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WEBCAM DELIVERY OF THE LIDCOMBE PROGRAM FOR PRESCHOOL CHILDREN WHO STUTTER: A RANDOMISED CONTROLLED TRIAL

Kate Bridgman

A thesis submitted in fulfilment of the requirements
for the degree of Doctor of Philosophy

March 2014

AUTHOR'S DECLARATION

I, Kate Bridgman, certify that this thesis, Webcam Delivery of the Lidcombe Program for Preschool Children who Stutter: A Randomised Controlled Trial, is my own work. It is based on original data gained from my own research. It contains no material that has been written or published by another person, except where acknowledgement is made. This work has not been submitted or accepted for the award of any other degree or diploma. All research practices involved with this thesis were approved by the University of Sydney and La Trobe University Human Ethics Committees.

Kate Bridgman

ACKNOWLEDGMENTS

Perhaps the greatest irony of this whole process has been my distance candidature. Like the webcam families in this RCT, I too have been reliant on Skype and its ability to allow me to work with my supervisors. Professor Mark Onslow. How humbling it has been to work so closely with you. You have pushed me further than I ever thought I could be pushed, and have challenged every fibre of my being. Thank you for seeing abilities in me that were far greater than what I could see in myself. Thank you for your patience, persistence and discipline in your teaching me to write, word by word, sentence by sentence, and finally chapter by chapter. I feel in recent months we have been co-parents of this thesis, with me only ever sleeping through the night when my precious document was with you.

Dr Sue O'Brian, working with you over Skype is how I knew we could expect rapport with our families to be the same as in clinic. I will always be grateful that you were a Skype call away. And Dr Sue Block, the ultimate clinical researcher or perhaps researching clinician. You impressed me as an undergraduate and were such a large part of the reason I agreed to this project. I am lucky to have worked with you as a mentor and now a colleague. Thank you to Mark Jones who provided valuable statistical support, your patient explaining was most appreciated.

To my thesis; I cannot imagine my life without you. For over 3 years you have been my sole focus. You have travelled with me around Europe, New Zealand, Bali and Australia. You have excited me, challenged me and had me search for discipline and resolve. I know you will never be perfect. I know that you will never be able to reflect everything that I have seen and learned during our time together. But as I write this, the time has come to let go and accept you as you are.

Finally, my greatest thanks go to the families who let me into their lives and trusted me with their precious preschoolers. Together we mastered Skype and learned to play in a virtual space. Thank you for seeing promise in this project, for giving it a go and welcoming me into your homes each week. Who knows where the future of webcam telehealth will lead, but let's hope we have started a process that will allow other preschoolers to be treated live in their homes, wherever they may live.

PAPERS AND PRESENTATIONS ARISING FROM THIS THESIS

Conference Presentations

Bridgman, K., Onslow, M., O'Brian, S., & Block, S. (2011). Lidcombe Program procedures have changed: The new ones. Paper presented at the 8th Asia Pacific Conference on Speech, Language and Hearing, Christchurch, New Zealand, January 2011.

Bridgman, K., Onslow, M., O'Brian, S., & Block, S. (2012). Preschool Telehealth: Treating children who stutter using the internet and webcam. Paper presented at the 7th World Congress of Fluency Disorders, Tour, France, July 2012.

Bridgman, K., Onslow, M., O'Brian, S., Block, S., & Jones. M. (2013). Lidcombe Program via Skype: Could you use it in your clinic? Paper presented at Speech Pathology Australia National Conference, Queensland, June 2013.

Journal Article

Bridgman, K., Onslow, M., O'Brian, S., Block, S., & Jones. M. (2011). Changes to stuttering measurement during the Lidcombe Program treatment process. *Asia Pacific Journal of Speech, Language, and Hearing, 14* (3), 147-152.

Non-peer-reviewed Publications to which the Candidate has Contributed Include:

Bridgman, K., Onslow., M & Jones., M. (2010). Important Lidcombe Program manual changes. *Lidcombe News, 38*, 10-17.

Bridgman, K. (2012). Research in progress: Telehealth trial of the Lidcombe Program. *Lidcombe News,42*, 11-14.

Bridgman, K. (2011). Webcam trial of the Lidcombe Program. *Speakout.* pp23.

Packman, A., Onslow, M., Webber, M., Harrison, E., & Bridgman, K. (2010). The Lidcombe Program of early stuttering intervention treatment manual, from http://sydney.edu.au/health_sciences/asrc/docs/lidcombe_program_manual_2010.pdf

Packman, A., Onslow, M., Webber, M., Harrison, E., Lees, S., Bridgman, K., et al. (2011). The Lidcombe Program of early stuttering intervention treatment guide, from http://sydney.edu.au/health-sciences/asrc/docs/lidcombe_program_guide_2011.pdf

CO-AUTHOR PUBLICATION STATEMENT

As co-authors of the publications arising from this thesis (see previous section) we confirm that Kate Bridgman has made the following contributions to all the publications: conception and design, analysis and interpretation of the findings, preparation of the manuscripts and critical appraisal of contents for all publications, and submissions of manuscripts for publication.

Signed: Date: 26 September 2013

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ABSTRACT

Early intervention provides children who stutter with the best opportunity to avoid the lifelong complications associated with stuttering. Access to effective treatment, in particular, the Lidcombe Program, provides preschool children with the best chance to overcome their stuttering. Currently many children are unable to access such efficacious treatment due to distance and lifestyle factors. One solution to this problem is to deliver the treatment via webcam over the internet. This service delivery model was designed to increase access to timely, best-practice intervention for those who are currently unable to access treatment. That model was thought to be able to produce efficiency rates similar to those of traditional clinic treatment. Further, it provides a method of service delivery that: (1) improves access to evidence-based best-practice stuttering treatment for children, (2) improves access to specialist speech pathologists and quality services, (3) reduces costs and resources involved with outreach service provision, (4) provides more convenient home-based treatment for young children, and (5) ensures more equitable service delivery for rural and remote preschool children and their families.

A Phase I study showed that webcam delivery of the Lidcombe Program was a viable treatment delivery model (O'Brian, Smith & Onslow, 2012). This thesis further investigates delivery of the Lidcombe Program for preschool children using the internet and a webcam. The modification in this project, compared to previous, low-tech telehealth (phone and mail) trials of the Lidcombe Program, allowed the principles of standard delivery of the Lidcombe Program to remain relatively unchanged. This was due primarily to the use of a webcam and live videoconferencing. The speech pathologist-parent-child triad was preserved, with all parties having clinic contact. Real-time measurements, observation and education for parent implementation of the program were also achieved through this medium. Thus, treatment could be delivered mostly in accordance with the program treatment guide (Packman et al., 2011, p. 1).

The design for this project was a parallel, open plan, Phase III noninferiority randomised controlled trial (RCT). The control group received standard delivery of the Lidcombe Program (Packman, et al., 2011) in a traditional clinic setting. The experimental group received the Lidcombe Program within their homes using a computer, a webcam, the internet and a live video calling program (Skype).

The primary outcome measures – the number of consultations and speech pathologist hours to attain entry into Stage 2 – evaluated treatment efficiency. The secondary outcomes – stuttering reduction as measured by parent evaluated severity ratings, investigated treatment efficacy, as did quantitative and qualitative data obtained from parent questionnaires. The number of weeks to attain Stage 2 entry was also measured.

Initially, 66 children were assessed for this trial. Eleven were ineligible and six withdrew during the assessment process, with 49 participants being randomised. Of these, 24 were assigned to the control arm and 25 to the experimental arm.

Due to time restrictions associated with the student's candidature, not all 18-month data were collected in time for inclusion in this thesis. Pretreatment data are reported for all 49 participants. Data for all 43 participants active in the trial 9 months postrandomisation are also reported. Stage 2 entry data are available for the 35 participants (71% of the total cohort) who reached Stage 2 by December 31st 2012.

Results for both groups showed no significant difference between the number of consultations and the number of weeks to Stage 2. Efficacy measures showed no significant difference between the groups in stuttering reduction. A further secondary outcome measure was parent responses to a questionnaire at entry into Stage 2. Similarly, there was no significant difference between the two groups when asked about speech pathologist-child rapport, speech pathologist-parent rapport, ease of learning treatment, severity ratings and ability to adapt treatment. Further, two-thirds of clinic families said they would choose webcam treatment in the future.

Webcam parents reported no difficulty in seeking out their own resources and did not feel treatment within their home was invasive. Webcam families listed convenience and comfort as the main advantages of webcam treatment, with technical difficulties as the main disadvantage. All webcam families would choose this same method for future stuttering treatment.

The thesis concludes with a discussion of the speech pathologist's role, consultation logistics and additional qualitative observations from the webcam group. These include convenience, treatment readiness, defining clinical space, trends in clinical transfer, clinical application, limitations and future directions. Overall, this thesis demonstrates that the findings from a Phase III RCT investigating the efficiency and efficacy of stuttering treatment for preschool support the use of webcam and internet to increase access to timely and appropriate stuttering intervention.

The potential for community translation of these findings is considerable; children as young as 3 years of age can receive the same stuttering treatment within their homes as they would within a clinic; they can expect no difference in outcomes or experience. This is significant given that children as young as 2 years of age can be negatively affected by their stuttering. No longer do children who stutter need to be disadvantaged by where they live or by the skills of the closest speech pathologist. They can now access evidence-based treatment within their homes.

ABBREVIATIONS

%SS	Percentage of syllables stuttered
RCT	Randomised controlled trial
SRs	Severity ratings

CONTENTS

PART 1: INTRODUCTION

CHAPTER 2: OVERVIEW OF EARLY STUTTERING INTERVENTION

CHAPTER 3: EQUITY AND ACCESS TO PAEDIATRIC SPEECH PATHOLOGY SERVICES

CHAPTER 4: TELEHEALTH

PART 2: METHOD

CHAPTER 5: TRIAL DESIGN

PART 3: RESULTS

CHAPTER 6: RESULTS

PART 4: DISCUSSION

CHAPTER 7: DISCUSSION OF RESULTS

CHAPTER 8: CLINICAL TRENDS

CHAPTER 9: TRANSLATIONAL CONSIDERATIONS

CHAPTER 10: TRIAL STRENGTHS, LIMITATIONS AND FUTURE DIRECTIONS

APPENDICES

LIST OF FIGURES

LIST OF TABLES

PREFACE

The Phase I trial of webcam delivery of the Lidcombe Program, as detailed in Chapter 4, was designed by Dr Sue O'Brian and was conducted by Ms Kylie Smith, through the Australian Stuttering Research Centre. This Phase I trial was a feasibility study, investigating the use of webcam and the internet to deliver the Lidcombe Program to preschool children within their homes. The manuscript of that study is currently under review (O'Brian, Smith, & Onslow, 2012). Personal communications with the authors allowed the candidate to read this manuscript to access the study design, methodology, results and discussion. Such access provided justification for the candidate to embark upon the Phase III study as detailed in this thesis.

The Phase III RCT, as detailed in Chapter 5, was designed by Professor Mark Onslow and Drs Sue O'Brian, Susan Block, Mark Jones and the candidate. The parent questionnaires were designed by Mark Onslow, Sue O'Brian, Susan Block and the candidate. Dr Mark Jones designed the randomisation schedule. The trial received ethics approval from the University of Sydney and La Trobe University. The project was funded under the NHMRC Program grant (#402763) titled "Equitable access to stuttering treatments: Developing distance and self-managed treatment models".

The candidate attended a Lidcombe Program Trainers Consortium Lidcombe Program Workshop before commencement of the trial. All treatment was delivered by the candidate as per *The Lidcombe Program Early Stuttering Treatment Guide* (Packman, et al., 2011). Beyond-consultation recordings and questionnaires were completed by research assistants to ensure lack of bias. All data were analysed and interpreted by the candidate, with assistance from Dr Mark Jones.

All guardians of the participants consented to their data being collected and analysed, with the knowledge it might be reported at conferences or in publications. There was no charge for the treatment provided to participants.

This thesis is the original work of the candidate. It has not been submitted to any other institution.

PART 1: INTRODUCTION

CHAPTER 1: STUTTERING

Overview

"Fifteen million of our fellows throughout the world, one million in our own land, speak with words whose wings are broken" (Johnson, 1955, p. 83). Johnson opened his seminal work with this striking and confronting statement. Today, over 50 years later, his words still ring true worldwide.

Defining Stuttering

At a global level, the World Health Organization describes stuttering as a speech disorder that disrupts the rhythm of speech. Disruptions may be in the form of repeated or prolonged sounds, syllables or words. They may also include hesitations or pauses (World Health Organization, 2010).

Identifying and Describing Stuttering

There are two main methods for stuttering identification: behavioural and perceptual (Onslow & Packman, 2007). The former method incorporates a list of behaviours, such as "part-word repetitions", "dysrhythmic phonation" and "tense pauses". Johnson (1959) prepared the first of such taxonomies which were subsequently adapted by others in research and clinical contexts (Onslow, 1996).

The perceptual method was initially formulated by Bloodstein (1990). He proposed that stuttering was whatever an observer perceived it to be, provided the listener shared reasonable agreement with others (p.392). Onslow and Packman (2007) have argued that many studies with preschool children who stutter as participants have, in effect, amalgamated the behavioural and perceptual methods of identification. Researchers in the studies concerned became aware of potential participants because of parent perception and their consequent referral. Stuttering status was confirmed subsequently with a behavioural taxonomy. Further, it might be argued that speech pathologists also amalgamate the two methods; parents come with a belief that their child is stuttering, and subsequently the speech pathologist draws on his or her previously acquired behavioural taxonomy for confirmation.

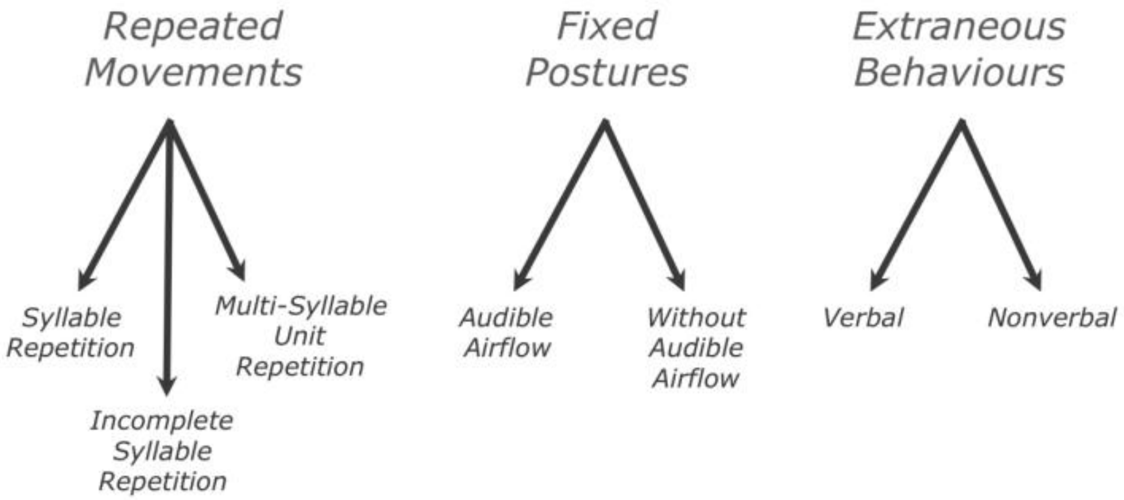
Categorising Stuttering

Many taxonomies have consisted of a combination of behavioural and nonbehavioural descriptions. The Lidcombe Behavioural Data Language (LBDL) was developed to describe stuttering behaviours, avoiding the ambiguity of existing taxonomies that were to some extent nonbehavioural (Teesson, Packman, & Onslow, 2003). There are three categories of stuttering within the LBDL: repeated movements, fixed postures and superfluous behaviours. Each category is comprised of two or three descriptors. Repeated movements consist of syllable repetitions, for example "I-I-I like" and "um-um-umbrella"; incomplete syllable repetition, for example "u-u-u-umbrella" and multisyllable unit repetitions, for example, "umbre-umbre-umbrella," where multiple syllables are repeated. Thus, this taxonomy distinguishes among the various forms of repetition.

The fixed posture category refers to any stutter in which the articulators stop moving. The two descriptors in this category refer to airflow. The descriptor "fixed postures with airflow" describes prolongations or the stretching out of a sound as in "liiiiiiiiiiiiike". "Fixed postures without airflow" refers to static articulatory movement with no audible airflow. This has been commonly referred to as a block (Van Riper, 1971).

Finally, the superfluous behaviours category refers to additional or nonspeech behaviours that occur in association with stuttering. These include additional behaviour that is not part of the intended message. This may be verbal: for example filler words like "um" or other generic phrases. It can also be nonverbal, such as facial, body or limb movements. This category encompasses features that have in the past been referred to as secondary behaviours or associated characteristics (Prasse & Kikano, 2008).

Figure 1.1. The Lidcombe Behavioural Data Language. Adapted from Teesson, K., Packman, A., & Onslow, M. (2003).



Prevalence and Incidence

Today, there is consensus that the prevalence of stuttering is 1% (Bloodstein & Bernstein Ratner, 2008). That implies more than 70 million children, adolescents and adults worldwide who stutter. The impact of the disorder is even greater in terms of lifetime incidence, which is currently considered to be around 5% (Yairi & Ambrose, 2005). Bloodstein and Bernstein Ratner (2008) considered this estimate to be conservative. They concluded a review of the issue by stating, "it would seem that a plausible figure for the lifetime incidence of all those who at some time either consider themselves or are considered by their parents to be stutterers is at least as high as 10 percent" (p. 91). Based on current population figures, this would equate to over 2.3 million Australians and nearly 680 million people worldwide. Such estimates of incidence and prevalence are confronting and concerning. They highlight just how common this disorder is and, more importantly, how many people might require intervention.

A more precise estimate of stuttering incidence can be obtained from community cohorts ascertained prior to stuttering onset. Using such a method, Reilly et al. (2009) reported a 36-month cumulative stuttering incidence of 8.5% for a cohort of 1,910 young children. This suggests that the prevalence and incidence of early stuttering may indeed be higher than previously thought.

Onset

Bloodstein and Bernstein Ratner (2008) reviewed reports of stuttering onset according to parent recall. That data-set contains 10 reports of reported mean onset, with a grand mean of 3.3

years, and four reports of median onset of 3.9 years. It appears, then, that stuttering typically begins some time during the fourth year of life.[1]

Stuttering can be highly variable following onset (Onslow, 2004; Packman & Attanasio, 2004). There can be a gradual emergence of stuttering behaviours over days, weeks or months. Conversely, stuttering onset can occur more suddenly, in a matter of hours or days (Reilly et al., 2009; Wyatt, 1969; Yairi & Ambrose, 2005). Yairi and Ambrose, for example, reported that 40% of children had an onset reported to occur over 1-3 days. A further 33% of children were reported to have started stuttering during 1-2 weeks. Onset was reported for the final 27% of children as gradual; occurring between 3 to 5 weeks; and 6 or more weeks.

The most common features of early stuttering appear to be repeated movements (Ambrose & Yairi, 1999; Andrews & Harris, 1964; Reilly, et al., 2009; Van Riper, 1982; Wyatt, 1969). These behaviours may change in type and frequency, even disappearing for periods (Packman & Attanasio, 2004; Yairi & Ambrose, 2005). The somewhat immediate onset with varying severity is a stark contrast to other speech and language disorders, as there is no forewarning of stuttering onset (Onslow, 2004; Packman & Attanasio, 2004; Yairi & Ambrose, 2005). By comparison, children with articulation difficulties or language delay demonstrate these features from the beginning of their speech and language history. Further, neither the presence of such disorders nor their severity fluctuates. Conversely, a child who stutters may have spoken with an intact system for some time before the speech disorder begins. Thus, the parents are used to hearing children speak fluently, until stuttering onset.

Natural Recovery

Following onset, some preschool children continue to stutter, and some stop without formal intervention. There have been different estimates of the rate of natural recovery during the preschool years, based on prospective methods. The first of these was from the 1000-family study in the United Kingdom (Andrews & Harris, 1964) that identified 43 stuttering children, of whom 18 (42%) reportedly recovered by 6 years. Mansson (2000) observed 1,021 Danish preschoolers. During their 3-year old screening, 51 preschoolers were identified to be stuttering. Only 15 of these preschoolers were identified as still stuttering at 5 years of age. That is, 71% were thought to have recovered by 5 years of age. In contrast but with similar findings, Yairi and Ambrose (1999) used a clinical cohort of 84 children who stuttered, 74% of whom were reported to have recovered during a 4 year period post-onset.

Regardless of what the natural recovery rate may be, there is doubt about whether it is truly natural. Ingham (1983) argued that children who stutter live in environments that contain potentially helpful input about their stuttering. In particular, Ingham cited evidence that spontaneous verbal contingent stimulation by parents can occur. As discussed in Chapter 2, there is good reason to believe that the latter can change or at least influence early stuttering.

Suggested clinical responses to the existence of natural recovery have changed markedly during past decades. Andrews' (1984) position was that intervention with preschool children is likely to be cost-ineffective and should be avoided. However more recent practices favour early intervention (Jones et al., 2005; Langevin, Packman, & Onslow, 2009; Langevin, Packman, & Onslow, 2010). This is further supported with the emerging knowledge of the mental health issues associated with

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[1] Naturally, this excludes cases of what are considered by some to be late onset stuttering of neurogenic origin (Van Borsel & Taillieu, 2001; Lebrun, Leleux, & Retif, 1987).

chronic stuttering, and the possibility that they begin early in life (Iverach, Menzies, O'Brian, Packman & Onslow, 2011).

Impact of Stuttering

For preschool children who do not experience natural recovery, or who are not treated effectively, stuttering can continue indefinitely. When untreated, stuttering becomes a chronic condition that can have significantly impact across social, educational, occupational and wellbeing domains of life.

Preschool Children (younger than 6 years)

Parent Reports

Stuttering is reported to affect preschool children who stutter and their parents. Due to the children's age and limited ability to self-report, parental reports of observations and perceptions of their child's speech are common (Bernstein Ratner, 1997; Cooper & Cooper, 1996; Langevin, Packman, & Onslow, 2010; Yairi, 1983). Beginning from onset, the majority of parents report that their children exert force and tension when they speak (Yairi, 1983). For themselves, parents of children who stutter report concern and distress over the disorder (Bernstein Ratner, 1997). Further, a significant number of speech pathologists have reported that parent counselling of preschool children is critical in early intervention (Cooper & Cooper, 1996).

Children as young as 2 years of age are reported to be affected by their stuttering. Yairi (1983) interviewed parents of 22 children who stuttered, aged between 2-years 1-month and 3-years 7-months. Most parents reported that their children's stuttering behaviours were associated with "some degree of tension and force" (Yairi, 1983, p. 176). Further, four of the children (18%) were reported to be bothered by their stuttering.

More recently, in a study of parent perceptions of the impact of stuttering on children, Langevin and colleagues (2010) reported that of the 77 parent participants, 81.8% reported noticeable frustration in their children, 42.9% reported negative impacts on mood, 34.2% reported decreased self-confidence in their children, 25% reported that their children communicated less since stuttering onset and 23.4% reported that their children withdrew from communicating. Parent reports of the social impacts of stuttering in preschool children were also investigated, with 27% of parents reporting that their children were teased by their peers (Langevin, et al., 2010). Parents also reported that in social situations with peers, their children were not given time to finish speaking, their sentences were interrupted or completed by others, and their peers walked away while their children were stuttering.

Peer Reactions

The direct impact of stuttering on preschool children was also explored by Langevin et al. (2009) who recruited four preschool children who stuttered. Peer responses to incidences of their stuttering were covertly video-recorded. Although the majority of peer responses to episodes of stuttering were found to be neutral or positive, there were negative social consequences in response to the stuttered speech. This was evident in one participant who received negative peer responses to 28% of his stuttered utterances. These responses included reduced efficacy and ability of communication, and lost meanings of stuttered utterances. Further difficulties experienced by the children who stuttered concerned retaining the attention of a listener, leading peers in play, resolving conflicts, and participating in class discussions and pretend-play scenarios with peers.

The development of children's awareness of stuttered speech has also been investigated (Ezrati-Vinacour, Platzky, & Yairi, 2001). A group of 79 children who did not stutter were asked to discriminate between the speech of two puppets, one who demonstrated stuttered speech and one who did not. The results showed that children as young as 3 years of age could discriminate. This discrimination, and therefore identification of different speech patterns, namely stuttering, increased with age. Most 5-year-old participants were able to discriminate accurately. While the children could discriminate between stuttered and stutter-free speech, they could not label stuttered speech as stuttered. By the age of 4 years, however, the majority of participants identified stuttered speech as a "not good" speech pattern and selected the nonstuttering speaker as their choice of preferred friend. This finding is significant in terms of the potential social isolation that preschool children may begin to experience when in peer groups of children who do not stutter.

Communication Attitudes

Communication attitudes of preschool children have been investigated using the KiddyCAT questionnaire (Cardell, 2010). The KiddyCAT (Vanryckeghem, Brutten, & Hernandez, 2005) is a 12-question test that elicits yes or no responses from children in relation to six positively worded and six negatively worded questions about their speech. This self-report tool was administered to children aged between 3 and 6 years of age.

KiddyCAT outcomes were reported for the 63 children who stuttered and 43 same-age children who did not. A statistical significance was found in the communication attitudes of the children who stuttered compared to those of their nonstuttering peer group (Cardell, 2010).

Anxiety

To date there are no peer-reviewed publications investigating or reporting anxiety in preschool children related to their nonstuttering peers. Such a study would require standard psychological measurement (Onslow, 2013). A recent study reviewed the literature related to temperament and early stuttering (Kefalianos, Onslow, Block, Menzies, & Reilly, 2012). The authors reviewed 10 studies and concluded that further longitudinal studies were required to determine a link between anxiety experienced in early stuttering and temperament.

Primary School Children (7-12 years)

As children who stutter progress to the primary school years the disorder becomes more complex than in earlier childhood (Conture & Guitar, 1993). It is assumed that school-aged children who stutter have limited chance of natural recovery. This is thought to be because the neural pathways become more established with age (Wohlert & Smith, 2002). The implication, or at least an interpretation of this is that it results in the disorder becoming less tractable. It also implies that treatment may need to be more complex or last longer. The phenomenon of relapse may indicate that treatment merely temporarily controls stuttering rather than actually facilitating the repair of neural pathways.

Peer Reactions

As children who stutter grow older it is likely that their stuttering features worsen, becoming more obvious (Craig, 2005). Whether the disorder has been treated or left untreated, school-aged children who stutter can be increasingly aware of how their speech differs from that of most of their peers (Cook & Botterill, 2005). Their awareness of peer reactions and the potential for negative peer reactions and bullying increase significantly. It is common, therefore, for this age group to develop avoidance strategies or learn to minimise or disguise their stuttering (Cook & Botterill, 2005). Further, peer reactions can impact negatively on treatment outcomes and attendance (Leith, 1984).

Children who stutter may withdraw from or reject treatment if their peers have a negative view of such intervention (Leith, 1984). Thus, whereas increasing severity and social implications can motivate some children to address their stuttering, peer reactions to treatment may affect some children's treatment motivation or persistence.

Social Implications

School-aged children who stutter may also experience social difficulties. In order to be accepted by their peers, school-aged children who stutter are more likely to limit their communication in an attempt to reduce their chance of stuttering. This, in turn, may increase their feeling of being different or their perception that they will be less popular or targeted by bullies. This can result in their appearing withdrawn or shy (Davis, et al., 2002; Hugh-Jones & Smith, 1999). Research indicates that stuttering places school-aged children at a significantly higher risk of being bullied or teased than their nonstuttering peers (Langevin, Bortnick, Hammer, & Wiebe, 1998). Such teasing and bullying are reported to have a direct effect on how children who stutter perceive themselves, causing them to feel upset. Further, there is a strong correlation reported between childhood bullying leading to anxiety in adulthood (Gladstone, Parker, Gordon, & Malhi, 2006; McCabe, Antony, Summerfeldt, Liss, & Swinson, 2003; Roth, Coles, & Heimberg, 2002).

Although not all school-aged children who stutter are bullied, their peers do consider them to be different from their nonstuttering peers (Davis, et al., 2002; Langevin, et al., 1998). The impact of stuttering on the social status and behaviour of school-aged children was explored by Davis, Howell and Cooke (2002). They concluded that, compared to nonstuttering controls, children who stutter are more often rejected by their peers, are less likely to be among the average social grouping, are less likely to be nominated leaders and more likely to be the victim of bullying. Such increased chance of being subject to bullying was further supported by Langevin et al. (1998), who reported that the incidence of bullying of children who stuttered was significantly higher than the incidence in the general school population. The negative impact for both the stuttering victims and their parents was also confirmed.

Bullying during the school years has been associated with stuttering for decades. In a retrospective study, Hugh-Jones and Smith (1999) surveyed 276 people who stuttered. Given that the age of respondents ranged from 7 to 75 years of age, experience of bullying at school by this group could date back to the 1920s. The incidence of bullying was reported to be 83%, with nearly one-fifth of respondents recalling daily bullying and over two-thirds recalling weekly bullying across their school years. The respondents reported avoiding verbal communication as a short-term effect of their bullying experiences. A further 46% reported long-term, lasting effects of bullying on their ability to form relationships. This ability to form friendships proved to be a significant variable in determining the severity of bullying.

Communication Attitudes

Children who stutter have more negative attitudes towards communication than their nonstuttering peers (De Nil & Brutten, 1991). In fact, children as young as 7 years of age can have a negative opinion of their ability to communicate. This stuttering group is thought to become increasingly self-aware of their problematic communication skills with age (De Nil & Brutten, 1991).

Self-Esteem

Not all school-aged children who stutter have reduced self-esteem. Yovetich and Leschied (2000) investigated the self-perception and self-esteem of 25 school-aged children who stuttered, aged between 7 and 12 years. This subset of children was found to possess average or above average self-esteem. Further, they were found to be no more defensive than their same-aged peers. Thus, for some children at least, stuttering itself may not always be the direct cause of reduced self-esteem. Rather, it may be the bullying or negative peer reactions that are more prevalent in school-aged children who stutter that subsequently affect their self-esteem.

