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Evidence for the treatment of co-occurring stuttering and speech sound disorder:
A clinical case series

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Abstract

Purpose: There is a paucity of evidence to guide treatment for children with co-occurring stuttering and speech sound disorder. Some guidelines suggest treating the two disorders simultaneously using indirect treatment approaches, however the research supporting these recommendations is over 20 years old. In this clinical case series, we investigate whether these co-occurring disorders could be treated concurrently using direct treatment approaches supported by up-to-date, high-level evidence, and whether this could be done in an efficacious, safe and efficient manner.

Method: Five preschool aged participants received individual concurrent, direct intervention for both stuttering and speech sound disorder. All participants used the Lidcombe Program, as manualized. Direct treatment for speech sound disorder was individualized based on analysis of each child's sound system.

Results: At 12 months post commencement of treatment, all except one participant had completed the Lidcombe Program, and were less than 1.0% syllables stuttered on samples gathered within and beyond the clinic. These four participants completed Stage 1 of the Lidcombe Program in between 14 and 22 clinic visits, consistent with current benchmark data for this Program. At the same assessment point, all five participants exhibited significant increases in percentage of consonants correct and were in alignment with age expected estimates of this measure. Further, they were treated in an average number of clinic visits that compares favorably with other research on treatment for speech sound disorder.

Conclusion: These preliminary results indicate that young children with co-occurring stuttering and speech sound disorder may be treated concurrently using direct treatment approaches. This method of service delivery may have implications for cost and time efficiency and may also address the crucial need for early intervention in both disorders.

These positive findings highlight the need for further research in the area and contribute to the limited evidence base.

Keywords: *Lidcombe Program, childhood stuttering, speech sound disorder, concurrent treatment*

Stuttering is a neurophysiological disorder that interrupts the flow of speech and causes frequent, unpredictable breakdowns in communication. The prevalence of stuttering has been estimated at around 1% (Bloodstein & Ratner, 2008), although the disorder's cumulative incidence has been reported to be as high as 8.5% during the preschool years (Reilly et al., 2009). With typical onset occurring in early childhood (Yairi & Ambrose, 1999), if left untreated, stuttering may become chronic (Bloodstein & Ratner, 2008). The negative impacts of stuttering across the lifespan are well documented. In the preschool years, stuttering may elicit negative peer responses, and may impact on children's social interactions (Langevin, Packman, & Onslow, 2009). Further, preschool children who stutter may be more prone to social withdrawal and increased frustration levels (Langevin, Packman, & Onslow, 2010). During the school years there are increased rates of teasing and bullying of children who stutter, which can have negative psychological, emotional, social, and physical effects (Blood & Blood, 2007). Retrospective reports from adults who stutter have substantiated these findings, with adults reporting difficulty establishing friendships later in life as a result of teasing and bullying during the school years (Mooney & Smith, 1995). For adolescents and adults who stutter, there is increased risk of developing social anxiety, generalised anxiety disorder, depression, and other mood disorders (Iverach et al., 2009). Stuttering also may affect educational achievement (O'Brian, Jones, Packman, Menzies, & Onslow, 2011) which may then impact on vocational opportunities (Klein & Hood, 2004).

Speech sound disorder (SSD) is an umbrella term for disorders that affect the production of speech sounds. It includes typical errors made by children learning to speak that persist past age-expected norms (American Speech-Language Hearing Association, 2014). The term SSD encompasses primary disorders of speech sounds (i.e. with an underlying phonetic or phonological deficit) and disorders caused by organic factors (e.g. cleft palate). Phonetically (articulatory) based errors involve the motoric ability required for

production of sounds, while phonological errors are rule-based (linguistic) in nature (Dodd, 2005). Similar to stuttering, the long-term negative implications of SSD are well-documented. If SSD persists into the school years, children may be at risk for poorer literacy outcomes compared to their peers (Nathan, Stackhouse, Goulandris, & Snowling, 2004). For these children, other academic areas/skills may also be affected including mathematics, comprehension, and phonological awareness (Denne, Langdown, Pring, & Roy, 2005; Gierut, 1998; Holm, Farrier, & Dodd, 2008; Leitão & Fletcher, 2004; Rvachew & Grawburg, 2006). The following variables are also reported to be associated with SSD in children: cognitive difficulties, issues with forming peer relationships, lowered self-perception and esteem, increased risk of school attrition, vocational impact, and parent/sibling report of feelings of guilt, anxiety, and/or protectiveness (McCormack, McLeod, McAllister, & Harrison, 2009). If children with SSD are not treated by school-age, some of these negative implications may persist into adulthood (Law, Boyle, Harris, Harkness, & Nye, 1998). The prevalence of speech sound disorder is reported to be high during the preschool years (Law, Boyle, Harris, Harkness, & Nye, 2000) with some estimates as high as 10-15% (Williams, McLeod, & McCauley, 2010).

In isolation, both stuttering and SSD have potential to result in negative long-term impacts on individuals. Therefore, early intervention for both disorders is considered to be crucial. Regarding stuttering, the disorder becomes less tractable as children get older (Lincoln, Onslow, Lewis, & Wilson, 1996) and it is reported that the most effective period for treatment is before the child reaches school-age i.e. around six years of age (Packman & Onslow, 2012). For children with SSD, research has established a Critical Age Hypothesis whereby children with persisting SSD past the age at which they are learn to read—around five and a half years—may have poorer literacy outcomes than their peers (Bishop & Adams,

1990; Nathan et al., 2004). Similarly then, intervention for SSD in the preschool years is essential and known to be very effective (Law, Garrett, & Nye, 2003).

