Contents lists available at ScienceDirect





# Journal of Fluency Disorders

journal homepage: www.elsevier.com/locate/jfludis

# Lidcombe Program telehealth treatment for children 6–12 years of age: A Phase II trial



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ARTICLE INFO

Keywords: School-age Stuttering Treatment

#### ABSTRACT

*Background:* For children older than 6 years who stutter, there is a gap in clinical research. This is an issue for speech-language pathologists because the tractability of stuttering decreases and the risk of long-term psychological consequences increase with age.

*Purpose*: To report a Phase II trial of a telehealth version of the Lidcombe Program with school-age children.

*Methods*: Participants were 37 children who stuttered, 6–12 years of age, from Australia, New Zealand, Hong Kong, and Singapore. Parents were trained by video telehealth how to deliver the Lidcombe Program to their child. Primary and secondary outcomes were stuttering severity and psychosocial functioning measured pre-treatment and at 6 months and 12 months after starting treatment. Parents submitted two 10-minute recordings of their child speaking in conversation, and three measures of anxiety, impact of stuttering, and communication attitude.

*Results*: Six months after starting treatment, seven children (18.9%) attained Lidcombe Program Stage 2 criteria, 25 children (67.6%) showed a partial response to treatment, and five children (13.5%) showed no response. By 12 months, 12 children (32.4%) had reached Stage 2 criteria. Psychosocial improvements were observed 6 and 12 months after starting treatment.

*Conclusions*: The Lidcombe Program may eliminate or nearly eliminate stuttering for about one third of children 6–12 years of age. Randomized controlled trials with this age group involving the Lidcombe Program are warranted. In the interim, the Lidcombe Program is a clinical option clinicians can implement with this age group to reduce stuttering and its psychosocial impacts.

## 1. Introduction

#### 1.1. The importance of early childhood stuttering intervention

Stuttering is a genetically influenced speech disorder associated with anomalies of brain structure and function in areas subserving spoken language (Chang & Guenther, 2020; Chow et al., 2020; Garnett et al., 2019; Koenraads et al., 2020; Packman et al., 2022;

https://doi.org/10.1016/j.jfludis.2024.106057

Received 15 June 2023; Received in revised form 5 March 2024; Accepted 1 April 2024

Available online 7 April 2024

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Thompson-Lake et al., 2022). A community cohort study reported a median onset age of 31 months and a 4-year cumulative incidence of 11.2% (Reilly et al., 2013). Natural recovery occurs for approximately two thirds of cases by adulthood but is less than 10% between 12 to 18 months post onset (Carey et al., 2021; Jones et al., 2008; Reilly et al., 2013; Yairi & Ambrose, 2004). Aside from observable speech disruptions, people who stutter are at much higher risk of developing a mental health disorder than the general population, particularly social anxiety disorder (Briley et al., 2021; Iverach et al., 2009). Those mental health disorders originate with noxious social experiences from peers that can occur shortly after onset and continue into the school years and beyond (Blood & Blood, 2016; Davis et al., 2002; Langevin, 2015; Langevin et al., 2009; Vanryckeghem et al., 2005). The social anxiety of those who stutter is connected to challenges reaching full educational and occupational potential (Berchiatti et al., 2020; Gerlach et al., 2018; Klein & Hood, 2004; McAllister et al., 2012; O'Brian et al., 2011). School-age children are required to participate in class discussions and group projects, as well as perform oral presentations. For children who stutter, speech-related anxiety associated with these classroom activities can lead to poorer academic performance (Berchiatti et al., 2020; O'Brian et al., 2011) and poorer school attendance (Boyle, Decouffe, & Yeargin-Allsopp, 1994). Chronic stuttering is associated with quality of life scores equivalent to those of conditions such as cardiovascular disease, HIV, and cancer (McAllister et al., 2017; Omori et al., 2021; Zhou et al., 2021).

#### 1.2. The Lidcombe Program

Because of its potential lifetime impact, there is consensus that early intervention during the pre-school years is best practice for managing stuttering (Brignell et al., 2021). There are three childhood stuttering interventions with randomized controlled trial evidence—the Lidcombe Program (Brignell et al., 2021), RESTART-DCM (de Sonneville-Koedoot et al., 2015), and syllable-timed speech (Trajkovski et al., 2019). A Cochrane Review indicated that the best available evidence is from four randomized trials of the Lidcombe Program compared with a waitlist control group (Sjøstrand et al., 2019). The Lidcombe Program is an operant treatment that does not require children to change their natural speaking patterns to achieve fluency. Instead, parents provide verbal contingencies to their child for stuttered or stutter-free speech in daily practice sessions and during everyday conversations. For children younger than 6 years of age, the odds ratio for recovery from stuttering, 6 months after the start of treatment, is 7.5 compared with no treatment (Onslow et al., 2012). This program also shows psychological benefits for young children (Woods et al., 2002) and no negative effects on parent–child communication (Bonelli et al., 2000). In fact, Woods et al. (2002) reported improvements in Child Behavior Checklist scores (Achenbach, 1991) relating to anxiety, aggression, withdrawal, and depression post treatment.

If the Lidcombe Program is the best available evidence for treating young children who stutter, then it seems intuitive that researchers and clinicians should know if it can be efficacious for older children as well. The only prospective group study of this treatment with children older than 6 years of age occurred two decades ago (Lincoln et al., 1996). That study used a preliminary version of the current *Lidcombe Program Treatment Guide* (Onslow et al., 2023) with 11 school-age children. The authors reported a mean stuttering reduction of 65% from pre-treatment to 12 months follow-up. Apart from the encouraging outcomes reported in Lincoln et al. (1996), the efficacy of the Lidcombe Program has been documented in four other case reports of children 6–12 years of age (Bakhtiar & Packman, 2009; Hewat et al., 2020; Koushik et al., 2009; Yandeau et al., 2021). The 15 children included in these case reports showed clinically meaningful stuttering reductions. Psychosocial outcomes after treatment were not reported in any of these preliminary studies, so it is not known whether any such benefits are associated with the program for older children.

