

Stuttering treatment for a school-age child with Down syndrome: A descriptive case report

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ABSTRACT

Background: Little is known about optimal treatment approaches and stuttering treatment outcomes for children with Down syndrome.

Aims and method: The purpose of this study was to investigate outcomes for a child with Down syndrome who received a combination of fluency shaping therapy and parent delivered contingencies for normally fluent speech, prolonged speech, and stuttered speech.

Results: In-clinic speech measures obtained at post-treatment and at 4 months follow-up reflected improvements in fluency of 89.0% and 98.6%, respectively. The participant's beyond-clinic follow-up sample reflected an improvement of 95.5%. Following treatment, the participant demonstrated improved self-confidence, self-esteem, and improved participation and functioning at school.

Conclusions: Findings suggest that fluency shaping with parental contingencies may be a viable treatment approach to reduce stuttering in children with Down syndrome. Future research using an experimental research design is warranted.

Educational objectives: Readers will be able to describe (a) prevalence estimates of stuttering in individuals with Down syndrome, (b) the main components of a fluency shaping program for a child with Down syndrome who stutters and has co-occurring speech and language delays, and (c) speech and parent-, teacher-, and self-report treatment outcomes.

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1. Introduction

Little is known about optimal stuttering treatment approaches and outcomes for children with Down syndrome (DS) despite prevalence estimates of stuttering among this population that range from 21% to 48% (Bloodstein & Bernstein Ratner, 2008), literature that suggests that those with DS may also exhibit cluttering (Van Borsel & Vandermulen, 2008; Van Borsel & Tetnowski, 2007), and the frequent presence of co-existing speech and language delays (Manning, 2001; Paul, 2001). This is the same for children who have co-existing intellectual disabilities or other genetic syndromes (e.g. fragile X syndrome or Prader-Willi syndrome) (Van Borsel & Tetnowski, 2007).

In terms of stuttering treatment approaches used with children with DS, survey evidence (Bray, 2003) suggests that speech-language pathologists use an array of approaches that range from direct fluency shaping or behavioural contingencies as in the Lidcombe Program of Early Intervention (Onslow, Packman, & Harrison, 2003) to indirect methods that

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include managing the communication environment and parent–child interaction therapy (Bray, 2003). With regard to fluency shaping, there is debate as to whether children with DS should learn fluency-skills. Bray (2003) reported that clinicians were concerned about the lack of generalization when fluency techniques were taught to children with DS and cautioned that fluency control techniques might be contra-indicated because the pressure to achieve control of fluency in the face of compromised linguistic and self-monitoring abilities could result in “failure, self-doubt and reduced self-esteem.” Similarly, Eckardt (2008) cautioned that “easy onsets” or “other stuttering modifications” might be too difficult and could result in “frustration” and “shame of stuttering”. However, she did suggest that teaching a slow rate of speech with phrasing and pausing techniques could be helpful. In contrast, others have more fully supported the direct teaching of fluency-enhancing techniques. For example, drawing from Cooper (1986), Manning (2001), and Ryan and Ryan (1995), Healey, Reid, and Donaher (2005) recommended that fluency-enhancing techniques (e.g., prolonged speech and gentle phonation onsets) can be used in treating children with DS. In addition, they suggested that length and complexity of utterances be manipulated, that substantial practice in structured and unstructured situations (i.e., treatment sessions, and home or school situations, respectively) be carried out, and that parents, teachers and other personnel be engaged to assist in maintenance of fluency skill use. Although there appears to be more support for the use of fluency enhancing techniques to treat children with DS than there are cautions, there is a surprising lack of research into the effectiveness of this treatment approach.

1.1. The current report

The aim of this descriptive case report is to provide a detailed description of the stuttering treatment programming provided to a school-age girl with Down syndrome and co-occurring speech and language delays and to report on her progress in therapy. Progress was measured in terms of reductions in stuttering and parent and child perceptions of the effects of treatment on self-confidence, self-esteem, anxiety, and social participation. We were particularly interested in learning if there were any negative effects of treatment on the latter variables.

