Down Syndrome and Childhood Apraxia of Speech:

Matching a Unique Psycholinguistic Profile to an Effective Treatment Program

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Abstract

Three single subject experiments were conducted to explore the response by individuals with Down Syndrome (DS) to different treatments for the remediation of speech impairments. Although the speech impairment in DS is typically described as dysarthria, a growing body of research suggests that there may be concomitant childhood apraxia of speech (CAS) and phonological disorders that explain poor speech accuracy and intelligibility in these individuals. The appropriate treatment to remediate speech impairment in DS may depend upon each individual's psycholinguistic profile. Three single subject randomized experiments were conducted with participants ranging in age from 10-20 years. For each experiment, two treatment conditions were compared to a control condition: the experimental conditions addressed the underlying deficits associated with either CAS (a motor planning impairment) or Inconsistent Phonological Disorder (a phonological planning impairment). High intensity treatment was undertaken with each participant receiving a total of 18 treatment sessions in six to nine weeks. In each study, the results indicate improvement in speech accuracy but with varying response across participants to specific treatment approaches. Follow-up assessments with each participant demonstrate maintenance of learning over time. Each participant's specific response to treatment is discussed in relation to their psycholinguistic profile as revealed by their pre-treatment assessment results.

Résumé

Trois études de cas unique ont permis d'explorer l'effet de différents traitements visant à remédier aux troubles de la parole associés au syndrome de Down. Ces troubles sont typiquement décrits comme de la dysarthrie, cependant un nombre croissant de recherches suggère que l'apraxie verbale développementale concomitante avec des désordres phonologiques pourraient expliquer la pauvreté de la précision articulatoire et de l'intelligibilité chez ces individus. Le traitement approprié pour remédier aux troubles de la parole du syndrome de Down dépend du profil psychologique de l'individu. Trois expériences aléatoires ont été conduites sur chacun des participants âgés de 10 à 20 ans. Deux conditions de traitements ont été comparées à une condition contrôle dans chacune des études : les conditions expérimentales ciblaient des déficits sous-jacents associés avec l'apraxie verbale développementale (trouble de la planification motrice) ou bien avec des troubles de la planification phonologique. Chaque participant a suivi un traitement intensif au cours de 18 sessions réparties sur une période allant de six à neuf semaines. Les résultats de chaque étude indiquent une amélioration de la précision articulatoire, avec cependant des réponses variables selon les différents types de traitements. Après deux mois, une évaluation de suivi de chaque participant a permis de démontrer le maintien des apprentissages sur le long terme. Les réponses de chaque participant aux différents traitements sont analysées en relation avec leur profil psycholinguistique, tel qu'évalué avant le début du traitement.

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Preface

This thesis research was conducted in the Child Phonology Laboratory Research Clinic that provides experimental interventions to children and young adults with severe speech disorders. In this clinic interventions are provided by a collaborative team that includes the lab director Dr. Susan Rvachew, Ph.D., S-LP(C), and doctoral student, Ms. Tanya Matthews MA, CCC, SLP, who supervise McGill University student S-LPs who provide the interventions, graduate student research assistants who process all the video and audio research data collected in the experiments, and volunteers who assist with the recording of data and other routine tasks. I contributed to this research by developing the research proposal, recruiting the three participants, providing the interventions during my final internship as an M.Sc. applied student, and conducting the primary video coding, as well as the reliability coding of the probe data. Dr. Rvachew selected treatment goals and randomly assigned treatments to targets and sessions using blinded procedures. Tanya Matthews supervised the provision of the interventions. Lizzie Carolan, and Omar Obregozo Zalava, transcribed the outcome data using procedures to ensure that coders were blinded to the treatment condition. Data analysis was conducted jointly by myself, Tanya Matthews, and Dr. Rvachew. I wrote the thesis independently.

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Intelligible speech is an essential factor for successful verbal communication. It can be categorized into information that is signal dependent, and information that is signal independent: the former referring to the sound signal itself; the latter contextual information such as facial expression and syntax (Miller, 2013; Yorkston, Strand, & Kennedy, 1996). Both of these broad categories are of concern for Speech-Language Pathologists, who seek to maximize their clients' verbal and nonverbal communication abilities. Low speech intelligibility has deleterious effects on communicative capability, and restricts the individual's ability to participate fully in conversation (Miller, 2013).

Individuals with speech sound disorders, an umbrella term for those with linguistic or articulation level difficulties, generally have poor speech intelligibility. The predominant goal of speech therapy for these individuals is to improve the clarity and accuracy of the acoustic speech signal, thereby positively influencing the intelligibility of the speaker (Namasivayam, et al., 2013; Kumin, 2006). This area of speech therapy typically addresses the signal dependent component of intelligibility. In the majority of cases of poor speech intelligibility, the speaker mispronounces certain natural classes of speech sound, producing predictable patterns of error. For example the individual might produce all the tongue tip sounds with the tongue slightly between the teeth resulting in a distortion of these sounds, e.g., 'the sunshine state' →[ðə s̪ʌnʃ̪ɑɪn s̪teɪt̪]; in this case the individual presents with an articulation disorder characterized by poor phonetic knowledge of specific articulatory gestures. Alternatively the individual might lack phonological knowledge of entire classes of phonemes, for example, representing all liquid sounds as glides, as in 'red lollypop' → [wɛd wɑwipap]; in this example, the presence of predictable patterns of phoneme substitution errors, called phonological processes, signal the presence of a phonological disorder (Rvachew and Brosseau-Lapré, 2012).

Less frequently, speech sound disorders will be caused by motor dysfunction (see Namasivayam et al., 2013; Murray, McCabe, and Ballard, 2015; Skinder, Strand, and Mignerey, 1999 for examples). Generally, motor speech disorders fall into two basic types, dysarthria and apraxia. Dysarthria is a movement disorder at the level of the central nervous system or the peripheral nervous system (lower motor neurons): therefore, execution of movements is affected (Duffy, 2013) so that rate, resonance, phonation and respiration may all be impaired. In contrast, apraxia is a motor speech disorder that affects speech during the planning and programming stages, prior to execution of movements themselves (Duffy, 2013), leading to speech errors are likely to be unpredictable and inconsistent. In some cases, an individual may present with characteristics of both types of motor speech disorders, and in this case the presenting disorder may be referred to as 'not otherwise specified' meaning that the cluster of presenting symptoms is not specific to either dysarthria or apraxia of speech (Shriberg, Strand, & Mabie, 2016).

Low speech intelligibility may also be caused by structural differences: in some cases the structure of the speech system is atypical to the extent that it directly causes speech errors such as in cleft palate (Ysunza, 2015; Cleland, Wood, Hardcastle, Wishart & Timmins, 2010). However, some individuals compensate very well despite structural differences. The impact of structural impairments may vary depending upon the effectiveness of surgical corrections or the presence of concomitant disorders such as co-occurring phonological or motor speech disorders (Cleland et al., 2010; Kent & Vorperian, 2013).

Some individuals present with poor intelligibility stemming from widespread impairments such as comorbid phonological disorder, motor speech disorder, and structural differences: individuals with Down syndrome (DS) may be included in this classification. Feeding challenges, drooling, and tongue thrust have also been reported in this population suggesting impaired coordination and low oral sensitivity (Kumin, 1994; Cleland et al., 2010).

Speech intelligibility was identified as a major concern by parents of 937 individuals with DS, surveyed by Kumin (1994): 58% of the survey respondents reported that low speech intelligibility was a *frequent* issue for their child; 37% noted this same problem *sometimes*. Only 5% of respondents reported that speech intelligibility was not an issue for their child. In the same survey over 80% of respondents reported articulation as an area of difficulty for their child (Kumin, 1994).

It has long been recognized that poor speech intelligibility in individuals with DS results from a combination of structural differences in combination with dysarthric components (Shriberg, Strand, & Mabie, 2016; Silverman, 2007; Stoel-Gamon, 1997). However, it has recently been suggested that apraxia also plays and important role in this population (Rulpela & Manjuela (2010); Rupela, Vellman & Andrianopoulos, 2016; Shriberg, Strand, & Mabie, 2016). Identifying motor speech disorders such as Childhood Apraxia of Speech (CAS) in the DS population is particularly difficult, as a primary diagnosis of DS includes inherent hypotonia/dysarthric components, as well as craniofacial differences. Further, heterogeneity is well documented within the DS population, with respect to long term trajectories of independence, (Fredricks, 1988), and levels of cognitive, speech, or language function (Rupela, Vellman & Andrianopoulos, 2016; Shriberg, Strand, & Mabie, 2016; Silverman, 2007; Stoel-Gamon, 1997). In this thesis research project, I investigated the effectiveness of an intervention targeting CAS for individuals with DS. My goal was to improve speech accuracy in participants with DS by better matching treatments to their underlying psycholinguistic profile.

Literature Review

General Concerns in Down syndrome

Down syndrome is among the most common developmental disabilities with genetic causation (Secretariat of World Health Organization, 2010). DS is caused by one of the following: a) complete trisomy of the 21st chromosome system wide, b) translocation of part of the 21st chromosome onto another chromosome, or c) complete trisomy of the 21st chromosome in only some of the system's cells (National U.S. Library of Medicine, 2015). The current incidence of DS is one in every 700 live births (Centre for Disease Control, 2014). DS is typified by a range of lifelong health concerns, including but not limited to a high prevalence of congenital heart defects (40-50%), hearing impairments (75%), vision problems (60%), obstructive sleep apnea (50-75%), and otitis media (50-70%) (Bull, 2011; Centre for Disease Control, 2014). The health concerns listed above require consistent monitoring and frequent medical intervention in order to maximise the health, wellbeing, and longevity of individuals with DS. Due to medical and educational advances the mean life expectancy of individuals with DS has increased by nearly 500% in the last seventy years and was reported to be age 47 in the year 2007 (Centre for Disease Control, 2014).

In addition to the medical concerns outlined above, individuals with DS also have a mild to moderate cognitive delay, and moderate to severe deficits in speech and language which exceed the level of cognitive impairment (Chapman, 1998; Cleland et al., 2010). Further, researchers have found differences in oral control and swallowing function, which also implies that individuals with DS may present with sensorimotor coordination issues Cleland et al. 2010; Kumin, 2006). Due to the complexity of needs in the areas of health, learning, speech and language, individuals with DS regularly have a team of therapists to support their development. The areas of speech and language are of particular concern because they affect the individual's ability to communicate with others and thereby maximise their independence (Fredericks, 1988; Goldstein, 1989).

Speech Development in Down syndrome

Some researchers opine that an individual's speech demonstrates a delay if it follows the same developmental pattern as a typically developing (TD) child, whereas a disorder denotes an atypical developmental sequence (Kent & Vorperian, 2013). Individuals with DS present with a mix of delayed and disordered speech: an uncommon trajectory is evident from the earliest junctures of speech development. For example, Kent & Vorperian, (2013), reported more nonspeech-like sounds and fewer speech-like sounds than TD individuals in infanthood: though babbling in DS is delayed and has different features than babbling in TD infants, but it is not as delayed as gross motor milestones such as sitting or crawling, which suggests the beginning of a unique developmental profile (Kent & Vorperian, 2013).

Adolescents with DS appear to present with persistent consonant errors and phonological processes, though the data is sparse. Sommers, Patterson & Wildgen (1988) reported that the frequency of errors was dependent in part on whether the individual with DS produced spontaneous or imitated speech, with the highest number of errors and phonological processes found in connected speech. In addition, they found that the frequency of speech errors was contrary to what would be expected of typical phonological development including high incidence of errors for nasal phonemes, liquids and stop consonants. Kent & Vorperian (2013) supported this idea by noting that seven of the ten most common error phonemes in the DS data were for the alveolar place of articulation, despite the fact that these are very early phonemes to develop in TD children. Given this information, it is evident that a unique profile of speech is maintained in adolescence for the DS population, well past the point where TD children have achieved accurate adult-like productions (Rvachew & Brosseau-Lapré, 2012).

The origin of speech deficits in Down syndrome. Many individuals with DS demonstrate speech deficits, the cause of which is widely agreed to be multifactorial (Cleland et al., 2010; Kent & Vorperian, 2013; Kumin, 2006; Patterson, Rapsey and Glue, 2013). As depicted in *Figure 1,* limited verbal short term memory, limited auditory short term memory, cognitive impairment, language impairment and physical differences all contribute to the broad speech profile of the DS population (Cleland et al., 2010; Kent & Vorperian, 2013; Kumin, 2006; Patterson et al., 2013). However, despite the presence of multiple adverse factors, the extent to which any given individual is affected varies widely (Patterson et al., 2013).

In agreement with this idea, Kent & Vorperian (2013) stated in a review of the DS phonology literature that while a subgroup of the population with DS may present with nondevelopmental errors, they do not inevitably occur across the population. However, a subgroup of individuals with DS *is* strongly impacted by the impairments outlined in *Figure 1*: this combined impairment is positively correlated with low overall speech intelligibility (Kumin, 2006). There is debate in the literature as to whether the interaction of the factors in *Figure 1* is sufficient to explain speech delay in DS (Stoel-Gamon, 1997), or whether a deficit in speech motor control, specifically apraxia of speech, is an additional explanatory factor (see Kumin, 2006; Rondal & Edwards, 1997; Cleland et al., 2010 for examples).

Figure 1: General Factors Contributing to Speech Deficits in Down Syndrome

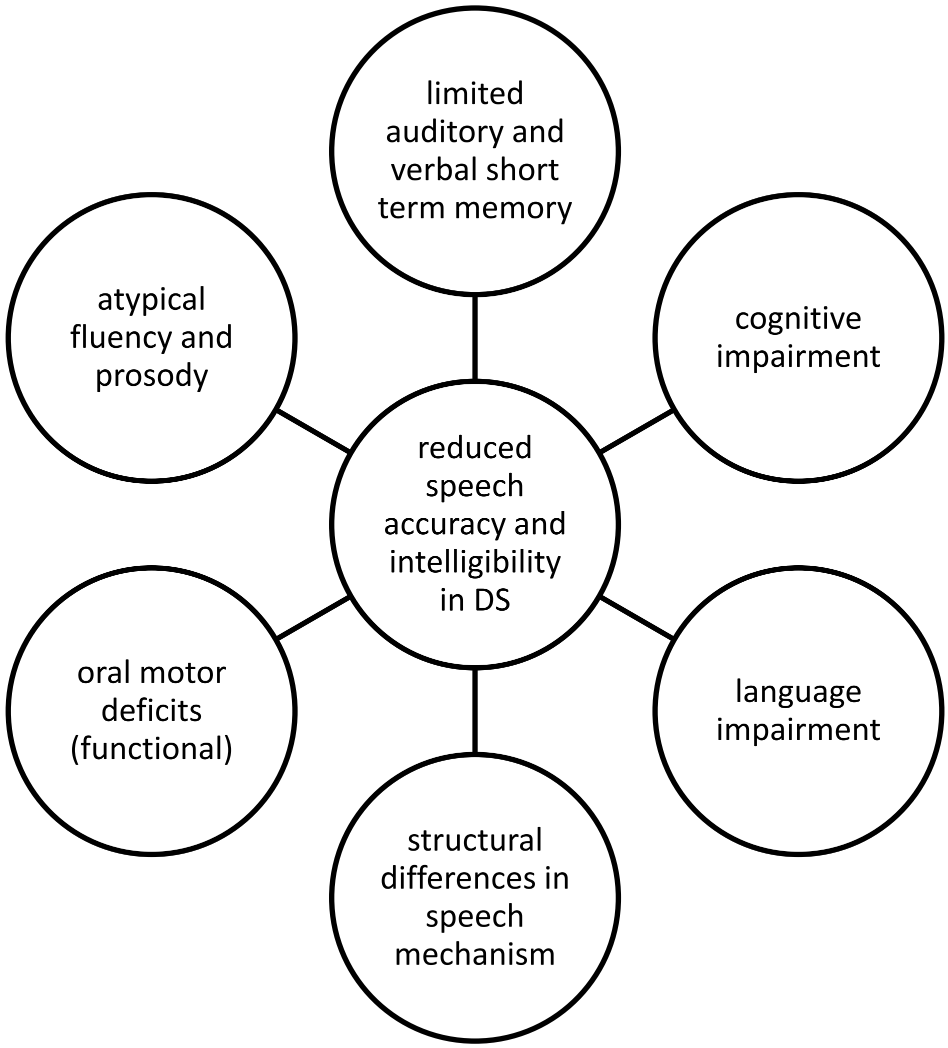

Figure 1. Factors reducing the speech accuracy and intelligibility in Down syndrome.

Deficits Underlying CAS

Increasingly, the speech disorder in DS is being described as a form of Childhood Apraxia of Speech (CAS) that interacts with structural anomalies, dysarthria, and phonological disorder (Cleland et al., 2010; Kumin, 2006; Stoel-Gamon, 1997; Shriberg, Strand & Mabie, 2016, Rupela, Vellman, & Andrianopoulos, 2016). For each case, it is essential to understand the primary challenge at any given developmental juncture in order to design an effective treatment plan (Rupela et al., 2016). Developing an assessment plan that is appropriately targeted at identifying the primary level of breakdown in the individual's communicative system requires an understanding of models of speech motor control.

