Pennington L, Lombardo E, Steen N, Miller N.

Acoustic changes in the speech of children with cerebral palsy following an intensive program of dysarthria therapy.

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Abstract

Background: The speech intelligibility of children with dysarthria and cerebral palsy has been observed to increase following therapy focussing on respiration and phonation.
Aims: To determine if speech intelligibility change following intervention is associated with change in acoustic measures of voice.
Methods and Procedures: We recorded 16 young people with cerebral palsy and dysarthria (9 girls; mean age 14 years, SD 2; 9 spastic type, 2 dyskinetic, 4 mixed; 1 Worster Drought) producing speech in two conditions (single words, connected speech) twice before and twice after therapy focusing on respiration, phonation and rate. In both single word and connected speech we measured vocal intensity (RMS), period-to-period variability (Shimmer APQ, Jitter RAP and PPQ) and harmonics to noise ratio (HNR). In connected speech we also measured mean fundamental frequency, utterance duration in seconds and speech and articulation rate (syllables per second with and without pauses respectively). All acoustic measures were made using Praat. Intelligibility was calculated in previous research.
Outcomes & Results: In single words statistically significant but very small reductions were observed in period-to-period variability following therapy: Shimmer APQ -0.15 (95% CI -0.21 to -0.09); Jitter RAP -0.08 (95% CI -0.14 to -0.01); Jitter PPQ -0.08 (95% CI -0.15 to -0.01). No changes in period-to-period perturbation across phrases in connected speech were detected. However, changes in connected speech were observed in phrase length, rate and intensity. Following therapy, mean utterance duration increased by 1.11 seconds (95% CI 0.37 to 1.86) when measured with pauses and by 1.13 seconds (95% CI 0.40 to 1.85) when measured without pauses. Articulation rate increased by 0.07 syllables per second (95% CI 0.02 to 0.13); speech rate increased by 0.06 syllables per second (95% CI <0.01 to 0.12); and intensity increased by 0.03 Pascals (95% CI 0.02 to 0.04). There was a gradual reduction in

mean fundamental frequency across all time points (-11.85 Hz, 95% CI -19.84 to -3.86). Only increases in the intensity of single words (0.37 Pascals, 95% CI 0.10 to 0.65) and reductions in fundamental frequency (-0.11 Hz, 95% CI -0.21 to -0.02) in connected speech were associated with gains in intelligibility.

Conclusions & Implications: Mean reductions in impairment in vocal function following therapy observed were small and most are unlikely to be clinically significant. Changes in vocal control did not explain improved intelligibility.

What is already known on this subject: Motor disorders of cerebral palsy affect control and coordination of movements for respiration, phonation, resonance, articulation and prosody. Therapy focussing on respiration, phonation and rate has been associated with increases in speech intelligibility for children with cerebral palsy and dysarthria.

What this study adds: Therapy focussing on respiration, phonation and rate may lead to a stronger vocal signal and increased coordination of speech movements. However, these acoustic changes are small and are unlikely to be perceived by listeners. Vocal changes have not been found to account for observed increases in intelligibility.

Clinical implications of this study: Intervention targeting respiration, phonation and rate could be offered to children with dysarthria and cerebral palsy who use speech as their main means of communication but whose intelligibility is limited. Therapy outcome should be measured at the level of activity, that is, intelligibility of speech.

Introduction

Cerebral palsy has a prevalence of 2-3 per thousand live births and is the most common cause of physical disability in childhood (Himmelmann and Uvebrant, 2014, Kirby et al., 2011, Reid et al., 2011, Cans et al., 2008). It is estimated that around 22% of children with cerebral palsy have speech intelligibility limitations due to dysarthria (Nordberg et al., 2013, Parkes et al., 2010, Stanley et al., 2000). The communication difficulties which arise from limited intelligibility have important consequences for children's social development and wellbeing. Children and young people with cerebral palsy who have communication difficulties are at greater risk of poor quality of life and limited social participation than their peers with cerebral palsy who can communicate well and those who are typically developing (Dang et al., 2015, Fauconnier et al., 2009, Colver et al., 2014, Dickinson et al., 2007).