Anxiety

Anxiety in school-age children has been investigated in two studies. Craig and Hancock (1996) administered the State and Trait Anxiety Inventory to 96 children who stuttered and 104 who did not. Children were aged between 9 and14 years. Thus the participants included some early adolescents. Results indicated no significant difference between the groups in relation to state or trait anxiety. Blood and Blood (2007) administered the Revised Children's Manifest Anxiety scale to 18 children who stuttered and 18 children who did not stutter. The children who stuttered were more likely to be bullied than their nonstuttering peers. In relation to anxiety, although more children who stuttered scored one standard deviation below the mean when compared to their controls, the scores themselves were not significant.

Adolescents and Adults (older than 17 years)

As people age, their stuttering is likely to become more complex and severe (Craig, 2005). The likelihood of natural recovery for adolescents and adults is significantly less than that for young children (Cook & Botterill, 2005), leaving this older group to manage a chronic condition and all its associated implications.

Adolescents (12-17 years)

In adolescents, sense of self develops as adulthood approaches (Rustin, Cook, & Spence, 1995). During this transitional phase between childhood and adulthood, there can be many pressures from peers, teachers, parents and society (Rustin, et al., 1995). Moreover, adolescents who stutter may perceive themselves to be poor communicators, resulting in lowered self-esteem (Blood & Blood, 2004).

Stuttering can place adolescents at risk of being bullied (Blood & Blood, 2004). The impact of peer rejection and bullying on adolescents who stutter can be lifelong. Peer acceptance and relationships contribute to adolescent personal identity, social belonging and status (Blood & Blood, 2004). Further, adolescents who stutter perceive themselves to have poor communicative competence (Blood & Blood, 2004).

The perceptions of adolescents and young adults who stutter have been investigated (Hearne, Packman, Onslow, & Quine, 2008). Thirteenparticipants were involved in focus group interviews about their adolescent experiences of stuttering. The groups reported that their friends were usually accepting of their stuttering. Despite this acceptance, adolescents who stuttered reported they did not want to discuss their stuttering with their peers. However, 15% of participants reported experiencing stuttering-related bulling during their adolescence. Other participants agreed that bullying and teasing had occurred during their primary school years. Additional trends that emerged were interest in providing schools with education about stuttering and lack of insight and understanding from parents.

Treatment motivation also emerged as a theme. The group agreed that the individual had to be ready to seek treatment and that there were often additional motivators to seek treatment. These included entry to university or seeking employment. They also reported enjoying group treatment sessions which provided some social connectedness with like peers.

Adults

Adults who stutter report feeling different from their nonstuttering peers, identifying their stuttering as a negative behaviour (Cream, et al., 2003). Cream et al. interviewed 10 adults who stuttered about their experiences following speech restructuring treatment. Several key themes were apparent from these interviews. First, the adults who stuttered experienced a need to protect themselves from situations with a high risk of stuttering. They did this by employing avoidance strategies that could include avoiding specific communication situations, topics of conversations or words. Second, despite attending therapy and experiencing some success, the adults might still have negative feelings about their stuttering. Third, the adults who stuttered were acutely aware of their listeners' reactions during a communication exchange. Fourth, when using speech restructuring strategies to control stuttering, the adults who stuttered still felt different in comparison to their nonstuttering peers. Further, they felt different from their former selves, as the use of their techniques might not allow them to express themselves as they would have previously.

Stuttering also appears to affect expressive language production in adults (Cream, et al., 2003; Spencer, Packman, Onslow, & Ferguson, 2009). Spencer et al. (2009) investigated the effect of stuttering on adult communication. The authors compared language use between 10 adults who stuttered and 10 who did not. Their results indicated that stuttering affected the language production of the adults who stuttered within this study. The experimental subjects produced shorter utterances with less syntactic complexity than their nonstuttering controls. They also demonstrated decreased modality use. Modality refers to the way speakers use language to communicate their opinions, attitudes, politeness, and how they engage with their communication partners. A reduction in modality use could limit the expression of a speaker's opinions, interests or attitudes when communicating, for fear of stuttering (Spencer, et al., 2009). Such limiting expression is likely to impact on communicative effectiveness and general participation in social interactions (Spencer, et al., 2009). This could also include a reduction of conversational engagement.

Stuttering also provides a financial burden for adults who stutter (Blumgart, Tran, & Craig, 2010). Blumgart and colleagues investigated the direct personal costs incurred by adults in Australia who stuttered. At least 50% of the participants reported that their stuttering had a negative impact on their employment opportunities. Although the direct cost of this cannot be quantified, it is highly likely these participants felt that their earning capacity was limited by their stuttering. As a direct cost, the mean expense of stuttering was found to be around $5,500 over 5 years. Expenses included treatment for stuttering and stuttering-related conditions, travel, technology and attendance at conferences. The authors added that the cost could be higher if indirect costs such as reduced or lost income related to stuttering were calculated.

Anxiety

There is a higher prevalence of anxiety-related mental health disorders in adults who stutter than in those who do not (Craig & Tran, 2005b; Ezrati-Vinacour & Levin, 2004; Ingham, 1984; Iverach et al., 2009; Kraaimaat, Vanryckeghem, & Van Dam-Baggen, 2002). In a clinical cohort of adults seeking treatment for stuttering, Iverach et al. (2009) found adults who stuttered had between a six and seven times greater chance of having any anxiety disorder than their nonstuttering controls. Further, the likelihood of social phobia was between 16 and 34 times greater, generalised anxiety disorder was four times greater and the likelihood of having a panic disorder was six times greater than in the general population.

Kraaimaat et al. (2002) administered a social anxiety inventory to 89 adults who stuttered and 131 who did not. Higher levels of emotional tension were reported by the participants who stuttered, with more than 50% scoring within the criterion for highly socially anxious psychiatric patients. This group also reported significantly fewer social responses when compared to the control group.

Ezrati-Vinacour and Levin (2004) investigated the types of anxiety experienced by adults who stuttered compared to a control group. With a sample size of 47 men in each group, those who stuttered were found to demonstrate higher levels of anxiety than their controls. Further, the experimental group was found to have higher trait anxiety. There was no relationship between stuttering severity and trait anxiety. Anxiety related to social communication or to specific communication situations was found to be higher in those who had more severe stuttering than in those who had mild stuttering or who did not stutter at all. Craig and Tran (2005) further confirmed the higher levels of trait anxiety exhibited by people who stuttered, adding that it might increase with age.

Educational and Vocational Impact

Educational outcomes and vocational choices may be affected for adolescents and adults who stutter (Cream, et al., 2003; O'Brian, Jones, Packman, Menzies, & Onslow, 2010). O'Brian et al. (2010) reported a negative relationship between educational attainment and stuttering severity. In particular, the subgroup that did not complete high school had a stuttering frequency six times greater than those who completed a postgraduate degree. Further, Cream et al. (2003) reported that following their education, adults who stutter may not apply for jobs that require particular verbal skills.

A recent review was completed investigating how Australian public universities provide information for prospective students who stutter (Meredith, Packman, & Marks, 2012). The website content of all 39 Australian universities was reviewed and found to have very little information relevant to supporting potential students who stutter. Overall, the authors concluded that current information provided by Australian universities was not sufficient to assist prospective students who stutter when considering which university would support their needs and successful participation.

Negative perceptions of stuttering with regard to employment opportunities have been investigated. Klein and Hood (2004) surveyed 232 people aged 18 years and older who stuttered. They reported that over 70% of participants believed their stuttering negatively affected their opportunity of employment or promotion. Further, over one-third cited stuttering as interfering with their ability to complete their job. Nearly 40% thought they would have a better job if they did not stutter. One-fifth cited their stuttering as the reason they had not accepted a new job or promotion, and 50% of participants reported that they were attracted to jobs that required minimal verbal communication.

Employers' attitudes towards stuttering have also been investigated. Hurst and Cooper (1983) surveyed 644 employers. They reported that 85% of employers agreed that stuttering decreases employment opportunities. As a group, however, the majority reported that stuttering did not affect job performance. More than one-third of the employers agreed that people who stuttered should apply for jobs that required minimal speaking. This study supports the perceptions of people who stutter regarding their employment.

Summary

Stuttering is a speech disorder that usually begins between the ages of 2 and 4 years. Early stuttering can affect both the child and the parents. For children who do not experience natural recovery, their stuttering can continue to affect them as they progress through school and adolescence. At school, they have an increased chance of experiencing teasing, bullying and negative attitudes towards communication. During adolescence and adulthood, people who stutter are more likely to experience anxiety-related disorders, and have limited educational and vocational opportunities. Given the potentially dire impact of stuttering on children, adolescents and adults, early intervention is recommended to provide the greatest opportunity to reduce or limit the impacts of stuttering.

CHAPTER 2: OVERVIEW OF EARLY STUTTERING INTERVENTION

This chapter provides a review of clinical trial evidence for the three early stuttering interventions published to date. The intervention with the greatest clinical evidence, the Lidcombe Program, in then reviewed further, including additional non-clinical-trials evidence.

What is Reviewed Here?

Defining a Clinical Trial

When reviewing the literature relating to early stuttering treatment, clinical trials were considered entry level evidence for treatment efficacy. The Onslow, Jones, O'Brian, Menzies, and Packman (2008) definition of a clinical trial was used, and includes: a clinical trial is a (1) "prospective attempt" (p. 404) to (2) determine the "efficacy of an entire treatment" (p. 402) with (3) at least a 3-month follow-up period, with the (4) primary outcome measure based on blinded assessment of beyond-clinic speech samples.

Clinical Trial Development Phases

Clinical trials summarised in this review are grouped by the clinical trials developmental phases, as defined by Robey (2005). Phase I trials are preliminary, feasibility studies, with the purpose of determining a possible treatment, developing a treatment protocol, and determining whether participants comply with the treatment. Phase II trials further develop treatment and protocols, and test outcome measures using a larger sample. Phase III trials are RCTs that explore treatment effects.

The Treatments

Verbal Response Contingent Stimulation

Verbal response contingent stimulation is the application of a verbal contingency to specific speech behaviour. In this context, specific verbal contingencies can be applied to either stuttered or stutter-free speech. In some instances, specific verbal contingencies may be applied to both (Onslow, 2003).

Phase I Clinical Trials

The first clinical trial to apply verbal response contingent stimulation to stuttering exhibited by a child used an ABA design with two preschool-age children who attended weekly 20-minute sessions (Martin, Kuhl, & Haroldson, 1972). This design is now considered to be a clinical trial. A reduction in stuttering severity was evident for both participants during treatment and at around 12 months posttreatment, within and beyond the clinic. A second trial with multiple baselines for two preschool-aged participants also demonstrated stuttering reduction during treatment and 8 months posttreatment (Reed & Godden, 1977). This design would be considered a clinical trial in this context.

A Phase I trial of what was to become known as the Lidcombe Program of Early Stuttering Intervention was conducted (Onslow, Costa, & Rue, 1990) with four preschool participants who maintained significant stuttering reductions 9 months posttreatment, based on beyond-clinic speech recordings. That preliminary study was significant in terms of favourably indicating that further Phase II trials were warranted. Consequently, as detailed below, there have been further clinical trials of the Lidcombe Program. Since 1990, the basic structure of the program has changed, although the key elements have remained the same.

Phase II Clinical Trials

A Phase II trial of the Lidcombe Program was completed (Onslow, Andrews, & Lincoln, 1994) with 11 participants who were randomised to a treatment and no-treatment group. Treatment results were positive, showing a mean of 11.5 sessions to complete Stage 1 of the program and achieve little to no stuttering. Twelve-month posttreatment assessment data indicated that all participants retained their stuttering reductions. These outcomes were not compared to the control participants as the latter withdrew throughout the study.

The next Phase II clinical trial of the Lidcombe Program (Lincoln & Onslow, 1997) was a long-term outcome study of participants from the initial studies (Onslow et al., 1990, 1994) and 34 additional children who had also received stuttering treatment. Participants from the Onslow et al. (1990) and Onslow et al. (1994) study exhibited less than 1.5 percent of syllables stuttered (%SS) across all speaking situations. The second group, who had not experienced previous Lidcombe Program treatment, exhibited a mean stuttering rate of less than 0.5%SS across the same speaking situations. An additional parent questionnaire was also completed by 79 parents of children who had received Lidcombe Program treatment in the past 1-4 years. One-third of the parents reported that their child had exhibited some stuttering in the past year, but only 5% reported that others had commented on the stuttering. The overall data-set from this study supports the claim of long-term outcomes of near-zero stuttering following Lidcombe Program treatment for up to 7 years.

Rousseau et al. (2007) explored language, phonology and the Lidcombe Program. Several independent predictors were identified. First, for every 1% increase in pretreatment stuttering as measured by %SS, the number of sessions to complete Stage 1 increased by 16%. Second, for every 10-point increase in the receptive language score on the Clinical Evaluation of Language Fundamentals (fourth edition), the number of sessions to complete Stage 1 increased by nearly one-third. The authors interpreted this finding with caution, however, reporting that it may be a type one error. Third, for every one unit increase measured in the mean length of utterance assessment, the number of sessions to complete Stage 1 decreased by almost 20%. No remarkable predictors were found to be associated with phonological development in isolation. The combination of phonological development and mean length of utterance did, however, prove remarkable. When combined, 42% of the required treatment time to complete Stage 1 could be attributed to speech and language abilities. Additional findings in this study included the replication of those of Jones et al. (2000) and Kingston et al. (2003), both of which reported pretreatment stuttering severity to be a predictor of Stage 1 treatment time.

The most recent Phase II clinical trial of the Lidcombe Program was a follow-up of 20 participants from the Phase III Jones et al. (2005) RCT who had received treatment (Jones et al., 2008). Participants were reassessed an average of 5 years after their initial randomisation. Of the 20 treatment group participants, 80%, or 16 participants, exhibited zero or near-zero stuttering levels in the speech samples provided. The other 20%, or 4 participants, were considered to have relapsed, as their speech samples were rated to have more than 1.1%SS. Thus, these results suggest two findings: that the long-term outcomes of the Lidcombe Program are favourable, and that a small number of participants experience relapse despite successful Lidcombe Program treatment.

Phase III Clinical Trials

The previously cited Phase III RCT of the Lidcombe Program by Jones et al. (2005) was conducted across two clinical sites in different countries. Fifty-four participants were recruited; 27 participants who received treatment completed the study, compared to 20 participants who did not receive treatment. Comparison of assessment pretreatment and 9 months following randomisation and the commencement of treatment showed the treatment group had a 77% stuttering reduction, compared to a 43% stuttering reduction in the no-treatment group. It should be noted, though, that the data were contaminated by five participants in the no-treatment group who sought treatment during this time. An odds ratio of 7:1 was reported, meaning that children who received treatment were seven or eight times more likely to reduce stuttering to near-zero levels than those who had no treatment.

Four additional Lidcombe Program studies listed in the Onslow et al. (2008) review of clinical trials for preschool children are the telehealth studies. They are reviewed in Chapter 4 (Harrison, Wilson, & Onslow, 1999; Lewis, Packman, Onslow, Simpson, & Mark, 2008; Wilson, Onslow, & Lincoln, 2004; O'Brian, Smith & Onslow, 2012).

Family-Based Therapy

Family-based therapy for stuttering is a multifactorial treatment. In contrast to the previous two treatments which incorporate verbal contingencies for stuttered or fluent utterances, the primary goal of family-based therapy is not to achieve zero stuttering. Rather, the program goals are to increase parents' ability to manage their child's stuttering and increase their fluent speech (Millard, Nicholas, & Cook, 2008). This is achieved by targeting parent communication acts as well as environmental and linguistic factors that might be facilitating stuttered speech (Lasan, 2012). Parent-child interaction is an example of family-based therapy.

Phase I Clinical Trials

The efficacy of parent-child interaction therapy was investigated by Millard et al. (2008) with six preschool participants. Results demonstrated stuttering reduction for four participants during treatment, which was maintained 12 months posttreatment.

Phase II Clinical Trials

A Phase II RCT was conducted with the Palin Parent-Child Interaction therapy (Millard, Edwards & Cook, 2009) using a single study replicated for 10 participants. Participants were randomised to a treatment and a no-treatment group. Four of the six participants who received treatment demonstrated clinically significant stuttering reduction during treatment. They were also reported to maintain these low levels during follow-up assessment that occurred between 7 and 12 months following initial assessment.

Phase III Clinical Trials

There has not yet been a Phase III trial completed or published. Such a trial would need to consider the treatment style and consider which outcomes relate to goals of parent management, rather than use direct measurement of speech outcomes.

Syllable-Timed Speech

Syllable-timed speech is a speech pattern that requires the speaker to say each syllable on the beat of a rhythmic pattern (Trajkovski et al., 2011). It has been used as a treatment for adults (Andrews & Ingham, 1973; Packman, Onslow & Menzies, 2000) and children (Greenberg, 1970; Coppola & Yairi, 1982).

Phase I Clinical Trials

The first report detailing the use of what is now called the Westmead Program was a single case study (Trajkovski, Andrews, O'Brian, Onslow, & Packman, 2006). A child aged 3 years and 2 months achieved near-zero levels of stuttering following seven treatment sessions. The use of syllable-timed speech in this study did not include programmed instruction or a metronome, as used in previous adult studies. This study supported the feasibility of the next Phase I study. A second Westmead Program study was conducted with three preschool children (Trajkovski et al., 2009). The children achieved near-zero stuttering in a mean of 8.6 sessions. Outcome data for these participants were collected just under 3 months posttreatment, but are considered in this review in keeping with Onslow et al. (2008) criteria.

Phase II Clinical Trials

A Phase II study further investigating the Westmead Program has been completed (Trajkovski, et al., 2011) with 17 preschool children. Nine children withdrew from the study during the treatment process. The authors report this is typical of early intervention studies. Eight children required a mean of 12.4 sessions to achieve near-zero stuttering, with a mean clinician time of 8 hours. Further, the near-zero stuttering rates were maintained 12 months following treatment. Posttreatment speech samples were independently judged to be natural sounding.

Phase III Clinical Trials

No Phase III trial of syllable-timed speech has been published to date.

Conclusions

Different Treatment Styles

Clinical trials evidence to date supports three early stuttering interventions to different degrees. Early success with two laboratory studies of verbal response contingent stimulation resulted in the development of the Lidcombe Program, which was found to be efficacious in Phase I, II and III trials in reducing stuttering to near-zero levels with long-term stuttering reductions maintained. Similarly, the use of syllable-timed speech in the Westmead Program has resulted in stuttering reductions, as evidenced by Phase I and II trials. Further outcome data is required to determine the long-term reductions associated with this treatment. In comparison, family-based therapy has limited clinical evidence but reports some stuttering reductions. This reduction is not the main objective of the program, so it cannot be compared to the other two interventions described.

A Treatment with the Greatest Evidence: The Lidcombe Program

Following the clinical trial evidence for the Lidcombe Program, it was selected as the intervention for this RCT. The research suggests it currently has the most evidence when compared to alternative preschool treatments. Additional non-clinical-trial Lidcombe Program evidence is listed below.

Additional Lidcombe Program Treatment Research

As well as the Phase I, II and III clinical trials evidence reviewed earlier in this chapter, many additional studies of the Lidcombe Program have been published. These include:

- a clinical case study (Hayhow, Kingston, & Ledzion, 1998),
- a retrospective file follow-up (Miller & Guitar, 2009),
- retrospective file audits (Jones et al., 2000; Onslow et al., 2002; Kingston, Huber, Onslow, Jones, & Packman, 2003; Koushik, Hewat, Shenker, Jones & Onslow, 2011),

- randomised experiments (Harrison, Onslow, & Menzies, 2004; Franken, Kielstra-Van der Schalk, & Boelens, 2005; Lattermann, Euler, & Neumann, 2008),
- optimal treatment intensity (Packman & Onslow, 2012),
- long-term follow-up posttreatment (Lincoln & Onslow, 1997; Jones et al., 2008(Koushik, Shenker, & Onslow, 2009),
- parent experiences (Goodhue, Onslow, Quine, O'Brian, & Hearne, 2010; Hayhow, 2009; Goodhue, et al., 2010),
- problem-solving (Trajkovski, Andrews, & Packman, 2010),
- clinical translation (Rousseau, Packman, Onslow, Dredge, & Harrison, 2002; O'Brian et al., 2013),
- treatment fidelity (Carr Swift et al., 2011; Swift, O'Brian, Onslow, & Packman, 2012),
- social validity (Lincoln, Onslow, & Reed, 1997),
- psychological impact (Woods, Shearsby, Onslow, & Burnham, 2002),
- speech timing (Onslow, Stocker, Packman, & McLeod, 2002),
- speech and language outcomes (Bonelli, Dixon, Ratner, & Onslow, 2000; Lattermann, Shenker, & Thordardottir, 2005),
- application with school-aged children (Bakhtiar & Packman, 2009; Lincoln, Onslow, Lewis, & Wilson, 1996),
- application with adults (Blumgart et al., 2001).

These studies investigated various aspects of the Lidcombe Program. Findings from these studies have assisted with; (1) further developing clinical benchmarks; (2)informing the program designers about how well the treatment process can be applied to nontarget populations and (3) how it might affect other outcomes.

Perhaps the best way to quantify the net outcomes of the Lidcombe Program research to date is the meta-analysis of randomised, controlled evidence and studies (Onslow, 2013). The participant group included 620 preschool children from Australia, Germany, Sweden, Holland, Canada, Britain and New Zealand. Efficiency outcomes showed that a mean of 15.4 weekly clinic visits was required for children to achieve near-zero stuttering levels. Further, an odds ratio of 7.5 was reported. Such findings further support the Lidcombe Program as the strongest evidence-based treatment option for preschool children who stutter.

The Present Thesis

The Need for Early Stuttering Intervention

The Problem

The impact of stuttering on a person's social and emotional wellbeing can begin as young as at 3 years of age, and potentially last a lifetime. Due to the myriad of negative lifelong difficulties that can be experienced by a person who stutters, it is paramount that young children who stutter receive timely access to evidence-based intervention.

When the current incidence of early stuttering is considered, a conservative prediction would be that close to 120,000 of the 1.4 million Australian children younger than 5 years of age (Australian Government, 2009) require intervention or at least access to a speech pathologist at any given time. Even if a more liberal view is taken of providing treatment for the 40-75% of children who stutter and who do not experience natural recovery, access to ongoing treatment would be required for a possible 30,000- 72,000 preschool children in Australia at any given time.

This, however, is currently unachievable due to the range of access barriers and service limitations preventing children from receiving intervention from appropriately trained speech pathologists.

The Solution: This Thesis

Given the overwhelming evidence in the literature of the difficulties faced by children, adolescents and adults who stutter, this thesis investigates a model of service delivery that will increase access to current evidence-based treatment for early intervention, namely the Lidcombe Program.

First, the current access and equity of paediatric speech pathology services is explored to highlight just how difficult it is for some families to access treatment. Then, telehealth, an emerging solution to access difficulties, is reviewed. Previous telehealth trials providing stuttering treatment are reviewed, including earlier low-tech Lidcombe Program studies. Then, consideration is given to current technology that is available to children in their homes. Finally, the feasibility of home-based webcam and internet treatment is considered.

Overall, this thesis aims to investigate and develop a service delivery model that increases access to timely, best-practice stuttering intervention. Telehealth using the internet and webcam could be a method of service delivery that: (1) improves access for children to evidence-based best-practice stuttering treatment, (2) improves access to specialist speech pathologists and quality services, (3) reduces costs and resources involved with outreach and home-based service provision, (4) provides more convenient home-based treatment for young children, and (5) ensures more equitable service delivery for young children and their families in rural and remote areas. The ultimate goal of this service model is to decrease the incidence of chronic stuttering and therefore reduce the number of school-age children, adolescents and adults who experience negative peer responses, have limited educational and vocational opportunities, develop social anxiety, and are greatly limited by the impact of their stuttering.

CHAPTER 3: EQUITY AND ACCESS TO PAEDIATRIC SPEECH PATHOLOGY SERVICES

Equity

At a global level, health is considered a fundamental right for all. Equity in health care for each country and its population is listed as a basic value of the current World Health Organization's Health for All policy framework (World Health Organization, 1998). In relation to speech pathology services in Australia, Speech Pathology Australia's Code of Ethics addresses service planning and provision within its standards of practice, stating, "At all times we endeavour to ensure our services are accessible and there is equity of access to services for our clients; such equity being determined by objective consideration of need…" (Speech Pathology Australia, 2010, p. 2). Such a code supports the theoretical World Health Organization framework that all Australians should have equal access to evidence-based speech pathology services. This, however, is not the current reality.

Access

Access Considerations for Rural Children

Equity and access to speech pathology services in Australia were reviewed and investigated by Wilson, Lincoln, and Onslow (2002), and then more recently by Verdon, Wilson, Smith-Tamaray, and McAllister (2011). These reviews are detailed below.

The Wilson et al. review

Background

In their literature review, Wilson et al. (2002) reported inequitable access to health care between rural and metropolitan areas, suggesting that this could lead to less desirable health outcomes for populations with limited access to treatment. This is consistent with a later review of evidence-based practice within Australian speech pathology, where McLeod (2008) cited distance and geographical isolation as factors that are reported to affect the equitability of Australian speech pathology intervention.

Methods

Wilson, Lincoln, and Onslow (2002) examined the inequitability of speech pathology services for a paediatric population. They conducted semi-structured interviews of 12 speech pathologists who provided services to children in rural areas. The speech pathologists were considered to be generalists who provided a broad range of intervention to children of various ages and speech pathology needs. Children who stuttered would be considered within this broader group, and the generalist speech pathologists would be responsible for providing intervention for stuttering.

Results

Overall, 11 of the 12 participants reported that they believed speech pathology intervention differed between metropolitan and rural populations. Information relating to speech pathology intervention for rural children was explored further. First, all participants reported that they or their service provided outreach services to more than one town. Service to these towns was reported to vary in frequency. Access for children within these towns, or surrounding areas, depended on the family's ability to travel to the town. The service delivery method was reported to vary from individual to group centre-based intervention; home, preschool or school visits; consultation to educational settings; drop-in clinics; and service via telephone. The frequency of intervention also varied greatly. Some consumers received weekly treatment; others received a set number of sessions or intensive blocks of treatment. Participant speech pathologists reported that children who were unable to access their service were provided with home or school programmes.

The use of telehealth consultations was reported by three participants. One reported use of the telephone as a supplementary, ad hoc service option. Two participants reported using the telephone either weekly or monthly as a means of providing direct service intervention to children who could not access outreach sites. Seven of the participants discussed the quality of intervention provided to isolated children. Five reported that intervention quality and efficacy was lower for children who could not engage in centre-based services regularly. This would suggest that rural children did not have equitable access to service, as service delivery options were largely determined by access rather than by the need of their disorder or by the evidence base for supplying the appropriate speech pathology intervention via the recommended delivery model.

Client access to service was a significant issue. Access was reportedly influenced by distance from services, transport disadvantage and socioeconomic factors that included the costs associated with travelling to outreach sites. A secondary consideration was also the time speech pathologists spent travelling rather than providing direct service.

Study Implications

The authors proposed that for health care to be equitable it should be available to all consumers, who in turn should have equal access to it. Further, they recommended that the service provided should be of appropriate standards, providing satisfactory outcomes. This is consistent with evidence-based best practices. The authors concluded by recommending alternative service models that might include the use of technology to assist with increasing equity and access to speech pathology intervention for children living in rural areas of Australia.

The Verdon et al. review

Access to speech pathology services for children living in rural areas has been more recently investigated in Australia (Verdon, Wilson, Smith-Tamaray, & McAllister, 2011). That review investigated access and service provision from the perspective of speech pathologists.

Methods

Seventy-four speech pathologists working in rural Victoria and New South Wales were interviewed about their workplace or service location and frequency of intervention offered. Their responses were used to map and calculate the availability of rural paediatric speech pathology services.

Results

The results showed that 98.6% of rural locations were underserviced, offering public services less than once a week. Another finding was that 50 kilometres was the maximum distance people would travel to access paediatric outpatient services. Of further interest, the majority of speech pathology positions were full-time and occupied by a single speech pathologist. Often, this single speech pathologist was reported to spend up to 4 hours of a working day driving to sites, leaving only 4 hours to provide treatment. Consequently, she or he could treat a only limited number of children during any single visit. The frequency of service for children in such regions could be considered even lower, considering they might not all be seen each time the speech pathologist visited.

The frequency of service in the above study varied. Speech pathologist visits ranged from daily to only when required. Service was often provided in blocks of 10 weeks. This differed greatly from the speech pathologists' perceptions of how often sites should be serviced. Nearly 75% stated that weekly visits would be optimal.

Study Implications

Such findings indicate issues of concern about access to services for children who stutter. First, they cannot access service at the recommended frequency of current evidence-based practices. Among sites that received at least weekly services, only 27% of site were within 25 kilometres of consumers. The access did, however, increase with distance, as 57% had access within a 50 kilometre radius and 83% within a 100 kilometre radius. However, 17% of consumers had no access to weekly services even within a 100 kilometre radius of their home. Second, even if a child who stuttered could access a speech pathologist, it is possible that the speech pathologist might not have the appropriate training, experience or desire to treat early stuttering. Furthermore, the speech pathologist might not attend the site on a sufficiently regular basis.

Access Considerations for Speech Pathologists

The issue of access extends far beyond physical location. It also includes resources, the type and frequency of the service, and the skills and expertise of speech pathologists. These issues have been investigated in Australian speech pathology and are reviewed below.

