other than stuttering suggest that an important concept is client "readiness for change" prior to therapy and "stages of change" during therapy. With these concepts in mind, individual interviews and focus groups of both clients and therapists have suggested three treatment targets: (1) changing stuttering and talking, (2) reducing negative thoughts and feelings, and (3) reducing avoidance behaviours related to stuttering. Further studies are being conducted with teens to determine their readiness to undertake change in these three areas. In addition, scales are being developed to use with adolescents to help them develop goals for therapy in each of these areas and to assess their readiness to make changes in each area. It is hypothesised that the collaboration between adolescents and clinicians and the buy-in by the adolescents will improve treatment outcomes for this age group.

Clinical importance of the topic

A challenging age group

Treatment of adolescents who stutter can be highly challenging but it is also critically important. The urgent need for treatment at this age is because by the teenage years the window for spontaneous recovery has essentially closed. Almost all spontaneous recovery occurs within 4 years after onset (Yairi & Ambrose, 1999) and by the teenage years stuttering is essentially chronic.

Treatment for teens is also vital because they can suffer a great deal if their stuttering continues during this key period of their development. They report being bullied and teased by peers, they are socially isolated, and they can be morbidly anxious (Blood & Blood, 2004; Blood, Blood, Tellis, & Gabel, 2003; Huber, Packman, Quine, Onslow, & Simpson, 2004). If stuttering is not remediated by the end of the teen years and continues into adulthood, even more dire consequences can result. Adults who stutter can be severely affected socially, emotionally, educationally, and vocationally. They feel stigmatised, self-conscious, have poor self-esteem, social phobia, and report poorer quality of life (Bloostein & Ratner, 2008; Iverach et al., 2009; Yaruss & Quesal, 2004). Thus, if effective treatment can occur before adulthood, much pain and suffering can be relieved.

However, treatment for adolescents who stutter has long been regarded as difficult. One of us was taught by a professor who avowed that he never treated adolescents who stuttered—he just waited for them to become adults and ready for therapy. Indeed, several clinicians

attest to the difficulty of treating teenagers (Conture, 1996; Daly, Simon, & Burnett-Stolnack, 1995; Van Riper, 1973).

Why are teens so difficult to treat effectively? Some of the answers may be in the teenage brain. Or, rather, the stage of development reached by the brain in adolescence—between ages 10 to 25 (Steinberg, 2014). During the teen years, some parts of the brain are maturing faster than others. The teenage brain is more excitable, especially in response to emotional stimuli (Jensen & Nutt, 2015). Therefore, it appears to us that learning at this age can take place quickly. For example, classically conditioned responses to the negative emotions associated with stuttering will be strong, making the teenage years a prime time for developing escape and avoidance behaviours in response to actual or anticipated stuttering. In addition to the problem of teens easily learning maladaptive stuttering behaviours, teens are not good at "hanging in there" when therapy becomes demanding. In the teen brain, nerve fibres connecting the frontal cortex and pre-frontal cortex to the rest of the brain have not been fully myelinated, thus those areas are not yet "on line." This means that it is difficult for teens who stutter to endure the hard work needed for their successful treatment, because they cannot use the executive functions of those pre-frontal areas to both plan for the future and to realise that hard work now can mean easier speaking later.

The data about therapy for teens reflect these difficulties. Many authors have commented on the fact that a high percentage of teens are either unable to complete a treatment program or they relapse after treatment (Craig, 1998; Craig, Hancock, & Cobbin, 2002; Hearne, Packman, Onslow, & O'Brian, 2008). A prime example is by Hearne, Packman, Onslow, & O'Brian (2008), who conducted a speech restructuring treatment with three teenage boys. One did not respond to the treatment, one withdrew from treatment, and only one did well with the treatment.

The need to persevere

Despite the evidence that treatment of teens who stutter is difficult, there are also strong arguments for persevering in the search for effective approaches. One is that the teenage brain may be more ready for change than previously thought. Neuroscientists have recently discovered that the brain's plasticity is almost as great during adolescence as it is from birth to age 3 (Steinberg, 2014). Pruning of synaptic connections is at a peak during adolescence, but at the same time rewiring of the brain is occurring, providing opportunities for growth (Chugani, 1996; Tyler & van Harreveld, 1942). In fact, studies have shown almost five times the rate of formation of new neurons during adolescence compared to adulthood (He & Crews, 2007). This makes adolescence a prime time for new learning. One of the changes in the adolescent brain that can influence learning is how receptive the teenage brain is to reward as shown by large responses in the ventral striatum to dopamine (Galvan et al., 2006). There are conflicting reports as to whether only large rewards make a difference to teens or whether small rewards such as sweets can have a big impact (Desor & Beauchamp, 1987). In any case, the implications are that stuttering therapy with teens is likely to be more powerful if strong rewards are used for progress. Conversely, the teen brain is known to be relatively unresponsive to negative consequences (Crone et al., 2008; Gunther Moor, Crone, & Van der Molen, 2010), suggesting the use of even mild punishment is unlikely to be useful.

As the foregoing studies suggest, the teenage years may be the last opportunity that individuals have to make radical changes in their stuttering. Rather than wait until adulthood, as that professor advocated, the best time for treatment may be now. Recently, some new

approaches have given reasons for hope that therapies for teens can be improved. We will now describe these approaches.

Research findings about the topic

The table gives details about several recent experimental treatments for teens who stutter, some showing promise of new approaches. One older study (Boberg & Kully, 1994) has been included to indicate that a long intensive program with careful long-term follow-up can be successful. Later studies have tried to develop approaches that are less resource intensive.

Study	N	Ages	Treatment	Outcome
Boberg & Kully 1994	25	11–17 years	Intensive 2-week speech restructuring, fear and avoidance reduction, social skills training.	Mean of 3.9 %SS at 12 months post-treatment.
Hearne et al. 2008	3	13, 14, 16 years	Non-intensive in-clinic speech restructuring.	One participant benefited at 12 months post-treatment.
Fry et al. 2014	3	17, 17, 18 years	Intensive 10-day speech restructuring, CBT, and communication skills training.	Only one participant benefited at 5 months post-treatment.
Carey et al. 2012	3	13, 15, 16 years	Non-intensive webcam speech restructuring.	Mean 74% reduction of stuttering severity at 12 months post-treatment. One participant reduced avoidance of speaking.
Carey et al. 2014	16	12–17 years	Non-intensive webcam speech restructuring.	Mean of 2.8 %SS at 12 months post-maintenance. Mean avoidance of speaking reduced by one third.

Note: %SS = percentage syllables stuttered, CBT = cognitive behaviour therapy

Clinical application of the research findings

The most recent approaches to treatment of teens who stutter suggest that certain treatment components can improve long term outcomes. One important innovation has been delivery of treatment with webcam so that adolescents can work on their speech from home using their own laptops (Carey et al., 2012; 2014). The Camperdown program appears to be a good match for this approach, although many other therapies may be suitable for webcam delivery. Because teens are often very comfortable with technology, the future use of computer games and virtual reality activities for transfer may be engaging and effective.

Information gathered from teens about their preferences for therapy makes it clear that teens like to work in a group with other teens (Hearne, Packman, Onslow, & Quine, 2008). The teen group environment may have been an important reason for the relatively good

long-term success of the participants in the Boberg and Kully program (1994) listed in the table. If a group environment can be combined with webcam delivery, as suggested by Carey et al. (2014), a powerful new approach could be developed. Currently online group stuttering therapy is being conducted in Germany (Euler, Von Gudenberg, Jung, & Neumann, 2009).

Another component of the Boberg and Kully program that may have contributed to the relative success of their clients was the work on reduction of fear and avoidance. Research about treatment with adults who stutter suggests that unless avoidance is reduced considerably, long-term outcome is in doubt (Guitar, 1976). Reports on the webcam trials of therapy for teens (Carey et al., 2012; 2014) indicate that avoidances of speech situations was not reduced for many of the participants. This may have contributed to relapse, therefore future treatments should consider incorporating avoidance reduction as a component of treatment.

Readiness for change

Recently, there have been proposals that a major factor underlying treatment outcomes in stuttering therapy for adolescents is "readiness for change: (Floyd, Zebrowski, and Flamme 2007. Research in behavioural health psychology across range of different clinical populations has shown that one's readiness to take action in changing behaviour is a dynamic cognitive process consisting of sequential steps or "stages" that lead to active and observable attempts at behavioural change (Prochaska, 1999). Equally important is the observation that movement between stages, and towards action, is facilitated or hampered by the relative weight that one assigns to the pros and cons of making a change (decisional balance), as well as the degree of confidence that one has in being able to "stay the course" in difficult situations (self-efficacy). Results from numerous studies by the second presenter, currently in preparation for publication, have shown that these two key cognitive constructs–decisional balance and self-efficacy–are responsive to intervention and thus, along with stage of change, should be considered within the therapeutic framework.

Research in progress

The first step in understanding how to identify stage of change, decisional balance and degree of self-efficacy in any clinical population is to develop the means to assess the current status of each construct within the group of interest. Using an evidence-based sequential and, iterative process, the second author and her colleagues have created and validated self-report scales for the three core constructs of change readiness: stage of change, decisional balance, and self-efficacy, and are presently collecting responses to these scales from 300 adolescents and young adults who stutter. All three of these scales hinge on the development of a standard definition of stuttering management, one that emerged from a qualitative analysis of individual interviews and focus groups consisting of adolescents who stutter and stuttering specialists.

Each of these interviews and focus groups began with the investigator posing this question: "What would someone be doing if they were managing their stuttering?" The second of us, with research that is currently being prepared for publication with Naomi Hertsberg, presented a thematic analysis of associated interview and focus group transcripts, showing that these two groups of teens who stutter and stuttering specialists, define the construct of "stuttering management" as comprised of three parts:

> *(1) Learning and practicing strategies to change either talking or stuttering, or both,*

(2) Changing negative thoughts and feelings about speaking and stuttering, and
(3) Decreasing avoidance of speaking and stuttering.

The significance of this three-part definition of stuttering management to our ideas about acceptable treatment outcomes seems clear. That is, both adolescents who stutter and the clinicians who work with them agreed that effective stuttering management consists of not only behavioural change, but cognitive and affective change as well. And, it may very well be the case that adolescents who stutter may differ in their readiness to make changes in all three domains at the same time. This definition was used in development of a Stages Survey, which asks teens and young adults who stutter to indicate whether they are thinking about doing one or more of the three components that define "stuttering management" outlined in (1)–(3) above, and when they think they will be ready to do them. For example, "I am thinking about doing one or more of these things in the next six months," "I am planning to do one or more of these things in the next month," "I have been doing one or more of these things for more than or (less than) 6 months." If a stage-based model fits the responses collected, factor analyses will show that respondents fall into non-overlapping groups, or stages of readiness to manage their stuttering.

The arguments in this line of work in preparation by the second author are that while understanding where a teen is in readiness to manage stuttering is key in establishing treatment goals, this can only take a client so far. As previously discussed, the cognitive constructs that characterise stage and are predictive of stage movement are the active ingredients in therapy. Therefore, the second author developed a Decisional Balance Scale which represented the pros and cons of managing stuttering and a Self-Efficacy Scale to assess the degree that participants felt confident that they could continue to do something to manage stuttering in difficult or challenging contexts. The definition of "stuttering management" that was used for the Stages Survey was also used to develop these two scales. The same process of key informant interviews and focus groups was used to elicit items for these scales, and a thematic analysis was conducted. As an example, the results of this analysis yielded 62 distinct items that adolescents who stutter and stuttering specialists endorsed as representative of the pros and cons of managing stuttering. Field-testing of this scale was conducted in which a subset of teens who stutter were asked to provide feedback on the clarity and readability of the scale, and were also instructed to use a five-point Likert scale to rate the personal importance of each item to their decision to manage their stuttering. Examples of scale items that participants were asked to rate for personal importance included "I would be able to say what I want to say when I want to say it," "I might not be able to change the way I speak," and "my friends might not like that my speech is changing." Presently, the second author and colleagues are finishing analysis of stage, decisional balance and self-efficacy scales from a large group (N=300) of adolescents and young adults who stutter. The long-range goal of this work is to develop a computer-assisted "expert system" for both identifying and predicting stage and stage movement for this population, and for developing and testing stage-based therapeutic tools or processes that facilitate the changes in decisional balance and self-efficacy needed for readiness to manage stuttering.

Motivational interviewing

One useful clinical tool, available now, for exploring readiness for change and supporting therapy compliance is motivational interviewing (Miller & Rollnick, 2012). This is a style of interpersonal communication that therapists can use to help teens to explore what changes they want to make in managing their stuttering, and their readiness to make them.

Motivational interviewing assumes that motivation is a dynamic psychological state that reflects ambivalence toward change. The goal of motivational interviewing, then, is to resolve ambivalence by helping the teen to articulate beliefs about both the benefits and drawbacks of doing something to change stuttering: the behaviours of stuttering or the thoughts and feelings related to stuttering. In speech-language pathology, motivational interviewing has been shown to be helpful in uncovering resistance to change in people with voice disorders (Behrman, 2006).