Stuttering and SSD have been reported to co-occur at a rate as high as 30-40% (Nippold, 2004b), although prospective epidemiological research is needed to support these figures because current estimates of co-occurrence are based on research utilising parent and/or speech-language pathologist (SLP) report or retrospective file audit. However, the two disorders do co-occur in the clinical environment and when this occurs, clinicians have reported that this may pose a clinical challenge for them (Unicomb, Hewat, Spencer, & Harrison, 2013). One reason for this may be because that although clinicians are aware that early intervention is essential for both disorders, there is little scientific evidence available to guide treatment when they co-occur. In a commentary on the subject, Ratner (1995) provided a framework for service delivery for this caseload. Several service delivery options were discussed which broadly included treating stuttering either serially (sequentially), cyclically, or concurrently. To further define these options, a serial approach addresses one disorder to a specified level of recovery before moving on to treat the other disorder. A cyclic approach involves treating one disorder for a period of time before moving on to address the other disorder for an equal amount of time, continuing cyclically. Concurrent treatment approaches involve treating both disorders at the same time, however there are two variations within a concurrent approach. These variations are a) treatment goals are delivered simultaneously during the same activities; or b) treatment goals are delivered discretely, that is, in the same session with different activities. For the purposes of this paper, the definition of a ‘simultaneous’ approach will draw upon the first variation, while the definition of a ‘concurrent’ approach will draw on the latter variation.

To date, just one study has been published that documents a treatment approach for co-occurring stuttering and SSD, and it was published over 20 years ago. Conture, Louko,

and Edwards (1993) investigated the efficacy of providing simultaneous treatment to children with co-occurring stuttering and SSD, using predominantly indirect treatment approaches. While direct treatment approaches aim to directly modify speech output, indirect approaches aim to adjust the person's environment and/or provide models to facilitate speech output. Conture et al. conducted a pilot study for which they recruited eight participants, who were divided into two groups. Group one were four children (mean age 69.7 months) who stuttered and had a dual diagnosis of SSD (phonologically based). Group two were four children who stuttered only (mean age 71.5 months). For both groups, treatment occurred once weekly for 45 minutes over the course of a university calendar year, during semester-time only. The sessions were run in a group format. Across both groups, stuttering treatment involved the use of procedures such as modelling speech at a slower rate, without physical tension, and enforcing conversational rules e.g. turn-taking. The children in the stuttering plus SSD group also received treatment for their speech sounds that used procedures such as auditory bombardment, modelling, and reinforcement of correct target production. For this group, the stuttering and SSD goals were delivered simultaneously and blended into the same activities. Parent-training and education also took place alongside the children's therapy sessions. Results indicated that three of the children in the stuttering only group showed an overall reduction in stuttering frequency (range 15%-55%). One child in this group had a 42% mean increase in stuttering frequency. Only two of the children in the stuttering plus SSD group had an overall reduction in stuttering frequency (range 27%-37%), while the other two children in this group had an overall increase in stuttering frequency (range 2%-17%). The children with SSD had an average decrease of 25% in targeted phonological processes across the group, however three of the four children in this group had an increased usage of some targeted processes, including interdental and lateral productions. One participant exhibited no change to any phonological process. All pre-treatment samples (for stuttering and SSD) were

gathered solely from conversations conducted within the clinic. Determination of SSD was based on the use of age-inappropriate and/or disordered (atypical) phonological processes uttered during conversation. Whilst the results were promising, the research was not without methodological limitations. In relation to sampling procedures, without information from beyond-clinic data and an extensive single-word naming speech sound assessment alongside observations in connected speech, full representation of the child's sound system is not realized. Due to the lack of controls employed in the study design, it is also not possible to state how much of the treatment effect was attributable to the various therapeutic procedures employed.

Despite the paucity of empirical research on intervention for children with these co-occurring disorders, several guidelines have been published that suggest approaches to service delivery which are based on experts' opinions. Some guidelines have suggested treating stuttering and SSD simultaneously using indirect treatment approaches, no doubt drawing Conture et al.'s (1993) research (Byrd, Wolk, & Lockett Davis, 2007; Ratner, 1995; Wall & Myers, 1995; Wolk, 1998). Others have suggested that consideration be given to the evidence base for treatments, and recommended, more direct procedures (Nippold, 2004a). Nippold argued that there was no reason to avoid using direct treatment approaches with this co-occurring caseload given the lack of evidence to support an interaction between stuttering and phonology. Guitar (2006) also recommended using direct treatment approaches for both disorders, and suggested a serial form of service delivery. However, Ratner (1995) argued that treating serially could compromise the optimal window period for early intervention that is considered crucial for each disorder. Further, treating serially may impose greater financial burden and/or result in therapeutic burnout for both service providers and families/clients alike (Unicomb et al., 2013).

Since the findings of Conture et al. (1993) were published, treatment approaches for both stuttering and SSD have evolved, and advanced. For both disorders in early childhood, there are high levels of evidence for direct treatment approaches. There are a number of treatment options for preschool-age children who stutter that are supported by research evidence (De Sonnevile-Koedoot, Stolk, Rietveld, & Franken, 2015; Millard, Edwards, & Cook, 2009; Trajkovski et al., 2011). However, the Lidcombe Program of early stuttering intervention (Onslow & O'Brian, 2013) is an approach supported by the highest level of evidence (Arnott et al., 2014; Jones et al., 2005; O'Brian et al., 2013). Similarly for SSD, examples of direct treatment that have high-level evidence include minimal pairs (Dodd et al., 2008), multiple oppositions (Williams, 2006), and traditional articulation therapy (Pamplona, Ysunza, & Espinosa, 1999).

Unicomb et al. (2013) reported that clinicians are implementing these direct approaches to children with co-occurring stuttering and SSD, however they reported that selecting an appropriate method for service delivery for this caseload caused a clinical dilemma. Clinicians were uncertain about whether to treat the disorders serially or concurrently, and did not know whether directly treating one disorder would affect the other. This uncertainty was a consequence of the paucity of scientific evidence in the area, as well as anecdotal reports that addressing just one disorder may potentially exacerbate the other, or that addressing both disorders concurrently may be detrimental to either disorder (Unicomb et al., 2013; Wall & Myers, 1995).

