Follow-up of 6–10-year-old stuttering children after Lidcombe Program treatment: A Phase I trial

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Abstract

Purpose: This Phase I trial sought to establish (1) whether the Lidcombe Program is viable for school-age children, (2) whether there is any indication that it requires modification for school-age children, (3) whether treatment effects are durable, (4) how many treatment sessions appear to be required to significantly reduce stuttering frequency and (5) whether there is an association between follow-up period and relapse tendency.

Method: Twelve children were treated, and one required an addition to the Lidcombe Program. The results for this child were excluded from group analysis, leaving a group of 6–10 year-olds. A retrospective method was used using routine pre-treatment clinic recordings. At follow-up, all children were telephoned and audio-recorded three times at random times during the day within a 7–10-day period.

Results: A blinded observer’s mean percent syllables stuttered score pre-treatment was 9.2 and 1.9 at follow-up. No association was found between follow-up period and stuttering rates. The mean syllables per minute score pre-treatment was 145.8 and 179.3 at follow-up. These results were attained in a median of eight clinic visits with a range of 6–10 visits.

Conclusions: Procedurally, the Lidcombe Program is viable for school-age children and parents report enjoyment in administering it. There appears to be a treatment effect that can be attained in a reasonable number of clinical hours. These results compel continued exploration with young school-aged children in subsequent Phase II and III studies.

Educational objectives: The reader will be able to: (1) summarize the status of clinical trials for stuttering school-age children, (2) describe the phases of clinical trial development, (3) evaluate outcomes the Lidcombe Program for a school-age population in terms of stuttering reduction and treatment time, (4) evaluate the suitability of the Lidcombe Program with population of school-age stuttering children, and (5) provide an interpretation of the finding of no correlation between follow-up and post-treatment stuttering rates.

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1. Introduction

It appears that stuttering is most tractable during the preschool years and that responsiveness to treatment decreases with age, presumably because of the accompanying decrease of neural plasticity (Wohlett & Smith, 2002). On balance, available data indicate that stuttering becomes less responsive to treatment after the preschool years (see Bothe, 2004; Bothe, Davidow, Bramlett, Franic, & Ingham, 2006; Bothe, Davidow, Bramlett, & Ingham, 2006; Ingham, 1984a, 1984b; Ingham & Cordes, 1999; Onslow, 1996; Onslow & Packman, 1997; Onslow & Packman, 1999; Prins & Ingham, 1983). The school-age period from 6 to 12 years of age might then be thought of as an important time of life for the receipt of an efficacious stuttering treatment. Without an efficacious treatment during the school years, the likelihood that stuttering will persist may increase with age.

As a child enters the school years the demands on life naturally increase in terms of school work and social activities. Therefore, finding time to treat school-age children may be problematic as they have limited time to dedicate to treatment approaches. Other factors to consider for school-age children are a longer history of stuttering, previous treatment failures and the possible suffering of teasing and bullying. For these reasons efficient treatment approaches are important for this population of children.

Onslow, Jones, O’Brian, Menzies, and Packman (2008) argue that methodological requirements for a clinical trial should be prospective evaluation of an entire treatment, where speech data are collected outside the clinic with a follow-up period of at least 3 months if an effect is found. Further, Onslow et al. argue that discussion of effect sizes for different stuttering treatments is currently not appropriate; most stuttering trials at present are non-randomized, and their results invariably introduce biases that inflate effect sizes (Kunz & Oxman, 1998).

There have been nine published clinical trials for school-age children, involving 131 school-age children. These include preliminary Phase I trials of Gradual Increase in Length and Complexity of Utterance (Ryan & Van Kirk Ryan, 1995) and regulated breathing (De Kinkelder & Boelens, 1998). There have been two Phase I trials (Ryan & Van Kirk Ryan, 1983; Ryan & Van Kirk Ryan, 1995) and one Phase II trial (Lincoln, Onslow, Lewis, & Wilson, 1996) of verbal response contingent stimulation. A successful Phase II trial of EMG biofeedback has been reported, (Craig et al., 1996; Hancock et al., 1998), although there has been a failure to replicate those results (Block, Onslow, Roberts, & White, 2004). Speech restructuring involves the reduction of stuttered speech by changes in various aspects of speech production, such as reduced rate, prolonged vowels, “soft” articulatory contacts, and “gentle” vowel onsets (Packman, Onslow, & Menzies, 2000). The most popular of these programs establish stutter-free speech at a slow speech rate. Commonly, speech is then shaped to near-normal rates through programmed instruction and transferred to everyday situations. There have been three Phase I trials of this procedure with school-age children (Boberg & Kully, 1994; Budd, Madison, Izkowitz, George, & Price, 1986; Kully & Boberg, 1991; Ryan & Van Kirk Ryan, 1995) and two Phase II trials (Craig et al., 1996; Hancock et al., 1998). No Phase III trials, incorporating randomized clinical trials evidence, have been reported.