At the University of Iowa intensive program for teenagers who stutter, we begin with a motivational interview framed by, but not limited to, questions that elicit the teen's desire to make a change, ability to engage in change in difficult situations, and the reasons and need for making changes in what they do about stuttering. Examples include:

> *I am really looking forward to working together with you this week. What would you like to tell me so I can understand what you'd like to learn or change?*
>
> *Is there anything else that you think I need to know? Do you have any questions for me, or is there anything you would like to know about me?*
>
> *Tell me about the therapy you have had for stuttering? What was helpful? What was not helpful?*
>
> *How do you think stuttering has affected your life? How "big a deal" is stuttering? To get started, think of a scale from 1 to 10, where 1 is "it's no big deal and hasn't affected me at all," and 10 is "it affects me every day." If you choose a number above 5, can you give me an example of how it has affected you, or affects you now.*
>
> *Imagine it's the end of the week and you think, "I'm happy that I worked on this because I am doing much better." What would you be doing differently, or what would be changed about yourself?*
>
> *How would other people, for example your friends and family, see that something has changed? What would they notice that was different?*
>
> *I want you to think of another time in your life where you wanted to learn something, or change something. Can you think of an example? What did you do to try to learn or change? What happened?*
>
> *Who are the people you talk to when you need help with something?*
>
> *Let's take a minute and draw your support system. We can work together, or you can work yourself. (The clinician draws a circle with the teen's name in the middle). Draw a line for each person you can think of who you go to for advice or support.*
>
> *Tell me how each of these people helps and supports you.*
>
> *What do your friends and family say that you are good at?*
>
> *What do you think you are good at?*

Once we complete this discussion, we summarise with the teen what has been said to develop a change plan that will serve as the guide for therapy moving forward, both during the intensive program and beyond. This change plan is clearly articulated by using the following organisational structure, as developed by Miller & Rollnick (2002, p. 137) and refined and reported by Behrman (2006, Appendix G) with voice clients:

> *The changes I want to make are …*
>
> *The most important reasons why I want to make these changes are …*
>
> *The steps I plan to take in changing are …*
>
> *The ways other people (clinician, parents, friends, teachers) can help me with my plan are …*
>
> *What I will see and feel if my plan is working …*
>
> *Things that will make it hard for me to follow my plan are …*
>
> *Things I can do if my plan isn't working are …*

Our experience with over 20 years of using motivational interviewing with teenagers who stutter is that these early conversations at the beginning of therapy are critical to the early establishment of a strong collaboration between the teen and therapist. Further, deep listening and problem-solving support from the therapist helps to pave the way for the adolescent to attempt meaningful changes in doing something about stuttering, whether that is behavioral speech modification, attitude change, or an increase in self-advocacy and assertive behavior.

Questions from discussion leaders

It could be difficult to have access to a multidisciplinary team for stuttering intervention. What would you suggest as the minimal standard of care team for adolescents who stutter?

It depends very much on what the adolescent wants. Many teens prefer to work on their own. They emphatically don't want their parents involved. But those who don't want parents or teachers involved can be enthusiastic about working with a group of their peers. If teens are amenable to it, then some parent and teacher involvement can help. But this only works if the teen can make the decisions about who is involved and how much.

From your 20 years experience of motivational interviewing with adolescents, what take-home message would you like students of speech-language pathology to have?

The most important take-away message is that decades of research have shown that therapy compliance and durable change happen when people and their health care providers work together to establish the goals and direction of treatment. This collaborative work begins with the motivational interview.

What do you think is the most important research finding about this topic that speech-language pathologists can apply to clinical practice?

Past reports of successful treatment are important for future clinical practice. Publications by Boberg and Kully (1994) and Carey et al. (2014) strongly suggest that group therapy is powerful for teens, that internet-delivered treatment can be successful for teens, and that fear and avoidance reduction may be important to include in treatment along with techniques for increasing fluency.

Your statement that "the brain's plasticity is almost as great during adolescence as it is from birth to age 3 (Steinberg, 2014)" seems at odds with the fact that stuttering seems a lot less tractable during adolescence than during the pre-school years. Could you comment?

Treatment of stuttering for teens can take advantage of findings about the teenage brain's plasticity by using strong rewards and no punishment. The changes that occur in the brain during adolescence involve not just growth, but reorganisation. Importantly, this reorganisation includes the development of a tighter relationship between arousal, reward seeking behaviour, and self-regulation. So, while teens are more sensitive to and easily aroused by the environment and experience, they are also more consistent and efficient in their ability to "put on the brakes" through increased problem solving and decision making skills.

Simply put, teens are better positioned than younger children to both notice and evaluate their own behaviour and make changes that they view as rewarding. While this scenario would suggest that stuttering treatment for teens would see a greater degree of success,

here the age and chronicity of the stuttering problem and the development of negative cognitive and affective consequences come into play. So, although learning is faster and more durable, it takes more time and effort to make behavioural change. Another explanation for the relatively high rate of relapse and intractable stuttering in teens may be that conventional therapy approaches are a poor fit for many adolescents. Simply put, they are not ready to use what they've got. Research in the second presenter's lab on readiness to change in adolescents who stutter is exploring ways to predict readiness and move teens toward action in doing something about their stuttering. Use of peer groups and helping teens feel that they have some control of the elements of treatment can make a difference. The same is true for the use of motivational interviewing with teens.

It should also be noted that adolescence is a period when sensitivity, self-consciousness, and emotionality are at a peak (Elkind & Bowen, 1979). Thus, stuttering may be most devastating to teens compared to other age groups. Because of their self-consciousness, teens want nothing more than to fit in with peers and not show any differences. Stuttering is an obvious difference that sticks out like a sore thumb or a second head on their body. Because of this, despite the plasticity of the teenage brain, treatment for adolescents will always be challenging. Previous approaches that have used peer group therapy have been successful, probably because of teens' desire to fit in with their peers. However, because teens' self-consciousness is so high, a strong focus must be placed not just on fluency, but also on self-acceptance and desensitisation to fear of stuttering.

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Anxiety and Bullying Management with School Age Children and Adolescents who Stutter

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Abstract

The aim of this presentation is to summarise findings in relationship to peer victimization (bullying), and anxiety in children and young people who stutter, and to consider their practical clinical applications. Children who are bullied are more at risk for experiencing a range of mental health difficulties, which can have long-term consequences. Children and young people who stutter are more at risk of being bullied by their peers and this may contribute to the vulnerability that some adults who stutter have to heightened social anxiety. Speech-language pathologists need to be knowledgeable about the effects of bullying and routinely consider psychological wellbeing and psychological aspects of stuttering as part of holistic assessment and treatment protocols. Therapy approaches which integrate standard fluency techniques with psychological therapies such as cognitive behaviour therapy and which support the development of resilience, is indicated by the research.

Clinical importance of the topic

Bullying, or peer victimisation, refers to repeated aggressive, intentional acts, carried out by one or more persons, with the intent to inflict harm on an individual who is physically or psychologically weaker (Olweus, 1999; Slonje & Smith, 2008). It can be physical, such as hitting or punching, or verbal, such as name-calling or taunting. Bullying can also be, relational which involves undermining the individual's sense of belonging in a group such as by exclusion, rejection, or spreading rumours or stories. Bullying can be direct or face-to-face, or it may be indirect, conducted through a third person or persons. A more recent phenomenon is bullying by digital media, which is known as cyber-bullying (Slonje & Smith, 2008). Bullying is different from teasing which is not intended to cause harm and which occurs between peer equals. However when teasing is, or becomes, repeatedly and intentionally hurtful and when there is an implicit power balance in the relationship or interaction, then it is bullying.

Bullying has been described as one of the most significant problems confronting children and adolescents (Turner, Exum, Brame & Holt, 2013). It is known that children worry about being targeted by bullies (Silverman, Greca, & Wasserstein, 1995) and see it as a significant threat to wellbeing (Goswami, 2012). Prevalence rates vary depending on how bullying is defined and measured (Haynie et al., 2001) however examples of prevalence include 18–20% of children in England (Boulton & Underwood, 1992), 25% of children in Australia (Slee, 1994), and an estimated 20% of children in America (Kaufman et al., 1998).

There is consensus that children and adolescents who are bullied are more at risk than others in the short-term for mental health problems such as depression, anxiety, loneliness and social anxiety (Arseneault et al., 2006; Hawker & Boulton, 2000). This may also translate into vulnerability for anxiety and social anxiety in adulthood (Arseneault, Bowes, & Shakoor, 2010; Boulton, 2013; La Greca & Harrison, 2005).

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It is clinically and theoretically important to understand the experience of children who stutter in respect to bullying and anxiety. Adults who stutter are more at risk for anxiety, and in particular social anxiety (Blumgart, Tran, & Craig, 2010; Craig & Tran, 2014; Iverach, et al., 2009; Iverach & Rapee, 2014; Smith, Iverach, O'Brian, Kefalianos, & Reilly, 2014; Stein, Baird, & Walker, 1996) however the mechanism by which social anxiety develops, the age when it emerges, and the reason why some adults are more at risk than others is not known (Smith, Iverach, O'Brian, Kefalianos, & Reilly, 2014). Developmental models of social anxiety propose that being bullied is one of several contributing factors, together with genetics, temperament, exposure to negative life events, family influences, cognitive styles, and cultural context (Rapee, Schnieiring, & Hudson, 2009; Wong & Rapee, 2016). In line with this, it has been proposed that if children and young people are bullied about their stuttering, this could be a negative social experience that contributes to risk (Blood et al., 2011).

The more that is understood about the pathway by which social anxiety emerges for some individuals who stutter, the more it will be possible to intervene earlier for those who are at risk, and potentially minimise its development (Messenger, Packman, Onslow, Menzies, & O'Brian, 2015; Smith et al., 2014). At a theoretical level, exploring the relationship between bullying, stuttering, and anxiety may enrich our understanding of the phenomenology of stuttering and help to define best practice for children who stutter.

Research findings about the topic

Psychological impact of bullying

Cross-sectional studies have demonstrated that, in the general population of children and adolescents, there is an association between being bullied by peers and experiencing mental health difficulties, including depression, anxiety and social anxiety (Hawker & Boulton, 2000; La Greca & Lopez, 1998; Storch, Brassard, & Masia-Warner, 2003). Longitudinal studies have demonstrated a causative pathway, showing that peer victimisation predicts subsequent increases in anxiety, depression, or social anxiety in children and adolescents (Acquah, Topalli, Wilson, Junttila, & Niemi, 2016; Bond, Carlin, Thomas, Rubin, & Patton, 2001; Siegal et al., 2009; Tillfors, Persson, Willén, & Burk, 2012). This is further supported by retrospective studies showing that social anxiety in adults is associated with recollections of being bullied (Boulton, 2013; Levinson, Langer, & Rodebaugh, 2013; McCabe, Antony, Summerfeldt, Liss, & Swinson 2003).

Relational bullying in childhood has been found to be the strongest predictor of anxiety in adults (Boulton, 2013). Adolescents, and girls in particular, are more likely to experience social anxiety after being subject to relational bullying than overt bullying (Bond et al., 2001; La Greca & Lopez, 1998; Siegel, La Greca, & Harrison, 2009; Storch et al., 2003), and low peer-acceptance and integration has been found to increase vulnerability to being bullied (Hodges & Perry, 1999).

Interpersonal models have led to an interest in whether social anxiety and aspects of peer relationships are bi-directionally related (Tilfors et al., 2012) and there is increasing evidence that this is the case. Being bullied not only predicts social anxiety, but appearing socially withdrawn or having less effective social skills is likely to elicit bullying. This may heighten social anxiety, affect interpersonal behaviour, and increase the likelihood of being targeted further (Tilfors et al., 2012). The way that individuals respond to being victimised may also mediate the outcome of bullying (Fox & Boulton, 2005; Levinson et al., 2013). Graham and Juvonen (1998) found that children who blamed themselves for being bullied, attributing it

to there being something inherently wrong with them, had significantly higher social anxiety than those who did not make this attribution. Behaviourally, higher reactivity to instances of bullying may serve to "reward" the perpetrator and increase further bullying, whereas an appearance of not caring may be a disincentive (Levinson et al., 2013).

An alternative perspective to exploring risk is to consider what makes some young people more resilient to the experience of being bullied. Resilience, or the ability to cope with adverse life events, is thought to be underpinned by environmental factors as well as individual coping styles. Environmental factors include warm and supportive family relationships and a positive atmosphere at home, the opportunity to experience success in other domains of life, the opportunity to cope with stressors successfully (Bowes, Maughan, Caspi, Moffitt, & Arseneault, 2010; Rutter, 2013), and having pro-social friends (Ttofi, Bowes, Farrington, & Lösel, 2014). Intrinsic skills include the ability to plan and self-reflect, having effective social skills and friendships, and having a flexible, optimistic attributional style (Yeager & Dweck, 2012). The more that is understood about the role of resilience, the more professionals supporting children who stutter may be able to work with them, their families and their teachers in a way that builds this capacity to cope.

Stuttering and bullying

Children have been found to exhibit negative attitudes to or reactions to stuttering from a young age (Ezrati-Vinacour, Platzky, & Yairi, 2001; Langevin, Packman, & Onslow, 2009). Parents of preschool children who stutter report that teasing and mimicking occurs (Langevin, Packman, & Onslow, 2010) and while preschool children's reactions to stuttering may be neutral or positive, negative reactions may also be encountered, particularly when stuttering is more severe (Langevin et al., 2009). In a recent study Weidner, St. Louis, Burgess, and LeMasters (2015) found that while showing evidence of "pre-prejudice" towards stuttering itself, preschool and kindergarten children expressed positive perceptions of the person who stutters together with strong convictions that it would be wrong to laugh at a child who stutters.

In the school age population Davis, Howell & Cooke (2002) found a trend for children who stutter to be described by peers as withdrawn and a "bully-victim," to be viewed as having lower social status, and to be socially rejected. Children who stutter have been described as increasingly less "likely to fit in" and more "likely to be teased" the more severe their stuttering (Evans, Healey, Kawai, & Rowland, 2008) and more likely to receive negative comments the more severe their stuttering (Panico, Healey, & Knopik, 2015). However, Evans et al. (2008) found that teasing was considered less likely when the speaker showed the most severe stuttering, suggesting a degree of diversity awareness, while Panico et al. (2015) found that severity of stuttering did not affect children's willingness to include a child who stutters in their friendship group. Further, two studies of adolescents who stutter have found that, as a group, they did not feel that their stuttering affected the degree to which people liked them or wished to socialise with them (Blood, Blood, Tellis, & Gabel, 2003; Erickson & Block, 2013). It is possible that school age children, in particular, may encounter varying degrees of peer acceptance and responses to stuttering, which in itself may create a challenge that social situations are uncertain and ambiguous.