When treating this caseload, only one method of service delivery has been investigated scientifically, that is, simultaneous treatment using indirect therapy approaches (Conture et al., 1993). Other models of service delivery now need to be explored given that research for both disorders in isolation has evolved in the past 20 years. Further, serial

treatment may jeopardise the ideal window for early intervention and be a less efficient way to treat this caseload. Therefore, the aim of this research was to:

1. Trial a new method of service delivery for co-occurring stuttering and SSD, that of concurrent treatment (with discrete goals) using direct therapy approaches that are supported by up-to-date empirical evidence; and
2. Investigate the efficacy, safety, and efficiency of this method of service delivery.

Method

This research was approved by the University of Newcastle's Human Research Ethics Committee in June, 2012 (reference H-2011-0383).

Participants

Participants were five boys with ages ranging from 3;6 and 4;7 years at the start of treatment, and their primary caregivers (Table I). All participants were recruited from the waiting list from the Stuttering Clinic, University of Newcastle, Australia. Potential participants were informed of the research if they were diagnosed with stuttering and flagged by the assessing SLP as potentially having difficulties with speech sound production. To be eligible for enrolment into the current research program, participants were diagnosed as stuttering by consensus between the assessing clinician and research clinician, had stuttering levels greater than 2% syllables stuttered (%SS) based on samples gathered within and beyond the clinic (Table I), and were reported to be stuttering for a minimum of 12 months prior to entry into the study.

<Insert Table I>

Participants were diagnosed with SSD or delay if they presented with two or more age-inappropriate (developmental) speech sound errors, or at least one atypical (non-

developmental) error, based on samples of single-word productions (Diagnostic Evaluation of Articulation and Phonology (DEAP) (Dodd, Crosbie, Zhu, Holm, & Ozanne, 2003)), and connected speech (Table II). Additionally, manual independent and relational analyses of speech production were conducted to gain a thorough understanding of each participant's sound system.

<Insert Table II>

All participants and primary caregivers spoke English as their first language. All had hearing levels and oral motor structures and function that were within normal limits (WNL). All participants underwent a receptive and expressive language screening assessment with the Preschool Language Scale Fourth Edition (PLS-4) Screening Test (Zimmerman, 2005). All but one participant passed this screening assessment. The participant who did not pass the screener was subsequently administered a full standardised language assessment, the Clinical Evaluation of Language Fundamentals: Preschool Edition-2 (CELF-P2) (Wiig, Secord, & Semel, 2006). The results of that assessment indicated that this child had receptive and expressive language abilities that were within normal limits. No participant had received treatment for either disorder prior to recruitment into the study. Two participants had a family history of stuttering (participants 3 and 5). Two participants also had a positive family history of SSD (participants 2 and 3).

Research Design

Study design was a clinical case series. Case series are non-experimental research designs whereby a group of participants undergo treatment with outcomes measured at pre- and post-treatment. Case series are often used in speech-language pathology to report on, and give detailed description regarding, novel treatment approaches. The information gained from clinical case series are useful to generate hypotheses that may lead to larger-scale

experimental research (Chan & Bhandari, 2011). This study was also considered a Phase I clinical trial using the protocol designed by Onslow, Jones, O'Brian, Menzies, and Packman (2008) for evaluating stuttering research. Using a broader framework employed by the American Speech-Language Hearing Association and adapted from the Scottish Intercollegiate Guidelines (American Speech-Language Hearing Association (ASHA), 2004), the study was classed as level III (case studies).

Treatment

The treatment of stuttering and SSD was delivered concurrently to each participant. Direct therapy approaches were used for both disorders, and session goals were delivered discretely. That is, both disorders were treated within the same session, using different activities for different treatment goals. Treatment was delivered over a period of 9 consecutive months at a university clinic. Treatment and assessment were conducted by an experienced SLP, trained in the use of the Lidcombe Program, who had extensive experience providing treatment to children with stuttering and SSD.¹

Stuttering treatment. All participants completed the Lidcombe Program (LP) of early stuttering intervention as manualized (Packman et al., 2011)². The LP is a direct, behavioral treatment, and is delivered to parents who conduct daily therapy at home with their child in their natural environment. A key component of the treatment is the parent's delivery of verbal contingencies to their child during structured practice sessions and, subsequently, in natural conversations. Conducted in two stages, the aim of Stage 1 is to reduce the child's stuttering to very low levels, and the aim of Stage 2 is to maintain this reduction for a clinically significant period. Entry to Stage 2 is performance-contingent, and based on measures of

¹ For more detailed information regarding the treatment procedures for each participant in this research study, please use the following link to download a copy of the first author's PhD thesis:
<http://hdl.handle.net/1959.13/1296498>

² This study was conducted using an earlier (2011) version of the LP training manual. The current version (Packman et al., 2016) no longer includes %SS as a compulsory weekly/clinical measure.

stuttering within and beyond the clinic over three consecutive weeks. More detail on the implementation of the LP as well as specific criteria for entry into Stage 2 can be found in the 2016 treatment manual {Packman et al., 2016).

Speech sound disorder treatment. All participants received individualized direct treatment for their SSD, with goals based on the results of their independent and relational analyses. Either traditional articulation therapy and/or minimal pairs therapy was used with all participants, with treatment selection based on aforementioned goals. Participants who displayed functional speech errors (e.g. articulatory based errors such as a lateral lisp) were treated using traditional articulation therapy following procedures set out in Bernthal, Bankson, and Flipsen Jr. (2013). Children who presented with phonologically based speech errors were treated using minimal pairs therapy as outlined by Baker (2010).

