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Effects of an intensive voice treatment on articulatory function and speech intelligibility in children with motor speech disorders: A phase one study

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ABSTRACT

Producing speech that is clear, audible, and intelligible to others is a challenge for many children with cerebral palsy (CP) and children with Down syndrome (DS). Previous studies have demonstrated the effectiveness of using the Lee Silverman Voice Treatment (LSVT LOUD®) to increase vocal loudness and improve speech intelligibility in individuals with dysarthria secondary to Parkinson's disease (PD), and some research suggests that it also may be effective for individuals with dysarthria secondary to other conditions, including CP and DS. Although LSVT LOUD targets healthy vocal loudness, there is some evidence of spreading effects to the articulatory system. Acoustic data from two groups of children with secondary motor speech disorders [one with CP (n = 17) and one with DS (n = 9)] who received a full dose of LSVT LOUD and for whom post-treatment intelligibility gains have been previously reported, were analyzed for treatment effects on: *1)* vowel duration, *2)* acoustic vowel space and *3)* the ratio of F2/i/ to F2/u/. Statistically significant changes in vowel duration and acoustic vowel space occurred pre-treatment to 12 weeks post-treatment in the CP group, and increased acoustic vowel space was observed in 5 of the DS participants. The present study provides preliminary evidence of intensive voice treatment spreading effects to the articulatory system in some children with CP and children with DS consistent with previous findings in other populations.

1. Introduction

Decreased speech intelligibility is often a challenge for both individuals with Down syndrome (DS) (see reviews in: Bunton & Leddy, 2011; and Wood, Wishart, Hardcastle, Cleland, & Timmins, 2009), and individuals with cerebral palsy (CP) (see reviews in: Pennington, Miller, Robson, & Steen, 2010; and Watson & Pennington, 2015). The results of one large group survey using parent report suggest that as many as 95% of children with DS have difficulty being understood at least some of the time, and 80% have difficulties specific to articulation (Kumin, 1994). According to one recent study of over 1300 individuals notified to the Northern Ireland Cerebral Palsy Register, approximately 36 % of children with CP have secondary motor speech impairments (Parkes, Hill,

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Platt, & Donnelly, 2010). However, estimates of prevalence vary widely in the literature (e.g., 21–53 % (see Mei et al., 2016; Nordberg, Miniscaco, Lohmander, & Himmelmann, 2013). Prevalence of DS has been estimated at 14.1 per 10,000 live and still births (Public Health Agency Of Canada, 2013), and prevalence of CP at 2.11 per 1000 live births (Oskoui, Coutinho, Dykeman, Jetté, & Pringsheim, 2013). Although exact figures are not available, it is clear that a significant number of children are affected by motor speech disorders secondary to DS or CP.

In pediatric populations, communication impairments interfere with overall development, and may be associated with depression, reduced quality of life, and increased risk of social isolation and academic difficulties (Fuhrman, Equit, Schmidt, & Von Gontard, 2014; Pennington, Miller, & Robson, 2009). However, it may be that interventions resulting in improved speech intelligibility of children with DS or CP would positively impact participation in life activities, and may have substantial short and long-term benefits for emotional and social well-being (Boliek & Fox, 2016; Pennington, Rauch, Smith, & Brittain, 2019).

There is no general agreement on which factors are responsible for the reduced speech intelligibility nearly always associated with DS, or whether it is best characterized as a motor speech disorder or a developmental phonological delay (Bunton & Leddy, 2011; Mahler & Jones, 2012; Wood et al., 2009). Many acoustic and perceptual observations of this population reported in the literature are consistent with motor speech deficits, which suggests that such deficits do at least play a contributory role in decreased intelligibility for at least some individuals with DS (Bunton & Leddy, 2011; Mahler & Jones, 2012), although they may not be easily categorized due to overlapping symptoms of dysarthria, childhood apraxia of speech and otherwise unspecified motor speech disorders (Rupela, Velleman, & Andrianopoulos, 2016). Reported perceptual features of voice and speech in individuals with DS include: reduced loudness, both hypo- and hypernasality, imprecise articulation of consonants and vowels; atypical pitch patterns and prosody; and breathy, hoarse or harsh voice quality (Mahler & Jones, 2012; see reviews in Roberts, Price, & Malkin, 2007; and Venail, Gardiner, & Mondain, 2004). Possible underlying causes are: structural characteristics that may impact articulation, such as abnormal facial musculature, and a small oral cavity together with a normal-sized tongue; physiological characteristics, such as low muscle tone and abnormal innervation of the articulators; phonological errors; and/or motor speech programming difficulties (see reviews in: Bunton & Leddy, 2011; Mahler & Jones, 2012; Martin, Klusek, Estigarribia, & Roberts, 2009; Roberts et al., 2007; and Wood et al., 2009). Further, DS almost always involves cognitive impairment, and is associated with a high incidence of hearing loss: both of these factors can contribute to an increased risk of overall speech and language delay (see reviews in: Martin et al., 2009; and Roberts et al., 2007).

The most prevalent form of speech disorder in individuals with CP is dysarthria, which may present as imprecise articulation, low pitch, reduced pitch variation, harsh voice, hypernasality, and/or deficient breath control for speech, all of which can contribute to reduced intelligibility (see reviews in Pennington et al., 2010; and Watson & Pennington, 2015). The perceptual characteristics of dysarthrias associated with spastic and dyskinetic types of CP are similar, although the severity is generally greater for individuals with the dyskinetic form (Pennington et al., 2010). While not as prevalent as in DS, cognitive impairment is present in approximately half of children with CP, and may impact speech and language development (Parkes et al., 2010; Surman et al., 2009; Watson & Pennington, 2015).

1.1. Vowel acoustics in motor speech disorders

In studies of motor speech disorders, acoustic vowel space and other measures of the first and second formants of vowels (F1 and F2) have been of interest both because of their importance for speech perception (see, e.g., Delattre, Liberman, Cooper, & Gerstman, 1952) and therefore intelligibility to listeners, and because of their relationship to the articulatory system as products of its anatomy and physiology (see, e.g. review in Hixon, Weismer, & Hoit, 2014, 415–454). F1 and F2 are affected by the coordination, strength, and range of motion of the articulators as they alter the shape and space of the oral cavity. In particular, F1 has been shown to decrease as tongue height increases, which may be affected by jaw movement as well as by independent lingual movement. F2 has been shown to increase as the tongue moves forward, though movement in the vertical plane may also affect F2 to a lesser extent. Both F1 and F2 have been shown to decrease with lip rounding, with F2 showing the most pronounced effects (Hixon et al., 2014; Lee, Shaiman, & Weismer, 2016; Stevens & House, 1955). Formant values also are affected by overall vocal tract structure both directly, as with the smaller size of children's vocal tracts, which are associated with higher formant frequencies (see, e.g. Lee, Potamianos, & Narayanan, 1999), and indirectly, as with structural anomalies such as the relatively small oral cavity associated with DS, which may restrict the working space of the articulators (Bunton & Leddy, 2011). It has been suggested that both compressed acoustic vowel space and reduced formant transitions may be common characteristics across dysarthrias (Weismer & Kim, 2010). Several studies have found decreased acoustic vowel space in speakers with dysarthria, as well as correlations between smaller acoustic vowel spaces and reduced intelligibility (see, e.g., Higgins & Hodge, 2002; Kim, Hasegawa-Johnson, & Perlman, 2011a; Kim, Kent, & Weismer, 2011b; Lansford & Liss, 2014a, 2014b; Lee, Hustad, & Weismer, 2014; Liu, Tsao, & Kuhl, 2005; Wenke, Cornwell, & Theodoros, 2010). Shallower F2 transition slopes also have been associated with dysarthric speech and reduced intelligibility (see, e.g., Kent et al., 1989; Lee et al., 2014; Lansford & Liss, 2014b).

As yet there has been little research using non-perceptual methods to assess the functioning of the articulatory system in individuals with DS. However, some limited evidence from acoustic and physiological studies suggests that this population has a reduced vowel working space (see review in Kent & Vorperian, 2013). Moura et al. (2008) observed that children with DS had an overall smaller acoustic vowel space than age-matched controls, and in particular smaller differences between F1 of /a/ compared with the F1 of /i/ and /u/, and a smaller ratio between the F2 of /i/ and /u/. The authors interpreted the F1 findings to reflect a more limited jaw movement and mouth opening, which would restrict the tongue from descending in order to produce the higher F1 frequency typical of low vowels. The smaller F2i/F2u ratio was interpreted as reflecting restricted tongue movement in the high back position, resulting in a higher than typical F2 value for /u/. Another recent study using both acoustic measures and X-ray microbeam tracking of tongue movements showed decreased acoustic vowel space, decreased articulatory space, and slower articulatory movements in two adults with DS compared with healthy controls (Bunton & Leddy, 2011).