In terms of prevalence of bullying, Hugh-Jones and Smith (1999) found that 83% of adults who stutter recalled being bullied at school, however researchers have since attempted to investigate the experience of children and young people more directly. Blood and Blood (2007) found that 61% of school age children who stutter reported being bullied, compared

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with 22% of their fluent peers. Being bullied also emerged as a predominant subtheme in a qualitative study of the experience of school age children who stutter (Lau, Beilby, Byrnes, & Hennessey, 2012), and dealing with teasing emerged as a basic theme in a study exploring children's hopes for therapy (Berquez, Hertsberg, Hollister, Zebrowski, & Millard, 2015). Blood and Blood (2004) found that 43% of adolescents who stutter reported being bullied in the previous week compared to 11% of their fluent peers, and Blood et al. (2011) found that 44% of adolescents met the criteria for "clinically significant bullying" compared to 14% of adolescents who did not stutter. Erickson and Block (2013) found that 53% of their participants had experienced bullying, most often verbal bullying, and most typically during late school age or early adolescent years. In contrast Hearne, Packman, Onslow, and Quine, (2008) found that 15% of adolescents reported being teased or bullied about their speech. However, as a rule there is robust evidence that children and adolescents who stutter are more at risk of being bullied and also that the experience is linked with anxiety (Blood & Blood, 2007), low self-esteem, less optimistic life orientation and lower overall life satisfaction (Blood et al., 2011), and sadness or depression (Erickson & Block, 2013).

Within this context, Blood, Boyle, Blood and Nalesnik (2010) found that speech-language pathologists rated verbal and relational bullying as less serious and less in need of intervention than physical bullying. Given that verbal and relational bullying may be the two forms of victimisation that children and adolescents who stutter are most at risk for experiencing, this suggests a need for greater awareness about bullying within the profession, coupled with a more proactive response when bullying in any form is disclosed.

Stuttering and anxiety

Preschool children who stutter do not differ from fluent controls in measures of temperament typically associated with anxiety (Alm, 2014; Kefalianos, Onslow, Ukoumunne, Block, & Reilly, 2014) which supports the view that anxiety develops as a consequence of stuttering rather than having a causative role (Kefalianos, Onslow, Block, Menzies, & Reilly, 2012; Smith et al., 2014). Investigations into anxiety within school age and adolescent populations have yielded contradictory findings. Some studies have not found significant differences between children or adolescents who stutter and controls or population norms (Craig & Hancock, 1996). Others have reported significantly higher scores on anxiety measures (Blood & Blood, 2007; McAllister, Kelman, & Millard, 2015; Mulcahy, Hennessey, Beilby, & Byrnes, 2008), but without these necessarily exceeding the normal range (Blood, Blood, Maloney, Meyer, & Qualls, 2007). There has been a shift towards using more sensitive measures that enable specific domains of anxiety, such as social anxiety, to be investigated more precisely. However, Gunn et al. (2014) and Messenger et al. (2015), using the Revised Children's Manifest Anxiety Scale, found that all subscale scores for their cohorts of 12–17 year olds and 6–18 year olds, respectively, were also within the normal range. In their review, Smith et al. (2014) found that approximately half of studies into anxiety in children and adolescents who stutter reported significant findings and half did not.

There is a tendency for adolescents who stutter to score more highly on measures of anxiety than younger children, and for children and adolescents who stutter to score more highly than controls, although this may be at a subclinical level (Messenger et al., 2015). Additionally, adolescents with concomitant speech and language disorders seem to score more highly for anxiety than controls (Blood et al., 2007), and for other domains of anxiety, such as generalised anxiety, also to be elevated for some individuals (Gunn et al., 2014; McAllister et al., 2015). There is not necessarily a correlation between psychological test

scores and severity of overt features of stuttering, and stuttering severity should not be taken as an indicator of the psychological impact of stuttering on the individual (Gunn et al., 2014). Results may be affected by children and young people having poor insight into their emotions (McAllister et al., 2015), or under-reporting problems due to reluctance to disclose (Messenger et al., 2015), which means that it is essential to obtain obtaining supporting information from parents (Gunn et al., 2014; McAllister et al. 2015). The relationship between stuttering and anxiety in children and adolescents has been described as complex (Iverach et al., 2009), the research findings as inconclusive, inconsistent, and equivocal (Smith et al., 2014), with a need for more research addressing the methodological limitations of previous studies (Gunn et al., 2014; McAllister et al., 2015; Smith et al., 2014).

Methodological discussion points have included the need to use age appropriate and sufficiently sensitive measures, such as childhood measures of social anxiety, and to recruit larger cohorts (Smith et al., 2014). Addressing these concerns, Iverach et al. (2016) compared 75 children who stuttered aged 7–12 with matched controls, using social anxiety measures complemented by a structured diagnostic interview and parental report. While scores on symptom measures fell within the normal range, diagnostic assessment using a structured clinical interview found that the cohort of children who stuttered had a six-fold increased odds for social anxiety along with seven-fold increased odds for subclinical generalised anxiety disorder. They also had significantly higher self-reported bullying related to "language," which the authors suggested refers to stuttering.

Clinical application of research findings

The increased risk for children and young people who stutter of being teased or bullied and of experiencing heightened anxiety has implications for how speech-language pathologists approach assessment and treatment, with a holistic approach to both being required (Millard, 2011; Yaruss & Quesal, 2006). Cognitive, affective, behavioural and interpersonal features of stuttering, as well as its surface features, need to be explored and addressed, and the individual's experience within the context of family, peer and school environments understood. Assessments such as the OASES battery (Yaruss & Quesal, 2010), psychological screening or assessment tools such as those for anxiety and social anxiety, and structured interviews, together help clinicians understand the unique and potentially complex experience of stuttering for the individual.

Bullying takes many forms and clinicians should ask about all of these, including relational bullying or social exclusion (Juvonen & Gross, 2008; Siegel et al., 2009). Speech-language pathologists need to enquire further when bullying is reported by children or their parents, in order to understand how often bullying occurs, how transient or chronic it is (Arseneault et al., 2010), how the individual copes, what the contributing risk factors are, and what protective factors are present. Speech-language pathologists also need to be alert to parents or teachers reporting changes in behaviour which might be indicative of a child being bullied, such as sleeping badly, becoming uncharacteristically aggressive, complaining of headaches or stomach aches, becoming withdrawn or talking about feelings of helplessness, as well as exhibiting obvious signs of physical bullying.

Given that being bullied is defined in part by an imbalance of power in peer interactions, it follows that therapy should focus on empowering individuals and increasing their sense of agency or self-efficacy when bullying is identified. Langevin (2000) and Murphy, Quesal, Reardon-Reeves, & Yaruss (2013) outline a range of coping strategies which are aimed at

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empowering or "bully-proofing" children. Children may be taught to give neutral or matter-of-fact responses, for example calmly saying "that's right, I stutter," as these are less likely to elicit further bullying than more reactive or retaliatory responses (Levinson et al., 2013; Siegel et al., 2009). Teaching problem-solving skills also empowers children by enabling them to clarify the problem, consider possible ways of responding, and to weigh up the advantages and disadvantages of each before selecting and trying out those that are most likely to be effective. Being supported in informing others such as class peers about stuttering—for example by giving a presentation—may also serve to empower children and young people who stutter.

Lau et al. (2012) concluded that children and young people want to be involved in deciding how bullying is managed, and Rutter (2013) warns that they can easily become disempowered when professionals "drive the process." Children, young people and their parents need the opportunity to try out their own ideas, drawing on their own skills and resources as well as their experience of what has worked in the past (Rutter, 2013). This requires speech-language pathologists to adopt a collaborative, strengths-based therapeutic style, and this is seen by many as being fundamental to the process of nurturing resilience and enabling individuals who stutter to communicate confidently (Fry, Millard, & Botterill, 2014; Millard, 2011).

Given that friendships, peer support and pro-social behaviour are protective and may reduce the likelihood of being bullied in the first place, and that more socially withdrawn or timid behaviour may increase the risk of being bullied, then including social skills training in therapy programmes, and creating opportunities for these skills to be developed in the context of group therapy, is critical (Siegel et al., 2009). Caughter and Dunsmuir (2017) found that being able to practise skills within the safety of the group was a factor contributing to change and increased resilience in a group of 10–14 year old children who stutter.

Cognitive behaviour therapy (CBT) is effective in reducing anxiety in adults who stutter (Menzies et al., 2008) as well as in fluent children with anxiety disorders (Barrett, Duffy, Dadds, & Rapee, 2001) and it should be integrated into therapy with children and young people who stutter when heightened anxiety is identified (Craig & Tran, 2014). CBT or other psychological approaches which strengthen psychological flexibility and resilience are particularly important when individuals have negative or self-blaming core beliefs, when they may have a bias towards misinterpreting ambiguous social cues, and when the nurturing of a more resilient and optimistic "mindset" is desirable (Yeager & Dwek, 2012).

Given the integral role of the family in building resilience and self-esteem and buffering children from stressors (Rutter, 2013), it follows that, where developmentally appropriate, it is crucial to involve families fully in therapy (Millard, 2011). Intensive, group therapy programmes for school age children and adolescents in which standard fluency management skills are integrated with social skills training and CBT in the context of family involvement are described by Fry et al., (2014), Millard (2011) and Caughter and Dunsmuir (2017). Individual family-based treatment incorporating CBT is described by Kelman and Wheeler (2015).

Finally, within a broader social context, there is an ongoing need for education and awareness-raising about stuttering in order to promote positive peer attitudes towards children who stutter (Weidner et al., 2015) and there are comprehensive published resources

such as the Teasing and Bullying Programme (Langevin, 2000) as well as online resources which are available to support this.

Summary and conclusions

Bullying is no longer viewed as an inevitable part of growing up and it is not, to borrow from Shakespeare, "much ado about nothing". There is considerable evidence that being victimised during childhood has significant psychosocial implications, and that along with other factors contributes to the risk for developing anxiety. Children and young people are at greater risk of being bullied, however more research is needed to explore the dynamics between victimisation and anxiety for children and young people who stutter, as well as the role of protective factors such as resilience. The onus is on clinicians to respond in a knowledgeable and proactive way when issues related to anxiety or bullying emerge for any individual, particularly when this occurs at critical developmental stages. The more that is understood about the role of protective factors and what enhances individual resilience, the more these can be routinely incorporated into therapy approaches, according to what each individual needs, in order to minimise future risk. It is increasingly evident from the research that these are the keys to minimising that risk: integrating psychological, social skills and fluency management therapy approaches, involving families in therapy, working collaboratively with schools to raise awareness about stuttering, working against negative stereotypes, increasing diversity awareness, together with a focus on strengths and empowerment.

Questions from discussion leaders

Although you outlined some differences between them, are teasing and bullying generally interchangeable terms?

Bullying is clearly defined but "teasing" is an ambiguous term. It describes interactions which are intended to be good-natured and are experienced as such by the recipient or which help in resolving conflicts. However it is also used to describe interactions which are intended to be, or are experienced as hurtful by the recipient and which could otherwise be referred to as verbal bullying. This ambiguity can trivialise the victim's experience and shield perpetrators when challenged: "It was only teasing."

Murphy et al., (2013) support drawing a clearer line between teasing and bullying, proposing that when teasing is experienced negatively it is "inappropriate teasing," or bullying. Ultimately what matters clinically is that when clients refer to being teased or bullied we explore the context in order to give appropriate support. Aspects of the context of teasing or bullying include the relationship in which it occurs, its function, intent, and its impact on the individual.

How can bullying and anxiety in children who stutter be dealt with in countries where parents are hesitant or unlikely to seek help?

We need to understand the barriers that confront parents in many contexts. If parents feel intimidated or unsure that their views will be respected, services need to be promoted in a culturally sensitive, respectful and jargon-free way. Word-of-mouth within a community, straightforward referral processes, and the use of professional interpreters may increase accessibility. Utilising a presence in schools to meet parents informally or offering parent workshops may also help.

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When parents cannot be involved, curriculum-based programs that target bullying and promote emotional well-being may be sufficient to meet a child's needs, with school-based counselling as a further support. Individual or group therapy within schools may enable young people to cope with anxiety, strengthen social networks, and to cope better with bullying. Speech-language pathologists can advocate for the child and liaise with other professionals so that bullying is addressed.

Obviously, there is much basic research about the effects of bullying on school age children and adolescents. What then is the take-home message for speech-language pathologists about how to apply that research to resolving any problems from bullying?

Firstly, explore young peoples' experience holistically. Routinely ask questions about cognitive, affective and interpersonal aspects, and support this with information from parents. Use screening tools of anxiety or emotional wellbeing to develop a richer clinical impression, and follow through when needed. Work actively with school staff and other professionals who are involved with the child, and remember that while some children will benefit from referral for support with emotional wellbeing, speech-language pathologists can effectively support many children affected by bullying. An important research finding is that having effective social skills and being socially well-integrated helps to protect young people from being targeted by bullies and also helps them respond more effectively if they are targeted. Social communication skills training with young people who stutter, and with their parents, is therefore an important part of therapy.

You mentioned CBT as part of a clinical response to bullying. Is that a realistic option for speech-language pathologists to administer?

When young people have emotional health problems such as anxiety or depression, they should be under the care of a mental health professional, particularly if issues extend beyond stuttering. However, many young people who stutter may benefit from CBT whether or not they have clinical anxiety, and in some countries speech-language pathologists are trained to use CBT during professional preparation, although recognising the limits of their skills.

Formal CBT training for speech-language pathologists requires a commitment that may not be realistic, and there are other counselling approaches that speech-language pathologists may use. There are options for developing psychological skills and because stuttering is "not just a speech disorder" this is essential for those specialising in the field. The development of on-line computerised CBT for adults and adolescents who stutter will resolve the issue for many, however speech-language pathologists also have a unique opportunity to blend psychological skills with an in-depth understanding of stuttering.

Could you elaborate on the concept of "resilience" and how it can be established?

Psychological resilience means being able to respond positively, emotionally, and behaviourally to adversity or challenges. It is a capacity that can be learned and nurtured.

Resilience in children is fostered by them having positive family and peer relationships, a positive self-view, the capacity to plan how to approach situations and to regulate emotions, and an optimistic mindset. Resilience is also fostered by experiencing challenges and discovering that they can be coped with.