This research employed a most-knowledge approach to target selection of speech sounds for all participants. There is some evidence that the most-knowledge approach to target selection (i.e. selecting stimutable, earlier developing targets) may be more appropriate for this co-occurring caseload (Sasisekaran, 2014; Wall & Myers, 1995). Therefore targets were developmental in nature (working from earlier to later acquired), were stimutable, and used minimally contrasting word-pairs. When atypical speech errors were observed (e.g. lateral lisp), these were also a target for treatment. When choosing SSD targets, this study used normative data described by Grunwell (1987; 1997). Although there is evidence supporting the selection of more complex sounds to facilitate change in SSD, there has been no research that has studied the impact of doing so when a child has co-occurring stuttering and SSD. Indeed, the selection of more complex sound targets may increase linguistic demand on a child. One well-known theory of the cause of stuttering implicates the potential unhelpful effect of increasing linguistic demand beyond a child's capacities (Starkweather & Gottwald, 1990). Therefore caution was applied when selecting sound targets for children

with co-occurring disorders. In addition, one child (participant 1) also received Stimulability Intervention (Miccio & Williams, 2010) as an adjunct to his direct treatment because several sounds were missing from his inventory.

Each child and their parent/carer attended weekly clinic visits. Each clinic visit was separated into two parts. In the first half of the session, the clinician delivered the LP; in the second half, the clinician delivered treatment for SSD.

Parents were required to complete daily home practice for both stuttering and SSD. They were advised that home practice should be delivered separately, that is, home practice sessions for the LP were conducted each morning and home practice for SSD therapy was conducted in the afternoon, where possible. Home practice for the LP was as manualized (Packman et al., 2011), whereby treatment is initially delivered for 10 to 15 minutes in structured conversations, with the gradual introduction of additional parental feedback at other times throughout the day in unstructured conversations. Home practice for the SSD was individualized depending on the nature of participants' SSD and treatment approach/es, and recommended to be around 10 minutes duration.

Research protocol. While the treatment for stuttering and SSD was delivered concurrently to participants during their clinic visits, the stuttering component mostly involved parent training on how to deliver the LP to their child in their natural environment. For this reason, the research protocol involved stuttering treatment only for the first two to three weeks, to ensure the parent was conducting this treatment in a safe, effective manner. Concurrent treatment for SSD was introduced into clinic visits from session two or three and continued in all subsequent sessions.

For ethical reasons, a stopping criterion was used. If a participant's treatment progress for stuttering was slower than expected and they had not reached Stage 2 of the LP after 22

clinic visits, then service delivery was altered to the administration of the LP only and SSD intervention was discontinued. This decision was made in collaboration with the caregiver. This criterion is consistent with worldwide benchmarks for the LP which have established that 90% of children reach Stage 2 by between 11 and 22 clinic visits (Packman et al., 2015).

If participants reached discharge criteria for SSD during the research protocol, they would continue to receive LP treatment in isolation for their stuttering. If participants reached Stage 2 of the LP—therefore no longer required weekly clinic visits for stuttering—but still required treatment for SSD, then weekly SSD treatment continued to be delivered, with the stuttering therapy occurring concurrently with this but only in line with the prescribed scheduling of clinic visits for Stage 2 of the LP.

Outcome Measures

Data were collected on four assessment occasions: (i) pre-treatment, (ii) at entry into Stage 2 of the LP, (iii) 9 months after treatment commencement, and (iv) 12 months after treatment commencement. Additionally, stuttering progress measures were collected on a weekly basis to guide treatment goals and decision-making.

Within-clinic speech samples were gathered at each of the four assessment occasions for both stuttering and SSD (connected speech) measures. Samples were no less than 300 syllables in length and were generated during imaginative play-based conversations; the same stimuli were used on all four occasions. Samples were recorded with a Tascam DR2D digital handheld recorder, and Sennheiser EW112 lapel wireless microphone attached to participants' collars to ensure quality recordings and enable discrimination of finer phonetic detail where possible. Recordings were in 16bit WAV format in stereo at a sampling frequency of 44.1KHz (MacWhinney, 2000). Beyond-clinic speech samples were obtained by the participants' parents at each assessment occasion using a Sony ICD-UX512F hand-

held digital voice recorder. Parents were requested to provide two 10-minute samples, one parent-child interaction in the natural home environment, and one with the child and a different familiar adult outside of the home environment.

Primary outcome measures. To determine the efficacy of direct, concurrent treatment of stuttering and SSD, the following primary outcome measures were used:

Stuttering. At each of the four assessment occasions, %SS were measurements were made of samples gathered both within and beyond the clinic (as previously described). Percentage of syllables stuttered were measured on a standard two-button counting device. All within and beyond-clinic measures were generated by the first author. All of the within- and beyond-clinic speech recordings were randomly arranged and presented to an observer (an experienced SLP) who was unaware of the nature of the study, in order to establish interjudge agreement. A secondary outcome measure (collected for the purposes of clinical treatment data) was a 10-point severity rating (SR) scale (Packman et al., 2011) (where 1 = no stuttering, and 10 = extremely severe stuttering)³. Correlation between the two observers was very high ($r=0.95$) for %SS, and good ($r=0.88$) for SR. To establish intrajudge agreement, 10% of the samples were re-rated by the first author six months after completion of the research protocol and were considered high for both measures ($r=0.99$ for %SS, $r=0.96$ for SR).

Speech sound disorder. At each of the four assessment occasions, the percentage of consonants correct (PCC) metric was utilized (Shriberg & Austin, 1997). This was measured on connected speech and single-word naming test samples as previously described. This process was aided by use of the Profile in Phonology (PROPH) component of the Computerized Profiling software (Long, Fey, & Channell, 2006), which provided

³ Note that in the revised version of this treatment manual, the SR scale has changed to a 9-point scale (where 0 = no stuttering and 9 = extremely severe stuttering) (Packman et al., 2016).

comprehensive independent and relational analysis of each child's sound system. This was supplemented with a manual analysis of the data (Baker, 2004).