To date there also has been little acoustical or physiological research on the articulatory function of children with CP, although there is some evidence that of all the speech subsystems, the articulatory subsystem may be the most determinative of intelligibility in this population (Lee et al., 2014; Levy et al., 2016; Nip, 2017; Scholderle, Staiger, Lampe, Strecker, & Ziegler, 2016), and that patterns of acoustic correlates of dysarthria in children are similar to those of adults (Lee et al., 2014). A recent study of the speech acoustics of children with CP and dysarthria found significantly smaller acoustic vowel space, longer vowel durations and shallower F2 transition slopes in diphthongs (i.e., in the words “pipe” and “toys”) and labiolingual glides (i.e., in the word “whip”), compared with control groups of typically developing children and children with CP but without dysarthria (Lee et al., 2014). Further, these acoustic measures of articulatory function were found to predict 58 % of the variance in intelligibility measures. The longer vowel durations and shallower F2 transition slopes of the CP-dysarthria group were interpreted by the authors as reflective of slower and reduced articulatory movement. This is consistent with the results of a recent kinematic study, which found reduced ability to coordinate articulatory movements of the jaw and lips among children with CP, and significant correlations between reduced interarticulatory coordination and decreased intelligibility (Nip, 2017).

1.2. Treatment approaches

There is currently no consensus on best practices for treatment of speech disorders in children with DS and CP, and the literature on the effectiveness of interventions for pediatric dysarthrias is sparse (Pennington, Parker, Kelly, & Miller, 2016). For individuals with DS, the traditional approach has been articulation therapy to target disordered phonological processes and sound errors (see, e.g. Martin et al., 2009). More recent exploratory work has involved electropalatography, (Wood et al., 2009), and intensive therapy targeting the laryngeal system (Boliek et al., 2012), both of which show some promise. Non-speech oral motor treatments also have been attempted. However, current evidence does not support their efficacy (Lee & Gibbon, 2015).

Treatment approaches to improve speech intelligibility of children with CP are similarly varied, and also have included articulation therapies and non-speech interventions of questionable effectiveness (Pennington et al., 2016). Some recent single subject and small group studies provide evidence that intensive therapies based on motor learning principles and a singular target of healthy vocal loudness (2016, Boliek & Fox, 2014; Fox & Boliek, 2012; Levy, Ramig, & Camarata, 2012) or multiple targets of breath support, phonation and speech rate (2013, Levy et al., 2012; Pennington et al., 2010) may produce better intelligibility gains for this population. These findings are consistent with neuroplasticity and motor learning principles that are increasingly informing overall rehabilitation strategies for individuals with CP (Garvey, Giannetti, Alter, & Lum, 2007).

The Lee Silverman Voice Treatment (LSVT LOUD®) is a short-term intensive therapy with a single target of achieving healthy vocal loudness. It was originally developed to treat individuals with Parkinson’s disease (PD) and dysarthria. Several studies have demonstrated the effectiveness of LSVT LOUD at increasing vocal loudness and improving the intelligibility of speech in individuals with PD (Sapir, Spielman, Ramig, Story, & Fox, 2007; Sauvageau, Roy, Langlois, & Macoir, 2015), and some small group studies suggest that it also may improve speech intelligibility for individuals with dysarthria secondary to other conditions, including adults with DS (Mahler & Jones, 2012), children with DS (Boliek et al., 2012), and children with CP (2016, Boliek & Fox, 2014; Fox & Boliek, 2012; Levy et al., 2012).

Whereas traditional treatments for dysarthria targeting multiple systems (breathing, laryngeal, velopharyngeal and oral articulatory) separately or in combination, may be effective at improving speech intelligibility in some school-aged children with CP (Levy et al., 2012; Pennington et al., 2010), LSVT LOUD, which has a single treatment target of healthy vocal loudness, and relies primarily on modeling rather than verbal instruction to elicit target behaviour, may be better suited to individuals with cognitive impairments (2013, Fox & Boliek, 2012; Pennington et al., 2010; Wenke et al., 2010; Youssef, Anter, & Hassen, 2015) and preschool children. This limited cognitive load makes it particularly promising for those children with CP and children with DS who also have reduced intellectual functioning, and also may allow for earlier interventions with preschool-aged children.

Although LSVT LOUD only directly targets the phonatory system, there is some evidence based on perceptual and acoustic measures that it also improves articulation in individuals with PD, individuals with non-progressive dysarthrias, and individuals with flaccid dysarthrias, which may explain some of the gains in intelligibility that cannot be accounted for by increased loudness alone (Sapir et al., 2007; Sauvageau et al., 2015; Wenke et al., 2010; Youssef et al., 2015).

Sapir et al. (2007) found significant post-LSVT LOUD changes in individuals with PD in vocal sound pressure levels, the F2 of the vowels /i/ and /u/, the ratio F2i/F2u, and perceptual vowel goodness ratings. No significant changes were observed in control groups of healthy individuals and individuals with PD who did not receive treatment. Acoustic vowel space was measured, but did not change significantly from pre- to post-treatment. Results from Wenke et al. (2010) study indicated that individuals with non-progressive dysarthrias secondary to stroke or traumatic brain injury made significant gains in both acoustic vowel space and perceptual intelligibility ratings post-LSVT LOUD treatment and at six months’ follow up. None of the acoustic or perceptual outcomes were significantly different from those of a control group of individuals who received a traditional dysarthria therapy based on multiple targets. Sauvageau et al. (2015) reported increased acoustic vowel space in individuals with PD who received LSVT LOUD, as well as greater post-treatment distinctiveness in consonant-vowel coarticulations. Youssef et al. (2015) study of individuals with flaccid dysarthria secondary to stroke or traumatic brain injury found statistically significant changes to F1 and F2 of /a/ and /u/, with a tendency for higher F1 values and lower F2 values post-treatment.

1.3. Purpose

To summarize, motor speech disorders are prevalent among individuals with DS and individuals with CP, affecting many children and adults. LSVT LOUD is an intensive therapy with a single target of healthy vocal loudness that has been shown to increase intelligibility in individuals with Parkinson's disease, through gains not only in the directly targeted phonatory function, but also in the functioning of the articulatory subsystem as evidenced by changes to measures of acoustic vowel space. Recent studies suggest that LSVT LOUD also may produce intelligibility and vocal loudness gains in children with motor speech disorders secondary to DS or CP. However, the possibility of treatment spreading effects to the articulatory subsystem in these populations has not yet been investigated.

The purpose of the present study was to undertake a retrospective analysis of acoustic data from two previous LSVT LOUD treatment studies, one of children with CP (Boliek & Fox, 2016), and one of children with DS (Boliek et al., 2012, 2010; Boliek, Hardy, Halpern, Fox, & Ramig, 2016), to test for gains similar to those found in studies of individuals with PD and non-progressive dysarthrias. It was predicted that both the CP group and the DS group would show significant post treatment increases in vowel duration, acoustic vowel space, and changes in the ratio of F2i/F2u formants.

2. Method

A within-group, repeated measures design was selected to test for post-treatment changes to acoustic measures.

2.1. Participants

Selection criteria for all participants included: a) presence of a perceptible speech or voice disorder; b) hearing within normal limits (aided or unaided); c) absence of vocal fold pathology; d) cognitive ability to follow directions and perform the voice and speech tasks of the study protocol; and e) medical stability. Exclusion criteria included a) severe velopharyngeal incompetence; and b) severe structural disorders of the speech mechanism. Table 1 provides details of individual participant characteristics for both participant groups.

Table 1
Description of participants with cerebral palsy and particpants with Down Syndrome including sex, age, speech diagnosis, rating on the Gross Motor Function Classification System (GMFCS- CP only), and cognitive level (CP only).

Participant	Sex	Age	CP Diagnosis	Speech Diagnosis	GMFCS	Cognitive Level
Cerebral Palsy						
F0601E	F	6	Spastic Quadriplegia	Spastic-Flaccid Dysarthria, Severe	V	Average
LSVTM5	M	8	Spastic Quadriplegia	Spastic Dysarthria, Mild-Moderate	V	Average
LSVTM8	M	8	Spastic Diplegia	Spastic-Flaccid Dysarthria, Moderate-Severe; Dysfluency, Mild	II	Below Average
F0801E	F	8	Spastic Quadriplegia	Spastic-Flaccid Dysarthria, Mild	I	Above Average
F0802E	F	8	Spastic Quadriplegia	Spastic Dysarthria, Mild	II	Average
F1001E	F	10	Spastic Quadriplegia	Spastic Dysarthria, Moderate	V	Average
F1201E	F	10	Spastic Quadriplegia	Spastic Dysarthria, Mild	III	Above Average
F1202E	F	12	Spastic Quadriplegia	Spastic-Flaccid Dysarthria, Severe; Dysfluency, Severe	II	Average
M0901E	M	10	Spastic Quadriplegia	Spastic-Flaccid Dysarthria, Moderate	III	Average
M1001E	M	10	Spastic Quadriplegia	Spastic-Flaccid Dysarthria, Severe	V	Average
LSVTM2	M	11	Spastic Diplegia	Spastic-Ataxic Dysarthria, Mild	II	Average
LSVTF3	F	12	Spastic Triplegia	Spastic-Flaccid Dysarthria, Mild; AOS, Mild-Moderate	II	Average
LSVTM4	M	12	Spastic Quadriplegia	Spastic-Flaccid Dysarthria, Mild	V	Below Average
LSVTF1	F	13	Spastic Diplegia	Spastic-Flaccid Dysarthria, Moderate	III	Above average
LSVTM6	M	13	Spastic Diplegia	Spastic-Flaccid Dysarthria, Moderate-Severe; AOS, Moderate-Severe	IV	Below Average
LSVTM9	M	13	Spastic-ataxic Quadriplegia	Spastic-Ataxic Dysarthria, Moderate-Severe; AOS, Mild	IV	Average
LSVTF7	F	16	Spastic Diplegia	Spastic-Flaccid Dysarthria, Severe; AOS, Severe	III	Average
Down Syndrome						
S24	F	4	Trisomy 21	Flaccid Dysarthria, Moderate; AOS, Mild		
S23	F	5	Trisomy 21	Spastic-Flaccid Dysarthria, Moderate; AOS, Mild		
S21	F	6	Trisomy 21	Flaccid Dysarthria, Moderate; AOS, Mild		
S25	M	7	Trisomy 21	Flaccid Dysarthria, Moderate		
S28	F	7	Mosaic	Flaccid Dysarthria, Mild		
S22	F	8	Trisomy 21	Spastic-Flaccid Dysarthria, Moderate-Severe		
S26	F	8	Trisomy 21	Flaccid Dysarthria, Moderate		
S27	F	8	Trisomy 21	Flaccid Dysarthria, Moderate; AOS, Mild		
S29	F	8	Trisomy 21	Flaccid Dysarthria, Mild-Moderate; AOS, Mild		