Numerous computational and psycholinguistic models of speech motor control have been proposed (Baker, Croot, McCleod & Paul, 2001; Stackhouse & Wells, 1993; Shriberg et al., 2012; Popple & Wellington, 1996; Levelt, 2001; Meyer, Huettig, & Levelt 2016; Terband, Maassen, Guenther & Brumberg, 2009). *Figure 2* outlines an amalgam- the speech chain as it is often conceptualized within a psycholinguistic framework, notwithstanding variations in details across specific models. Shriberg et al. (2012) identified four processes necessary for articulate speech including encoding, memory, transcoding, and execution. Each of these stages in speech production encompasses a number of sub-processes, which require the functioning of interconnected speech neural substrates. The functioning of the speech chain is influenced by individual factors such as age, gender, processing speed and level of phonological awareness (Shriberg et al., 2012).

Figure 2. The Speech Chain

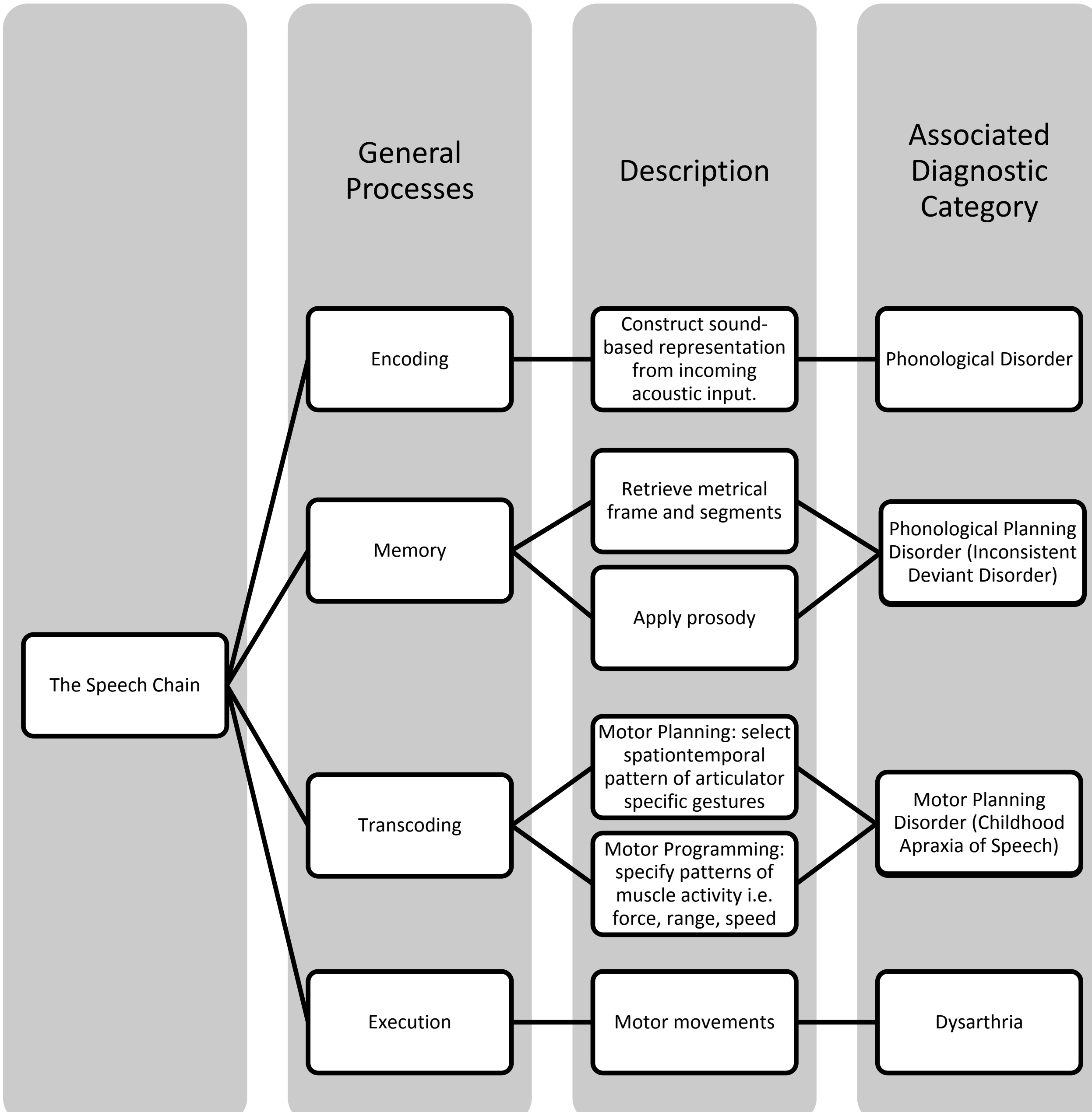

Figure 2. A basic schematic of the speech chain adapted from (Shriberg, Lohmeier, Strand, & Jakielski, 2012; Roelofs, 1997; Dodd, 1995*),* depicting the four major processes involved in speech production, their description, and associated impairments.

According to Shriberg et al., (2012) encoding is a process whereby lexical representations are abstracted from the incoming acoustic signal. Evidence from imaging research links the auditory association areas of the brain, specifically the anterior and middle planum temporale with this stage of the speech chain (Markiewicz & Bohland, 2016; Hickock & Poeppel, 2004). Individuals with encoding deficits present with phonological disorders (Shriberg et al., 2012; Rvachew and Brosseau-Lapré, 2012). For example, an individual with encoding deficits may not attend to the third formant cue in the input signal, and consequently present with a consistent pattern of gliding liquids in their speech output (e.g. 'red licorice' → [wɛd wɪkəwɪʃ]).

Memorial processes involve storage and retrieval of linguistic representations. Both the left supra-marginal gyrus and angular gyrus have been linked to working memory in tasks involving serial recall (Markiewicz & Bohland, 2016). During nonsense syllable repetition tasks, Bohland, Bullock, and Guenther (2010) suggest the pre-supplementary motor area maintains abstract syllable frames, while the left inferior frontal sulcus is responsible for the construction of phoneme sequences (Markiewicz & Bohland, 2016). Phonological memory deficits create problems for lexical access while comprehending speech, and for phonological planning while producing speech. These deficits reveal themselves in the form of lexical and phonemeic paraphasias leading to inconsistent speech errors, such as our observation of 'helicopter' → [ɛtatɚ], [hatdag] and [hʌtatɚ] by a young child with CAS. The research of Shriberg et al., (2012) using the syllable repetition task (SRT) indicates that deficits in the memory stage of the speech chain may contribute to severe speech and language impairments including CAS.

The third stage of the speech chain includes transcoding processes that intervene between the phonological plan and execution of the planned utterance. Whereas Shriberg et al. (2012) proposed a single stage, transcoding, which encompasses both motor planning and programming processes; a similar framework outlined by van der Merwe (2008) outlined two distinct stages, exemplifying the divergent nature of many speech production theories. Ventral pre-supplementary motor cortex activation is responsible for motor planning at the syllable level (Markiewicz & Bohland, 2016). Efficient integration of both sensory and motor information in both the motor and sensory cortices is essential for the functioning of the transcoding process. Speech motor control is dependent upon the development of well-tuned internal models that map the relations between acoustic-phonetic targets, vocal tract shapes, articulatory movements and the somatosensory and auditory consequences of those articulatory gestures (Shiller, Rvachew, & Brosseau-Lapré, 2010). Some models propose that deficits in the processing of sensory feedback explain the primary characteristic of motor planning and programming disorders, specifically disrupted transitions between segments, syllables, and morphemes and poor spatiotemporal coordination of multiple articulators (Terband et al., 2009; Shriberg et al., 2012). Transcoding errors manifest as addition errors, voicing errors, and oral/nasal contrast errors, such as our observation of ‘van’ → [vɑt] by an adolescent with DS.

Frequently children diagnosed with CAS may produce symptoms of both phonological planning disorders and motor planning disorders, though as noted above, the specific underlying deficit may occur at different levels of the speech chain. Despite differences in terminology, both Shriberg et al. (2012) and Ozanne (1995, 2005) agree that the primary deficit in CAS is at the transcoding/ motor planning and programming stage of speech production. Furthermore, there is some agreement that CAS may be accompanied by additional deficits in phonological memory and planning and/or execution.

The final stage of the speech chain, the execution process, relies upon bilateral activation of the primary sensory and motor cortices (Lotze, Seggewies, Erb, Grodd & Birbaumer, 2000). Further, articulation also relies upon appropriate activation of cranial nerves V, VII, IX, X, and XII (Duffy, 2013). Impairment in speech execution, termed dysarthria, may arise from impairment in the central nervous system or the neuromuscular junctions in the peripheral nervous system and may affect speech precision, prosody, fluency, or coordination of speech (Duffy, 2013; Lotze et al., 2000). Within the DS population signs of dysarthria include low range of movement of articulators, abnormal posture or pitch when speaking, hypotonia of oral structures, and distorted speech (Rupela, et al., 2016; Pennington, Miller & Robson, 2010). Rupela et al. (2016) also indicated a number of speech symptoms that are indicative of apraxia and dysarthria, in particular, dysfluency.

Psycholinguistic accounts differ in their conception of interaction between levels of the speech chain. One account postulates discrete impairments, where single or multiple levels of the speech chain may be affected, but do not interact. In other words, lower level processes are not susceptible to higher level errors: each process is discretely activated (Goldrick & Blumstein, 2006). By this view, even when an individual presents with deficits in encoding and motor planning, exemplified by incomplete lexical representations and disrupted syllable transitions, it is assumed that the higher level encoding deficits do not negatively impact subsequent stages of the speech chain.

A contrasting view suggests a cascade effect, where a problem with higher levels of the speech chain overflows onto all lower levels of processing. For example, partial activation of a lexical representation may result in delays in phonological planning, so that the plan contains mis-ordered segments or mis-specified prosody, subsequently resulting in motor planning errors due to poor processing of feedback relative to a poorly specified target. Goldrick & Blumstein (2006) pointed out that a discrete account of speech production errors cannot explain the full range of errors and error patterns in individuals with speech impairments. As noted by van der Merwe (2008), interaction between the stages of speech production is both intricate and essential, given parallel processing and shared neural structures among theoretically discrete processes. Importantly, both the discrete activation account and the cascading activation account allow for errors at multiple levels of speech production (Goldrick & Blumstein, 2006; Goldrick 2006), which is an idea that is congruent with work by Shriberg et al., (2012), van der Merwe (2008) and Ozanne (1995, 2005).

In brief, psycholinguistic and computational models explain the observable surface characteristics by proposing unique impacts of disruptions in each of four important processes involved in the production of speech. In this view CAS is considered a multi-deficit disorder characterized by a primary deficit in transcoding, with possible accompanying impairments in encoding, phonological planning or execution (Ozanne, 1995, 2005; Shriberg et al., 2012; Terband et al., 2009; van der Merwe, 2008).

Evidence for a dual DS-CAS diagnosis. Evidence from multiple sources supports the idea that a subgroup of individuals with DS present with phonological planning and/or motor planning disorders, in possible combination with dysarthria and/or phonological disorder. Typically phonological and motor planning disorders are subsumed under the title of CAS because inconsistent speech errors are a common symptom across these two categories. Research into diadochokinesis characteristics, error patterns and severity, phonological development, and the tractability of speech errors in DS provides the foundation for the conclusion that such a subgroup exists. This section briefly outlines the research in this area.

Data on complex neurodevelopmental disorders and their intersection with motor speech disorders produced by Shriberg, Strand, & Mabie (2016) supports the idea that individuals with DS may present with a variety of motor speech phenotypes. In a study of 50 individuals with DS where the Madison Assessment Battery was administered, 28% of the participants presented with unspecified motor speech disorders, 40% with a primarily dysarthric profile, 10% with a purely CAS profile, and 20% with a mixed dysarthric and CAS profile. In total, Shriberg et al. (2016) estimate a prevalence of 30% of individuals with DS presenting with CAS. This conclusion to varied profiles is congruent with recent findings by Rupela, et al. (2016) who also described a variety of motor speech profiles in the DS population.

Rupela & Manjuela (2010) compared the scores of the diadochokinesis alternate and sequential motion rate tasks between a group with DS, a mental age matched typically developing group, and a non-DS cognitive impairment group. Productions were scored for their rate, consistency, accuracy, and number of attempts required to complete the task (Rupela & Manjuela, 2010). The researchers found that individuals with DS demonstrated significantly lower performance for all tasks compared to the other groups, which suggests a speech profile indicative of childhood apraxia of speech (Rupela & Manjuela, 2010).

In a review of the literature, Stoel-Gamon (1997) cited slow and frequently atypical development, heterogeneity, and supra-segmental errors as the primary differences between the phonological development of people with DS and TD children. She posited hearing loss and structural differences as sources for deficits in articulation, which subsequently result in atypical phonological developmental patterns. However, she also noted the possibility of deficits in speech motor control (Stoel-Gamon, 1997).

Dodd & Thompson (2001) compared individuals with DS whose speech was characterized by inconsistency to a group who also had inconsistent productions but no intellectual disability (specifically, a diagnosis of 'inconsistent phonological disorder'). While the two groups appeared superficially similar in the overall number of errors, closer investigation revealed that the two groups produced different patterns of speech errors (Dodd & Thompson, 2001). They concluded that the children without intellectual disability produced inconsistent phonological errors due to a deficit in phonological planning and memory, accounting for the higher number of phoneme changes within words and the greater range of substitution errors for any given phoneme (Dodd & Thompson, 2001). They further suggested that the inconsistent errors produced by the participants with DS may be due to deficits in encoding processes and incomplete lexical representations, explaining inconsistent within word errors that varied within a constrained range.

Several inconsistencies in Dodd and Thompson's report necessitate considerable caution when interpreting the clinical significance of their findings. First, the groups were poorly matched, specifically with regards to the mean age and PCC scores. The mean ages were 127 months for the DS group, and 55 months for the inconsistent phonological disorder group. In addition, four of the DS participants had PCC scores that were too low to allow for appropriate pairwise matching. The scores from the DS group ranged from PCC 23-82, but the inconsistent phonology group ranged only from PCC 55-89. This demonstrates that though the means between the groups were not significantly different, there was a sizeable difference in the range of scores for each group. Most crucially, oro-motor skills were assessed only for the inconsistent phonological disorders group. Therefore the conclusion that oro-motor skill can be ruled out as an explanation for the differences in speech error patterns between the two groups is invalidated.

Cleland et al., (2010) also pointed out that Dodd & Thompson (2001) did not specifically measure oro-motor skills in their research, and thus the conclusions drawn there are incomplete: motor planning cannot be ruled out as the source of inconsistent production. Cleland et al. (2010) noted in their own research that many of the participants with DS did in fact present with oral-motor difficulty as evidenced by poor diadochokinesis rates, which suggested difficulty in combining and sequencing phonemes. As such, they concluded that structural and muscular anomalies and phonological disorders cannot entirely account for speech deficits in these cases, leaving childhood apraxia of speech as a possible cause. Though a diagnosis of apraxia usually excludes individuals who have dysarthria, a widely accepted list of diagnostic criteria is not decided upon. In rare cases CAS and dysarthria can be concomitant in the same individual (ASHA, 2007). As noted by Bunton, Leddy & Miller (2007), individuals with DS can present with error patterns consistent with dysarthria, apraxia, or both.

Consistent with the ASHA (2007a) definition, vowel errors are widely reported in DS speech literature (Kent & Vorperian., 2013; van Bysterveldt, Gillion & Foster-Cohen, 2010). In a review of the literature, Kent & Vorperian (2013) pointed out that many authors have reported contradictory findings regarding whether the acoustic properties of vowels produced by individuals with DS are different than TD individuals. The authors suggest that some of this disagreement may stem from the temporal variability found in DS speech (Kent & Vorperian, 2013). Temporal variability in itself night be indicative of CAS although it can also be associated with dysarthria and therefore the source of vowel errors in DS is unclear.

Current CAS intervention strategies. The multi-deficit nature of CAS poses a challenge for intervention: children presenting with CAS are often described as difficult to treat, and slow to progress in therapy (Ozanne, 1995, 2005). Maas et al. (2008) suggested that individuals with CAS learn new motor movements slowly, which may be a contributing factor to their protracted progress in therapy.

Treatment for CAS uses the integral stimulation hierarchy as well as the principles of new motor learning to present the target word or sequence at an appropriate level of difficulty (Maas, Gildersleeve-Neumann, Jakielski, & Stoeckel, 2014; Maas et al., 2008; Kaipa & Peterson, 2016; Rvachew & Brosseau-Lapré, 2012). Variables such as massed vs. distributed practice, variable vs. consistent target elicitation, knowledge of results vs. performance feedback, direct vs. summative feedback are all carefully manipulated in an effort to create an effective treatment programme (Maas et al., 2008; Austermann Hula et al., 2008). Further, the inclusion of pre-practice procedures is recommended in treating CAS: this portion of therapy is designed to orient the client to the expectations of the session, and to identify acceptable responses (Maas et al., 2008). High intensity practice has been found to be more effective than therapy with moderate intensity: sessions containing 100-150 trials of target forms resulted in short and long term gains (Edeal & Gildersleeve-Neumann, 2011). Not only did children receiving high intensity therapy learn their targets faster, they also demonstrated greater generalization to untrained words (Edeal & Gildersleeve-Neumann, 2011).