The evidence for speech restructuring is the strongest available for school-age children, with four independent replications, and no published failures to replicate. However, there may be some concerns with this style of treatment that make it problematic for school-age children. In the first instance, this style of treatment is based on multi-day, intensive treatment formats that were developed for adults. These methods typically require more than 100 clinical hours (Andrews, Guitar, & Howie, 1980; Onslow, Costa, Andrews, Harrison, & Packman, 1996). It certainly is possible for such a treatment format to be applied to school-age children with success. In fact two of the participants in the Onslow et al. (1996) trial were of school-age, and one of them, 7-year old Participant DT, had favorable outcomes at 9 years post-treatment (Onslow, O’Brian, Packman, Rousseau, 2004).
Cobbin (2002) found it necessary to develop and test a relapse management procedure for children in this age group.

The Lidcombe Program of Early Stuttering Intervention was developed for preschool-aged children. A meta-analysis of randomized clinical evidence for 34 children showed an odds ratio of 7.5 (Onslow, Jones, Menzies, O’Brian, & Packman, in press). However, the treatment has had limited trialling with school-age children. The Lincoln et al. report was based on a much earlier version of the treatment. Verbal response contingent stimulation is non-programmed and does not involve control of stuttering with a speech pattern. As such it is potentially suitable for school-age children. The present study contributes to the development of that treatment with a Phase I trial of this treatment for school-age children, in order to determine whether Phase II and Phase III clinical trials are justified.

Specifically, this Phase I trial seeks to establish whether (1) the Lidcombe Program is viable for school-age children, (2) whether there is any indication that it requires modification for school-age children, (3) whether treatment effects are durable over time, (4) how many treatment sessions appear to be required to significantly reduce stuttering frequency and (5) is there any association between follow-up period and relapse tendency. The Lidcombe Program focuses on speech outcomes (Australian Stuttering Research Centre, 2008), hence the present Phase I trial of the Lidcombe Program focuses on speech outcomes. If the Lidcombe Program appears to be associated with positive speech outcomes, then Phase II and III trials can explore whether important non-speech outcomes—such as speech-related anxiety and bullying rates—accompany its intended treatment effects. Randomized Phase II and III clinical trials designs would be necessary to make such a determination, and would guide future clinical trials development for this population.

2. Method

2.1. Participants

Participants were 12 children taken in order from the treatment waiting list at the Montreal Fluency Centre, a speech and language clinic in Montreal, Canada. Participant 12 received an addition to the Lidcombe Program therefore was removed from the group analysis. The children were between the ages of 6 years 8 months and 10 years 8 months (mean = 9 years 0 months) at the start of treatment. Ten were boys and two were girls. Stuttering was diagnosed by the presence of unambiguous stuttering (Jones et al., 2008) in the clinic sample according to the judgement of the treating speech-language pathologist (SLP) and confirmation by the parent. Exclusion criteria were pre-treatment stuttering less than 2.0%SS as determined by the assessing SLP, first language not English, and most proficient language not English. Language proficiency was determined with the Bilingual Language History and Proficiency form (Roberts & Shenker, 2007).

2.2. Assessments

On the day of the assessment, the clinician measured stuttering severity in the clinic with the standard Lidcombe Program scale of 1 = no stuttering, 2 = extremely mild stuttering and 10 = extremely severe stuttering (Jones et al., 2005). The use of this scale is standard during the Lidcombe Program, and clinician severity scores are used at each clinic visit to calibrate parent severity scores. Additionally, pre-treatment audio-recorded speech samples of the 12 children were obtained within the clinic on the day of assessment, 1–2 weeks prior to treatment. One 10-min, pre-treatment audio recording was obtained for each child during conversation with the clinician in the presence of the parent/s. These audio recordings were routine practice in the clinic.

Follow-up assessments were conducted with random telephone calls to the children at home, in English. This method was chosen for its validity in preference to parents and children returning to the clinic for follow-up assessment. The latter method would incur potential bias from discriminated learning to the clinic of stutter-free speech, and participant selection bias of those parents willing to travel to the clinic. Additionally, unpredictable telephone calls were chosen because of their validity in preference to having parent audio- or video-recording the children in conversation. The latter method allows participants to respond to “demand characteristics” (Orne, 1962)—cues that inform participants what behaviour is sought from researchers—by self selection of speech sampling situations. The audio sampling procedure was considered suitable in light of evidence with children that audio only percent syllables stuttered (%SS) scores will not underestimate stuttering rates of post-treatment samples (Rousseau, Onslow, Packman, & Jones, 2008). The assessment requirements for the study incurred little participation bias because pre-treatment audio recordings
were routine in the clinic. At follow-up, children needed to be available for three telephone calls in their home environment.