It is apparent that clinicians have a wealth of evidence available to them to effectively treat stuttering during the pre-school years. Beyond this age group though, there is a gap in clinical research (Brignell et al., 2021; Nippold, 2011). There are several reasons why filling this knowledge gap is critical for the field of speech-language pathology. First, the clinical tractability of stuttering decreases during the school years (Bothe et al., 2006; Ingham & Cordes, 1999; Koushik et al., 2009; Onslow & Packman, 1997). Presumably, this is due to decreasing cortical plasticity of speech related areas during the period between 6 and 12 years of age, making these neural patterns more complex to shift. By adolescence, stuttering is clinically intractable. Second, there is no safe period of exposure to negative social experiences. Children as young as 3 years of age experience negative social consequences of stuttering (Vanryckeghem et al., 2005), and this escalates once a child reaches school age (Blood & Blood, 2007; Blood et al., 2011; Blood et al., 2010; Guttormsen et al., 2015). Hence, the school years could be the last opportunity for effective clinical control of stuttering to offset the possible psychological effects of a lifetime of communication difficulty.

If the Lidcombe Program can effectively reduce stuttering as well as the negative psychosocial impacts of stuttering, for preschoolers as well as school-age children, this will be useful knowledge for speech-language pathologists. This is because, at present, there is no gold standard practice for treating children older than 6 years of age, leaving clinicians challenged as to how to help them (Nippold, 2011). The Lidcombe Program does not require the child or the parent to change the way they speak to achieve fluency. Hence, it can be seamlessly integrated into the family lifestyle. This also means that fluency achieved in this program is unlikely to attract unwanted peer attention. As it stands, the Lincoln et al. (1996) trial is the only prospective group study of more than 10 participants supporting Lidcombe Program use with school-age children. Clinical trial research of the Lidcombe Program with school-age children is needed to advance treatment efficacy knowledge for this under-researched age group.

#### 1.3. The present trial

In summary, there is a concerning clinical research gap when it comes to managing stuttering when children are between 6 and 12 years of age. To date, the Lidcombe Program is regarded as a key method for treating stuttering close to onset (Sjøstrand et al., 2019) and could, therefore, be a viable option for older children. To investigate this, in this study we initiate a program of research about the efficacy of the Lidcombe Program with school-age children, using a non-randomized pre-to-post-treatment Phase II trial with video

telehealth. We assess short- and longer-term outcomes for improving psychosocial impacts as well as reducing stuttering.

#### 2. Methods

#### 2.1. Participants

The study was approved by the University of Melbourne Human Ethics Committee (ID: 2057373). Inclusion criteria were (a) diagnosis of developmental stuttering by a speech pathologist, (b) 6-12 years of age at recruitment, and (c) child and parent with proficient spoken English to participate in the treatment. Exclusion criteria were (a) language or intellectual disability and/or (b) stuttering treatment received within the previous 6 months.

Seventy-five families from Australia, New Zealand, Hong Kong, Singapore, and North America expressed interest in participating. Of those 75 families, 32 were not included in the study because of parents choosing not to commence the program (31.3%), failure to submit pre-treatment assessments (25.0%), unconfirmed stuttering diagnosis (15.6%), child not within the age range of 6–12 years (12.5%), or a preference for in-clinic service rather than the video telehealth treatment format offered in this study (6.3%). Of those 32 families, three families (9.4%) who were managing more than one sibling who stuttered were also excluded. The reason for those exclusions was the parent burden of managing multiple treatment programs. Of the 43 children included in the treatment group, six families decided to withdraw shortly after beginning the program. The reasons for withdrawing from the study included not committing to program requirements (50.0%), the parent deciding the child no longer required intervention (16.7%), or other developmental priorities (33.3%). The baseline information for the 37 children who completed at least 6 months of the program is presented in Table 1.

#### 2.2. Treatment

The first author is a speech pathologist with 6 years of clinical experience, who received Lidcombe Program Consortium Training in 2021. The children were treated by the first author using the *Lidcombe Program Treatment Guide* (Ver. 1.3) (Onslow et al., 2021). This treatment is a parent-delivered program, where parents learn how to implement the therapy at home. This training occurs in weekly, 45–60-minute clinical appointments with the speech-language pathologist. In these appointments, parents are taught by the clinician to provide verbal contingencies in response to their child's stuttered or stutter-free talking. Parent verbal contingencies occur in 10–15-minute practice sessions as well as everyday conversations with the child to positively reinforce stutter-free speech or gently correct stutters; an example of the former is "Great smooth talking", and examples of the latter are "I heard a bumpy word" and "Can you say that again without the bumps?" The clinician also coached parents on how to manage their child's language output during daily 10–15-minute practice sessions to optimise stutter-free speech practice. The structure of these practice sessions was guided by the severity of the child's stuttering, so parents could alter the target utterance length and complexity as needed to facilitate stutter-free speech. For example, for a child with a high stuttering severity rating, an appropriate activity would be a single-word picture naming game. However, a child with a low stuttering severity rating might participate in an unstructured Lego-building activity. Managing the length and grammatical complexity of children's utterances during daily practice time increases the likelihood of children regularly attaining stutter-free speech during those sessions. Beyond these 10–15-minute daily practice sessions, children's language output was not managed in any way.

The aim of the Lidcombe Program for children to meet the following two criteria for three consecutive clinical appointments to progress from Stage 1 to Stage 2: (a) parent daily severity ratings are 0–1 during the preceding week with at least four of the daily ratings being 0, and (b) clinician severity ratings do not exceed 1 during clinical appointments. Stage 2 is a maintenance phase, where treatment is systematically withdrawn, contingent on sustained clinical gains achieved in Stage 1.

The treating clinician conducted clinical appointments with the children and parents by video telehealth. Screen sharing and online activities were utilized in appointments as part of the online format. Children and parents also utilized resources available at home or at school to show on screen to the treating clinician during appointments. With pre-schoolers, telehealth has been shown to be non-inferior to in-clinic appointments (Bridgman et al., 2016). Accordingly, clinical appointments could occur when the child was at home or at school, at the convenience of the child and parent. Telehealth also allowed for international recruitment of participants.