Speech Pathology Resources

Speech pathologists are the professionals trained to treat stuttering (Onslow, Jones, Menzies, O'Brian, & Packman, 2012). It is difficult to accurately quantify the number of trained speech pathologists in Australia who work with people who stutter. A conservative estimate would be around 1000. This is based on close to 20% of the speech pathologists listed on the Speech Pathology Australia website who have listed stuttering as clinical practice area. This figure is, however, a conservative estimate, given that not all speech pathologists subscribe to Speech Pathology Australia, or choose to be listed on the website. Yet it could be accurate as there are thought to have been around 4,500 speech pathologists in Australia in 2009 (Speech Pathology Australia, 2009), and now there are over 5000 Speech Pathology Australia members (Speech Pathology Australia, 2013). Regardless of the exact figure, there is a shortage, with the need for service exceeding the demand.

An Original Speech Pathology Australia Report

Access, locality and service provision were reviewed by Speech Pathology Australia in 2002. While the data are now 12 years old, trends reported can still be considered relevant as no subsequent report has been released. The 2002 Speech Pathology Australia Membership Survey (Lambier, 2002) provided interesting data pertaining to paediatric consumers of speech pathology. First, 65.1% of respondents identified preschool children as the main population with whom they worked. This was the second largest area. Second, time spent in speech pathology was greatest for school-age and preschool-age children. Third, the state governments were the largest employing bodies of speech pathologists, with community health employing a high proportion of these respondents within the state health sector. This underlines the importance of the data reported by the early intervention sector, as it was the largest clinical area represented in the survey.

Work locality data also provided an insight into access issues for clients who stutter (Lambier, 2002). There is a caveat to this information, however, as it was based on a response rate of only 52.5% (1514 members) of the association's membership, which itself is not mandatory. Thus, we can only be guided by the following information. Approximately 85% of respondents worked in highly accessible areas: an area defined as having access to a variety of goods, services and opportunities.

It is worth noting, however, that one-third of these speech pathologists were self-employed or worked for private practices or organisations. At the time of this survey, the cost of a treatment session was reportedly $70-$100 per hour. This in itself could be a barrier to service, as such cost, particularly on an ongoing basis, might be beyond the means of many families. A further 9-10% of respondents worked in areas classified as accessible; that is, having some restricted access to goods, services and opportunities, which in this study translates to restricted access to speech pathologists (Lambier, 2002). In contrast, only 1.5% of respondents worked in areas classified as remote or very remote, with significantly restricted access to goods, service and opportunities. This figure comprised solely self-employed speech pathologists, again adding cost of service as a barrier to access service for this demographic.

Speech Pathology Stuttering Expertise

From reviewing the Speech Pathology Australia workforce data (Lambier, 2002) it becomes evident that access to evidence-based stuttering intervention is likely to be further affected by the fact that not all speech pathologists are trained in early stuttering intervention. Consequently, the issue of access to appropriate intervention for early stuttering is complex.

First, families need to be able to have reasonable access to a speech pathologist. In this case, access relates to timely, regular service that can be accessed without significant difficulty, time or expense by the family. Second, the speech pathologist to whom families have access needs to have the clinical skills and training to deliver evidence-based intervention. This includes access to a service model that permits the recommended frequency and duration of treatment. It is this second aspect that is often overlooked. As a result, access to appropriate, timely, evidence-based treatment on a regular basis is a problem faced by both rural and metropolitan families. In reality, then, true access to evidence-based stuttering intervention could only be reported by a locality study of trained speech pathologists. This access issue is compounded by other factors. Even if a service has appropriately trained speech pathologists, secondary barriers can include long waiting lists in community services, or service models which may not be conducive to the recommended amount of treatment.

In keeping with the need for all children who stutter to have access to suitably skilled and trained speech pathologists, it can be argued that the minimal service that rural and remote populations can currently access may be reduced still further if the closest or visiting speech pathologist is not suitably skilled, or is not able to offer appropriate, regular, ongoing treatment sessions. In response to these issues, alternative service delivery methods are being developed to increase access to intervention for stuttering. These include telehealth adaptations and internet programs of the current evidence-based stuttering intervention, the Lidcombe Program.

Ethical Considerations with Speech Pathology Access

Equity of service has been reported as an ethical dilemma faced by new graduates and experienced speech pathologists in Australia (Kenny, Lincoln, Blyth, & Balandin, 2009). Kenny et al. conducted a study of 10 new graduate speech pathologists and 10 experienced speech pathologists who worked in public and private service in New South Wales, Australia. The speech pathologists were interviewed using a narrative approach to elicit reports of ethical dilemmas in their professional roles. Both groups reported that decisions around negotiation of intervention and the need to discharge clients were challenging. Further, 70% of new graduates and 100% of experienced speech pathologists spoke of the ethical dilemma they experienced in relation to service delivery. This included the quantity and quality of intervention provided. Equity of service provision was reported as a significant challenge to both groups, who identified providing intervention to large caseloads as the greatest confounding factor to providing equitable care to the clients who could access their services.

Paediatric Service Delivery: The Ruggero et al. Report

Background

Delivery of paediatric speech pathology services was recently reviewed in Australia using parent surveys (Ruggero, McCabe, Ballard, & Munro, 2012). The purpose of this study was to determine whether paediatric speech pathology services were consistent with evidence-based practice and parent preferences for treatment. In their literature review, Ruggero et al. (2012) reported that many Australian speech pathologists cited time and resources as barriers to providing evidence-based practice intervention to their caseloads. Limited staffing, large caseloads and availability of service were also reported to be factors impeding Australian children from receiving timely, evidence-based intervention. This, of course, would include young children who stutter. Finally, parent reports indicated greatest satisfaction with frequent service provided to their children across longer periods of time rather than less frequent services, or shorter periods of time.

Method

The online survey was completed by 154 parents who had at least one child waiting for or receiving speech pathology intervention, or whose children had received intervention. Demographics collected from the survey demonstrated responses from parents in all states and territories in Australia, including small and large cities and towns and with participants from low, middle and high socioeconomic backgrounds. Children engaged in services ranged in age from younger than 2 years to 18 years of age.

Results

Overall, waiting times for an initial assessment were reported to range from less than 1 week to over 1 year. The median time was between 2 and 6 months. Following assessment, children waited between 1 and 6 months for intervention. Children in Queensland often waited for longer than 1 year. Almost half of the children represented in the survey received intervention in private practices, and one-third received intervention in community health settings. Children in capital cities attended private practices more than rural and remote children; 75% of services were reported to be conducted at clinical sites, with 85% of children attending individual sessions.

Over one-third of the parents reported not receiving the service type they wanted for their child. Almost half of the parents attending university settings and private practices reported being given a choice in intervention planning, compared to less than one-third who attended community or hospital settings. This trend was similar for session frequency. Over half of the parents attending university settings and private practices reported receiving weekly treatment sessions, compared to less than one-third who attended hospital or community settings. Overall, the median length of sessions was 30-44 minutes. This was consistent with parent preferences. Close to half of the parents wanted weekly treatment sessions for their children, with 80% wanting individual sessions. Treatment breaks and discharge were also reported. Close to 75% of parents reported that they were not involved in decisions relating to breaks in treatment or discharge. For children on breaks or discharged from treatment, 52% had been given home programs to complete. The completion rate of these home programs was high, with 85% of parents reporting they had completed set work.

In relation to technology, 97% of parents reported having a computer at home, with 92% having broadband internet. This is significant when considering the use of telehealth services using the internet for both metropolitan and regional families, as most would have access to the required technology. It was also reported that 89% of parents stated that access to services for their child was insufficient or unavailable, resulting in some families travelling considerable distances to city-based services.

Ruggero et al. (2012) also collected parent suggestions for changes to current speech pathology services. Again, almost half of the parents recommended increasing services. This included increasing the frequency of sessions and the length of treatment blocks, having more follow-up sessions and better access to services for rural and regional families. Close to one-third of the parents suggested increasing the number of speech pathologists, and one-fifth suggested shorter waiting times for initial assessments. Additional suggestions included increased subsidies and lower costs for private services, more collaboration between speech pathologists with parents and teachers, retention of rural-based speech pathologists, greater flexibility of service locations, times and therapy styles, having speech pathologists in every school and early years setting and more research and speech pathologists specialising in childhood apraxia of speech.

Study Implications

In conclusion, the authors recommended that speech pathologists should implement evidence-based best-practice intervention in service models individualised for each family. Ruggero et al. (2012) also concluded that distance models of treatment, namely telehealth, should be included in mainstream service planning rather than being presented as an alternative to standard treatment options. This finding is significant to this thesis, in which telehealth is investigated as a stand-alone service delivery model for preschool children who stutter.

Speech Pathology Lobbying for Access

Following on from parent concerns about access, the peak professional body in Australia for speech pathologists, Speech Pathology Australia has also identified access to services for all children as a leading lobbying issue in 2007 (Speech Pathology Australia, 2007a; 2007b). The association prepared three issue statements that were used for lobbying the Federal Government. The first statement proposed "mandated speech pathology services for all children with speech and/or language disorders" (Speech Pathology Australia, 2007a). The second statement further identified the issues associated with the service barriers faced by Australians living in rural and remote areas, and proposed "improved access to speech pathology services in rural and remote Australia" (Speech Pathology Australia, 2007b).

Both papers cited that untreated speech and language disorders can be associated with reduced employment outcomes, increased social and emotional difficulties, mental health problems and increased occurrence of criminal behaviour resulting in juvenile detention or imprisonment. The paper relating to rural and remote Australians further stated that to access service, this group faces significant financial strain and may even be forced to leave remote locations to reside closer to services.

In 2009, Speech Pathology Australia campaigned further about "Speech Pathologist' (sic) Role in Early Childhood Development and Education" (Speech Pathology Australia, 2009). This issue statement suggested that the government plan targeted speech pathology services for at-risk populations. Speech Pathology Australia again cited the dire long-term outcomes associated with ongoing speech and language delays, stating in addition that "oral language competency in the young child is a strong protective measure against negative outcomes in later life" (Speech Pathology Australia, 2009).

While such position statements are broad, children who stutter clearly belong in the category of ongoing speech disorders that impact on oral competency. Children who stutter can thus be identified as a specific subset of the population that should receive targeted speech pathology services. Children who stutter also reside in rural and remote areas, areas that are underserviced and greatly isolated (Speech Pathology Australia, 2007b).

Summary

Equitable access to speech pathology intervention is not universal for Australian children. Rural populations are reported to receive service that is less than and different from that of their metropolitan peers. The impact is great for rural children who stutter. For evidence-based intervention for preschool stuttering to be truly effective, its target population must be able to access it. The issue of access for preschool children who stutter is twofold: (1) clinics need to have appropriately trained speech pathologists who can offer the required treatment sessions and (2) children who stutter need to be able to access these clinics. Telehealth, using internet and webcam, could be a viable option to overcome some of these access barriers.

CHAPTER 4: TELEHEALTH

Definition

Telehealth refers to the use of technology by health professionals to assess, treat or consult with their clients remotely. It can be an economical method of service delivery, eliminating travel costs for providers and consumers.

Telehealth in Australia

In the 2011-12 Australian budget, the Federal Government committed $352.2 million dollars to telehealth service provision. The investment was in recognition of the limited access of rural, remote and regional Australians to primary care and specialist services (Australian Government, 2011). In addition to this investment, the government also added telehealth consultations to the schedule of items that can receive a rebate from Medicare. Moreover, financial incentives were to be given to specialists, consultant physicians or psychiatrists who registered for this method of service delivery (Medicare Australia, 2010). Such an investment and initiative could certainly be interpreted as an endorsement of the validity, desirability and future of telehealth within the Australian health care system.

The Scope of Telehealth

Telehealth is currently used for a wide range of health issues (DePalma, 2009). An international telehealth rev iew was published in 2006 (Koch, 2006). This review included 578 papers published between 1990 and 2003. Koch (2006) reported that the development of telehealth services in developed countries is a result of the increased demand for accessible, efficient health care, aging populations and the difficulty retaining home-visiting health professionals. In summary, Koch reported that telehealth had become a more affordable treatment option due to low-cost technologies and the availability of the internet. The limitations of such advances were reportedly related to the logistics and sustainability of the technology. Such issues include the lack of protocols, guidelines and standards; the need for evaluation; ethical considerations; usability and economic considerations.

Telehealth as Viable Healthcare

A review of clinical outcomes and processes, utilisation and costs was completed for clinical rehabilitation services delivered using telehealth in 2009 (Kairy, Lehoux, Vincent, & Visintin, 2009). Papers that had an experimental or observational design, detailing telehealth rehabilitation for participants with physical impairment were included. Interventions included rehabilitation of community adults, follow-up treatment for spinal cord injury, and neurological, cardiac and speech-language impairment rehabilitation. Service locations also varied between clients' homes, rooms in health care facilities and group settings in the community.

This review of clinical processes reported that attendance and adherence to treatment recommendations was positive for telehealth rehabilitation programs. Further, both health care professionals and consumers reported satisfaction and positive gains for this service delivery model, with convenience listed as a common theme. Only one study reported only moderate satisfaction with rapport developed between the consumer and health professional using telehealth. Health professionals identified technical difficulties as a disadvantage of the telehealth service model, with shy or active children reported to be a more difficult group to engage. Rural health professionals were reported to be satisfied with this model also.

The authors concluded that there was a positive trend supporting the efficacy and efficiency of telehealth for the rehabilitation sector. They reported that clinical outcomes were similar to those of more traditional service models and that consumer acceptance and satisfaction were high. Five of the 28 studies reported a cost analysis of telehealth service delivery, with four reporting lower costs associated with telehealth. Consequently, the authors concluded that further research into cost-effectiveness was warranted.

Telehealth and Speech Pathology

Background

Telehealth has been formally endorsed by the American Speech-Language-Hearing Association (ASHA), which has stated that it is an acceptable method of service delivery (American Speech-Language-Hearing Association, 2005). Australia's professional body for speech pathology is yet to produce a position statement on telehealth, which is currently under development in 2013. The professional bodies for speech pathologists in Australia, the United Kingdom, Asia Pacific, India, Africa, Canada and Europe do not have such position statements or guidelines. Some of those countries do, however, have restrictions related to registration and locations in which speech pathologists are able to treat. In Canada, for example, speech pathologists must register in each province in which they work. Consequently, Canadian speech pathologists are restricted to using telehealth with clients who live in the same province, or they must register to practise in several provinces (Langevin, personal communication, 30th June 2011).

Guidelines and Protocols

Skill Sets

ASHA has produced a position statement (American Speech-Language-Hearing Association, 2005a) and knowledge and skills document (American Speech-Language-Hearing Association, 2005b) that recommends that speech pathologists possess a specific skill set before commencing telehealth service delivery. These skills include (1) ability to assess whether the service delivery method is conducive to the intervention that is required by the client, (2) ability to maintain professional competency, (3) ability to access appropriate support for technology-related issues and to provide these to the client, (4) adherence to a client's cultural beliefs and linguistic abilities, (5) ability to continually assess the effectiveness and outcomes of treatment via this medium, and (6) ability to adhere to client confidentiality protocols.

Privacy

Privacy relating to speech-language intervention using internet-based videoconferencing software has been reviewed (Cohn & Watzlaf, 2011). The increasing availability of free or low-cost internet-based software led the authors to review privacy policies of companies providing video conferencing technologies. That paper is particularly relevant to this thesis, as Skype was listed as an example of the medium the authors reviewed. Skype is the medium used for the RCT described in this thesis.

In summary, Cohn and Walzlaf (2011) stated that speech pathologists using internet-based technologies need to be informed of their responsibilities and to investigate the software they intend to use. They should inform their clients of the potential risks associated with certain software, allowing the clients to give informed consent. A tool to assist speech pathologists with this is a risk-analysis checklist (Watzlaf, Moeini, & Firouzan, 2010). This checklist addresses the aspects of privacy and security that speech pathologists should consider.

Legal Considerations

Speech pathologists are also required to consider legal and regulatory requirements when employing telehealth as a service model. Cason and Brannon (2011) considered frequently asked questions about telehealth practice in America for speech pathologists. Questions related to the licensing requirements in the respective state in which speech pathologists treat clients, the different media of telehealth, billing of telehealth in comparison to clinic services, professional and malpractice insurance, and the requirements of individual speech pathologists versus entire services. Although most of these recommendations relate to legislation and regulations specific to America, this paper highlighted generally how new practice models were not yet supported by current health policy.

In 2010, the American Telemedicine Association released a blueprint for telehealth guidelines. The aim of these guidelines was to educate and provide recommendations for health practitioners to use telehealth in a safe and effective manner (Brennan et al., 2010). Speech pathology was listed as a clinical area that was recommended to adhere to a series of administrative, clinical, technical and ethical practice guidelines.

The Mashima and Doarn Review

Mashima and Doarn (2008) completed an overview of the use of telehealth in the discipline of speech pathology. They reported published accounts of this practice in Australia, Canada, Greece, Ireland, Japan and the United Kingdom. Treatment areas included neurogenic communication disorders, dysphagia, paediatric speech and language disorders, voice disorders and stuttering. Mashima and Doarn (2008) concluded that “real-time” (p. 1105) interaction was recommended with equipment that supported audio and visual information to allow both parties to see and hear each other. The authors also reported that participants in telehealth trials had received “favourable patient feedback” (p. 1105). Further, speech pathologists were reported to have “initial scepticism” (p. 1105) but subsequent acceptance of telehealth.

Mashima and Doarn (2008) overviewed the advantages of telehealth. These included access to rural and unserviced regions, reduced service delay, increased cost-effectiveness of service for provider and consumer, allowance for additional support between visits, and the inclusion of family members. The challenges that emerged in this review include lack of professional standards and guidelines supporting the implementation of telehealth, the cost of purchasing initial equipment, lack of technical support and infrastructure, and ethical and legal issues regarding client privacy.

As reported in the reviews, there are many telehealth studies relating to speech pathology. For the purpose of this thesis, several papers have been selected that directly relate to the topic of this thesis or aspects of the methodological design. Studies specific to stuttering are reviewed later in the chapter.

The Hill and Miller Review

A recent survey determined the current clinical use of telehealth as a service model by Australian speech pathologists (Hill & Miller, 2012). In their literature view, Hill and Miller (2012) reported increasing availability of literature to support the use of telehealth for speech pathology assessment, but less for intervention. Overall, data were obtained from 57 respondents from Queensland, Victoria, New South Wales, Western Australia and the Northern Territory, with more than 75% of respondents reporting working in rural areas. More than 75% of respondents were under the age of 45 and 70% worked full-time. All respondents used telehealth as a method of service delivery in their current workplace. These included 57.9% in public health settings, 22.8% in private practice, 12.3% in public education settings, 10.5% in community settings, 8.8% in specialist services, 5.3% in private education settings, and 1.8% in private health services or nursing homes. Seventy percent of consumers were reported to receive the telehealth service within their homes, 21.1% from a medical centre, 21.1% at school and 10.5% at work.

Various technologies were reported to be used as part of telehealth services. Relevant to this thesis, 23% of metropolitan speech pathologists used stand-alone videoconferencing technology compared to 60.5% of rural speech pathologists. The use of the audiovisual program Skype was reported by over 30% of respondents. Telehealth was used by 40% of speech pathologists for informal and formal assessments, and by 86% for treatment. Paediatric caseloads were the largest consumer of telehealth speech pathology services, with 73.6% of respondents servicing this population compared to 52.6% who serviced adult populations.

Overall, 71.9% of speech pathologists reported being confident or very confident with their use of this method, and being satisfied or very satisfied with the service that consumers received. Reported benefits themes included access, time and cost efficiency, client focus and caseload management. Limitations or reported barriers to telehealth or speech pathology services were technical difficulties and lack of assessment and resources conducive to assessment and treatment via telehealth. Speech pathologists' suggestions to overcome these barriers included formal training and professional development, demonstrations by other speech pathologists, access to electronic resources, funding to establish service, ethical guidance, a position paper from Speech Pathology Australia, patient education, and inclusion in university speech pathology syllabuses.

Telehealth and the Hanen Program

Telehealth delivery of the speech pathology Hanen Program, It Takes Two to Talk, has been investigated (Loomes & Montgomery, 2012). Three different methods of videoconferencing were trialled with different participating families. First, the use of videoconferencing suites was trialled. Four families were required to travel to these sites to participate in group sessions. Participants reported satisfaction with the technology, but some of the families stated that travelling to the sites was difficult. These families reported that they would have preferred home-based services.

The second group of four families participated in group sessions using web-based group conferencing software from their homes, with their own computers and internet. The use of this software allowed all families to simultaneously see the presenter, PowerPoint slides and video examples. Family reports were positive about this medium; there was, however, a higher incidence of transmission and technical difficulties. The third method combined residential, on-site group sessions with home-based telehealth sessions. Specialised videoconferencing equipment was provided to three families. As a result, higher audio and visual quality was reported for this group.

Loomes and Montgomery (2012) reported that few modifications had to be made to the treatment program. These related to adapting small group exercises. The authors concluded that this method of service delivery for this specific parent education program was "comparable" (p. 125) to standard delivery. Treatment effect or postintervention language outcomes were not reported. Further research was recommended to directly compare satisfaction and child communication outcomes using a RCT.

Telehealth and Speech Sound Intervention

Telehealth delivery of speech sound intervention for school-aged children has been compared with standard delivery (Grogan-Johnson et al., 2011). Seven children aged between 7 and 12 years received traditional speech sound intervention using videoconferencing software at their school. An "e-helper" was trained to assist the child with the technology while a speech pathologist implemented all of the treatment. Six children aged between 6 and 11 years received the same intervention from the speech pathologist who visited them at school. The mean length of sessions was 22.9 minutes for telehealth and 20 minutes for the standard, school-based sessions. Attendance was also comparable, with 76% of telehealth sessions completed and 75% of school-based sessions completed. Treatment outcomes were reported to be significant for both groups as measured by pre- and postintervention scores using standardised assessment. Individual performance on the students' quarterly education plans was higher for the telehealth group.

Overall, the authors concluded that this model could be an effective and efficient way to provide speech sound intervention to school children. They did, however, cite their small sample size and nonlaboratory environment as reasons that larger scale research trials need to be conducted.

The Grogan-Johnson Project

In further consideration of school-based telehealth services in America, Grogan-Johnson (2012) reported on a pilot project under way across two rural school districts. Over 300 children from kindergarten to high school received telehealth services for speech pathology intervention. After 5 years of development of the telehealth project, Grogan-Johnson described a five-step plan and some practical advice when reviewing the school-based model.

The first recommended step was to investigate. This included researching the relevant policies, legislations and regulations, and then observing and trialling different telehealth options. The second step was to plan. This included considering the scope of practice, the technology needed, the training and implementation of technology use, appropriate facilities, support staff and policies and procedures. The third step was implementation. Trial sessions were recommended to identify possible difficulties that may arise. Orientation was also recommended for all parties involved, which should include education and the opportunity for information to be explained and discussed. The fourth step was to evaluate. This was considered crucial in further developing or expanding the use of telehealth. Evaluation should be sought from all stakeholders and parties involved with information used for the final step, reworking the model. This fifth step may include adapting processes, protocols and refining interventions.

Finally, following the five-step process of evaluation and revision, Grogan-Johnson shared three lessons learned. The first was to establish cooperation with the school, creating a cohesive working relationship. Second, selecting the "right students" (p. 3) was recommended. This included considering family preferences and the student's type of communication disability. The third recommendation was to commit to completing the project and working through challenges.

Grogan-Johnson (2012) cited data from a 2007 (Grogan-Johnson, Alvares, Rowan, & Creaghead, 2010) pilot model, in which 93% of the 29 students and 82% of the 22 parents interviewed reported satisfaction with this telehealth model of receiving speech pathology intervention. The model was considered "very good" or "above average" in targeting their child's communication abilities by 95% of parents. In contrast, 55% of the staff surveyed reported limited knowledge or understanding relating to the intervention students received, or how their skills developed. Teachers reported, however, that in their opinion, students who were involved in the project had a "very good" attitude towards the telehealth model.

Videoconferencing Software for Telehealth

Background

Currently, the greatest development in the use of telehealth appears to be the increasing use of low-cost or free software that is available on the internet and allows videoconferencing between the speech pathologist and the consumer. Such advances mean that consumers now readily access telehealth services using their own computers rather than travelling to purpose-built telehealth suites. Given this advance, a clinical review of one such software program, Skype, has been completed (Armfield, Gray, & Smith, 2012). This review is of particular importance to this thesis as Skype was the software used for this Phase III RCT.

The Armfield et al. Review

Armfield et al. (2012) initially sought studies that had been completed during 2006 or later, with large sample sizes with a randomised control design, and that provided direct care rather than use for training or administration. However, no studies were found that matched all those criteria. Rather, their literature search found case reports, pilot studies, uncontrolled studies and experimental demonstrations. This is significant for this thesis, as the Phase III RCT using Skype was considerably larger and more scientific than the body of research for Skype speech pathology intervention to date.

Rather than review these smaller studies, Armfield et al. (2012) composed a list of features that future RCTs investigating Skype as a medium for telehealth should consider. Again, the relevance to this thesis is significant as many of these elements were considered by the candidate in designing this study, prior to the publication of the Armfield et al. paper. These include considering the impact of image quality and types of webcameras used, the benefits and limitations of using home- and office-based technology and environments, consistency of internet connection and quality across sessions, treating patient directly, security and privacy. Overall, Armfield et al. concluded that using Skype could be advantageous for low-risk consultations, particularly when either party had limited resources to provide or access distance intervention. They cautioned, however that there was as yet no clinical trials evidence to support its use by health professionals. This provides further support for the study described in this thesis.

Interactive Computer-Based Therapies

Following the development of videoconferencing software that allows live clinical interactions, Theodoros (2008) suggested that specific interactive software be further developed. Such purpose-built software should allow both clinical interactions and simulated environments. Theodoros also recommended that research into the cost, application and professional development needs to be considered with this proposed model of care, to maintain appropriate clinical standards.

Internet Access

For telehealth using webcam and the internet to be a feasible service option, families require reasonable access to the internet. Internet access is reportedly increasing at a national and international level.

International Internet Usage and Access

At a global level, close to one-third of the world's population is reported to use the internet (Miniwatts Marketing Group, 2013). This ranges from 60 to 80% for Europe, North America, Oceania and Australia, 30 to 50% for the Middle East, Latin America and the Caribbean and 15 to 30% for Asia and Africa.

Quantifying access is reported less frequently than usage. A report from 2009 cited worldwide access data in relation to Australia (Pink, 2009). When access figures were compared for households in over 30 countries across North America, Europe, Asia and Australia, Australia ranked 12th. Further, Australia ranked ninth when access to broadband internet was compared. Interestingly, in both comparisons, access of Australian household was notably higher than that of American households. This is worth considering in light of America's acceptance of telehealth as an equitable service delivery method for speech-language interventions (American Speech-Language-Hearing Association, 2005a).

Australian Internet Access

Webcam telehealth is a feasible service option for Australian consumers. The most recent figures, reported at the end of June 2011, were 10.9 million individual internet subscribers in Australia, with 80% being household subscribers (Australian Bureau of Statistics, 2012). The use of broadband connection had increased to 95% of subscribers. Therefore, the technology required for webcam telehealth services is readily available to many Australians.

Australian Households

Household use of technology in Australia has been reviewed. The Australian Bureau of Statistics completed a detailed report based on data obtained during 2008-2009 (Pink, 2009). The findings of this report are significant when considering the application of webcam telehealth within the Australian health care system. First, 72% of households were reported to have internet access. Second, an estimated 5 million Australian households were reported to have access to broadband internet connection. This equates to 62% of all Australian households, and 86% of those with the internet. This was a significant increase in the past decade, with only 16% of Australian households having access to the internet in 1998. Australian households more likely not to have access to the internet were those without children under 15 years of age, those who lived outside metropolitan areas or who had a lower household income, considered to be less than $40,000 per annum. Third, 65% of nonmetropolitan households had access to the internet. This was only marginally less than the 76% of metropolitan households who have access. This could suggest that it is not distance or locality alone that acts as a barrier to internet service. This was consistent with by the figures of households accessing the internet using broadband connections: 88% of metropolitan households compared to 82% of nonmetropolitan households.

Children

Children's use of the internet was also reviewed (Pink, 2009). Children aged five to 14 years of age who lived in private homes were recruited for the 2009 Children's Participation in Cultural and Leisure Activities survey. Of the 2.7 million children aged 5 to 14 years in 2009, 79% were reported to use the internet. The family home was the main site of internet access for 73%, or some 2 million Australian children. Children who had less access to the internet were those with unemployed parents (64% of this demographic had access), children from Tasmania (64% access) and the Northern Territory (62% access). Children living in metropolitan households had a higher incidence of access than their remote peers, who had an incidence of 70% household internet access. Given the increase in household use between data reported in 2009 and 2011, it can be assumed that children's usage and access figures would have increased. These have not yet been reported.

The Australian National Broadband Network

The Australian Government has committed to further enhancing internet through the National Broadband Network. It is planned that the network will provide coverage to all Australians by 2020. The current plan for the network is that 93% of Australians will have access to optic fibre technology that is anticipated to be up to 100 times faster than the current broadband speed. The final 7% will receive their broadband via wireless and satellite technologies (Australian Government, 2011). Such an increase in access would certainly be conducive to further establishing webcam telehealth as a viable service option for many Australians.

Telehealth and Stuttering Intervention

Background

Telehealth is reported to have been used extensively for people who stutter. Research investigating the use of telehealth to provide treatment to people who stutter has largely been conducted in Australia and Canada, due to the reasonably small populations living across an expansive country (Packman & Meredith, 2011). To date, the technology used in telehealth studies has varied from low-tech telephone studies, to high-tech videoconferencing suites. In more recent times, studies have used the internet and webcams as a more cost-effective option. Telehealth is reported to be a feasible service option for people who stutter. Relevant literature is reviewed here.