GMFC = *Gross Motor Function Classification System* (expanded and revised scale; Palisano, Rosenbaum, Bartlett, & Livingston, 2008) (higher number indicates greater severity). Note: Diagnosis was made by a pediatric neurologist, GMFCS was determined by a physical therapist, cognitive level was determined by a licensed psychologist as reported in the participant record, and speech/voice diagnoses and their severity were determined by licensed speech-language pathologists who specialize in pediatric motor speech disorders.

2.1.1. CP group

Participants were 17 children with CP, aged 6 to 16 years (mean age of 10.6 years, sd = 2.52 years), and were recruited and treated in Edmonton, Alberta, Canada. All participants had Western Canadian English as their first language. Informed written consent and assent were obtained in accordance with the requirements of the Health Research Ethics Board at the University of Alberta, which approved the study. All participants were diagnosed by certified SLPs who specialized in the diagnosis and treatment of pediatric speech disorders. Based on individualized diagnostic protocols, this population was characterized as having spastic (n = 4), spastic-ataxic (n = 2), or spastic-flaccid (n = 11) dysarthria, ranging from mild to severe. Four participants also had apraxia of speech diagnoses, and two had dysfluency diagnoses.

2.1.2. DS group

Participants were 9 children with DS, aged 4–8 years (mean age of 6.8 years, sd = 1.48 years), and were recruited and treated in Denver, Colorado. In addition to the exclusion criteria described above, participants in this group were excluded if they had a severe articulation disorder, and /or a concomitant speech disorder (e.g., dysfluency). Consent and assent were obtained in accordance with the requirements of the human research ethics board at the University of Colorado, which approved the study. Further ethical approval was provided for the transfer of de-identified data to the University of Alberta for analysis and interpretation. All participants were diagnosed with mixed dysarthria by consensus of three certified SLPs.

2.2. Procedures

2.2.1. Recordings

Recordings for both groups were made within one week prior to treatment (PRE), and within one week following treatment (POST). With the exception of one participant, the CP group was also recorded at twelve weeks follow-up (FUP). Recording sessions ranged from 30 min to one hour. CP group participants had a single session at each time. DS group participants had from 1 to 3 sessions at PRE, and from 1 to 2 sessions at POST. The individuals who collected the recordings were not associated with the treatment or data analysis phases of the studies, and were trained to be consistent across participants. Recordings of CP participants were made in a quiet room; recordings of DS participants were made in an Industrial Acoustics Company sound-treated booth. All recordings were collected using either a lapel unidirectional microphone (Shure 185: CP group), or a small omni-directional condenser microphone (Audio-Technica, Model AT 803b: DS group) secured to participants' foreheads to maintain consistent mouth to microphone distances (8 cm for the DS group and 10 cm for the CP group). Signals were sent to a digital audiotape (DAT) recorder [Panasonic Digital Audio Tape Deck, Model SV-3500, 44.5 kHz: DS group; Tascam DA-P1 DAT, 44.1 kHz, or directly to computer using TF32 software (Milenkovic, 2004): CP group. All acoustic data were converted to WAV files.

2.2.2. Speech samples

A summary of the speech samples used is presented in Table 2. Speech samples include single words from the Test of Children's Speech Plus (TOCS+) (Hodge, Daniels, & Gotzke, 2006) for the CP group, and the *Goldman-Fristoe Test of Articulation 2* (GFTA) (Goldman & Fristoe, 2000) for the DS group; and the sentences, "The potato stew is in the pot," and "The blue spot is on the key" for both groups. The TOCS + words were elicited by computer software, which simultaneously displayed a picture and played a recording of a word for the participant to repeat. Most of the GFTA words (97% of those used in this study) were produced spontaneously by the participant (i.e., without first hearing someone else say the word), after being asked to name a picture. The TOCS + and GFTA words were elicited only one time at each sitting, with the exception of six TOCS + words (5% of those used in this study), which were produced twice. All of the sentences were produced in direct imitation of a Western Canadian English speaker (children in the CP cohorts) and in direct imitation of a Western US English speaker (DS cohort). Sentences were modelled using a conversational speaking rate. Sentences were repeated three times at each session. In most cases, participants were asked to repeat phrases rather than the full sentences (e.g., "the blue spot" / "is on the key"). Participants' total number of repetitions of each token word at each time varied from a minimum of one to a maximum of nine. None of the words used as tokens were trained during the treatment phase.

2.2.3. Treatment protocol

Each participant received a full dose of LSVT LOUD treatment from a certified SLP. Consistent with the standard protocol for LSVT LOUD (Ramig et al., 2001), treatment consisted of 16 one-hour sessions, delivered over a period of four weeks (four days per week) as well as daily homework assignments (one per day on treatment days, and two per day on non-treatment days). The CP group also participated in a maintenance program of at-home practice during the 12 weeks following the treatment. All treatment sessions followed the same protocol during the first 30 min: i) at least 15 repetitions each of "long ah" (maximum phonation duration), "high ah" (maximum f_0 range), and "low ah" (minimum f_0 range); and ii) at least five repetitions each of 10 functional phrases chosen by the participants and their parents. The second 30 min included individualized practice based on topics of interest, and progressing over the course of the month from short to longer utterances (individual words to paragraphs) and from simple to more complex (repetition without distractors to reading and conversation with distractors). Homework included repeating the exercises and practice used in the sessions, as well as extra assignments such as talking to someone on the phone.

Table 2
Description of speech samples and tokens.

Data set	Source of speech samples	Number of speech samples at each time point	Tokens	Number of productions of each token per sample/participant	Type of speech
CP – Single Words	*Test of Children's Speech Plus* (each participant was randomly assigned one of 3-word lists)	Pre: 1 Post: 1 FUP: 1	each participant was assigned one of the following sets: "jaw", "bee", "two" "paw", "tea", "two" "top", "bee", "boo" Note: "pooh" was substituted for "two" for one participant.	1 - 2	Imitative.
CP – Sentences	"The blue spot is on the key" and "The potato stew is in the pot."	Pre: 1 Post: 1 FUP: 1	"blue", "key", and "pot"	3	Imitative.
DS – Single Words	*Goldman-Fristoe Test of Articulation 2*	Pre: 1-3 Post: 1-2	"blue", "tree", and "watch"	1	Spontaneous: 97% Imitative: 3%
DS - Sentences	"The blue spot is on the key" and "The potato stew is in the pot."	Pre: 1-3 Post: 1-2	"blue", "key", and "pot"	3	Imitative.

"CP" refers to the cerebral palsy group; "DS" refers to the Down syndrome group; "FUP" refers to follow-up. Where more than one speech sample was collected for a single time point (e.g., 3 pre-treatment samples), each sample was collected on a separate day.

2.3. Perceptual ratings of single word intelligibility

Single word intelligibility data is from previously reported studies of these same groups of participants (Boliek & Fox [in preparation], Boliek & Fox, 2016; Boliek et al., 2016, 2012; Boliek et al., 2010). Tokens for the CP group were words and phrases from TOCS+. Tokens for the DS group were words from the GFTA. All listeners in those studies were tested for normal hearing, were aged between 18 and 60 years, had English as a first language, and no training in speech-language pathology or experience with dysarthric speech. Listeners were randomly assigned subsets of recordings to evaluate, they heard each token one time, and recorded what they heard each speaker say on a form or typed directly into the computer using an open set procedure. Mean intelligibility for each speaker was calculated across the responses of five listeners for each speaker.