Treatment fidelity was assured with regular supervision of the treating clinician by a speech-language pathologist with 30 years of specialist experience treating stuttering and who is a member of the Lidcombe Program Trainers Consortium. The specialist speech-language pathologist observed 27 h of treatment session video recordings and met for discussion with the treating clinician fortnightly for the first 9 months of the treatment period. Fidelity of parent treatment delivery was monitored in weekly clinical appointments as part of standard Lidcombe Program procedure (Onslow et al., 2021).<sup>1</sup> During appointments, parents demonstrated 5–10 min of home practice to the clinician, and then the parent and clinician would discuss and implement necessary modifications to optimise treatment delivery. The benefit of telehealth also meant that the treating clinician could observe an accurate representation of the parent delivering treatment in their own environment with their own resources.

<sup>&</sup>lt;sup>1</sup> The present Phase II trial was conducted using the *Lidcombe Program Treatment Guide* (Ver.1.3) (Onslow et al., 2021) before the publication of the updated *Lidcombe Program Treatment Guide* (Ver.1.5) (Onslow et al., 2023) was published.

# Table 1

| Baseline characteristics of the participant sample. |   |
|---|---|
|   | _ |

| Referral source  |            |
|--|------------|
| School   | 15 (40.5%) |
| Speech pathologist   | 17 (46.0%) |
| Other childhood services   | 5 (13.5%)  |
| Male   | 35 (94.6%) |
| Age range at recruitment (years;months)                                | 6;0-11;9   |
| 6;0-8;11   | 26 (70.3%) |
| 9;0–11;9   | 11 (29.7%) |
| Mean reported age of stuttering onset (years;months) (SD) <sup>a</sup> | 4;7, (1.7) |
| Mean stuttering severity at onset (SD) <sup>b</sup>                    | 4.0, (2.1) |
| Children with comorbidities  |            |
| Speech sound disorder or delay   | 9 (24.3%)  |
| Language disorder or delay   | 4 (10.8%)  |
| Other neurodevelopmental diagnosis                                     | 1 (2.7%)   |
| Children who received previous stuttering treatment                    | 10 (27.0%) |
| Family cultural background   |            |
| Caucasian  | 20 (54.1%) |
| Asian  | 7 (18.9%)  |
| Eastern European   | 5 (13.5%)  |
| Arabic   | 3 (8.1%)   |
| Brazilian Portuguese   | 1 (2.7%)   |
| Indigenous Australian  | 1 (2.7%)   |
| Bilingual speaking   | 13 (35.1%) |
| Single parent  | 3 (8.1%)   |

<sup>a</sup> This is based on parent report from case history survey.

<sup>b</sup> Parents used a stuttering severity scale where 0 = no stuttering, 1 = extremely mild stuttering,

and 9 = extremely severe stuttering.

#### 2.3. Outcomes

Primary and secondary outcomes were measured pre-treatment, at 6 months, and 12 months after starting treatment.

#### 2.3.1. Clinician-reported severity ratings

The primary outcome was stuttering severity measured by an independent, blinded speech-language pathologist, using a scale where 0 = no stuttering, 1 = extremely mild stuttering, and 9 = extremely severe stuttering. This scale is described in detail in the *Lidcombe Program Treatment Guide* (Ver. 1.3) (Onslow et al., 2021).<sup>2</sup> There is empirical support for measurement of clinical change with stuttering severity ratings (Karimi et al., 2014; Onslow et al., 2018), and the measure has been reported as an outcome in several clinical trials (O'Brian et al., 2020). The independent, blinded clinician had over 20 years of experience with childhood stuttering. These measurements were made from two 10-minute parent-recorded speech samples of the child speaking at each of the three assessments: pre- treatment, 6 months after starting treatment, and 12 months after starting treatment. At each assessment, parents collected two 10-minute audio recordings of their child talking in conversation beyond the clinic environment with either a parent, a non-family member, or on the phone with a friend. The independent, blinded clinician then assigned a severity rating to each child speech sample, and the mean severity rating was calculated for each assessment.

To support parents to collect a representative sample of their child's speech, during the initial research consultation parents were instructed to encourage conversational speech from their child. This included strategies such as using open-ended questions and exploring topics of interest to the child.

#### 2.3.2. Psychosocial questionnaires

To determine whether the Lidcombe Program alters psychosocial impacts of stuttering, families were given instructions to access an online platform so that parents and children could submit the following questionnaires. These questionnaires were completed pretreatment, 6 months after starting treatment, and 12 months after starting treatment:

The Overall Assessment of the Speaker's Experience of Stuttering–School-age (OASES-S) is an instrument to evaluate the adverse impacts of stuttering (Yaruss & Quesal, 2016). The OASES-S is a 60-item questionnaire completed by the child. The child responds to questions on a Likert scale based on the following sections: (a) general perspectives and feelings about stuttering, (b) affective, cognitive, and behavioural reactions to stuttering, (c) functional communication difficulties in daily situations, and (d) impact of stuttering on quality of life in relation to communication at school, socially, and at home. The responses, therefore, provide data about whether a child is experiencing negative impacts associated with stuttering and also the degree to which stuttering is affecting quality

<sup>&</sup>lt;sup>2</sup> The present Phase II trial was conducted using the 9-point severity rating scale recommended in the *Lidcombe Program Treatment Guide* (Ver.1.3) (Onslow et al., 2021). The updated *Lidcombe Program Treatment Guide* (Ver.1.5) (Onslow et al., 2023) now recommends using a 10-point stuttering severity rating scale.

#### of life.

The Communication Attitudes Test (CAT) is a survey that is completed by the child (Brutten & Dunham, 1989). It includes 35 statements, which require children to respond with either "true" or "false" in relation to their own thoughts and feelings about their communication. The scores are totalled, with higher scores indicating negative communication attitudes and lower scores indicating positive communication attitudes.