The Kully Report

Kully (2002) reported on the use of telehealth at the Institute for Stuttering Treatment and Research (ISTAR) in Canada. Kully cited access to the centralisation of specialist services as the main reason telehealth was introduced as a service delivery model. At the time of the report, Kully reported that over 80 telehealth sessions had been administered to clients who ranged in age from 3 to 38 years. In all but one of the cases, telehealth was offered in combination with clinic-based treatment rather than as a stand-alone method. Most participants were required to travel to telehealth sites; these were often community centres or schools that had appropriate equipment. The set-up costs of these sites and the cost of travel for clients to satellite sites were reported as limitations of telehealth

Kully (2002) reported positive observations from the introduction of telehealth at the ISTAR. Clients and speech pathologists reported that treatment goals were met and both parties were satisfied with sessions. Technology was reported to be adequate for the majority of the sessions, with visual transmission particularly important during early sessions and when working with smaller children. The Lidcombe Program was reportedly used with preschool children, but no specific outcome data were reported for this treatment. Speech pathologists working with preschool children and families reported difficulty directly modelling treatment for the families. Consequently, speech pathologists relied on their ability to verbally explain specific treatment aspects to parents. Kully reflected that not sharing a physical space had less of an impact in the treatment interaction than anticipated. Kully concluded by raising issues related to the need for client privacy and confidentiality, together with consent, liability and licensing.

The Packman and Meredith Review

Packman and Meredith (2011) posed significant considerations for speech pathologists who offer telehealth. First, they cautioned about the safety of distance intervention, citing privacy and confidentiality as issues to consider. Second, they warned about the legal and ethical issues that might occur if speech pathologists treated across state or national borders. This is because different states and countries have different telehealth legislation and professional boards of governance. Third, the authors considered it imperative for speech pathologists to continue to seek and employ effective evidence-based treatment models. Packman and Meredith concluded by recommending professional guidelines for standards of care for people who stutter.

Adults

A Phase I Trial

A Phase I trial of 10 participants receiving the Camperdown Program, a behavioural treatment using prolonged speech, was reported by O'Brian (2008). Consultations occurred using the telephone. As a group, there was an 82% reduction in stuttering frequency immediately following treatment and a 72% reduction 6 months posttreatment. A mean number of 8 clinical hours was required per participant. This was fewer than the clinic Camperdown Program group. Thus, not only was telehealth efficacious, it was also efficient in terms of hours required by the speech pathologist to treat each participant.

Compliance in that study was high, with no participants withdrawing. This might be attributed to the sessions being conducted at locations that suited the participants, as they did not need to attend a clinic. Many of the participants attended sessions while at work, during their lunch break, in a park or the car park, on holidays or on their days off. Such compliance and convenience was a notable feature.

A Phase II Trial

A Phase II RCT was then completed to further determine the efficiency of this service delivery method (Carey, O'Brian, Onslow, Block, Packman & Jones, 2010). For the 40 participants, both clinic and telephone delivery were found to be efficacious. Further, speech pathologist time required per participant was less for the participants who were randomised to the experimental group. Participants who received treatment over the phone required a mean of 2 hours less than the control group. That was a 20% saving per participant. Such efficiency is significant in terms of costs associated for both clients and speech pathologists: time off work and treatment fees for the client and clinical hours and resources for the speech pathologist. Both studies retained the principles of the Camperdown Program, using exemplar tapes to provide speech models and having participants self-evaluate their speech (Carey et al., 2010; O'Brian, 2008).

Adolescents

A Phase I Trial

Telehealth delivery of the Camperdown Program has been investigated with adolescents, similarly receiving the treatment via webcam (Carey, O'Brian, Onslow, Packman & Menzies, 2011). Results from a Phase I trial with three participants were positive. As a group, the participants demonstrated an 83% reduction in stuttering frequency immediately following treatment, a 93% reduction 6 months posttreatment and 74% reduction 12 months posttreatment. Further, the participants preferred their telehealth experience to the clinic-based sessions that they had experienced previously.

Children: Videoconferencing

The first study to use videoconferencing for stuttering treatment was completed with four children aged between 3 and 12 years of age, and two adolescents who were 17 and 19 years old

(Sicotte, Lehoux, Fortier-Blanc, & Leblanc, 2003). The treatment was delivered from a tertiary centre to a remote satellite clinic. The remote clinic was equipped with resources for treatment and a speech pathologist to support the parent and child. Families were required to travel to this clinic to receive treatment. The exact nature of the treatment was not detailed; the authors reported that it was informed by evidence-based practices at the time. Treatment consisted of 12 weekly treatment sessions initially, and then a schedule of less frequent sessions during the maintenance stage of the treatment process.

Treatment outcomes were reported in relation to stuttering reduction. Pretreatment stuttering levels ranged from 13 to 36%SS. Posttreatment stuttering levels reduced to a range of 2-26SS%, and post-follow-up stuttering levels ranged from 4 to 36%SS. All participants were reported to have reduced stuttering levels immediately posttreatment and to some degree at the follow-up assessment.

Overall, the treating speech pathologist was satisfied or highly satisfied with the clinical quality of 81% of sessions. Sessions that were judged as less satisfactory included times when the speech pathologist found it difficult to engage with a child moving around or one who was shy. The technical quality was reported to be moderately good, with image reported as the least successful technical component. Participant opinion was obtained using questionnaires. Technical and clinical quality was rated highly by five of the six participants. All participants reported confidence in the quality of treatment. In conclusion, Sicotte et al. (2003) reported that their study supported the use of videoconferencing as a viable stand-alone service delivery method for children who stutter.

Preschool Children: Low-tech Lidcombe Program Telehealth Studies

The Lidcombe Program is the only preschool stuttering intervention program that has published clinical trial evidence in support of telehealth (Harrison, et al., 1999; Lewis, et al., 2008; Wilson, et al., 2004). The studies are reviewed below.

The Harrison et al. Phase I Trial

The first study involved an Australian speech pathologist treating a single child living in the United Kingdom who had a long history of stuttering and failed treatment (Harrison et al., 1999). The child successfully achieved entry into Stage 2 following 25 phone consultations over 277 days. The treatment effects were still evident 23 months after entry to Stage 2.

To administer treatment in this unconventional way, Harrison et al. (1999) altered the standard delivery of the Lidcombe Program considerably. Standard one-hour treatment sessions were replaced with scheduled phone calls. These consultations did not adhere to the prescribed weekly sessions, as time was needed to allow the parent training tapes and taped samples of the client's speech to be mailed back and forth between Sydney and the United Kingdom. Consequently, sessions occurred at a mean rate of every 11.5 days. No sample of the child's speech was measured for %SS by the speech pathologist and parent at the beginning of each session, nor was the child present for the majority of the sessions. Instead, tapes of the child's speech were sent to the speech pathologist to measure %SS. Severity rating training occurred over the telephone, with parent-speech pathologist agreement achieved by rated samples sent via post. The speech pathologist was not able to model the treatment for the parent with the child, nor was the speech pathologist able to observe the parent implementing the treatment live. Instead, video samples were sent to the speech pathologist of the parent implementing treatment with the client. This allowed feedback to be given and adjustments to be made to the treatment program retrospectively. The telephone consultations were also usually shorter than the prescribed 1 hour, varying from 20 to 65 minutes.

Harrison et al. (1999) reported that the cost-effectiveness of this method of service delivery was a considerable limitation. Apart from the treatment consultation, considerable time was spent rating the video samples and observing the parent's treatment. This additional time was not required during clinic treatment as these tasks were incorporated within the treatment sessions. However, despite the further limitation that this was a single case study, the outcomes supported the viability of this treatment method. Overall, the outcomes were satisfactory and attested to the flexibility and robustness of the Lidcombe Program treatment principles.

The Wilson et al. Phase II Trial

The second telehealth study was a Phase II trial investigating telephone delivery of the Lidcombe Program with five Australian-based participants (Wilson, et al., 2004). Again, considerable changes were made to the treatment process. These were: (1) clinic sessions were replaced with phone consultations, (2) real-time measurement was removed from these consultations and replaced by sample audio tapes sent to the speech pathologist on a regular basis, (3) severity rating agreement was obtained by the parent and speech pathologist using taped audio samples rather than live clinic samples, and (4) real-time observation of parent treatment was replaced by taped samples sent to the speech pathologist for delayed feedback. Regular contact, treatment and direct speech sampling with the client were removed from almost every consultation. Training was provided during phone consultations and was supplemented with exemplar tapes and information sheets that were also sent to the families. Parent education for identifying ambiguous and unambiguous stuttering occurred following viewing of tapes of the parent implementing treatment sample. The parents were reported to be able to use severity ratings accurately. Thus, this method of parent training was deemed adequate.

Participant selection criteria included isolation from weekly Lidcombe Program treatment elsewhere. Interestingly, this did not result in an uptake of only regional or rural clients. Two participants lived 4 hours from the closest service, but the remaining three lived in areas that were classified as "highly accessible". These participants reported dissatisfaction with their local service, or lack of service provision. Speech pathologists at their local service did not offer the Lidcombe Program.

The total speech pathologist time was measured for each participant in the study. This time included all noncontact tasks such as preparing materials and reviewing tapes. The mean session times ranged from 22 to 41 minutes, with the total speech pathologist time for each session calculated to 33-68 minutes. The frequency of contact ranged from 8 to 38 days. Wilson et al. (2004) reported that for four of the five participants, treatment sessions exceeded the benchmark data for clinic treatment time of 11 sessions (Jones, 2000). The authors noted that this could be in part related to the predictor variable of stuttering severity, as increased stuttering severity is reported to increase treatment time required. In this study, four of the five participants were considered to present with more severe stuttering, which is associated with a known increase in treatment time.

Wilson et al. (2004) concluded that although telephone service delivery was not an efficient model of service, a distance alternative for delivering the Lidcombe Program could be efficacious. They recommended further investigation of ways to increase efficiency, including videoconferencing that would allow direct contact and observation between the speech pathologist and parent and child. This method could be conducive to live measurement, modelling treatment, observations and coaching.

The Lewis et al. Phase III Trial

The third study investigating this form of service delivery for the Lidcombe Program was a Phase III trial RCT (Lewis et al, 2008). This trial compared telehealth delivery to no treatment. Thirty-seven participants were initially recruited, but 15 did not commence. The majority of drop-outs reported that the stuttering had stopped or that alternative treatment had commenced elsewhere. One participant withdrew as the parent did not want to adhere to the requirements of the study, one for an unknown reason and one was uncontactable. This resulted in 22 participants being randomised to a control arm of no treatment for the initial 9-month period or the experimental arm. Unequal randomisation occurred, with 9 in the experiment arm and 13 in the control arm. Further withdrawals resulted in 9-month data being analysed for 8 experimental participants and 10 control participants.

The Lidcombe Program treatment process was adjusted in the same way as for the previous Phase I and II trials. Similarly, it proved safe and effective, with poorer efficiency. The results showed it took three times longer when compared to the existing benchmarks for clinic treatment. Using a larger sample size, the authors reported a mean number of 49 consultations over 62.9 weeks to complete Stage 1. However, Lewis et al. (2008) were more able to adhere to the recommended weekly sessions, with a mean of 7.7 days between consultations. Their consultation time varied from the manualised recommended 45-60 minutes, with the mean consultation time being 33.1 minutes. In the Phase III trial the total clinical time was a mean of 77.3 minutes per session. Information obtained from parents via questionnaires indicated that despite the considerable time taken to achieve near-zero stuttering, parent satisfaction was high, with 87% of participant parents reporting their telehealth experience to be positive. Further, all parents rated themselves as "very satisfied" with their child's fluency.

There is a caveat to this study. The treatment process differed from the manualised version on which clinic benchmarks are based. The treatment was not delivered weekly and the speech pathologist was unable to observe treatment implemented live, rate the child's speech live, or engage in subsequent problem-solving and treatment alterations live. Such logistics changed the Lidcombe Program treatment process considerably.

Preschool Children: High-tech Lidcombe Program Telehealth Studies

The O'Brian et al. Phase I Trial

More recently, another Phase I trial was completed (O'Brian, Smith, & Onslow, 2012). The recommendation from Wilson et al. (2004) that treatment efficiency could be significantly improved if live teleconferencing was used was addressed. Use of a different process allowed the principles of standard delivery of the Lidcombe Program to remain relatively unchanged. It enabled the speech pathologist-parent-child triad to remain, with all parties having clinic contact. Real-time measurements, live observation and coaching for parental implementation of the program remained intact. Three preschool children received the Lidcombe Program, delivered in real time using the internet and a webcam in their own homes. Overall, the mean stuttering reduction achieved was 83% over a mean of 34 treatment sessions to complete Stage 1. During the treatment process, minor modifications were reportedly made due to the service delivery model. These included using verbal training techniques or explanations and role plays with the parent rather than demonstrating directly with the child. The treating speech pathologist reported less direct engagement with the children than in clinic Lidcombe Program treatment sessions.

Despite this, parent reports for this service model were positive. Parent reports in favour of webcam delivery of the Lidcombe Program related to the benefits of less time and cost, the comfort of being in their own homes, and less interruption to siblings' routines. A limitation was reported to be occasional technical difficulties.

The stuttering reduction outcomes and parental reports confirm the safety, validity and convenience of this method of delivery for the Lidcombe Program. The efficiency was less than the clinic benchmarks, but better than the previously reported low-tech trials. The authors suggested that a larger trial would investigate treatment efficiency more accurately as the three participants in this study might not be representative.

Summary

Telehealth provides an alternative service delivery option for people who have difficulty accessing treatment sites. Speech pathologists offering telehealth services need to be aware of legislation, regulations and possible limitations associated with internet-based software. Stuttering treatments for adults and adolescents have had positive outcomes that confirm the efficacy and efficiency of using telehealth methods. For younger children who stutter, low-tech trials using telephones were found to be efficacious but to take considerably longer than clinic treatment. Published trials with young children who stutter relate to the Lidcombe Program and other nonspecified treatments. More recently, preliminary data show that webcam telehealth delivery of the Lidcombe Program may provide a viable delivery model with efficiency rates closer to clinic benchmarks.

Part Two of this thesis outlines the methodology for a Phase III RCT that investigated the delivery of the Lidcombe Program using webcam, compared with traditional clinic delivery.

PART 2: METHOD

CHAPTER 5: TRIAL DESIGN

Noninferiority

Trial Design

The design for this project was a parallel, open plan, Phase III noninferiority RCT with the question: Are outcomes following Skype delivery of the Lidcombe Program at least as good as those following clinic delivery? Given the significant advances in technology since the previously published Lidcombe Program telehealth trials (Harrison, et al., 1999; Lewis, et al., 2008; Wilson, et al., 2004), use of webcam and the internet was considered the preferred medium for current telehealth services. This was further supported by the Phase I feasibility study detailed in Chapter 4. The trial design conformed to consort guidelines (Moher et al., 2012; Schulz, Altman, & Moher, 2011)

Clinic Lidcombe Program Control Arm

The control group received standard delivery of the Lidcombe Program (Packman et al., 2011) in a traditional clinical setting at the La Trobe Communication Clinic, La Trobe University, Melbourne, Australia. This group is hereafter referred to as the clinic group.

Webcam Lidcombe Program Experimental Arm

The experimental group received the Lidcombe Program within their homes, using a computer, a webcam, the internet and the live video calling program Skype. This group is hereafter referred to as the webcam group. The candidate provided all treatment and was located in an office at La Trobe University.

Primary Outcomes

There were two primary outcomes: (1) number of consultations to Stage 2, and (2) speech pathologist hours to Stage 2. These outcomes measured treatment efficiency and were collected at Assessment 2.

Consultations to Stage 2

The number of consultations to Stage 2 entry was recorded for each individual participant following their completion of Stage 1.

Speech Pathologist Hours to Stage 2

The duration of each consultation was recorded for each participant at the completion of the session. The total speech pathologist hours was then calculated for each participant by adding the Stage 1 session durations. This measure also allowed for comparison between groups and with previously published benchmark studies.

Secondary Outcomes

There were four secondary outcomes:

Weeks to Stage 2

The number of weeks to Stage 2 entry was recorded for each individual participant following their completion of Stage 1.

Parent Severity Ratings (SRs)

Parent-reported SRs were obtained at each assessment using an outcome questionnaire (see Appendix B for the Outcome Questionnaire). These ratings were collected independent of the candidate of the candidate within 2 weeks of the assessment of the assessment date.

Parent Satisfaction Ratings

Parent-reported satisfaction with child fluency was obtained at each assessment using the outcome questionnaire (Appendix B). These results were collected independent of the candidate.

Parent Survey Responses

Parent questionnaires were administered at Assessment 2. These questionnaires sought information about parent attitudes, experiences and satisfaction with the treatment process and outcomes. The questionnaires were completed via telephone with a research assistant independent of the treatment, transcribing the parent's responses to both 5-point scale and short answer questions (see Appendices C and D for these questionnaires).

Assessments

Participant information, primary and secondary outcomes were collected at a series of assessments during the course of this study. The assessment schedule and outcomes are detailed below.

Assessment 1

The first assessment occurred pretreatment. The purpose of assessment was to determine eligibility and to gather pretreatment information. Initial, pretreatment questionnaires were completed as part of the assessment battery. These included standard case history information and an Outcome Questionnaire (see Appendix A for pretreatment participant data). Outcomes were in the form of parent-reported SRs for the past week, reflecting the children's typical and most severe stuttering, where 1= no stuttering, 2= extremely mild stuttering and 10= extremely severe stuttering. Parents were also asked to rate their satisfaction with the level of fluency on a scale from 1 to 10, where 1= extremely satisfied and 10= totally dissatisfied. Two 10-minute beyond-clinic recordings were also completed during the assessment process, one with a familiar person and one with an unfamiliar person. Finally, a Skype test was completed to ensure adequate real-time audio and visual connection (see Appendix E for the Skype quality test).

Assessment 2

The second assessment occurred on completion of Stage 1 of the treatment process. The number of hours, sessions and weeks was recorded for each individual child to determine the efficiency outcomes. Parents were asked to complete another Outcome Questionnaire, as detailed in Assessment 1. This was to determine treatment efficacy by calculating stuttering reduction. A parent questionnaire was completed with each parent, gathering information about their experience, learning the treatment components and receiving consultations in the clinic or via webcam (see Appendices C and D).

Assessment 3

The third assessment occurred for each participant 9 months following randomisation and the commencement of treatment. Parents completed the Outcome Questionnaire, reporting SRs and fluency satisfaction ratings (as for Assessments 1 and 2).

Assessment 4

The fourth assessment occurred for each participant 18 months following randomisation and commencement of treatment. Parents completed the Outcome Questionnaire, reporting SRs and fluency satisfaction ratings (as for Assessments 1 and 2). This was to determine long-term treatment outcomes.

Hypotheses

The noninferiority hypothesis of this study was that outcomes following Lidcombe Program treatment using webcam would be at least the same as outcomes following clinic treatment. In relation to this thesis and the outcomes, it was proposed that (1) the number of sessions and weeks for the webcam group would be no more than for the control group, and (2) at 9 months postrandomisation, webcam participants would have the same stuttering reduction as clinic participants.

Participants

Preschool children who stuttered and their parents were recruited from the stuttering waiting list at La Trobe Communication Clinic, La Trobe University. A total of 49 participants were randomised, 24 in the clinic group and 25 in the webcam group. The participant group consisted of 37 boys and 12 girls, aged 3 years to 5 years 11 months at the time of assessment. The mean age of the clinic group was 4 years 2 months and for the webcam group 4 years 5 months. The mean time since onset was 15 months; 16 months for the clinic group and 14 months for the webcam group. The mean typical SR for the clinic group was 4.0 (range of 1-7) and for the webcam group it was 4.7 (range 2-9). Given that stuttering severity is a known predictor for treatment time (Jones et al., 2005), it was important to determine whether the groups differed in pretreatment severity. An unpaired t-test showed no significant difference in pretreatment parent-reported stuttering severity between the clinic group (M=4.0, SD=1.6) and the webcam group (M=4.7, SD=1.8); t(38)=-1.4, p=.2. This was important when interpreting efficiency measures, as severity could be excluded as a variable when comparing treatment time between the clinic and webcam groups. Efficiency outcomes could therefore be attributed to the service delivery method or variables other than severity.

Participant Assessment Procedures

A maximum of three assessment sessions was required to complete pretreatment questionnaires and determine eligibility. These were conducted at the La Trobe Communication Clinic with a speech pathologist independent of the treatment. This was to ensure that the candidate could not develop prior rapport with families who would be randomised to the webcam group. Therefore, webcam families met the candidate during their first webcam session, and were not permitted to visit the candidate at the clinic during the 18-month treatment process. Similarly, clinic families met the candidate at their first treatment session in the clinic.

Inclusion Criteria

All participants were children and their parent/s who met the following inclusion criteria: (1) aged between 3 years and 5 years 11 months at commencement of treatment; (2) stuttering for longer than 6 months (to reduce the likelihood of natural recovery occurring); (3) diagnosis of stuttering by consensus from the parent and assessing speech pathologist, confirmed by speech pathologist identification during assessment; (4) participant speech and language skills deemed functional for the purposes of the treatment process, and (5) in-home access to Skype with adequate audio and visual qualities.

Exclusion Criteria

The exclusion criteria were: (1) parent or participant without functional English for assessment and treatment; (2) parent report of child ADHD, intellectual disability or any other co-morbidity or complicating factor.

There were 11 children who were deemed ineligible during the assessment process for the reasons shown in Table 5.1.

Table 5.1

Ineligible Participants

Reason	Number
Behavioural concerns	2
Pragmatic disorder	2
Significant other speech disorder	1
Significant language delay	2
Failed Skype test	4

Withdrawals

A further six children withdrew from the assessment process for the reasons shown in Table 5.2.

Table 5.2

Assessment withdrawals

Reason	Number
Pursue articulation treatment	1
Death in family	1
New baby	1
Seeking private treatment	1
No reason provided	2

Groups

Randomisation

Eligible participants were randomly allocated to one of the two treatment groups and commenced treatment within 4 weeks of their initial assessment. Randomisation occurred independently of the candidate and participants at an interstate site. The children's names and the mean %SS of their beyond-consultation speech samples were sent to the Australian Stuttering Research Centre, The University of Sydney, where they were randomised according to a predetermined schedule that was prepared by a biostatistician on another site using dynamically balanced randomisation.

Stratification

Participant pretreatment beyond-consultation speech samples were stratified based on a baseline severity of above and below 5.0% SS. The rationale for this stratification was to ensure that both groups had similar baseline severity, as severity would be correlated with time taken to attain entry into Stage 2 (Jones, et al., 2005; Kingston, Huber, Onslow, Jones, & Packman, 2003; Rousseau, Packman, Onslow, Harrison, & Jones, 2007). This measurement was taken from the two 10-minute beyond-consultation speech samples that were recorded for each participant during the assessment process. The two measures were then pooled for the stratification. Each measurement was completed by the candidate to ensure reliability. Overall, 7 participants were randomised above 5.0%SS and 42 participants stratified below 5.0%SS.

Webcam Group: Logistics

Testing the Webcam Connection

For the purpose of the eligibility process, all participants had to pass a webcam test. This test ensured that minimum audio and visual standards were met; it also allowed families to trial the use of a webcam if they were unfamiliar with it. For many this was their first time. While the process was relatively straightforward for most families, encouragement and support were provided to some families who had not used the technology before.

Contingency Plan for Webcam Failure

At the beginning of the first consultations, parents were asked to communicate at any time if the audio or visual quality of the consultation was compromised. It was agreed that if at any time connection was lost, the candidate would always be the party to call back; this was to avoid both parties trying to call each other causing the call to be engaged.

Initiating a Webcam Consultation

To maintain consistency with standard clinical practice, the candidate established with the webcam families that the candidate would always initiate the call for the treatment session. Families were asked to be logged onto webcam, and to have tested their equipment 5 minutes before their scheduled call. The candidate would then initiate the webcam call at the designated time. Parents were asked to have their SR graph, resources for the session and participating children ready with them at this time. Families were asked to communicate in a timely fashion if they were unable to attend, or if their technology was not working.

Webcam Treatment Resources

Parents were told to have a simple picture story book with them during the first consultation. Discussion occurred during the first consultation to find out the kind of resources families had within their homes. Throughout the course of the treatment program the families were instructed to have certain resources with them for the webcam consultation. Parents could source a particular item during a webcam consultation if the one they had prepared was not suitable for the treatment conversation. Parents were also encouraged to have resources available for children to play with while they were not engaged in direct parent-speech pathologist discussion.

On occasion, the candidate emailed paper-based resources to families who did not have appropriate resources within their own homes. This occurred for both webcam and clinic families. Resources included matching pictures for common card games, lotto games and picture description scenes.

The Treatment

Lidcombe Program Treatment Process

The Lidcombe Program is a behavioural treatment for preschool children who stutter. The aim of the treatment is to reduce stuttering to zero or near-zero stuttering levels during Stage 1 of the treatment, and then to sustain these levels while treatment is withdrawn during Stage 2. The Lidcombe Program treatment guide (Packman, et al., 2011) can be downloaded from the Australian Stuttering Research Centre website. The guide is summarised briefly.

Stage 1

Overview

During Stage 1 the child and parent[2] attend weekly 45-60 minute consultations with a speech pathologist. During initial consultations the parent is trained to identify stuttered and stutter–free speech, and then to apply parent verbal contingencies. A typical Stage 1 consultation begins with the speech pathologist and parent discussing the SRs and treatment of the previous week. The child is then engaged in a conversation so that a representative speech sample can be obtained and a SR can be assigned. Agreement within one scale value between the speech pathologist and parent is required.

Next, during a typical consultation, the speech pathologist observes and supports the parent demonstrating verbal contingences with the child. Treatment modifications are trialled and program adjustments are made. Once the parent demonstrates an ability to apply verbal contingencies and facilitate appropriate conversations, beyond-clinic treatment can begin. Presentation of verbal contingencies for children differs depending on the nature of the stuttering, the parent-child interaction, parenting style, preference of the child and the child's reaction to treatment. Thus, program elements are individualised.

Measurement

Speech measures during the Lidcombe Program treatment process are used to determine stuttering severity, to determine whether treatment is reducing stuttering, and to judge when children have met criterion speech performance. Speech measures are also used to monitor change over time and to assess clinical progress against published benchmarks (Jones, et al., 2000; Kingston, et al., 2003; Rousseau, et al., 2007).

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[2] The Packman et al. (2011) program guide uses the term *parent*; this includes parents, caregivers and guardians.

Severity Ratings

A measurement of severity encompasses the frequency and duration of stuttering episodes, together with the degree and type of stutters during a speech sample (O'Brian, Packman, Onslow, & O'Brian, 2004). The SR scale is a 10-point scale, where 1= no stuttering, 2=extremely mild stuttering and 10= extremely severe stuttering. The speech pathologist and parent use this scale during the Lidcombe Program treatment process to identify severity changes, within and beyond the clinic. Usually, parents chart their child's typical daily severity. In some cases they may record several SRs in different situations or to reflect variability.

Percentage of Syllables Stuttered

Percentage of syllables stuttered (%SS) is a measure of the frequency with which stuttering occurs in a speech sample (O'Brian, et al., 2004); an overall percentage of stuttered syllables is calculated. This can be done by the speech pathologist, typically during a speech sample that is obtained at the beginning of a treatment session. In previous Lidcombe Program manuals, this was a mandatory speech measure during the treatment process. As a result of a review completed recently, it is now an optional speech measure that may be used during the Lidcombe Program (Bridgman, Onslow, O'Brian, & Block, 2011; Bridgman, Onslow, O'Brian, Block, & Jones, 2011).

Verbal contingences

Parent verbal contingencies are applied to the child's stuttered and stutter-free speech throughout the Lidcombe Program. Application of parent verbal contingencies has been empirically associated with stuttering reduction (refer to Chapter 2); thus they are presumed to be the treatment agent. The specific contingencies are outlined below.

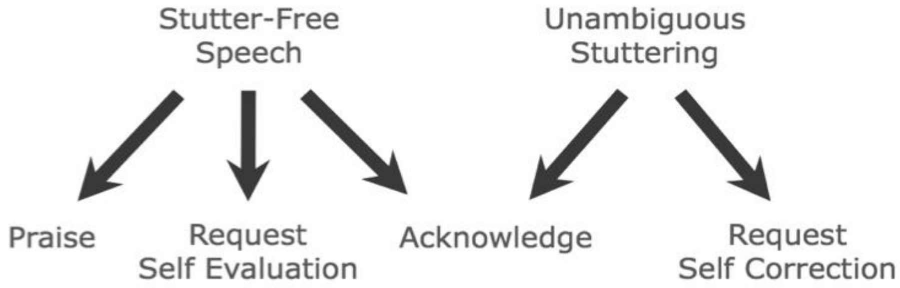
Stutter-free Speech

There are three verbal contingencies that can be applied to stutter-free speech. First, the speech may be praised specifically, for example, "super smooth words", "nice smooth talking", "so smooth". Second, stutter-free speech may be acknowledged, for example "that was smooth", "I heard your smooth talking", "you are being smooth", "you sound smooth today". The final verbal contingency for stutter-free speech is a request for self-evaluation. In this instance, following a stutter-free utterance from the child, the parent may ask the child whether the utterance was stuttered or not. Such phrases may include "was that smooth?", "were you smooth then?" This final contingency is not essential. Parent verbal contingencies applied to stutter-free speech should occur more often than for stuttered speech. This reinforces the desired speech behaviour and ensures that the experience remains positive for the child.

Stuttered Speech

There are two contingencies that can be applied to unambiguous stuttering. The first is acknowledgement, for example "that was bumpy", "I heard a bump", "you got stuck at the start". The second is request for self-correction. This is when the child is asked to correct a stuttered syllable, word or phrase. The child may be asked to "say that again". If there is uncertainty about whether a speech moment is stuttered or stutter-free, it is deemed ambiguous, and no parent verbal contingency of any kind is applied. Figure 5.1 depicts the parent verbal contingencies applied to stuttered and stutter-free speech as previously detailed.

Figure 5.1: Parental Verbal Contingencies, from Packman, A., Onslow, M., Webber, M., Harrison, E., Lees, S., Bridgman, K., et al. (2011). The Lidcombe Program of early stuttering intervention treatment guide. Reproduced with permission from the Lidcombe Program Trainers Consortium.