2. Method

2.1. Participant

Sarah, aged 8 years, 10 months, presented with profound stuttering characterized by part word repetitions, silent prolongations, and audible prolongations. She often stuttered on several syllables within a word. Secondary features included visible lip tension and open mouth posturing during moments of stuttering. During the fluency assessment that occurred two months prior to starting therapy, Sarah’s stuttering ranged from 29.8% syllables stuttered (%SS) in a reading sample to 54.7%SS in conversation. Sarah’s responses on the Self-Rating of Effects of Stuttering – Children (Langevin & Kully, 1997) questionnaire indicated that stuttering was having a negative impact on her home and school life. Similarly, during the assessment, her mother reported that stuttering was causing Sarah significant frustration and that Sarah was engaging in situational avoidances. Given these stuttering characteristics and the absence of a fast rate of speech (see Fig. 1), a diagnosis of genuine stuttering was given. Readers are referred to Van Borsel and Vandermulen (2008) for a discussion of differential fluency diagnosis in individuals with DS. When referred to the clinic, Sarah had been stuttering for just under one year. She had no history of prior treatment for stuttering but had been receiving treatment since preschool for concomitant speech and language delays. Formal assessment at age 5 years 2 months had revealed a mild phonological delay. Formal language assessment with the *Clinical Evaluation of Language Fundamentals – Preschool 2* (CELF-P2; Wiig, Secord, & Semel, 2004) at 5 years 9 months revealed moderate expressive (6th percentile) and moderate receptive (3rd percentile) language delays with a language structure score in the severe range (first percentile). Sarah’s mother also reported that, since undergoing surgery to remove her tonsils and adenoids at age 7, Sarah’s speech tended to be hypernasal. While some speech sound distortions and substitutions were present in Sarah’s speech during the fluency assessment, her speech was generally intelligible. Sarah’s mother reported that she had received speech therapy to address articulation concerns up to Grade 2. Subsequent therapy focused on supporting Sarah’s expressive and receptive language skills during functional tasks.

At the beginning of treatment, Sarah was a Grade 3 student in a community classroom with program adaptations to support her learning needs. According to parent report, psychoeducational assessment completed at the end of grade 3 yielded an IQ score of 69, representing a mild intellectual impairment (American Academy of Pediatrics Committee on Genetics, 2011). Sarah’s communication and learning needs were well supported by her parents. Sarah was the oldest of two children.

2.2. Treatment

A sequential model of treatment was used (Bernstein Ratner, 1995) in that language treatment was suspended until treatment for stuttering was nearing completion. However, in Phase V of Sarah’s treatment program (see below) consultative support was given to her mother to assist her in supporting Sarah’s continued language development.

Treatment programming was primarily based on the Comprehensive Stuttering Program for School-aged Children (CSP-SC) (Kully & Boberg, 1991; Langevin, Kully, & Ross-Harold, 2007). The CSP-SC is an integrated program that directly addresses stuttering and associated social and emotional sequelae. It also incorporates training for client self-monitoring and parents

Table 1
Overview of the treatment program phases and components.

Phase (number of sessions and phase duration)	Components
I. Establishment (7 sessions over 4 weeks)	<ul style="list-style-type: none"> ○ Prolongation (called “stretch”) at a rate of 40–60 syllables per minute (SPM) ○ Easy breathing, gentle starts, smooth blending, and light touches ○ Self-corrections to modify moments of stuttering ○ Daily parent severity ratings ○ Parent participation in fluency skill practice in the clinic ○ Daily home practice that consisted of a warm-up of fluency skills and practice using fluency skills in simple activities that mirrored the length and complexity achieved in the clinic (e.g., “The car” or “I found a ball”)
II. Intensive Fluency Skill Practice (5 sessions in 1 week)	<ul style="list-style-type: none"> ○ Prolongation at a rate of 60–90 SPM with systematic increases in utterance length and complexity ○ 3T’s: a strategy to assist with language formulation and resisting time pressure that has the following three sequential elements: think, take a breath, and talk using stretch ○ Parent modelling of fluency skills during in-clinic sessions ○ Daily home practice that consisted of a warm-up and a stretch activity during which the parent praised Sarah for using stretch and requested corrections of stutters and non-stretched speech (using a ratio of 10 praises to 1 request for correction) ○ Parent praise for spontaneous stretch in naturally occurring exchanges (i.e., off-task talking that occurred outside of practice activities)
III. Transfer (7 sessions over 13 weeks)	<ul style="list-style-type: none"> ○ Prolongation at 90–120 SPM ○ Transfer activities that included scavenger hunts and surveys with unfamiliar adults and participation in simulated school sessions with other children who stutter ○ Attitudinal/emotional support as needed ○ Participation in a teasing and bullying discussion ○ Home practice that included a warm-up of fluency skills and the provision of contingencies for stretch and stuttered speech during practice activities. ○ Praise for fluent speech achieved with or without fluency skills – the ratio of praise to correction was changed to 5 praises to 1 request for correction ○ Prolongation at a rate of approximately 120 SPM
IV: Consolidation of Fluency Skills and Training of Classroom Support Staff (16 sessions over 28 weeks)	<ul style="list-style-type: none"> ○ Family support provided regarding continued implementation of fluency skill practice and contingencies for smooth and stuttered speech ○ Consultative support provided to Sarah’s school team (teacher and educational assistant) ○ Concept of being “calm and cool” introduced to facilitate self-regulation ○ Praise for remaining “calm and cool” given by clinician and parents ○ Praise for Sarah’s use of 3T’s provided by the parent ○ Adults in Sarah’s environment were taught to model a slower rate of speech, a calm and relaxed body, and to show they were thinking first before speaking
V: Refinement of Fluency Skills and Home Programming (6 sessions over 29 weeks)	<ul style="list-style-type: none"> ○ Continued in-clinic sessions that included fluency skill warm-ups and practice using stretch and other fluency skills ○ Focused skill practice in functional situations (e.g., practice with reading aloud, giving a presentation, or answering questions) ○ Parent provided with strategies to strengthen emerging language skills ○ Transfer planning continued ○ Consultative support was provided to facilitate language development
VI: Maintenance and Follow-up (no sessions over 20 weeks)	<ul style="list-style-type: none"> ○ Parental contingencies for stretched/smooth speech were withdrawn gradually ○ Use of fluency skills and prolongation continued as needed ○ Consultation with treating clinician occurred as needed