Dysarthria is associated with spastic, dyskinetic, and ataxic types of cerebral palsy (Bax et al., 2006) and children with the different types have been noted to share similar perceptual speech characteristics (Workinger and Kent, 1991). All speech systems: respiration; phonation; resonance; prosody; and articulation are usually involved, although the degree of impairment in the function of each subsystem can vary between individuals (Workinger and Kent, 1991). Limited respiratory control has been observed in shallow tidal volume, use of paradoxical breathing patterns, and difficulties coordinating exhalation with phonation (Solomon and Charron, 1998, Kwon and Lee, 2013, Kwon and Lee, 2014). Impairment in laryngeal function has been perceived in harsh and breathy voice qualities, reduced pitch range, and unwanted pitch breaks (Workinger and Kent, 1991, Ciocca et al., 2002). Difficulties regulating and coordinating respiration and phonation can create an impaired and often weak vocal signal with low signal (harmonics) to noise ratio (Fox and Boliek, 2012, Jeng et al., 2006, Patel, 2003, Wit et al., 1993). Impairment of the movements of the articulators can reduce the phonemic repertoire; perceived voice, place and manner errors are common (Lee et al., 2014, Ansel and Kent, 1992, Whitehill and Ciocca, 2000). However, acoustic analysis suggest that phonetic contrasts such as fricative-affricate and voice-voiceless contrasts may be marked but not perceived by listeners (Ansel and Kent, 1992), possibly because of the weak aero-acoustic signal. Recent research suggests that articulatory, respiratory, and prosodic impairment are most strongly associated with intelligibility for children and adults with cerebral palsy who have dysarthria (Lee et al., 2014, Schölderle et al., 2016).

Texts commonly advocate that interventions for dysarthria should focus on improving respiratory support (Kent and Read, 1992, Yorkston et al., 1999) and increasing the coordination of respiration with phonation, to reduce air wastage. Following the source filter model of speech production (Kent and Read, 1992, Fant, 1960), it is hypothesized that together, these actions will create a clearer voice signal, which can support longer utterances. The resulting voice may be perceived as consistently louder. Improved respiratory and phonatory coordination may also enable speakers to increase the variation in their pitch range (Duffy, 2005, Strand, 1995). The two interventions which have been tested most extensively with young people with cerebral palsy are Lee Silverman Voice Therapy LOUD (LSVT) (Fox and Boliek, 2012, Boliek and Fox, 2016, Fox et al., 2008) and the Speech Systems Approach (Pennington et al., 2013, Miller et al., 2013, Pennington et al., 2010). Both programmes are intensive, requiring 16-18 sessions over 4 to 6 weeks. Both use motor learning principles of high intensity practice, randomisation of target speech behaviours and fading of feedback (Schmidt and Lee, 2005, Maas et al., 2008). One of the main differences between the two programmes is that LSVT Loud® focuses on respiration and phonation and self-monitoring, and the Speech Systems Approach additionally targets rate, so that speakers have sufficient time to move from one articulatory positon to the next within words and leave sufficient time between words to mark lexical and phrase boundaries(Duffy, 2005). A further difference between LSVT LOUD and the Speech Systems Approach, is that in the latter not all children are encouraged to increase their loudness. Children who have generally weak, quiet voices are encouraged to speak more loudly across a phrase, whereas those whose speech shows inappropriate variability in loudness/intensity across a phrase are encouraged to use a ‘smooth’ or ‘nice and easy’ voice to maintain even respiratory effort across a phrase.

In early phase trials of LSVT with children who have spastic type cerebral palsy, Fox and Boliek have observed change in acoustic and perceptual measures of voice quality post therapy. Statistically significant gains in maximum performance tasks including loudness (dB SPL) of sustained phonation and period-to-period variability of fundamental frequency (jitter) and amplitude (shimmer) have been found in two studies (Boliek and Fox, 2016, Fox and Boliek, 2012). However, gains have not been observed across all voice characteristics; average fundamental frequency and loudness (dB SPL) were not observed to change in sustained phonation, and gains made in loudness in untrained phrases were not maintained. Although intelligibility increased by an average of 7% at 6 weeks post therapy, gains were not maintained at 12 weeks follow-up. Nevertheless, perceptual changes in voice and speech characteristics were observed by trained clinicians blinded to the time of voice recordings. These expert listeners preferred the speech of children in post therapy recordings over pretherapy recordings in terms of speech loudness, loudness variability, pitch, pitch variability, overall voice quality and articulatory precision (Boliek and Fox, 2016, Fox and Boliek, 2012). On average, children’s parents rated their child’s speech as louder and sounding more natural and less hyponasal, hypernasal and strained after therapy. Parents also reported that children were more likely to start conversations and were less frustrated by their speech following therapy (Boliek and Fox, 2016).