2.4. Acoustic analyses

2.4.1. Tokens

See Table 2 for a summary of the tokens used. Tokens were selected to provide samples of the vowels /ɑ/, /i/ and /u/. As three separate word lists were used with participants in collecting the TOCS + words, three sets of tokens were used for this data set: jaw/bee/two (n = 6), paw/tea/two (n = 5; in one case “two” was replaced by “pooh” because a recording of “two” by that participant was cut off by an external sound), and top/bee/boo (n = 6). The three tokens used in the GFTA data set were: “watch”, “tree”, and “blue”. The three tokens used for the sentence production datasets were “pot”, “key” and “blue”. Tokens produced in isolation in the recordings of sentences (e.g., “key”, instead of “on the key”), were excluded from further analysis. Vowel boundaries were marked on spectrograms using Praat software. Glides and liquids immediately preceding the vowel, including those produced in error (e.g., as in [twi] for “tree”), were included within the vowel boundaries. Tokens were excluded at the boundary-marking stage for one of two reasons: *a)* the vowel was cut off by an ambient sound, such as a page turning or the voice of another person in the room; and *b)* the vowel was whispered.

2.4.2. Measurement of formants

Formants were initially measured using a custom-made Praat script which extracted average F1 and F2 values over the 30 ms midsection of each vowel. Fig. 1 provides a sample spectrogram illustrating how vowel boundaries were marked and where formant measurements were taken. Spectrograms of individual speakers were manually reviewed using Praat’s formant tracker to verify the number of formants setting that produced the most accurate fit for an individual speaker. Maximum formant frequency was set at 5500 Hz for all speakers. Boxplots were produced for all measured formant values for each of the vowels for each of the data sets. A second manual review of 180 spectrograms (20 %) was conducted to verify outliers from the boxplots. Following these reviews: 55 of a total of 882 tokens (6 %) were removed entirely from the data due to the difficulty of identifying formants. Values for 32 tokens (4 %) were manually measured at midpoint

sections of the vowels where the reviewer could clearly identify the formants and they appeared stable. Twenty-one tokens were excluded from acoustic vowel space analyses in the TOCS + data. Tokens were removed because the acoustic vowel space variables required a set of valid measurements for all three vowels at each time point. Missing or incomplete token sets were spread fairly evenly among participants and can be seen by the symbol (-) in Table A1 in Appendix A.

2.4.3. Calculation of variables

Both acoustic vowel space and the F2i/F2u ratio were calculated using the average F1 and F2 values for each participant from the 30 ms midsections of the vowels. Acoustic vowel space was calculated as the area of a triangle formed by the locations of the three vowels in the F1-F2 space using absolute values of the equation $\Delta = 0.5(-x_2y_1 + x_3y_1 + x_1y_2 - x_3y_2 - x_1y_3 + x_2y_3)$, with x_1, x_2, x_3 =

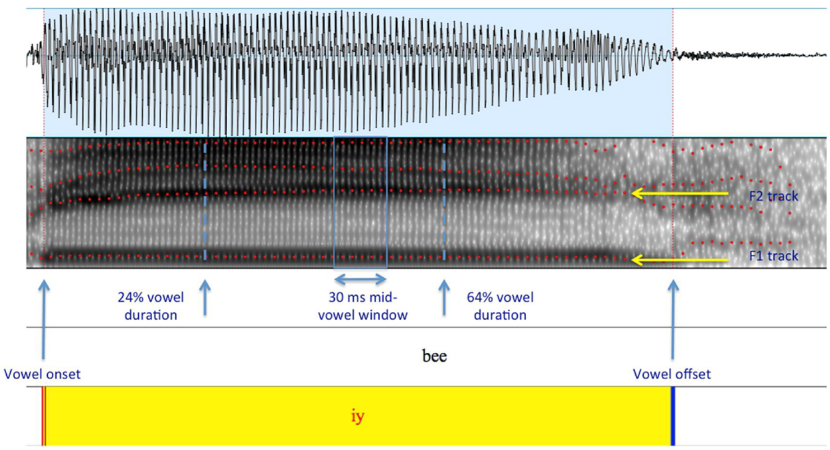

Fig. 1. Sample spectrogram of the token “bee” indicating vowel boundaries and formant measurement locations.

mean F1 values in Hz, and y_1, y_2, y_3 = mean F2 values in Hz. The F2i/F2u ratio was calculated using $\log_{10}$ transformed frequency values.

2.5. Reliability

To assess intra-measurer reliability of acoustic measurements, 10% of tokens were randomly selected and vowel boundaries were re-marked by the original measurer. The formant measures used to calculate the variables were derived using the Praat script with the same settings used for each token as in the original measurements. The intraclass correlation coefficients (ICCs) obtained for acoustic vowel space variables were: Vowel space area, $r = .78$; ratio of F2i/F2u, $r = 0.85$. It should be noted that the ICCs for these variables reflect agreement in all but six formant measurements and two formant measurements, respectively.

To assess inter-measurer reliability of acoustic measurements, 10% of tokens were randomly selected and vowel boundaries were re-marked by a second person trained in the protocol for determining vowel boundaries. The formant measures used to calculate the variables were derived using the Praat script with the same settings used for each token as in the original measurements. The following ICCs were obtained for acoustic vowel space variables: Vowel space area, $r = 0.60$; ratio of F2i/F2u, $r = 0.72$.

2.6. Statistical analyses

Data for intelligibility, averaged dB SPL and averaged vowel durations were tested for change from PRE- to POST- and PRE- to FUP-treatment (CP cohorts) using *t*-tests[1] . For acoustic vowel space and F2i/F2u, data for each participant at each time point were averaged to provide mean formant values, which are shown in Appendix A. Friedman's tests were used to compare CP group variables at PRE, POST and FUP, as all datasets either included outliers, or did not meet the assumption of normality for parametric tests, or both. Post hoc comparisons were made using Wilcoxon tests for paired samples, with no corrections for multiple comparisons. This approach is consistent with Robey's (2004) suggestion that Phase I research should adopt a liberal tolerance for Type 1 error. A second set of comparisons was made for CP group variables in the phrases condition excluding three participants (F1202, M6 and F7) with comorbid apraxia of speech or dysfluency diagnoses of moderate or greater severity (CP group dysarthria-only). These comparisons were not made in the single word condition, because of the already smaller sample sizes due to the reduced number of available tokens. Only four DS group participants had sufficient data to yield acoustic vowel space measurements for sentence productions, and these results are reported descriptively only. All other DS group variables were compared using Wilcoxon tests for paired samples, as all datasets either included outliers, or did not meet the assumption of normality for parametric tests, or both.

A Cohen's d statistic (Cohen, 1988) or r statistic was applied to treatment-related outcome measures (i.e. pre- to post-treatment values and pre- to 12-week follow-up values). Whereas Beeson and Robey (2006) suggest caution when assigning a categorical value to effect sizes in early phases of treatment research, we adopted a standard categorization of effect size of $d = 0.2$ as "small", $d = 0.5$ as "medium", as $d = 0.8$ or greater as "large"; and a categorization of effect size of $r = 0.1$ as "low", $r = 0.3$ as "medium", and $r = 0.5$ or greater as "large" (Cohen, 1988).

For all analyses, we considered a liberal α of < 0.10 consistent with Phase I research (Robey, 2004). Using this approach, we indicate when significance was met using the more standard $p < 0.05$ criteria and also when values "approached" significance, that is, if α was between $p = .06$ and $p = 0.10$, perhaps indicating worthy of further investigation.

3. Results

3.1. Overview of results

3.1.1. Intelligibility

3.1.1.1. CP group. Group averaged intelligibility data are reported separately for two cohorts of participants: LSVT I (F601, F801, F802, F1001, F1201, F1202, M901 and M1001), and LSVT II (F1, M2, F3, M4, M5, M6, F7, M8, M9) in Table 3. The results indicate significant PRE to POST increases in single word intelligibility for the LSVT I cohort ($t = 3.02$, $p < .01$) and significant PRE to POST and PRE to FUP gains in sentence intelligibility for the LSVT II cohort ($t = 4.08$, $p < .01$; $t = 2.57$, $p = .03$, respectively). Both cohorts showed gains in single word intelligibility from PRE to FUP that approached statistical significance (LSVT I: $t = 1.69$, $p = .09$; LSVT II: $t = 1.91$, $p = .09$).

3.1.1.2. DS group. Intelligibility data for the DS group are reported in Table 3, and indicate PRE to POST gains in single word intelligibility that approached statistical significance.

3.1.1.3. dB SPL outcomes. The data from dB SPL derived from produced phrases are shown in Table 4. Significant PRE to POST increased in dB SPL were found for the CP LSVT I cohort ($t = 2.5$, $p = .05$) and the DS cohort ($t = 2.5$, $p = .05$). PRE to POST

[1] Because one of the participants from the CP cohort did not attend a follow-up session, the mean value of all the other participants was computed at FUP for each variable, and inserted into the data set for that participant for the purposes of comparing means between the three time points. The Pre- vs Follow up-comparisons were not affected statistically either way so we opted for increased statistical power especially on the important Pre- vs Post- comparisons.

Table 3
Intelligibility outcomes for children with cerebral palsy (CP) (n = 17; 8-LSVT I and 9-LSVT II) and Down Syndrome, (DS) (n = 9).