are trained to deliver the therapy at home. Fluency skills and corrections of stuttered speech are used to achieve reductions in stuttering. In the CSP-SC parents have always provided contingencies for fluent speech achieved with the use of fluency skills and contingencies for stuttering (i.e., they were asked to fix the stutter). Parents also provided daily global ratings of their child’s beyond clinic speech. In 2004, contingencies for fluent speech produced without fluency skills were added and global ratings were changed to numerical parent severity ratings. These changes were drawn from the Lidcombe program (Onslow et al., 2003).

Sarah’s treatment program consisted of six phases, with the frequency of treatment varying according to the phase of treatment. An overview of her treatment program including the number of sessions and weeks in each phase is shown in Table 1. In general, Phases I–V focussed on establishment, transfer, and consolidation and refinement of fluency skills. In Phase VI (the maintenance period) in-clinic sessions were suspended and parent delivery of treatment at home was gradually withdrawn.

In total, over the course of 17 months Sarah received 43 h of treatment in 41 treatment sessions. She also participated in a simulated classroom transfer. In addition, Sarah’s parents attended one consultation visit made to the school by the treating clinician (the first author). Follow-up measures were gathered at 4 months post treatment.

2.2.1. Phase I: establishment of fluency skills

In Phase I Sarah learned to prolong speech (i.e., “stretch” speech) at a rate of 40–60 syllables per minute. She also learned the following fluency skills which are more fully described in Langevin et al. (2007): easy breathing, gentle starts, smooth blending and light touches. Briefly, prolonged speech in the CSP-SC is achieved by prolonging vowels. Transitions through consonants are slowed but not prolonged. Easy breathing establishes a relaxed inspiration–expiration cycle. Inspiration has a diaphragmatic locus and careful attention is paid to ensure that the inspiration does not exceed the child’s normal lung capacity. Learning to break speech into breath groups (i.e., more manageable phrases) is also part of the easy breathing skill. Gentle starts are used at the beginning of each new breath group or utterance. They are characterized by an easy relaxed initiation of speech with a gradual increase in loudness occurring within the first syllable of the utterance. No differentiation in onset is made between vowel or consonant initiated words and voiced or voiceless beginning sounds. That is, the manner of sound productions is preserved. When using smooth blending, airflow is continuous and syllables within the breath group are linked as would occur in normal co-articulation. The voicing characteristics of phonemes within the utterance are preserved. When using light touches, consonants within breath groups are produced with lightened articulatory contacts. Again, normal voicing of sounds is preserved.

During this phase of treatment and in the intensive practice in Phase II (see below), the length of utterances and the complexity of language progressed from short conceptually simple utterances to more complex and abstract conversation that did not exceed Sarah’s developing language skills. Spontaneity was also varied, beginning with highly structured speaking tasks and moving to natural exchanges. To facilitate the transition to using fluency skills in natural conversation, Sarah was reinforced (i.e., praised) for spontaneously using “stretch” in off-tasking talking that occurred alongside the focussed practice activity or before or after it.