Treatment during Structured Conversations

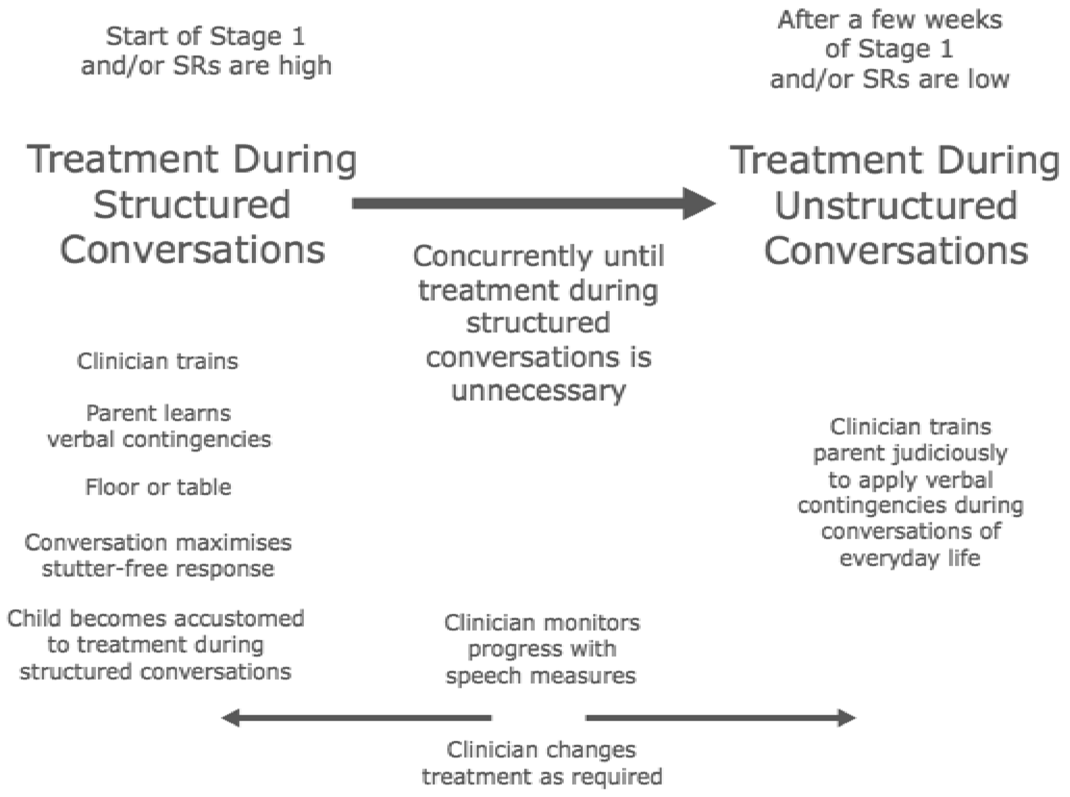
Parents administer the Lidcombe Program treatment in their children's natural environments. Once trained, the parent applies verbal contingencies to the child's speech during daily 10-15 minute conversations. Initially, these conversations are structured. The parent may engage the child while playing a game or sharing a book, or any activity that may be conducive to structured interactions. The purpose of structured conversations is to maximise the child's ability to produce stutter-free speech, and for the child to become used to receiving verbal contingencies.

Treatment during Unstructured Conversations

The structure of treatment conversations can decrease as a child uses more stutter-free speech. Such conversations may then be held during daily routines and interactions. As Stage 1 progresses, context-specific incidents of increased stuttering may be targeted with treatment conversations. For example, if the child's stuttering tends to increase during dinner time, or when engaged in imaginative play with siblings, the parent may facilitate conversations and apply verbal contingencies during those times.

The level of structure required to facilitate stutter-free speech is likely to vary at any time. This is due to the stuttering severity at that time. Transitions between structured and unstructured conversations are initially introduced by the speech pathologist, who teaches the parent how to adapt conversation levels beyond the consultation accordingly. The relationship between treatment during structured and unstructured conversations is illustrated in Figure 5.2.

Figure 5.2. The Relationship of Treatment in Structured and Unstructured Treatment Conversations, from Packman, A., Onslow, M., Webber, M., Harrison, E., Lees, S., Bridgman, K., et al. (2011). The Lidcombe Program of early stuttering intervention treatment guide. Reproduced with permission from the Lidcombe Program Trainers Consortium.



Program Speech Criteria

A set of criteria must be reached during Stage 1 to enter Stage 2 of the Lidcombe Program; hence both stages are considered performance-based. A child must obtain beyond-consultation parent SRs of 1-2 with the majority of scores being 1 to enter Stage 2. The consultation rating must also be rated 1 or 2 by the speech pathologist. Both criteria must be obtained for three consecutive weekly consultations.

Stage 2

Overview

Regardless of how effective Stage 1 of the Lidcombe Program might be, children are required to complete Stage 2 in order to have the greatest chance of long-term maintenance of Stage 1 treatment effects (Webber & Onslow, 2003). Stage 2 sessions consist of the parent and speech pathologist reviewing beyond-consultation SRs and reduction of beyond-consultation parent verbal contingencies. Parents monitor children's speech for signs of relapse, and may need to temporarily increase the use of verbal contingencies accordingly.

Consultation schedule

Weekly consultations are replaced with a programmed maintenance schedule of 2 weeks, 2 weeks, 4 weeks, 4 weeks, 8 weeks, 8 weeks and then 16 weeks. This schedule may alter if the speech pathologist thinks that withdrawal of treatment needs to be observed more closely or if at any time the child returns to Stage 1.

Program speech criteria

During Stage 2, children must continue to satisfy Stage 2 entry criterion of SRs of 1-2s with majority of 1s. Parents are asked to record SRs for the week preceding the scheduled consultation. If criterion is not met, a child may return to Stage 1.

Lidcombe Program Experimental Arm

Previous low-tech telehealth studies have produced less favourable efficiency outcomes compared to clinic treatment. This could be attributed to the deconstruction of the Lidcombe Program treatment process in which speech pathologists used role plays, video examples and gave delayed speech pathologist observations of parent treatment or ratings of speech samples from recordings. It is the hypothesis of this candidate, therefore, that if Lidcombe Program treatment sessions are conducted via webcam exactly as they are within the clinic, there should be no difference in the outcomes of both arms of the trial. Given this hypothesis, the decision was made not to adapt or supplement the current treatment process for webcam delivery in any way. Consequently, all parents and children receiving treatment via webcam participated live from their homes, just as they would within the clinic. No additional supplementary materials were used for the experimental group, nor did the clinician alter treatment practices in any way. Webcam parents and children were engaged with directly, just as with the clinic parents and children.

There is no standard clinic benchmark for the use of supplementary materials, program deconstruction or the use of recordings in lieu of real-time treatment. Therefore, including them would have added two variables to the experimental group.

Summary

A Phase III RCT was undertaken to investigate the question, Are outcomes following webcam delivery of the Lidcombe Program at least as good as those following standard clinic delivery? Forty-nine participants were randomised into either a clinic or webcam treatment group. Efficiency measures were reported in relation to the number of sessions, weeks, and clinician hours to attain entry into Stage 2 of the Lidcombe Program. Stuttering severity was measured by parent-reported SRs before the participants commenced treatment, at completion of Stage 1 of the Lidcombe Program, and 9 and 18 months after randomisation. These outcome measures were used to determine efficacy by comparing stuttering reduction in the two groups. Qualitative and quantitative data from parent questionnaires were also obtained.

PART 3: RESULTS

CHAPTER 6: RESULTS

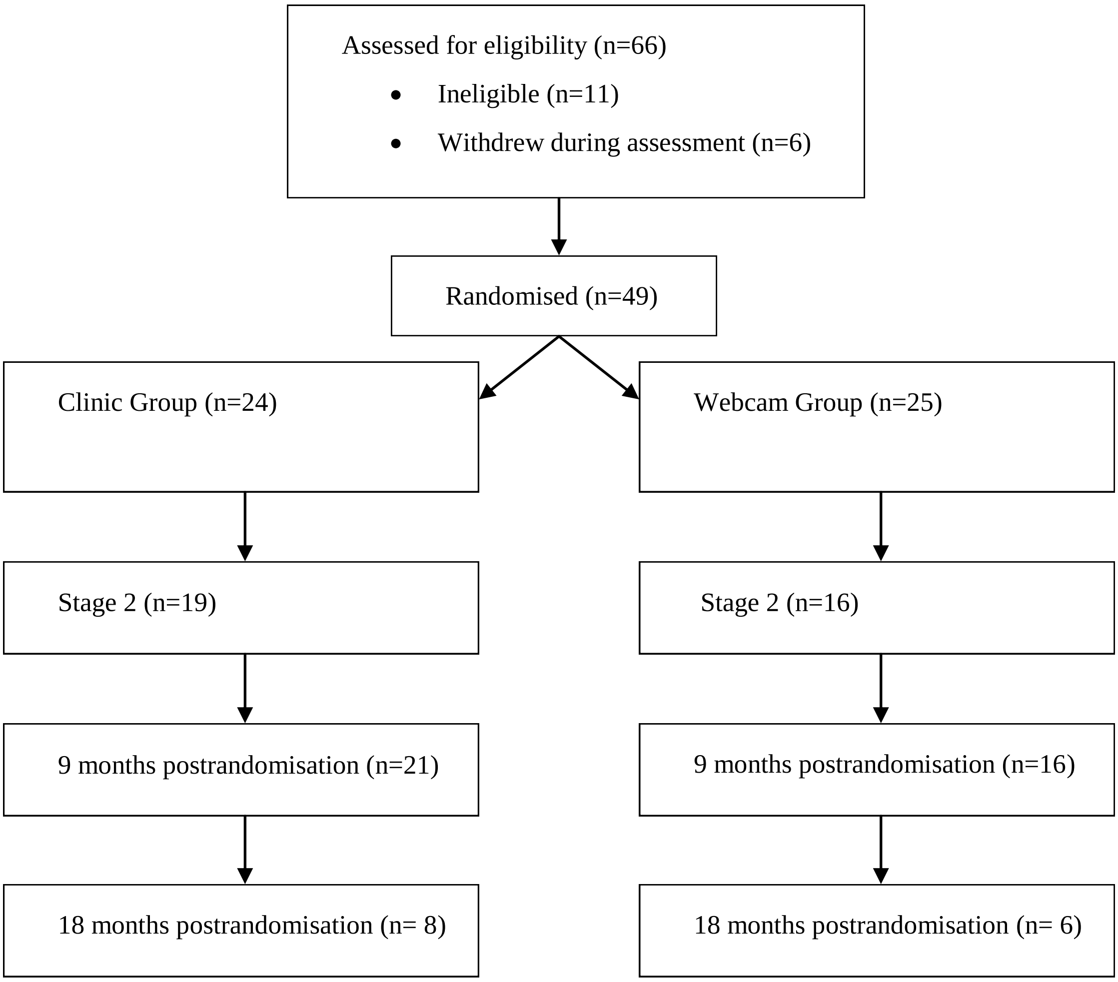
Trial Progress

Data-Set

This thesis reports on the following data-set. Pretreatment data are reported for all 49 participants. Data for all 43 participants active in the trial at 9 months postrandomisation are also reported. Stage 2 entry data was available for 35 participants who reached Stage 2 by December 31, 2012. In early January 2013, 5 participants remained in Stage 1 and 9 participants had withdrawn from the RCT. Due to time restrictions associated with the student's candidature, not all 18-month data were collected in time for inclusion in this thesis. Individual data used in this thesis are tabled in Appendix F. Figure 6.1 depicts the participant flow for the data-set used in this thesis.

Trial Flow Chart

Figure 6.1. Trial Flow Chart



Ineligible Participants

Eleven children did not satisfy the eligibility criteria for this study. They were ineligible for the following reasons: failed Skype test (n=4), diagnosed pragmatic disorder (n=2), significant language disorder (n=2), significant behavioural difficulties (n=2), and significant speech sound disorder (n=1).

Withdrawals during Assessment

Six children withdrew during the assessment process. Parents of the withdrawing children reported the following reasons: death in the family (n=1), wanting to pursue articulation treatment (n=1), new baby in the family (n=1), pursuing private treatment (n=1), and no reason (n=2)

Withdrawals during the Trial

Nine participants withdrew during the course of this RCT. Table 6.1 details the group, number of weeks and consultations until withdrawal, number of cancellations and appointments not attended, and the reason families provided for withdrawing.

Table 6.1

Summary of Participant Withdrawals

Group	Number of weeks	Number of consultations	Number of consultations cancelled	Reason
Clinic	0	-	-	did not want to travel to clinic
Clinic	16	0	0	treatment demands/difficulty attending regular consultations
Clinic	21	16	5	difficulty attending regular consultations
Clinic	40	23	17	treatment demands/difficulty attending regular consultations
Webcam	4	1	1	treatment demands/difficulty attending regular consultations
Webcam	6	3	2	child noncompliant
Webcam	12	5	6	treatment demands/difficulty attending regular consultations

Webcam	44	20	22	treatment demands/difficulty attending regular consultations
Webcam	77	52	17	treatment demands/difficulty attending regular consultations

Clinical Progress

Consultation Attendance

The clinic group cancelled a mean of 40% more consultations than the webcam group. An unpaired t-test showed there was no significant difference in attendance between the clinic group (M=5.6, SD=5.16) and the webcam group (M=3.43, SD=2.7); t(29)=-1.71, p=.1.

Altered Program Criteria

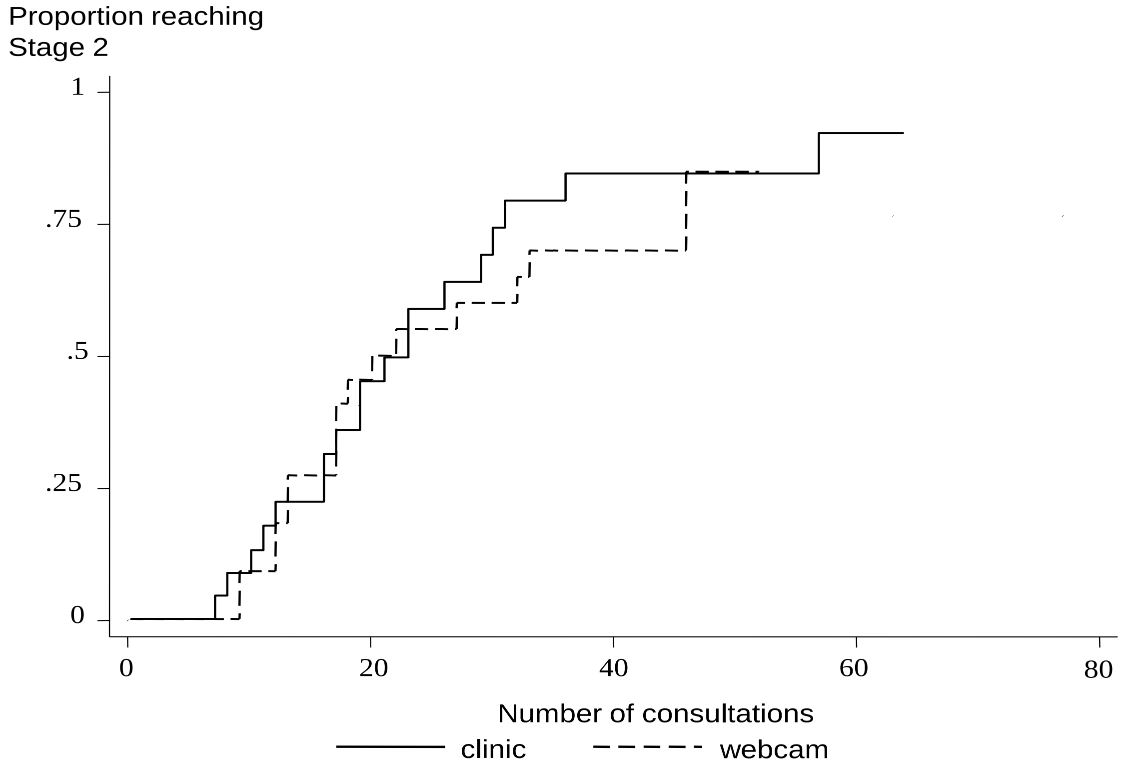
An amendment to the treatment protocol was made during the course of the trial. When participants were not demonstrating treatment progress and families were experiencing burnout, a new Stage 2 entry criterion was introduced. Participants progressed to Stage 2 if after the 9-month assessment the mean of the daily SR remained unchanged for 12 consecutive weeks. The goal of Stage 2 was then to sustain these severity levels while withdrawing daily treatment and decreasing the frequency of clinical consultations, as per the Lidcombe Program treatment guide. The candidate believed that further stuttering reduction was unlikely to occur following 9 months of Lidcombe Program treatment and 3 months of no change. Rather than have these participants withdraw and be counted as drop-outs, the altered criterion allowed them to remain in the trial. This criterion change is unprecedented. It is not detailed in the treatment guide, nor has it been published in any Lidcombe Program study to date. Altering the criterion in these circumstances has, however, been recommended by Lidcombe Program experts in the clinical domain (Sheedy, Erian, & Nikolas, 2012).

Primary Outcomes

Number of Consultations to Stage 2

The Kaplan-Meier plot of consultations to Stage 2 in Figure 6.2 shows similar distributions for both groups, with a median of 23 consultations for the clinic group and 20 for the webcam group.

Figure 6.2: Kaplan-Meier Plot of Consultations to Stage 2



An unpaired t-test showed no significant difference for the number of consultations taken between the clinic group (M=21.6, SD=11.9) and webcam group (M=19.8, SD=10.1); t(33)=.49, p=.63.

Speech Pathologist Hours to Stage 2

An unpaired t-test showed no significant difference between the number of speech pathologist hours taken by the clinic group (M=14.6, SD=8.9) and webcam group (M=11.4, SD=6.4); t(33)=1.23, p=.23.

Secondary Outcomes

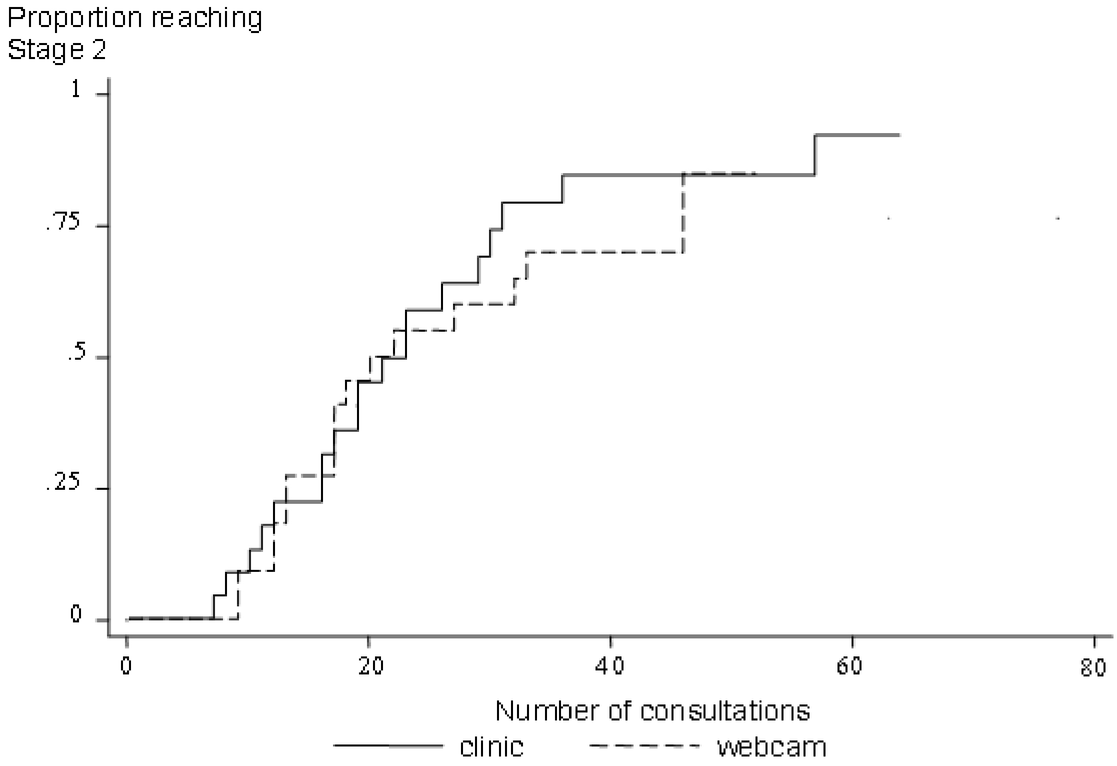
Consultation Duration

An unpaired t-test showed a statistically significant difference in the mean duration of Stage 1 consultations between the clinic group (M=40.5, SD=5.2) and the webcam group (M=33.7, SD=4.8); t(33)=4.01, p<.001, 95% CI [3.3, 10.23]. The mean webcam consultation duration was 17% less than the mean clinic consultation duration.

Weeks to Stage 2

Sensitivity analysis showed similar results for the number of weeks for both groups compared to the number of sessions, p=0.94. Analysis for the censoring for the altered criterion show no difference. Consequently, results reported herein are not sensitive to the altered Stage 2 criterion. The Kaplan-Meier plot of number of weeks for Stage 2 entry shows a similar distribution for the clinic and webcam treatment group. The plot in Figure 6.3 shows that the webcam group had fewer children than the clinic group in Stage 2 at 40 weeks.

Figure 6.3: Kaplan-Meier Plot of Number of Weeks to Stage 2



An unpaired t-test showed no statistically significant difference in the number of weeks required to reach Stage 2 entry between the clinic group (M=27.3, SD=14.7) and the webcam group (M=23.4, SD=11.0); t(33)=.90, p=.37.

Parent Severity Ratings

Comparison of pretreatment entry SRs showed no significant difference between the clinic group (M=4, SD=1.6) and webcam group (M= 4.6, SD= 1.8), p=0.21. There was no evidence of difference in mean SRs reported at 9 months postrandomisation between the clinic group (M=1.8, SD=1.6) and the webcam group (M=1.9, SD=1.0), p=0.88. A 95% CI on the difference is -0.56 to 0.49, showing no evidence of a difference between groups. Further, unpaired t-tests showed no significant difference between control girls (M=2, SD= 0.67) and webcam girls (M=1.6, SD=0.3); t(5)=0.84, p=.21, or between control boys M=1.8, SD=0.4) and webcam boys (1.9, SD=1.35); t(21)=-0.3, p=.75 at 9 months postrandomisation.

Parent Satisfaction with Child's Level of Fluency

Unpaired t-tests showed no significant difference between the clinic group (M=2.3, SD=1.47) and the webcam group (M=2.8, SD=2.39); t(33)=-.62, p=.54 at 9 months postrandomisation, or at 18 months postrandomisation; control (M=1.5, SD=0.76), webcam (M=1.3, SD=0.49); t(12)=0.66,p=.52.

Parent Questionnaire

Unpaired t-tests showed no difference between the mean response scores for the first five questions common to each group questionnaire. Table 6.2 shows the mean response scores, t-value and p value for each group.

Table 6.2

Mean responses to Parent Questionnaire (1= totally agree, 2= agree, 3= neither agree/disagree, 4= disagree, 5= totally disagree)

	Clinic	Webcam	t value	p value
Speech pathologist-parent relationship	1.4	1.1	1.5	.14
Speech pathologist-child relationship	1.3	1.3	0.42	.68
Learning severity rating scales	1.9	1.6	1.18	.25
Learning treatment	1.8	1.6	0.83	.41
Modify treatment	1.8	1.6	1.03	.31

Clinic group

When asked about the ease of travelling to the clinic each week, 55% of clinic parents strongly agreed or agreed that travelling to the clinic was easy, whereas close to one-third of control parents disagreed. The mean weekly travel was 66 minutes (range 15-180). The mean maximum travelling time parents reported that they would consider was 99 minutes (range 10-300). Two parents reported no maximum, stating they would travel as far as required. Two-thirds of the parents strongly agreed or agreed that they could receive treatment via webcam, 26% were neutral and 11% reported they would not consider receiving treatment via webcam.

Webcam group

Among the webcam group, 44% of parents reported that they had not used Skype before; 85% of webcam parents strongly agreed or agreed that webcam made attending sessions easier, the remaining 15% neither agreed nor disagreed. None of the parents agreed that webcam treatment in their home was invasive; 94% either disagreed or strongly disagreed. Among the webcam parents, 81% reported that the quality of Skype allowed them to see and hear the speech pathologist clearly. The remaining 12.5%, or two parents, reported that the quality was not adequate to see and hear the speech pathologist. There was no clear relationship between internet provider and Skype quality, as evident in Table 6.3.

Table 6.3

Internet Providers Compared with Parent Response to "The quality of the Skype connection allowed me to see and hear the clinician well at all times".

Internet Provider	Totally agree	Agree	Neither agree/disagree	Disagree	Totally disagree
Telstra	1	4	1		
Optusnet	1	2	1		
iiNet	1		1	1	

Netspace	1	
Primus		1

Additional Webcam Parent Responses

Additional responses from the 16 webcam parents whose children had entered Stage 2 are summarised below. A full transcript of the responses is listed in Appendices H and I.

The most common response listed from the parents was convenience. Four of the parents used the word "convenience", and a further eight made comments related to not needing to travel and saving time. Two of these parents also made comments about "minimal effect on work-life balance" (L434[3]) and being "able to work afterwards" (L434). Three additional parents made comments related to siblings, with two commenting on scheduling treatment time during baby's sleep time and one parent commenting on "not having to take young children to clinic" (L432).

Additional comments were made in relation to the child attending treatment while in their home. These included "easier at home; child might have felt more comfortable at home" (L450), "comfortable" (L419), "treatment place is home, so 'normal' as possible" (L451), "comfortable for child, at home with own things" (L458), "being in own environment, having his own belongings; helped him keep up a conversation because familiar items" (L434), "home becomes pseudo-clinic" (L454).

The main disadvantage commented on related to the quality of webcam or internet connectivity. Comments relating to this varied. Some were mild: "only difficulties were when there were glitches getting the connection or sound established but we easily overcame with Kate giving instructions" (L402), "sometimes service would drop out but it was good 98% of the time" (L427), "occasionally bad line" (L430), "occasional connection problems" (L451) and "child occasionally frustrated about not having whole visual of some activities" (L458). Two other parents reported connection difficulties that had greater impact: "poor quality connection were a problem" (L434) and "two to three times webcam connection really disappointed us" (L419).

Additional comments appeared related to the child's attention or behaviour. These included: "child might have concentrated better face to face with eye contact" (L450), "you have to keep hold of the child to keep in front of the camera. Might be easier in clinic as child can get up and move around without disrupting consultation" (L454) and "child could run around and out of the room due to personality" (L419). Finally, three parents commented on the child-speech pathologist interaction or relationship. One parent reported "sometimes not as personal as face to face" (L443). In contrast, another reported that the child "really wants to meet Kate" (L434).

Additional Analyses

Predictor Variables for Treatment Time

Additional analysis of log-normal censored regression found no evidence of an effect of age on the number of consultations to Stage 2 entry (p=.18). Evidence supported the expectation that higher pretreatment parent-reported SRs were associated with greater number of consultations to Stage 2 entry. This increase was calculated to be 13% for each additional unit increase in severity rating p=.03, 95%CI [.01,.27].

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[3] This is the de-identified participant number.

Unpaired t-tests compared individual case history or demographic variable with efficiency outcomes to determine if there were any other possible prognostic indicators. A table detailing the mean weeks, consultations and speech pathologist hours required for entry into Stage 2 for the clinic and webcam groups is in Appendix J. No statistically significant difference for treatment times (weeks, consultations, speech pathologist hours) was found between boys and girls; stuttering onset less than or greater than 12 months; negative or positive family history of stuttering; previous treatment; assessment only or no previous treatment; or combined family income $30,000-$59,999 or $60,000- $80,000 plus.

Summary of Results

The results reported in this chapter support the hypothesis of this thesis: that Skype delivery of the Lidcombe Program is at least as good as clinic treatment in terms of both efficiency and efficacy. The outcomes are summarised:

Primary Outcomes

- There was no statistically significant difference between the number of consultations required by each group to complete reach Stage 2 entry.
- There was no significant difference between the number of speech pathologist hours required by each group to reach Stage 2 entry.

Secondary outcomes

- There was a significant difference in the mean consultation duration required by each group to reach Stage 2 entry.
- There was no difference in the number of weeks required by each group to reach Stage 2 entry.
- There was no difference in parent-reported SRs between the two groups at the 9-month assessment.
- There was no difference in parent satisfaction with child's level of stuttering between the two groups at the 9-month assessment.
- There was no difference in rapport, parent learning of SRs, implementing and modifying treatment between the two groups.

Additional Findings

- Higher pretreatment stuttering severity was associated with increased treatment time required to reach Stage 2 entry.
- Age was not associated with treatment time required to reach Stage 2 entry.
- No other prognostic indicators were found to be significant.

The next part of this thesis discusses the results of this RCT. Additional clinical trends observed by the candidate throughout the course of the trial are also discussed, followed by considerations for translation and future research.

PART 4: DISCUSSION

CHAPTER 7: DISCUSSION OF RESULTS

Webcam service delivery of the Lidcombe Program could allow greater and more timely access to early stuttering intervention for preschool children. Early access to effective treatment could reduce the number of children who continue to stutter into their school years, adolescence and adulthood. Such prevention of chronic stuttering could reduce the possible lifelong impacts of stuttering on wellbeing and life participation. The results of this trial are discussed.

Data-Set: Connectivity

From the 49 participants recruited for this Phase III RCT, outcome data are reported for 35 participants at entry to Stage 2 of the Lidcombe Program, and 43 participants 9 months postrandomisation. During recruitment, 11 children were found not to be eligible. It is worth noting that only 4 of these (37%) were ineligible due to poor Skype connectivity. That was based on a Skype test administered 3 years ago. It is thought that connectivity will continue to improve as technology increases and the National Broadband Network is implemented in Australia. A further consideration relating to the data-set is the number of withdrawals from the clinic group. All four withdrawing families cited travel and difficulty attending regular appointments as a reason they withdrew. This suggests that travel can be a burden not just for distant or isolated families but also for metropolitan families.