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Speech-language pathologists can support resilience by including social communication skills and self-esteem training in therapy, and by working in groups to build support networks and working with parents. The use of praise, confidence-building and problem-solving are often included in social skills programs. Flexible perspective taking and helpful or kind self-talk are also introduced by many speech-language pathologists. Importantly, helping children approach and deal with challenges skilfully helps to build their resilience.

We would like to follow up on your response to the last question where you endorsed resilience training for adolescents. Could you outline the empirical support for that approach? What clinical trials have been done?

The effectiveness of resilience-focussed programs amongst clinical and non-clinical populations is a burgeoning field of research, driven by evidence that psychological resilience is associated with emotional wellbeing. Methodological challenges such as variations in the way that the construct of resilience is operationalised, and the heterogeneity of training programs are discussed in the literature. However, resilience interventions are grounded in CBT, which has strong randomised controlled trial evidence.

Two systematic reviews of the efficacy of resiliency training in adults by Leppin et al. (2014) and Macedo et al. (2014) have concluded that there is a degree of evidence of effect but that more research is needed and training protocols need to be refined. Research into the impact of resiliency training in children and adolescents is in its infancy and consists primarily of pilot studies. However this is a rapidly developing field within CBT.

In the field of stuttering, the role of resilience has been discussed in relation to adults who stutter (Craig, Blumgart, & Tran, 2011) and school age children who stutter (Caughter & Dunsmuir, 2017), with the authors of both of those studies concluding that the construct of resilience has utility.

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Speech Treatments for Adults who Stutter

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Abstract

Although it obviously is not the only goal of treatment, adults who stutter frequently seek treatment to reduce or modify their stuttering. For those requiring behavioural control of stuttering, there are clinically useful recent empirical advances with techniques, programs and delivery models. Recent reviews have confirmed that speech restructuring remains not only the most researched but the most efficacious treatment for reducing stuttering. Positive outcomes can be achieved regardless of whether the treatment is offered in individual, group, weekly, or intensive formats. Advances in technology have also meant that similar outcomes can be achieved with webcam service delivery, with computer-driven feedback about speech production and, to a limited degree, with clinician-free Internet programs. Effective programs exist for use by generalist clinicians with the treatment guide and resource materials available on the Internet. This presentation discusses the research evidence for and clinical application of these various treatments and delivery formats for adults who stutter. It concludes with a discussion of how to potentially make treatment even more convenient for our clients and less costly to health care systems.

Clinical importance of the topic

The stuttering of adults and the consequences of the disorder are obviously different from stuttering in young children. First, the disorder in adults is unlikely to go away naturally. It is more resistant to treatment in the sense that the speech behaviours become more difficult to reduce. Stuttering reductions, when achieved, often require constant vigilance and effort to maintain over time and hence relapse following treatment is a common problem (Andrews, Guitar, & Howie, 1980; Craig & Hancock, 1995; Martin, 1981). Consequently, many adults seek treatment a number of times throughout their lives (Yaruss, Quesal, & Murphy, 2002; Yaruss et al., 2002; Venkatagiri, 2009). This may be to try to regain previous benefits or sometimes to try a different approach, perhaps because of disillusionment with prior treatment or maintenance. Then there are the psychological, social, educational, occupational and financial issues that are related to living with a frustrating and unpredictable disorder for many years (Blumgart, Tran, & Craig 2010; Klein & Hood 2004; O'Brian, Jones, Packman, Menzies, & Onslow, 2011; Tran, Blumgart, & Craig, 2011). And finally, there are the purely practical issues for adults trying to negotiate time off work to attend treatment and to find time within busy schedules to practice the skills taught in treatment. Thinking about these points, it isn't hard to imagine that motivation to persevere with treatment and general disillusionment with available treatments may well be issues for many adults who stutter.

When adults present to a clinic for assessment, they may do so for a number of different reasons (Venkatagiri, 2009). Clearly some want to learn skills that can be used to reduce or modify their stuttering behaviours. They may want to use these skills to control stuttering all the time or perhaps just have access to skills they can use in specific or important situations. Others may wish to address the anxiety associated with speaking. This could be at the same time as or instead of reducing stuttering. Other clients may seek assistance to become more accepting of themselves as someone who stutters. And finally, there are those who remain

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disillusioned with all traditional therapy—or more likely the effort it requires—and are hopeful of just finding something new and better.

In serving client needs, it is important for the speech-language pathologist to provide those who stutter with up-to-date information about various forms of treatment and to help them to decide what it is they want and need from treatment. Then they can provide the most appropriate treatment or combination of treatments to meet those needs. At best, a "one size fits all" approach is not going to be very satisfactory for many clients, and clinicians need to be able to either offer different approaches, or know where to refer clients for different approaches.

This presentation focuses on the evidence and issues relating to behavioural control of stuttering. This is important because according to at least two sources of evidence (Venkatagiri, 2009; Yaruss et al., 2002), more than half of our clients do seek this type of treatment. However, it should be read in conjunction with other papers in this edition that focus on how adult stuttering clients process information when speaking and on psychological issues, anxiety in particular, which are associated with stuttering. Again, there is no intention here to suggest that behavioural control of stuttering is the most important or most relevant goal of treatment, merely one option for part of treatment.

Research findings about the topic

There have been several comprehensive reviews of adult stuttering treatments during the last few years (Baxter et al., 2015; Bothe, Davidow, Bramlett, Franic, & Ingham, 2006; Bothe, Davidow, Bramlett, & Ingham, 2006; Onslow, Jones, O'Brian, Menzies, & Packman, 2008). All have confirmed that speech restructuring remains not only the most researched but the most efficacious treatment for reducing stuttering. The term speech restructuring simply covers the range of treatments that directly target stuttering reduction by teaching the client a new way of speaking. Examples would include prolonged speech, smooth speech, easy speech and fluency shaping. Most programs begin by teaching the client an unnatural-sounding speech technique to control stuttering. This speech is then shaped into more natural-sounding speech while still controlling stuttering.

Despite most of the evidence favouring speech restructuring treatments, there is limited evidence for some other treatment approaches as well. In evaluating this literature, the clinical trial remains the most fundamental and useful means of interpreting treatment outcomes. Therefore, the Onslow, Jones, O'Brian, Menzies, & Packman (2008) definition of a clinical trial is used: (1) a prospective attempt to investigate (2) an entire treatment with (3) at least one pre-treatment and one post-treatment outcome after at least 3 months, and (4) where outcomes involve assessments in everyday environments independent of treatment. A discussion follows of those treatments for which, according to this definition, there is evidence available.

Time-out

In simple terms, time-out involves the speaker pausing briefly after a moment of stuttering. This pause can be self-imposed or can occur after clinician feedback. What is surprising is that, despite many laboratory studies investigating various aspects of time-out (see Hewat, Onslow, Packman, & O'Brian, 2006, for a review), there have been few clinical trials using this procedure with adults who stutter. Two of these reports (Hewat, O'Brian, Onslow, & Packman, 2001; James, 1981) involved only one participant each. The third report (Hewat et

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al., 2006) was a phase II trial with 30 participants. The 22 participants who completed the first two stages of the program showed a wide range of responsiveness from virtually no response to more than 90% stuttering reduction. This and other studies have indicated that time-out is likely to be more effective with more severe stuttering and with those clients who have had previous speech restructuring treatment (James, Ricciardelli, Rogers, & Hunter, 1989; Franklin, Taylor, Hennessey, & Beilby, 2008). Presumably this could be because the time-out allows the client to access previously learned strategies for reducing stuttering. Interestingly, neither the frequency nor the duration of the time-out pause appears to affect outcomes (James, 1976).

Self-modelling

Another technique with limited evidence is self-modelling. This simply involves clients listening to, or better still watching, recordings made of themselves when speaking without stuttering. With adults, there has been one laboratory study of self modelling used alone (Webber, Packman, & Onslow, 2004) and two clinical trials (Cream et al., 2010; Cream, O'Brian, Onslow, Packman & Menzies, 2009), one a randomised controlled trial of self modelling used in conjunction with speech restructuring. Results suggest that the procedure is best used in combination with speech restructuring treatment, particularly when used to deal with relapse.

Machine-driven treatments

The most researched of the machine-driven treatments is Altered Auditory Feedback. This term refers to treatments or devices that change the speech signal, typically in pitch and/or timing, so that clients hear their voices differently. There has been much laboratory research done with such treatments (see Finn, Bothe & Bramlett, 2005, for a review). The most common methods investigated recently are delayed auditory feedback and frequency altered feedback, with the SpeechEasy device being the most studied. Until recently, most of the published evidence came from laboratory studies rather than treatment trials. However, the authors of the only clinical trial, conducted by researchers independent of the treatment, concluded that although there was significant individual variation, their "sample did not benefit significantly from the device" (p. 525) (Pollard, Ellis, Finan, & Ramig, 2009). It seems that wearing these devices can certainly reduce stuttering for some speakers, at least initially. But what still appears to be lacking is clinical trial evidence with significant numbers showing long, or even medium term benefits, and evidence that gains can be transferred to everyday speaking environments.

The other machine-driven approach that has been investigated recently is the Modifying Phonation Intervals treatment program. This is a computer program which, in contrast to speech restructuring techniques, gives feedback to clients with the aim of reducing the number of short phonation intervals during their connected speech. Clinical trial evidence is still unavailable but data based case studies suggest that it has the potential to be an efficacious treatment (Ingham, Ingham, Bothe, Wang, & Kilgo, 2015; Ingham et al., 2001).

Speech restructuring treatments

As noted previously, speech restructuring treatments have the most evidence for their efficacy with positive outcomes confirmed over many years. However, three speech restructuring programs for adults stand out for their rigorous clinical trial evaluation this century. These are the Comprehensive Stuttering Program at the Institute for Stuttering Treatment and Research in Canada (Langevin, Kully, Teshima, Hagler, & Narasimha, 2010;

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Langevin et al., 2006; Langevin & Kully, 2012), the Smooth Speech Program at La Trobe University, Melbourne, Australia (Block, Onslow, Packman & Dacakis, 2006; Block, Onslow, Packman, Gray & Dacakis, 2005) and the Camperdown Program developed at the Australian Stuttering Research Centre at The University of Sydney (Carey et al., 2010; Cocomazzo et al., 2012; Erickson et al., 2012; 2016; O'Brian, Cream, Onslow, & Packman, 2001; O'Brian, Onslow, Cream, & Packman, 2003; O'Brian, Packman, & Onslow, 2008). Outcomes from clinical trials of these programs have all shown stuttering reductions in everyday environments, in some cases for up to 5 years post-treatment, with reasonable speech naturalness and client speech satisfaction. However, there were issues with maintaining speech outcomes for all three programs.

So, if the evidence shows that speech restructuring programs generally have the best outcomes, what are some of the issues worthy of further discussion? Both the Comprehensive Stuttering Program and the Smooth Speech program are routinely run as multi-day group intensive programs requiring many clinicians and 90 and 59 hours of treatment per client, respectively. The Camperdown Program, by contrast, can be implemented by a single generalist clinician during weekly clinic visits with a mean of 20 hours of treatment. So is there any evidence to support intensive over weekly programs, and is more necessarily better? Unfortunately, there is limited direct evidence to answer these questions.

A study compared speech restructuring treatment over four consecutive 8-hour days with treatment over two 2-hour sessions for 8 weeks (James, Ricciardelli, Rogers, & Hunter, 1989). They found no difference between groups for stuttering frequency, efficiency or compliance. One other study (Carey et al., 2010) compared a combined one-day intensive and weekly appointments with just weekly appointments, and again found no difference in stuttering outcomes. So intensive compared to weekly possibly makes no difference. As for whether more is better, there is no empirical evidence that has answered this question. A meta-analysis of treatments conducted back in the 1980s (Andrews et al., 1980) suggested that the more hours in treatment the better the outcome, however this analysis was across different treatments with an average of 80 hours.

Speech restructuring treatments and technology

So how have advances in technology, in particular the Internet, influenced speech restructuring treatments? Undoubtedly, the biggest benefit of technology has been the increased access to quality treatment for all clients, regardless of distance or lifestyle factors. Instead of clients needing to attend a clinic, treatment can now be taken to the client. The first reports of telepractice for adult stuttering were around the turn of the century with reports of the telephone for remotely located clients to facilitate maintenance and follow-up sessions (Kully, 2002). However, there was no clinical trial of a telepractice procedure for adult stuttering clients until some years later for the Camperdown Program presented by telephone (O'Brian et al. 2008). Despite being published only 9 years ago, this would now be considered very low-tech by today's standards. In this trial, the clinician never met any of the clients face-to-face. The positive outcomes of this phase I trial—around 80% reduction of stuttering—at least confirmed the viability of such a delivery format. However, there were two especially interesting findings from this study.

First there were no drop-outs from the trial which is rare for any clinical trial. Admittedly, study numbers were quite small but the authors noted that telephone treatment made it easier for clients to comply with treatment demands when their circumstances changed. For

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example, moving cities, changing jobs or even illness, all of which typically have an effect on compliance for in-clinic treatment, were accommodated with this delivery format.

The second interesting finding was that the outcomes suggested that telepractice delivery may in fact be more efficient than in-clinic delivery, with a mean of only eight contact hours to reach the maintenance stage of the program. The authors commented that this was due to much of the speech practice, typically done in the clinic, in fact being done at home. But it also stands to reason that treatment conducted outside the clinic may well generalise better or faster to everyday situations.

A follow-up randomised controlled trial compared the telephone delivery service with in-clinic delivery and with significantly greater numbers (Carey et al., 2010). This trial showed that there were no significant differences in stuttering outcomes between the two delivery models in the short or long term. This direct comparison also confirmed that telephone delivery was in fact more efficient than in-clinic delivery; it required on average close to 3 hours less clinician time than did in-clinic delivery.

The other significant finding was about convenience. As this was a randomised clinical trial, all the participants needed to live locally in case they were allocated to the in-clinic group. Regardless, the telepractice group attained significantly higher convenience scores compared to the in-clinic group, especially in relation to time savings associated with needing to travel to treatment. This finding was not compromised by poorer clinician rapport or satisfaction with treatment, which were equivalent for both groups. In other words, telepractice was confirmed as not just a delivery model to effectively deal with remote client location, but also as a preferred method in terms of convenience for local clients.