	PRE-POST Comparison				PRE-FUP Comparison			
CP % Whole Word correct	Mean differences (SD) [95% CI]	*t*-value	*p*-value	Cohen's *d*	Mean differences (SD)	*t*-value	*p*-value	Cohen's *d*
LSVT I – single words (TOCS+)	7.28% (6.36%) [18.27, 5.13]	3.02	< .01*	0.35	4.71% (7.22%) [20.79, 1.29]	1.69	.09†	0.42
LSVT II – single words (TOCS+)	1.37% (6.77%) [7.29, 3.40]	0.61	.56	0.05	4.48% (7.03%) [7.299, 0.47]	1.91	.09†	0.18
LSVT II – Sentences (TOCS + sentences)	9.61% (7.06%) [15.10, 4.20]	4.08	< .01*	0.31	6.76% (7.88%) [12.82, 0.71]	2.57	.03*	0.22
DS								
Single words - GFTA	4.44% (8.6%) [11.58, 6.4]	1.55	.08†	0.27				

Percent whole world correct (n = 5 listeners/speaker); FUP refers to "follow-up"; "TOCS+" refers to the *Test of Children's Speech Plus*, GFTA refers to *Goldman-Fristoe Test of Articulation* 2. Note: only single words were evaluated for intelligibility in the LSVT I cohort. 95% confidence intervals [CI] are reported for upper and lower intervals of the differences. *Statistically significant. † Approaching statistical significance.

Table 4
Results for dB SPL from spoken sentences for both LSVT CP cohorts (n = 17; 8-LSVT I and 9-LSVT II) and the cohort with DS (n = 9).

Group	PRE(SD)	POST(SD)	FUP(SD)	PRE-POST		PRE-FUP	
				t	*p*	*t*	*p*
CP LSVT I	57.98 (17.81)	68.11 (16.58)	68.51 (11.35)	2.5 [16.27, 0.19]	.05*	2.3 [16.27, 0.19]	.06†
CP LSVT II	70.22 (6.31)	72.18 (7.54)	79.10 (7.26)	2.2 [6.38, 3.54]	.06†	2.6 [1.6, 14.41]	.03*
DS	52.58 (11.9)	61.65 (11.5)		2.5 [4.93, 0.45]	.05*		

FUP refers to 12 weeks follow up. 95% confidence intervals [CI] are reported for upper and lower intervals of the difference. *Statistically significant. † Approaching statistical significance.

increases in dB SPL approached statistical significance for the LSVT II cohort ($t = 2.2$, $p = .06$). PRE to FUP increases in dB SPL were found for the CP LSVTII cohort ($t = 2.6$, p = .03), and approached statistical significance for the CP LSVTI cohort ($t = 2.3$, $p = .06$)

3.2. Vowel duration outcomes

An analysis of the vowel duration data, summarized in Table 5, found only one significant comparison. The CP children durations

Table 5
Vowel duration outcomes for words and sentences for children with cerebral palsy (CP) (n = 17) and Downs Syndrome (DS) (n = 9).

	PRE-POST Comparison				PRE-FUP Comparison			
	Mean differences (SD)	*t*-value	*p*-value	Cohen's *d*	Mean differences (SD)	*t*-value	*p*-value	Cohen's *d*
CP								
Single words (TOCS+)	0.02 s (0.15) [0.04, 0.002]	0.71	.48	0.14	0.04 s (0.15) [0.06, 0.02]	1.3	.20	0.27
Sentences (TOCS + sentences)	0.06 s (0.16) [0.08, 0.04]	3.1	< .01*	0.38	0.04 s (0.15) [0.06, 0.02]	1.92	.06†	0.24
DS								
Single words (GFTA)	0.01 s (0.12) [0.06, 0.02]	0.02	.85	0.04				
Sentences (GFTA)	0.03 s (0.10) [0.06, 0.003]	1.24	.21	0.19				

FUP refers to "follow-up"; "TOCS+" refers to the *Test of Children's Speech Plus;* "GFTA" refers to the *Goldman-Fristoe Test of Articulation 2.* Children with Down Syndrome did not participate in a FUP session. Note: Differences are in the direction of longer durations at the second point in time (i.e., POST, FUP). 95% confidence intervals [CI] are reported for upper and lower intervals of the difference. *Statistically significant. † Approaching statistical significance.

Table 6
Vowel acoustic space measures and F2i/F2u ratios for children with cerebral palsy.

	PRE	POST	FUP	Time Main Effect		Post-hoc Contrasts PRE-POST		PRE-FUP		POST-FUP	
	Median (lower quartile, upper quartile)			χ^2	*p*	*Z*	*p*	*Z*	*p*	*Z*	*p*
Sentences Full Vowel Space Area (Hz^2) n = 16	250933 (123264, 354,813)	182,678 (62293, 250,643)	149,295 (53978, 214,606)	6.13	.05*	−1.34	.18	−2.84	< .01*	−1.34	0.18
Sentences Dys Vowel Space Area (Hz^2) n = 13	256381 (146549, 358,472)	182,928 (35524, 238,589)	155,105 (54089, 206,317)	6.00	.05*	−1.71	.08†	−2.90	< .01*	−0.66	0.51
Single words Vowel Space Area (Hz^2) n = 12	223575 (72053, 256,222)	238,445 (77332, 287,080)	275,190 (230003, 351,533)	4.67	.09†	−0.31	.75	−2.12	.03*	−1.33	0.18
SentencesFull F2i/F2u n = 16	1.08 (1.05, 1.09)	1.06 (1.05, 1.08)	1.07 (1.05, 1.07)	0.87	.65						
SentencesDys F2i/F2u n = 13	1.081 (1.06, 1.10)	1.065 (1.04, 1.09)	1.070 (1.06, 1.08)	1.08	.58						
Single words f2i/f2u n = 12	1.05 (1.04, 1.09)	1.06 (1.03, 1.09)	1.08 (1.07, 1.10)	0.67	.72						

"Full" refers to the full cerebral palsy (CP) group; "dys" refers to the dysarthria-only CP subgroup, "single" refers to the single words condition, "FUP" refers to follow-up. Vowel acoustic space values are in Hz^2; F2i/F2u is the ratio of the mean F2 values of /i/ and /u/ in log_{10}Hz. Vowel acoustic space values are in Hz^2; F2i/F2u is the ratio of the mean F2 values of /i/ and /u/ in log_{10}Hz. *Statistically significant. † Approaching statistical significance.

for POST- were significantly longer than the PRE-treatment durations for the sentence production data ($t = 3.1, p < .01$). However, PRE to FUP comparisons approached significance, suggesting longer vowel durations for sentence productions at FUP ($t = 1.92, p = .06$). All other comparisons were non-significant (see Table A3 in Appendix A).

3.3. *Acoustic vowel space*

Acoustic vowel space and the ratio of F2i/F2u, were measured at each time point. The number of participants contributing to vowel space area and ratio of F2i/F2u analyses are reported for each task. See Tables A1 and A2 of Appendix A for F1 and F2 values for each of the vowels used in the acoustic vowel space calculations.

3.3.1. *CP group*

Results are reported in Table 6, which includes the medians, lower and upper quartiles and results of statistical tests for acoustic vowel space area and F2i/F2u. For sentence productions, statistically significant decreases were found for acoustic vowel space following treatment. This was the case for both the full CP group (n = 16, $\chi^2(2) = 6.13, p = .05$) and the dysarthria-only CP sub-group (n = 13, $\chi^2(2) = 6.00, p = .05$) (see Fig. 2). For single word productions, increases in acoustic vowel space approached statistical significance (n = 12, $\chi^2(2) = 4.67, p = 0.09$) following treatment. Post-hoc analysis with Wilcoxon signed-rank tests was conducted. Median acoustic vowel space for sentences produced at PRE-, POST- and FUP-treatment revealed that the only significant differences were between PRE- and FUP-treatment, with medium effect sizes in both the full group ($Z = 2.84, p < 0.01, r = .41$) and the dysarthria-only sub-group ($Z = 2.90, p < 0.01, r = .46$). However, PRE- to POST-treatment also approached significance in the dysarthria-only sub-group ($Z = 1.71, p = 0.08$). Post-hoc analysis for single word acoustic vowel space, using Wilcoxon signed-rank tests, showed a significant increase from PRE-to FUP-treatment with a medium effect size ($Z = 2.12, p = 0.03, r = .35$). No significant results were found for F2i/F2u. *Post-hoc* follow up analyses were conducted on each formant (i.e., F1 and F2) for each vowel from single word and sentences comparing PRE-to POST and PRE-to FUP-treatment. These paired *t* tests revealed one significant difference and three differences approaching statistical significance for vowels from sentence productions in the CP cohort (i.e., F1 /a/ Pre-FUP ($t = 1.54, p = .10$); F2 /a/ Pre-FUP ($t = 1.68$. $p = 0.10$); F1 /i/ PRE-POST, ($t = 2.56, p = 0.02$); and F1 /u/ PRE-POST ($t = 1.85, p = 0.08$). No differences were found for word productions.