Altered Program Criteria

Efficiency calculations found no statistical difference between participants who entered Stage 2 with an altered program criterion when compared to the standard criterion. This should be interpreted with caution, however, as this finding it limited to the data-set of this RCT. To determine whether this altered criterion is safe for translation, long-term follow-up data are needed to determine whether participants sustained their Stage 2 entry severity levels. This will be assessed when the 18-month follow-up data are complete and reviewed.

Primary Outcomes

Number of Consultations to Stage 2 and Hours to Stage 2

There were no statistical differences between the number of consultations and speech pathologist hours taken to Stage 2 entry between the clinic and webcam group. This is significant in relation to efficiency. First, this outcome supports the noninferior hypothesis of this thesis. Second, this is the first telehealth Lidcombe Program study that has not produced lower efficiency outcomes. Thus, use of this method could be viable within the community.

Secondary Outcomes

Consultation Duration

There were two interesting findings in relation to mean consultation duration. First, the mean for both groups was less than the 45-60 minutes recommended by the manual. This finding suggests that, for some participants at least, sessions may be shorter. This could be due to the change in speech sampling. This was the first study to adopt the 2011 treatment guide recommendations of assigning a SR to a representative speech sample rather than completing a %SS rating. The difference could also be attributable to the candidate's individual practices.

The second finding of interest was that the mean duration of Stage 1 consultations was significantly different between the two groups, with webcam sessions being 17% shorter. While this could be view as an efficiency saving, this finding should be interpreted with caution. It is the impression of the candidate that some sessions at least were shortened at the request of parents who wanted to attend to competing demands. On the other hand, several participants consistently required sessions longer than the mean. Further studies are required to determine optimal session duration for both clinic and webcam delivery.

Weeks to Stage 2

The efficiency finding of no difference between the groups for the number of weeks to Stage 2 has similar significance to the number of consultations taken to Stage 2 entry. It also suggests that, although webcam permitted greater attendance and treatment continuity when families were unwell or away, it did not lead to completing the program in fewer weeks.

Parent Severity Ratings

The parent SRs attested to the efficacy of the program. The finding of no difference between groups in parent-reported SRs, and consequently stuttering reduction, also supports the hypothesis of this thesis. Furthermore, pretreatment SRs for both groups demonstrated that participants' severity ranged from mild to more severe stuttering. This is important to consider in relation to webcam. These findings suggest that low pretreatment stuttering severity is not required for successful outcomes using webcam. Rather, regardless of the delivery method, children with any level of pretreatment stuttering severity can be expected to achieve results that would not differ.

Parent Satisfaction with Child's Level of Fluency

Like the SRs, parent-reported satisfaction ratings did not differ between the two groups. This finding further supports the hypothesis of this thesis and perhaps suggests that parents' perceptions of the service delivery did not affect their perception of their children's stuttering.

Parent Questionnaire

The results of the parent questionnaire showed no difference in parental reports about developing rapport and learning the main Lidcombe Program components. These findings are significant. First, one of the greatest misconceptions reported to the candidate during the course of this trial from speech pathologists is that rapport cannot be developed with a parent and child using webcam to the degree that it can in the clinic. These findings of this parent questionnaire do not support that perception. Second, previous studies have deconstructed the Lidcombe Program and taught key components in alternative ways, rather than adhering to the standard manual. Yet these findings from the parent questionnaire suggest that not only can Lidcombe Program components be taught to and understood by webcam families in the same way as clinic families, there does not appear to be a need to provide webcam families with supplementary materials when standard Lidcombe Program treatment practices are adhered to.

Clinic parent responses confirmed that travel can be a burden for metropolitan families. The range of maximum travel times that parents indicated they would consider was interesting. While the mean was 99 minutes, and two families said they would travel as far as they had to, one family stated 10 minutes as the greatest travel time they would accept. Such a range adds further support to travel and distance barriers being just as significant for metropolitan families as distance or remote families.

Webcam parent responses added to the positive efficacy and efficiency findings. Reports supported the notion that the webcam made attending sessions easier and that previous use of Skype was not required for successful webcam sessions. Information relating to poor connectivity and internet providers suggests that locality appears to be the crucial factor that affects connectivity rather than specific providers. Again, this should be less of an issue as connectivity increases in coming years.

Ancillary Investigations

Predictor Variables for Treatment Time

Analysis of possible predictor variables for treatment is important. It can allow speech pathologists to identify specific factors that might cause treatment time to take longer than anticipated. Given that age was not found to be a variable, it can be said that a child of any age (between 3;0 to 6:0) is suitable for webcam delivery. The opinion that some children are too young was not supported by any evidence from this trial. Further, the finding that income was also not a predictor supports the contention that webcam delivery is feasible for a broad demographic rather than a certain income level.

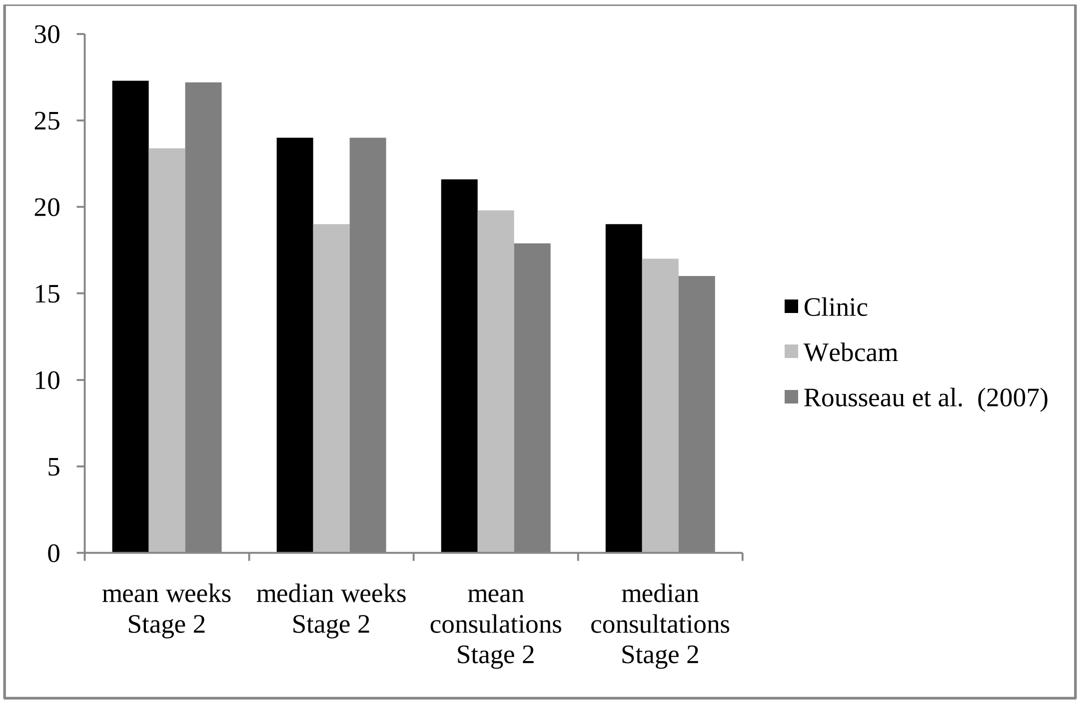
Comparison to Previous Lidcombe Program Studies

Given the positive efficiency and efficiency outcomes, together with the parent reports, the strength of this study can be further examined by comparing the outcomes to previous Lidcombe Program studies.

Efficiency Measures

Efficiency measures are the main outcomes reported from Lidcombe Program treatment studies. The median number of weeks and number of consultations required in this RCT to achieve Stage 2 entry, calculated using both the Kaplan-Meier plot with survival analysis and descriptive statistics, produced results similar to those obtained by Rousseau et al. (2007). The mean and median numbers of weeks and sessions to reach Stage 2 are comparable to the previously published outcomes study completed by Rousseau et al. (2007). Figure 7.1 provides the relevant comparisons.

Figure 7.1: Mean and Median Weeks and Consultations for Groups in Webcam or Clinic Delivery of the Lidcombe Program and Rousseau et al. (2007)



Severity

The finding in this study that pretreatment severity was a predictor for treatment time is consistent with findings reported in previous Lidcombe Program studies (Jones et al., 2000; Kingston et al., 2008; Rousseau et al., 2007).

Community Translation Study

The recent Lidcombe Program publication (O'Brian, Iverach, Jones, Onslow, Packman, & Menzies, 2013) reported outcome data obtained from 31 speech pathologists and 57 preschool Lidcombe Program clients from community clinics. The relevance of the community translation study to this RCT is that most speech pathologists adhered to the treatment guide, with the exception of consistent 45-60 minute sessions and requests for parent demonstration of treatment. The finding of O'Brian et al. about session duration is consistent with the consultation length outcome data for this RCT. The difference with regard to parent demonstration is not consistent with this RCT. In fact, the concern of O'Brian et al. over the lack of parent demonstration is of considerable interest to this RCT, which is the first Lidcombe Program telehealth study to include live parent demonstration of treatment. In comparison, this RCT reported similar data in relation to both the number of weeks required to reach Stage 2 and the parent-reported SRs 9 months postrandomisation.

Telehealth Studies

Methodology

Previous Lidcombe Program telehealth studies have proven effecacious but not as efficient as clinic models. In reviewing these studies the initial emphasis was on the medium used for delivery.

As reviewed in Chapter 4, the initial telehealth studies deconstructed the Lidcombe Program treatment process. They used telephone consultations, exemplar tapes and video recordings in lieu of live, real-time treatment consultations (Harrison, Wilson, & Onslow, 1999; Wilson, Onslow, & Lincoln, 2004; Lewis, Packman, Onslow, Simpson, & Mark, 2008). The most recent telehealth Lidcombe Program trial used Skype to allow live, real-time interactions with both the parent and child (O'Brian, Smith, & Onslow, 2012). The treatment was still modified from standard practice, however, as the parents were taught treatment mostly via speech pathologist explanation and role-play rather than live speech pathologist demonstration with the child. Overall, the children were reported to be less involved in the webcam consultations than children would be in a standard clinic consultation (O'Brian, Smith, & Onslow, 2012).

Efficiency Outcomes

When efficiency outcomes are compared, all four previous studies report far longer treatment durations compared to standard clinic benchmarks. The initial three studies (Harrison, et al., 1999; Lewis, et al., 2008; Wilson, et al., 2004) cited the low-tech telehealth method as the likely reason for this discrepancy. On initial analysis, it appears that all authors placed emphasis on the relationship of the outcomes to the service delivery, citing telehealth as the reason poor efficiency outcomes were achieved. Yet an additional variable to consider is the impact of the treatment program modifications on outcomes. All studies deviated from standard Lidcombe Program practice and all studies took longer. This would suggest that the modifications to key Lidcombe Program treatment components could at the very least contribute to reduced efficiency outcomes. This notion is further supported by the efficiency outcomes of this study; in order to compare clinic service delivery to webcam delivery of the Lidcombe Program, the treatment program itself remained unaltered. Unlike the previous Lidcombe Program telehealth trials, webcam participants in this study received Lidcombe Program treatment as per the treatment guide, with no modifications. This was done to limit the variables to the service delivery method only.

The rest of this chapter therefore reviews the current treatment guide, dismantling the Lidcombe Program to report how the planned treatment fidelity was achievable, and to report any amendments required for the webcam group such as were evident in the previous telehealth studies.

Lidcombe Program Components Using Webcam

Parent Verbal Contingencies

As detailed in Chapter 5, parent verbal contingencies form the basis of Lidcombe Program treatment. In this study, parents in both groups were taught how to apply parent verbal contingencies during live demonstrations with their child, as detailed below under Stage 1. The candidate then observed this use and coached the parent accordingly.

Parent reports from the questionnaires relating to learning how to apply Lidcombe Program treatment showed no significant difference between the webcam and clinic groups. This is therefore evidence of the safety and efficacy of teaching parents how to use parent verbal contingencies via webcam with their children.

This finding could also underscore the importance of this Lidcombe Program treatment component in relation to treatment efficiency. This is the only Lidcombe Program telehealth study that consistently demonstrated the parent verbal contingencies live with the child in each consultation rather than in role plays (O'Brian, Smith, & Onslow, 2012) or video examples (Wilson, Onslow, & Lincoln, 2004; Lewis, Packman, Onslow, Simpson & Jones, 2008).

Measurement of Stuttering

As discussed in Chapter 5, measurement is a vital component of the Lidcombe Program treatment process. Measurement is used to evaluate treatment efficacy, progress, and variability beyond treatment consultations, and is necessary to determine stuttering reduction and entry into Stage 2. The treatment guide details the need to train the parent to use the 10-point severity scale, to consistently calibrate ratings between the parent and speech pathologist, for parents to complete daily SRs and for the speech pathologist to record these ratings, together with consultation severity ratings.

Data from the parent reports showed no significant difference in parent-reported ability to learn how to use SRs. This is consistent with the candidate's opinion of being able to teach both groups equally.

Additional speech measures using %SS counts were taken at times for both groups. Clinic participants could see the candidate rating their speech so this could possibly be viewed as an overt rating. Webcam participants could not see the candidate rating; this rating could therefore be considered as a covert rating. It is not known whether covert rating could affect the clinic participants' fluency, resulting in the speech sample being less representative.

Interestingly, though, clinic parents often reported that the speech samples obtained within the clinic were not representative of beyond-consultation speech, stating that their children were often more fluent in clinical consultations. Webcam families did not report this. This could therefore be suggestive of the fluency-inducing nature of clinical spaces contrasted with the more natural and therefore representative communication environment in a child's home. As this is the first trial to provide children with home-based treatment, there is no precedent for this comparison.

Weekly Clinic Consultations

Stage 1 Lidcombe Program consultations were scheduled weekly for all participants. Weekly consultations are recommended to take between 45 minutes and 1 hour. This component of the Lidcombe Program treatment guide was used when booking treatment consultations. However, it was not adhered to for either group, with the consultations finishing once the typical Lidcombe Program consultation components were completed. As discussed in Chapter 6, this resulted in mean consultation lengths for both groups of less than 45minutes. The webcam consultations were significantly shorter than the clinic consultations. This is evidence for the efficiency of the webcam delivery.

Treatment during Structured and Unstructured Conversations

There was no difference between the groups as to the structure or dose of conversations during Stages 1 or 2. Parent reports from both groups showed no significant difference in their ability to modify their child's treatment.

Programmed Maintenance

Stage 2 was managed in the same way for both groups. Consultations were scheduled less frequently and treatment was systematically withdrawn. The available follow-up data at 18 months postrandomisation supports this process. Both groups adhered largely to the recommended schedule in the treatment guide.

Stage 1

The current treatment guide (Packman et al., 2011) details 13 components of a typical Stage 1 clinic consultation. As discussed previously, not all telehealth studies have implemented all components consistently. Both groups in this RCT did, however, receive a consultation that included all 13 components, as detailed below.

Measurement

The initial three Stage 1 consultation components relate to eliciting a speech sample that the parent and speech pathologist can then rate using the severity scale. Typically for the clinic group this occurred by the candidate engaging the child in conversation during play. At times, a parent was asked to evoke conversation from a child who did not engage with the candidate. For three clinic participants, the candidate regularly left the room and observed the child speaking with the parent from behind an observation window, as the child did not speak initially in the presence of the candidate.

During webcam consultations the children typically engaged in direct conversation with the candidate. At times, resources would be used to help elicit a sample. This involved either the candidate using some toys, or the child sharing some toys from the home environment. Parents were sometimes involved in eliciting a speech sample if the child was less talkative with the candidate. There were no instances of the child not speaking in front of the candidate. This suggests a comparative strength of the home webcam-based speech samples: children may be more comfortable to speak within their own environment, using their own resources, rather than in a clinic.

The next two Stage 1 consultation components relate to the recording of SRs. In both groups parents were able to simply relay their previous week's daily SRs for the candidate to enter into the child's file. The only difference between the groups was that clinic parents sometimes left their graphs at home. This resulted in the candidate being unable to adapt treatment as accurately or engage in problem-solving discussions as specifically as if the data were present. Given that the webcam parents were within their homes, they always had access to their daily SRs. Such compliance with presenting daily SRs within home-based consultations could be seen as advantageous for webcam treatment.

Parent Demonstration of Treatment

Both groups of parents demonstrated treatment procedures with their children live in front of the candidate during each consultation. Clinic families mostly used resources present within the clinic. They would be asked to select a resource that was most similar to the treatment activities they had completed at home. While the parent was able to demonstrate card games with cards provided, or structured turn-taking games with games provided, this demonstration often required the parent and child to adapt to a new or unfamiliar resource. In contrast, the webcam families used their own resources. This resulted in the candidate observing more accurately the exact resources that were used for daily home-based treatment.

This latter point supports the use of observation by speech pathologists of clinic families with their own resources, by asking parents regularly to bring items from home, or to provide recordings of treatment that occurs within the home, as recommended by Swift, O'Brian, Onslow, & Packman (2012).

The candidate engaged in discussion with both groups of parents about the treatment and resources and suggested changes for the coming week. This included discussing available resources with both groups. At times, both groups were provided with paper-based activities if they did not have access to appropriate resources. All webcam families had access to printers, so these resources could be emailed to them for immediate use. Clinic families were provided with the resources in the consultations or via email following the consultation.

Speech Pathologist Demonstration and Teaching

The candidate demonstrated treatment changes routinely with children in both groups. Despite previous studies omitting this component from telehealth service delivery, this component was feasible for all webcam children. Overall, all resources that were used within the clinic were also used via webcam. The only difference was that with webcam the child could not physically touch the resources. Despite this, webcam children still engaged in typical structured conversations using cards, turn-taking games and books. For less structured conversations they engaged in play sequences with pretend-play sets, picture description, narrative story activities and general conversation.

For both groups, the candidate was able to teach the parent treatment changes. The parents were then able to demonstrate these changes. The clinic families were able to use the original resource the candidate had demonstrated with. The webcam families were able to use the same resource too if the candidate helped with holding it up in front of the camera. They were also able to source similar items from their home and practise with these resources. This was considered advantageous as on many occasions the candidate could assist with adapting the family's resources for the purpose of treatment. This could also be done for clinic families, but only through discussion rather than direct observation.

Problem-Solving

Planning and problem-solving discussions occurred with parents from both groups at the end of each consultation. The content and detail of these discussions was the same for both groups. At times, clinic discussions were brief or shortened due to the behaviour of the children and their siblings. This was not a problem for webcam families as the children were often dismissed from the consultation and the parent and candidate could engage in the discussion without interruption. Thus it was also possible for parents to raise issues or concerns that they did not want to discuss in front of their children. These issues ranged from concerns about their child's stuttering, behaviour, bullying or progress to the parent's personal stressors about finances, relationships, attitudes and wellbeing. These personal stressors were always raised in the context of factors that were affecting treatment compliance or the child.

Such opportunities to engage in parent-speech pathologist conversations without children present could be seen as an advantage of webcam treatment. It could also be a component that is added to standard clinic practice.

Preparing for and Entering Stage 2

Both the clinic and webcam groups prepared for Stage 2 entry during their consultations. The application of the standard and altered criteria for entry were the same for both groups. There was no deviation in this practice for webcam children.

Stage 2 Procedures

Children from both groups were prescribed the scheduled Stage 2 consultations. Several children from both groups required adjustment to this schedule to sustain criterion. As for the Stage 1 consultations, parents in both groups presented the required SRs, a speech sample was measured with a SR by the parent and candidate, and treatment decisions were discussed.

Individualising the Lidcombe Program

The Lidcombe Program treatment guide details child age and behaviour, stuttering severity, parent and child personalities, family circumstances and parent treatment history as factors to consider when individualising the treatment process. Due to the RCT design, these factors could not be accounted for when assigning participants to treatment groups. The only exception to this was the stratification for severity which occurred to ensure that different severity levels were represented in both groups.

As a result of this design, both groups contained children with different age, behaviour, speech and linguistic ability, personality, temperament and stuttering severity. Similarly, parents with different personality, treatment history and family circumstances were represented in each group. Treatment history for several parents in both groups related to their previous experience with delivering Lidcombe Program treatment rather than being adults who stuttered or had stuttered.

The randomisation allowed for individualisation of the program within the clinic and webcam service model as detailed. Individualisation was comparable between both groups for all factors with the exception of family circumstances.

Webcam delivery allowed for far greater individualisation for family circumstances. Many families were able to occupy other children within the home; they scheduled consultations for sleep times of younger children or set older children up with activities in other areas of the house. In several cases, the second parent came home in time for the consultation to start, but could not have come home early enough for the treating parent to leave and travel to the clinic with the participant.

Absence of the need for transport and travel was significant for one of the webcam families, who did not have access to a car and would have spent nearly 2 hours each way with a 4-year-old on public transport each week to attend consultations. With webcam treatment they simply walked into their lounge room. Cancellations due to limited transport access were evident in four clinic families who each cancelled appointments several times due to unreliable or problematic cars.

Webcam delivery was also more conducive to coping with health difficulties within families. Many webcam participants still attended consultations when the parents or siblings were sick. Clinic families cancelled appointment in these instances. Furthermore, two clinic families had weekly attendance significantly interrupted due to the treating parent's health. In both cases the parents requested their consultations be conducted via webcam as they felt they could have attended the consultation within their own home, but were not able to drive and travel to the clinic.

Such family circumstances provide a range of issues when we consider ability to access service. These issues are far broader than the initial rationale for telehealth, which was to provide access for remotely based populations. In many examples discussed above, families were unable to attend the clinic and consequently cancelled consultations. In these instances, however, as they all had to pass a webcam test as part of the assessment criteria, they could have been offered a webcam consultation in lieu of their clinic consultation. Such situations suggest the desirability of a hybrid model of service delivery, with families attending either in the clinic or via webcam, depending on the situation at any given time. Further support of this model is the parent report data: no differences were reported for rapport, learning measurement or treatment, so a hybrid model should not affect treatment progress or outcomes.

Summary

Primary and secondary results support the hypothesis of this thesis. Efficiency outcomes were consistent with previous Lidcombe Program clinic-based treatment studies and better than previous Lidcombe Program telehealth studies. This could in part be attributable to the methodology of the studies, and the use of webcam allowing standard Lidcombe Program treatment processes to be implemented. Additional clinical observations are discussed in the following chapter.

Chapter 8: Clinical Observations

The efficiency and efficacy trends, together with the parent reports, support the hypothesis of this thesis. While pleasing, this finding was not overly surprising. There were, however, some less predictable clinical observations that emerged during this RCT. Such observations are discussed in relation to this trial, and are then considered in relation to translating the findings from this RCT.

Unexpected Observations During Webcam Delivery

Convenience

The first unexpected observation that emerged during this study relates to convenience. The rationale for webcam treatment was related to increased access to service for those who cannot readily access it. The interesting trend in this study, however, was that despite having access to local speech pathology services, this metropolitan sample still found webcam treatment more convenient. The convenience extended beyond increased access; it provided a family-friendly service option that was easier for families with young children.

Travel

As previously discussed, webcam parents did not have to pack the children into the car and travel to the clinic. Rather, they just had to turn on their computer and bring the child into the designated area of their house. The negative impact of travel was reported by clinic families, despite many living within a 10-kilometre radius of the clinic site. This further supports the argument that distance can be a barrier or limitation of any clinic-based service.

Siblings

During the trial, webcam consultations were most often scheduled while other siblings were out or occupied. Hence they interfered less in webcam consultations.

Illness

Many webcam families attended consultations when the participating parent or siblings were unwell. Some families even attended when the participant was sickIn three instances children could still attend while they had contagious conditions that would have otherwise stopped them attending the clinic.

In one case, a webcam parent was diagnosed with a progressive degenerative neurological disorder that affected the parent's health and ability to drive daily. Webcam allowed greater attendance as the parent and child could still attend consultations on days the parent would not have been able to drive. This differed greatly from the situation of a clinic parent who had multiple sclerosis. This parent had to cancel clinic consultations due to health, inability to drive, and the need to travel interstate periodically for treatment. On each occasion the parent and child travelled interstate they asked if they could attend the treatment consultations via webcam. Due to trial protocol they could not, but clinically allowing webcam attendance would have permitted better attendance continuity and progress throughout treatment.

Holidays

The majority of the clinic families cancelled at least one consultation due to being away on a holiday. In two instances, clinic participants went on extended holidays overseas during Stage 2. Consequently the children had significant breaks in treatment consultations and returned from holidaying with increased SRs. In contrast, most webcam families attended their treatment consultations while on holidays.

The Impact of Convenience

Thus far the convenience of webcam has been discussed in relation to the flexibility it provided to assist in better, more sustained attendance. There were, however, some clinical differences that were observed among a small group of webcam families who at times suggested that webcam was too convenient. Although these are examples of single consultations or occurrences, they are occurrences that did not ever occur with the clinic families. Examples include:

- one family attended a webcam consultation while still in their pyjamas, not having eaten breakfast;
- one child was woken from an afternoon nap during the treatment consultation;
- several families attended the consultation while they had visitors in the house;
- one family regularly came home straight from being out at the time of the appointment;
- one parent tried to set the child to speak for the initial speech sample while the parent finished preparing dinner;
- some parents attended with a cup of coffee; one family sometimes gave the child and sibling lunch during the consultation;
- two families had their after-school snack during the consultation;
- one family was found to be out shopping when they were contacted after not being live at the time of their consultation.

Over the course of the treatment process it was difficult to determine whether such different behaviours were perceived as a disadvantage of webcam. Given the positive efficiency and efficacy outcomes, however, observations of such behaviour could be seen as giving the speech pathologist a more accurate or representative view of family dynamics within the natural environment. Such observations were insightful for the candidate to better understand how daily treatment was implemented beyond consultations. It could also be suggestive of the overall priority or importance given to the stuttering treatment within the parent and child's life. Such observations led to the candidate wondering how representative the behaviours of clinic families were of their home-based behaviours.

Clinical Behaviour Issues

Apart from some different within-consultation behaviours, overall behaviours towards the treatment process differed for a small group of webcam families. This was related to beyond-consultation behaviour and are detailed further.

Attendance

Statistically, there was no difference in consultation attendance between the groups. However, the behaviour relating to cancellations differed for some webcam families. Most cancellations occurred on the day of the scheduled consultations, a large proportion being within 15 minutes of the consultation. Webcam families were also less likely to inform the candidate if they were running late. It is unclear if this was related to the issue of treatment readiness, discussed later in this chapter, or whether they valued this service delivery less. It is also possible that these families would have behaved in the same way if they had been randomised to the clinic group. An additional point to consider is the lack of fees or cancellation policies that would more commonly support attendance and cancellations in a timelier manner. It is the experience of this candidate that when working in both public and private settings, such policies result in families cancelling fewer appointments, and with greater notice.

Consultation Duration

The quantitative data showed a shorter mean length of consultations for webcam delivery. It is interesting to report that a small group of families at times requested shorter consultations to allow them to attend to competing lifestyle demands. This request was most often made at the beginning of the treatment consultation. This rarely occurred for clinic families. If clinic families had competing demands, they negotiated starting their appointment earlier or an alternative treatment time prior to the consultation.

Consultation Times

To ensure that all families were given equal opportunities, and to avoid bias to either group, consultation times were offered between the standard operation hours of the treatment clinic: 8am to 6pm on weekdays. For some working parents in both groups this meant they had to alter their working hours or days depending on the availability of consultation times at the time they entered the trial. Interestingly, parent responses to available treatment times differed between the groups. Clinic parents either swapped days of work, attended the appointment with the child on the way to childcare or school, or they arranged to finish work early and bring the child to their appointments late in the afternoon. Webcam parents as a group appeared less flexible. They often requested times outside the designated working day or made the appointment as early or late as they could.

Such requests could be related to parent perception of convenience and consequently, the negative impact on the children's compliance and possible progress. Clinic children who attended 5pm or 5:30pm appointments were compliant and engaged for the duration of the consultation. In these cases the children had not been home, and so a late appointment was an extension of their day outside of the home. Webcam children treated at 5pm or 5:30pm differed greatly despite comparable ages. These children had returned home from their day at school or childcare, and accordingly wanted to play within the home, spend time with their other parent or siblings, or eat their dinner. Consequently, these early evening webcam consultations were shorter in duration. On two occasions the candidate felt that the time of the consultation had too great an effect on its quality, and an earlier consultation time was negotiated.

Such differences also occurred for clinic and webcam appointments at 8am or 8:30am. Clinic families were on time, with the children dressed, having eaten breakfast and prepared for the consultation and their day outside the home. Webcam families were often late to these early appointments; on occasion the children or parents were not dressed, they had not eaten breakfast or were in the process of eating it. Such behaviours and lack of readiness again affected the quality of the treatment consultations, resulting in the candidate negotiating readiness with one family, and making the consultations later for another family.

Finally, several webcam families requested treatment consultations at weekends. The reasons related to difficulties with attending consultations around work, childcare, siblings and general lifestyle demands. The interesting point, however, is that due to the randomised nature of the trial, these demands were presumably similar for the clinic group, but not a single clinic parent requested a consultation outside the standard clinical hours. Such a difference could relate to the direct versus indirect clinical boundaries, or assumptions that relate to these boundaries. It could also somehow relate to the perceived convenience and flexibility for webcam families of webcam delivery, or to the lack of boundaries they thought were in place for the candidate. Although such requests were denied during this trial, such a flexible option could suit both consumers and speech pathologists within the community.

Communication

All participant parents in the trial were given the candidate's office phone number and email address. Many families from both groups were also given the candidate's mobile phone number. Throughout the course of the trial a clear trend emerged as to methods of communication. While the candidate communicated with all families via telephone, webcam parents most commonly communicated via email or at times by text message. Several webcam families also used the messaging function on Skype. Clinic families most commonly phoned either the office or the mobile phone. No clinic families sent text messages.

This trend is interesting. Given the randomised nature of the trial, it cannot be said that webcam families were initially more technologically minded or based, but somehow treating them via webcam suggested or reinforced communicating via alternative means. This trend could be related to convenience or the way they perceived the candidate, being technologically minded, or communicating via these media because the treatment was offered using technology.