In addition to the above fluency skills, Sarah also was taught to modify moments of stuttering with self-corrections. To carry out a self-correction, she would release tension, inspire and then begin speaking with a gentle start. This skill requires the ability to self-monitor fluency, in particular, the ability to independently detect and modify moments of stuttering.

In order to conduct Sarah’s daily home practice and to facilitate generalization of fluency skills to the home environment, Sarah’s mother learned to model the fluency skills and learned to carry out skill practice activities in the home. In this phase, activities consisted of the completion of a short warm-up of fluency skills in single words and short phrases followed by a fluency skill practice using talking activities that were at the same level of length and complexity as in the in-clinic treatment sessions. As soon as Sarah began to spontaneously use stretch in structured practice in both the clinic and home sessions, she was praised for doing so. Sarah’s mother also learned to rate Sarah’s stuttering severity (1 = no stuttering, 10 = the most severe stuttering imaginable) and recorded daily severity ratings. In this phase Sarah’s father also attended a session to become familiar with the fluency skills.

2.2.2. Phase II: intensive fluency skill practice

Sarah and her mother attended one week of daily one-hour treatment sessions (i.e., 5 sessions). During these sessions, prolongation practice was carried out at a rate of 60–90 SPM. The length and linguistic complexity of utterances was increased (e.g., answering “wh” questions and describing pictures etc.). Sarah’s mother learned to model prolongation at the rate of 60 to 90 SPM and to provide contingencies for non-stretched (e.g., “can you tell me that in stretch?”) and stuttered speech in daily home practice activities (i.e., she asked for a self-correction using terms such as, “let’s smooth that out” or “can you stretch that”). A ratio of 10 praises for stretched speech to 1 correction for non-stretched or stuttered speech was used. Sarah’s mother also began to praise spontaneous use of stretched speech in naturally occurring exchanges. To facilitate language formulation and the ability to resist time pressure, Sarah was taught the 3Ts skill. The 3Ts is a fluency enhancing skill that helps children deal with language demands and time pressures. It is comprised of three sequential elements: **Think** before speaking, **Take** time to breathe, and **Talk** using fluency skills (or ‘smooth speech’ in the later stages of therapy). During the “Think” stage children are encouraged to think about taking time to start talking; what they want to say and how they will say it; and what speech skills they will use.

2.2.3. Phase III: transfer

During this phase Sarah received 9 h of therapy in 7 sessions. Fluency skills were practiced at 90–120 SPM in a variety of settings with a variety of speaking partners. For example, Sarah completed scavenger hunts and surveys with clinic staff members and attended two simulated school-days with other children who stutter. Sarah’s mother was also trained to praise fluent speech that was achieved with or without fluency skills. For example, she would say, “that was nice and smooth” if Sarah’s speech was fluent. Sarah’s mother learned to plan and carryout transfer activities with Sarah. Because Sarah had been teased by a peer in her classroom, she participated in a discussion with other children who were receiving treatment in a group intensive treatment program. The CSP-SC generally includes discussions about teasing and bullying (e.g., what bullying is, why kids bully, how it feels to be bullied, and potential response strategies). Parents also participate in a discussion that focuses on how they can help their child deal with bullying.

2.2.4. Phase IV: consolidation of fluency skills and training of classroom support staff

Sarah attended 16 twice monthly sessions over 7 months and the treating clinician attended a meeting with Sarah’s family and school team. During this phase, fluency skills were practiced at approximately 120 SPM. Clinic sessions and home practice activities focussed on extending Sarah’s fluency to a variety of settings and conversational partners. In addition,

a strategy to assist with self-regulation was introduced. Sarah was taught to remain “calm and cool” and her mother was taught to cue and praise Sarah for remaining so. Sarah’s mother continued to model and reinforce Sarah’s use of 3Ts. Adults in Sarah’s environment also were taught these strategies. In addition, they were taught to prolong speech so they could model a slowed rate of speech to support Sarah’s fluency at home and at school. Sarah’s father and an educational assistant attended sessions during this phase and the treating speech-language pathologist attended a meeting at Sarah’s school to provide support to Sarah’s teacher.