The psycholinguistic framework outlined in the previous section aides in the categorization of underlying deficit or deficits that result in CAS, as well as correctly identify appropriate therapy strategies and materials. Currently, treatment strategies differ depending on the suspected level of underlying deficit. When a deficit at the levels of phonological memory and planning is suspected, treatment focuses on helping the individual to create their own phonological plans-- integrating the demands of intonation, syntax, phonology and morphology into a metrical frame (Rvachew & Brosseau-Lapré, 2012, p.

181). To this end, imitative prompts are avoided, and other cues such as graphics, sign, and text are used to help the individual create stronger linguistic representations (Gordon-Brannon, & Weiss, 2007). This kind of treatment has been shown to increase production accuracy in individuals with Down syndrome (Knight, Kurtz, & Georgiadou, 2015). This therapy is output oriented, but draws on high level phonological representations as well as phonological planning processes. Treatment strategies include both forward and backward chaining, target segmentation, and verbal instructions for articulatory placement (Gordon-Brannon, & Weiss, 2007).

In contrast, therapy for motor planning/transcoding level impairment focuses on auditory motor integration. Stemming from Van Riperian therapy (1978), ear training is central to this therapy method (Rvachew & Brosseau-Lapré, 2012). This therapy is initially input oriented followed by output oriented practice with the focus on helping the individual improve their ability to plan speech movements in time and space. Some research suggests that while individuals with speech sound disorders may be aware of their own inaccurate productions, they have difficulty processing and integrating sensory feedback with motor plans to adjust their productions online and during future attempts (Terband, van Brenk, & van Doornik-van der Zee, 2014). A key feature of this therapy is attention to correct versus incorrect productions of target words. Auditory bombardment and focused stimulation techniques are used to provide access to the correct forms of the target words which serves to establish strong internal models for the target segments/segment sequences (Rvachew & Brosseau-Lapré, 2012). Use of amplification during auditory bombardment and production practice creates an enhanced signal to noise ratio. The auditory motor integration techniques prepare the individual to correctly monitor their own productions during high intensity speech practice (Rvachew & Brosseau-Lapré, 2012).

Objectives

Currently, little is known regarding best practice in speech therapy for the population with concomitant DS and CAS. However, new assessment measures allow us to identify the primary underlying factors with increased confidence. Treatment procedures that are better targeted at the specific needs of individuals with DS and CAS may generate greater improvements in articulatory accuracy with the extended goal of positively impacting speech intelligibility. Therefore, this investigation sought to answer the following question:

> Will individuals with Down syndrome and Childhood Apraxia of Speech respond preferentially to an intervention that targets phonological planning or an intervention that targets motor planning in comparison to a control intervention that focuses solely on high intensity speech practice with no special pre-practice procedures to treat planning deficits?

To answer these questions, three individuals were enrolled in a single subject randomization experiment in which they each received intervention for three different, individually selected, speech goals. For each goal, a set of 5 words (including thematically related pseudowords and real words) were taught in meaningful contexts using a randomly assigned treatment approach, either the control, phonological planning or motor planning intervention. Six intervention sessions were provided for each goal using the assigned intervention with session order determined randomly. Outcomes were assessed using imitation probes constructed from lists of real words containing the taught target sounds and structures. Nonparametric randomization tests were used to determine whether each child responded preferentially to one or more of the three interventions.

Methods

Participants

Participants for these studies were selected according to a number of inclusionary and exclusionary criteria. Inclusionary criteria were as follows: all participants had Down syndrome, and presented with severe speech sound disorders and inconsistent errors according to their school speech-language pathologist. Further, participants and their families agreed to participate in the project for the duration of the treatment sessions and follow-up assessment. Finally, each participant was able to actively engage with the therapist for a 45 minute period. Participants were excluded from this study if: (a) they did not have Down syndrome, (b) they did not present with severe speech impairments, (c) they were unable to attend all treatment and assessment sessions, or (d) they were unable to engage or pay attention for the duration of each 45 minute treatment session.

Participants for this research were recruited by requesting referrals from the local speech-language pathology community, especially those working in schools for students with special needs. Three participants were solicited, and subsequently recruited for this study. The participants were recommended for this study due to the severity and intractability of their speech production deficits.

Characteristics of CAS were confirmed through the following assessments: Syllable Repetition Task (Lohmier & Shriberg, 2008), Diagnostic Evaluation of Articulation and Phonology (Dodd, Zhu, Crosbie, Holm & Ozanne, 2002), in particular the Word Inconsistency Assessment, and low PCC scores. Additional assessments, as described in Table 1, were conducted to confirm compliance with the inclusion criteria and complete the characterization of the participant's underlying psycholinguistic profile.

Table 1

Pre-treatment assessments to identify presence of CAS and eligibility for treatment

Assessment Name	Assessment Details
Kaufman Brief Intelligence Test II (KBIT-2) (Kaufman & Kaufman, 1997)	• A norm referenced measure of verbal and non-verbal intelligence. • Norms for ages 4;00-90;00.
Diagnostic Evaluation of Articulation and Phonology (DEAP) (Dodd, Zhu, Crosbie, Holm, & Ozanne, 2002)	• A norm referenced test battery designed to differentially diagnose articulation disorders from phonological disorders. • Includes five subtests: a Diagnostic Screen, Oral Motor Screen*, Articulation Assessment, Phonology Assessment and Word Inconsistency Assessment* • Norms for ages 3;00 -8;11.
Peabody Picture Vocabulary Test III, IV (PPVT -3, -4) (Dunn, Dunn, & American Guidance Service, 1997)	• A norm referenced measure of receptive vocabulary ability. • Norms available for 2;06-90;00.
Free Speech Sample* (FSS)	• Uses the book Flotsam (Wiesner, 2006) or the book Good Dog Carl (Day, 1989) to elicit a speech sample from the participant. • Measures Percent Consonants Correct (PCC), Percent Vowels Correct (PVC), and Mean Length of Utterance (MLU) in a natural setting.
Syllable Repetition Task (SRT) (Lohmeier & Shriberg, 2008; Shriberg et al., 2011)	• An 18 item computer administered non-word repetition task using early developing consonants /m,b,n,d/ and the vowel /ɑ/. • Identifies encoding, memorial and transcoding deficits. • Norms for age 3-17, scores matched to receptive language age as determined by PPVT scores
Maximum Performance Tasks MPT* (Rvachew, Hodge, Ohberg,2005; Kent et. al. 1987; Fletcher, 1972; Robbins & Klee, 1987)	• A norm referenced assessment of speech motor control targeting maximum phonation duration, fricative duration, single syllable repetition rate and multisyllable repetition rate. • Uses the mean data from a number of tasks to differentially diagnosis of CAS and dysarthria. • Norms for 6;00-13;00 (Fletcher, 1972); 2;06-6;11 (Robbins & Klee, 1987)
Phonological Awareness Test (Bird, Bishop, Freeman, 1995; Rvachew & Grawburg, 2006 Rvachew, 2007)	• A norm referenced test of implicit phonological awareness with norms for prekindergarten, kindergarten, and first grade. • Comparisons are made with cognitive age from KBIT-2
Speech Assessment and Interactive Learning System II (SAILS-2) (Rvachew S. , SAILS: Speech Assessment and Interactive Learning System, 2014)	• A computer based norm referenced measure of aural speech perception. • Examinee listens to a word over headphones and makes a judgement as to whether they heard X or Y. • Norms for ages 4;06-7;06

Specifically, one or more of the following symptoms was identified in each participant's' speech: (1) transcoding or memory errors during SRT (Lohmier & Shriberg, 2008) performance; (2) difficulties with prosody and/or transitioning between segments and syllables in connected speech, as observed via acoustic analysis of the free speech sample; or (3) inconsistent errors on the DEAP Word Inconsistency Assessment. Thus, the symptomology of each participant was aligned with the CAS characteristics identified by ASHA (2007a). Information about the assessments was provided to each family before assessing the potential participant, and each family provided their informed consent to participate in the study.

Assessments and participant profiles. Table 1 presents the assessments that were administered to each potential participant to determine whether they were eligible for the study, and to delineate a cognitive profile for each participant. Tests marked with * indicate assessments that were considered in determining if the participant was eligible for the study. Given that the diagnosis of DS is indicative of a cognitive delay, the age equivalence data obtained from the KBIT-2 (Kaufman & Kaufman, 1997) was used in the selection of appropriate norms for the rest of the tests when applicable. As such, these test scores were non-standardized, and were therefore only used as a guide in decision making.

The cognitive and linguistic profiles of each study participant are outlined in Table 2. The test scores were obtained from the pre-treatment assessments. Each participant was given a participant code to maintain their anonymity throughout the treatment and analysis.

Table 2
Participant profiles and pre-treatment assessment results

		Participant		
Assessment title	Assessment Score	TASC DS 35	TASC DS 37	TASC DS 38
Participant Demographic Information	Age	15;08	20;03	10;01
	School attendance	Full time	Full time	Full time
	Hearing status	OK	OK	Fluctuating Loss
	Fluency status	Stuttering	Stuttering	OK
Kaufman Brief Intelligence Test II (KBIT-2) (Kaufman & Kaufman, 1997)	Verbal IQ standard score	40	40	40
	Percentile Rank	<0.1	<0.1	<0.1
	Non-verbal IQ standard score	58	44	57
	Percentile Rank	0.3	<0.1	0.2
	Significant verbal/non-verbal gap	Yes	No	Yes
Peabody Picture Vocabulary Test III, IV (PPVT -3, -4) (Dunn, Dunn, & American Guidance Service, 1997)	Standard Score	40	21	Did not achieve baseline
	Percentile Rank	<0.1	<0.1	
	Age Equivalent	3;06	4;04	
Free Speech Sample* (FSS)	MLU	2.0	3.58	1.98
	Percent Consonants Correct	42%	71%	38%
	Percent Vowels Correct	67%	92%	74%
Diagnostic Evaluation of Articulation and Phonology (DEAP) (Dodd, Zhu, Crosbie, Holm, & Ozanne, 2002)	Phonology Error Score	52	29	73
	Articulation Error Score	61	17	54
	Percent Consonants Correct	22%	79%	29%
	Percent Vowels Correct	75%	92%	68%
	Percent Phonemes Correct	41%	83%	42%
	Oral Motor Score	30	36	Refused
	Word Inconsistency Assessment (WI) Score	52%	56%	84%

Table 2 contd.
Participant profiles and pre-treatment assessment results

		Participant		
Assessment Title	Assessment Score	TASC DS 35	TASC DS 37	TASC DS 38
Syllable Repetition Task (SRT)* (Lohmeier & Shriberg, 2008; Shriberg et al., 2011)	Competency Score	26.00	54.35	34
	Z score	-2.02	-1.58	-1.60
	Encoding Score	33.33	85.71	29.41
	Z score	-0.69	1.12	-1.02
	Memory Score	47.68	79.52	47.68
	Z score	1.09	0.33	-0.17
	Transcoding Score	61.11	0.0	22.22
	Additions	63	0	6
	Z score	-2.45	0.27	-0.51
Maximum Performance Tasks MPT* (Rvachew, Hodge, Ohberg, 2005; Kent et al., 1987; Fletcher, 1972; Robbins & Klee, 1987)	Max Phonation Duration	6.35 sec	12.99 sec	Refused to participate
	Max Frication Duration	-----	7.64 sec	
	Max Repetition Rate (Monosyllabic reps/second)	3.85	4.22	
	Max Repetition Rate (Trisyllabic reps/second)	-----	-----	
Phonological Awareness Test (Bird, Bishop, Freeman, 1995; Rvachew & Grawburg, 2006; Rvachew, 2007)	Rime Matching Score	29%	28%	Refused to participate
	Onset Matching Score	10%	30%	
	Segmentation & Matching	30%	20%	
	Total Score	21%	27%	
	Z score	-3.14	-2.70	
Speech Assessment and Interactive Learning System II (SAILS-2) (Rvachew S., SAILS: Speech Assessment and Interactive Learning System, 2014)	R- Score mean	50%	50%	Refused to participate
	C- Score mean	50%	60%	
	L- Score mean	80%	50%	
	S- Score mean	60%	36.66%	

---- indicates that the participant was engaged, but was unable to complete the task

TASC DS 35 assessment profile. TASC DS 35 was a male adolescent with a history of multiple therapies including occupational therapy, massage therapy, physiotherapy, and speech-language therapy. His speech dysfluency was not measured in detail in this study, though it was regularly observed. At the time of the study he attended a “minor” speech-language therapy group at school. His mother reported that TASC DS 35 occasionally used an AAC device at school because his poor intelligibility caused him to become frustrated with communication. A history of P-E tubes, and difficulties with swallowing and drooling was reported by his mother in his case history form. As indicated by his results on the PPVT-3, the participant demonstrated a severe delay in receptive language. He produced little spontaneous output during the free speech sample apart from labeling the items he saw in the pictures of Good Dog Carl (Day, 1989), along with some prescribed phrases such as “all about___”. His labeling was frequently accompanied by rudimentary signs, and this greatly increased his ability to be understood in this context.

In the pre-treatment assessment TASC DS 35 presented with extremely low speech intelligibility marked by numerous articulation errors on both vowels and consonants, as well as consonant and syllable harmony. For example, the participant produced ‘shark’ /ʃa˞k/ → [ka˞k], ‘zebra’ /zibɹə/ → [łæłæ]. When words of three or more syllables were elicited in the DEAP WI test and SRT, the participant frequently inserted numerous syllables indicating increased phonological planning deficits for longer words, for example, ‘umbrella’ /ʌmbɹɛlə/ → [ʌbələlədidə], [ʌmbəbəbəłæłə]. The participant also demonstrated a great deal of inconsistency in his speech productions across assessments, characterized by oral/nasal errors, voicing and vowel errors which suggested disrupted transitions between segments and syllables, for instance, ‘jump’ /d͡ʒʌmp/ → [map], [bap] and poor coordination of multiple articulators. These speech error patterns, in combination with addition errors, suggested the presence of both phonological and motor planning disorders as outlined in the psycholinguistic framework reviewed in the previous section.

TASC DS 35's profile was suggestive of a primary motor planning impairment with additional encoding, phonological planning, dysfluency, and dysarthria. A motor planning (transcoding) deficit was confirmed by the SRT test on which he produced 63 addition errors and his MPT performance, during which he was unable to sequence [pataka] despite achieving reasonable single syllable repetition rates. On the SRT, his phonological memory score was within normal limits because his score for two and three syllable items was similar (a low memory score reflects significantly poorer performance as item lengths increase). Yet, it is difficult to actually interpret the memory score because his performance was so poor at the shortest item length, i.e., two syllable nonsense words. As previously indicated, his performance on the Word Inconsistency Assessment suggested difficulties at both the phonological and motor planning levels, but overall the impression is of a primary deficit in motor planning. Therefore it was hypothesized that the participant would benefit from a treatment targeting motor planning by focusing on auditory-motor integration.

TASC DS 37 assessment profile. TASC DS 37 was a male with a history of treatment for speech, namely fluency treatment, though he was not longer in treatment. His parents noted difficulties with fine motor skills and feeding and drooling issues in childhood, but not adolescence. At the time of the study, he attended group activities focusing on the use of the iPad during school. TASC DS 37 was able to produce full sentences spontaneously, and describe pictures with reference to objects and actions during the free speech sample. This participant presented with significant difficulty in coordinating subject verb agreement, as well as choosing the appropriate articles, and struggled to use appropriate morphemes in spontaneous speech. For example, the participant produced “well, is what happens the waves” when describing a picture.

A number of articulation impairments were observed for TASC DS 37 which were markedly more severe in connected speech. This participant presented with increased difficulty with multisyllabic words, voicing errors, and vowel distortions, for example ‘zebra’ /zibɹə/→ [sibɹə], ‘television’ /tɛləvɪʒən/→ [tʌwɪʃən] suggesting a mixed phonological planning and motor planning impairment. He demonstrated a great deal of inconsistency in his speech regardless of word length, for example ‘tongue’ /tʌŋ/→ [tɔn], [tʌŋ], [tʌn]. TASC DS 37 presented with a low competency on the SRT task, but did not present with transcoding errors (i.e., additions). Rather, he performed markedly worse on the four syllable items than the three syllable items which emphasized deficits in phonological memory. This decline in performance was not reflected in his memory test score that compares two syllable items and the three syllable items. This combination of error types suggested that the participant had wide spread impairments across phonological planning and motor planning processes of speech production.

The participant’s scores on the maximum performance task were indicative of planning deficits rather than dysarthria: he was able to produce monosyllabic sequences, but not the trisyllabic [pataka] sequence. Based upon the results of his SRT, MPT and WI tests, the overall impression for this participant is that his primary deficit is at the level of phonological planning, given within word inconsistency, the prominence of vowel errors, difficulties with increased utterance length and inability to sequence multisyllables but the absence of transcoding errors. Therefore, it was hypothesized that the participant would benefit from a treatment designed for disorders in phonological planning, though it was unclear whether or not the participant would also benefit from a motor planning treatment.

TASC DS 38 assessment profile. TASC DS 38 was a male whose case history form indicated conductive hearing loss ranging from mild-moderate-severe, which was managed with bone anchored hearing aids (BAHA). The participant had numerous medical issues with his BAHAs, and his use of the devices was intermittent. His parents also indicated issues with oral control, namely the inability to spit, but frequent drooling. Issues with pocketing food were observed informally during assessment, indicating low oral control and hyposensitivity of the oral structures. TASC DS 38 was receiving language therapy, physiotherapy, and occupational therapy at school.