Interjudge agreement was established using procedures similar to those described in Dodd et al. (2008). The same independent observer mentioned earlier was presented with 10% of the connected speech and single-word speech samples to establish point-to-point agreement of the first 100 intelligible words in each sample after 2.5 minutes of audio had elapsed. These words were transcribed phonemically. For the purposes of research, point-to-point agreement of PCC using phonemic transcription has been established as reliably adequate (Shriberg & Austin, 1997; Shriberg & Lof, 1991). For the connected speech and single word naming test samples, interjudge agreement was considered high at 94%, and 96% respectively. To establish intrajudge agreement, 10% of the speech samples were re-transcribed six months after completion of the research protocol. Point-to-point calculations of these data established high intrajudge reliability at 98% across both sampling contexts.

Data Analysis

Analysis of the primary outcome measures (%SS, PCC) were conducted using 'reliable change with 95% credible intervals' (Unicomb, Colyvas, Harrison, & Hewat, 2015). Statistical analysis of single case studies has been criticized as less rigorous and less statistically robust than that applied to experimental designs. Jacobson and Truax (1991) proposed the reliable change index to measure change within an individual, a method that is widely used in the field of clinical psychology. Reliable change index indicates whether changes observed in the individual are both statistically and reliably improved due to the condition or treatment being administered. However, the reliable change index does have certain limitations. For example, it assumes the measures used have a normal distribution or spread. It assumes knowledge of normative data for the population under study, and relies on

the use of continuous data. The primary outcome data used in this study were categorical in nature, with binary outcomes, therefore a modification to the original method proposed by Jacobson and Truax was designed. This modification was used to analyze the data in this study, and is presented in detail in Unicombe et al., (2015). However, to summarize, the reliable change index with 95% credible intervals determines whether changes observed in an individual's pre-treatment and post treatment data are statistically significant. This approach involves estimating the difference between two proportions (i.e. pre- and post-treatment data), and the 95% credible interval for that difference. This was calculated using a Bayesian approach to determine the credible interval (also known as a Bayesian confidence interval). As with confidence intervals, credible intervals provide a range in which this true but unknown difference is expected to be found. Results are typically visualised graphically, where any significant changes observed are significant to the level of .05.

Treatment efficiency. In order to assess efficiency of treatment, measures of the timing and number of clinic visits were collected and recorded in file notes. For stuttering, number of clinic visits and number of weeks to complete Stage 1 of the LP were recorded. For SSD, number of clinic visits to achieve observable outcomes were recorded. Further, average session length (in minutes) was recorded for each component of the session (i.e. stuttering and SSD) and these are presented in Table III.

<Insert Table III>

Results

Stuttering

One participant (participant 5) did not complete Stage 1 of the LP while enrolled in the study. In collaboration with this participant's caregiver, a decision was made during clinic visit 22 to cease treatment for SSD, and continue the LP only (as described above).

Analysis of the primary outcome measure for stuttering, %SS, on samples collected within-clinic revealed a significant improvement ($p < .05$) in stuttering levels for four participants (Table IV). Participant 5 exhibited increased stuttering levels from pre-treatment to 12 months post-treatment, however this increase was not found to be significant based on within clinic data (Figure I).

Analysis of the primary outcome measure for stuttering, %SS, on samples conducted beyond-clinic revealed a significant improvement ($p < .05$) in stuttering levels for all except one participant (Table IV). Participant 5 exhibited significantly ($p < .05$) increased stuttering levels from pre-treatment to 12 months post-treatment (Figure I).

<Insert Table IV>

<Insert Figure I>

Speech Sound Disorder

One participant (participant 2) reached discharge criteria for SSD early in Stage 2 of the LP. From that point forward, this participant continued Stage 2 LP only (as described above). For the remaining three participants still receiving treatment for SSD, weekly treatment for SSD continued throughout both Stages of the LP. Upon completion of the research protocol, if participants still required treatment for SSD, relevant referrals were made to ensure continuity of service. These referrals were made for four of the five participants in this study.

Analysis of primary outcome data for SSD, PCC, obtained from single-word naming test samples revealed that in all cases there was a statistically significant improvement ($p < .05$) in this outcome measure from pre-treatment to 12 months post-treatment (see Figure 3). Further analysis of this measure revealed that for one participant (4), significant improvement in this outcome measure did not occur between assessment occasions 1 and 2, with the most improvement being observed once Stage 1 was completed (Table V).

Analysis of the primary outcome data for SSD, PCC, based on samples derived from connected speech again revealed that in all cases there was a statistically significant improvement ($p < .05$) in this outcome measure from pre-treatment to 12 months post-treatment (see Figure II). As described above, significant improvement in this outcome measure for participant 4 occurred once Stage 1 was completed (Table V).

<Insert Table V>

s<Insert Figure II>

Treatment Efficiency

Stuttering. As previously mentioned, participant 5 did not complete Stage 1 of the Lidcombe Program. At 12 months post commencement of therapy, participant 5 had completed 40 Stage 1 clinic visits. He continued treatment in the University of Newcastle's Stuttering Clinic at completion of the research protocol (i.e. at 9 months), however his therapy was suspended temporarily in order to avoid burn-out.

The remaining four participants completed Stage 1 of the LP in an average of 17 clinic visits (Table III). Taking into account sessions that were missed for personal reasons (e.g. illness, holiday periods etc.), these four participants completed Stage 1 in an average of

20.3 weeks. Participant 4 appeared to take longer to complete Stage 1 of the LP than the others, however this was not analysed for significance.

Of the four participants who completed Stage 1 of the LP, all continued Stage 2 at the University of Newcastle's Stuttering Clinic after their 9 month assessment occasion.