2.2.5. Phase V: refinement of fluency skills and home programming

In this phase Sarah attended 6 one-hour treatment sessions over an 8 month period (approximately monthly sessions). Treatment sessions focused on refining fluency skills and, in consultation with Sarah’s mother, further adaptation of programming to meet Sarah’s needs. Sarah’s mother continued to plan transfer activities designed to help Sarah be more consistently fluent in more settings. Consultative support was also provided to Sarah’s mother to help her adapt practice activities to facilitate Sarah’s language development in terms of emerging language structures and concepts. For instance, Sarah’s mother provided scaffolding during practice activities targeting Sarah’s ability to provide more detailed descriptions and Sarah’s understanding and use of questions (e.g., “how”, “why”, and “when”). Post-treatment measures were completed at the end of this phase.

2.2.6. Phase VI: maintenance and follow-up

In this period, Sarah’s mother began to phase out direct skill practice and withdrew praise for fluent speech. The treating speech-language pathologist was available to the family for additional consultation as needed. Follow-up measures were gathered at the end of maintenance which was completed at 4 months post-treatment.

2.2.7. Child initiated program adaptations

According to parent report Sarah often engaged in independent warm-up and fluency skill practice. That is, she initiated and carried out her own practice. This continued throughout the treatment program. She also was often overheard using fluency skills while talking aloud during pretend play (e.g., while playing school).

2.3. Outcome measures

2.3.1. Speech measures

Measures of percent syllables stuttered (%SS) and syllables spoken per minute (SPM) were obtained from in-clinic conversation and reading video-samples obtained at pre-treatment, post-treatment, and follow-up. As well, a home sample was obtained at pre-treatment (audio-sample) and follow-up (video-sample). Frequency counts of stutters and syllables spoken were made on an electronic button-press event and timing device (Boberg & Kully, 1985). Each syllable was counted only once as stuttered or non-stuttered. Formulative pauses of more than 1 second were excluded. Speech rate was calculated by dividing total syllables spoken by total speaking time. To examine reliability of speech measures, 75% (6 of 8) of the speech samples were rated by a research assistant who was independent of the therapy program and blind to the purpose of this study. Speech samples were presented in random order. Before beginning to analyse the samples in this study, the research assistant had been trained to rate samples using guidelines established at the Institute for Stuttering Treatment and Research (ISTAR) (Kully, 1986) and was required to establish 90% inter-rater agreement with the executive or clinical director of ISTAR. Intra-class correlations were used to assess inter-rater reliability. Correlations were 0.99 for both %SS and SPM.

2.3.2. Severity ratings

Sarah’s mother made daily severity ratings, using the 10 point scale described above. Inter-rater reliability of severity ratings made by both Sarah’s mother and the treating clinician was examined. From the 14th session onward, severity ratings were made independently at the beginning of each treatment session and then discussed. Data from both the clinician and the parent were available for 79% of these sessions (i.e., 22 of the 28 subsequent sessions). The intra-class correlation was .96.

2.3.3. Self-, parent-, and teacher-report

The Self Rating of Effects of Stuttering – Children (SRES-C) (Langevin & Kully, 1997) was administered to Sarah before treatment and at follow-up. The SRES-C uses a 7 point scale to measure the impact that stuttering has on various aspects of home and school life (e.g., “How much does your stuttering interfere with or affect...talking with family...talking with friends...your school work?”). Response options range from 0 to 6 (0 = not at all, 1–2 = a little bit, 3–4 = quite a bit, and 5–6 = a lot).

A post-treatment questionnaire developed for this study was used to obtain parent perceptions of Sarah’s participation at follow-up relative to pre-treatment. It also examined the degree to which therapy affected confidence, self-esteem and the degree to which therapy was enjoyed or stimulated anxiety. Response options for rating degree of participation were “never”, “sometimes”, “often” and “always”. Response options for rating anxiety were “no”, “little”, “some” or “a lot” of anxiety. Response options for rating the effect of treatment on self-esteem and self-confidence ranged from “greatly lowered”

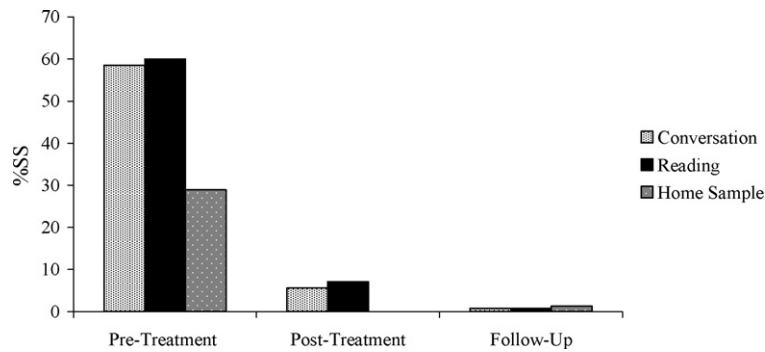


Fig. 1. Percent syllables stuttered (%SS) at pre-treatment, post-treatment and follow-up.

to “greatly increased” confidence or self-esteem. Sarah’s mother also was asked in this questionnaire to comment on the challenges faced during the treatment process.