Throughout the study TASC DS 38 presented with low intrinsic motivation, and was reluctant to participate in assessments or therapy sessions. Behaviour difficulties included non-compliance, minimal responsiveness, emotional lability, and consistent under-performance. As a result, many assessments cannot be considered valid as they did not reflect his true capability. This participant produced little spontaneous output, aside from refusals and requests for food, electronic games, and break time. However, he did label objects during the free speech sample when prompted.

TASC DS 38's scores on the SRT indicated widespread impairments in encoding, memory, and transcoding: age 3 norms were used as the participant did not achieve a PPVT-4 baseline. The low competency and encoding scores on the SRT were indicative of a phonological disorder, not surprising given his hearing loss and early stage of language development. The memory score is not low because his performance was poor at all item lengths. Further, the number of additions present in the SRT task signalled a motor planning impairment (the z score is not low because the standard deviations are very high in the 3-year-old norms; however our experience indicates that 4 or more additions at any age is cause for concern). The combination of hearing loss, lack of participation, and widespread deficits precluded the construction of a specific hypothesis about which treatment condition would be most appropriate for this participant. However, given the severity of the participant's impairments the participant was included in the study with the hope that high intensity therapy would be beneficial.

The issue of participant age. A systematic review of the changing nature of cognition in individuals with DS concluded that the timeline of cognitive development seen in DS is unique to this condition (Patterson et al., 2013). Specifically, current research does not clearly define periods of cognitive development or decline in this population which would negate the validity of this study based upon the ages of the participants. However, Patterson et al. (2013) did stipulate in their review that an appropriate control measure must be included in research to account for age effects versus treatment effects. The parameters of the control condition are outlined in the study overview section.

Goal selection. The speech goals and target words selected for each participant were chosen according to their individual profile of errors from the intake assessments. A quick multilinear analysis (Rvachew & Brosseau-Lapré, 2012) was performed on the data collected from the DEAP Phonology and Articulation subtests (Dodd et al., 2002) and the free speech sample in order to identify potential speech goals at three potential levels: (1) prosodic level, such as specific stress patterns or word shapes (e.g., W-s-W words, CCVC words); (2) feature contrasts, such as glides vs. liquids, voiced vs. voiceless, or coronal vs. dorsal; and (3) segments, such as /f/. Individual speech goals were assigned to one of three thematic routines: the thematic routines provided a meaningful context through which the target words were instructed. A list of goals for each participant is provided below.

Table 3
TASC DS 35 speech goals, routines, and target words

Monster Routine	**Garden Routine**	**Dog Routine**
Goal: suppress vowel and consonant harmony in CVCVC and CVCCVC words	Goal: establish major sound class contrasts glide vs. stop vs. liquid	Increase consistency in production of /f/ while reducing confusion with /p/ across syllable positions
Target Words: tamdow, boonday, downup, pogtog, peanut	Target words: weet, loot, dirt, water, yod	Target Words: Puff, Fap, Fooey, Woof, Food

Table 4
TASC DS 37 speech goals, routines, and target words

Monsters Routine	**Wizards Routine**	**Fish Routine**
Goal: 3 syllable words with strong-weak-strong stress pattern	Goal: word/phrase internal abutting consonants	Goal: voicing contrast in the onset position
Target Words: Bobbytak, Goobertoom, meediking, nubiting, wonderful	Target words: Pog wizard, Tam Wizard, Pog team, Tam team, Darklord	Target Words: zander, zingle, zug, jig, sea

Table 5
TASC DS 38 speech goals, routines, and target words

Monsters Routine	**Dogs Routine**	**Clowns Routine**
Goal: $C_1V_1C_2V_2$ trochee, contrasting manner at the labial place of articulation (suppress consonant place harmony)	Goal: Establish major manner classes in onset of CVC words, specifically: C_1VC_2 C_1= /n,j,r,d/, C_2=S	Goal: /f/ in a CV(r) syllable
Target Words: Meewa, Wobey, meebing, wamming, happy	Target Words: Yos, Ness, doos, rice, nice	Target Words: Foo, Fay, fa(ll), fou(r), fa(r)

The target words themselves consisted of phonemes or syllable structures for which the participant was stimulable. Further, the target words did not have more syllables than the participant produced meaningfully in spontaneous speech. All three treatment conditions used a mix pseudowords and real words as targets, assigned to semantically related concepts (for example, the names of two monsters and their favourite activities). This was done to minimize previous experience or learning effects while allowing for the practice of words in meaningful contexts. All target words complied with the phonotactic constraints of Canadian English. Speech goals and target words were selected without knowledge of the participants' psycholinguistic profile or the hypotheses for treatment response.

The speech goals for TASC DS 35 were as follows: (1) at the prosodic level, reduction of consonant place harmony across 2 syllable words and phrases; (2) feature level, establish major sound class contrasts, as this participant demonstrated confusion across sound classes in his initial assessment; and (3) segment level, accurate production of /f/, a phoneme for which the participant was stimulable although he did not use it correctly in context.

Goals for TASC DS 37 were as follows: (1) prosodic level, establish consistent production of the strong-weak-strong stress pattern, as this was only intermittent during the initial assessment; (2) word shape level, maintain syllable codas and onsets in a word -or phrase-internal abutting context, as the participant was often observed to omit one or more of the consonants in this context; and (3) feature goal, the voicing contrast for /z/ vs. /s/ and /t͡ʃ / vs. /d͡ʒ/.

Due to TASC DS 38's hesitancy to participate and his limited phonemic repertoire, uncomplicated goals and target words were assigned, as follows: (1) prosodic level target, establish two syllable words with a trochaic stress pattern while reducing consonant harmony; (2) word shape level, establish major sound classes by practicing nasals, liquids, glides and stops in the onset of words that have a fricative in the coda position; and (3) segmental level, establish /f/ for which the participant was stimulable but did not use correctly in any context.

Experimental conditions. Effective treatment should include high intensity practice and also an element of pre-practice wherein the individual is oriented to the task and the clinician's expectations regarding acceptable productions (Maas et al., 2008). In this research we applied experimental conditions to the pre-practice portion of treatment sessions, while maintaining roughly uniform duration, intensity, and structure for the practice portion. We compared two experimental conditions-- phonological memory and planning (PMP) pre-practice (targeting phonological planning deficits) and auditory motor integration (AMI) pre-practice (targeting motor planning deficits), to a control condition in which no pre-practice was done. As outlined in Table 6, the pre-practice procedures for each experimental condition were compiled to address disparate deficits in the speech motor system while still conforming to the broader goals of pre-practice (i.e. task orientation, and provision of expectations).

As discussed above, current treatment in CAS aims to remediate the underlying deficits in the speech chain: for example, treatment for an individual with a primary impairment in motor planning should strengthen that individual's ability to integrate auditory and motor signals. Therefore, the motor planning intervention in this study included four auditory-motor integration procedures specifically targeting knowledge of the target and processing of auditory feedback while speaking, that were implemented during the prepractice half of each treatement session, as described in Table 6. During the practice portion of the session, a strong emphasis was placed on providing auditory-visual models prior to the participant's production attempt, and on delaying feedback after the attempt so that the participant had an opportunity to process, integrate and respond to self-produced feedback.

In contrast, a treatment targeting phonological planning deficits should strengthen the individual's knowledge of the phonological structure of the target and encourage the learner to produce and implement their own phonological plan for target words. Therefore, the phonological planning intervention in this study used procedures such as segmentation and chaining, again implemented during the prepractice half of treatment sessions, to help the participant identify the segments and segment order for each target, as described in Table 6. During the practice portion of the session, visual prompts were used when necessary but imitative models were avoided as a means of stimulating production practice of the target words.

High intensity practice was applied across after the pre-practice section was completed (further details about the structure of sessions are provided in the *Study Design* section). The integral stimulation hierarchy (Maas et al., 2014; Rvachew and Brosseau-Lapré, 2012) was used in the practice portion of the PMP and AMI sessions to maintain practice at the challenge point for each participant. Rvachew and Brosseau-Lapré (2012) posit that the appropriate challenge point is 80% correct responding. If the learner is practicing above or below the challenge point, learning does not occur. For example, if the participant is achieving 100% correct responses, performance during the session is high but this kind of practice does not permit generalization to the extra-clinic environment or to similar targets because there is no opportunity to practice adapting motor plans to changing circumstances. Maintaining practice at the challenge point requires that the SLP monitor the learner's performance from trial to trial and adjust pretrial stimulation, the difficulty of the target, or post-trial feedback as needed.

The control condition differed from the two experimental conditions in two important ways. First there were no prepractice procedures. Second, the high intensity practice procedures were implemented in a uniform fashion rather than adjusted on a trial-by-trial basis to ensure challenge point performance. For example, in the two experimental conditions the frequency of feedback was manipulated depending upon the amount of support that the participant needed to practice the target word at the challenge point; therefore, if the participant was achieving a high level of correctness feedback frequency was reduced but if the participant was struggling to achieve correct performance, feedback frequency was increased. In comparison, the feedback frequency in the control conditions was held constant at a rate of 60% of all trials in congruence with the suggestion by Austermann Hula, Robin, Maas, Ballard & Schmidt (2008) that increased external feedback precludes learning of the core features of a movement pattern. A similar intensity of practice was expected in all three conditions however (at least 100 practice trials per 20 minute practice session).

The speech goals and their selected thematic routines were randomly assigned to experimental conditions using list randomizer software at www.randomizer.org to reorder the goals independently for each participant, resulting in the assignments shown in *Figure 3*. Again, this process was completed with no knowledge of the participant's psycholinguistic profile.

Table 6

Experimental Conditions and their Description

Experimental Condition	Pre-Practice Procedures	High Intensity Practice Procedures
Auditory Motor Integration	• Auditory bombardment: SLP produces stories containing many exemplars of the target, using props and a meaningful context, to ensure that the participant has a rich acoustic-phonetic representation of the target words. • Target detection: participant learns to associate SLP's production of the target with a motor response. • Error-detection tasks: participant produces distinct motor responses to identify SLP and self-produced versions of target vs misarticulated target, therefore learning to self-monitor and self-correct mismatches between produced speech and the target. • Focused stimulation: SLP plans activities that encourage the participant to produce the target words while providing opportunities to highlight mismatches or matches between the participant's output attempts and the acoustic-phonetic characteristics of the target.	• Practice in a drill and/or drill-play context provides opportunities to practice selecting and executing the motor plans required to produce the target words. • Imitative cues are provided as needed. • Delayed feedback about results is preferred so that the participant is encouraged to evaluate his/her own responses. • Use integral stimulation hierarchy and principles of motor learning to ensure that a challenge point of 80% correct trials is maintained.
Phonological Memory and Planning	• Target words are segmented into phonemes and syllables and the participant is taught to pair the segments with visual cues. • The participant learns to produce the individual segments in the target words, with articulatory placement and verbal instructions provided as need, and visual cues used to help the participant recall the plan. • Subsequently forward and backward chaining of the phoneme sequences is used to teach the target words. • Imitative cues are provided at the segment and syllable levels for the purpose of teaching, but avoided at the word level.	• Practice in a drill and/or drill-play context provides opportunities to practice constructing and implementing a phonological plan. • Visual cues (print or sign), but not imitative verbal cues, may be provided so that the participant is obliged to create their own phonological plan. • Use integral stimulation hierarchy and the principles of motor learning to ensure that a challenge point of 80% correct trials is maintained.
Control	• No pre-practice: games or other assessments were completed during this time.	• Practice in a drill and/or drill-play context to teach accurate production of the target words. • Target complexity and variability within blocks of trials was increased gradually according to a predetermined schedule • Pre-trial stimulation was gradually reduced according to principles of motor learning • Feedback was restricted to occur on 60% of trials.

Figure 3. The Randomization of Speech Goals to Experimental Conditions.

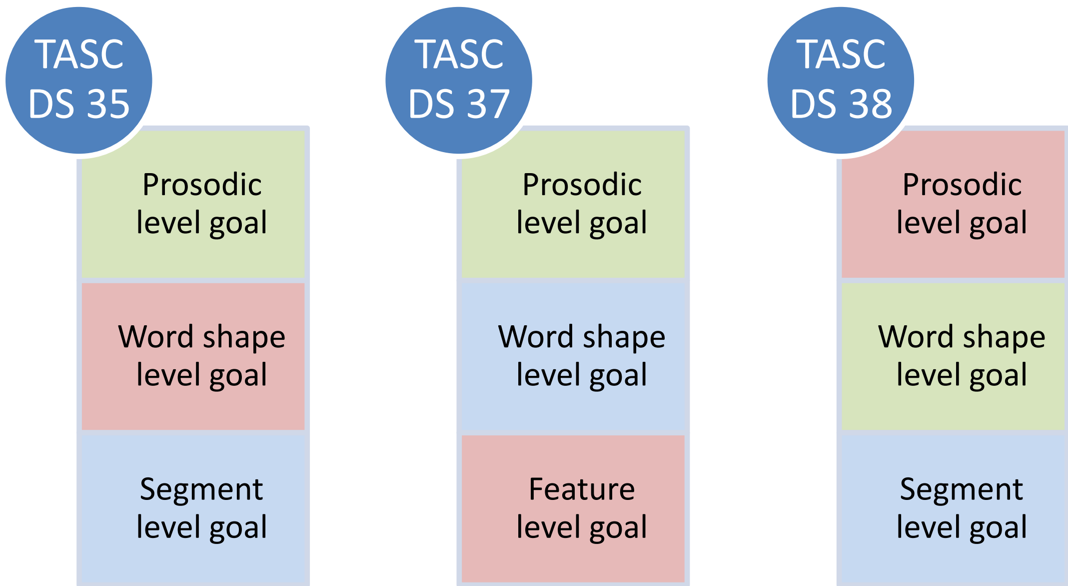

Figure 3: Speech goals were randomly assigned to experimental conditions: red (☐) indicates assignment to the auditory motor integration (AMI) condition, blue (☐) indicates assignment to the control condition, and green colouring (☐) indicates assignment to the phonological memory and planning (PMP) condition.

Study Overview

Study design. This study used a single-subject block randomized alternation design, replicated across three participants, within a pre-test-treatment-post-test framework. In other words, this study was designed to measure within-participant change. The customizability of this design was an advantage when considering the rarity of this combination of clinical characteristics, and the substantial heterogeneity within this small population (Onghena & Edgington, 2005). In addition, n-of-1 trials with alternating design and randomization eliminate selection, maturation, and history effects, yielding data with high internal validity (Rvachew, 1988; Rvachew & Matthews, Submitted).

This research design is sensitive to treatment effects within the individual as they serve as their own control through exposure to all treatment levels (Onghena & Edgington, 2005). In this way it allows for the identification of the most effective treatment for the individual while maintaining rigorous experimental controls (Ledford et al., 2016) The design is also well suited to individuals who do not present with a stable baseline, such as the participants in this particular series of research experiments (Rvachew & Matthews, submitted; Edeal & Gildersleeve-Neumann, 2011; Smith, 2012)

Single-subject block randomized alternation design allows for repeated observation points for each treatment condition as found in a time series study. In this case, three treatment conditions were provided within each block with random assignment of condition to session within blocks (see Appendix A for a depiction). Nonparametric resampling tests were used to compare within-participant outcomes on the dependent variables across conditions. There were two dependent variables for each participant: Same Day Probes that assessed performance on the targeted treatment goal at the end of each treatment session; and, Next Day Probes that assessed maintenance of learning to the beginning of the next session. Both of these probes assessed transfer from treated words/pseudo-words to untreated real words.

This research study consisted of eighteen sessions over the course of six to nine weeks, equivalent to two or three sessions per week depending on the participant's scheduling constraints. Before the treatment sessions begin, each participant underwent a pre-treatment assessment, and a follow-up assessment took place approximately two to three months after the final session. The general schedule is depicted in *Figure 4*.

Session Structure*:* The duration of each session was forty-five minutes in length: within each session Next Day probes, pre-practice, practice, and Same Day Probes were completed. The timeline of a single session is outlined graphically in Figure 5. Participants were given a visual schedule of each session, and their progress through each section was shown with stickers or by crossing out the task. Each session provided two sampling opportunities to measure performance and maintenance through the Same Day Probes and Next Day Probes, for a total of 36 samples from each participant.

Next Day Probes measured short term maintenance and learning from the previous session in a closed context through ten imitated phrases. The pre-practice section was approximately 15 minutes in length and applied one of the experimental treatment conditions, which were outlined in the previous section. Within the pre-practice portion, the principles of motor learning and the integral stimulation hierarchy informed the adaptations that were made online to the presentation, instruction, and elicitation of the target words assigned to the PMP and AMI conditions.