*The Spence Children's Anxiety Scale (SCAS)* is a 44-item questionnaire completed by a parent, and another version is completed by the child (SCAS Child version: Overview, 2021). This tool measures six domains, broadly corresponding to the dimensions of anxiety disorders in the *Diagnostic and Statistical Manual of Mental Health Disorders – Fifth Edition* (DSM-5) (American Psychological Association, 2013). These domains include generalized anxiety, panic, social phobia, separation anxiety, obsessive compulsive disorder, and physical injury fears. Responders of the SCAS rated the degree to which they experience each questionnaire item using a 4-point Likert scale (e.g., 0 = never to 3 = always). The summation of each item calculated gives a total score where higher scores suggest increased frequency and severity of symptoms. A large body of evidence supports the psychometric properties of both the parent and child versions of the SCAS (Essau et al., 2011; Nauta et al., 2004; Spence, 1998; Whiteside & Brown, 2008). The simplicity of its application is also a benefit for its use with young people.

#### 3. Results

A summary of the means and standard deviations for clinician-reported severity ratings and the three questionnaires at each of the three assessments are presented in Table 2. For each participant, a mean clinician-reported stuttering severity rating was calculated from two 10-minute speech samples at each assessment. One-way repeated-measures analysis of variance (ANOVA) was used to assess changes over time in severity ratings, impact of stuttering, communication attitude, and anxiety symptoms in response to Lidcombe Program treatment.

#### 3.1. Clinician-reported severity ratings

#### 3.1.1. Inter-judge reliability

To assess inter-judge reliability, 16% of the 10-minute audio recordings, taken 6 months after starting treatment, were presented to another independent, blinded clinician with 6 years of clinical experience. The clinician used the same stuttering severity scale to rate the recordings. Absolute reliability was high, with 22/23 (95.7%) severity scale scores agreeing by +/- one scale value with the original clinician. Relative reliability was high, with a Pearson correlation between the two sets of scores at r = .92. Another sample (13%) was re-presented to the initial clinician to establish intra-judge reliability. Absolute reliability was high, with 24/28 severity scale scores agreeing by +/- one scale value with the original rating. Relative reliability was also high, with a Pearson correlation between the first and second ratings at r = .96.

#### 3.1.2. Six months after starting treatment

Seven children (18.9%) attained Stage 2 criteria 6 months after starting treatment. The mean pre-treatment stuttering severity rating for the group reduced from 3.6 to 1.1. This represents a mean stuttering severity rating reduction of 69%. The two participants with the largest changes in stuttering severity ratings from pre-treatment to 6 months after starting treatment had stuttering severity rating reductions of 6 and 7. Of the children who did not reach Stage 2 criteria, 25 children (68%) had a partial response to the program of greater than 1 severity rating decrease. Clinician-reported severity ratings for five of the children (13.5%) did not change after 6 months suggesting no treatment effect. No children showed an increase in stuttering severity over the first 6 months of treatment.

#### 3.1.3. Twelve months after starting treatment

Including the seven children who met Stage 2 criteria within 6 months, a total of 12 children (32.4%) from the sample reached Stage 2 criteria 12 months after starting treatment. The mean stuttering severity rating for the treated group had decreased from 3.6 to 1.2 which represents a mean stuttering severity rating reduction of 67%. Of the children who had not reached Stage 2, six children (16%) showed further reductions greater than one stuttering severity rating between 6 and 12 months of treatment. Seven children

#### Table 2

Summary of group mean and standard deviations for clinician-reported severity ratings and psychosocial questionnaires at pre-treatment, 6 months, and 12 months after starting treatment.

|                            | Group mean and standard deviation (parentheses) |                   |                      |  |  |  |  |  |  |
|----------------------------|---|-------------------|----------------------|--|--|--|--|--|--|
|                            | Pre-treatment $n = 37$                          | 6 months $n = 37$ | 12 months $n = 35^*$ |  |  |  |  |  |  |
| Stuttering severity rating | 3.57 (1.70)                                     | 1.15 (1.53)       | 1.17 (1.51)          |  |  |  |  |  |  |
| CAT                        | 13.73 (8.84)                                    | 9.73 (7.38)       | 7.60 (7.41)          |  |  |  |  |  |  |
| OASES-S                    | 2.43 (0.61)                                     | 2.04 (0.47)       | 1.97 (0.54)          |  |  |  |  |  |  |
| SCAS (child) Total Score   | 24.70 (11.42)                                   | 21.49 (11.92)     | 19.31 (10.42)        |  |  |  |  |  |  |
| SCAS (parent) Total Score  | 18.86 (10.58)                                   | 16.37 (8.46)      | 13.00 (5.66)         |  |  |  |  |  |  |

*Note:* \* Of the 37 participants who completed 6 months of the program, two participants dropped out of the program before data collection 12 months after starting treatment.

(19%) showed an increase in stuttering severity rating between 6 and 12 months of treatment with a mean stuttering severity rating increase during this time of 1.6; only one child (3%) had a stuttering severity rating increase beyond pre-treatment scores.

#### 3.1.4. Statistical results for clinician-reported severity ratings

A one-way repeated-measures ANOVA showed that the changes in clinician-reported stuttering severity ratings significantly decreased at the 0.5 level, F(2, 70) = 47.74, p < .001. The effect size measured by partial eta-squared was  $\eta_p^2 = .58$ , which is considered to be a large effect. A post-hoc Tukey pairwise comparison test showed a significant reduction in severity ratings between pre-treatment and 6 months ( $M_{difference} = -2.4$ , 95% CI [-3.3, -1.5], p < .001) and pre-treatment to 12 months ( $M_{difference} = -2.4$ , 95% CI [-3.3, -1.5], p < .001). There was no significant difference between severity ratings at 6 months and 12 months,  $M_{difference} = .02$ , 95% CI [-0.9, 0.9], p = .99.

#### 3.2. Statistical results for the psychosocial outcomes

#### 3.2.1. CAT

Fig. 1 reflects the changes in group mean total scores for the CAT at each outcome assessment. The highest possible negative communication attitude score was 35 if the child responded negatively to all 35 statements presented to them on the test. A one-way repeated-measures ANOVA suggested a significant reduction in total scores between assessments at the 0.5 level, F(2, 70) = 13.89, p < .001. The effect size was large,  $\eta_p^2 = .28$ . A post-hoc Tukey pairwise comparison test showed a significant reduction in total scores between pre-treatment and 6 months a ( $M_{difference} = -4.0$ , 95% CI [-6.51, -1.49], p = .001) and pre-treatment and 12 months ( $M_{difference} = -5.4$ , 95% CI [-7.95, -2.82], p < .001). Six months after starting treatment, the group mean CAT score decreased from 13.7 to 9.7 which is a score reduction of 29%. Twelve months after starting treatment the group mean score had further decreased to 8.3 which is a score reduction of 39%. There was no significant change in the group mean CAT total score between 6 months and 12 months,  $M_{difference} = -1.4$ , 95% CI [-3.95, 1.18], p = .402.