These are the results for low-tech methodology but what about high-tech methods? The Internet has now been around for more than 40 years and, according to the Australian Bureau of Statistics, by the end of 2015 the number of Australian households connected to the Internet was 86%. This is probably reasonably representative of developed countries and the percentage is likely to be substantially more by now.

Webcam technology would appear to have advantages over telephone delivery. The addition of real-time visual images of the clinician and client may assist in the development of clinician-client rapport or provide more opportunity for real-time demonstration and evaluation of specific skills. And yet, despite clinical trials of webcam treatment with adolescents and children, there have been no clinical trials of webcam treatment for any treatment for adult stuttering clients.

Finally, we tend to think of telepractice as provision of treatment or assessment by a clinician at a distance, but is the clinician really necessary? The term eHealth has recently been coined to include self-managed computer-driven interventions such as standalone—clinician free—Internet treatment programs. What could be the advantage of a clinician-free program? Such programs have all the convenience advantages of telepractice, plus they require no clinical infrastructure and hence are cost free. Better still, clients can access their treatment at any hour of the day or night to suit their needs, and when using these programs clients are routinely accessing the specialist clinical skills of the developers.

There have been two reports of a clinician-free Internet speech restructuring program, loosely based on the Camperdown Program (Erickson et al., 2012; 2016). Participants in

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these trials progressed through the program over the Internet, at their own pace, in their own time, in their chosen environments. There was no clinician support available. The first study was a pilot with just two participants designed to assess the safety and viability of such a program. Both participants completed the program entirely on their own, one in 4 weeks and the other in 6 weeks. Both participants achieved around a 60% stuttering reduction.

After modifications to the program, a second study was conducted (Erickson et al., 2016) involving 20 participants. Five of these participants completed the entire program, taking between 8–24 weeks, with four of them reducing stuttering frequency by more than 50%. Another five of the participants completed more than half the program, with two achieving similar reductions to those who completed the program. Two other participants reduced their stuttering severity by more than 80%. In general, the participants who accessed more of the program had better outcomes.

Feedback from participants about the program was interesting. Most of them agreed that it was beneficial, however, and perhaps this is unsurprising, only three of the participants admitted to actually using the program in the manner they were supposed to, even those who accessed all of the program. Several of the participants reported forgetting to log on, so later recruits were given email reminders that appeared to help. Some, again unsurprisingly, also cited motivation as an issue.

In sum, more than a third of the participants across the two Internet studies achieved a stuttering reduction greater than 50%. This was achieved with zero clinician hours of input, without participants needing to take time off work or travel to a clinic, and they could have continued access to their therapy at any time. Perhaps time and further studies will help to decide whether a reduction of 50% is an acceptable or a poor result given the circumstances.

Drawing all this information together, positive outcomes seem to be achievable with speech restructuring techniques regardless of whether the treatment is implemented daily or weekly, in groups or individually. Advances in technology have also meant that similar outcomes can be achieved with telephone service delivery and to a limited extent with standalone Internet programs. Limitations to all of this research, however, include: lack of long-term outcomes for most programs, small numbers in many studies, no trials conducted against a no-treatment control group, lack of knowledge about which clients might benefit most from different treatment methods or delivery formats, and no research about the economic health benefits of most interventions. Nor have the legal or ethical considerations associated with eHealth delivery of stuttering treatments been seriously considered.

Clinical application of the research findings

Time-out

The limited studies show that time-out has potential for use as a treatment agent with some adults. To assist generalisation and maintenance, the time-out obviously needs to be self-imposed, not just in response to clinician feedback. Individual responsiveness to time-out can be determined quickly and easily in the clinic by taking a baseline stuttering rating without using time-out, and then introducing clinician imposed time-out for a short period (Franklin et al., 2008). If stuttering reduces, the treatment can be simple, and easily implemented by generalist clinicians. It doesn't require the use of an unnatural speech pattern or ongoing vigilance over speech technique by the client. Additionally, it is cost

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effective because it requires few treatment hours and no equipment. There is a description of how to conduct this treatment in the Hewat et al. (2006) paper.

Self-modelling

This is a simple, cheap and time efficient procedure that can be effective for some clients without the need for clinician support or oversight. With improvements in technology, it is now quick and easy to make high quality recordings of clients for self-modelling use. For example, you can make a video of the client speaking and then edit out all the stuttered utterances so that the client only sees a video without stuttering. Another way to achieve this result, as with the Cream et al. (2009) study, is to record the client using the speech restructuring technique in a natural manner to control stuttering. A third way might be to rehearse questions and answers until the client is stutter-free and then record these productions. This method was used in the Webber et al. (2004) study. Regardless of the method, the client is then encouraged to watch these stutter-free videos daily and to try to speak like the model. The evidence suggests that as little as 1 hour of clinical time may be needed to assist relapse prevention (Cream et al., 2009).

Machine-driven treatments

Given that specialist training is needed to accurately fit and calibrate the Speech Easy device, that it is costly to purchase and the one clinical trial outcome is poor, it is difficult to recommend it for use by generalist clinicians. The Modifying Phonation Intervals treatment is not supported by clinical trials, and has another significant limitation of limited availability. The software and hardware are only available for use by specially trained clinicians, although a smartphone application is available.

A stepped care approach

For many years, treatment of adult stuttering has remained predominantly in the domain of specialist stuttering clinics, typically involving many hours of intensive treatment, multiple clinicians and significant infrastructure. While there is no intention to undermine the powerful effects of such treatments, there will never be enough such clinics to service the stuttering population in a timely, economical, and widely accessible manner. Nor is it possible for many clients to spend 3 weeks in treatment due to family, work or study commitments. So it is imperative for generalist clinicians to fill this gap. Most generalist clinicians have neither the time, the resources, nor the training to conduct multi-day programs.

At present only one of the three evidence-based programs discussed earlier offers a freely accessible treatment guide and resource materials that are readily downloadable on the Internet for generalist clinicians to use without training. That treatment is the Camperdown Program. The downloadable treatment guide (O'Brian et al., 2017) has been developed specifically for generalist clinicians to conduct the treatment one-on-one with a client during weekly clinic visits or by telepractice. While this program can go some way towards addressing the shortfall of specialist clinicians, and telepractice delivery can deal with many of the access issues, can research findings be used to make treatment even more convenient for our clients and less costly to health care systems without sacrificing outcomes?

This could be possible with an integrated stepped care model (Bower & Gilbody, 2005). The stepped care model aims to minimise health care costs and contains two fundamental principles. First, clients initially receive the simplest and most cost efficient efficacious treatment to meet their individual needs. Second, clients progressively escalate to more

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resource intensive and more costly models when or if they are needed. So in this scenario what would be the simplest and most cost efficient efficacious treatment?

Undoubtedly, that would be an easily accessible, clinician-free, cost-free, Internet program similar to the one just described (Erickson et al., 2012; 2016). The speech-language pathologist could simply direct the client to the website and treatment would be self-directed from there. We know from the evidence that this could potentially help as many as a third of our clients, particularly those who have had previous treatment and are looking for a "booster." And if effective, clients would have ongoing access to the program to help maintain the benefits of their treatment over time.

But what about the others who do not obtain enough benefit from a standalone clinician-free treatment website? That is where the stepped care approach comes in. No health care expenses have been directed at the client so far, so the next logical step would be to direct some resources to the client. This could involve initial access to minimal clinician support, perhaps once a month, in conjunction with the Internet program, or perhaps intermittent clinician treatment supplemented with practice apps. Then, if minimal clinician contact combined with technology is not enough, the next step would be weekly webcam or in-clinic consultations, with generalist clinicians, employing whatever treatment approach seems best for the individual client. Or this could be the point to consider "boosters" for those who have previously had success in treatment. With this process, many clients may benefit from treatment simply and cost effectively. The limited resources that remain could then be directed to those who really need intensive treatment with specialist clinicians. And obviously, the management of anxiety and other client support could also be integrated into this stepped care model.

Summary and conclusions

In conclusion, it is imperative that speech-language pathologists are able to offer clients treatment choices, so that they get the treatment they want and need, when they need it, and for as little cost, inconvenience, and time commitment as possible. There are many alternatives to the traditional intensive programs: ones that generalist clinicians can easily implement and which may better suit at least some clients. Unfortunately, the research is still lagging behind many of these alternatives.

Questions from discussion leaders

If a person has severe stuttering, would you advocate direct speech intervention before or after psychological intervention?

We have no evidence to suggest that people with more severe stuttering generally have more significant mental health issues. Therefore I think the timing of psychological intervention should be based on the severity of the mental health problems and how much they interfere with the person's daily life and/or with speech treatment. A speech-language pathologist will typically assess or at least screen for psychological problems during assessment. If this indicates that any psychological problem is likely to interfere with beginning or complying with treatment, then I would refer to a psychologist to deal with this first. If the stuttering appears to be the greater problem, then I would begin treating this directly and introduce simple cognitive behaviour therapy strategies or refer to a psychologist when needed to deal with any psychological issues during treatment.

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Regarding the notion of stepped care with least clinician involvement first, is there evidence that people who stutter will remain motivated and committed if they require more than one treatment model?

There is no evidence for this, however, there are two points to consider. First, a client does not necessarily begin at the simplest level if there is a belief that the client will not respond to this treatment or will gain more benefit from starting with more support. This could be assessed before beginning any treatment. Second, if a client is showing little or no response to a treatment, you would not persevere with completing that treatment. So the notion that a client would be expected to complete one treatment after another is typically not justified.

Do you think that self-help groups can help to prevent post-treatment relapse?

I don't believe that self-help groups alone can prevent relapse. However, they can help in the sense that they can provide two forms of support: psychological support and opportunities for speech practice. Ideally, self-help speech practice would be monitored by a speech-language pathologist to ensure the appropriateness of some of the practice routines. Sometimes exercises can be performed out of habit rather than as a result of determining what strategies might work best, setting goals and evaluating the consequences. For best results, speech practice needs to constantly change to meet demands rather than being a static programme.

The value of speech restructuring treatment depends on how many clients really want it. Is there any evidence about how many clients who come to speech clinics really want to control their stuttering with speech restructuring?

There have been a couple of studies that have tried to answer this question although, in both cases, they were surveys where the study respondents were self-selected. However, that aside, it seems that more than half of our clients do seek this type of treatment, particularly young men. In many cases they seek this type of treatment several times throughout their lives, despite the effort required to control stuttering and to comply with treatment demands, and even though relapse can be a substantial problem. It seems clear that many clients, at least initially, do seem to want to try to reduce their stuttering rather than learning to live with it.

Can webcam telepractice with speech restructuring obtain as much compliance and motivation as when done in the clinic?

Certainly the evidence suggests that this is the case and in fact there is some evidence that compliance might be better with telepractice in general. Studies conducted with both adults and adolescents have shown that development and maintenance of clinician rapport is the same for both formats. However, clients overwhelmingly report that the convenience of telepractice—not having to take travel time from work or other commitments—outweighs that of clinic visits. Also there is evidence from some trials that drop-outs are minimal and clinician contact time is less for telepractice, so this might suggest better compliance and motivation for this format.

Overall, how would you describe the impact on quality of life when adults use speech restructuring successfully?

First I will need to define what I understand by the expression "use speech restructuring successfully." This will be individual for each client and involves using a fluency technique

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that sounds and feels acceptable to the client while controlling stuttering to an acceptable level in everyday situations. Research has shown that this is an attainable goal for many clients. The most common measure of quality of life used in research is the OASES (Yaruss & Quesal, 2010). Studies have shown reductions not only in the total impact score following treatment but also on individual section scores, including reactions to stuttering, communication in daily situations and quality of life. However, it should also be noted that one qualitative study (Cream, Onslow, Packman, & Llewellyn, 2003) has reported clients having an ongoing fear that they will lose control of their stuttering and therefore they continue to experience a need to protect themselves from the hurtful consequences of stuttering. These issues will likely affect quality of life.

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Information Processing and Stuttering

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Abstract

Many adults seeking treatment for stuttering will be diagnosed with social anxiety disorder. According to cognitive theorists, social anxiety disorder is maintained by information processing biases. Due to the high rates of social anxiety disorder associated with those who stutter, it is likely that they too will display similar information processing biases. The purpose of this presentation is to review the research evidence for information processing biases with social anxiety disorder and to describe the mechanisms for anxiety maintenance. Emerging evidence with adults who stutter suggests that they display information processing biases similar to those with social anxiety disorder. However, the effects of such information processing biases on anxiety maintenance or speech treatment outcomes with stuttering is unclear and further research is needed.

Clinical importance of the topic

Relapse after treatment for stuttering

The most efficacious treatment for adults and adolescents who want to control stuttering is speech restructuring (Andrews, Guitar, & Howie, 1980; Bothe, Davidow, Bramlett, & Ingham, 2006; Ingham, 1993; Ingham & Andrews, 1971; Onslow, Costa, Andrews, Harrison, & Packman, 1996; Packman, Onslow, & Menzies, 2000). Speech restructuring involves altering the way a person speaks with the use of a novel speech technique.

Many adults who receive speech restructuring treatment will fail to maintain their treatment benefits (Andrews & Craig, 1988; Block, Onslow, Packman, & Dacakis, 2006; Craig, 1998; Craig & Calver, 1991; Craig & Hancock, 1995; Martin, 1981). Based on percentage syllables stuttered, reported relapse rates vary from 23% (Boberg & Kully, 1994) to 40% (Howie, 1981). Using self-report data, rates up to 72% have been reported (Craig & Hancock, 1995). Pre-treatment severity is one factor that has most consistently been related to relapse (Andrews & Craig, 1988; Block et al., 2006; Craig, 1998; Huinck et al., 2006). Also, those with less favourable attitudes to communication may be less likely to maintain treatment gains (Guitar, 1976; Guitar & Bass, 1978), although Block et al. (2006) were unable to replicate this finding. Locus of control has been associated with relapse (Craig & Andrews, 1985), but did not predict long-term treatment outcomes in the De Nil and Kroll (1995) or Block et al. (2006) studies.