3.3.2. *DS group*

There were no significant results in the single words condition for either acoustic vowel space or F2i/F2u (see Table 7). However, all four participants showed PRE- to POST-treatment gains in both acoustic vowel space and F2i/F2u in the sentence production

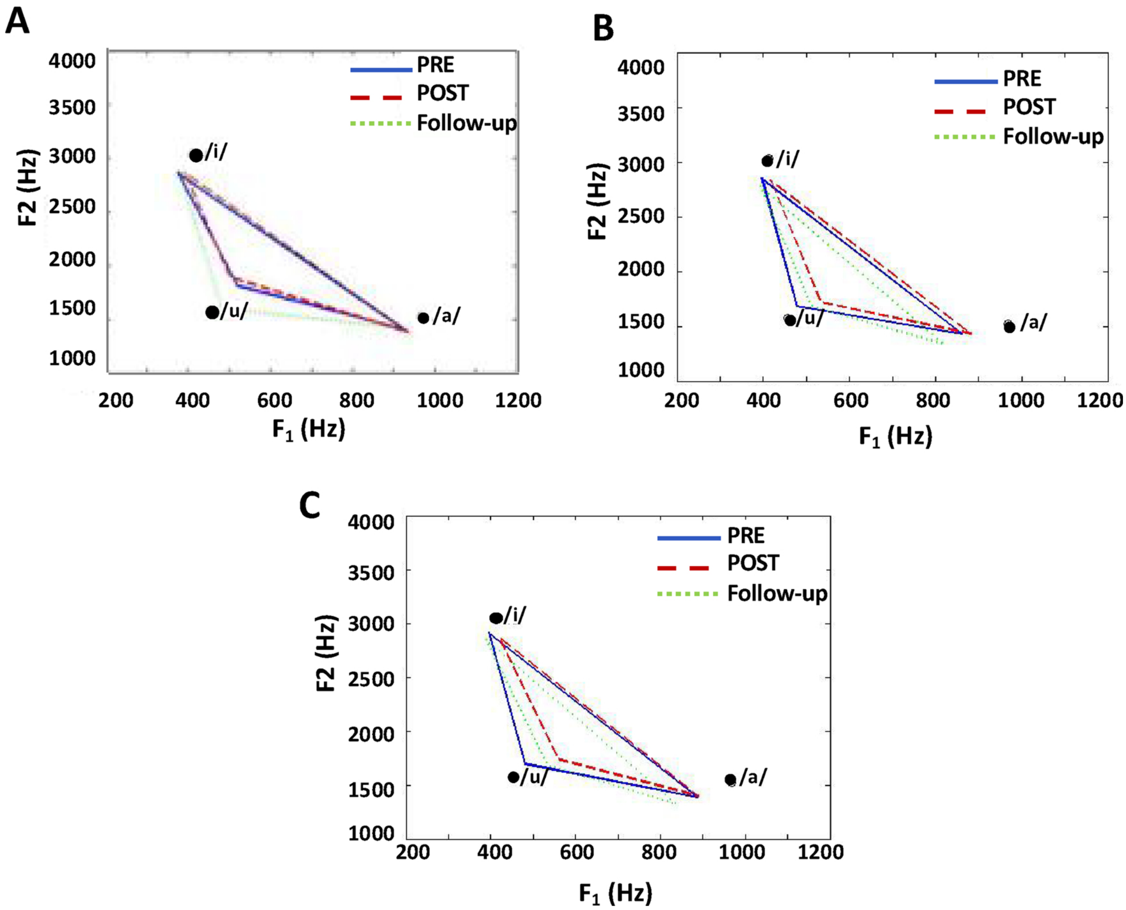

Fig. 2. Vowel triangle area for children with CP before treatment (PRE), immediately following treatment (POST), and 12 weeks after treatment (Follow-up). Panel A represents the vowel area for single words. Panel B represents the vowel area for sentences. Panel C represents the vowel area for sentences produced by the dysarthria-group only. Black solid circles represent normative values for eight-year-old males with no known speech pathologies reported in Lee et al. (1999).

condition (see Table 8 and Figs. 3 and 4 for description of results). Results for the sentence production condition are reported descriptively, as only four participants had sufficient valid tokens to calculate the variables, and no statistical tests could be carried out. *Post-hoc* follow up analyses were conducted on each formant (i.e., F1 and F2) for each vowel from single word and sentences comparing PRE-to POST and PRE-to FUP-treatment. These paired *t* tests revealed no significant differences for formants from vowels produced in single words or sentences.

Table 7
Vowel acoustics space measures and F2i/F2u ratios from single word productions for children with Down syndrome.

	PRE	POST	PRE-POST Comparison	
	median (lower quartile, upper quartile)		*Z*	*p*
Single Words Vowel space area (Hz^2) n = 8	264578 (64661, 374623)	173214 (68904, 314172)	0.70	.48
Single Words F2i/F2u n = 8	1.06 (1.02, 1.07)	1.04 (1.03, 1.06)	0.00	1.00

Vowel acoustic space areas are in Hz^2; F2i/F2u is the ratio of the mean F2 values of /i/ and /u/ in log_{10}Hz.

Table 8
Vowel acoustic space measures and F2i/F2u ratios from sentence productions for children with Down syndrome.

Participant	PRE Vowel acoustic space	POST Vowel acoustic space	PRE F2i/F2u	POST F2i/F2u
S25	282845	381906	1.08	1.09
S26	251872	825336	1.05	1.13
S28	134960	157386	1.04	1.05
S29	214446	393453	1.06	1.09

Vowel triangle areas (VOWEL SPACE AREAs) are in Hz^2; F2i/F2u is the ratio of the mean F2 values of /i/ and /u/ in $\log_{10}$Hz.

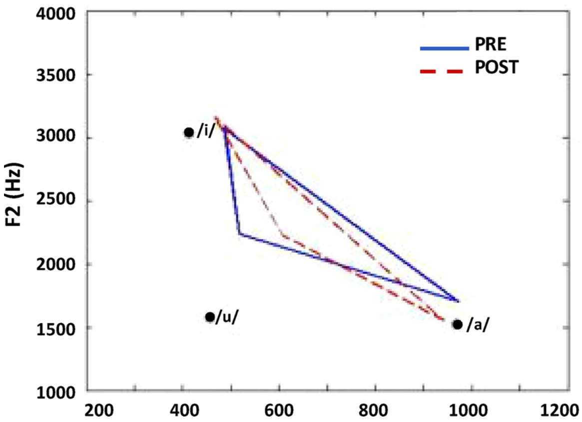

Fig. 3. Vowel triangle area for single words produced by children with DS before treatment (PRE) and immediately following treatment (POST). Solid black circles represent normative values for eight-year-old males with no known speech pathologies reported in Lee et al. (1999).

4. Discussion and conclusions

The purpose of this Phase I treatment study was to test for acoustic changes, specifically to vowel duration, acoustic vowel space and F2i/F2u, in the speech of two groups of children with dysarthria and cerebral palsy (CP) or Down syndrome (DS), who received full doses of LSVT LOUD, and who had previously shown indications of improved speech intelligibility following treatment. As is typical with Phase I research, we stress the preliminary nature of these results especially when considering findings that were marginally significant using Phase I statistical criteria. Statistically significant PRE- to POST- (CP and DS groups) and PRE- to 12 weeks follow up (FUP)-treatment (CP group only) gains in percent whole word correct, one measure of speech intelligibility, have previously been observed in the same participants. In the CP group, statistically significant results from the acoustic measures tested in the present study were: 1) PRE- to POST-treatment increases in vowel durations for sentence productions, 2) PRE- to FUP-treatment decreases in acoustic vowel space for sentence productions and 3) PRE- to FUP-treatment increase in acoustic vowel space for single word productions. Changes in acoustic vowel space found for sentence productions retained their significance when retested in a sub-group of children with CP, excluding those with moderate or greater comorbid dysfluency or apraxia of speech. There were no statistically significant results in the DS group, possibly as a result of the small sample size. However, all four participants in sentence production conditions showed PRE- to POST-treatment gains in acoustic vowel space, which were reported descriptively.

4.1. Treatment effects

The present findings suggest that following intensive voice treatment, both children with CP and children with DS demonstrated positive gains in vocal dB SPL and measures of intelligibility. Moreover, children with CP appeared to produce sentences with increased vowel durations immediately following treatment. The combined findings with respect to acoustic vowel space in both the CP and DS groups following LSVT LOUD treatment suggest potential changes in jaw and tongue movement. This is particularly true in the context of the low vowel /ɑ/ and high vowel /u/ (i.e., especially after 12-weeks follow up in the CP cohort), and to some extent, the high back vowel /i/. The evidence for changes in production of the vowel /ɑ/ is especially interesting in light of the emphasis on repetitions of “ah” in the treatment protocol, and previous findings that /ɑ/ may be particularly difficult to distinguish in dysarthric speakers (e.g. Levy et al., 2016). Repeated practice producing “ah” at various pitches, a task initially designed to target the laryngeal system, may also have the effect of improving coordination and extent of jaw opening. In dysarthric speakers with DS, the tongue may lower with the jaw, resulting in higher F1 /ɑ/ values closer to canonical targets, and overall increased vowel working space. In dysarthric speakers with CP, the tongue might initially retract rather than lower with the jaw, resulting in lower F2 /ɑ/ values closer to canonical targets. While the exact physiological changes behind the acoustic changes, and the exact mechanisms by which these changes might account for improved intelligibility, are not entirely clear, the present findings do together provide some initial