Figure 4. Study Schedule Outline

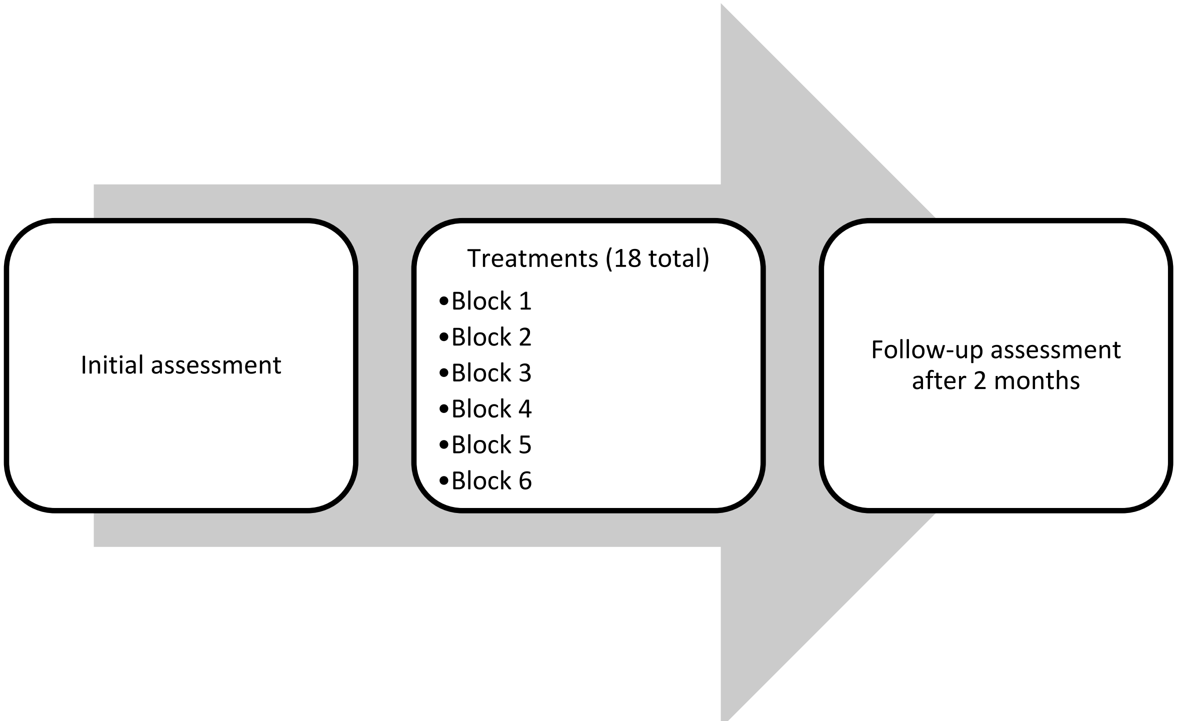

Figure 4. A general outline depicting the progression of each individual through the study from intake assessment to treatments to follow-up assessment.

Figure 5. Session Timeline

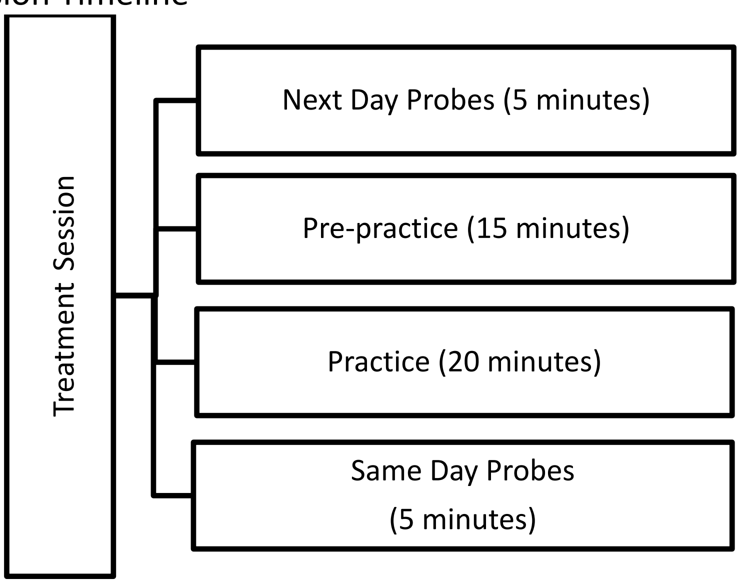

Figure 5. Each session is divided into four major sections.

Following pre-practice, 20 minutes of high intensity practice was completed in which the participant was expected to complete a minimum of 100 trials of the target sounds/words. The principles of motor learning were applied in the practice portion of the treatment sessions: specifically, the complexity of the elicited response, the type and amount of feedback provided were adjusted according to the participant's performance. The integral stimulation hierarchy was also used to maintain an appropriate challenge point.

Within the practice section, a set referred to a group of five trials: Within these sets, targets might be practice in blocks or according to a variable schedule depending upon the participant's performance (e.g., dirt, dirt, dirt, dirt, dirt when the performance was low; weet, loot, dirt, water, yod when performance was high), since Neel, Boyd, Carrol, and Sanchez (2015) found that greater generalization results from variable practice. The amount of stimulation provided before each trial was also manipulated as a means of maintaining an appropriate level of challenge for the participant with an effort made to progress from imitative to spontaneous responses as treatment progressed. The type and amount of feedback were also manipulated as a function of the participant's performance. As stipulated by Rvachew and Brosseau-Lapré (2012), feedback focused on accuracy of specific speech movements (knowledge of performance feedback) when performance was below challenge point and on the overall accuracy of trials (knowledge of results feedback) when performance was at or above challenge point. Finally, Same Day Probes concluded the session, as a measurement of session performance: this probe, too, was imitative consisting of 10 short phrases.

Outcome measures. These research studies included four outcome measures to determine the extent of the change in participant performance and learning, and maintenance of those changes over time. The first measure analyzed performance through trial-by-trial treatment data recorded from each session. The second outcome measure assessed transfer, specifically the transfer of skills learned from the target words to untreated words. This was measured through the Same Day Probes. The third outcome measure assessed maintenance in the short term, or how well the transference of skills to untreated words was maintained from one session to the next. This outcome measure was captured through the Next Day Probe data. Finally, long term maintenance of change was assessed through the two month follow-up assessment. The follow-up assessment consisted of a repeat demonstration of the DEAP articulation test and word inconsistency test, free speech sample, and a complete set of probe phrases from each of the experimental conditions.

General Procedures.

This series of experiments was carried out at the School of Communication Sciences and Disorders at McGill University. Assessments and treatment were carried out by student clinicians in the Speech-Language Pathology Master's degree program who were supervised by an experienced clinician. Goals were determined by agreement among the student clinicians, the clinical supervisor and the lab director. Randomization of treatment conditions to speech goals was carried out by the lab director with the online software random.org/lists. Time stamps provided by the software were used as a record of the randomization process. Subsequent randomization of treatment sessions within blocks was done with the randomizer.org software by the lab director.

Videos were coded in their entirety for the following information: (1) number of trials; (2) trials correct; (3) level of the integral stimulation hierarchy used by the clinician; (4) the level of complexity of the target elicited; (5) whether the trials elicited were uniform or varied in each set; (6) the type of feedback provided by the clinician; and (7) the amount of feedback provided. I coded the videos of each session across experiments: 10% of the videos were also coded by a paid research assistant who selected the videos at random.

The procedures used by the student clinicians for the AMI or PMP protocols were also logged during video coding as a measure of treatment fidelity which was calculated by scoring the observed protocols for each pre-practice condition against the expected protocols. For the AMI condition the expected protocols were: (1) auditory bombardment; (2) focused stimulation; (3) target detection; (4) error detection; and (5) self-monitoring of the participant's own productions. For the PMP condition the expected protocols were: (1) segmentation of the target words into syllables and individual phonemes; (2) chaining of segments and syllables to systematically build a phonological plan; (3) multisensory prompts including tactile stimulation as needed; (4) visuals supports; and (5) phonetic placement instructions. The control condition did not have pre-practice procedures, thus treatment fidelity was not calculated for these sessions.

Transcription of the probes was done by a paid research assistant who was blind to the assessment results, speech goals, the assignment of treatment conditions, and probe scoring criteria. I also performed transcription reliability coding on a minimum of 10% of probes for each participant. In order to be transcribed and subjected to inter-rater scoring and agreement measures, each production was required to meet the standards for production of a canonical syllable. These criteria include a range of no more than 30dB within a single syllable, full oral resonance for vowels, syllable duration of 500 ms or less, and a transition from consonant to vowel lasting between 25 and 120ms (Rvachew & Brosseau-Lapré, 2012). Vocalizations that did not comply with this criteria, singleton nasals, whispers, and yells were not included in inter-rater agreement measures. After transcription was complete, any discrepancies between the two transcribers were rectified by mutual agreement.

The lab director and I scored the transcribed probes separately without reference to the assigned treatment conditions or the participant's psycholinguistic profile. Criteria for scoring were discussed and agreed upon by both scorers. In subsequent comparisons of the scores, any discrepancies were discussed and resolved given the agreed upon criteria. Probe scoring criteria is reported in Appendix C. This scoring procedure resulted in a score for each probe out of a maximum score of 10 points. For each participant and each probe type (i.e., Same Day probe, Next Day probe) there are 6 probes for each condition and three conditions for a maximum of 18 probe scores.

Analysis tools. The sessions were video recorded with a Sony HDR-XR150 camera mounted on a tripod. The video camera was operated by a volunteer who was present in the treatment room, at a distance of approximately 1.5 meters from the participant. The camera was zoomed in to capture the participant's head and torso on the screen. Video coding logs were created using Microsoft Excel 2010.

The probes were also audio recorded with a Zoom H2 Recorder. The Zoom H2 recorder was kept at a distance of 53-71cm from the participant, and was set to 4-way stereo recording. The probes were transcribed using the PHON software, version 2.1.8 (2016) to ensure blind transcriptions by multiple coders.

Materials. Different materials were used for each thematic routine: this method allowed for the selected goals and their assigned target words to be embedded within simple games and activities that made the session more engaging for the participant. Materials included colour paper clipart images for each target word, Melissa and Doug ® Reusable Sticker Pads for the AMI condition, and Super Duper ® photo cues cards for the PMP condition. Photos of a research assistant were used as photo cues for vowel phonemes. Photos cues were printed in black and white with colour lines indicating articulatory cues added with coloured pens. Graphemes were placed below the photos for additional visual cues. Example cards and target word images are provided in Appendix B. In addition, a number of board games and toys were used to provide a context for breaks between blocks of trials. Games included Connect 4, Pig goes Pop, Bowling, ball games, puzzles, and simple board games constructed on paper from clip art. Adapted game pieces were used to facilitate participation where fine motor skill was an obstacle. Game pieces were made from foam and paper with pipe cleaner loops attached for grasping.

Results

Results are presented in the following sections for each individual experiment with regard to each of the four outcome measures, namely measures of performance, skill transfer, short term maintenance/learning, and long term maintenance. Cumulative treatment intensity was calculated for each treatment condition, where cumulative treatment intensity equals dose frequency times the total intervention duration times the mean dose. Statistical analyses were conducted separately for each set of probes (Same Day probes, Next Day probes). Random permutation of probe scores was used to determine if there was a significant association between treatment condition and outcomes, with the test statistic being *F* but the significance value determined by resampling test (this is a form of nonparametric repeated-measures ANOVA that is appropriate for assessing the results of a randomized single subject alternation design because the validity of the test is not dependent upon the assumptions of the parametric test such as normality or independence of the data points; the significance value is based on the actual distribution of the data obtained in the experiment, and not a theoretical distribution of *F* statistics; Huo, Edgington, & Onghena 2006). For pairwise planned comparisons of the probes, correlated two-tailed t-tests were calculated for each pair of treatment conditions among the three provided, with significance again determined via resampling from the distribution of random permutations of actual obtained data (Huo, Edgington, & Onghena 2006; Rvachew, 1988). Cohen's d_z was used in calculation of effect sizes (Lakens, 2013).

TASC DS 35

TASC DS 35 presented with a complex profile demonstrating multiple areas of deficit in encoding, phonological planning, motor planning, and execution. However, motor planning deficits predominated in his pre-treatment assessment profile: specifically, he added 63 phonemes when repeating nonsense words on the SRT, indicating a major problem with syllable transitions, also observed in his spontaneous speech; he was unable to produce a trisyllable sequence accurately during MPT testing; and, spatiotemporal coordination of articulators was a clear issue as manifested by oral-nasal confusions in his speech. Therefore, the hypothesis for this case was that he would respond most favourably to the auditory-motor integration intervention that was designed to mitigate problems in the area of motor planning.

Based upon his individual profile on the intake assessment, three speech therapy goals were selected to represent different level of the phonological hierarchy: specifically, (1) suppression of place harmony in 2-syllable CVC.CVC words, (2) major manner class contrasts, i.e., accurate production of stop vs. glide vs. liquid in the onset position of single syllable words; and (3) consistent production of the /f/ phoneme in the onset or coda of single syllable words. These speech goals were each randomly assigned the experimental treatment conditions: auditory motor integration pre-practice (goal 2), phonological memory and planning pre-practice (goal 1), and no pre-practice (goal 3). Treatment goal/treatment condition pairs were randomly assigned to sessions, within blocks so that all three goals were treated within each week and all goals were treated six times over the six week study.

Reliability and treatment fidelity. Inter-rater agreement for the narrow transcription of probes was 86% for vowels, and 85% for consonants. The inter-scorer agreement for probe item correctness was 97.7%. The criteria for probe scoring is noted in Appendix C. Treatment Fidelity was 100% for the PMP condition meaning that all expected PMP procedures were used in all 6 pre-practice sections, specifically segmentation of phonemes in target with linking to visual cues, chaining of segments to teach production syllables and whole word target, multisensory instruction including visual and tactile when needed but with avoidance of imitative models, and verbal instructions and phonetic placement to teach individual phonemes when necessary. Treatment fidelity was 73% for the AMI condition which suggests that auditory bombardment, target detection tasks, error detection tasks, were used in all six pre-practice sections but the participant was rarely asked to self-monitor his productions, which deviated from the expected treatment protocol. The primary requirement for treatment fidelity during the practice portion of the session is maintenance of a high number of practice trials, which will be reported in the next section.

Treatment data. Session level performance data, obtained from the video recordings of each treatment session, is presented for TASC DS 35 in *Figures 6* and *7*. *Figure 6* shows that the requirement for intense practice was met with a mean of 110 total trials across sessions and a generally increasing number of practice trials with time, regardless of condition. Furthermore, the mean number of correct trials per session was roughly similar across weeks and among all conditions although the percentage of correct trials indicates

Figure 6. TASC DS 35 Treatment Session Data

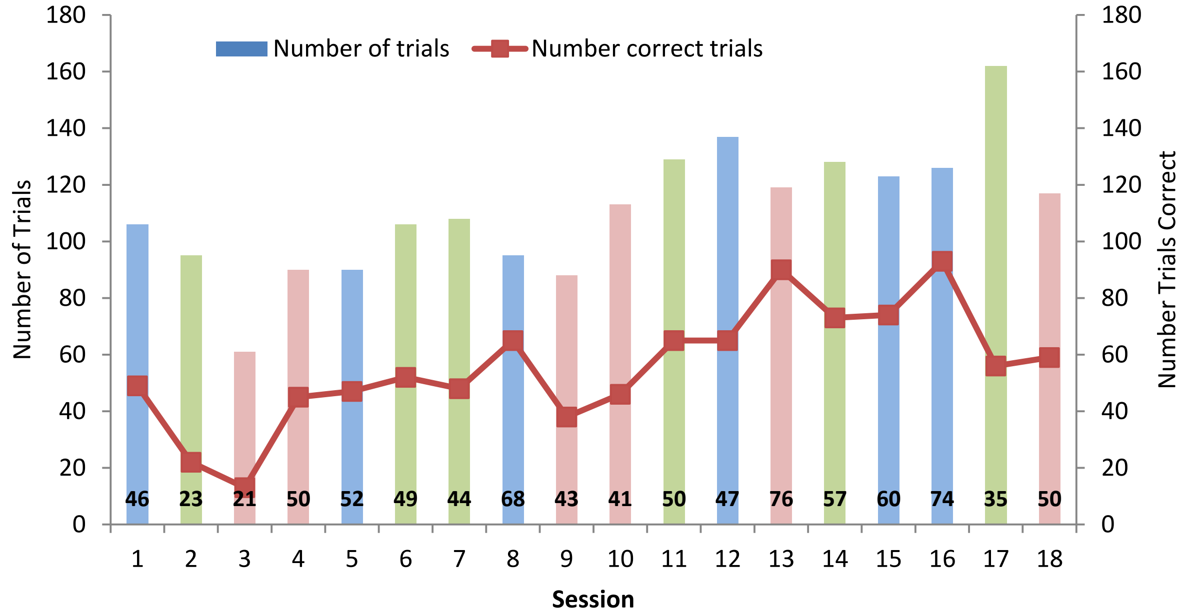

Figure 6. Number of trials, number of trials correct, and percent trials correct (data label) by session and condition. In this figure AMI is depicted as ☐, Control as ☐, and PMP as ☐. Total Trials: AMI *M=98 (SD=22.6),* Control *M=113 (SD= 18.69),* PMP *M=121 (SD= 23.9)* Percent Trials Correct: AMI *M=46% (SD=17%),* Control *M=58% (SD= 11%),* PMP *M=43% (SD= 12%)*

Figure 7. TASC DS 35 Progression of Target Complexity

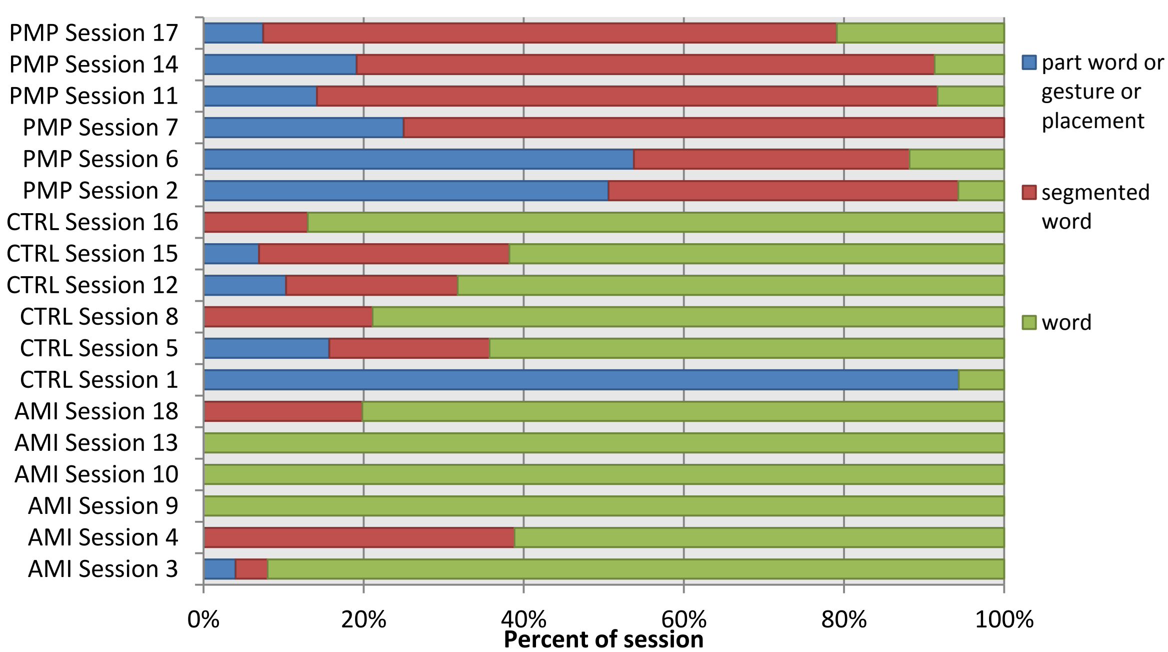

Figure 7. TASC DS 35 progression of target complexity shown as a function of condition and session.

that the participant was performing well below the desired challenge point of 80% correctness for all sessions. However, the cumulative treatment intensity varied widely among treatment conditions: scores were 588 for AMI, 678 for control, and 726 for PMP, indicating that the PMP was by far the most intensive treatment.