#### 3.2.2. OASES-S

Fig. 2 shows the changes in group mean total scores for the OASES-S at each outcome assessment. OASES-S scores above 3.7 reflect severe stuttering impact, and scores below 1.5 reflect mild stuttering impact. A one-way repeated-measures ANOVA indicated a significant reduction in the group's total impact score between assessments at the 05 level, F(2, 67) = 22.53, p < .001. The effect size was large,  $\eta_p^2 = .40$ . Six months after starting treatment, the group mean OASES-S total score had reduced from 2.4 (moderate impact) to 2.0 (mild-moderate impact). A post-hoc Tukey pairwise comparison test showed that this reduction was significant,  $M_{difference} = -0.4$ , 95% CI [-0.54, -0.21], p < .001. Twelve months after starting treatment, the OASES-S total score reduced further to 1.9 (mild-moderate impact) which was significant [Tukey comparison:  $M_{difference} = -0.43$ , 95% CI [-0.60, -0.23], p < .001. There was no significant change in the total impact between 6 months and 12 months,  $M_{difference} = -0.5$ , 95% CI [-0.22, 0.17], p = .745.

#### 3.2.3. SCAS (child and parent versions)

Fig. 3 and 4 reflect the changes in group mean total anxiety scores for the SCAS parent and child versions at each outcome assessment. One-way repeated-measures ANOVA showed significant total score improvements at the.05 level for the child (F(2, 70) = 5.29, p = .007) and parent versions (F(2, 70) = 10.17, p < .001). There was a medium effect size for changes in child anxiety scores ( $\eta_p^2 = .13$ ), and a large effect size for parent-reported scores ( $\eta_p^2 = .23$ ). Clinically meaningful improvements in child and parent total anxiety scores for the SCAS were observed after 6 months of treatment, with reductions of 3.2 (13%) and 2.6 (14%) points respectively. A similar pattern was observed after 12 months with reductions of 5.4 (22%) and 5.9 (31%) points respectively for child and parent total anxiety scores. Despite these improvements, a post-hoc Tukey pairwise comparison test showed that the child ( $M_{difference} = -3.22$ , 95% CI [-7.17, -0.74], p = .133) and parent ( $M_{difference} = -2.59$ , 95% CI [-5.59, 0.40], p = .102) total anxiety score improvements



Fig. 1. Communication Attitude Test total scores and standard error for the participant group measured at each outcome assessment.



Fig. 2. OASES-S Overall Impact Scores and standard error for the participant group measured at each outcome assessment.



Fig. 3. Spence Children's Anxiety Scale scores and standard error for the child-version measured at each outcome assessment.



Fig. 4. Spence Children's Anxiety Scale scores and standard error for the parent version measured at each outcome assessment.

were not statistically significant after 6 months. However, 12 months after starting treatment, the total anxiety score improvements had reached statistical significance for both child ( $M_{difference} = -5.44$ , 95% CI [-9.48, -1.40], p = .005) and parent responses ( $M_{difference} = -5.75$ , 95% CI [-8.82, -2.69], p < .001).

A subscale test of the Spence Children's Anxiety Scale, social phobia also showed statistically significant improvements 12 months after starting treatment, F(2, 70) = 8.91, p < .001. This subscale test is of particular importance as it is reflective of changes in social anxiety symptoms.

#### 4. Discussion

This non-randomized pre-to-post-treatment Phase II trial explored the efficacy of the Lidcombe Program—a key evidence-based stuttering treatment for pre-schoolers (Sjøstrand et al., 2019)—with school-age children. The findings are consistent with a preliminary report published over two decades prior to this publication (Lincoln et al., 1996). In the present trial, most stuttering children 6–12 years of age had a clinically significant response of greater than one scale value reduction on the severity rating scale 6 months after starting Lidcombe Program treatment. In addition, approximately one third of the sample eliminated or nearly eliminated their stuttering within 12 months. A stuttering severity score of 0–1 is considered equivalent to typical childhood speech (Lincoln et al., 1997). Over half the children in this cohort showed clinically significant treatment gains 12 months after starting treatment, with improvements of more than one stuttering severity score value achieved by 62% of the group.

Video telehealth delivery facilitates parent access to specialist speech-language pathology services because it does not require inperson clinic attendance. This is specifically appealing for families of busy school-age children who encounter barriers attending inclinic appointments around school hours. This trial suggest that video telehealth delivery of the Lidcombe Program may minimize those barriers to healthcare access for school-age children who stutter. The results are broadly consistent with a randomized controlled trial with pre-school children that showed video telehealth delivery of the Lidcombe Program can achieve the same treatment outcomes as in-clinic services (Bridgman et al., 2016).

School-age children are at heightened risks of harmful social experience because of their stuttering (Blood et al., 2011; Davis et al., 2002). An intervention that might impact the psychosocial impacts of stuttering is therefore a priority at this age, to mitigate the risks of longer-term psychological disorders that are highly prevalent in adolescents and adults who stutter (Briley et al., 2021; Iverach et al., 2009). In this study, there was evidence that the overall impact of stuttering, measured with the OASES-S, changed after treatment from moderate to mild-moderate. Children's attitude to communication, measured with the CAT, also improved substantively. Although total scores for the SCAS reduced but did not show significant improvements at a statistical level after 6 months, anxiety symptom improvements were significant by 12 months, indicating anxiety symptoms continued to improve. Social phobia subscale scores also significantly improved after 12 months of treatment. These gains are of interest because social anxiety disorder affects about 60% of adults who stutter (Blumgart et al., 2010; Iverach & Rapee, 2014). It is likely that these speech-related anxiety patterns are emerging during the school years (Iverach et al., 2016), so the Lidcombe Program may contribute to minimising potential anxiety patterns developing into clinically significant social anxiety in later years.