Within a 7–10-day period, each of the children was telephoned three times, approximately every second day, at random times during the day. All follow-up speech samples were audio-recorded with an Olympus Digital Voice Recorder, using a telephone jack device. The conversations were approximately 10-min in duration and the child was asked open-ended questions in order to collect sufficient speech samples. The mean period post the conclusion of Stage 1 that the 11 children were followed up was 70 weeks with a range of 9–187 weeks.

On the day of the first follow-up telephone call, each parent was interviewed about their experience with the Lidcombe Program. The interviews were conducted by telephone before the conversations with the children and the responses were reported on a standard form and subsequently collated. The parent interviews and follow-up telephone calls were conducted by a graduate student in speech-language pathology who was blinded to the purposes of the study and who had no knowledge of the parents and children.

Two of the children had English as their dominant language with French as a second language introduced at 4 years or older. At the time of treatment, these children had very limited exposure to French and thus were categorized as English dominant. Three children were bilingual English with French, Hebrew or Italian introduced simultaneously from birth. The predominant language spoken by these three children was English. The remaining six children were introduced to a second language at 4 years or older with English as their predominant language and second language of Italian, Greek or Portuguese. Parents reported stuttering in all languages spoken. Of these 11 children, seven did not present with any other speech or language concerns, one presented with a receptive and expressive language disorder, one with an articulation disorder and one with an expressive language disorder and diagnosed attention deficit hyperactivity disorder.

All children received treatment in English and verbal contingencies applied by parents beyond the clinic were in English. For those children presenting with other speech or language concerns, remediation of those concerns followed treatment for stuttering. Details of the children are presented in Table 1.

2.3. The treatment

The treatment was the Lidcombe Program of Early Stuttering Intervention (Jones et al., 2005; Onslow, Packman, & Harrison, 2003). The Lidcombe Program is a behavioral treatment developed for preschool children who stutter. It involves verbal response contingent stimulation treatment and is administered by parents. The Lidcombe Program incorporates weekly visits to the speech clinic by the child and parent, during which the SLP trains the parent to present three verbal contingencies for the child’s stutter-free speech, and two verbal contingencies for unambiguous stuttering. The verbal contingencies for stutter-free speech are acknowledgment, praise and request for self-evaluation. The contingencies for unambiguous stuttering are acknowledgement and request for self-correction. Initially, the parent presents these verbal contingencies in short, structured conversations each day that are designed to maximize the occurrence of contingencies for stutter-free speech. The parent then administers the contingencies in everyday conversations. Clinician speech measures in the clinic and parent speech measures beyond the clinic are used to guide the clinical process. Criteria for ending Stage 1 of the treatment and beginning Stage 2 (maintenance) are “(1) Percent syllables stuttered (%SS) less than 1.0 within the clinic, and (2) severity ratings (SRs) scores for the previous week of either 1 or 2, with at least four of these being 1” (Australian Stuttering Research Centre, 2008, p. 8).

For all children, the Lidcombe Program was conducted in the child’s strongest language, which was English. Verbal contingencies applied beyond the clinic were also provided in English. The criteria for ending Stage 1 of treatment and beginning Stage 2 were more stringent than for manualized for preschool children (see above). Since the clinical response of these children was much quicker than for preschool children (see below), it was felt that imposing more stringent criteria for entry into Stage 2 would create a hedge against relapse during the early weeks of Stage 2. In the present study, the criteria for entering Stage 2 were (1) %SS less than 1.0 within the clinic, and (2) SR scores for the previous week of “1” each day for either three consecutive visits or two consecutive clinic visits spanning 3 weeks during which there was an intervening non-attendance at the clinic (3-week period).

2.4. Outcome measures

The primary outcome measure was percent syllables stuttered (%SS). A secondary speech outcome was syllables per minute (SPM). A non-speech secondary outcome was the number of clinic sessions to reach Stage 2. The latter
Table 1
Details of the participants. Stuttering scores in the last two columns are from the blinded, independent observer.