Participants 1, 3 and 4 completed all Stage 2 scheduled visits and were subsequently discharged from this service. Participant 2 had completed all Stage 2 clinic visits with the exception of his final scheduled appointment; clinic notes revealed he failed to attend the final maintenance session before being discharged from this service.

Average session length for Stage 1 clinic visits (stuttering component only) was 50.1 minutes (range 30-78 minutes). Average session length (stuttering component only) for Stage 2 appointments was 35.7 minutes (range 20-51 minutes).

Speech sound disorder. The four participants were treated in an average of 22.3 clinic visits for their SSD, after which they were all in alignment with age-expected estimates of PCC (Table V). One participant (2) met criterion for discharge for his SSD after 20 clinic visits. All four of these participants had a reduction in the numbers of speech errors from pre-treatment to post treatment (Table II). Participant 5, whose treatment for SSD was abandoned under the research protocol also exhibited a reduction in the number of speech errors used from pre-treatment to post treatment. The residual errors displayed across all five participants were considered age appropriate or delayed, with no atypical errors being used.

Average duration of the SSD component of each session was 29.7 minutes (range 20-48 minutes).

Discussion

This clinical case series has established that for some children with co-occurring stuttering and SSD, direct and concurrent treatment of the two disorders may be an effective, safe and efficient means of service delivery.

Analysis of the primary outcome measure, %SS, for stuttering revealed that four of the five participants showed statistically significant improvements in their fluency levels from pre-treatment to post-treatment. These four participants were measured at below 1%SS both within and beyond the clinic, which is consistent with other Lidcombe Program research outcomes (Jones et al., 2008). As previously mentioned, one participant (5) increased stuttering from pre-treatment to post-treatment. This participant exhibited challenging behaviours during the course of the study, preventing strict adherence to the treatment manual for the LP beyond the clinic. Although referrals for further investigation in relation to these behaviours were put in place during the course of the study, this was not followed up by the caregiver at that time and no formal diagnosis was made in relation to these behaviours. However, at the 12 month assessment occasion it was reported that upon commencing Kindergarten, this participant was subsequently referred for cognitive and psychological evaluation after consultation with school educators. Although completion of LP Stage 1 data has not been formally investigated, previous studies have also reported episodes of non-completion. For example, a North American file audit of 165 children under 6 years of age and who had LP treatment reported that 13.5% of children did not complete Stage 1 for various reasons (Koushik, Hewat, Shenker, Jones, & Onslow, 2011).

Analysis of the primary outcome for SSD, PCC, revealed that all five participants showed statistically significant improvement from pre-treatment to 12-months post-commencement of treatment. This occurred across both sampling contexts, single-word naming test and connected speech.

Participant 4 differed from the others in that he did not follow the same trajectory of improvement and this may be due to the nature of his SSD. Although this participant exhibited phonological difficulties, the error having the most impact on this child's intelligibility was mostly motoric (e.g. lateral lisp) in nature. The majority of time in his SSD treatment targeted these motoric errors using a motor-based treatment approach. It is possible that the competing demands of working on two separate speech disorders, each with underlying speech motor involvement, limited his capability for concurrent improvement in both. It is also possible that this child took longer to respond to treatment for SSD in general, as previous research (Baker & McLeod, 2011) has established that traditional articulation therapy may take longer to complete than phonologically based interventions. Despite this participant's limited improvement in SSD treatment process measures prior to entry into Stage 2 of the LP, it is worth noting that neither disorder was exacerbated by the direct concurrent treatment of the other, and that the overall post-treatment result was a significant improvement in both disorders for this child.

Apart from participant 4, for the other participants, whose underlying SSDs were primarily phonological in nature, the two disorders were successfully treated directly and concurrently. That is, the two disorders were treated at the same time within the same sessions yet discretely, using treatment approaches that are direct in nature. Further, the results of these participants suggest that greater gains may be related to SSD treatment that is linguistically based (e.g. minimal pairs) where the aim of therapy is to highlight homonymy and to teach the child the function of sounds (Barlow & Gierut, 2002).

Although there are some recommendations in the literature regarding treatment of co-occurring stuttering and SSD, many of them recommend the use of indirect treatment approaches to address stuttering, and/or the SSD (Byrd et al., 2007; Ratner, 1995; Wall & Myers, 1995; Wolk, 1998). Others propose that there is no need to avoid direct treatment for

this caseload (Guitar, 2006; Nippold, 2004a) particularly as there is insufficient empirical evidence to support an interaction between the two disorders. This research contributes to the recommendations of the latter authors who propose that both disorders may be treated concurrently using direct treatment approaches.

In terms of treatment safety, the results of this clinical trial revealed no evidence that treating both disorders at the same time using direct treatment approaches was detrimental to either disorder. In other words, treating the stuttering did not appear to exacerbate the SSD, and vice versa, for any participant. One participant (5), did increase stuttering from pre-treatment to 12 months post-treatment, however this did not occur until well after the treatment for the SSD had been withdrawn in accordance with the research protocol. Alongside this, participant 5 also continued to show improvements in primary outcome measures for SSD, PCC, at each assessment occasion.

Another consideration for this study was to assess whether direct and concurrent treatment of stuttering and SSD could be completed in a timely and cost efficient manner, particularly compared with a serial form of service delivery for these disorders. This is an important consideration given the universal advice in favor of early intervention for both disorders in isolation. Stuttering treatment data showed that the four participants who successfully completed Stage 1 of the LP did so in an average of 17 clinic visits. This is in line with established worldwide benchmarks. Research about LP treatment (in isolation) has indicated that the median time to reach Stage 2 is around 16 clinic visits (Rousseau, Packman, Onslow, Harrison, & Jones, 2007), and established worldwide benchmarks show that the majority of children complete Stage 1 in between 11 and 22 clinic visits (Packman et al., 2015). Further, completion of Stage 2 of the LP is expected to take an additional 11 months (Packman et al., 2015). Although participant 4 completed Stage 1 of the LP, this took 22 clinic visits, which aligns with the higher end of the benchmark median. It is also

noteworthy that this participant demonstrated more severe pre-treatment stuttering compared to the others. Previous research (Jones, Onslow, Harrison, & Packman, 2000) suggested a correlation between higher severity levels of stuttering and longer time taken to complete LP Stage 1. However, due to the nature of the research design used in this study, it is not known whether either stuttering severity or the nature of this participant's speech errors were factors that may have caused longer treatment time, however this is a consideration that warrants further investigation.