The reasons for these psychosocial improvements can only be speculative at this early phase in this research. However, it is possible that for these children a reduction or elimination of stuttering may have reduced negative peer reactions to speech, and hence reduced avoidance of speaking situations. This may have improved their overall communication experiences. None of the psychosocial measures were at clinically concerning levels before treatment, so at present it is unknown whether the Lidcombe Program might be associated with psychosocial improvements of clinically significant levels pre-treatment.

For the present Lidcombe Program stuttering reductions with school-age children, mechanisms of action can also only be speculative because there is still no clarity about this feature of the treatment with pre-school children, for whom it was originally developed. Amato Maguire et al., 2023 reviewed this topic, citing evidence that neither post-treatment acoustic nor linguistic changes are involved in reported treatment effects, and citing an ambiguous empirical literature about whether the parent verbal contingencies are a mechanism of action. Amato Maguire (2023) introduced an additional empirically-driven suggestion that parent interturn speaker latency may be a mechanism of action for the Lidcombe Program. Theoretical speculation about this matter by Zebrowski & Arenas (2011) included the idea that common factors underlie all treatments (Asay & Lambert, 2004; Wampold et al., 1997). Accordingly, Zebrowski and Arenas suggested that Lidcombe Program effects can be explained by "the quality of the client-clinician relationship" and "the client and the clinician's hope or expectation that change can and will occur" (Zebrowski & Arenas, 2011, p. 148). Shenker et al. (2023) offered theoretical speculation that childhood stuttering interventions "reap the benefits of heightened neuroplasticity in early childhood" and that they stimulate "brain development toward more typical growth patterns" (p. 3). Clearly, then, there is much empirical work remaining to be done to resolve this issue definitively.

The results of this study are encouraging for school-age children. However, the pre-to-post-treatment design potentially overestimated effect sizes. Without a no-treatment control group, we cannot be certain if the changes in the outcome variables are indeed a treatment effect. The study design does not exclude possible influence of other variables, such as natural progression of the disorder. The participant group included a range of ages, with 70% of ages between 6–8 years. According to prospective reports, during that period natural recovery may still be occurring (Ambrose et al., 2015; Kefalianos et al., 2017; Yairi & Ambrose, 1999). The influence of natural recovery is therefore worth considering when interpreting the effect size of stuttering reduction post-treatment. To evaluate the effectiveness of the Lidcombe Program for school-age children more accurately, the findings from this study provide support for a randomized controlled trial comparing telehealth Lidcombe Program to no treatment as the next phase of this program of research.

There is also a need for longer-term follow up to determine whether Lidcombe Program treatment effects can be sustained for school-age children. This is especially important considering the increasing risk of relapse after successful treatment reported in older age groups (Block et al., 2005; Craig & Hancock, 1995; Cream et al., 2009). Families from the present trial will be assessed in 2–5 years to establish whether treatment gains were maintained.

Compared with pre-school children, natural recovery from stuttering without intervention is less likely for children 6–12 years of age because the tractability of stuttering decreases with age (Chang, 2014; Kaller et al., 2017). For the period until a randomized trial is available, parents of children 6–12 years of age who stutter, and who present at speech clinics for help with their child's speech, can be empirically informed that the Lidcombe Program provides a chance of halting further stuttering development for over two thirds of these children. The school-age years are a period of worsening tractability (Bothe et al., 2006; Ingham & Cordes, 1999; Koushik et al.,

2009; Onslow & Packman, 1997), but at least for some school-age children, this treatment could help prevent or minimize lifelong quality of life impairment associated with chronic stuttering. Funding Statement.

### Funding

This research was partly funded by an Australian Government Research Training Program Scholarship.

#### CRediT authorship contribution statement

Mark Jones: Writing – review & editing, Methodology, Investigation, Formal analysis, Data curation. Elaina Kefalianos: Writing – review & editing, Supervision, Resources, Methodology, Conceptualization. Georgina Johnson: Writing – review & editing, Writing – original draft, Visualization, Validation, Resources, Project administration, Methodology, Investigation, Formal analysis, Data curation, Conceptualization. Mark Onslow: Writing – review & editing, Supervision, Resources, Methodology, Conceptualization. Brenda Carey: Writing – review & editing, Validation, Supervision, Resources, Methodology, Investigation.

# **Declaration of Competing Interest**

None.

#### Data availability

The data that has been used is confidential.

#### Acknowledgements

The authors would like to thank Damien Liu-Brennan for his scientific copyediting contribution to this publication. The authors would also like to thank two anonymous reviewers for their useful input in developing this manuscript.

#### Appendix A

Participant descriptive information.