Pennington, Miller and colleagues found that following the Speech Systems Approach parents of children and adolescents with spastic and/or dyskinetic cerebral palsy also reported increases in communicative participation (Pennington et al., 2013, Thomas-Stonell et al., 2010)) Following the Speech Systems Approach the proportion of words understood by familiar and naive listeners in single word and connected speech increased, with mean absolute gains in intelligibility of 10 to 15% across groups and conditions and maintenance of effects at 6 and 12 week follow-up (Pennington et al., 2013, Pennington et al., 2010). However, for the Speech Systems Approach there was no association between change in intelligibility and communicative participation as measured by the FOCUS (Pennington et al., 2013, Thomas-Stonell et al., 2010). In their study with adolescents (Miller et al., 2013), trained therapists rated all vocal characteristics as impaired using the GRBAS scale (Hirano, 1981, Miller et al., 2013) at the start of therapy. Following therapy reductions in asthenia were on the borderline of statistical significance, but no other characteristics were perceived to have changed in severity. Only reductions in ratings of asthenia were associated with gains in intelligibility, and this association was weak, with one unit change on asthenia rating on the GRBAS scale being associated with 11% gains in intelligibility.(Hirano, 1981)

Thus, current research into therapy focussing on respiration and phonation shows promising results in terms of activity (intelligibility) and participation level outcomes. However, the effects on impairment at the level of body function are less clear cut. It has not yet been identified if acoustic phonetic changes are responsible for (some) improvements in intelligibility. To gain further understanding of the impact of the Speech Systems Approach on vocal function we conducted analysis of the speech of the adolescents who participated in our original group study reported by Pennington et al (2010) and Miller et al (2013) using acoustic measures relating directly to respiratory and phonatory control. We then examined the association between these acoustic impairment measures and gains in percentage of words understood by listeners.

We hypothesized that following therapy improved respiratory and phonatory control would be evidenced in increased intensity of voice, greater signal to noise ratios, and increased regularity in vocal fold vibration (i.e. reduced period-to-period variability). We also hypothesized that increased control would enable children to produce longer utterances in connected speech. As speaking rate was targeted in therapy, we expected this to reduce. Finally, we hypothesized that changes in vocal parameters would explain previously observed gains in intelligibility.

Design

This phase II study (Robey and Schultz, 1998, MRC, 2000) uses an interrupted time series design in which participants acted as their own controls. Voice and intelligibility measures were taken on two separate days at each of the following time points: six weeks before therapy; one week before therapy; one week after therapy; and six weeks after therapy. The pre-therapy measures acted as a baseline. The study was approved by Sunderland Local National Health Service Research Ethics Committee. Young people aged 16 years and above provided written consent. The guardians of children aged below 16 years provided written consent to their participation in the study and children also gave written or verbal assent. Findings on intelligibility and perceptual measures of voice impairment severity have previously been reported (Miller et al., 2013, Pennington et al., 2010) for this cohort. Here we present data on acoustic measures of their speech and test associations with intelligibility. All methods reported here, except acoustic measures and analysis, have been presented in previous reports (Miller et al., 2013, Pennington et al., 2010).