Treatment Preparation

During the RCT, an interesting trend began to emerge in relation to preparing for a consultation. This was observed to varying degrees and is explored below.

Being Present

The majority of webcam families were present and ready live at the time of their scheduled call. There was, however, a small group of families who were not as listed previously on page 72. An overall trend that appears to emerge from these issues relating to presence concerns organisation: although webcam consultations required less organisation, the negative of that could be that parents did not organise accordingly to attend.

Parents

Lidcombe Program treatment requires the parent to deliver treatment appropriately to the child in everyday conversations. Attendance at treatment consultations is also the sole responsibility of parents, as young children cannot do this independently. For these reasons, preparation for webcam consultations was required by parents. First, the parents needed to organise their day and children around being at home for their scheduled appointment. Second, parents needed to source the appropriate resources and the severity rating sheet to have it accessible at the computer prior to the consultation start. Finally, parents needed to prepare their children.

Child

For the majority of webcam consultations, the candidate would place the call and be greeted by the participating parent and child ready to start. Some parents preferred to have initial discussions without their child and then call the child into the room when required. When parents had prepared their children, telling them in advance they would be having the consultation, the children would come immediately and be compliant. When parents had not pre-warned their children about the consultation, the children would often protest about being removed from their game, siblings, television show or computer game. This then appeared to affect compliance for the rest of the consultation. Sometimes the parent took the lead in trying to coax the child to stay; in many instances, however, the responsibility was left to the candidate to engage the child. A social story was provided for one particular family, resulting in far greater readiness and compliance.

Resources

Parents who had resources with them at the computer to occupy their child and then use in treatment were more successful in keeping their child at the computer for the duration of the consultation. Parents who did not provide their child with something to do during parent-speech pathologist discussion were often interrupted by the child, or the child would leave the room. This also occurred at times if the parents had not pre-planned their treatment resources. They would either select items they could quickly access or try to treat the child without games or resources. This often led to the child being uninterested and the conversation dissolving. Such demonstration also limited the feedback the candidate could give the parent about treatment, as it was often not representative of the treatment they had completed during the week.

The Impact of Not Being Prepared

When parents and children did not appear prepared for consultations or treatment, the candidate supported them in being better prepared by providing suggestions and engaging in problem-solving discussions. Recommendations included ensuring that parents were home with their children with enough time to prepare for the consultation. This involved allowing time for snacks and going to the toilet, and pre-warning the children that the consultation was going to take place; usually with a general comment in the morning when discussing the day and then again 15 or 30 minutes prior to the consultations. In some cases play tasks or activities were discussed prior to consultations, with the emphasis being on not removing a child from doing something, but rather naturally finishing the task in time for the consultation to begin. Parents were also encouraged to prepare the resources they required prior to the consultation. In some instances special activities were brought out during treatment consultations.

Readiness of Clinic Children

It became apparent during the course of the trial that the trend of treatment readiness was largely implied for clinic families. The children were told where they were going when they were put in the car. They then sat and waited in the car, most doing nothing while they were driven to their clinic treatment consultation. They entered the treatment clinic and often waited again for the candidate. Finally, once they entered the consultation, they had often been prepared and waiting, but had also not been engaged in anything particularly exciting immediately prior to treatment so were naturally more willing to engage in the consultation.

It can also be assumed that clinic parents undergo a similar preparation process. They prepare their child for the consultation, travel to the consultation and sit and wait in reception. This contrasts to webcam parents who did not necessarily have to prepare for the consultation more than 5 minutes prior to the scheduled time.

Clinic families most often did not need to prepare resources in advance; the candidate provided them in the clinic. For most families who attended the clinic with siblings, the candidate also provided resources for the siblings.

Treatment Representativeness

The differences in treatment readiness between the two groups could again raise questions about the representativeness of clinic families. This is because the logistics of travelling to the consultation naturally prepare the parent and child. Further, the candidate most often took responsibility for all the resources used in the consultations. Therefore, although initial trends suggested that webcam treatment might be too convenient and not conducive to focused, sustained treatment, the consultations provided greater insight into how parents approached daily treatment at home.

Defining the Clinical Space

A further difference that became apparent for the webcam group throughout the treatment process was defining the clinical space. Like treatment readiness, this was indirectly implied for the clinic group. The clinic group attended a treatment clinic, received treatment in a clinic room with clinic resources. As many of the parents and families would have undergone similar routines when visiting other health professionals for appointments, the clinical space and rules or boundaries were largely pre-defined. The clinic families were naturally conditioned by past experiences to attend on time, wait in the waiting room, focus on the candidate and respond to candidate-led pragmatic or behavioural cues during consultations. The children themselves appeared to just know to ask before they used something, only played with toys that were presented, stayed in the room and responded to candidate instructions. The clinic parents rarely attended to outside interruptions and always asked before they could leave the room.

This behaviour was consistent with that of the majority of webcam families, but some families did not adhere to common or assumed clinical boundaries. Several parents often left their child with the candidate without warning. They were observed to answer their phone more often and attend to non-treatment-related tasks.

Beyond the consultation, this same group of webcam families was less likely to cancel consultations or communicate if they were running late. They forgot several consultations or asked to have consultations shortened due to competing lifestyle demands. Such behaviour suggests the need to explicitly define clinical boundaries when using webcam as the service delivery model.

Behaviour Management

The greatest challenge for the candidate was behaviour management. This issue was twofold. First, webcam participants at times appeared on the surface to be less compliant. Superficially, this could just be related to the service delivery model. When the "difficult" children were observed more closely, however, there appeared to be some common variables: their parents had not prepared them for the consultation and were not prepared themselves. These children were often not seated correctly and were not given things to do during the initial parent-speech pathologist discussion. In these parent-child dyads, the parents appeared less direct or controlling in their general management of their child, allowing the child more freedom in behaviour and compliance. It became evident that with these families, the success of the consultation was largely based on the parent's ability to control their child's behaviour. While these children were usually compliant for the candidate, it was often when the parent was responsible for behaviour that compliance diminished.

On reflection, it was these incidents that highlighted a difference in candidate, parent and child behaviour across the two settings. Within the clinic the candidate was direct with respect to the rules of the clinic room, how the resources were to be used and what behaviour was acceptable. During webcam consultations, however, the candidate did not have a physical space to assist with setting the boundaries. Rather, the treatment space, for the children at least, was within the family home. Consequently, the children tended to behave in the same manner they did at home.

Again the questions are raised: was this a negative of webcam delivery?A common occurrence in home-based treatment? Or simply a more representative insight into family dynamics that are not controlled or perhaps apparent within a clinical environment?

Speech Pathologist Responsibility

The second issue related to clinical space concerned safety and duty of care. Within the clinical environment, if a parent leaves a child with the speech pathologist while the parent steps out of the room, the speech pathologist is responsible for what happens to the child in her care. The speech pathologist and child share the same environment so the speech pathologist can intervene, physically if required. During a webcam consultation, however, the speech pathologist cannot do this. This raises concern as to the speech pathologist's responsibility for a child who is left unattended during a webcam consultation. Although all parents were told to remain with their children during the consultation, a small number of parents often left their child unattended at the computer speaking to the candidate. The candidate spoke to the parents about this on each occasion. These instances raised the alert about a need to further develop a contingency plan for such an occurrence, to protect the speech pathologist and further define duty of care. Possible contingencies could include the speech pathologist stating that she will immediately hang up a call if a child is left unattended, abdicating any responsibility for supervising the child, or stating that the child is the sole responsibility of the parent for the entirety of the consultation.

Consultation Attendees

A final incident related to treatment space concerned consultation attendees. Any additional attendees to clinic consultations were obviously visible to the candidate. In such instances the third party and candidate were introduced.

During a webcam consultation, however, it was less obvious to the candidate who was present during the consultation. Only people in view of the camera could be seen. At times siblings could be seen and heard. This did not impact on the consultation or the candidate. During a particular consultation, however, a third party was heard laughing in response to a candidate instruction; when asked, the participating parent reported that a second adult had been observing the consultation from the beginning. The candidate was surprised that the person had not been introduced, but the parent just stated the third party was watching. Following this incident families were asked to disclose all consultation attendees at the beginning of each consultation.

Neutral Space

During the course of the RCT it became apparent that there was not a shared concrete treatment space for the webcam group. Rather, the interaction took place between the two sites. In most consultations this neutral treatment space allowed both parties to be comfortable and safe in their own environments, supporting the balance of the parent-speech pathologist relationship. Given the use of technology, this could be considered a "virtual" treatment space.

Webcam Relationships

As reported in Chapter 5, the parent outcome questionnaire detailed parents' views on how well rapport was developed with the candidate via webcam. During the course of the trial, these reports seemed consistent with what was experienced by the candidate. Both groups contained parents who engaged in general discussion and who spoke only of their children and the treatment.

During the course of Stage 1, parents from both groups reported difficulties, spoke of stressors, and demonstrated emotional responses during these discussions. While this occurred in both groups, a difference emerged when the candidate reflected on how well the parent could be supported. Often, at times of upset or distress, clinic parents would avoid eye contact or direct the candidate toward their child. It was evident that they did not want to further discuss their difficulties or be upset in front of their child. On several occasions the candidate phoned the parent later in the day but the time had passed to discuss how they were feeling and on all occasions the parents dismissed their earlier behaviour.

In contrast, webcam families engaged in further discussion of the challenges of treatment or life stressors affecting their ability to apply treatment, or they expressed their fears and concerns for their child who stuttered. At these moments, the parent would dismiss the child from the room, start the consultation without the child, or wait until the consultation was finished and they could send the child off to do something else. Such discussions were viewed by the candidate as part of the problem-solving element of the Lidcombe Program treatment process. In allowing the parent to communicate those feelings and discuss in detail and length the challenges or emotions, the candidate could then support the parent and adapt the program accordingly.

This ability to extend parent discussions via webcam is not just a benefit of this service model. It also highlights the potential need for standard Lidcombe Program treatment to include some exclusive parent-speech pathologist discussion time. Such feelings of difficulty and emotion as expressed by the webcam parents are also consistent with the previously reported study by Goodhue et al. (2010) of mothers' experiences.

Nontreating Parent

During the course of the trial, more nontreating parents attended webcam consultations than clinic consultations. This was often requested in advance by the families who reported that the other parent wanted to know what the child was doing, how they could be involved, or that they would be home from work and could observe. Two webcam families had both parents attend regular webcam consultations and learn to treat their child, a third family alternated between parents depending on the parents' shift-work roster. This did not occur in the clinic, although one clinic family changed the treating and attending parent twice during the treatment process, due to work and family commitments.

Participants

During initial consultations, the webcam children often asked where the candidate was,. As the consultations progressed and the children became familiar with the consultation routine they asked eagerly what resources the candidate had to share with them. The children also often prepared their own resources. They were also more likely to relate the candidate's comments or questions to items they had in their home, leaving the computer to source the desired item. Clinic children rarely bought personal items from home to share with the candidate during clinic consultations. These observations support the individualisation of the Lidcombe Program as recommended in the standard treatment guide, as it also allows the speech pathologist to better know and understand the childs interests and everyday life.

Siblings and Family Members

Extended family and the alternate parents would often walk past the computer and greet the candidate. Although siblings were usually kept occupied during the consultations, both younger and older siblings sought interaction with the candidate. Siblings as young at 10 months responded to the candidate's interaction. Older siblings often asked if they could have a consultation with the candidate. They tended to be satisfied with a brief chat at the end of the consultation.

Summary

Clinical observations were also made during this RCT. Unexpected clinical observation related to convenience, clinical behaviour, treatment preparation, defining the clinical space, behaviour management and developing relationships. Such observations are significant as they do not appear to have been previously reported in the telehealth literature.

CHAPTER 9: TRANSLATIONAL CONSIDERATIONS

The main objective of this RCT was to determine if webcam was a noninferior service option for the Lidcombe Program. Based on the positive quantitative and qualitative findings of this trial, this chapter recommends how this model could be translated. Translation is considered in terms of providing supplementary information to the Lidcombe Program treatment guide and professional development for speech pathologists. Technological requirements and client suitability for webcam service delivery are also considered. Such translation may need to be investigated in further translational studies.

Supplementary Information for the Lidcombe Program Guide

The information detailed below is recommended to supplement the Lidcombe Program treatment guide for webcam delivery. These recommendations follow the format of the current guide (Packman et al., 2011).

Recommendations for Lidcombe Program Consultations

Assessment Clinic Consultation

Although assessments were not completed via webcam for this project, the following process is recommended for webcam stuttering assessments.

Initial Contact

During the initial contact with the parent the speech pathologist may offer a webcam assessment if the parent and child are unable to attend the clinic. The speech pathologist will explain that this service can be offered provided a sufficient internet connection between both parties will allow for quality audio and visual real-time exchange using webcam. The parent is required to have a computer, the internet and a webcam.

Webcam Test

To ensure the quality of the webcam is sufficient to conduct a real-time consultation, a webcam test is recommended between the parent and the speech pathologist, with both parties testing from the sites and using the equipment they intend to use for the webcam consultation. It is recommended that this test occurs prior to the assessment consultation; this allows both parties time to attend to any difficulties that arise during testing. Refer to Appendix E for a Skype Quality Test Template.

Preparing for the Consultation

To help parents prepare themselves and the child for the consultation it is recommended that the speech pathologist talk in advance about the likely structure of the consultation. First, the parent and speech pathologist will engage in discussion regarding the nature of the stuttering and the child's general case history. During this time the parent is recommended either to have someone else assist with entertaining the child or have an activity set up for the child close by. Then, once the child is required to engage in a speech sample, the parent is asked to have prepared some toys or items of interest that the parent or speech pathologist can use to obtain a speech sample from the child. The speech pathologist may suggest that the parent prerecord an audio or video sample that demonstrates the child stuttering. This can be shared with the speech pathologist via email or a secure file sharing website. This is currently recommended in the standard treatment guide.

The speech pathologist may also engage in a brief discussion about positioning, recommending that seating be considered to allow the child to sit on his or her own chair. If using a laptop, the speech pathologist and parent may discuss where would be appropriate to set up for the call, usually a contained room away from other family members and household distractions rather than open living spaces.

Webcam consultation rules should also be explained. These include: (1) the speech pathologist will place the call at the scheduled time, (2) the parent must remain with the child at the computer; the child is not to be left alone, (3) the parent is asked to cater for siblings during this time to avoid disruption and the parent is asked not to attend to anything else during the consultation. Finally, parents are asked to contact the speech pathologist in a timely fashion if they are unable to attend the consultation. Speech pathologists may take this opportunity to explain the attendance and cancellation policy of their workplace.

Placing the Call

At the time of the call, the speech pathologist will place a video call. First, the rules of a webcam consultation will be explained. This includes that (1) either party should communicate to the other at any time if the audio or visual connection is not sufficient, (2) in the case of a disconnection or ended call the speech pathologist will always be the person to reinitiate the call, (3) the parent is required to attend throughout the consultation, and (4) the child will not be left alone at the computer at any time.

Speech Pathologist-Parent Discussion

Once the consultation is underway, the speech pathologist and parent can engage in the clinical interviewing and discussion as listed in the Lidcombe Program treatment guide.

Obtaining a Speech Sample

Following the speech pathologist-parent discussion, the parent will be asked to call the child and seat the child in view of the camera. The speech pathologist will engage in discussion to elicit a speech sample. If the child does not readily engage with the speech pathologist, the parent may be asked to elicit a representative speech sample and the speech pathologist can listen and rate accordingly. If either the parent or speech pathologist feels the speech sample is not representative of the child's stuttering, an audio or video sample can be shared with the speech pathologist. This is currently recommended in the standard treatment guide. Once a representative sample is obtained or a recording is reviewed, the child can be dismissed from the consultation.

Clinical Recommendations

The speech pathologist and parent can then return to discussion, with the speech pathologist providing information as listed in the standard treatment guide.

Stage 1

As reviewed in Chapter 7, a typical Stage 1 consultation was not altered for webcam delivery. All 13 components as listed in the standard treatment guide were implemented. The recommendations below may help speech pathologists unfamiliar with webcam delivery to implement these components.

Measurement

Measurement can be applied to the real-time speech sample obtained during the webcam consultation. This process is described above in assessment recommendations. A sample may be elicited by the speech pathologist or the parent. Either party may use items to elicit a natural speech sample. Children at home might like to share a special toy or a piece of artwork, or the speech pathologist might like to use available clinic resources. Such resources could be similar to pretend-play sets that would be presented at the beginning of a standard consultation.

To assist with stuttering identification during this speech sample it is recommended that the parent either look at the child's face directly, or watch the video image on the screen. This will allow for identification of blocks or any secondary facial features. Both parent and speech pathologist are encouraged to maximise the image of the child on their screen during this sample to allow better observation. If the child becomes distracted by the image then it can be reduced.

Severity ratings can be taught and reviewed just as they would be in the clinic, with speech pathologist explanation and rating of real-time child speech samples. Parent SRs can easily be exchanged by the parent reading them out to the speech pathologist, or the parent entering them into a shared document as detailed in the standard treatment guide.

Parent Demonstration of Treatment

Parents are asked to have with them the resources that they used with their child for treatment during the week. These resources can then be readily drawn upon for parent demonstration of treatment. If positioning is not conducive to the task, the parent may complete the demonstration on the floor, ensuring that both parent and child are in view of the camera. If using a laptop computer, parents may consider setting the consultation in the area of the house where treatment activities were completed during the week. This could include in the kitchen if snack time, cooking or unstacking the dishwasher were treatment activities, or in the child's bedroom with a doll house or train set.

Speech Pathologist Demonstration and Teaching

The speech pathologist should prepare a range of resources to demonstrate treatment conversations directly with the child. These resources can be the same as those that would be presented in a clinic consultation. The speech pathologist may adapt the activity for webcam delivery, taking the child's turn at touching, opening or manipulating a resource. For example, in structured tasks this may include lifting the flaps in simple picture books, holding up the two cards for the child's turn in a memory game or spinning the spinner or rolling the dice in a turn-taking game. For less structured activities in construction or pretend play, the child may inform the speech pathologist which pieces or items should be placed where.

Problem-Solving

For review or problem-solving discussion parents may decide to allow their children to leave the room first to allow for uninterrupted, more in-depth discussion. Alternatively, they may set their child up with a task or play at the desk or on the floor before they start the discussion.

Stage 2

Preparing for Stage 2 and the Stage 2 procedures were consistent with the treatment guide and the previously detailed real-time speech sampling and parent-speech pathologist discussion.

Individualising the Lidcombe Program

Offering webcam delivery in itself may be a way of individualising Lidcombe Program treatment for families who have difficulty accessing clinics, have competing lifestyle demands, or may prefer the convenience of home-based treatment consultations.

Community Translation

The recommendations listed above, together with the standard Lidcombe Program treatment guide, could be used by Lidcombe Program trained speech pathologists to implement this service option within the community. Such recommendations will form the basis of a treatment guide once this RCT is complete.

There may, however, be important service and logistic considerations for speech pathologists before offering this service model, particularly if their workplace does not currently offer telehealth services. Some factors to consider are detailed below.

Service Provider

The potential advantages in offering webcam Lidcombe Program treatment are considerable for service providers. These could include increased cost efficiency of outreach services, increased direct treatment time offered, resulting in shorter waiting lists and greater throughput. It could allow public services in particular to offer more flexible, inclusive treatment options to families who face various access barriers. Such technology would also allow service providers to offer a consultation service or to develop specialist central clinics that can service undefined geographical areas or populations.

To establish a new service delivery model within a service, however, consideration needs to be given to the logistics. This may include developing service policies for speech pathologists using telehealth that may relate to competency, privacy and, in some cases, insurance. Policies may also include attendance and compliance agreements with consumers and minimum standard technical requirements for both parties.

The considerations for service providers are also relevant for individual speech pathologists. Benefits for individual speech pathologists from this service model include greater flexibility in work hours or location, as they do not need to be located at a clinic site. Occupational health and safety may also be considered for speech pathologists: they can use this service model to avoid working in isolated and potentially volatile environments; they would not be exposed to sick children or families; and they may be able to reduce or avoid the amount of driving required to outreach service provision.

A speech pathologist’s clinical skills should also be considered. The outcomes from this trial were achieved by a Lidcombe Program Consortium trained community speech pathologist who had access to specialist Lidcombe Program clinical supervision. Such outcomes could support community-based Lidcombe Program trained speech pathologists using webcam. The development of webcam skills or competency may, however, not occur at the same rate within the community. The caseload of the candidate in this RCT included Lidcombe Program clients for four full clinical days per week, whereas other community speech pathologists might only have a few Lidcombe Program clients on their caseload.

Considerations for Potential Clients

Technological Requirements

Requirements for webcam Lidcombe Program treatment relate to technology, the internet and resources. Potential families are required to have a computer and webcam, with internet that supports real-time audio and visual connection. This can be determined via the previously described webcam test. Webcams can be purchased from most general purpose department stores, technology stores and even some supermarkets. Alternatively, they may be purchased online.

Families who do not have access to such technology or the internet in their homes may be able to access suitable technology at a family member's house, the local school, community centre or neighbourhood house. Each of these has implications for confidentiality. It is recommended that the viability of such an option be carefully considered, especially whether accessing a computer and internet outside the home would be sustainable on a weekly basis for a number of months.

Parent

The success of Lidcombe Program treatment is largely due to the parent's implementation of the daily treatment requirements. For the webcam treatment group, the success of the consultations appeared largely related to the parent. Parents who prepared their child for consultations, organised their resources and could manage their child's behaviour during consultations appeared to have more productive consultations, with often more efficient outcomes, than parents who did not prepare or manage their children. It is recommended, therefore, that the parent requirements for a webcam consultation are explicitly discussed when considering offering webcam treatment. Parents should be informed what will be expected of them in terms of preparing resources and managing their child's behaviour. More general discussion of the parent's behaviour management style and ability to sustain their child's attention may help to determine if webcam is a suitable option for a family.

Such discussion also provides transparency for the speech pathologist. If expectations and requirements are clearly discussed and agreed to initially, it may be easier for the speech pathologist to engage in discussion with the parent about not preparing or managing the child. The speech pathologist could assist in problem-solving discussions should these issues arise. Finally, for families who may be unsure, or if a speech pathologist has reason to question the parent's ability, a trial consultation or block of consultations may be arranged to determine whether webcam consultations are sustainable.

Child

As referred to previously, the greatest factor in a child's compliance and participation in webcam consultations was viewed to be the parent's ability to prepare the child and manage the child during the consultations. No other significant predictor variables or hallmarks were discerned in the types of children being more or less likely to have successful webcam consultations. Age and severity were not found to be factors. In two cases, webcam participants were given the role of being responsible for the technology. This approach seemed to appease these two children who were observed to have a very "own agenda" presentation.

Resources

Given that resource materials are required for daily treatment in the Lidcombe Program, webcam families were not required to have any more resources than clinic families. Part of the initial weeks for both groups involves discussing the resource materials available to respective families and adapting them for treatment accordingly. The speech pathologist used general paediatric clinical resource materials for both groups interchangeably. These included books, cards, turn-taking games, magnetic and felt board, pretend-play sets, figurines and construction toys.

Webcam Treatment Models

While the outcomes are related to each respective treatment model as stand-alone methods of service delivery, clinical application could result in webcam being used in many ways for Lidcombe Program treatment.

Stand-Alone

Stand-alone webcam Lidcombe Program treatment may be offered to families who are unable to access clinics due to distance or lifestyle factors. It may be that webcam is the only option, or that it is an easier option. This could be related to transport, illness, siblings, time constraints, or simply a preference to stay at home.

Lidcombe Program treatment may be sought by families who want to access Lidcombe Program treatment specialists. This may involve speech pathologists treating clients in different suburbs, cities, states or even countries. Such speech pathologists may offer the entire treatment program; others may provide consultations or second opinions.

Hybrid

While the findings of this trial are limited to each method of service delivery independent of the other, community translation could include a hybrid model of delivery. A hybrid model would allow families the flexibility to use both models, attending some sessions in clinic and some using webcam. Although no supportive outcome data are available, it could be assumed that this model would be effective given that the outcome data for both methods were comparable. Such a hybrid model could be used to individualise the Lidcombe Program for many reasons. These may include, but are not limited to: general convenience; overcoming daily access barriers like illness in families, difficulty with transport, childcare or time constraints, or the birth of a new baby; support for treatment continuity while families are holidaying. Clinically, it could be beneficial to obtain live speech samples from beyond the clinical environment or to observe treatment within the home, using the family's own resources, This may also allow for treatment across various sites to support transfer or fluency in the child's environment.

Webcam may be offered to a family as a way of reinvigorating a lengthy Stage 1 treatment process. Finally, the use of webcam would allow clients to remain in the clinical care of their treating speech pathologist even if they move or need to be absent during the course of the treatment program.

Alternative Use

Alternative uses for webcam Lidcombe Program treatment may include using webcam to train others adults in the child's life who spend considerable time with them. Consultations could be conducted with child care workers, aides or grandparents responsible for weekly care. It could allow school speech pathologists to treat from a central site and offer more treatment consultations rather than travelling to schools. In such cases, parents may attend the webcam consultation with their child at school. Webcam technology allows a second parent to observe and participate in discussion from a second site; that parent may be unable to leave work to attend a consultation in the clinic or at home, but may be able to webcam into the consultation from a workplace for part or all of the time.

Beyond direct clinical use, webcam could allow specialist fluency speech pathologists to support or consult to community speech pathologists who are having difficulty. In this instance the parent and child might attend the primary speech pathologist's clinic and have a webcam consultation with a speech pathologist who is a specialist in stuttering treatment and management. This would allow the consultant to observe both the parent and speech pathologist demonstrating treatment, and for the consultant speech pathologist to make suggestions that can be immediately observed and implemented. The consultant speech pathologist could also directly model treatment approaches or changes with the child.

Summary

The findings of this RCT are positive, and support community translation of webcam service delivery for the Lidcombe Program treatment process. Such translations should include considerations related to service providers, speech pathologists and potential consumers, that include service logistics, resources, parent skills and speech pathologist skills. Outcome data support clinical application when the treatment process is considered by suitably informed, prepared and supported parents and speech pathologists.

CHAPTER 10: TRIAL STRENGTHS, LIMITATIONS AND FUTURE DIRECTIONS

The findings of this Phase III RCT support the noninferiority hypothesis: that outcomes following webcam delivery of the Lidcombe Program are at least as good as those following clinic delivery. The strength of these outcomes and indeed this RCT can in part be related to the RCT study design. A retrospective review is completed below.

Methodological Review

To assist with objective review of this Phase III RCT, Ost's (2008) recommendations for RCTs to provide empirical evidence will be applied. Although this framework was designed following a systematic review and meta- analysis for behaviour therapies, it is applied to this study as no comparable work has been published addressing RCTs in speech pathology. In the paper, Ost provided 15 recommendations to consider when planning RCTs. Those recommendations are intended for the design of studies that produce empirical evidence to support the experimental treatment, and are listed in Appendix K. In relation to this RCT, 12 of Ost's recommendation are satisfied, and are discussed below.

RCT Recommendations by Ost (2008)

First, Ost (2008) recommended appropriate control groups. Using "waiting list controls" or "treatment as usual" (p. 314) controls is not recommended. Rather Ost recommended using an "active" control (p. 314) treatment that "has been established as effective for the disorder in question" (p. 314). These three recommendations were achieved within this RCT: the control group participants were recruited for the purposes of the study, and their service delivery method had clinical trial evidence as detailed in Chapter 5. Next, a power analysis was recommended. This was completed, as detailed in Chapter 5.

Further, Ost (2008) recommended that the participant group be representative and that diagnosis occur by a suitably trained professional. As described in Chapter 5, this project used a community-ascertained participant group, with stuttering confirmed by a speech pathologist working in the area of stuttering. Next, Ost recommended that the participant group be randomised by an independent party and that researchers within the study be blinded to the randomisation. Participants in this project were randomised by a third party using a randomisation schedule.

Following randomisation, Ost recommended that measurement outcomes be "reliable and valid" (p.314); being both "specific to the disorder and general" (p.314). Given that the primary outcome measure related to efficiency, general measures of time were used: minutes, hours and the number of consultations taken to achieve entry to Stage 2. The secondary outcome measure used to determine efficacy was parent-reported SRs. They are considered reliable as they are they are the only mandatory measure listed in the Lidcombe Program treatment guide, and have research to support their reliability (Kully & Boberg, 1988; O'Brian, Onslow, Cream, & Packman, 2003).

Next, it was recommended that "at least a 1-year follow-up" be included in the project, with any additional interventions in this time being declared by the participant. It is unclear if this 1-year period should follow completion of treatment or initial participation. Follow-up assessments were completed during the course of this project for all participants. These assessments occurred 9 and 18 months after initial randomisation. Due to the nature of the Lidcombe Program treatment schedule, participants would have been at varying stages of their treatment during these assessments.

Ost (2008) then recommended that concomitant treatments be controlled for participants. This was addressed upon recruitment to the project, when participants were asked not to engage with or seek treatment from a speech pathologist independent of the project for the duration of their participation in the project. The impact of prior treatment or treatment knowledge was considered upon recruitment, with participants needing to have had at least a 6-month break from previous stuttering treatments. Participating parents with prior experience of Lidcombe Program treatment were disclosed.

It was further recommended that attrition rates be detailed when reporting results and data, and that drop-out analysis include all randomised subjects with an intention-to-treat analysis. In relation to withdrawals, the number, reasons and data available for these participants were reported. Intention-to-treat analysis was completed for efficiency and efficacy outcomes.

The final recommendation by Ost (2008) was to assess the clinical significance of the primary outcome measures. This was done by comparing the efficiency outcomes of number of consultations and weeks to Stage 2 with previously published benchmarks.

Overall, the design of this Phase III randomised clinical satisfied 12 of the 15 recommendations Ost (2008) provided for RCTs that are relevant to this RCT. The three Ost recommendations that were not satisfied by this trial are reported.