As indicated in *Figure 7*, TASC DS 35 did not progress beyond the word level with the target words in any condition. This suggests that the participant was able to achieve more trials per session as well as increase the number of times the target words were produced correctly, but that the complexity of the target did not greatly increase over the course of the treatment program. When combined with the low percentage of correct trials achieved, as shown in *Figure 6*, the treatment data suggests that this participant was struggling to achieve all three treatment goals.

Same Day Probes. Same day probes for TASC DS 35 are shown in *Figure 8*, as an indicator of generalization from treated nonsense words to untreated real words and phrases. A significant treatment effect was obtained for the same day probes overall, (F=5.71, p= 0.035) indicating an association between treatment condition and variation in generalization from treated to untreated words. Pairwise comparisons and effect sizes are reported in Table 5. The effect sizes suggest a much larger effect of the AMI treatment on Same Day probe performance in comparison to the PMP and Control condition but none of the pairwise comparisons were statistically significant. This may be due to the small sample size for each comparison (six sessions per condition).

Figure 8. TASC DS 35 Probe Scores

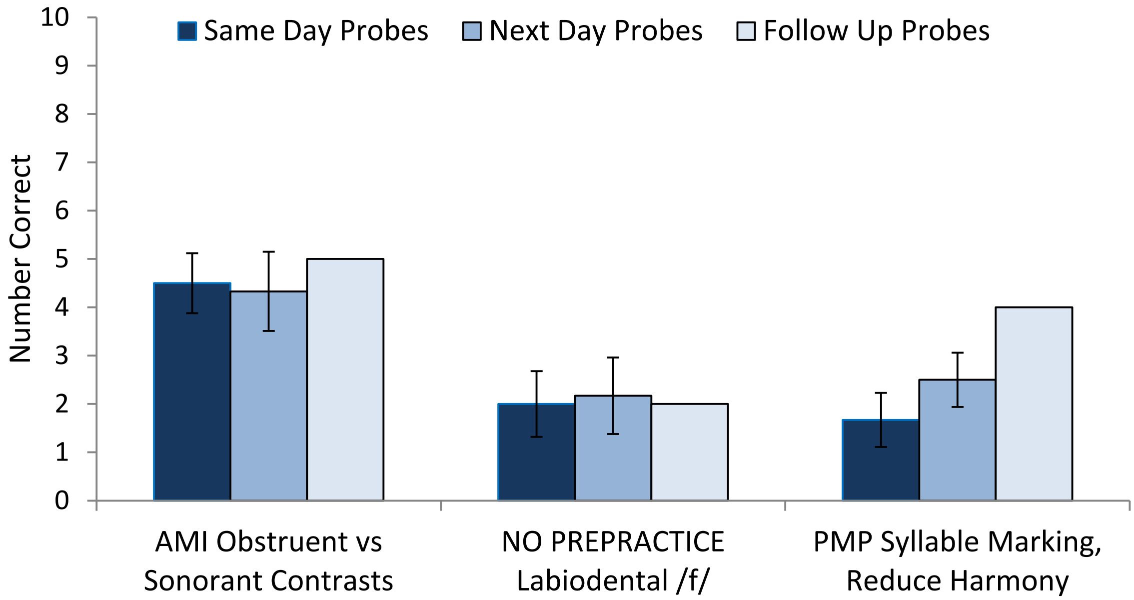

Figure 8. Probe scores for Same Day, Next Day, and Follow-up Probes.
Same Day Probes F=5.71, p=0.035 Next Day Probes F=4.15, p=0.056

Table 7
TASC DS 35 pairwise comparisons of treatment conditions

		Condition Comparison		
Probe Set	Calculation	AMI/Control	AMI/PMP	PMP/Control
Same Day Probes	t value	2.521	4.029	-0.326
	p value	0.097	0.058	0.935
	effect size (d_z)	2.384	2.336	0.354
Next Day Probes	t value	2.892	3.379	0.316
	p value	0.065	0.062	0.874
	effect size (d_z)	1.666	1.827	0.302

** Indicates a significant comparison*

Next Day Probes. The Next Day probes, indicating short-term maintenance of learning, are also shown in *Figure 8*. Although Next Day probe scores were numerically larger in the AMI condition, these probe scores were highly variable across weeks and conditions, and statistical significance was not achieved overall ($F=4.15$, $p= 0.056$), or for the pairwise comparisons. Again, the effect sizes suggest a larger effect of the AMI treatment on Next Day probe score performance in comparison with the PMP and Control conditions.

Follow-up assessment. A follow-up assessment was administered 84 days after the last treatment session, providing information about long-term maintenance of learning. Follow-up DEAP articulation and word inconsistency test performance is presented in Table 8. Although his word inconsistency score did not improve, percent consonant and vowel correct scores at the single word level, improved markedly, with 16% and 11% increases respectively. This improvement in segment accuracy was also noted in connected speech despite the fact that his MLU did not increase. Furthermore, these improvements appeared to reflect the goals of his treatment program in that the major class contrasts were better maintained at follow-up (e.g. ‘elefant’ /ɛləfənt/ →[ɛɫəbələ] at intake, but [alɛləvɪt] at follow-up), and consonant and syllable harmony were reduced (e.g. ‘this’ /ðɪs/→ [gɪk] at intake, [dɪs̺] at follow-up).

Visual analysis of the follow-up probes taken in the follow-up assessment (see *Figure 8*) shows improvement in both the AMI condition, and the PMP condition, but a slight decline in the probe score for the control condition. This suggests that the treatment effect seen during treatment was maintained over the three month period post treatment. In addition, the improved scores in the AMI and PMP conditions suggest increased generalization of the skills learned in treatment during this three month period.

Table 8
TASC DS 35 Follow-up assessment results

Assessment title	Assessment Subtest	Assessment Score: Intake Scores	Assessment Score: Follow-up Scores
Diagnostic Evaluation of Articulation and Phonology (DEAP) (Dodd, Zhu, Crosbie, Holm, & Ozanne, 2002)	Word Inconsistency Score	52%	60%
	Articulation Error Score	61	42
	Percent Consonants Correct	22%	38%
	Percent Vowels Correct	75%	86.1%
	Percent Phonemes Correct	41%	55.5%
Free Speech Sample (Day, 1989)	MLU	2.0	1.63
	Percent Consonants Correct	47%	50%

The long term outcome measure emphasizes that the AMI treatment condition was best suited to the needs of the participant. Improvement in the PMP condition is also understandable, given the patient's deficits in the areas of phonological planning.

Discussion. In this case, the participant presented with a primary underlying deficit in the area of motor planning at this particular developmental juncture, not in phonological planning or dyarthria: performance, transfer and maintenance observations aligned with the hypothesis that the participant would benefit most from an intervention designed to meet the needs of a motor planning deficit. This participant also had a coexisting fluency disorder, but measuring the potential affects of the fluency problem on motor oucomes was beyond the scope of this study. The outcome reported above suggests that he did indeed respond to CAS intervention, and that the most beneficial treatment could be deduced from his pre-treatment assessment profile. Further, it appears that the skills that were learned and maintained over the course of therapy led to some modest improvement in the participant's speech production accuracy given his improved PCC scores.

It is interesting to note that the level of success in the sessions as determined by the percent trials correct is seemingly unrelated to how well the participant was able to generalize the skills that he was learning: while the control condition had the highest number of correct trials in sessions, it produced the lowest probe scores. Also, although he produced more practice trials in the PMP condition than in the other conditions, the probe scores from *Figure 8* indicate that simply having more practice opportunities is not a guarantee of greater generalization. This lends support to the idea that mass practice alone is not sufficient, rather the practice must be targeted to address the underlying deficits. Figure 8 further suggests that the treatment effect observed for the AMI condition is not simply due to the extra treatment provided during the prepractice portion of the session, relative to the shorter control session; rather the nature of the procedures that were provided during prepractice appeared to place a role in his learning, at least in the short term.

TASC DS 37

TASC DS 37 presented with a mixed psycholinguistic profile with impairments in memory, transcoding, and execution. He was also dysfluent in connected speech, but investigation of this area was beyond the scope of this study. Increased difficulty in producing four syllable sequences on the SRT test, tendency to omit weak word-internal syllables, and overall inconsistent productions evinced a primary phonological planning disorder, although inability to sequence /pataka/, and voicing errors also indicated a motor planning disorder. Based on the severity of the phonological planning disorder, the hypothesis for this case was that the participant would benefit most from phonological memory and planning intervention, though it was unclear whether he would also benefit from auditory motor integration treatment.

Three goals were selected for this participant based upon his intake assessment profile: (1) strong-weak-strong stress pattern, (2) accurate production of internal abutting consonants, and (3) voicing contrast between /s/ and /z/. These treatment goals and their associated routines were randomly assigned to experimental conditions: auditory motor integration (goal 3), phonological memory and planning (goal 1), and no pre-practice control (goal 2). Treatment goal and condition pairs were randomly assigned to sessions within blocks so that each condition was addressed once per block, for a total of six sessions of each type (see appendix A for a depiction).

Reliability and treatment fidelity. Inter-rater agreement for the narrow transcription of the probes was 87% for vowels, and 84% for consonants. Agreement for probe score correctness was 91.66%. Rules used to score probe items as correct or incorrect are presented in Appendix C. The lab director and I used different rules when scoring stuttering for this participant which led to discrepancies on select items. Mutual agreement was reached through discussion, and it was decided that stuttered items would be scored as incorrect for this participant. The percent agreement score reported above reflects the scoring prior to discussion regarding stuttering. The treatment fidelity was 100% for the PMP condition indicating that all of the expected protocols for the condition were used in all six of the PMP pre-practice sections. The AMI condition had 87% treatment fidelity with intermittent use of the expected self-monitoring protocol indicating that there was less conformity to the protocols of this condition. Treatment fidelity with respect to session performance is discussed in the next section.

Treatment data. *Figures 9 and 10* depict session performance, through analysis of trial by trial data. *Figure 9* demonstrates that the treatment expectation of high intensity practice was met with a mean of 108.3 practice trials per session. Overall, TASC DS 37 remained relatively close to the desired challenge point of 80% correct trials though he was producing the target words in increasingly complex contexts as the sessions progressed. This suggests that the participant achieved consistently improved performance and dynamic stability at greater levels of target complexity. Cumulative treatment intensity varied among conditions: a score of 606 was calculated for the AMI condition, 684 for the Control condition, and 660 for the PMP condition, indicating that in fact the Control sessions had the highest level of treatment intensity.

Figure 9. TASC DS 37 Treatment Session Data

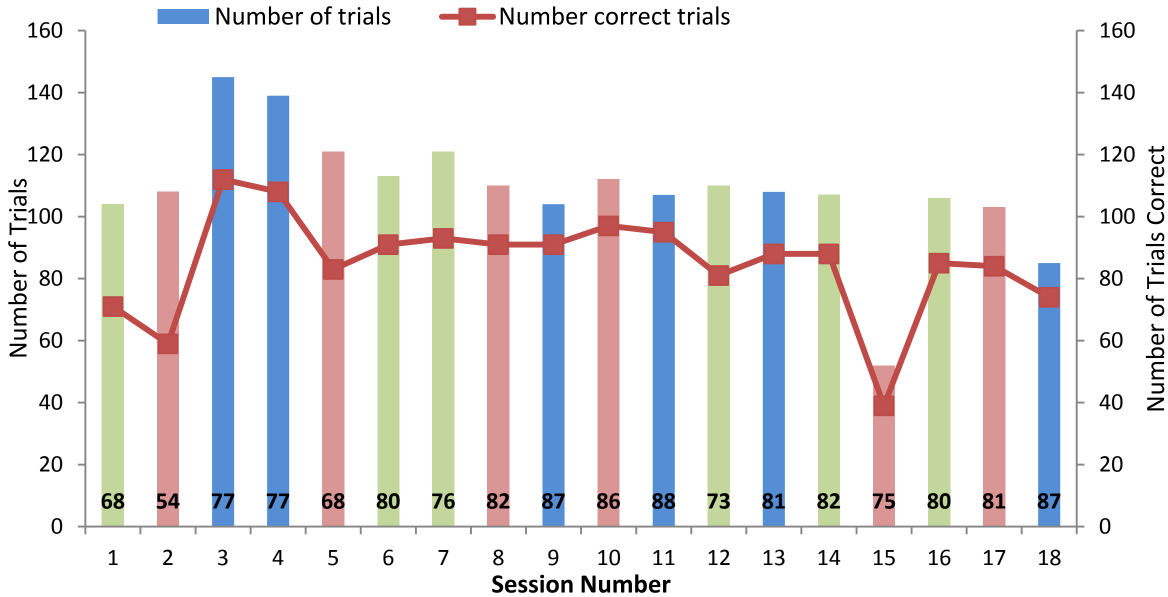

Figure 9. Number of trials, number of trials correct, and percent trials correct (data label) by session and condition. In this figure AMI is depicted as ☐, Control as ☐, and PMP as ☐. Total Trials: AMI: *M*=101 (*SD* 24.7), Control: *M*=114 (*SD* 22.8), PMP *M*=110 (*SD* 6.17) Percent Trials Correct: AMI: M=75% (SD11%), Control: M=83% (SD 4%), PMP M=77% (SD 5%).

Figure 10. TASC DS 37 Progression of Target Complexity

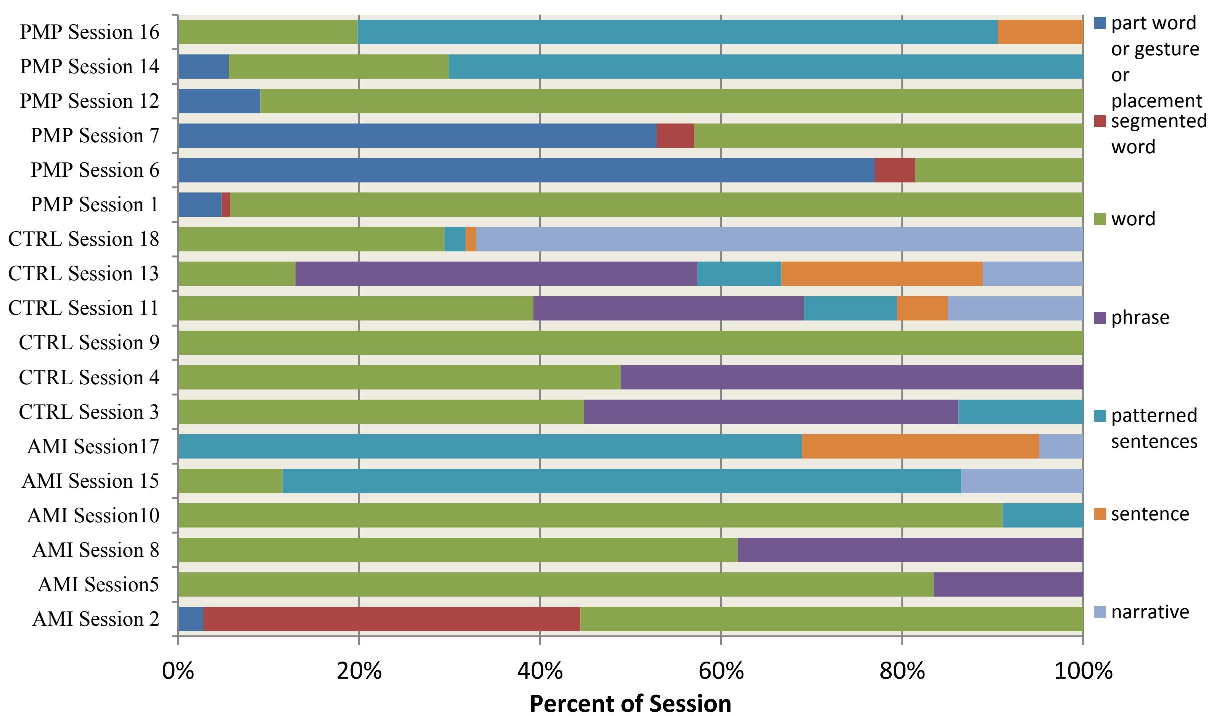

Figure 10. TASC DS 37 progression of target complexity shown as a function of condition and session.