Methods

Participants

Sixteen young people with cerebral palsy and dysarthria were recruited to the study (9 girls; age 12-18 years, mean = 14 years, SD = 2) via their local NHS speech language pathologists across the North East of England. As previously reported, nine participants had spastic type cerebral palsy, two had dyskinetic type, four had mixed (spastic and dyskinetic), and one had Worster Drought (Clark et al., 2009). All participants were native speakers of British English. Previous investigations by medical personnel determined that participants had no bilateral hearing impairments greater than 50 dB HL, no severe visual impairments, and no cognitive impairments that would prevent them from following task instructions. Gross Motor Function Classification Scale ER (Palisano et al., 2007) was used to classify children's mobility and posture. Scores ranged from 1 – 5, (*Med* = 4), indicating that most children in the study were unable to walk independently and required adaptive seating in order to maintain posture and facilitate hand control. The severity of dysarthria was rated as moderate to severe by referring speech language therapists. At the start of the study unfamiliar listeners could understand 18.7 to 62.0% of participants' speech in single words (*M* = 40.9%, *SD* = 13.4) and from 3.0 to 61.0% of their connected speech (*M* =28.6%, *SD* = 20.9) (Pennington et al., 2010). Perceptual ratings of the severity of the voice impairment using the GRBAS scale (Hirano, 1981) indicated that all participants had voice disorders. At the start of the study speech language pathologists who were blind to the study aims rated individual characteristics of voice impairment: Grade median = 2 (IQR = 1-2); Roughness median = 1 (IQR 0-1); Breathiness median = 1 (IQR 0-2); Asthenia median = 1 (IQR 0-2); Strain median = 1 (IQR 0-2). (0= normal, 1 = mild, 2 = moderate, 3 = severe) (Miller et al., 2013). Characteristics of individual participants are found in Table 1.

Insert Table 1 about here

Listeners

As previously reported in Pennington et al (2010) 128 adults who had no experience of interacting with children who have speech disorders were recruited from local business corporations to act as listeners in the study and rate the intelligibility of participants' speech. All listeners were aged between 18 and 58 years of age (43 Male; 85 female) and were familiar with the regional accent of the participants. All listeners provided written consent and confirmation that they did not experience hearing difficulties; for example, they did not need to increase the volume on television and radio compared their friends and family.

Intervention

As reported by Pennington et al (2010) each participant received a course of individual therapy which focused on stabilizing respiratory and phonatory effort and control, speech rate and phrase length/syllables per breath which was developed from previous therapy texts (Hodge and Wellman, 1999, Strand, 1995, Yorkston et al., 1999, Duffy, 2005). At the start of the therapy program participants were encouraged to coordinate expiration with phonation to generate a clear voice in an open vowel following a therapist's model. As there is variation in perceptual characteristics of dysarthria in cerebral palsy, the feedback given to help participants achieve a clear voice differed according to their predominant vocal characteristics. If children exhibited weak, breathy, quiet voices they were encouraged to use a louder, stronger voice. Those whose speech showed inappropriate variation in loudness were encouraged to use a smooth voice, to maintain even loudness across a phrase and conversational turn. The therapist used a short cue to achieve the new voice relating to the participant's perceptual target; for example, 'Good, that was strong'; 'Smooth all the way along.')

Once participants were able to generate a clear voice consistently in open vowels they named this voice (e.g. 'my clear voice', 'strong voice'). They then practised using the new voice in a hierarchy of exercises in which utterance length, cognitive effort and feedback from the therapist were graduated. The programme started with exercises for single syllable words and moved to multi-syllable words and phrases. At each target utterance length participants practised their new voice in repetition and moved to picture naming. At phrase level cognitive load was further increased by eliciting speech in question and answer formats and games such as ‘Who am I?’ The final level in the hierarchy was conversational speech. In longer phrases participants were encouraged to ‘chunk’ utterances into manageable breath groups that could be achieved with their target voice and rate. At each level in the hierarchy feedback from the therapist was faded from feedback after every single attempt to feedback after every ten attempts, in order to improve skill retention. Participants were also asked to reflect on their own productions, with questions such as ‘how did that feel?’ Participants progressed from one hierarchical level to the next when the therapist judged that they had maintained controlled respiration/phonation over the entire segment of speech in 90% of speech behaviours elicited in an exercise (e.g. 9 out of 10 single words repeated following the therapist’s model with feedback after each attempt; 9/10 three syllable phrases elicited in picture naming with feedback after every tenth production). Therapy sessions followed a set format: 1) open vowels following the therapist model; 2) repetitions of familiar phrases used by participants in everyday life; 3) practice at the target level; 4) random practice of any speech behaviour from current and previous target level (e.g. for a participant working on three syllable phrases: open vowel; picture naming of single syllable word; naming picture of three syllable word; repletion of single syllable word etc.). Participants received three 35-40 minute individual sessions of therapy per week for 6 weeks at school. Thus, the therapy followed motor learning principles of intensive practice, random presentation of speech targets, provision of knowledge of results and fading of feedback.