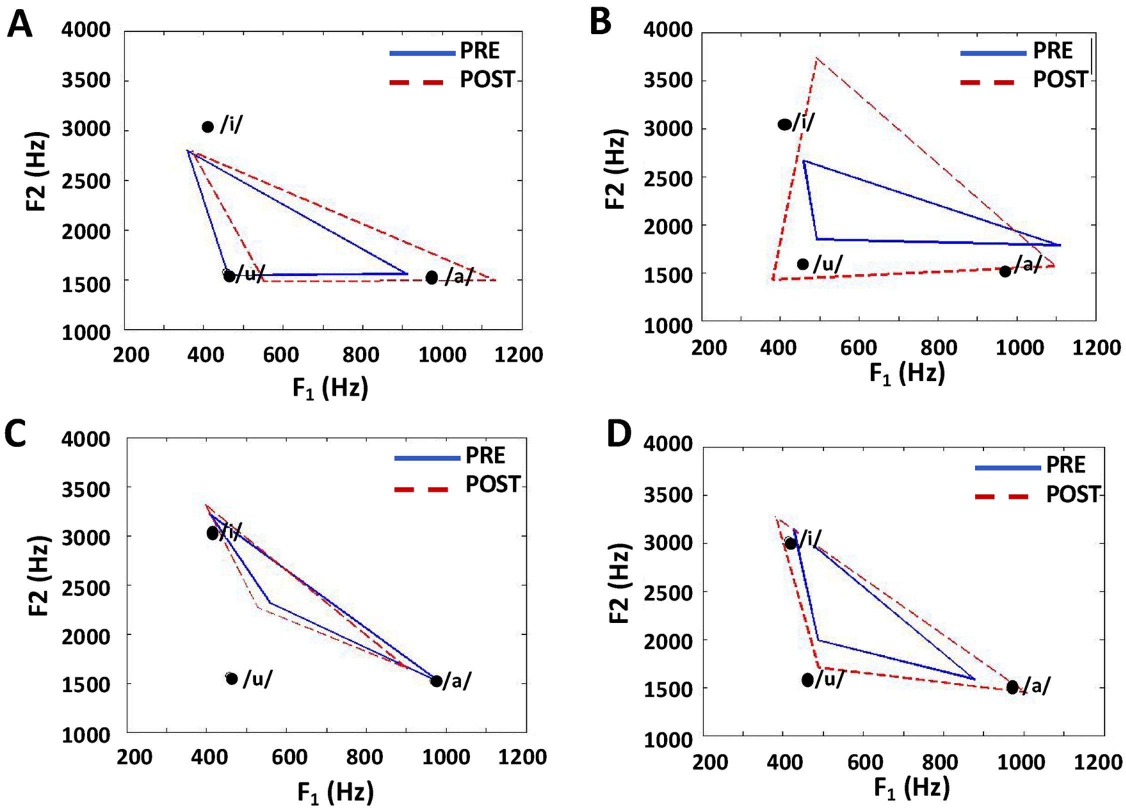

Fig. 4. Vowel triangle area for sentences produced by four individuals with DS before treatment (PRE) and immediately following treatment (POST). Panel A: S25; Panel B: S26; Panel C: S28; and Panel D: S29. Solid black circles represent normative values for eight-year-old males with no known speech pathologies reported in Lee et al. (1999).

evidence of treatment spreading effects on the articulatory system, adding to the body of similar findings in the literature on LSVT LOUD (Sapir et al., 2007; Sauvageau et al., 2015; Wenke et al., 2010; Youssef et al., 2015).

4.2. Acoustic vowel space

The CP group results for vowel acoustic space were surprising because the direction of change in the sentence production condition was a decrease in acoustic vowel space, which suggests a smaller vowel working space, and because this finding conflicted with that in the single word condition, which did show an increase in acoustic vowel space as predicted. As described above, previous dysarthria research has correlated reduced vowel working space with lower speech intelligibility (see, e.g., Higgins & Hodge, 2002; Liu et al., 2005), yet the participants in this study showed speech intelligibility increases both PRE to POST and PRE to FUP. One possibility, especially given the heterogeneity of the group's ages, neurological and dysarthria diagnoses, and dysarthria severity ratings, is that individual participants responded very differently to the therapy, and that the group study design obscures some treatment effects by aggregating results from strong responders, weak responders, and non-responders (see, e.g., discussions in Nip, 2017; Wenke et al., 2010; and Youssef et al., 2015). A second possibility is that the acoustic vowel space metric using the three corner vowels /ɑ/, /i/, and /u/, is not a sensitive enough measure of vowel working space; measures that incorporate formants from more vowels might provide more accurate representations of articulatory function (see, e.g., Lansford & Liss, 2014b; Sandoval, Berisha, Utianski, Liss, & Spanias, 2013).

It also is possible that for some participants in the sentence production condition, the treatment resulted in changes to speech physiology that, while they reduced vowel working space, were compensated for by greater articulatory precision. For example, Weismer and Kim (2010) describe a trading relationship between F1 precision and acoustic vowel space in one of their control participants: despite a smaller acoustic vowel space, the participant maintained vowel distinctiveness because of relatively little F1 variability. The conflicting findings within the present study, between single word and sentence production, could be related to task differences (i.e., repeating a sentence, which would have had increased breath support, articulatory demands and/or cognitive demands, as compared with repeating a single word), and/or to differences in the coarticulatory contexts for the target vowels. It is possible that, due to task demands, vowel duration was shorter in the sentence production condition than in the single words condition, particularly since two of the three tokens used in the sentences ("key" and "pot") were at the end of the sentence when breath support may have been taxed. If so, longer vowel durations in the single words condition might have allowed participants additional time to reach more distinctive articulatory targets, which would result in increased acoustic vowel space. This suggests the possibility that, as for the participant in Weismer and Kim (2010) study, a strategy of greater articulatory consistency might be more effective in running speech for at least some speakers with dysarthria and CP than a strategy of producing more distinctive vowels that also reduces speaking rate.

While the comparisons between formant values for participants in this study in the sentence production condition and the norms illustrated in Fig. 2 should be treated with caution due to differences in age, dialect, speech context, and other factors, the most apparent difference is the decreased F1/ɑ/ in the CP group compared with norms for healthy speakers at PRE, POST and FUP (as plotted) using Lee et al. (1999). The vowel /ɑ/ typically features a relatively high F1 and a relatively small gap between F1 and F2 (see, e.g., the values reported in Lee et al., 1999). A lower F1 would be expected to correspond to a higher tongue position (see, e.g., Hixon et al., 2014) and reduced distinctiveness of /ɑ/ from central and mid-back vowels. The lower F1/ɑ/ values observed in the present study are inconsistent with Higgins and Hodge (2002) finding of higher F1/ɑ/ values in children with dysarthria compared with controls, but are consistent with Levy et al. (2016) finding of a trend of /ɑ/ as having the lowest intelligibility of vowels in children with CP and dysarthria. Examination of Figs. 2 and 3 suggests that the statistically significant PRE to FUP change in acoustic vowel space was primarily a manifestation of decreased F1 and F2 values for /ɑ/ at FUP and, to a lesser extent, increased F1 values for /u/, with some change in the /i/ formants (i.e., F1 /i/ PRE-POST, $t = 2.56$, $p = 0.02$). The direction of movement in F1/ɑ/ from PRE to FUP is unexpected as it suggests a further reduction in vowel distinctiveness, and is inconsistent with Youssef et al.'s (2015) finding of higher F1/ɑ/ values post LSVT LOUD. However, the lower F2/ɑ/ and higher F1/u/ are both consistent with the results of that study. The decrease in F2/ɑ/ would be consistent with increased tongue movement toward the back of the oral cavity (see, e.g., Hixon et al., 2014), and might help to offset the low F1 values by narrowing the gap between the two formants, which could explain some of the improvement in intelligibility. This combination of formant changes might be consistent with a wider opening of the jaw, without a corresponding lowering of the tongue. Finally, the finding of significant changes PRE to FUP but not PRE to POST might be explained as the result of some treatment effects taking longer to manifest than others, perhaps as some participants continued to practice skills learned in therapy, or perhaps as slow phase learning of motor skills (Boliek & Fox, 2016). This result differs from the results of both Youssef et al. (2015) and Wenke et al. (2010), who found significant PRE to POST changes that were maintained at FUP only in the latter study. This inconsistency between the findings of the present study and those of Youssef et al. and Wenke et al. could reflect differences in how LSVT LOUD treatment effects occur in children and adults, and/or in participants with different types of dysarthrias.