A post-therapy speech performance questionnaire developed at ISTAR and used regularly, measured Sarah’s perceptions of her overall level of fluency and degree of self-confidence. Response options ranged from “not improved” to “greatly improved.” When the questionnaire was administered to Sarah, the response options were explained, the item was read to her and Sarah made her choice by circling her response. Sarah often added comments to support her answer (e.g., when choosing “very good” she commented that her speech was smooth).

Finally, a questionnaire developed at ISTAR and used regularly investigated teacher perceptions of Sarah’s fluency at school, her response to speech difficulties, difficulties associated with answering questions and responding to time pressures, and difficulties experienced in other situations. The pre-treatment questionnaire was completed by Sarah’s 3rd grade teacher and the post-treatment questionnaire was completed by her 5th grade teacher at follow-up.

3. Results

3.1. Speech measures and parent severity ratings

Sarah made substantial gains in fluency and speech rate (Figs. 1 and 2, respectively). In the in-clinic speech samples, her stuttering decreased from an average of 59.25%SS at pre-treatment to 6.35%SS immediately post-treatment and 0.80% at follow-up. Accordingly, her rate of speech increased from 33.40 SPM at pre-treatment to 149.75 at post-treatment and 182.85 at follow-up. Home samples showed a reduction in stuttering from 28.9%SS at pre-treatment to 1.3%SS at follow-up.

As shown in Fig. 3, there was much variability in parent severity ratings over the course of treatment; however, Sarah’s mean weekly ratings decreased from 8.2 at pre-treatment to 2 at follow-up. Her ratings in the final 5 weeks of maintenance (from the 92 to the 96th rating) reflected more stability in fluency, with four of the ratings being 2 and one being 3 (a mean of 2.25).

3.2. SRES-C

At follow-up Sarah’s ratings of impact showed substantial improvement in terms of the negative effects that stuttering was having in talking with friends and family (Table 2). She indicated that she was no longer bothered or worried about her

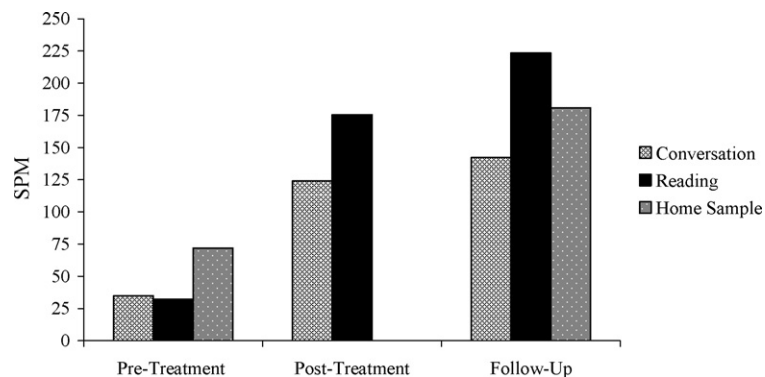


Fig. 2. Syllables spoken per minute (SPM) at pre-treatment, post-treatment, and follow-up.

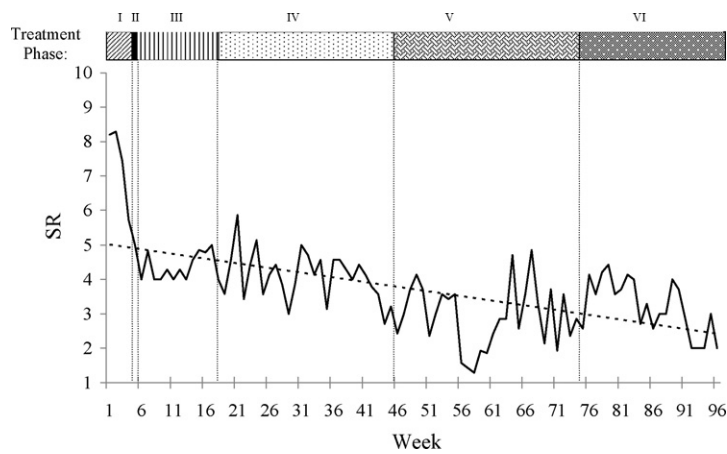


Fig. 3. Weekly mean parent severity ratings (SR).