Figure 9 is interesting because the trial number columns show a slight decrease in the total number of trials over time. This can be accounted for by considering the data presented in *Figure 10* which indicates that the participant moved from single word level complexity to sentences and narratives using the target words. This suggests that while the participant completed fewer trials per session as the treatment program progressed, the trials became more complex. The Control and AMI conditions reached the highest levels of complexity, with practice time devoted to embedding the target words within narratives. Practice did not progress to this level within the PMP condition: the participant progressed only as far as the sentence level for this condition.

Same Day Probes. The Same Day probe data charted in *Figure 11* shows an indicator of generalization from the treated words to untreated words and phrases. *Figure 11* denotes a statistically significant treatment effect, ($F= 4.657$, $p= 0.041$), demonstrating an association between condition and the transfer of skills from treated to untreated words. Planned pairwise comparisons and effect sizes are reported in Table 9. The effect sizes suggest a much larger effect of the PMP treatment on Same Day probe performance in comparison with the AMI and Control condition but the pairwise comparisons did not achieve statistical significance. As noted previously, this may be due to the small sample size for each comparison (six sessions per condition).

Figure 11. TASC DS 37 Probe Scores

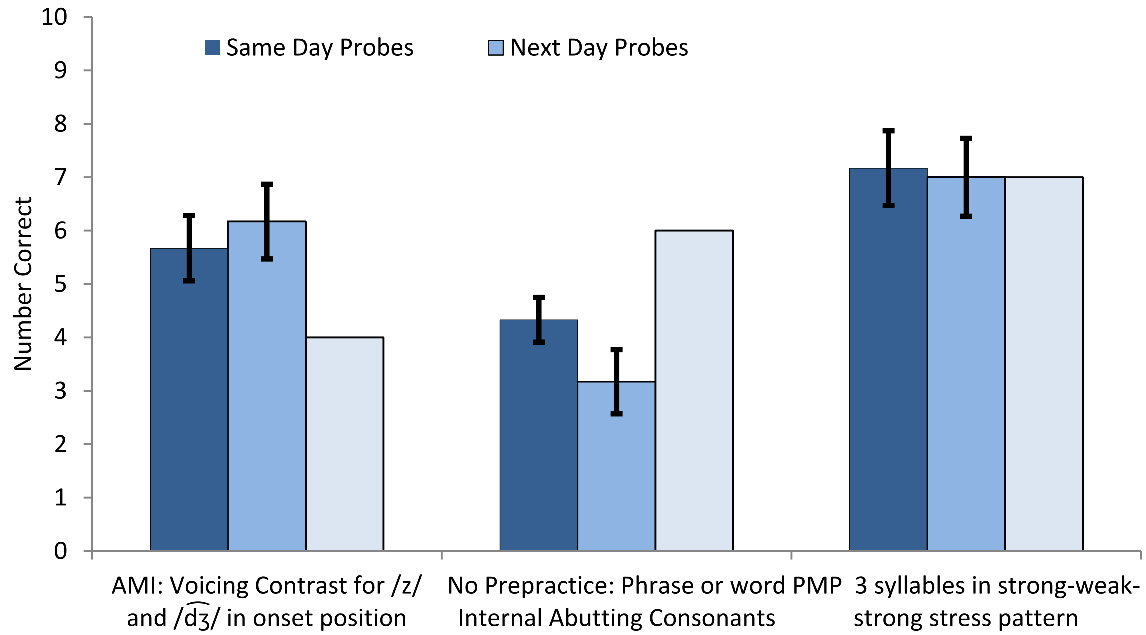

Figure 11. TASC DS 37 Probe scores for Same Day, Next Day, and Follow-up probes.
Same Day Probes F= 4.657, p= 0.041, Next Day Probes F= 7.491, p= 0.015

Table 9
TASC DS 37 pairwise comparisons of treatment conditions

		Condition Comparison		
Probe Set	Calculation	AMI/Control	PMP/AMI	PMP/Control
Same Day Probes	t value	1.397	-1.861	2.795
	p value	0.317	0.195	0.060
	effect size (d_z)	1.907	1.1303	3.577
Next Day Probes	t value	2.423	-2.076	3.608
	p value	0.089	0.188	0.060
	effect size (d_z)	3.554	0.264	4.386

** Indicates a significant comparison*

Next Day Probes. Next day probes were elicited as a measure of short term maintenance of skills, as well as learning. TASC DS 37 demonstrated numerically higher scores for both the PMP and AMI treatment conditions, compared to the Control, and a statistically significant treatment effect was found overall ($F= 7.491$, $p= 0.015$). Statistical significance in this comparison suggests an association between treatment condition and variation in scores for this short term maintenance outcome measure. Pairwise comparisons and effect sizes are reported in Table 9. The effect sizes suggest that the PMP condition had the greatest impact on the Next Day probe performance, followed by the AMI condition.

Follow-up assessment. As an indication of long term maintenance and learning, a follow-up assessment was administered 73 days after the last treatment session. The participant's follow-up assessment scores are presented in Table 10, showing an increase in the participant's MLU from the intake, but a slight decrease in the overall PCC count for connected speech. However, single word articulation scores have marginally improved from the intake assessment, with a 2% increase in PCC, PVC, and PPC scores. Furthermore, the follow-up word inconsistency test revealed a 4% decrease in the participant's production inconsistency. The improvements at the single word level appear to reflect the participant's treatment goals, for example, increased voicing contrast was observed so that 'zebra' /zibrə/ → [sibrə] at intake, but [zibrə] at follow-up, and 'jam' /d͡ʒæm/ → [t͡ʃæm] at intake, but [d͡ʒæm] at follow-up. Improvements were also noted in syllable marking, for example 'ladybug' /ˈleɪˌdiˈbəg/→ [ˈləˌləˈleɪˌdiˈbəg] in intake, but [ˈleɪˌdiˈbəgz] at follow-up.

Table 10
TASC DS 37 Follow-up assessment results

		Assessment Score	
Assessment title	Assessment Subtest	Intake Scores	Follow-up Scores
Diagnostic Evaluation of Articulation and Phonology (DEAP) (Dodd, Zhu, Crosbie, Holm, & Ozanne, 2002)	Word Inconsistency Score	56%	52%
	Articulation Error Score	17	15
	Percent Consonants Correct	79%	81%
	Percent Vowels Correct	92%	94%
	Percent Phonemes Correct	83%	85%
Free Speech Sample (Day, 1989)	MLU	3.58	4.21
	Percent Consonants Correct	71%	66%

Discussion. The results presented above suggest that the participant responded to treatments which addressed CAS. Further, the most beneficial treatment could be deduced Follow-up probe scores shown in *Figure 11* demonstrate that the participant was able to maintain the gains that he made in the PMP protocol: his score was exactly equal to his mean Next Day Probe score. However, the gains that were seen during treatment for the AMI condition for the short term outcome measure were not maintained at the point of follow-up in the probe phrases.

The follow-up probe scores establish that the hypothesis based upon the initial SRT result was correct: the participant demonstrated more long term benefits for treatment that specifically targeted phonological memory impairments. Interestingly, the participant made large gains in the skill practiced through the control condition, suggesting that some learning took place, or was integrated between the end of therapy and the follow-up assessment. This is understandable given that this treatment condition also included high intensity practice. through careful analysis of the participant's intake assessment. TASC DS 37 was hypothesized to respond best to a treatment for phonological planning disorders, and this was indeed the case from the short term outcome measures through the follow-up assessment despite the fact that this condition did not have the highest treatment intensity when compared to the control condition. Interestingly, this participant appeared to be the most engaged with the materials for the control condition, as wizards were one of his preferred conversation topics. Higher engagement and treatment intensity did not yield improvements in generalization as demonstrated in the Same Day and Next Day probe scores, however, the improvements seen in the follow-up probe scores suggest that some learning took place which is reasonable given the high treatment intensity.

TASC DS 38.

TASC DS 38 presented with a mixed profile of deficits throughout the speech chain including phonological disorder, phonological planning disorder, motor planning disorder, deficits in execution, and fluctuating hearing loss. The participant's hearing loss and lack of participation pre-empted the construction of specific hypothesis, though high intensity was warranted given the severity of his impairments.

Based upon the intake assessment profile, the selected goals were; (1) trochaic word stress with suppression of consonant harmony; (2) major manner contrasts in a CVC word shape, and; (3) consistent production of the /f/ phoneme. The first goal was randomly assigned to AMI, the second to PMP, and the third to the control condition. Subsequent randomization of treatment condition/goal pairs to sessions ensured that each treatment was addressed once in each block, for a total of six sessions per condition.

Reliability and Treatment Fidelity. Inter-rater agreement for the narrow transcription of speech sounds was 97% for vowels and 85% for consonants. Agreement for probe score correctness was 98.6%. Many of the sounds produced by this participant during the probes did not qualify as speech sounds given the criteria outlined in the methods section: these vocalizations were excluded. The treatment fidelity for the PMP condition was 93%, meaning that segmentation of the target word, visuals, phonemic placement, and multisensory prompting were used across all sessions, but chaining was only addressed in five of six sessions. AMI treatment fidelity was 56% due to the participant's refusal to engage in error detection or self-monitoring tasks in any session. Auditory bombardment, focused stimulation, and target identification were completed across sessions, however.

Treatment fidelity for session performance is reported in the next section. The participant would not tolerate feedback about correcting speech sounds in any condition which limited the clinician's ability to adhere the practice protocols, specifically those regarding the presentation of feedback.

Treatment Data. Session data presented in *Figures 12* and *13* depicts the participant's performance over the course of treatment. *Figure 12* demonstrates that the requirement for high intensity treatment was only marginally met, with a mean of 97.6 trials per session across treatment conditions. It is apparent that the participant was often below challenge point of 80% correctness indicating that the optimal setting for learning was not achieved through this treatment.

The participant achieved his highest level of success with PMP pre-practice condition sessions, save for the 13th session which had to be terminated due to a behavioural incident. The Control condition had the highest cumulative treatment intensity with a score of 654, followed by the AMI condition with a score of 594, and the PMP condition at 510.

Though the participant progressed beyond the word level across conditions, no marked decrease in the number of trials completed was observed, suggesting that the participant achieved enough dynamic stability in his productions to use the targets in more complex contexts.

Figure 12. TASC DS 38 Treatment Session Data

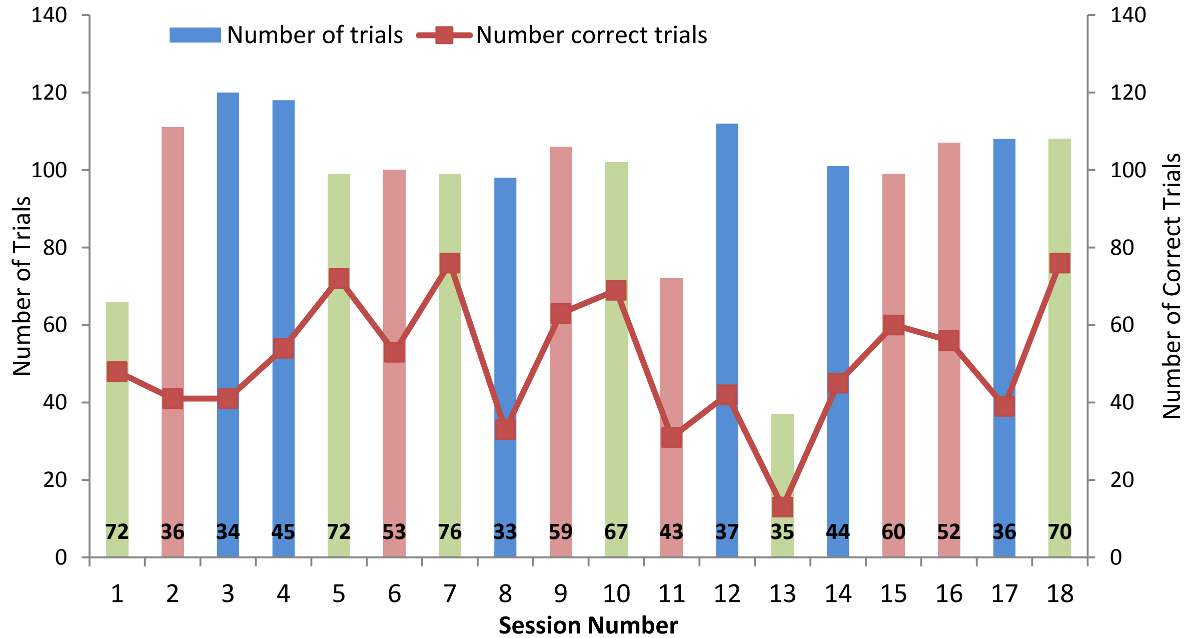

Figure 12. TASC DS 38 number of trials, number of trials correct, and percent trials correct (data label) by session and condition. In this figure AMI is depicted as ☐, control as ☐, and PMP as ☐.Total Trials: AMI *M*= 99 (*SD*=14.0), Control *M*= 109 (*SD*=8.89), PMP *M*= 85 (*SD*=27.8) Percent Trials Correct: AMI *M*= 51% (*SD*=8%), Control *M*= 38% (*SD*=4%), PMP *M*= 65% (*SD*=15%).

Figure 13. TASC DS 38 Progression of Target Complexity

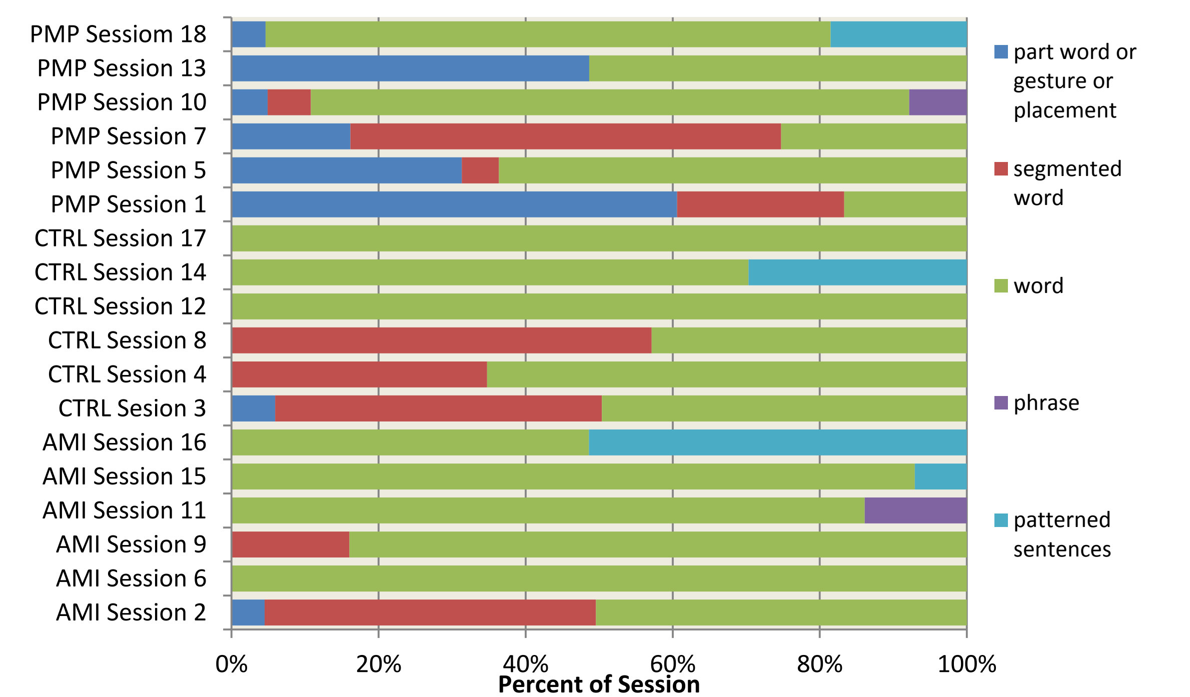

Figure 13. TASC DS 38 progression of target complexity shown as a function of condition and session.

Same Day Probes. Same Day probes are presented in *Figure 14* depicting generalization from treated words to untreated words. It is possible that the participant may frequently have underperformed for these probes: the participant was often observed to be unmotivated and frustrated by the end of the session. These scores showed a high level of variance, and the comparison was not significant (F=2.692, p= 0.1176). Thus, no association can be drawn between the treatment condition and the participant's probe score performance. Planned pairwise comparisons and effect sizes reported in Table 11. Again, pairwise comparisons did not generate statistically significant results. Comparisons with the AMI condition yielded the largest effect sizes for this outcome measure.