These same four participants who completed Stage 1 of the LP were also treated in an average of 22.3 clinic visits for their SSD, after which they were all in alignment with age-expected estimates of PCC, approximately 88.5% for children aged 4 to 5 years, and around 93.4% for children aged 5 to 6 years (McLeod & Bleile, 2003). One participant reached discharge criteria following only 20 clinic visits for SSD. In isolation, duration of treatment time for SSD is variable depending on the intervention approach being used. Traditional articulation therapy has been reported to take upwards of 40 months until discharge criteria are reached (Baker & McLeod, 2011). In contrast, interventions that are phonological in nature (e.g. minimal pairs) may require a minimum of 30 sessions to be effective (Williams, 2012).

In Stage 1 of the Lidcombe program the typical duration of clinic visits is between 45 and 60 minutes (Packman et al., 2016). In Stage 2 of the Program, clinic visit duration is typically shorter in length, usually around 30 minutes. In the current study, session duration for both Stages 1 and 2 was approximately 50 and 35 minutes respectively for the stuttering component only. Adding in session duration for the SSD component, approximately 30 minutes, average overall session duration during Stage 1 was approximately 80 minutes in length to treat both disorders. Weighing this information up against potential overall time taken in treatment using if treating both disorders serially (i.e. session duration + total

intervention duration), concurrent delivery may be a more efficient method of service delivery, although more research is required to confirm this. Efficiency is one variable that service providers could consider when considering concurrent versus serial treatment of these disorders, however financial implications on all parties, as well as minimizing the risk of therapy burnout for families are other factors that should also be considered

Although the findings from this clinical trial are largely positive, they are not without limitations. The research design employed for this study was a clinical case series (Chan & Bhandari, 2011) which was considered appropriate for this preliminary research where the safety of participants was paramount. This was important because there is a limited treatment evidence available for this caseload, and there are anecdotal reports that direct, concurrent treatment has the potential to exacerbate one or both disorders (Unicomb et al., 2013; Wall & Myers, 1995). This research design allowed for careful and thorough report on each participant individually, and involved no manipulation of variables. Therefore, an obvious limitation to this study is there is no inference of cause and effect because the design is not experimental in nature. The use of a true experimental design is, however, indicated and would allow for stronger conclusions to be drawn about the efficacy of the treatment delivery used in this study. The small participant numbers also mean that generalization of results should be regarded with caution. Also, due to the heterogeneous nature of both stuttering and SSD in each individual, the results of this study may not easily be replicable. However, the detailed description of treatment and statistical analysis may assist with replicability. Preliminary indications arising from this study are that concurrent treatment of stuttering and SSD may be feasible and effective, although further experimental studies involving larger numbers of participants are needed to substantiate these outcomes.

Of note is that the participants in this study all had mild or mild-moderate level of severity of SSD at entry into the study. Further research could also investigate whether similar outcomes could be achieved by children with a more severe disorders.

Conclusion

The aim of this study was to add to the evidence base for treatment of co-occurring stuttering and SSD, exploring a new method of service delivery involving concurrent treatment (discrete goals) using direct treatment approaches that are supported by scientific evidence for both disorders, and whether this could be done in an effective, safe, and efficient manner. The findings have revealed that treating in this manner may be safe and efficacious for some children with co-occurring stuttering and SSD, particularly those with phonologically based errors. Furthermore, treating these children directly and concurrently may have implications for cost and time efficiency compared to treating the disorders serially, particularly if the underlying nature of a child's SSD is mild or mild-moderate in severity, and phonological in nature.

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Table I
Participant age and pre-treatment information

Participant	Age	Within-clinic	Beyond- clinic	PCC Single-word	PCC Connected
		% SS(SR)	%SS/SR	(SEV)	speech (SEV)
1	4;1	2.2% (3)	2.5% (2,3)	75.5% (mild-mod)	84.8% (mild-mod)
2	3;6	3.9% (3)	4.3% (6,3)	69.4% (mild-mod)	81.5% (mild-mod)
3	4;4	3.5% (4)	3.7% (4,4)	76.9% (mild-mod)	78% (mild-mod)
4	3;7	14.6% (7)	9.3% (7,6)	72.3% (mild-mod)	85% (mild-mod)
5	4;7	4.2% (4)	3.0% (6,4)	77.8% (mild-mod)	86.7% (mild)

*%SS - percentage of syllable stuttered; SR - severity ratings; PCC - percentage of consonants correct

**Beyond clinic %SS based on average of two speech samples

***SEV = severity of SSD based on Shriberg and Austin (1997)

Table II

Summary of speech sound errors by participant at pre- and 12 months post-treatment⁴