Table 2
Self rating of effects of stuttering.

Item	Pre-treatment	Follow-up
How much stuttering interferes with or affects:		
Talking with friends	6	0
Making new friends	6	2
School work	6	4
Talking with family	6	0
How much stuttering bothers you	6	0
How much you worry about your stuttering	6	0

Note: Response options range from 0 to 6 with 0 = not at all, 1–2 = a little bit, 3–4 = quite a bit, and 5–6 = a lot.

stuttering. However, although there was improvement, stuttering was still having some effect on making new friends and her school work.

3.3. Parent and child report of the impact of therapy

Reports from Sarah and her mother indicated that therapy had a positive impact on Sarah in terms of improved self-confidence and use of skills outside of the clinic (Table 3). According to Sarah’s mother, there was substantial improvement in Sarah’s self-confidence, self-esteem and participation. She also reported that Sarah did not experience anxiety as a result of the therapy experience and that Sarah always enjoyed therapy.

3.4. Teacher’s evaluation of fluency

The teacher’s evaluation of Sarah’s fluency also showed substantial improvement in terms of the amount of stuttering that Sarah was exhibiting at school (Table 4). Sarah was reported to no longer have trouble answering questions or dealing with time pressures and she was no longer avoiding situations. In contrast she was self-correcting stutters. In terms of remaining difficulties, Sarah was still having some difficulty in oral reading in that she was tending to read too quickly which, presumably, resulted in some stuttering.

Table 3
Parent and child report of the impact of the therapy experience.

Item	Parent	Child
Self-confidence	Greatly improved	Greatly improved
Self-esteem	Greatly improved	n/a
Anxiety	No anxiety	n/a
Participation	Improved (sometimes participated previously, now often participates)	n/a
Enjoyment of therapy	Always enjoyed	n/a
Use of skills outside of clinic	Most of the time	Most of the time

Note: Response options were “never”, “sometimes”, “often”, and “always”.

Table 4

Teacher's evaluation of speech fluency.

Item	Pre-treatment	Follow-up
Amount of stuttering noted at school (1 = no stuttering, 10 = lots of stuttering)	7.5	2
How the child reacts to any speech difficulties present	Avoids some speaking situations, sometimes appears frustrated or distressed when experiencing difficulty	Self-corrects by slowing down (note: when oral reading has a tendency to read quickly)
Does the child experience more difficulties when:		
• Answering questions	Yes	No
• There are time pressures	Yes	No
Other situations in which the child has more difficulty	Raising her hand to participate in class discussions	On occasion when oral reading

3.5. Parent report of challenges experienced

Sarah's mother reported having difficulty finding time to devote to daily home practice and that learning to evaluate Sarah's fluency while simultaneously listening to content was challenging. She reported that "the significant amount of time spent focussed on Sarah's needs left little time for her other child". Finally, Sarah's mother indicated that feeling as if she was "the sole person" that Sarah relied on during this difficult time was challenging.

4. Discussion

Findings of this descriptive case report suggest that treatments that use a combination of direct fluency-skill training and parent administered behavioural contingencies may be a viable treatment approach for school-age children with Down syndrome and that therapy can be an enjoyable experience. Sarah's post-treatment measures reflected an 89.0% improvement in fluency immediately post-treatment and a 98.6% percent improvement at follow-up as measured in the in-clinic samples. Her beyond-clinic follow-up sample reflected an improvement of 95.5%. Concomitant increases in speech rate suggest that her improvement in fluency was not compromised by unusually slow speech and that she was more efficient in imparting information or conversing. Sarah's improvement in fluency is consistent with children who participated in a pre-post outcome study of the CSP-SC who, with the exception of stuttering, were typically developing. In Kully and Boberg (1991), (8) school-age children (aged 6, 9–11 years) who received either intensive or extended therapy showed improvements in fluency that ranged from 84% to 100% at the end of transfer. At 8–18 months follow-up, 6 of the 8 children were showing improvements in fluency that ranged from 79.0% to 98.5%. In Langevin et al. (2007), (3) of the 4 children reported on were showing improvements of 52% to 75% at 6–19 months follow-up. It is notable that (a) Sarah's pre-treatment conversational %SS (58.5%) well exceeded the pre-treatment range of 4%SS to 35%SS of all the school-age children in Kully and Boberg (1991) and the 3 children who had a pre-treatment range of 6.5%SS to 11.5%SS who were maintaining treatment gains in Langevin et al. (2007). It is also notable that, unlike the fourth child in Langevin et al. (2007) who had equally high pre-treatment stuttering (54.4%SS) but was in relapse at follow-up, Sarah was maintaining her treatment gains despite the severity of her earlier stuttering and despite having a mild cognitive impairment. It is possible that Sarah's extended treatment program contributed to better maintenance of stuttering reductions than the shorter 3 week intensive program undertaken by the fourth child in Langevin et al. As also reflected in the parent severity ratings at the end of maintenance, Sarah's residual stuttering was clearly mild in comparison to the profound impairment that she was experiencing at pre-treatment. According to parent and teacher report, it is also clear that at the end of treatment Sarah was functioning better at school and in social relationships. In contrast to therapy being onerous and anxiety provoking, it appears that therapy was an enjoyable experience for Sarah.