<table>
<thead>
<tr>
<th>Participant</th>
<th>Gender</th>
<th>Previous treatment</th>
<th>Details of previous treatment</th>
<th>Subsequent treatment</th>
<th>Details of subsequent treatment</th>
<th>Language background</th>
<th>Other speech/language concerns</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Boy</td>
<td>Yes</td>
<td>Articulation treatment</td>
<td>No</td>
<td></td>
<td>L1 = E; L2 = I;</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>L3 = F</td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>Boy</td>
<td>Yes</td>
<td>4–6 stuttering treatment sessions at 3 years of age</td>
<td>No</td>
<td></td>
<td>L1 = H; L2 = E;</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>L3 = F</td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>Girl</td>
<td>No</td>
<td></td>
<td>No</td>
<td></td>
<td>L1 = E; L2 = F</td>
<td></td>
</tr>
<tr>
<td>4</td>
<td>Boy</td>
<td>Yes</td>
<td>3–4 stuttering treatment sessions, articulation treatment</td>
<td>No</td>
<td></td>
<td>L1 = E; L2 = I;</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>L3 = F</td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>Girl</td>
<td>No</td>
<td></td>
<td>No</td>
<td></td>
<td>L1 = E; L2 = F</td>
<td></td>
</tr>
<tr>
<td>6</td>
<td>Boy</td>
<td>Yes</td>
<td>Stuttering treatment at school which involved exercises for breathing</td>
<td>No</td>
<td></td>
<td>L1 = E; L2 = I;</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>L3 = F</td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>Boy</td>
<td>No</td>
<td></td>
<td>No</td>
<td></td>
<td>L1 = E; L2 = F</td>
<td>Expressive language disorder</td>
</tr>
<tr>
<td>8</td>
<td>Boy</td>
<td>Yes</td>
<td>Stuttering treatment at school which involved breathing and pausing</td>
<td>No</td>
<td></td>
<td>L1 = F; L2 = E;</td>
<td>ADHD/expressive language disorder</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>L3 = P</td>
<td></td>
</tr>
<tr>
<td>9</td>
<td>Boy</td>
<td>No</td>
<td></td>
<td>Yes</td>
<td>1 follow-up session at Children’s hospital</td>
<td>L1 = E; L2 = F</td>
<td></td>
</tr>
<tr>
<td>10</td>
<td>Boy</td>
<td>Yes</td>
<td>Articulation treatment</td>
<td>No</td>
<td></td>
<td>L1 = E; L2 = F;</td>
<td>Articulation disorder</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>L3 = I</td>
<td></td>
</tr>
<tr>
<td>11</td>
<td>Boy</td>
<td>No</td>
<td></td>
<td>No</td>
<td></td>
<td>L1 = E; L2 = G;</td>
<td>Receptive and expressive language disorder</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>L3 = F</td>
<td></td>
</tr>
<tr>
<td>12</td>
<td>Boy</td>
<td>No</td>
<td></td>
<td>No</td>
<td></td>
<td>L1 = E; L2 = F</td>
<td></td>
</tr>
</tbody>
</table>

L1, first language; L2, second language; L3, third language. Language: E, English; F, French; G, Greek; H, Hebrew; I, Italian; P, Portuguese. Note: Participant 11 entered Stage 2 with speech criteria more liberal than the study criteria. Participant 12 received a speech-restructuring additive to treatment and was not included in the group report.

The follow-up measure is important in terms of cost efficiency of treatment delivery, and benchmarks are available for the Lidcombe Program. The collective findings of previous clinical trials (Jones et al., 2005; Onslow, Andrews, & Lincoln, 1994; Rousseau, Packman, Onslow, Harrison, & Jones, 2007) and large cohort recovery plot studies (Jones, Onslow, Harrison, & Packman, 2000; Kingston, Huber, Onslow, Jones, & Packman, 2003) convey that a median of around 12 treatment sessions are required for preschool children to be at the requisite low stuttering levels for entry to Stage 2 of treatment (maintenance). With the imposition of more rigorous criteria in the recent version of the treatment manual (Australian Stuttering Research Centre, 2008), Rousseau et al. reported a median of 16 clinic visits to attain Stage 2. Consistent with the methodological recommendations of Bothe, Davidow, Bramlett, Franic, et al. (2006) and Bothe, Davidow, Bramlett, and Ingham (2006), speech naturalness was not included as an outcome because the treatment does not involve the use of a novel speech pattern. Prior to therapy, there was no instruction for the children to use speech restructuring nor were they asked to change the way they spoke throughout the treatment.

A total of 44 speech samples (11 participants × 4 assessments) were obtained. The follow-up samples were pooled to provide 11 treatment samples, each of approximately 30 min duration. The mean number of syllables in the pooled follow-up samples was 1427 syllables (SD = 218, range 828–1287). The mean number of syllables in the pre-treatment samples was 439 (SD = 72, range 274–500). All speech samples were transferred to a Windows XP operating system.
Table 2
Details of follow-up samples.