The effect of comorbid mental health disorders on speech treatment

Research conducted during the past decade has produced an explanation for the problem of post-treatment relapse with stuttering. Several studies have shown that many adults seeking treatment for stuttering present with co-morbid mental health disorders (Iverach, O'Brian, et al. 2009; Iverach et al., 2010; Stein, Baird & Walker, 1996), including anxiety disorders such as social anxiety disorder and mood disorders such as depressionIverach, Jones, et al. (2009) implicated such disorders with failure to maintain speech treatment

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[†] Ross Menzies and Mark Onslow contributed to the material from which this paper was developed.

benefits. In that study involving 64 adults seeking treatment for stuttering, Iverach and colleagues reported that around two-thirds of participants were diagnosed with having one or more mental health disorders. Those participants failed to maintain the benefits of their speech treatment at the 6-month follow-up assessment. That finding occurred in the absence of any significant differences between the groups for stuttering severity pre-treatment or immediately post-treatment.

Social anxiety disorder

The most prevalent mental health disorder for adults who stutter is social anxiety disorder. Between 20–60% of participants seeking treatment for stuttering are diagnosed with the condition (Blumgart et al., 2010; Iverach, O'Brian et al., 2009: Menzies et al., 2008; Stein et al., 1996). Social anxiety disorder is one of the most common anxiety disorders, affecting up to 13% of the general population (Grant et al., 2005; Kessler et al., 2005; Ruscio et al., 2008). According to the Diagnostic and Statistical Manual of Mental Disorders (American Psychiatric Association, 2013), social anxiety disorder is characterised by extreme distress in social or performance-based situations where there is the potential for scrutiny from others. The central feature is an extreme fear of humiliation or embarrassment in social situations, which is excessive and irrational in comparison to the actual threat (American Psychiatric Association, 2013).

Social anxiety disorder is a distressing and debilitating condition. Social anxiety disorder can impact on overall quality of life (Sareen et al., 2006; Schneier et al., 1994; Stein & Kean, 2000; Stein et al., 2005). The condition affects educational and occupational attainment. Further, social anxiety disorder is associated with comorbid depression and substance use disorders (Kessler, 2003; Lampe, Slade, Issakidis, & Andrews, 2003; Marks, 1969; Turner, Beidel, Dancu, & Keys, 1986; Wittchen, Stein, & Kessler, 1999).

Let's consider a case example. Ivan is a 35 year-old man. He works for the public service in an administrative role. Ivan has been diagnosed with social anxiety disorder. He fears talking in front of others. He is afraid that he will say something stupid, and as a result his work colleagues and boss will develop a negative opinion about him. As a consequence, Ivan avoids specific work situations, such as talking on the phone in the presence of others. He waits for the caller to leave a message and then returns the call in a private office or when other people are not around to hear him make the call.

Avoidance and in particular, avoidance of particular situations is common for many people with social anxiety disorder. However while Ivan, as in the example above, and many others with social anxiety disorder avoid a range of situations, there are many situations that cannot be avoided. Typically, many of those situations that are not avoided, don't end in disaster either. In other words, the feared outcome does not occur or is not as catastrophic as predicted. So why do those with social anxiety disorder continue to experience such anxiety in the absence of feared outcomes? That is the puzzle.

Information processing biases

According to cognitive theorists, social anxiety disorder is maintained by information processing biases (Clark & Wells, 1995). Examples of information processing biases include self-focused attention, observer perspective memories, spontaneous imagery, and attentional biases. While this is not an exhaustive list, these four information processing biases are the focus of this paper.

Self-focused attention

Self-focused attention involves attending to negative thoughts or anxiety symptoms such as the heart beating fast. Let's consider our case example. Wen Ivan is anxious he notices his heart rate increases and he starts to feel shaky. However, focusing on this information can affect Ivan's ability to notice what is happening in the situation. For example, if Ivan focuses attention towards how he feels, he doesn't notice that the social situation he is in is working well. He doesn't notice that others appear unaware of his anxiety symptoms. The consequence of this is that when Ivan is recalling details about the situation, he can only remember the information that he was focusing on: his heart beating fast and his shakiness. Because of this self-focused attention Ivan has no recollection of what actually happened during the situation and how others perceived him. However, Ivan desperately wants to know how he was perceived by others. This is a common characteristic of those with social anxiety disorder. Therefore information from self-focused attention rather than accurate feedback from past social interactions is used to evaluate performance during those social interactions.

Observer perspective memories

For many people with social anxiety disorder, memories of the situation are often recalled from the perspective of others. This is referred to as the observer perspective. The observer perspective can be described as looking at the self from an external point of view. This is in contrast to the field perspective in which a people look at or view a situation from the perspective of themselves, looking out at the surrounding environment. There is much empirical evidence supporting an observer bias recall by those with social anxiety disorder for memories (Coles, Turk, Heimberg, & Fresco, 2001; D'Argembeau, Van der Linden, d'Acremont & Mayers, 2006; Wells & Papageorgiou, 1998).

What is intriguing about this observer perspective is that it is completely inaccurate. We don't actually see ourselves and how we are performing during social situations. The problem is that those inaccurate observer perspective memories are used as evidence for what happened during social encounters and typically that information confirms a person's fears. For instance, a typical fear by one who is socially anxious is that others will be able to observe their anxiety symptoms, which will lead to negative evaluation and disastrous consequences. When memories are recalled from the perspective of others and depict the person looking anxious, it is that information that is used as evidence of performance, hence confirming fears. It is no wonder then that those with social anxiety continue to feel anxious.

Spontaneous Imagery

Many people with social anxiety disorder see mental images that appear spontaneously during situations when they feel anxious. These images are scenes that typically depict a former social situation when the person was embarrassed or experienced negative evaluation. For example, Ivan recalled an event from his school years when a teacher asked him a question in class and he was not able to answer it. The teacher made a comment and the whole class laughed. Ivan was extremely embarrassed. Ivan now frequently recalls that scene when he feels anxious about an upcoming social event or during social situations.

Eye gaze avoidance

Avoiding eye gaze is commonly reported by those with social anxiety disorder and has recently been shown to occur in laboratory experiments (Horley, Williams, Gonsalvez, & Gordon, 2003; 2004). Avoiding eye contact with others might be used to avoid being

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included in conversations and therefore prevents the possibility of negative evaluation from others. However, avoiding eye contact can lead to biased information processing. As with our case example, if Ivan avoids eye contact with others, he won't be able to see their reactions. He won't benefit from learning that being part of a conversation may not result in disastrous outcomes. People might actually be showing an interest in what Ivan has to say. However, he won't be aware of that if he is avoiding eye contact.

What is important about avoiding eye gaze is that it can actually lead to negative reactions from others. Avoiding eye gaze can give the impression that Ivan is aloof and not interested in others, or not interested in the conversation. This can in fact lead to the feared outcome of negative evaluation from others.

Interactions among information processing biases

The information processing biases discussed above do not necessarily occur in isolation either. They often interact and impact on each other. For example, self-focused attention and avoiding eye contact can result in information from the social interaction not being noticed. So when recalling memories of the situation, the only information available will be from self-focused attention.

Research findings about the topic

Eye gaze avoidance with adults who stutter

One of the most common behaviours reported by people who stutter is that many avoid maintaining eye contact with others during interactions. Even early treatment programs for those who stutter incorporated procedures to encourage eye contact with others (Luper & Mulder, 1964; Sheehan, 1970).

The first direct investigation of eye gaze with adults who stutter was conducted by Lowe et al. (2012). In that study, adults who stutter and control participants gave a speech in front of an audience. The audience was filmed for the study and displayed on a television screen, and the audience members were trained to display positive, negative and neutral expressions and behaviours. An eye tracker was used to record participant eye gaze while giving the speech. Overall, participants in the stuttering group spent less time than controls looking at audience members while they were giving their speech. In addition, for the stuttering participants, reduced eye gaze towards positive audience members was associated with negative self-perceptions and anxiety during the speaking task.

Imagery and recalling scenes from an observer perspective

In the first study exploring imagery with adults who stutter, Tudor, Davis, Brewin and Howell (2013) reported that the participants who stuttered experienced recurrent images of themselves more than control participants. In addition, the images were linked to memories of previous situations. The themes common to both groups were disfluency, anxiety, negative evaluation, and self-focus. However, additional themes for the stuttering participants included helplessness, shame, sadness and frustration. In another study, adults who stuttered were more likely than control participants to see images of themselves that were distorted, negative and recalled from the observer perspective (Lowe, Menzies, Packman, O'Brian & Onslow, 2015). So, the adults who stuttered in that study were more likely to recall memories or images of themselves that were from the perspective of another person.

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Self-focused attention

There is evidence that those who stutter may focus on information about themselves during social interactions. For example the evidence from the imagery studies just mentioned suggests that during social interactions they focus on how they feel and how they are perceived by others. Further, the Unhelpful Thoughts and Beliefs about Stuttering scale (St Clare et al., 2009) is a self-report measure developed from a file audit of the negative thoughts of people who stutter who presented to an anxiety disorders clinic for treatment. Many items on the scale relate to self-focused attention: "I will stutter," "Everyone in the room will hear me stutter," "I'll block completely and won't be able to talk," "I can't think clearly because I stutter," and "My pauses are too long."

Clinical application of the research findings

Client needs and attentional biases

So how can these findings be applied to the work of speech-language pathologists? Many adults who stutter with mental health disorders will not have received treatment for their mental health condition. This is particularly important for speech-language pathologists who are often the first point of contact for those who stutter.

It is important that speech-language pathologists consider the needs of their clients seeking treatment. In the majority of cases such needs will relate to controlling or managing stuttering. However, speech-language pathologists are ethically bound to consider whether clients may benefit from the treatment that they seek. For those clients who want to control stuttering, speech restructuring techniques require significant attentional resources to learn and implement. So, for those clients who present with social anxiety, information processing biases such as self-focused attention may affect the ability to use speech techniques. For example, focusing on negative thoughts, spontaneous imagery or anxiety symptoms, may result in clients being unable to direct adequate attentional resources to implementing speech restructuring techniques.

In addition to this, while speech restructuring techniques can provide a means to control stuttering, they can feel unusual or unnatural for the client to use. So even though clients may be able to control stuttering, they may be concerned they will sound strange when using a speech technique and they may feel different when they speak that way (Cream, Onslow, Packman & Llewellyn, 2003). It is critical then for those who stutter to attend to external information during interactions with others. They need to look at others during social interactions to obtain accurate feedback. Feedback from others might help to disconfirm their fears of negative evaluation associated with using a speech technique to control stuttering.

Assessing attentional biases

Generally, speech-language pathologists are not trained to diagnose social anxiety disorder. That is the domain of psychologists. However, there are certain features that may alert speech-language pathologists to the presence of social anxiety. Further, there are simple procedures that can be used to identify those features.

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Memory recall

A simple technique can be to ask clients to recall and describe memories of social situations when they felt anxious. From those memories, the clinician can ascertain whether clients recall faulty memories from an observer perspective.

Let's consider a case example. Ross is a clinical psychologist. In an interview with Beth, he asks her to recall a recent social situation when she felt anxious. Ross asks Beth if she can picture a scene or an image of the situation, and if so, to describe the scene. Beth recalled a recent social event when she met some friends at a restaurant. She said that she couldn't remember much from the situation such as what the group talked about during the evening. Ross asks Beth if she could see herself or just the others in the situation. Beth noted that she saw herself in the scene and that her face looked red when she was talking to her friends. This recall was obviously the perspective of an observer.

Spontaneous images

In order to assess whether a client experiences spontaneous imagery, similar interview techniques can be applied. Let's return to the case example about Beth. Ross asks Beth a series of questions to explore whether she sees spontaneous images of herself. He asks Beth whether she was anxious on her way to the restaurant to see her friends. He then asks her what she was thinking about on her way. Beth recalled and described an event that occurred many years prior when was still at high school. Beth described a situation when she stuttered in front of a group of friends. People in the group laughed at her and this was a source of excruciating embarrassment for Beth. Beth noted that she frequently sees that image when she is about to meet with a group of people. Asking clients about their thoughts is another simple technique that can help to determine if spontaneous imagery occurs with clients.

Self-focused attention

Attending excessively to to negative thoughts, anxiety symptoms, and how one is coming across to others might indicate that clients engage in self-focused attention. Self-focused attention is associated with anxiety maintenance. Therefore is it important to identify if clients who stutter are excessively self-focused. Again this can be achieved through some simple interview questions. In the case example, the psychologist Ross, asks Beth to recall details about the experience in the restaurant with her friends. Beth couldn't recall specific details about the evening or about the conversations she had with her friends. Ross probes Beth further by asking her to think about why that might be. Beth noted that she tends to focus on how she is coming across to others, what she is going to say, how she sounds to others, what she said, even where to put her hands so she looks natural and relaxed. Ross pointsout to Beth that she is aware that she misses a lot when focusing on herself. In addition to this, worrying about how she is coming across to others is affecting Beth's interactions. Beth noted that her friends frequently comment that they often have to repeat information to her.

Eye gaze avoidance

Clients may not always avoid eye contact with speech-language pathologists in the clinic, so that may not be an accurate assessment of eye gaze habits. There are several other opportunities to observe eye gaze with clients. Speech-language pathologists may be able to observe their clients interacting with the receptionist when they arrive for their appointment. Interview topics at assessment can be revealing, such as strategies clients use during social interactions when feeling anxious, and how they manage situations when they

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feel anxious. It is always useful to ask clients to describe interactions with others in general terms. It is also useful to note that, rather than avoiding eye gaze, clients instead may break eye contact during moments of stuttering. If this occurs frequently, then they are likely to miss information and potentially positive or encouraging reactions from others when the experience moments of stuttering.