Measures and Procedures

Each participant was audio recorded on two separate days at four different time points: six weeks before therapy (Recordings A and B); one week before therapy (C and D); one week after therapy (E and F); and six weeks after therapy (G and H). Speech was elicited in two conditions: single words and connected speech. Single word speech was elicited using the Children's Speech Intelligibility Measure (CSIM) (Wilcox and Morris, 1999), in which children repeat a list of fifty single words. Items in each list of the CSIM are selected from a corpus of 600 words and lists are balanced in length and complexity. Different lists from the CSIM were randomly allocated to the children's recording sessions; no list was used more than once in the study. Cartoon story sequences containing four cartoons were used to elicit connected speech. Four sets of cartoon pictures were used, and were randomly allocated to each participant with the proviso that each sequence was used only twice with each participant. Recordings were made in quiet rooms at the participants' schools using an EDIROL R1 digital recorder and an AKG C420 head-mounted microphone. Speech files were then digitized using Creative Wave (Creative Technology Limited, 2008) software and sampled at 48 kHz.

1. Intelligibility. To calculate average intelligibility recordings were listened to by three listeners blind to the speaker and time of recording. Each listeners was randomly allocated three recordings, comprising single word and connected speech, with the constraint that they heard a child only once. Thus, recordings from the same speaker were heard by different listeners and each recording could be allocated to be heard first, second or third for individual listeners. The order of presentation of single word and connected speech was randomized for each recording for each listener. This allocation schedule was devised to maximize variation in listeners and reduce learning effects and required 128 listeners (each listener heard three recordings; each recording was heard by three listeners; 16 participants

recorded twice at four time points = 128 recordings in total).

For the listening task, listeners selected the single words they heard from lists of 12 phonetically similar items. In the connected speech condition listeners transcribed each phrase orthographically. Recordings were played to individual listeners from a Toshiba Satellite Pro M40 laptop computer using iTunes 8.2 (Apple Inc, 2008) through Logitech X120 speakers placed one metre from the listener. Recordings were played only once. All listeners were blind to the time points of the speech they were rating. Mean intelligibility was calculated across the three listeners for each recording for single word speech and connected speech separately. Intelligibility data were reported in Pennington et al (2010).

2. Objective measure of voice impairment. Single word and connected speech data were analysed acoustically. From each single word list nine words were analysed: two of the first five words, two of the last five words, and five words from the middle forty words were randomly selected. Conversational speech data were divided into 'pause groups', distinguished by periods of silence that exceeded the usual boundaries of periods of silence normally exhibited by individual speakers. Four pause groups were analysed. As participants could show variability in loudness performance across and within pause groups, we ensured that we captured this variation by selecting for analysis their phrase with the greatest average intensity and three other randomly selected exemplars. Acoustic analysis was conducted in PRAAT using waveforms and spectrograms (Boersma and Weenink, 2010). As standard in acoustic analysis, a combination of measures were used as no single measure corresponds to human auditory perception (Yiu et al., 2000). Table 2 provides an explanation of all measures and reference values for the typically developing population where available.

Insert Table 2 here

Inter-rater reliability

A second researcher independently analysed a randomly selected 15% of each participant’s single word and connected speech data blind to the first rater's measurements. For the analysis of single words the second researcher manually selected the whole vowel using PRAAT. Where a word had more than one vowel the stressed vowel was selected. Agreement between the two raters was high for all single word measures (see Table 2) For connected speech samples agreement between raters on pause group boundary position was not significantly different (X^2 = 0.439). Where the researchers disagreed on pause boundaries the second rater used the boundary of the first rater for reliability measurement. Agreement between raters on connected speech measures was high (see Table 2). It is notable that the agreement for single word data whilst still acceptable is lower than for connected speech. This is due to the agreement check for single words comprising both the selection of the waveform to be examined and measurement of its acoustic properties. For connected speech sections of the waveform to be analysed were agreed prior to measurement, following other research designs (e.g. Boliek and Fox, 2016) in which similar levels of agreement were observed.