<table>
<thead>
<tr>
<th>Participant</th>
<th>Gender</th>
<th>Age at onset of treatment</th>
<th>Number of weeks to Stage 2</th>
<th>Age at follow-up</th>
<th>Number of weeks at follow-up</th>
<th>Pre-treatment %SS</th>
<th>Severity rating at assessment</th>
<th>Mean %SS at follow-up 9–187 weeks</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Boy</td>
<td>10;3</td>
<td>6</td>
<td>10;7</td>
<td>9</td>
<td>1.9^b</td>
<td>4</td>
<td>1.0</td>
</tr>
<tr>
<td>2</td>
<td>Boy</td>
<td>9;5</td>
<td>7</td>
<td>9;10</td>
<td>12</td>
<td>16.1</td>
<td>8</td>
<td>3.6</td>
</tr>
<tr>
<td>3</td>
<td>Girl</td>
<td>6;9</td>
<td>8</td>
<td>7;8</td>
<td>38</td>
<td>12.5</td>
<td>5</td>
<td>0.9</td>
</tr>
<tr>
<td>4</td>
<td>Boy</td>
<td>10;5</td>
<td>10</td>
<td>11;10</td>
<td>59</td>
<td>2.0</td>
<td>4</td>
<td>3.8</td>
</tr>
<tr>
<td>5</td>
<td>Girl</td>
<td>8;9</td>
<td>6</td>
<td>12;9</td>
<td>187</td>
<td>2.1</td>
<td>4</td>
<td>0.4</td>
</tr>
<tr>
<td>6</td>
<td>Boy</td>
<td>8;11</td>
<td>6</td>
<td>11;1</td>
<td>101</td>
<td>11.1</td>
<td>5</td>
<td>2.1</td>
</tr>
<tr>
<td>7</td>
<td>Boy</td>
<td>7;4</td>
<td>10</td>
<td>8;10</td>
<td>66</td>
<td>6.9</td>
<td>6</td>
<td>1.3</td>
</tr>
<tr>
<td>8</td>
<td>Boy</td>
<td>10;3</td>
<td>9</td>
<td>11;5</td>
<td>12</td>
<td>6.9</td>
<td>5</td>
<td>1.3</td>
</tr>
<tr>
<td>9</td>
<td>Boy</td>
<td>9;4</td>
<td>6</td>
<td>11;8</td>
<td>108</td>
<td>2.3</td>
<td>4</td>
<td>0.2</td>
</tr>
<tr>
<td>10</td>
<td>Boy</td>
<td>9;8</td>
<td>8</td>
<td>12;5</td>
<td>125</td>
<td>12.5</td>
<td>6</td>
<td>2.8</td>
</tr>
<tr>
<td>11</td>
<td>Boy</td>
<td>6;8</td>
<td>8</td>
<td>7;11</td>
<td>54</td>
<td>27.3</td>
<td>8</td>
<td>3.4</td>
</tr>
<tr>
<td>12</td>
<td>Boy</td>
<td>10;8</td>
<td>7</td>
<td>14;1</td>
<td>155</td>
<td>8.3</td>
<td>6</td>
<td>2.5</td>
</tr>
</tbody>
</table>

^ Severity rating = a scale from 1 to 10, where 1 = no stuttering and 10 = extreme stuttering for that child. For this measure the clinician and parent jointly rated the severity of stuttering on the day of the assessment.

^b Exclusion criteria were pre-treatment stuttering less than 2.0%SS as determined by the assessing SLP. However, the blinded observer's score for this child differed slightly from the assessing clinician.

and stored in a media audio format. The observer of all speech samples in the study was an independent, New Zealand based SLP who was blinded to the purposes of the study. All speech samples were evaluated for %SS. A button-press counting and timing device (True Talk) was used to measure %SS and SPM for each sample while listening to the sample through headphones. The speech recordings were presented to the blinded observer, de-identified and in random order. The observer was not instructed to base stuttering counts on behavioral criteria of any kind. Instead, consistent with measurement procedures of the Lidcombe Program (Onslow et al., 2003), the observer was instructed to use a perceptual procedure and to count as stuttering any speech events that were considered unambiguously to be so. The observer was experienced with the Lidcombe Program and consequently experienced with this measurement method. The measures obtained by the blinded observer are reported as outcomes for the trial and are presented in Table 2 (Fig. 1).

3. Results

3.1. Clinical progress

Only one child (Participant 12) required a supplement to the Lidcombe Program. This boy received the addition of a speech-restructuring component in addition to the Lidcombe Program. His results were excluded from the group results presented below for the 11 children who received only the Lidcombe Program. Of the 11 participants in total,
eight had completed Stage 2 at the time of the follow-up telephone calls. Three participants (Participant 1, 2 and 9) had not completed Stage 2 at that time but all subsequently did so.