| Participant | Age at recruitment<br>(year; month) | Biological<br>sex | Age of onset<br>(year;<br>month) <sup>a</sup> | Family history of stuttering <sup>a</sup> | Stuttering treatment history | Languages spoken other than English | Parent who<br>administered the<br>treatment |  |
|-------------|-------------------------------------|-------------------|---|---|------------------------------|-------------------------------------|---|--|
| 1           | 10;3                                | Male              | 7;0   | No  | None                         | None                                | Mother                                      |  |
| 2           | 7;7                                 | Male              | 2;0   | No  | None                         | Lebanese                            | Father                                      |  |
| 3           | 8;5                                 | Male              | 8;5   | No  | None                         | Arabic                              | Mother                                      |  |
| 4           | 6;10                                | Male              | 4;5   | No  | None                         | None                                | Mother                                      |  |
| 5           | 8;6                                 | Male              | 6;0   | No  | None                         | None                                | Mother                                      |  |
| 6           | 8;3                                 | Male              | 4;6   | No  | None                         | None                                | Mother                                      |  |
| 7           | 6;3                                 | Male              | 3;0   | No  | None                         | None                                | Mother                                      |  |
| 8           | 6;1                                 | Male              | 3;0   | Yes                                       | None                         | None                                | Mother                                      |  |
| 9           | 7;1                                 | Male              | 6;5   | No  | None                         | None                                | Mother & Father                             |  |
| 10          | 6;5                                 | Male              | 4;0   | Yes                                       | Lidcombe Program             | None                                | Mother                                      |  |
| 11          | 6;10                                | Male              | 4;0   | No  | Lidcombe Program             | None                                | Mother                                      |  |
| 12          | 7;9                                 | Male              | 4;0   | No Lidcombe Program Brazilian             |                              | Mother                              |   |  |
|             |                                     |                   |   |   |                              | Portuguese                          |   |  |
| 13          | 6;2                                 | Male              | 6;0   | No  | None                         | Arabic                              | Mother                                      |  |
| 14          | 6;4                                 | Male              | 4;5   | Yes                                       | None                         | Telegu and Tamil                    | Mother                                      |  |
| 15          | 6;1                                 | Female            | 4;0   | Yes                                       | None                         | None                                | Mother                                      |  |
| 16          | 10;8                                | Male              | 4;0   | No  | Lidcombe Program             | Greek                               | Mother                                      |  |
| 17          | 6;7                                 | Male              | 3;0   | No  | None                         | None                                | Mother                                      |  |
| 18          | 10;11                               | Male              | 9;6   | No  | None                         | None                                | Mother                                      |  |
| 19          | 7;6                                 | Male              | 5;0   | No  | None                         | None                                | Mother                                      |  |
| 20          | 7;7                                 | Male              | 2;0   | Yes                                       | None                         | None                                | Mother                                      |  |
| 21          | 10;0                                | Male              | 3;0   | Yes                                       | Lidcombe Program             | None                                | Mother                                      |  |
| 22          | 6;5                                 | Male              | 3;0   | Yes                                       | None                         | Mandarin Chinese                    | Mother                                      |  |
| 23          | 9;11                                | Male              | 4;0   | Yes                                       | None                         | None                                | Mother                                      |  |
| 24          | 11;9                                | Female            | 4;0   | No  | None                         | None                                | Mother                                      |  |
| 25          | 10;0                                | Male              | 7;6   | No  | None                         | Mandarin Chinese                    | Mother                                      |  |
| 26          | 9;1                                 | Male              | 6;0   | Yes                                       | None                         | None                                | Mother                                      |  |

(continued on next page)

#### (continued)

| Participant | Age at recruitment<br>(year; month) | Biological<br>sex | Age of onset<br>(year;<br>month) <sup>a</sup> | Family history of stuttering <sup>a</sup> | Stuttering treatment history | Languages spoken other than English | Parent who<br>administered the<br>treatment |  |
|-------------|-------------------------------------|-------------------|---|---|------------------------------|-------------------------------------|---|--|
| 27          | 7;7                                 | Male              | 3;0   | No  | None                         | Vietnamese                          | Mother                                      |  |
| 28          | 8;1                                 | Male              | 5;0   | No  | None                         | None                                | Mother                                      |  |
| 29          | 6;7                                 | Male              | 4;5   | No  | None                         | None                                | Mother                                      |  |
| 30          | 7;8                                 | Male              | 4;7   | No  | Lidcombe Program             | None                                | Mother & Father                             |  |
| 31          | 6;0                                 | Male              | 3;0   | Yes                                       | None                         | None                                | Mother                                      |  |
| 32          | 9;4                                 | Male              | 4;0   | No  | None                         | Mandarin Chinese                    | Father                                      |  |
| 33          | 6;0                                 | Male              | 3;8   | No  | None                         | Turkish                             | Mother                                      |  |
| 34          | 6;1                                 | Male              | 3;0   | Yes                                       | None                         | None                                | Mother                                      |  |
| 35          | 10;1                                | Male              | 7;0   | Yes                                       | None                         | None                                | Mother                                      |  |
| 36          | 10;10                               | Male              | 3;6   | Yes                                       | Lidcombe Program;            | None                                | Mother                                      |  |
| 07          |                                     |                   | 1.0   |   | Syllable-timed<br>speech     |                                     |   |  |
| 37          | 7;8                                 | Male              | 4;0   | Yes                                       | None                         | Mandarin Chinese                    | Mother                                      |  |

<sup>a</sup>This is based on parent report from case history survey.

#### Appendix **B**

Participant raw scores for the primary and secondary outcome measures collected before treatment, at 6 and at 12 months after starting treatment .