Regarding parent challenges, it is evident that Sarah's mother experienced challenges that parents of typically developing children who stutter have anecdotally reported over many years. In particular, the most common challenge that parents encounter is finding time for daily home practice. This finding is consistent with that of Koushik, Shenker, and Onslow (2009) who found that parents of school-age children receiving the Lidcombe Program (Onslow et al., 2003) reported difficulty in finding time to carryout structured conversations each day.

4.1. Limitations and future research

Because an experimental case study design was not used in Sarah's programming, no direct relationship between therapy and outcomes can be concluded. However, it should be noted there was no improvement in stuttering in the two month interval between her assessment and the beginning of treatment. As well, generalization to other children with Down syndrome and in particular those with greater cognitive impairments cannot be made. However, there is evidence that children with cognitive impairments that were formerly labelled as "educable" responded favourably to a fluency shaping program that was a precursor to the CSP-SC (Boberg & Fong, 1980). In contrast, children with cognitive impairments labelled "trainable" as opposed to "educable" did not respond favourably even to a program that was radically modified. Nevertheless, it is possible that other children with Down syndrome who have cognitive functioning and self-monitoring abilities similar

to Sarah and similar or less severe pre-treatment stuttering severity may benefit from a fluency shaping based treatment program such as that used with Sarah. Experimental research of outcomes with the programming used in this study and with different treatment approaches is clearly needed for this population of children.

5. Conclusion

Findings support recommendations in the literature that suggest that fluency shaping may be successfully used to treat stuttering in children with Down syndrome.

CONTINUING EDUCATION

QUESTIONS

- 1) Published prevalence estimates of stuttering among individuals with Down syndrome range from:
 - a. 5–10%
 - b. 10–16%
 - c. 21–48%
 - d. 36–50%
 - e. 0–5%
- 2) When polled, speech-language pathologists reported they use the following methods to treat stuttering in children with Down syndrome:
 - a. indirect treatment approaches only
 - b. fluency shaping only
 - c. behavioural contingencies for smooth speech only
 - d. direct and indirect treatment approaches
 - e. environmental modifications only
- 3) Sarah's response to using fluency skills was:
 - a. consistent with typically developing children reported on in earlier CSP-SC outcome studies
 - b. atypical relative to typically developing children reported on in earlier CSP-SC outcome studies
 - c. characterized by frustration and anxiety
 - d. negatively impacted by her delayed speech and language skills
 - e. negatively impacted by her inability to respond to a model of fluency skills
- 4) The following were the essential components of Sarah's treatment program:
 - a. parent training to gather severity ratings
 - b. establishment of fluent speech through indirect methods
 - c. participation in transfer activities such as a mock school day and scavenger hunts
 - d. manipulation of length and complexity of Sarah's utterances within Sarah's developing language skills
 - e. prolongation and fluency skills, parent delivered contingencies for fluent and smooth speech, and environmental modifications
- 5) At follow-up, results showed that:
 - a. reductions in stuttering reflected less than 50% improvement and improved participation but no change in self-confidence
 - b. reductions in stuttering reflected between 60% and 89% improvement with minimal improvement in self-confidence and self-esteem
 - c. reductions in stuttering reflected 89% improvement and greater and improved participation, self-confidence and self-esteem improved
 - d. minimal change in fluency but improved participation, self-confidence and self-esteem
 - e. return to pre-treatment levels of fluency and no improvement in participation

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