Cognitive Behaviour Therapy

Cognitive behaviour therapy (CBT) is the leading treatment for social anxiety disorder. Among the standard procedures used during CBT are those that correct information processing biases. Although CBT is typically administered by psychologists or those who have received a formal qualification to do so, can speech-language pathologists target information processing biases during treatment with their clients? Menzies, Onslow, Packman, and O'Brian (2009) have described procedures that may be used by speech-language pathologists with the caveat that the procedures are implemented by those who have received relevant training and that training is recognised by the professional association in which they practice. Whether information processing biases are best treated before starting treatment, in combination with speech treatment, or after treatment remains to be shown with research. However, if a client presents with clinically significant levels of distress related to anxiety, then referral to a psychologist is warranted.

Speech treatment and self-focused attention

Clearly, behavioural treatments for stuttering work for some clients who want to control stuttering. However, does teaching a client speech restructuring techniques increase self-focused attention? It may do because speech restructuring draws attention to speech and speech performance. It is important then for speech-language pathologists to consider whether the techniques taught in some speech treatment programs increase self-focused attention and therefore contribute to the persistence of anxiety.

There is however clearly a potential problem. Clients may fail to maintain the benefit of their speech treatment because it increases self-focussed attention, particularly if this is associated with a mental health disorder. For such clients, behavioural treatment for stuttering may not be appropriate. Therefore it is important for speech-language pathologists to carefully assess clients who stutter before providing speech treatment that may not be suitable. For clients already receiving treatment, ongoing assessment as described in this paper is critical.

Summary and conclusions

It is clear that continued research is needed to determine the extent to which information processing biases are a characteristic of people who stutter in general, not simply a reflection of social anxiety. Additionally the effects of information processing biases for anxiety maintenance and the ability of clients to maintain speech treatment benefits need to be determined. Also, research is needed to verify whether information processing biases can be modified with the same techniques used for those with social anxiety disorder. The results of that research may inform the development of improved treatments for those who stutter to improve their overall quality of life.

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Questions from discussion leaders

Do you think it would be helpful for clinicians to assist people who stutter to increase eye-gaze?

Cognitive theories suggest that those with anxiety disorders fail to observe and process information from the social environment due to maladaptive attentional biases. Standard CBT procedures involve psycho-education, behavioural experiments, and cognitive restructuring. Those procedures may involve instructing clients to look at and observe the behaviours and reactions of others during social encounters to observe accurate information from the social encounter and evaluate situations and outcomes accurately. A CBT package developed for adults who stutter (Menzies et al., 2008) has been shown to improve overall functioning and wellbeing, reduce avoidance of speaking situations and remove diagnoses of social anxiety disorder. Therefore, if a client presents with signs of anxiety a psychological assessment is indicated to determine if CBT is warranted. That treatment may involve instruction to increase eye gaze during social interactions in conjunction with evaluation and cognitive restructuring procedures and that will be determined according to specific client needs.

Even when those who stutter maintain eye gaze, will they see negative things that are not there?

Spontaneous mental imagery is common with those who are socially anxious. Two studies reviewed (Lowe et al., 2015; Tudor et al. 2013) have reported that people who stutter experience negative mental imagery. In isolation, direct instruction to increase and maintain eye contact is unlikely to repair other maladaptive attentional processes that may be involved with anxiety maintenance. Treatment for those who are anxious involves CBT procedures that incorporate psycho-education, behavioural experiments, cognitive restructuring, and rescripting of memories and images. I suggest that clients who stutter displaying signs of anxiety require a comprehensive psychological assessment in order to identify information processing biases and behaviours that may maintain anxiety and impede speech treatment progress. Some may require treatment from a clinical psychologist.

Is it possible, then, that speech restructuring increases self-focused attention and in fact is psychologically harmful?

It is well established in the psychological literature that those who are anxious focus attention towards anxiety symptoms and ruminate on past events and performance. Treatment for adults who stutter to help them control their stuttering involves teaching clients to use a novel speech technique. In order to evaluate performance, clients rate their stuttering severity, evaluate how well they implemented the speech technique, and identify factors that may impact on their capacity to use such a technique. Those procedures require clients to focus on their performance during and after social encounters. It is currently not known if those procedures are psychologically harmful for those who stutter, that is, whether they contribute to the development or maintenance of anxiety. Research is required to determine if speech treatment approaches such as speech restructuring or specific procedures within those treatments contribute to the development and maintenance of anxiety for those who stutter.

Where is all this research going? During the 1980s authorities of the day pronounced that there was no relationship between stuttering and anxiety, and at the turn of the century the

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view began to change, and now it seems that those who stutter have mental issues that are as troubling as their speech issues. Can you project what you might be telling us in 20 years from now?

Psychological theories and models that emerged during the 1980s and 1990s form an important basis for researching the effects of anxiety with those who stutter. Research is beginning to unveil important information about the way in which adults who stutter process information during social encounters and their effects on anxiety maintenance. In 20 years from now we will have a better understanding of the factors that maintain anxiety with adults who stutter and the impact of those factors on stuttering and speech treatment outcomes. That research evidence will inform the development of treatments for those who stutter to improve their overall quality of life. Additionally, continued research exploring the mechanisms that contribute to the onset and maintenance of anxiety with those who stutter has the potential to inform the development of preventative programs and treatments.

What take-home message would you like students of speech-language pathology to have about this topic?

Students need to be aware that adults who stutter are at risk of developing mental health disorders, in particular anxiety disorders. The presence of mental health disorders can affect client's ability to maintain their speech treatment benefits. Speech-language pathologists should ensure their clients are aware of those risks and consider that evidence during treatment planning to determine whether it is appropriate to commence speech treatment in the first instance.

The take home message is that it is essential that any adult who stutters presenting for speech treatment should be assessed or screened for anxiety and mental health disorders. If a mental health disorder is suspected then a referral to a psychologist will be required.

To pursue your answer to that last question a little more, that seems to be onerous for students. They would be advised during their careers to not administer speech treatment to adult clients with a mental health disorder, and to screen for mental health disorders before beginning any speech treatment. Yet a great many—perhaps the majority— of their clients will fail a mental health screening. They will not be qualified to diagnose mental health disorders in the event of such screening failures, so they will be constantly dependent on clinical psychologists. Is that a fair assessment?

Evidence suggests that any adult who stutters could benefit from cognitive behaviour therapy. For instance Menzies et al. (2008) showed that Global Assessment of Functioning scores improved for the group of participants who received the CBT package. In addition the CBT package reduced speaking situation avoidance as well as eliminating social anxiety disorder. The CBT package has been translated to a standalone Internet treatment which is demonstrating similar outcomes.

One approach for speech-language pathologists is to direct clients to the standalone Internet CBT program before starting speech treatment. Alternatively, if it becomes apparent that anxiety is impacting on speech treatment progress, then clients could be directed to the Internet CBT program during speech treatment or speech treatment could be suspended temporarily while they are completing the program. Further research is needed however it is foreseeable that speech-language pathologists may be able to manage their clients without the need for training in psychological assessment or CBT

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procedures. Consequently, access to a clinical psychologist for many clients may not be required.

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Mental Health Measurement and Management with Stuttering

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Abstract

In the past, stuttering has been seen as a disorder falling solely within the clinical domain of speech-language pathology. However, it has become increasingly apparent that adults who stutter may suffer from a range of comorbid mental health disorders that require treatment for satisfactory speech outcomes to be achieved. This presentation explores the mental health difficulties among adults who stutter, and proposes solutions to the problems that these comorbid disorders present. Literature regarding anxiety, its assessment and treatment, and its relationship to stuttering is reviewed. Consistent findings have been reported for elevated rates of anxiety disorders among adults who stutter. Further, mental health diagnoses were associated with higher rates of relapse 6 months after treatment with a standard speech restructuring program. Fortunately, cognitive-behaviour therapy may adequately treat the anxiety symptoms typically associated with stuttering. Taken together, the findings strongly suggest that adults who stutter need to be screened for mental health disorders, particularly the anxiety disorders. Given that the presence of these disorders interferes with long-term outcomes from speech restructuring treatment, it is suggested that this screening is completed prior to undertaking speech treatment.

Clinical importance of the topic

In the past, stuttering has been seen as a communication disorder falling within the clinical domain of speech-language pathology. However, during the last decade it has become clear that those who stutter may suffer from a range of comorbid mental health disorders, particularly conditions with an anxiety basis (see Iverach, O'Brian et al., 2009). There are many reasons why this is one of the major developments for the field of stuttering in recent times.

The complexity of the disorder
First, the complexity of stuttering is greater than previously thought. The presence of anxiety-related disorders may complicate the clinical picture, and the response of the individual to speech treatment. In other areas of health, complex comorbidity tends to lead to poorer health outcomes (Belanger et al., 2016; Goddard, Wingrove & Moran, 2015; Wolitzky-Taylor et al., 2015).

Engagement with speech-language pathology services
Second, the presence of anxiety-based conditions may influence the way in which those who stutter engage with speech-language pathologists. For example, will anxious individuals potentially reject speech techniques? Will they use the speech techniques taught in speech rehabilitation across all situations? It is possible that those who stutter may prefer to produce repetitions and blocks than "odd" or unnatural sounding speech. In this regard, there is evidence that some who stutter have negative impressions of the sound of restructured speech (Cream, Onslow, Packman & Llewellyn, 2003). Will there be an interaction between the speech naturalness achieved by clients during treatment, their baseline anxiety levels, severity of stuttering and willingness to engage in speech restructuring treatments? This latter question, perhaps more than any other in the field of mental health and stuttering,

shows the increased clinical complexity of the disorder that arises from the discovery of comorbid mental health issues.

In a related concern, there may be aspects of current speech practice that might interfere with recovery from anxiety states. Given that speech-language pathologists have been unaware of these comorbid problems, they may have been inadvertently fostering the maintenance—or even the development—of anxiety-based problems. While this proposition is clearly speculative, it warrants investigation (Helgadottir, Menzies, Onslow, Packman, & O'Brian, 2014a).

Who shall provide treatment?

The presence of comorbid mental health issues among those who stutter raises questions about who is best equipped to treat these psychological difficulties. Can speech-language pathologists become involved in the management of these comorbid problems? Alternatively, should the treatment of these additional conditions simply rest with clinical psychologists and psychiatrists? On that front, are mental health professionals sufficiently trained to manage this particular population? Those who stutter may be very different from the anxious clients typically seen in mental health services. For example, unlike other populations with social anxiety disorder, those who stutter have not been shown to differ in temperament from the general population (see Kefalianos, Onslow, Block, Menzies & Reilly, 2012). Since the mental health issues of those who stutter seem to arise from early negative social conditioning rather than temperament, treatment response for this population may differ from other anxious groups.

Identification of mental health problems with stuttering

The presence of psychiatric illness in this population raises the question of how these mental health problems should be identified. If speech-language pathologists are the front line of treatment for this disorder, how will they go about identifying those with mental health issues? Should they be trained in the screening of mental health disorders? Is this feasible? Should every individual who stutters have a mental health screen?

Treatment priorities

If mental health issues are present with stuttering clients, what should the order of treatment be? Should speech treatment precede the treatment of mental health disorders, or should mental health be given the clinical priority? Can the treatments proceed in parallel or will this negatively impact on clinical outcomes? (see Menzies et al., 2008).

As discussed now, research has only recently begun to explore some of these complex questions.

Research findings about the topic

The presentation identifies research evidence published in refereed journals about the relationship between anxiety, mental health, stuttering and treatment outcomes. Where multiple papers report similar findings, preference is given to recent publications in this brief review. The purpose of the review is to explore the mental health difficulties among adults who stutter, and to propose solutions to some of the problems that these comorbid disorders introduce in treatment.

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The mental health of adults who stutter

Iverach, O'Brian et al. (2009) reported dramatically increased odds of a range of psychiatric disorders among treatment-seeking PWS, compared to community controls. Ninety-two clients seeking treatment for stuttering were compared to age and sex-matched individuals from the Australian National Mental Health Survey using the same psychiatric structured interview. The clients who stuttered were shown to have dramatically increased odds, compared to controls, of having social anxiety disorder: a 16 to 34-fold increased odds. In addition, increases were noted for generalised anxiety disorder, panic disorder, obsessive-compulsive disorder, and even the mood disorders (Iverach, O'Brian et al., 2009).

This was not the first, and certainly not the only, report to suggest that those who stutter experience high rates of anxiety disorders, particularly social anxiety disorder. Data from several research teams suggest that up to 60% of adults seeking treatment for stuttering suffer with social anxiety disorder (Blumgart, Tran, & Craig, 2010; Menzies et al., 2008; Stein, Baird, & Walker, 1996). This should not be surprising, given that social anxiety disorder centres on intense anxiety and fear of negative evaluation in social situations (American Psychiatric Association, 2013). The socially anxious individual will seek to avoid situations that may involve scrutiny and evaluation from others. Typical situations include public speaking, meeting new people, leading a group discussion, introducing other people, and conversations that may involve conflict or confrontation (American Psychiatric Association, 2013). Social anxiety disorder is disabling, as so many aspects of daily life involve social connection. Not surprisingly then, social anxiety disorder is associated with reduced educational and occupational achievement, unemployment, and lower socioeconomic status (Stein & Kean, 2000).

The high prevalence of social anxiety disorder in those who stutter, and the associated costs of the condition, are obviously concerning. However, even more significantly, in a separate report, Iverach, Jones et al. (2009) found that the presence of a mental health diagnosis is associated with higher rates of relapse 6 months after speech treatment. Of 64 clients who received speech treatment in this report, 18 (28%) had no mental health disorder and the remainder (72%) had one or more mental health disorders, most commonly social anxiety disorder. Notably, there were no significant pre-treatment differences in stuttering severity between clients with no mental health disorder and clients with one or more mental health disorders. At 6 months post-treatment, those without a mental health diagnosis maintained their treatment benefits. However, those with a mental health disorder were unable to effectively demonstrate a restructured speech technique in beyond-clinic phone calls. That is, they appeared to have lost their capacity to produce stutter-free speech. Put simply, they had begun to relapse.