3.2. Outcomes

3.2.1. Primary outcome

For the 11 school-age children who received the Lidcombe Program, the blinded observer’s mean %SS score pre-treatment was 9.2 (SD = 7.8) and 1.9 (SD = 1.3) at follow-up ($t_{[10]} = 5.77, p = .0002$). Six months after the original measurements, three pre-treatment and three follow-up recordings were randomly selected and re-represented to the blinded observer to determine intra-observer reliability. The tapes ranged in initial %SS scores of the blinded observer from 16.1 to 0. All second %SS scores differed from the first by 1.0%SS or less, with a correlation of .997 ($p < .001$). Despite large stuttering reductions at follow-up, three of the children’s follow-up stuttering rates were greater than 3.0%SS (see Table 2).

For inter-observer reliability purposes, these data were compared to those obtained using the same methods by the treating SLP in Canada. The unblinded treating clinician scored lower than the blinded observer, with mean %SS scores pre-treatment of 7.2 (SD = 5.1) and 0.9 (SD = 0.5). The difference between the two observers’ %SS scores pre-treatment was not significant ($t_{[10]} = 1.04, p = .33$). Difference between observers’ follow-up %SS scores was significant ($t_{[10]} = 5.31, p = .0003$). This result was predictable considering that the treating clinician was extremely familiar with the children. However, the %SS scores of the blinded and clinician observers correlated highly at .91 ($p < .001$).

3.2.2. Secondary outcomes

These results were attained in a median of eight clinic visits (Range 6–10 visits). For the 11 school-age children who received the Lidcombe Program, the blinded observer’s mean SPM pre-treatment score was 145.8 (SD = 22.7) and 179.3 (SD = 20.5) at follow-up ($t_{[10]} = 3.19, p = .0097$). The SPM scores of the blinded and clinician observers correlated significantly at .58 ($p < .05$). This result is consistent with improved verbal output associated with stuttering reduction. It also indicates that stuttering reductions were not attained at the cost of reduced speech rate.

3.2.3. Correlation between outcomes and follow-up period

As with any retrospective study, there was a wide range of follow-up periods as noted in Table 1. Pearson correlations were calculated for %SS scores at follow-up and number of weeks to follow-up in order to assess any tendencies for relapse during the post-treatment period. A nonsignificant correlation of .20 ($p = .51$) shows that the follow-up period explains only .04 of the variance in follow-up %SS scores. This indicates the absence of a relationship between relapse and duration of time post-treatment with this group of children.

3.2.4. Individual cases

One of the two children in the study with moderate language disorders (Participant 11) was admitted to Stage 2 with speech performance close to, but not below, the treatment criteria as stated above. This was the only child in the study with a receptive language disorder, however this did not appear to impede his understanding of verbal contingencies. However, receptive and expressive language difficulties seemed to be hindering his progress in attaining the study speech criteria. The clinician noted that when this child was formulating thoughts and ideas to explain something difficult, he tended to stutter more when he was unsure of how to formulate his utterances. Stuttering occurred when he had difficulty with vocabulary, sentence structure or the sequence of ideas. Further, he stuttered more on conjunctions that preceded new ideas. In linguistically undemanding conversational exchanges between this child and the clinician, %SS in the clinic was typically below 1.0. The parents of this child consistently assigned SRs of 1 and 2 for a period of three clinical sessions, without further progress. It was decided that this child would be admitted to Stage 2 with %SS measures in the clinic below 2.0 and a mean SR each week of 2 or below. During Stage 2, attempts were made to maintain these levels while he received language therapy. If the parent/s reported SRs higher than 2, they were asked to

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3 Because of small sample size, this and other paired t-tests in this report were log transformed prior to analysis, as recommended by Jones, Onslow, Packman, and Gebski (2006).
Table 3
Parent satisfaction with the Lidcombe Program.