|             |          |          |          |           | Total scores |           |                   |                   |                   |                  |                  |                  |                  |                  |                  |
|-------------|----------|----------|----------|-----------|--------------|-----------|-------------------|-------------------|-------------------|------------------|------------------|------------------|------------------|------------------|------------------|
| Participant | A1<br>SR | A2<br>SR | A3<br>SR | A1<br>CAT | A2<br>CAT    | A3<br>CAT | A1<br>OASES-<br>S | A2<br>OASES-<br>S | A3<br>OASES-<br>S | A1<br>SCAS-<br>C | A2<br>SCAS-<br>C | A3<br>SCAS-<br>C | A1<br>SCAS-<br>P | A2<br>SCAS-<br>P | A3<br>SCAS-<br>P |
| 1           | 1        | 1        | 3        | 3         | 4            | 5         | 1.79              | 1.75              | 1.75              | 6                | 18               | 17               | 0                | 7                | 7                |
| 2           | 7        | 0        | x        | 30        | 29           | x         | 2.92              | 2.64              | x                 | 24               | 21               | x                | 24               | 20               | x                |
| 3           | 3        | 0        | 2        | 4         | 8            | 1         | 2.06              | 1.71              | 1.49              | 25               | 26               | 17               | 37               | 25               | 15               |
| 4           | 4        | 1        | 4        | 5         | 2            | 6         | 1.72              | 1.92              | 1.78              | 7                | 6                | 8                | 8                | 8                | 12               |
| 5           | 3        | 2        | 1        | 17        | 2            | 5         | 2.38              | 1.56              | 1.80              | 27               | 39               | 20               | 25               | 28               | 14               |
| 6           | 2        | 0        | 2        | 23        | 15           | 12        | 2.74              | 2.43              | 2.38              | 23               | 17               | 21               | 24               | 17               | 16               |
| 7           | 1        | 0        | 0        | 1         | 1            | 2         | 1.57              | 1.26              | 1.49              | 13               | 16               | 16               | 14               | 16               | 12               |
| 8           | 5        | 0        | 0        | 4         | 1            | 0         | 1.79              | 1.60              | 1.46              | 17               | 13               | 14               | 21               | 17               | 21               |
| 9           | 6        | 1        | 0        | 27        | 18           | 9         | 3.37              | 2.33              | 1.77              | 20               | 12               | 7                | 14               | 13               | 8                |
| 10          | 6        | 2        | 2        | 1         | 5            | 3         | 1.58              | 1.78              | 1.58              | 24               | 26               | 24               | 13               | 19               | 13               |
| 11          | 4        | 1        | 0        | 5         | 12           | 4         | 2.31              | 2.55              | 2.05              | 26               | 21               | 12               | 15               | 27               | 14               |
| 12          | 3        | 0        | 0        | 4         | 5            | 0         | 2.02              | 1.65              | 1.44              | 31               | 18               | 19               | 25               | 11               | 13               |
| 13          | 4        | 3        | 4        | 17        | 15           | 10        | 1.64              | 2.15              | 1.69              | 28               | 21               | 13               | 16               | 13               | 10               |
| 14          | 3        | 1        | 0        | 16        | 3            | 2         | 2.20              | 1.53              | 1.81              | 19               | 15               | 10               | 12               | 5                | 7                |
| 15          | 5        | 2        | 1        | 22        | 3            | 5         | 3.25              | 1.78              | 2.05              | 48               | 11               | 21               | 43               | 13               | 20               |
| 16          | 3        | 1        | 2        | 24        | 18           | 22        | 2.98              | 2.53              | 2.57              | 25               | 26               | 15               | 24               | 23               | 17               |
| 17          | 4        | 4        | 4        | 1         | 2            | 6         | 1.71              | 1.36              | 1.76              | 31               | 25               | 33               | 20               | 15               | 22               |
| 18          | 3        | 0        | 1        | 26        | 10           | 19        | 3.26              | 2.73              | 3.25              | 44               | 57               | 48               | 27               | 32               | 17               |
| 19          | 6        | 4        | 3        | 22        | 19           | 27        | 2.71              | 2.75              | 3.43              | 48               | 40               | 39               | 25               | 22               | 10               |
| 20          | 4        | 0        | 0        | 2         | 2            | 3         | 1.74              | 1.30              | 1.22              | 23               | 15               | 15               | 13               | 4                | 6                |
| 21          | 5        | 5        | 6        | 15        | 8            | 21        | 2.75              | 2.35              | 2.57              | 18               | 9                | 10               | 16               | 9                | 7                |
| 22          | 8        | 0        | 0        | 15        | 16           | 9         | 3.14              | х                 | х                 | 27               | 25               | 24               | 24               | 22               | 23               |
| 23          | 6        | 2        | 1        | 26        | 19           | 11        | 2.81              | 2.50              | 2.12              | 32               | 27               | 20               | 32               | 25               | 17               |
| 24          | 2        | 1        | 2        | 21        | 21           | 11        | 2.83              | 2.82              | 2.75              | 42               | 37               | 28               | 42               | 22               | 18               |
| 25          | 5        | 0        | 0        | 11        | 4            | 2         | 2.28              | 1.86              | 1.45              | 39               | 23               | 17               | 18               | 12               | 10               |
| 26          | 4        | 1        | 0        | 22        | 11           | 6         | 3.21              | 2.35              | 1.98              | 16               | 13               | 28               | 22               | 15               | 11               |
| 27          | 1        | 0        | 0        | 11        | 1            | 2         | 1.96              | 1.51              | 1.49              | 10               | 4                | 7                | 7                | 7                | 6                |
| 28          | 4        | 3        | 1        | 11        | 12           | 6         | 2.04              | 1.83              | 1.86              | 21               | 22               | 16               | 11               | 8                | 15               |
| 29          | 3        | 1        | 1        | 5         | 9            | 1         | х                 | 2.07              | 1.38              | 15               | 24               | 6                | 9                | 14               | 6                |
| 30          | 4        | 2        | 0        | 10        | 1            | 0         | 2.05              | 1.61              | 1.43              | 13               | 11               | 4                | 8                | 13               | 8                |
| 31          | 4        | 1        | x        | 21        | 19           | x         | 3.00              | 2.81              | х                 | 23               | 21               | х                | 17               | 17               | x                |
| 32          | 3        | 1        | 1        | 11        | 12           | 16        | 2.43              | 2.31              | 2.38              | 25               | 38               | 37               | 21               | 27               | 21               |
| 33          | 2        | 1        | 2        | 17        | 13           | 3         | 3.63              | 2.07              | 2.13              | 11               | 21               | 29               | 10               | 14               | 19               |
| 34          | 4        | 1        | 0        | 4         | 3            | 2         | 1.63              | 1.39              | 1.33              | 6                | 6                | 4                | 6                | 3                | 3                |
| 35          | 7        | 7        | 4        | 24        | 17           | 24        | 3.40              | 2.53              | 2.64              | 41               | 4                | 34               | 6                | 2                | 4                |
| 36          | 2        | 2        | 0        | 11        | 3            | 7         | 2.72              | 2.25              | 2.08              | 41               | 48               | 24               | 41               | 39               | 22               |
| 37          | 3        | 0        | 0        | 19        | 17           | 4         | 2.49              | 1.80              | 1.95              | 25               | 19               | 19               | 8                | 18               | 11               |

*Note:* A1 = before starting treatment, A2 = 6 months after starting treatment, A3 = 12 months after starting treatment; SR = Clinician-reported stuttering severity rating using the Lidcombe Program rating scale (Onslow et al., 2021); CAT = Communication Attitude Test, OASES-S =  $(1 + 1)^{-1}$ 

Overall Assessment of the Speaker's Experience of Stuttering – School version (Yaruss & Quesal, 2016); SCAS-C = Spence Children's Anxiety Scale – Child version; SCAS-P = Spence Children's Anxiety Scale – Parent version (Spence, 1998).

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