The treatment of anxiety in adults who stutter

Given the high rates of mental health problems in those who stutter seeking treatment, and the effect of these disorders on stuttering treatment outcomes, it is imperative that effective treatment for anxiety-related disorders be developed. In terms of the treatment for these problems, cognitive-behaviour therapy (CBT) remains the gold-standard intervention in psychology. Menzies et al. (2008) designed the first CBT package specifically for adults who stutter and conducted the first randomised trial of this type of intervention. The results showed that CBT, when presented by a clinical psychologist, dramatically improved the global functioning of the clients and eliminated speaking situation avoidance. Two thirds of the experimental group (who received CBT) were diagnosed with social anxiety disorder, but

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none of that group retained this diagnosis at 12 month follow-up in blinded psychiatric interviews (Menzies et al., 2008). The psychiatric status of control participants (who received speech restructuring but did not receive CBT) remained unchanged at follow-up. Individuals who received the CBT intervention were clearly more engaged in everyday activities, and suffered less anxiety in these activities, than those who did not receive CBT (Menzies et al., 2008).

Notably, however, the therapy sessions in the randomised trial of CBT described above were conducted by a senior clinical psychologist with extensive experience in the management of stuttering. The generalisability of the findings to community psychological services is questionable, with few psychologists claiming expertise in stuttering. Accordingly, recently research attention has turned to developing and testing interactive, standalone Internet CBT treatment programs that require no psychologist. The economic advantages of clinician-free, standalone Internet treatment are obvious, and Internet treatments are emerging in a range of health domains. In social anxiety disorder, where avoidance of therapy itself (because of fear of scrutiny) can be significant, the advantages are particularly apparent (see Helgadottir, Menzies, Onslow, Packman, & O'Brian, 2009a). The standalone Internet CBT method allows treatment access to those in rural and remote areas, and the treatment can be accessed at any hour in the client's own home. In the present context, it is particularly appealing that speech-language pathologists are able to integrate CBT treatment with speech restructuring treatment without needing psychological training or access to a clinical psychologist.

Prior to developing their clinician-free standalone Internet CBT treatment package, the team at the Australian Stuttering Research Centre summarised the literature on published online CBT treatment programs for the anxiety disorders (Helgadottir, et al., 2009a). They concluded that existing Internet CBT programs had several significant deficiencies. First, no program at this time customised their clinical formulation for individual users. That is, each person using the program received essentially the same intervention, without consideration for the symptom picture of the particular case. Second, the existing programs did not incorporate human-like features to engage clients, such as faces and voices. It was suggested that the lack of these features was a major contributor to the poor compliance and completion rates associated with these programs (Helgadottir et al., 2009a). In general, Internet CBT programs at the time were essentially fixed, online workbooks that were failing to take advantage of the possibilities of computer-administered treatment.

The CBTpsych program

In response to these issues, researchers at the Australian Stuttering Research Centre adapted their standard, in-clinic CBT treatment for standalone Internet application: CBTpsych. The program they developed involves no clinician contact, but presents customised feedback to participants based on established CBT procedures for social anxiety. In order to emulate a real clinician experience, the CBTpsych program uses the voices and faces of a psychologist who talks to the user throughout by means of recordings (Helgadottir, Menzies, Onslow, Packman, & O'Brian, 2009b). Most importantly, the program individualises the treatment for each user based on pre-treatment assessments. No two users experience the same program, or complete the same set of homework exercises. In this way, CBTpsych mimics the clinical experience that clients receive in typical anxiety treatment units. That is, treatment progression, and the selection of homework procedures, depends on the individual's symptom picture and their weekly progress with individualised homework exercises.

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CBTpsych is divided into seven modules. Critically, prior to starting the treatment modules, an individualised profile for each user is built from an online battery of measures designed to assess negative thoughts, behaviours, and emotional responses. *Module One* presents basic concepts including the relationship between thoughts, attitudes and emotions. It is essentially psycho-education about the foundations of CBT. *Module Two* uses data from the online assessment battery to target the particular negative thoughts about stuttering that are driving anxiety in each user. The module requires the user to write at least 40 different cognitive challenges to particular unhelpful thoughts. Repetition of thought challenging is a cornerstone of the program.

Module Three extends the psycho-education provided in *Module One*. It expands the users understanding of the relationship between avoidance, thoughts, anxiety, safety behaviours, physiologically symptoms, and self-focused attention. The instructions show users how to build their own idiosyncratic anxiety formulation, incorporating each of these elements. To our knowledge, CBTpsych is the first fully automated internet-delivered program to achieve individualized anxiety formulations.

In *Module Four*, users select one of their avoided situations and the program creates a behavioural experiment for that situation, targeting the unhelpful thoughts responsible for driving avoidance and anxiety. The behavioural experiment is directly derived from the individual formulation constructed in the preceding module. Importantly, participants are asked to eliminate any safety behaviours that they regularly use to minimise stuttering in speaking situations. Typical examples of these are avoiding difficult words, rehearsing answers before speaking, overusing non-verbal communication, and avoiding eye contact. Safety behaviours have been shown to contribute to the maintenance of anxiety disorders in several trials (see Helgadottir et al., 2009b). Helgadottir et al. (2014a) reported that speech-language pathologists often recommend safety behaviours to clients who stutter during speech treatment. Further, self-reported use of these safety behaviours has been shown to be positively correlated with anxiety among clients who stutter (Lowe, et al., 2017).

Module Five of CBTpsych is designed to eliminate self-focused attention and an observer perspective of social interaction, which are common maintaining factors in social anxiety (see Helgadottir et al., 2009b). In *Module Six*, after an extensive voiceover introduction from the program, the user is asked to write a short essay on "why it doesn't matter what other people think of me." *Module Seven* covers relapse prevention and daily strategies for maintaining treatment gains.

Clinical trials of CBTpsych

In 2009, a Phase I trial of the CBTpsych with two clients with stuttering and social anxiety disorder provided proof of concept for the package (Helgadottir, Menzies, Onslow, Packman, & O'Brian, 2009b). Neither participant had a social anxiety disorder diagnosis at post-treatment, and both experienced clinically significant reductions in fear of negative evaluation and improved quality of life.

In 2014 a Phase II trial of CBTpsych attained similar results with 14 participants in a pre-post design (Helgadottir, Menzies, Onslow, Packman & O'Brian, 2014b). Seven participants were diagnosed pre-treatment with social anxiety disorder and all, with exception of two who did not complete all modules, lost that diagnosis at post-treatment. Significant improvements were reported for the Fear of Negative Evaluation scales, the Depression Anxiety Stress Scales, and the Unhelpful Thoughts and Beliefs About Stuttering scale. Nine participants

completed all seven modules, and a linear relationship was found between post-treatment scores and number of modules completed. In general, the results replicated the earlier positive findings of Menzies et al. (2008) with therapist-driven CBT.

Notably, these Phase I and II trials involved pre- and post-treatment assessment at a speech clinic. This contact may have increased compliance, commitment and adherence with the CBTpsych program, raising questions about the external validity of those trials. Given this, Menzies and colleagues conducted a large international trial of CBTpsych program with no contact of any kind from researchers or clinicians (Menzies, O'Brian, Lowe, Packman & Onslow, 2016). Two hundred and sixty-seven stuttering participants, recruited through speech-language pathology services in 23 countries, were given a maximum of 5 months access to CBPpsych. Completion of the program was associated with large, statistically and clinically significant reductions for all measures. The reductions were similar to those obtained in earlier trials of the program, and those obtained in trials of live, in-clinic CBT with an expert clinician. It was striking that the mean post-test Depression Anxiety Stress Scales scores were within the normal community range on all subscales (Menzies et al., 2016).

Clinical application of the research findings

The mental health status of those who stutter is an area of growing interest among researchers and clinicians alike. This brief review has shown that they have significantly impoverished mental health, with alarmingly high rates of social anxiety disorder and related conditions. Further, the presence of these comorbid mental health problems has been shown to interfere with speech restructuring treatment. Though the specific reasons for the relationship between mental health status and relapse have not been identified, it is clear that the presence of mental health disorders is a poor prognostic indicator for speech interventions for stuttering. Fortunately, as has been shown above, the anxiety-related problems of stuttering clients can be effectively treated with contemporary CBT procedures. The most recent data suggest that these techniques can be effectively delivered in an automated, standalone Internet program that does not require the active involvement of mental health practitioners or services.

Given that mental health problems are associated with poor medium-term outcomes, it seems imperative that speech-language pathologists advise clients who stutter of the need for mental health screening. Whether all clients should be screened prior to undergoing speech treatment is open to debate. Though such a policy may seem overly conservative, it is worth remembering that, in the Iverach, Jones et al. (2009) report, only only 28% of those seeking treatment had no mental health disorder and did not relapse after treatment.

A less conservative approach would involve speech-language pathologists administering screening tools and conducting a basic mental health assessment prior to undertaking speech treatment. A range of measures are available to assess anxiety, avoidance and related phenomenon among those with social anxiety (Iverach, Menzies, O'Brian, Packman & Onslow, 2011). However, few have been derived by examining the actual negative thinking style of the specific population of stuttering clients. Most scales, like the Fear of Negative Evaluation scale (Watson & Friend, 1969), have been based on responses of those with general social anxiety, rather than the combination of stuttering and anxiety. In fact, only two scales in the literature are directly derived from client responses involving those who stutter undergoing psychiatric interviews and treatment for anxiety. The first such scale to be

published was the Unhelpful Thoughts and Beliefs About Stuttering (UTBAS) scale (St Clare et al., 2009). It was developed by a review of treatment files of stuttering clients who had undertaken psychological treatment at a major anxiety clinic in Sydney, Australia. Sixty-six items reveal the negative thoughts experienced by such clients, such as "people focus on every word I say," "people will think I'm stupid if I stutter," and "I always embarrass the people I'm speaking too." The UTBAS scale has been shown to positively correlate with generic scales of social anxiety such as the Fear of Negative Evaluation scale, but has the advantage of being specifically designed to assess the mental health of those who stutter. The second scale to be published using this approach is the UTBAS-6 (Iverach et al., 2016) This scale contains six items from the original UTBAS scale that correlate well with the full scale. Available clinical data using the UTBAS-6 allow speech-language pathologists to compare an individual's severity of negative thinking with other treatment-seeking clilents.

The use of these scales is recommended because the speech-language pathologist can directly assess the extent to which any given case involves the thinking style of those who have previously been referred for psychological treatment or sought treatment for their stuttering. Notably, the UTBAS-6 provides percentile ranks allowing the speech-language pathologist to compare the individual client with other clients who stutter. As such, the use of the UTBAS-6 promotes open conversations with clients about their mental health, which is a topic that has for too long been simply ignored in clinical practice.

Questions from discussion leaders

Can reliving a traumatic situation that has caused distress to a client be harmful?

Talking to a health professional about past encounters that have caused distress is commonplace in mental health services, and dealing with negative intrusive memories is a standard part of CBT for anxiety-related problems. Exposure to past trauma is a standard part of CBT interventions for a variety of conditions. In general, this type of exposure has been shown to reduce anxiety and distress. Having said this, some individuals experience intrusive memories of past traumas in the presence of everyday cues in the environment. These intrusions into everyday life can temporarily increase anxiety and interfere with social performance.

Behavioural experiments are used as part of CBT to treat anxiety. What do behavioural experiments involve for people who stutter?

Behavioural experiments are designed to test the negative predictions about the world that anxious people make. These vary from person to person. Fears may be about different things: "the sales assistant will laugh at me," "I won't be able to get my point across," "the other person will hang up on me," "people will move away from me at the party." In CBT, the client who stutters is trained to identify the feared outcome and then test out the prediction by confronting the feared situation. After the behavioural experiment, the client is trained to write a brief summary of what actually occurred, such as whether the sales assistant actually laughed. In this way, the client is actively involved in collecting evidence for and against fearful predictions.

Can an online CBT program treat anxiety in stuttering as effectively as a face-to-face psychologist?

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As described in my presentation, the CBTpsych program appears to produce outcomes that are similar to those obtained in face-to-face CBT. Of course, randomised trials that directly compare the two ways of presenting CBT are needed to clarify the effectiveness of CBTpsych.

Do you think all these mental health problems will be obviated if stuttering is controlled effectively during the preschool years?

Unfortunately, there are no data that speak to this question. Having said this, there is no theoretical reason to suggest that the effects of negative social conditioning on an individual would disappear once the stutter is treated. That is, if the preschool child has had punishment experiences from peers, such as other children walking away from stuttered utterances, the effects of those experiences would remain. For this reason, removing stuttering as soon as possible after onset should be a clinical priority. Early intervention should certainly help minimise negative social conditioning events in a child's life. It is my view that delays in treatment of preschool children who stutter need to be avoided.

You made much of research about the negative effects of anxiety on speech restructuring, but that seems all to be based on one research publication. Is it too premature to be making so much of this?

This is an interesting point. In general, the confidence one can have in a research finding increases with the quality of the research itself, and replication of the finding. The publication that demonstrated the negative impact of mental health disorders on speech restructuring can be considered a high quality paper because (1) the sample size was large, (2) psychiatric diagnoses were made using a reliable, structured interview, (3) the speech-language pathologists involved in treatment were blinded to psychiatric status, (4) the stuttering severity data collected were from beyond-clinic phone calls that were recorded, and (5) the research staff involved in the collection of stuttering severity data were blinded to psychiatric status. Unfortunately, while replication from an independent research group would be ideal, it is possible that this may never occur. Large, well controlled studies in the field of stuttering are rarely replicated. Accordingly, speech-language patholoists may have to base their clinical decisions on this single report for many years to come.

We would like to pursue your response to the last question a little further. It may in fact be a long story, as you have outlined. But is the end of the story that, indeed, clinicians should not make too much of that one report?

Evidence-based decisions need to be made on the best available data. As I've said, the existing study can be regarded as a high quality paper. While it is possible that future studies might produce data that temper conclusions drawn from the present study, I wouldn't assume that this will be the case. I would encourage clinicians to act conservatively, and openly discuss the available data with their clients. I believe there is an ethical responsibility to inform clients who stutter that current data suggest that mental health disorders are associated with relapse.

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