Next Day Probes. The Next Day probes, an indicator of short-term maintenance of learning, are depicted in *Figure 14* revealing a significant treatment effect (F=5.511, p= 0.0268). This suggests that there is an association between treatment condition and the variation seen in probe performance for this outcome measure. Pairwise comparisons and effect sizes are charted in Table11: no statistically significant comparisons were found. However, the magnitude of the effect sizes for the AMI and PMP conditions in the Next Day probes is much greater than the Same Day probes suggesting a much larger effect of treatment condition for the Next Day probe measure.

Figure 14. Probe Scores for TASC DS 38

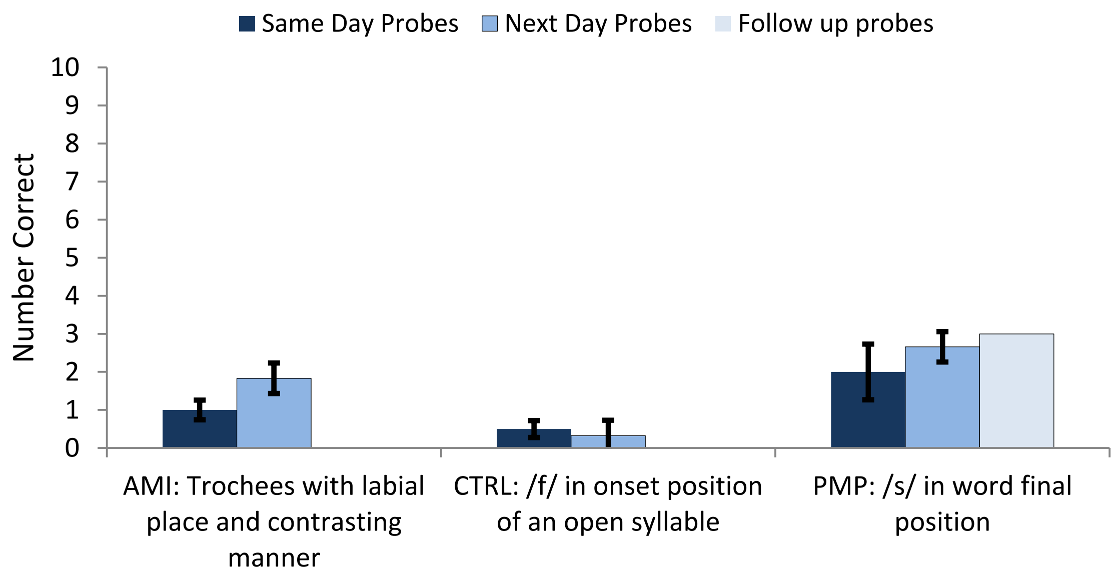

Figure 14. TASC DS 38 Probe Scores for Same Day, Next Day, and Follow-up Probes.
Same Day Probes: F=2.692, p= 0.1176: Next Day Probes: F=5.511, p= 0.0268

Table 11
TASC DS 38 pairwise comparisons of treatment conditions

		Condition Comparison		
Probe Set	Calculation	AMI/Control	AMI/PMP	PMP/Control
Same Day Probes	t value	1.168	-1.168	2.423
	p value	0.505	0.445	0.129
	effect size (d_z)	1.501	1.226	0.998
Next Day Probes	t value	3.000	-0.955	3.264
	p value	0.060	0.508	0.065
	effect size (d_z)	3.260	0.881	2.304

* Indicates a significant comparison

Follow-up assessment. Follow-up was undertaken 52 days after the last day of treatment: this follow-up assessment was completed earlier than the ideal time due to the participant's enrolment in another treatment protocol and the desire to prevent false inflation of treatment outcomes due to other treatments. Results of the follow-up assessment are presented in Table 12 showing a modest increase in the participant's PCC scores in connected speech, despite a lack of change in MLU. In addition, word level measures depict a decrease in the overall word inconsistency by 5% and an increase in single word consonant accuracy by 4% while vowel accuracy remained constant. The modest improvements observed at the single word level appeard to reflect the participant's treatment goals in that manner contrasts were somewhat more maintained, for example 'bird' /bɝd/→ [vøt] at intake, but [bɜwt] at follow-up.

This participant demonstrated maintenance of the skills used in the PMP condition only: his follow-up probe scores were zero for both AMI and control conditions. TASC DS 38 was engaged during the elicitation of the follow-up probes, and these scores are considered to accurate representations of his ability. This long term outcome measure suggests that the PMP condition was most favourably suited to the participant's needs.

Table 12
TASC DS 38 Follow-up assessment results

		Assessment Score	
Assessment title	Assessment Subtest	Intake Scores	Follow-up Scores
Diagnostic Evaluation of Articulation and Phonology (DEAP) (Dodd, Zhu, Crosbie, Holm, & Ozanne, 2002)	Word Inconsistency Score	84%	79%
	Articulation Error Score	54	53
	Percent Consonants Correct	29%	33%
	Percent Vowels Correct	68%	68%
	Percent Phonemes Correct	42%	45%
Free Speech Sample (Wiesner, 2006)	MLU	1.98	2.1
	Percent Consonants Correct	38%	44%

Discussion. As noted above, this participant was observed to be unmotivated to participate in therapy, and presented with numerous behavioural challenges. His level of engagement did not align with a particular experimental condition, but was seen to reflect his daily experience prior to arriving at therapy: his Mother's reports of whether it was a 'good day so far' were generally accurate predictors of his behaviour. The performance level data, as well as both probes show a higher level of variability for this participant than for the two previous experiments. Performance variability may reflect the behavioural and motivational state of the participant in that he was willing to engage to a much larger degree in some sessions compared to others.

Despite these challenges, the participant did make gains in therapy with regard to the learning and maintenance of speech goals, as is evidenced by his probe scores for the PMP condition. To be precise, this participant benefitted most from treatment that was multisensory in nature: this may be because learning from the visuals was not dependent on his fluctuating hearing ability. This finding is interesting because the PMP condition had the lowest cumulative treatment intensity overall, implying protocols of the treatment itself were crucial to the participant's success.

General Discussion

This dissertation presented three single subject randomized experiments investigating speech therapy outcomes for individuals with Down syndrome and Childhood Apraxia of Speech. Specifically, these studies investigated whether (1) individuals with DS and severe speech sound disorders respond to CAS treatment, and (2), whether a particular form of speech therapy intervention produced superior results with regards to target learning, generalization, and maintenance. Speech goals were randomized to treatment conditions within blocks: high intensity therapy was conducted three times per week, over six week-long blocks for a total of 18 45-minute sessions. Four outcome measures were elicited for each participant regarding performance, transfer, short term maintenance, and long term maintenance. These outcome measures were analyzed through inspection of the session level data, Same Day Probes, Next Day Probes, and follow-up assessment, respectively. All probes were scored for target accuracy with individualized criteria.

Successes

Program design. The use of single subject alternation randomization design allows for both visual and statistical analysis of treatment progress. Each session yielded two sampling opportunities to objectively measure learning and generalization through the Same Day Probes and Next Day Probes. This conforms to the agreed upon idea that the sampling method should be dynamic and sensitive to subtle changes over time (Smith, 2012; Krauth, 2000). The sessions were also video recorded and analyzed for treatment fidelity, number of completed trials, progression through the integral stimulation hierarchy, and progression of target complexity, which provided a measure of change in performance. The ingenuity of this research design and its sensitivity to change makes it particularly suitable for clinical research. The design is remarkably valuable in cases where group level study and analysis are impertinent. For example, it allowed for an in-depth analysis of TASC DS 38, where his behaviour might have resulted in his exclusion from group level studies. Practicing clinicians can utilize this design to determine effective treatments for individuals who present with a unique cluster of speech characteristics or behavioural challenges.

Recent publications report that participant characteristics are of critical importance with regard to treatment outcomes: the same conclusion can be drawn from this data. Ledford et al. (2016) point out that one potential benefit of single subject research is the possibility of drawing conclusions from the potentially equivocal data based upon participant characteristics. In other words, replication of single subject research allows for the analysis of heterogeneous data produced by seemingly homogeneous participants. In this research each participant responded differently to the treatment and control conditions despite a uniform label of severe speech sound disorders at the time of intake.

Participants. Significant treatment effects with large effect sizes for each participant indicate that using specialized pre-practice procedures is more beneficial for treatment outcomes, in comparison with high intensity treatment alone. This finding suggests that simply ensuring high numbers of practice trials is not sufficient for improving speech target generalization and maintenance: rather, therapy must remediate the underlying source of the speech sound disorder. For the participants presented in this dissertation, application of therapy techniques that address the underlying deficits in phonological memory and motor planning prior to high intensity practice allowed the individuals to benefit more from treatment.

Clinical implications. This research has generated three major clinical implications which relate to the overall problems of inquiry that drove these experiments. First and foremost, all three experiments found that the participants presented with CAS in combination with impairments at the other stages of speech production such as phonological disorders, dysarthria, and fluency disorders. Inconsistent productions, prosodic errors, slotting errors, disrupted syllable transitions, oral/nasal contrast errors, vowel errors, and voicing errors were widely observed in this research, though the symptomatology of each individual was unique. The observed error patterns demonstrated both phonological planning impairments and motor planning impairments which has implications for speech treatment in DS; it suggests that the speech profile for this population may be more unique and more complex than previously anticipated. Assessments of speech should include a motor speech component to identify whether phonological planning and/or motor planning impairments are present and to what extent they impact speech.

Second, though motor speech impairments were found across participants, no single uniform underlying deficit in speech production was identified. Indeed, each individual presented with a unique speech profile and was variably affected by impairments across the speech chain. Rather than attempting to treat all the observed impairments simultaneously, this research focused treatment on the most prominent impairment, namely phonological or motor planning deficits. Therefore, selection of appropriate treatment goals was dependent upon thorough assessment and analysis of the individual's unique psycholinguistic profile. This finding is congruent with that of Rupela et al. (2016), wherein the authors noted that identification of effective intervention can only be made possible through systematic assessment and analysis of the clinical presentation of the individual.

Finally, this research found that individuals with DS respond constructively to CAS treatment and that their performance improves with the use of specialized pre-practice protocols. For example, TASC DS 35 presented with a primary deficit in the area of motor planning and responded best to the AMI pre-practice condition, whereas TASC DS 37 presented with a primary deficit in phonological planning and responded best to the PMP pre-practice condition. These pre-practice protocols, in combination with high intensity practice, resulted in greater generalization and maintenance of the speech targets than high intensity practice alone.

Limitations

This series of studies contained a number of limitations, reflecting the small number of participants and the particular aims of this project. Although this research was focused on specificity of treatment for phonological and motor planning disorders, it is important to consider the possibility that the performance of the individuals presented here could have been influenced by their other speech profile characteristics. Other factors such as dysfluency and dysarthria could have impacted upon the individual's performance; however, measurement of those areas was beyond the scope of this study.

This research in is limited in statistical power for these participants who demonstrated small gains and variable performance over the six week duration of the study. Therefore, the pairwise analysis of treatment and control conditions was inconclusive even though significant associations between the experimental manipulations and probe scores were obtained overall. Although the ANOVA analysis revealed an association between treatment condition and performance variability, it was not possible to confirm that either of the experimental conditions was significantly more effective than the control or that there was a significant advantage to either of the experimental treatments relative to the other. It is reasonable to expect that these differences in treatment effectiveness would be small, given that the participants received opportunities for high intensity speech practice in all three conditions. Achieving sufficient statistical power to permit these finer level comparisons would require an adjustment to the design, specifically: (1) increase the number of treatment sessions for each participant, or (2) replicate the procedures with more participants to allow for pooling of p-values across participants.

While presenting three single subject randomized cases does provide some insight as to how a heterogeneous population may respond to treatment, further exploration by replicating these treatment procedures would be germane. Greater participant success in the PMP and AMI conditions suggests that individuals with DS and CAS respond to specialized pre-practice procedures. However, given the small number of studies completed at this time, it is unclear whether one of these treatments is more effective for generalization and maintenance than the other. Replications of this research would provide the opportunity for more in-depth analysis of whether one treatment is significantly more beneficial than the other for individuals in this population.

This research was limited to investigating treatment for older children and adolescents with DS: in consideration of this, it is unclear whether this type and intensity of treatment would be appropriate for younger individuals. Intensive speech therapy for CAS is often geared towards younger individuals with the goal of improving speech sound production at an earlier age, and this would be ideal for the population with concomitant DS and CAS. However, given the inherent complexity of this population, and the high demands of this type of therapy in terms of attention and engagement, it is unclear whether this diagnostic and treatment process would be transferrable to younger individuals. Further replications of this type of intervention with younger individuals would elucidate the treatment’s effectiveness for this scenario.

Another challenge of this type of clinical research is that it is quite resource heavy in regards to recording and processing the data: numerous volunteers and research assistants contributed to the data management of this project, from wielding the video camera to working on narrow transcription. Given the caseload and administrative demands of many practicing clinicians, this presents an obstacle in terms of replicating this type of research within the bounds clinical employment. A potential solution for clinicians who feel that this type of treatment research is warranted is to make use of volunteers who have experience in phonetic transcription and are looking to further expand their understanding of its clinical application.

Conclusion

Currently, the idea that individuals with Down syndrome may present with a varied profile of speech disorders is gaining more support. Traditionally, the diagnostic challenge is presented as one of differentiating phonological from motor speech disorders or dysarthria from apraxia. Individuals with Down syndrome experience a complex neurodevelopmental profile however; each individual may exhibit symptoms of phonological delay, phonological planning disorder, apraxia and dysarthria, sequentially or simultaneously. Therefore the challenge is to identify the primary deficit of the individual at any given development juncture. As the ability to identify the primary deficit improves with the use of new diagnostic testing, there is an increased need for intervention techniques that target the specific underlying deficit. This research was intended as an initial enquiry into the treatment of the motor speech disorder CAS in individuals with DS with the broad goal of identifying whether CAS oriented treatments are effective in this population. The findings presented here are encouraging, suggesting that specialized treatment measures can be more effective than high intensity practice alone in treatment. Further, this data suggests that it is possible to identify which specialized pre-treatment procedures will be most effective in treatment through administration of a thorough pre-treatment assessment and analysis of those results. It is my hope that these findings can guide further research into this area of speech therapy, specifically in applying this kind of assessment and treatment to younger individuals with DS. By replicating this research in adolescents and younger people, it will be possible to improve the quality of evidence based practice for this population.

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Appendix A:

Randomization of treatment conditions to sessions within blocks

Block Number	TASC DS 35	TASC DS 37	TASC DS 38
1	CTRL	PMP	PMP
	AMI	AMI	AMI
	PMP	CTRL	CTRL
2	AMI	CRTL	CTRL
	PMP	AMI	PMP
	CTRL	PMP	AMI
3	PMP	PMP	PMP
	AMI	AMI	CTRL
	CTRL	CTRL	AMI
4	CTRL	AMI	PMP
	AMI	CTRL	AMI
	PMP	PMP	CTRL
5	PMP	CTRL	PMP
	AMI	PMP	CTRL
	CTRL	AMI	AMI
6	CTRL	PMP	AMI
	PMP	AMI	CTRL
	AMI	CTRL	PMP

Treatment Conditions:
Auditory Motor Integration= AMI,
Phonological Memory and Planning= PMP,
Control = CTRL

Appendix B:

Examples of face cue cards used in the PMP condition for each participant. The dimensions of the cards were approximately 6cm x 11cm for TASC DS 35 and TASC DS 38, and 5cm x 8cm for TASC DS 37. Examples of target word images displayed below the photo cues.

Y o s

t i g er

Bling Four Fall Far

Zander Zug Zingle Jig Sea

Tam team Tam wizard Pog team pog wizard darklord

Bobbytak Goobertoom Wobey Meewa

Appendix C:

Criteria for probe scoring. Due to the unique profile of each participant, scoring rules were individually created.

Participant	AMI	PMP	CONTROL
TASC DS 35	Goal: sonorant/obstruent contrast across word Probe Scoring Criteria: must get at least one glide or liquid correct in each phrase with two Cs in different classes in the utterance	Goal:2 syllable word practice with trochaic stress, contrasting place of articulation with stop consonants Probe Scoring Criteria: 1. vowel accuracy 2. no consonant or vowel harmony 3. accurate number of syllables Disregard voicing and final consonant deletion	Goal: /f/phoneme with greater contrast between /f/ and /p/ across word positions Probe Scoring Criteria: Must produce /f/ phoneme in the correct word position.
TASC DS 37	Goal: voicing contrast in the onset position Probe Scoring Criteria: Segment must be +voice, +continuant, +coronal, and +consonantal	Goal: 3 syllable words with strong-weak-strong stress pattern Probe Scoring Criteria: Strong-weak-strong stress pattern, with no stuttering	Goal: word/phrase internal abutting consonants Probe Scoring Criteria: Must contain distinct abutting consonants, with no stuttering
TASC DS 38	Goal: C1V1C2V2 trochee, contrasting manner at the labial place of articulation Probe Scoring Criteria: Must have CV structure, must match place and manner: disregard voicing.	Goal: C_1VC_2 C_1=/n,j,r,d/, C_2=S Probe Scoring Criteria: Must match place and manner of articulation: disregard voicing.	Goal: /f/ in a CV(r) syllable Probe Scoring Criteria: Must match place and manner of articulation: disregard voicing.