<table>
<thead>
<tr>
<th>Parent questionnaire</th>
<th>%yes</th>
<th>%no</th>
</tr>
</thead>
<tbody>
<tr>
<td>Since your last visit to the clinic has your child’s fluency worsened?</td>
<td>20%</td>
<td>80%</td>
</tr>
<tr>
<td>Overall how would you rate child’s speech severity in terms of severity ratings (SRs), at this time?</td>
<td></td>
<td></td>
</tr>
<tr>
<td>SR 1 – 1 parent</td>
<td>10%</td>
<td></td>
</tr>
<tr>
<td>SR 2 – 6 parents</td>
<td>60%</td>
<td></td>
</tr>
<tr>
<td>SR 3 – 3 parents</td>
<td>30%</td>
<td></td>
</tr>
<tr>
<td>SR 4 – 0 parents</td>
<td>0%</td>
<td></td>
</tr>
<tr>
<td>Are you continuing to give feedback for child’s speech? (i.e. positive reinforcement for fluency, request for correction for stuttering)</td>
<td>80%</td>
<td>20%</td>
</tr>
<tr>
<td>Besides stuttering, do you have other speech and language concerns for your child?</td>
<td>20%</td>
<td>80%</td>
</tr>
<tr>
<td>Has your child been seen by another SLP for stuttering since leaving our clinic?</td>
<td>10%</td>
<td>90%</td>
</tr>
<tr>
<td>Did you enjoy participating in the LP?</td>
<td>100%</td>
<td></td>
</tr>
<tr>
<td>LP factors that were difficult?</td>
<td></td>
<td></td>
</tr>
<tr>
<td>finding 10 min a day for therapy</td>
<td>60%</td>
<td>40%</td>
</tr>
<tr>
<td>giving feedback to your child</td>
<td>30%</td>
<td>70%</td>
</tr>
</tbody>
</table>

increase their verbal contingencies in both 10-min structured and unstructured conversations until the SRs decreased to 2 or below. Participant 11, who received language treatment, was admitted to Stage 2 in eight clinic visits, with the above study criteria. The remaining 10 participants attained Stage 2 in a median of 7.5 clinic visits (mean = 7.6, range = 6–10 clinic visits).

For Participant 4, the %SS measures at follow-up were higher than pre-treatment conversation in the clinic, suggesting no treatment effect. With this child, Stage 2 goals for maintenance of stutter-free speech were not being met. During the parent interview, a number of compliance issues emerged. The parent was unique in providing verbal contingencies only for stuttered speech but no verbal contingencies—acknowledgement, praise and request for self-evaluation for stutter-free speech. This parent was one of several parents (see below) who reported difficulty in finding a short time each day for treatment in structured conversations.

3.3. Parent interviews

The parents of 10 of the school-age children in this study were interviewed about their satisfaction with the Lidcombe Program. The remaining 2 parents were not available for the interview. Using the Lidcombe Program SR scale (1 = no stuttering, 2 = extremely mild stuttering, 10 = extremely severe stuttering), seven (70%) parents assigned a score of 1 or 2 to their child’s speech, while three (30%) parents assigned scores of 3. No parents scored the child’s SR at 4 or greater. Eight parents (80%) reported that the child’s stuttering had not worsened since their last clinical visit and two (20%) reported a slight increase. Eight (80%) parents reported that they continued to provide occasional verbal contingencies to their children for stutter-free and stuttered speech. All parents indicated that they enjoyed participating in the Lidcombe Program, however six (60%) reported that they found it difficult to find time to conduct treatment in structured conversations each day, mainly because of busy home schedules. Table 3 summarizes the parent questionnaire responses.

4. Discussion

Given that stuttering is more tractable during childhood, it is clear that efficacious stuttering treatment for school-age children is of great importance. Verbal response contingent stimulation procedures such as the Lidcombe Program used in this study are not complicated by programmed instruction or the use of a novel speech pattern. Generally, this Phase I trial presented positive answers to the questions driving the research. Procedurally, the Lidcombe Program is viable for school-age children and parents report enjoyment in administering it. In all but one of the 12 children studied, it required no modification to the manualized procedures for school-age children. There is some reason to believe that
over a period of time, there is a stuttering reduction treatment effect that can be attained in a reasonable number of clinical hours. No association was found between follow-up period and stuttering rates, suggesting the durability of any treatment effects. These results compel continued exploration of the Lidcombe Program with young school-aged children in subsequent Phase II and III clinical trials.

For the 11 school-age children, the blinded observer’s mean %SS scores pre-treatment was 9.2 and 1.8 at follow-up. These results were obtained in a median of eight clinic visits. Four of those children at follow-up had %SS scores below 1.0, four had %SS scores below 3.0 and three had %SS scores below 4.0. The 11 children received treatment by a clinician in a different country than that in which the Lidcombe Program was originally developed. Therefore, this report independently substantiates the Lincoln et al. (1996) findings of the benefits of a much earlier version of the Lidcombe Program with this age group. The program is worthy of continued clinical trials development with this age group.

Although four of the 11 participants maintained an average %SS below 1.0% at follow-up, seven others did not achieve this criterion which suggests that there is more variability in Stage 2 outcomes for the school-age population compared to preschool children (Jones et al., 2005). This variability is comparable with the findings obtained for school-age children from Lincoln et al. (1996). This raises questions about what factors might contribute to this variability in the school-age population.