Ost (2008) Recommendations Not Met by This Trial

Three of Ost's (2008) 15 recommendations for RCT methodology were not satisfied in this RCT. They are considered limitations of this RCT and are now detailed.

First, following the use of outcome measures, Ost recommended that blind assessors be used, with regular measurement of inter-rater reliability to be measured throughout the course of assessment. Due to the data being parent-reported SRs, blind assessors were not needed for the data reported in this thesis.

The second recommendation relates to the candidate. It is recommended that "three or more properly trained therapists" deliver the treatment and that patients are subsequently randomised to these therapists "to enable an analysis of possible therapist effect on the outcome" (p.314). This limitation could be addressed in future research with replication studies independent of the original researcher.

Finally, the third recommendation not applied in this RCT was the assessment of treatment adherence and speech pathologist competence. Ost (2008) recommended that all treatment consultations be recorded, with 20% then independently reviewed. During this project all consultations were recorded. Due to the incomplete data-set for this thesis, however, treatment compliance and candidate competence will not be reviewed until the project is complete. Treatment compliance is largely addressed in Chapter 7 where Lidcombe Program fidelity and treatment guide compliance are detailed for webcam service delivery.

Future Directions

This RCT is the first known RCT of its size to trial webcam treatment for preschool children within their own homes. The findings are significant, as no significant difference was found in relation to efficiency and efficacy of treatment. This was also the first Lidcombe Program telehealth study to result in such promising efficacy. Although the outcomes are exciting, they are limited to this RCT. Future research resulting from this RCT is therefore considered.

Research

Lidcombe Program Treatment

Software

The findings of this Phase III RCT were technically limited to the use of Skype. Investigation and use of different software programs or live video streaming programs may produce different outcomes concerning quality, usability for the speech pathologists and families, and cost-effectiveness for both parties. This may also be a consideration in translation studies, as many health services do not permit the use of Skype.

Replication

The outcomes of this study are considerable. They are, however, limited to the ability of a single speech pathologist working within a research framework, supported by the initial Lidcombe Program designers. The single speech pathologist was completing this project as part of a doctoral study and so the potential of researcher bias must be considered. To provide further evidence of the efficacy and efficiency of this model, independent replications of this trial are recommended. This would test the robustness of the program application and outcomes when independent researchers or speech pathologists carry out the same trial with a different cohort, independent of the original program designers.

Rural, Interstate and International Populations

The efficacy and efficiency outcomes support community translation with various populations; however, the parent reports and clinical behaviours are limited to a metropolitan sample. Further research into the application of this model with a rural and remote population would provide insight into potential differences in attitudes, attendance, convenience and treatment readiness for this group. These aspects might also differ with investigation of interstate and international cohorts. The quality of the internet connection and the technology may also differ between metropolitan, rural, interstate and international locations.

Hybrid Models

The results of this project are constrained to each service delivery group being independent of the other. Yet, as discussed in Chapter 9, a hybrid model might be of great benefit and more practical use in the clinical domain. Currently, the efficiency and efficacy of such a model could only be assumed, given the noninferiority outcomes of this trial. A trial investigating the use of a hybrid model would provide evidence as to its efficacy and efficiency, as well as of trends and clinical behaviours.

Home-Based Intervention

During the course of this project it became clear that webcam was the delivery medium, but the location of the client's treatment was the home. It would be plausible, therefore, and more thorough to investigate treatment outcomes, compliance with treatment, attendance behaviours and attendance patterns if the speech pathologist was physically based in the home with the family. This would test the use of webcam and a remote speech pathologist delivering the Lidcombe Program in a purer form, as the single difference would be the medium, not the medium and location of service.

Stuttering Treatment

The findings of this study are limited to the Lidcombe Program, but future studies could compare the use of alternative stuttering treatments. These may include the Westmead Program and Parent Child Interaction Therapy.

Preschool Treatment

The success of engaging preschool children and their families using webcam, noninferiority outcomes for stuttering treatment may form a basis for further research with preschool interventions. Many treatment programs and methods for preschool speech and language intervention rely on a similar model to that of the Lidcombe Program: parent education, parent training, direct treatment of the child and observation of the parent applying treatment to the children. As treatment components have proven successful when delivered via webcam, future research into speech and language intervention programs might provide similar results. Even if the efficacy was similar, preschool speech and language intervention would increase access to services for families who cannot access it. Early intervention for speech and language disorders is also vital for preschool children, as poor oral language is a predictor for literacy difficulties, educational attainment and vocational success in later life (Speech Pathology Australia, 2009).

Clinical Application

As discussed in the clinical applications chapter, the future direction of Lidcombe Program via webcam is significant. With the number of Lidcombe Program trained speech pathologists increasing, webcam treatment can be offered to families who are unable to access services, or who would like to receive home-based treatment for a variety of reasons. The results supported use of the standard manual. It is therefore a recommendation of this study that this method could be translated into community populations, with Lidcombe Program trained speech pathologists using webcam.

Such translation could be significant in increasing access to the Lidcombe Program for families who are currently unable to do so. The potential impact is great, as the consequence could be fewer adolescents and adults who stutter and experience the chronic difficulties associated with stuttering, as detailed in Chapter 1.

Summary

Outcomes from this RCT supported the noninferiority hypothesis. A methodological review of the RCT design satisfied 12 of Ost's (2008) recommendations. Although outcomes from this Phase III RCT are significant, future research is required. This could include further investigation of different software options, hybrid models, and further trials using rural, remote, interstate and international populations. The outcomes of this trial also support investigation of a hybrid model of Lidcombe Program service delivery, and a home-based delivery given the additional clinical trends that emerged from webcam families. Finally, the evidence supporting engaging directly with a preschool child, using webcam, warrants further research into this method of service delivery with this population for other speech pathology interventions.

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APPENDICES

Appendix A.

Pretreatment Participant Details

Participant	Group	Gender	Age	Stratification (above or below 5%SS)	Time since onset	Family History	Previous Treatment	Parent experienced with Lidcombe Program
1	webcam	boy	4 years	below	12months	positive	no	no
2	webcam	boy	5 years 5months	above	4months	positive	yes	yes
3	webcam	girl	5 years 1month	below	9months	positive	assessment only	no
4	clinic	boy	3years 4months	above	12months	negative	no	no
5	clinic	boy	3years 4months	above	1months	positive	assessment only	no
6	clinic	girl	3years 9months	below	15months	positive	no	no
7	clinic	boy	3years 5months	below	9months	negative	no	no
8	clinic	boy	4years 3months	below	9months	positive	no	yes
9	webcam	boy	4years 3months	below	24months	positive	no	no
10	clinic	boy	4years	below	12months	positive	assessment	no

			1month				only	
11	webcam	girl	4years 3months	above	12months	positive	no	no
12	webcam	boy	5years 3months	below	21months	positive	assessment only	no
13	clinic	boy	4years 2months	below	12months	unsure	assessment only	no
14	webcam	girl	5years 2months	below	14months	positive	no	no
15	webcam	boy	4years 11months	below	18months	negative	yes	yes
16	clinic	boy	4years 5months	below	19months	positive	no	no
17	clinic	boy	5years 11months	below	24months	positive	no	no
18	clinic	boy	4years 5months	below	12months	negative	assessment only	no
19	clinic	boy	4years 1month	below	24months	unsure	no	no
20	webcam	boy	5years 1month	below	30months	positive	no	no
21	webcam	girl	3years	below	6months	negative	no	no
22	webcam	boy	4years 11months	below	6months	negative	no	no

23	webcam	boy	5years 4months	below	14months	positive	no	no
24	webcam	girl	3years 11months	below	21months	positive	no	yes
25	clinic	boy	4years 2months	below	18months	negative	no	no
26	clinic	boy	5years 1month	below	13months	positive	no	no
27	clinic	boy	3years 10months	below	22months	positive	no	no
28	clinic	girl	3years 7months	above	15months	negative	assessment only	no
29	webcam	boy	4years 5months	above	24months	negative	no	no
30	webcam	boy	3years 11months	above	14months	positive	yes	yes
31	webcam	boy	5years 2months	below	17months	positive	no	no
32	clinic	boy	4years 4months	below	24months	negative	no	no
33	webcam	boy	4years 4months	below	10months	negative	no	no
34	webcam	boy	4years 3months	below	13months	negative	no	no
35	clinic	boy	3years	below	15months	positive	no	yes

			9months					
36	clinic	boy	3years 1months	below	8months	positive	no	no
37	webcam	boy	5years 2months	below	24months	positive	no	yes
38	webcam	boy	3years 4months	below	10months	positive	no	no
39	clinic	boy	3years 8months	below	8months	positive	no	no
40	clinic	boy	5years 6months	below	30months	positive	no	no
41	clinic	girl	3years 3months	below	9months	negative	assessment only	no
42	webcam	boy	3years 3months	below	6months	positive	no	no
43	webcam	girl	4years 1month	below	6months	negative	no	no
44	clinic	girl	5years 11months	below	23months	positive	no	yes
45	webcam	girl	3year 9months	below	12months	unsure	no	no
46	clinic	girl	4years 11months	below	13months	negative	no	no
47	webcam	boy	3years 1month	below	6months	negative	no	no

48	experimental	boy	4years 2months	below	6months	negative	no	no
49	control	boy	3years 11months	below	17months	positive	no	no

Appendix B.

Outcome Questionnaire

The Australian Stuttering Research Centre, The University of Sydney

Outcome Questionnaire

(Parent to complete)

Client ID number:

Date Questionnaire Completed: / /

1. During the last week, what would have been your child’s *typical* stuttering severity on any day?

1	2	3	4	5	6	7	8	9	10
No									extremely
stuttering									severe stuttering

2. During the last week, what would have been your child’s *most severe* stuttering severity on any day?

1	2	3	4	5	6	7	8	9	10
No									extremely
stuttering									severe stuttering

3. How satisfied are you with your child’s present level of fluency?

1	2	3	4	5	6	7	8	9	10
extremely									totally
satisfied									dissatisfied

Appendix C.

Standard Lidcombe Program Parent Questionnaire

The Australian Stuttering Research Centre, The University of Sydney

Clinic Lidcombe Program Parent Questionnaire

Client ID number: Date Questionnaire Completed: / /

Dear ,

Thank you for taking the time to complete this questionnaire. The information you provide will help us to evaluate the current ways we are conducting treatment with preschool children who stutter. Your answers will be used to adjust our treatment and the Lidcombe Program for the future.

For the following questions, please *circle the word(s)* that best represents how you feel about the statement.

1. The clinician established and maintained a strong relationship *with me* during treatment.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

2. The clinician established and maintained a strong relationship *with my child* during treatment.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

3. Learning how to use the severity rating scale was easy.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

4. Learning how to implement treatment with my child was easy.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

5. I felt confident in being able to modify my child’s treatment according to their need.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

6. Travelling to weekly sessions at the clinic was easy for our family.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

7. On average how many minutes did it take you to travel to the clinic from your home and back again each week?

____

8. What is the maximum time you would consider travelling to attend treatment sessions?

____

9. I would consider receiving treatment using the internet and a web camera.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

10. Please tell us anything else you would like us to know about your experiences during your child’s treatment.

Thank you for taking the time to complete this questionnaire.

Adapted from Lewis, C., Packman, A., Onslow, J., Simpson, J., & Jones, M. (submitted). A Phase II trial of telehealth delivery of the Lidcombe Program of Early Stuttering Intervention.

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Appendix D.

Webcam Lidcombe Program Parent Questionnaire

The Australian Stuttering Research Centre, The University of Sydney

Webcam Lidcombe Program

Client ID number: Date Questionnaire Completed: / /

Dear ,

Thank you for taking the time to complete this questionnaire. The information you provide will help us to evaluate the use of the Skype technology to treat preschool children who stutter. Your answers will be used to help determine whether we continue to use Skype in the future.

For the following questions, please *circle the word(s)* that best represents how you feel about the statement.

1. The clinician established and maintained a strong relationship *with me* during treatment.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

2. The clinician established and maintained a strong relationship *with my child* during treatment.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

3. Learning how to use the severity rating scale was easy.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

4. Learning how to implement treatment with my child was easy.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

5. I felt confident being able to modify my child’s treatment according to need.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

6. Before starting this program I had previous experience with Skype.

☐ Yes ☐ No

7. Using the internet and webcam in our own home made receiving treatment easier.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

8. I felt that using the webcam in my own home was invasive.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

9. If my child required further treatment I would prefer ;

☐ Standard in clinic visits ☐ Skype sessions ☐ No preference

10. Please list the Internet provider you used throughout treatment;

____

11. The quality of the Skype connection allowed me to see and hear the clinician well at all times.

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

12. Finding my own toys and activities to use during the Skype treatment sessions was easy

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

13. Having the opportunity to speak with the clinician without my child in the room was beneficial

Totally agree	Agree	Neither agree nor disagree	Disagree	Totally disagree

14. What were the main advantages of using a webcam and the internet?

15. What were the main disadvantages of using a webcam and the internet?

Thank you for taking the time to complete this questionnaire.

Adapted from Lewis, C., Packman, A., Onslow, J., Simpson, J., & Jones, M. (submitted). A Phase II trial of telehealth delivery of the Lidcombe Program of Early Stuttering Intervention.
Lees, S., Onslow, M., Packman, A., O'Brian, S. & Block, S. (2010). Group Delivery of the Lidcombe Program: Is it more efficient?

Appendix E.

Skype Quality Test

Participant Name: **ID Number:**

Skype Contact Name:

Testing Clinician: **Date:**

Equipment

Confirm Broadband connection:	Yes	No		
Type of Computer:	Laptop	Desktop	PC	Mac
Type of Webcam:	External	Built in		
Have you used Skype before? :	Yes	No		
If yes, do you have any difficulties	Yes	No	Explain:	

Test 1: Participant- Ask the parent to talk for a few minutes about their child. Explain the purpose of this is to test the audio and video of connected speech.

Audio Quality:	*Good*	*Some delay*	*Significant interruptions*

*Comment/Problem solving*____

Visual Quality:	*Good*	*Some delay*	*Significant interruptions*

*Comment/Problem solving*____

Camera setting: (test all)	Close (head/shoulders)	Distant (Room/ floor)

*Comment/Problem solving*____

Test 2: Clinician- Tell the parent about the study. Explain the purpose of this is to test the audio and video of connected speech. Ask the parent the following questions based on what they can see/hear

Audio Quality:	*Good*	*Some delay*	*Significant interruptions*

*Comment/Problem solving*____

Visual Quality:	*Good*	*Some delay*	*Significant interruptions*

*Comment/Problem solving*____

Thank parent for their time, confirm pass OR ask them to try and correct identified difficulties and organise a second testing time.

Outcome: **PASS** **FAIL – reason:** **2nd Test – reason:**

Note any discussion / questions asked overleaf.

Appendix F.

Individual Participant Outcomes

Participant	Group	No. Consultations Stage 2	Speech Pathologist Hours to Stage 2	Mean Consultation Duration	Weeks to Stage 2	Pre-treatment Severity Rating	Entry to Stage 2 Severity Rating	9-month Severity Rating
1	webcam	27.0	15.5	34.4	32.0	4	2	1
2	webcam	withdrawal				9		2
3	webcam	20.0	13.9	41.6	21.0	6	2	2
4	clinic	36.0	23.4	38.9	44.0	1	2	2
5	clinic	did not achieve Stage 2 entry				5		2
6	clinic	16.0	12.6	47.1	18.0	5	1	2
7	clinic	non-starter				2		
8	clinic	10.0	7.7	45.9	12.0	4	1	2
9	webcam	23.0	10.1	26.3	26.0	6	1	1
10	clinic	32.0	19.2	35.9	35.0	3	1	1
11	webcam	13.0	8.7	40.2	19.0	5	2	missing
12	webcam	17.0	11.5	40.5	19.0	5	1	1
13	clinic	11.0	6.7	36.5	14.0	3	1	2

14	webcam	18.0	10.5	34.9	18.0	5	1	1
15	webcam	12.0	6.2	30.8	13.0	2	1	1
16	clinic	17.0	10.2	36.1	18.0	4	1	1
17	clinic	19.0	13.8	43.5	24.0	5	2	1
18	clinic	12.0	8.1	40.4	15.0	4	1	2
19	clinic	8.0	6.3	47.4	9.0	missing	1	1
20	webcam	22.0	10.0	27.3	27.0	4	1	1
21	webcam	withdrawal				3		2
22	webcam	12.0	6.2	30.8	13.0	4	1	2
23	webcam	withdrawal				6		
24	webcam	17.0	8.8	31.2	18.0	2	1	1
25	clinic	21.0	15.8	45.2	24.0	7	1	1
26	clinic	30.0	20.4	40.7	46.0	5	1	2
27	clinic	57.0	43.6	45.9	62.0	4	2	2
28	clinic	29.0	20.1	41.5	35.0	6	1	2
29	webcam	in Stage 1				5		4
30	webcam	in Stage 1				8		5
31	webcam	46.0	29.8	38.8	50.0	5	2	2

32	clinic	31.0	22.0	42.6	46.0	2	2	2
33	webcam	33.0	17.9	32.5	41.0	7	2	2
34	webcam	39.0			56.0	5		2
35	clinic	withdrawal				3		2
36	clinic	19.0	12.0	37.9	26.0	2	3	3
37	webcam	9.0	4.2	27.7	15.5	missing	1	2
38	webcam	13.0	7.6	35.2	15.0	6	3	2
39	clinic	26.0	14.6	33.7	41.0	5	2.0	2
40	clinic	16.0	10.5	39.3	21.0	4	1	
41	clinic	23.0	15.9	41.3	29.0	3	1	3
42	webcam	9.0	4.1	27.2	12.0	3	4	1
43	webcam	withdrawal				2		
44	clinic	7.0	4.5	38.6	9.0	2	1	1
45	webcam	17.0	8.5	30.1	26.0	5	1	2
46	clinic	withdrawal				6		
47	webcam	withdrawal				3		
48	experimental	in Stage 1				4		2
49	control	in Stage 1				4		3

Appendix G

Standard Lidcombe Program Parent Questionnaire: Question 10 Transcript

The Australian Stuttering Research Centre, The University of Sydney

Standard Lidcombe Program Parent Questionnaire

10. Please tell us anything else you would like us to know about your experiences during your child's treatment.

I very much appreciate Kate's hard work & positive attitude. Particularly when I became a bit frustrated or despondent with progress. Its a fantastic program .

I feel the success of the therapeutic relationships between the child & clinician depends on the child's personality. We have a child who is very shy and doesn't warm to those outside Mum & Dad easily. I feel this would have made sessions for the therapist quite tricky.

One thing I found really great was having resources at home and knowing which resources we could use for therapy. It was quite helpful that the clinician provided me with games & materials to take home. It was wonderful that the clinician put so much effort into the games & toys each week & remembering what they'd already played with so she didn't get bored. She enjoyed the craft activities too.

This child responded a lot faster that the previous child who had therapy, possible due to less severe stutter. Also family had conducted LP before so that got into program more quickly. No learning process.

Has been fantastic. Noticed an enormous change. Clinician is wonderful. Treatment worked very quickly. Severity dropped from 4-5 to 3 dramatically and consistently. Friends and family comment/feedback- changes to stutter. Increased awareness in awareness of severity in Thomas during treatment very quickly and then modified himself.

James has been very engaged & looks forward to the session.

Taught me to be better Mo in listening & focusing on playing and talking. Made Mo more ware of focusing 100% on child. Good instruction to learn to praise child for good speech.

Rewarding experience-program works. RA& clinician magnificent. Personality clash early on with SP: rectified. Mentally hard thing for Mo - physically being alert, travelling. Hard to hear child speak each day as at work. Emotionally difficult for Mo. Hard to be on task all time. Fell like miss out on natural conversation where listening to bumps; speech not content. Good to send report to all clients participating.

All very positive, Kate's been great. It's been less difficult than what I thought it would be & child has started to show improvement quite quickly.

Rapport great with Kate and Eli. Speech therapy was outing for family. 3 kids in the room - Kate worked with all really well - juggling kids. Happy with quickness of progress. Feel confident in own ability. Nothing negative to say.

Positive experience. Good for bonding with child as well - husband commented about this too. Positive experience. Kate very obliging - get stuck don't hesitate to call/email. Did not feel abandoned over Xmas period.

There has been progress.

Kate was sensational & very accommodating with siblings; very approachable; parent support would be good - eg for parents to discuss program/strategies & to know there are others out there & to offer support to each other.

Nothing as far as the treatment is concerned, but it is very time consuming. I sometimes feel guilty if he has a bad week because that may be my fault because treatment is so heavily reliant on the parent & sometimes doing enough treatment is difficult. I've felt a bit uneasy about possible changes in expectations etc due to change in clinicians.

Wanting more clinician time rather than having mum do the treatment at home. It didn't make her feel comfortable he was receiving the best treatment possible.

Two children in treatment, Skype would be so much better.

Nothing to add, the service has been fantastic.

When discussing with clinician regarding Hugo's progress Hugo appeared self conscious - felt scrutinised? Clammed up and didn't talk much at all during treatment sessions. This had been discussed with clinician. Representative of stutter overall but not Hugo's overall speech. Mo thought that Hugo couldn't get rhythm going to potentially stutter more in sessions. "Really good program". Hugo's speech variable before treatment commenced but stuttering severity decreased and didn't increase in severity again over course of treatment. Mo & child pleased with results as child will be going to school next year.

Couldn’t be happier with level of professionalism & support. Wonderful experience for child and mother
very effective, gave me the tools to help out at home.

Appendix H.

Webcam Lidcombe Program Parent Questionnaire: Question 10 Transcript

The Australian Stuttering Research Centre, The University of Sydney

Webcam Lidcombe Program Parent Questionnaire

14. What were the main advantages of using a webcam and the internet?

Convenient

Not having to go to the clinic with 8mth child. Could have baby occupied or sleeping during sessions. Like d the organised time each week. When experienced problems with connecting or sound - could send messages over Skype or call each other.

Not having to take 2 children to Bundoora each week.

At home, not having to go in clinic. Learnt how to use Skype.

Could do treatment at home whilst younger child sleeps, no need to disturb.

Saving time - travel, comfortable & convenient.

Convenient - time effective, personal.

No travel, no time taken out of day.

Time shorter, no travel, in comfortable environment, cheaper - no travel.

not having to take young children to clinic.

Didn’t have to leave home; easier at home; child might have felt a more comfortable at home.

No travel; accommodating other children etc is easier; treatment place is home - so 'normal' as possible; more windows to change appointment times when a problem arose with schedule.

Convenient, comfortable for child - at home with own things; Kate is an excellent SP and I'm really happy with her.

Being in own environment, having his own belongings- helped him keep up a conversation because familiar items; didn't lose much time - able to work afterwards.

Don't have to rush elsewhere to take child to clinic - home becomes pseudo-clinic, minimal effect on work-life balance.

Travel, especially once started school - distance 45-1hr each way to La Trobe.

Appendix I.

Webcam Lidcombe Program Parent Questionnaire: Question 10 Transcript

The Australian Stuttering Research Centre, The University of Sydney

Webcam Lidcombe Program Parent Questionnaire

14. What were the main disadvantages of using a webcam and the internet?

Drop-out sometime; technical problems.

None really. Only difficulties were when there were glitches getting the connection or sound established but we easily overcame with Kate giving instructions.

Process may have been slower.

No disadvantages.

Not really any - having younger child around to interrupt.

Child could run around & out of the room due to personality.

Wonder if sometimes it was harder for Max to develop a relationship with Kate .

Sometimes service would drop out but it was good 98% of the time.

Occasionally bad line, keeping Jack on task.

Tech difficulties now & then.

Child might have concentrated better face to face with eye contact; child could be distracted by equipment.

Amount of space to work in (tied to computer, child had to keep still); requires good internet connection - some 'fiddling' required; child occasionally frustrated about not having whole visual of some activities; change 'change up' the environment when child was distracted or activity wasn't working.

Occasional connection problems.

Drop-outs / poor quality connection were a problem; Jack really wants to meet Kate; not able to play with new clinic toys.

You have to keep hold of the child to keep in front of the camera - might be easier in clinic as child can get up & move around without disrupting session. 2-3 times Skype connection really disappointed us but child could have a bad day in clinic too.

Internet connection problems. Sometimes not as personal at face to face.

Appendix J.

Pre-treatment Variables and Stage 2 Entry Efficiency Data

Variable		Group	Weeks Stage 2	Consultations Stage 2	Speech Pathologist Hours St 2
			mean (Standard Deviation)		
Overall Outcomes		Clinic	27.3 (14.7)	21.6 (11.9)	14.6 (8.9)
		Webcam	23.4 (5.6)	19.8 (10.1)	11.4 (6.4)
		P Value	p= .38	p= .63	p= .23
Gender	Boys	Clinic (n=15)	28.5 (15.6)	22.4 (12.6)	15 (9.6)
		Webcam (n=11)	24.8 (13.1)	21.1 (12.0)	12 (7.9)
		P Value	p= .51	p= .79	p= .39
	Girls	Clinic (n=4)	22.8 (11.6)	18.8 (9.5)	14.2 (6.6)
		Webcam (n=5)	20.4 (3.7)	17 (2.5)	10.1 (2.6)
		P Value	p= .72	p= .74	p= .41
Time since onset	6-12months	Clinic (n=8)	25.9 (12.1)	20 (8.9)	12.3 (5.6)
		Webcam (n=11)	22.4 (10.1)	18 (8.3)	10.3 (4.9)
		P Value	p= .53	p= .65	p= .46
	13-24months	Clinic (n=10)	29.1 (17.6)	23.5 (14.5)	16.9 (11.1)

		Webcam (n=7)	24.1 (13.5)	21.6 (13)	12.9 (8.9)
		P Value	p= .52	p= .78	p= .42
	over 24months	Clinic (n=1)	27.0	22.0	10.0
		Webcam (n=1)	21.0	16.0	10.5
Family History	Positive	Clinic (n=11)	27.5 (15.9)	21.8 (13.4)	14.5 (10.5)
		Webcam (n=12)	23.5 (10.8)	20.3 (10.6)	12 (7.1)
		P Value	p= .04	p= .76	p= .51
	Negative	Combined (n=9)	28.9, 29	23.2, 23	15.1. 15.9
		Clinic (n=6)	32.2, 32	25.3, 26	17.5, 18
		Webcam (n=3)	22.3, 13	19, 12	10.1, 6.2
	Unknown	Clinic (n=2)	11.5 (3.5)	9.5 (2.1)	6.5 (0.3)
		Webcam (n=1)	26	17	8.5
Previous Treatment	No	Clinic (n=14)	28.6 (16.4)	22.4 (13.2)	15.5 (9.9)
		Webcam (n=14)	23.9 (11.7)	20 (10.9)	11.2 (7.1)
		P Value	p= .39	p= .61	p= .2
	Assessment Only	Clinic (n=5)	23.8 (9.1)	19.6 (7.8)	12.2 (5.6)
		Webcam (n=2)	20 (1.5)	20 (2.1)	12.7 (1.7)
		P Value	p= .42	p= .78	p= .86
	Yes	Clinic (n=0)			

		Webcam (n=1)	13	12	6.2
Parent Previously exposed to LP	No	Clinic (n=17)	29.3 (14.2)	23.2 (11.6)	15.6 (8.9)
		Webcam (n=13)	25.2 (11.5)	21.5 (10.5)	12.6 (6.8)
		P Value	p= .4	p= .67	p= .29
	Yes	Clinic (n=2)	10.5 (2.1)	8.5 (2.1)	6.1 (2.2)
		Webcam (n=3)	15.5 (2.5)	12.7 (4)	6.4 (2.3)
		P Value	p= .1	p= .2	p=. 89
Household income (n=32)	30000-39999	Clinic (n=0)			
		Webcam (n=1)	18.0	16.0	12.6
	40000-59999	Clinic (n=5)	38.6 (19.3)	30 (17.5)	21 (13.7)
		Webcam (n=3)	15.5 (3.5)	10.3 (2.3)	5.7 (2.7)
		P Value	p= .06	p= .07	p=.07
	60000-79999	Clinic (n=2)	23.5 (3.5)	19.5 (4.9)	10.3 (0.3)
		Webcam (n=3)	22 (8.9)	19 (7.2)	11.5 (3.9)
		P Value	p= .81	p= .93	p= .64
	80000+	Clinic (n=10)	22.7 (12.9)	18.7 (9.4)	12.9 (8.1)
		Webcam (n=8)	24.3 (12.7)	21.8 (11.6)	12.9 (8.1)
		P Value	p= .93	p=. 55	p= .94

Appendix K.

RCT Recommendations taken directly from Öst, L.-G. (2008).

(1) Do not use WLC as the control condition, since criterion I requires a placebo or another treatment.

(2) Do not use TAU as the control condition, since the methodological problems described above are so extensive.

(3) Use an active treatment as comparison, preferably one that has been established as effective for the disorder in question.

(4) Do a proper power analysis before the start of the study and adjust the cell size for the attrition that may occur.

(5) Use a representative sample of patients, diagnose them using suitable instruments in the hands of trained interviewers, and test the diagnostic reliability.

(6) Let an independent researcher or agency use an unobjectionable randomisation procedure, and conceal the outcome of it from all persons involved in the study.

(7) Use reliable and valid outcome measures; both the ones that are specific to the disorder and general ones.

(8) Use blind assessors and evaluate their blindness regarding treatment condition of the patients they assess.

(9) Train the assessors properly and measure inter-rater reliability on the data collected throughout the study (not just during training).

(10) Use three or more properly trained therapists and randomize patients to therapist to enable an analysis of possible therapist effect on the outcome.

(11) Include at least a 1-year follow-up in the study and assess any nonprotocol treatments that the patients may have obtained during the follow-up period.

(12) Audio- or videotape all therapy sessions. Randomly select 20% of these and let independent experts rate adherence to treatment manual and therapist competence.

(13) Insert procedures to control for concomitant treatments that patients in the study may obtain simultaneously as the protocol treatment.

(14) Describe the attrition, do a drop-out analysis and include all randomized subjects in an intent-to-treat analysis.

(15) Assess clinical significance of the improvement of the primary measures.