In the DS group, acoustic vowel space did increase PRE to POST as predicted in all four of the participants in the sentence production condition. The single-word condition did not produce a statistically significant result. However, four of eight participants also showed increased acoustic vowel space, including three of the four participants in the sentence production condition. The similarity of results occurred despite differences between the tokens used in the datasets, namely the single-word tokens were nearly all spontaneously produced (i.e., without the benefit of modeling) in contrast with the sentences, which were directly imitated by participants. Moreover, in the single word condition the token "watch" was used for /ɑ/, which might have influenced acoustic vowel space values through coarticulatory effects of the linguolabial glide persisting at mid-vowel where the formant measurements used in the calculations were taken. An examination of Fig. 4 shows striking variance in magnitude and direction of acoustic vowel space increase among the four participants in the phrase condition. Only one participant (S26, Panel B, Fig. 4) showed significant movement for the vowel /i/. In one of the three pre-treatment speech samples taken, this participant presented with exceptionally poor voice quality, which produced F2 values around 1300 Hz. As results were averaged across the three samples, the initial low value of F2 for /i/ illustrated in Fig. 4 is likely not representative of S26's typical performance. Three of four participants show noticeably higher F1 values for /ɑ/ at POST, and all four participants show lower F2 values for /u/, although these vary in magnitude. These results are consistent with previous findings with respect to F1/ɑ/ and F2/u/ post LSVT LOUD treatment (Wenke et al., 2010; Youssef et al., 2015), as well as Moura et al. (2008) observations in children with DS of 1) lower F1/ɑ/ values, which the author attributed to more limited jaw movement and mouth opening, and 2) smaller F2i/F2u ratios, which the author attributed to higher F2/u/ values due to restricted tongue movement in the high back position. As noted above, the higher F1 /ɑ/ value would be consistent with a lower tongue position and possibly a more open jaw, and a lower F2 /u/ value would be consistent with a less forward tongue position (see, e.g., Hixon et al., 2014). Both of these changes would be consistent with less centralization and more distinctive productions of the two vowels.

4.3. Limitations and future directions

As suggested above, one limitation of the present study is that the heterogeneity of the CP group in particular may have masked some treatment effects; future studies might use sub-groups chosen for common characteristics (e.g., severity of dysarthria diagnosis) with the aim of identifying factors that might predict strength of response to LSVT LOUD therapy (Boliek & Fox, 2014). Larger sample sizes also would be desirable to increase statistical power. The data used in this study were not originally collected for the purposes of analysis of formants. Thus, future studies designed for this purpose might control vowel contexts more carefully. For example, excluding tokens with glides and liquids preceding or following the vowel, or by excluding tokens which children are likely to make articulation errors on like consonant blends such as in the word "tree" (i.e., [tri], [twi], [ti], [tʃ i], and [fi]). Moreover, ensuring more repetitions of word and sentence tokens would improve the available acoustic samples for analyses. Tokens could also be chosen to allow for calculation of acoustic vowel space using an expanded set of vowels (e.g., Sandoval et al., 2013), which might be more sensitive to treatment effects. Analyses of different types of speech tasks, for example, conversational speech and read speech (for participants who have that capacity) would also be beneficial. Future research also could consider whether and how the current LSVT LOUD treatment protocol (e.g., intensity and program length), which was developed for individuals with PD, might be modified to maximize results for pediatric populations with dysarthria secondary to CP or DS (e.g., dose, maintenance), and whether or not treatment effects are similar to those of other therapies targeting multiple speech sub-systems (e.g., Pennington et al., 2010).

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Appendix A

Table A1
Children with cerebral palsy individual average formant values at 30 ms mid-section of vowels (Hz).

Participant	F1 ɑ PRE	F1 ɑ POST	F1 ɑ FUP	F2 ɑ PRE	F2 ɑ POST	F2 ɑ FUP	F1 i PRE	F1 i POST	F1 i FUP	F2 i PRE	F2 i POST	F2 i FUP	F1 u PRE	F1 u POST	F1 u FUP	F2 u PRE	F2 u POST	F2 u FUP
Single Words																		
F1001	983	1044	932	1396	1647	1544	334	304	287	2950	3088	3237	491	431	531	2187	2086	1476
F1201	1013	984	975	1118	1230	1166	453	554	543	2772	2564	2720	420	599	547	1486	1797	1280
F3	844	968	978	1323	1333	1487	404	494	434	3175	2900	2981	533	532	494	1601	1702	1632
F601	1110	1127	–	1447	1196	–	334	398	–	2982	2967	–	596	652	–	2130	2489	–
F7	1007	999	990	1644	1768	1821	323	275	318	2792	2978	2813	467	506	491	1833	1792	1739
F802	948	1015	960	1307	1378	1288	326	385	364	3279	3400	3415	509	561	600	2609	2125	1693
M1001	881	955	802	1863	1940	1713	420	405	488	2416	2827	2497	844	519	506	1672	2906	2017
M2	827	962	867	1077	1044	1166	431	474	401	2711	2537	2495	469	487	459	1416	1294	1357
M4	735	676	729	1465	1310	1205	286	277	282	3050	2605	2617	311	339	297	1346	1137	1498
M6	955	817	998	1341	1070	1351	403	349	303	3024	3025	2946	377	387	337	2297	2637	1445
M8	990	889	1007	1569	1549	1605	364	412	452	3275	3274	3299	702	481	565	1524	1286	1543
M901	851	810	915	1264	1088	1217	431	354	291	1948	2257	2565	527	532	478	1595	1397	1911
Sentences																		
F1	938	996	941	1336	1380	1282	392	455	402	3132	3209	3098	374	364	404	1344	1281	1296
F1001	929	860	719	1458	1277	1511	347	345	305	3256	2733	2753	414	454	476	1465	1690	1625
F1201	849	905	908	1235	1510	1238	479	492	452	2761	2569	2787	535	639	657	2255	2163	1947
F1202	611	627	516	1631	1642	1040	438	459	424	2535	2160	2157	395	320	385	1321	1445	1525
F3	857	881	837	1410	1308	1216	393	400	382	3021	3008	2984	513	503	500	1869	1884	1844
F601	1128	1092	–	1978	1785	–	453	549	–	3056	2792	–	639	965	–	1998	1992	–
F7	1025	1080	1040	1782	1792	1815	416	436	427	2762	2745	2785	488	492	495	1472	1490	1658
F801	1025	1001	906	1309	1407	1573	444	419	371	3255	3377	3013	494	553	740	2163	2019	2097
F802	971	1030	978	1375	1512	1357	330	408	514	3453	3634	3561	500	633	614	1749	1672	2013
M1001	764	666	768	1668	1691	1592	449	444	461	2414	2302	2317	561	537	529	2024	1865	1862
M2	832	885	816	1243	1152	1168	412	416	390	2543	2565	2462	486	533	474	1412	1635	1535
M4	725	599	685	1277	1534	1138	264	271	276	2743	2551	2638	353	358	362	1394	1140	1397
M5	998	969	833	1483	1269	1462	426	474	497	2817	3057	3282	519	622	599	1387	1658	1402
M6	617	845	692	1455	1337	1290	357	343	390	2439	3111	2550	519	468	380	2079	1943	1919
M8	856	850	923	1235	1190	1276	327	384	316	2836	2747	2678	383	546	539	1637	1986	1602
M9	677	849	735	998	1151	1075	423	389	289	2629	2859	2615	472	563	517	1345	1620	1544
M901	938	996	941	1336	1380	1282	392	455	402	3132	3209	3098	478	364	404	1344	1281	1296

"FUP" refers to follow-up.

Table A2
Children with Down syndrome individual average formant values at 30 ms mid-section of vowels (Hz).

Participant	F1 ɑ PRE	F1 ɑ POST	F2 ɑ PRE	F2 ɑ POST	F1 i PRE	F1 i POST	F2 i PRE	F2 i POST	F1 u PRE	F1 u POST	F2 u PRE	F2 u POST
Single Words												
S21	1465	1088	1799	1583	529	573	3894	3357	616	572	2296	
S22	777	716	1581	1391	460	451	2570	3180	493	427	2595	2406
S23	1142	893	1817	1760	499	404	3071	3095	449	714	1966	2337
S24	998	789	2074	1237	545	513	3378	3497	552	774	1893	2781
S25	987	921	1544	1526	354	400	3019	2888	419	352	1797	1614
S26	1163	1211	2050	1928	419	401	3395	3565	562	638	2608	2715
S28	739	798	1281	1231	546	552	2789	3047	577	704	2546	2332
S29	848	1039	1464	1593	484	428	2972	3014	508	551	1791	1885
Sentences												
S25	912	1134	1554	1490	358	371	2798	2793	462	553	1543	1482
S26	1111	1100	1783	1563	459	492	2664	3737	492	380	1846	1423
S28	988	903	1502	1651	407	399	3225	3236	558	528	2313	2267
S29	879	1008	1583	1443	426	382	3147	3272	487	489	1990	1704

"FUP" refers to follow-up.

Table A3
Average vowel durations (s) for children with cerebral palsy (CP) and children with Down syndrome (DS).

	PRE			POST			FUP		
	ɑ	i	u	ɑ	i	u	ɑ	i	u
CP Group (full) - Sentences	0.288	0.305	0.403	0.274	0.386	0.435	0.281	0.424	0.507
CP Group (dysarthria only) - Sentences	0.203	0.289	0.389	0.266	0.349	0.447	0.272	0.329	0.454
CP Group – Single Words	0.383	0.536	0.639	0.379	0.538	0.569	0.400	0.535	0.676
DS Group –Sentences	0.218	0.256		0.228	0.298				
DS Group – Single Words	0.314	0.281	0.353	0.267	0.288	0.414			

"FUP" refers to follow-up.

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