Of course, these results pertain to speech improvements. Should these results be replicated with prospective, randomized clinical trial designs, it will be of interest subsequently to determine whether any non-speech improvements can also occur as expeditiously. There are clinical trials data, for example, that provide some indication of the clinical time required for efficacious management of speech-related social anxiety (Menzies et al., 2008). It is likely that such anxiety is present with the age group studied here. In which case, it may be that efficacious anxiety management with the present age group can be an expedient matter.

In the case of Participant 12 the treating clinician judged that progress was not satisfactory with the Lidcombe Program alone and supplemented it with traditional speech-restructuring targets. This decision was made because the treatment was not suitable for the child’s parent. The parent found it difficult to give the verbal contingences. Additionally, this parent reported only providing occasional contingencies for stuttered speech. Therefore, the clinician introduced speech-restructuring components so that Participant 12 would not need to rely completely on parental verbal contingencies for the control of his stuttering. With the aid of the speech-restructuring supplement, Participant 12 did reach Stage 2 in seven clinic visits (see Table 2). The blinded observer’s %SS score pre-treatment was 8.3 and 2.5 at follow-up. For this child, the combination of the two treatment styles did not result in a superior outcome to the other participants in the study.

One of the two children with language disorders (Participant 11) who received the Lidcombe Program needed to enter Stage 2 with more liberal criteria as judged by the treating clinician. The reason for this was because language problems were felt to be a barrier for attaining the study speech criteria. Nonetheless, this child appeared to benefit from the treatment up until the time of follow-up. However, this raises an issue for future research about the characteristics of school-age children for whom the Lidcombe Program is suitable, and the factors such as the presence or absence of language disorders that may affect treatment outcomes for children with these profiles.

One of the encouraging findings for the children in this study is the number of treatment sessions required. If 16 clinic visits is considered a reasonable benchmark for the time to attain speech criteria for admission to Stage 2 (see Section 1), the present report of half that number, with a median of eight, is encouraging. One reason it may have occurred is because of the enhanced cognitive functioning of school-age children and their increased capacity to self-direct their treatment. A meta-analysis of available data (Kingston et al., 2003) suggest that treatment may be shorter for older children in the preschool age range, and the present results may represent an extension of that effect into the school-age years. However, the present finding is based on a small cohort of children and requires replication.

The present results of a verbal response contingent treatment program for stuttering school-age children are encouraging. However, the study was preliminary and retrospective, and hence it is likely to contain positive bias associated with participant selection. Another limitation of the present report is that it compared recordings in the clinic pre-treatment with telephone recordings at follow-up. However, as noted in Section 2 (see Section 2.2), this was a less bias-prone procedure than having the children return to the clinic. Further, as is the case with the majority of clinical trials of stuttering, there was a risk of underestimating follow-up stuttering rates with audio observations. The present study used telephone post-treatment speech samples. However, at least for non-telephone audio samples, the risk of error appears minimal (Rousseau et al., 2007). The above limitations aside, the study was of an age group not renown
for natural recovery, hence the occurrence of that event with the children would not have been a likely source of bias. Overall, the findings justify the conduct of Phase II and III clinical trials to establish treatment effects of the Lidcombe Program for school-age children. It may well prove to be a valuable contribution to stuttering treatment that is greatly needed for this age group of children who stutter.

CONTINUING EDUCATION

Follow-up of 6–10-year-old stuttering children after Lidcombe Program treatment: A Phase I trial

QUESTIONS

1. According to Onslow et al. (in press), which of the following is not a clinical trial of a stuttering treatment:
   (a) A fundamental interpretable unit of treatment efficacy
   (b) At least one pre-treatment and one follow-up outcome measure
   (c) An attempt to determine outcome with retrospective data
   (d) An attempt to determine outcome with prospective data

2. What phase of clinical trials is known as the “gold-standard” evidence for efficacy?
   (a) Phase I
   (b) Phase II
   (c) Phase III
   (d) All of the above

3. The Lidcombe Program:
   (a) Is a behavioural treatment approach developed for school-age children who stutter
   (b) Is administered only by a speech-language pathologist
   (c) Involves verbal response contingent stimulation
   (d) All of the above

4. Treatments for school-age children are needed urgently
   (a) Because there are none
   (b) Because there has been only one clinical trial
   (c) Because they are known to be bullied during the school years because of stuttering
   (d) None of the above

5. This study found
   (a) Clinical trials evidence in favour of the Lidcombe Program for school-age children
   (b) Evidence to warrant clinical trials of the Lidcombe Program with that age group
   (c) No suggestion of a treatment effect at all
   (d) None of the above

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References


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