Participant	Age appropriate speech sound errors	Non-age appropriate + atypical speech sound errors
	Pre-treatment	
1	Gliding, labiodental realisation of /θ/ and/or /ð/, gliding of fricatives, interdental realisation of /s/ and /z/	Velar assimilation, cluster reduction, backing, final consonant deletion, velar fronting, late stopping, glottal substitution, weak syllable deletion, early stopping
2	Cluster reduction, gliding, labiodental realisation of /θ/ and/or /ð/, late stopping, weak syllable deletion, palatal fronting	Final consonant deletion, early stopping, context sensitive voicing
3	Gliding, labiodental realisation of /θ/ and/or /ð/	Cluster reduction, late stopping, weak syllable deletion, palatal fronting, velar fronting, palatisation, alveolarisation, lateral production of /s/
4	Cluster reduction, gliding, labiodental realisation of /θ/ and/or /ð/, late stopping, weak syllable deletion, interdental production of /s/	Velar fronting, early stopping, lateral production of /s, z, ʃ/
5	Gliding, labiodental realisation of /θ/ and/or /ð/	Cluster reduction, early stopping, weak syllable deletion, deaffrication, interdental production of /s/
12 months post-treatment		
1	Labiodental realisation of /θ/ and/or /ð/	Cluster reduction, gliding
2	Gliding, labiodental realisation of /θ/ and/or /ð/, f, late stopping	-
3	Labiodental realisation of /θ/ and/or /ð/	Late stopping, palatal fronting
4	Gliding, late stopping	Interdental production of /s, z/, cluster reduction, palatal fronting
5	Labiodental realisation of /θ/ and/or /ð/	Gliding

⁴ These errors were interpreted as being age appropriate, delayed, or atypical in line with available normative information (Grunwell, 1987, 1997).

Table III

Time taken in treatment for stuttering and SSD by participant

Participant	No. Sessions		
	to complete Stage 1 LP	No. Weeks to complete Stage 1 LP	No. sessions SSD treatment
1	14	17	25
2	15	18	20
3	17	19	20
4	22	27	24
5	n/a ⁵	n/a	17

⁵ Note that data is missing for participant 5, as they did not complete Stage 1 of the LP while enrolled in this research study

Table IV

Primary outcome data within-clinic (WC) and beyond-clinic (BC) for stuttering for participants at each assessment occasions (AO)

Participant	Pre-treatment (A01)		Entry Stage 2 LP (A02)		9-months post (A03)		12-months post (A04)	
	WC	BC	WC	BC	WC	BC	WC	BC
	%SS(SR)	%SS(SR)	%SS(SR)	%SS(SR)	%SS(SR)	%SS(SR)	%SS(SR)	%SS(SR)
1	2.2 (3)	2.5 (2,3)	0.9 (2)	0.7 (2,2)	0.4 (2)	0.3 (1,1)	0.4 (2)	0.5 (2,1)
2	3.9 (3)	4.3 (6, 3)	1.0 (2)	0.9 (1,2)	0.0 (1)	0.3 (2,1)	0.0 (1)	0.1 (2,1)
3	3.5 (4)	3.7 (4, 4)	0.0 (1)	0.2 (1,2)	0.2 (1)	0.1 (2,1)	0.4(1)	0.0 (1,1)
4	14.6 (7)	9.3 (7,6)	0.3 (1)	0.5 (2,2)	0.0 (1)	0.1 (1,2)	0.2 (2)	0.1 (1,1)
5	4.2 (4)	3.0 (6, 4)	4.2 (5)	2.0 (4,2)	2.8 (4)	2.0 (3,-)	6.4 (5)	5.9 (6,5)

**%SS - percentage of syllable stuttered; SR - severity ratings*

***Beyond clinic %SS based on average of two speech samples*

Table V

Primary outcome data single-word (SW) and connected speech (CS) for SSD across participants, assessment occasions (AO)

	Pre-treatment (A01)		Entry Stage 2 LP (A02)		9-months post (A03)		12-months post (A04)	
Participant	PCC SW(SEV)	PCC CS(SEV)	PCC SW(SEV)	PCC CS(SEV)	PCC SW(SEV)	PCC CS(SEV)	PCC SW(SEV)	PCC CS(SEV)
1	76% (mild-mod)	85% (mild-mod)	88% (mild)	90% (mild)	94% (mild)	98% (mild)	94% (mild)	97%(mild)
2	69% (mild-mod)	82% (mild-mod)	82% (mild-mod)	88% (mild)	87% (mild)	95% (mild)	91% (mild)	96%(mild)
3	77%(mild-mod)	78%(mild-mod)	79% (mild-mod)	89% (mild)	93% (mild)	96% (mild)	94% (mild)	98.9% (mild)
4	72%(mild-mod)	85%(mild-mod)	75% (mild-mod)	86% (mild)	86% (mild)	85% (mild)	91% (mild)	93.2% (mild)
5	78%(mild-mod)	87%(mild)	83% (mild-mod)	94% (mild)	91% (mild)	95% (mild)	97% (mild)	97% (mild)

**PCC = percentage of consonants correct*

***SEV = severity of SSD based on Shriberg and Austin (1997)*

Participant	Pre	Post	Diff	lcl	ucl	Lower bar	Upper bar		Pre	Post	Diff	lcl	ucl	Lower bar	Upper bar
1	2.244	0.42	-1.82	-3.68	-0.32	1.86	1.51		2.46	0.50	-1.95	-3.48	-0.77	1.52	1.18
2	3.86	0	-3.86	-5.66	-2.27	1.80	1.59		4.34	0.08	-4.26	-5.25	-3.31	0.99	0.95
3	3.54	0.40	-3.14	-5.16	-1.41	2.02	1.73		3.70	0.058497	-3.65	-4.44	-2.91	0.79	0.74
4	14.58	0.22	-14.36	-17.44	-11.35	3.08	3.02		9.31	0.11	-9.20	-10.44	-7.99	1.24	1.21
5	4.25	6.47	2.22	-1.39	5.89	3.62	3.67		2.97	5.90	2.93	1.31	4.57	1.61	1.64

Figure I. Modified RC method to determine statistical significance (reliable change) in stuttering outcome measure of %SS within- and beyond-clinic for all participants from pre-treatment to 12 months post-treatment.

Figure II. Modified RC method to determine statistical significance (reliable change) in SSD outcome measure of PCC at the single-word and naming test and connected speech levels.