Statistical Analyses

As in our original study, we calculated the mean for each acoustic measurement for each participant in both single word and connected speech for each recording. We used repeated-measures multi-level analysis of variance assuming a normal error structure to analyse change in the mean acoustic measures. We fitted contrasts to examine three potential effects: 1) An intervention effect: a difference between pre-post intervention (recordings A, B, C, D vs recordings E, F, G, H); 2) A change within each of the pre and post periods (A, B, E, F versus C, D, G, H) in conjunction with contrast 1, to assess if there was a trend over the period of the study rather than a step change in voice characteristics following the intervention; 3) A systematic change between first and second recordings within each time point (A, C, E, G versus B, D, F, H). We report interval estimates of differences in acoustic measures corresponding to each of these effects below; confidence intervals were estimated using 200 bootstrap samples. We investigated the impact of phonetic voice quality on speech intelligibility using a regression model with mixed effects. Speech intelligibility was the dependent variable. Variation in intelligibility between children and within children (between visits) were modelled as random effects. The impact of therapy was then entered as fixed effect giving a "reference model" to which each of the acoustic measures was entered in turn as a fixed effect.

Results

Change in acoustic measures of voice impairment

In our comparisons of pre-therapy recordings versus post-therapy recordings of single word speech we observed slight, statistically significant mean reductions in period-to-period variability in amplitude (shimmer APQ) and frequency (jitter RAP% and PPQ%). Vocal intensity (RMS) and harmonics to noise ratio of single word speech did not change from pre to post therapy. s. The duration of breath groups and rate of speech and articulation increased post therapy. Mean duration of breath groups with and without pauses increased by approximately 1 second, articulation rate (rate without pause) increased by 0.7 syllables per second per breath group, and speech rate (rate with pauses) increased by 0.06 syllables following therapy. We observed no changes in period-to-period variability or harmonics to noise ratio in connected speech Results of pre-post therapy comparisons are shown in the left-hand columns of Table 3. Changes within pre-intervention and post-intervention periods are also shown in Table 3 (middle columns) and between pairs of recordings at each time point (right-hand columns). With the exception of fundamental frequency, which reduced by approximately 10 Hz across each of the four time points, and RMS which reduced from recording F (second recording one week after therapy), results showed no significant change within pre- and post-intervention periods or between pairs of recordings.

Insert table 3 about here

Prediction of change in intelligibility by acoustic measures

Our previous research (Pennington et al., 2010) showed that intelligibility of single words and connected speech increased following therapy. We used regression techniques to investigate the impact of acoustic measures on intelligibility change. The results of the regression models are shown in Table 4. The first row in each section of the table corresponds to a “reference model” with the impact of therapy as a single fixed effect. It shows the estimated impact of speech and language therapy was an increase of 15% in single word intelligibility ($F(1,125)$ 14.95, p<0.001, R^2 0.65)and 17.0% in connected speech intelligibility ($F(1,127)$ 16.99, p <0.001, R^2 0.68). Each of the acoustic measures was entered in turn to this reference model, as a fixed effect. The estimated change in intelligibility corresponding to a unit increase in the phonetic variable is given in the left-most column of numbers in Table 4. In general, the confidence intervals associated with the estimated change in intelligibility include zero which would suggest that the acoustic variables have no impact on the change in intelligibility associated with intervention. There are two exceptions. An increase of one Pascal in RMS would be associated with 37% increase in intelligibility of single words following intervention (NB mean change observed was 0.06 Pa; change of 1 Pa would be very large in conversational speech). An increase in one Hz in Mean F0 would be associated a with a 0.11% reduction in intelligibility of connected speech following intervention. However, the estimated impact of the intervention is still statistically significant and only slightly reduced when adjusted for the effect of RMS ($F(2,125)$ =11.87, p<0.001) and mean Mean F0 ($F(2,123)$ =14.69, p<0.001) when compared with the effect of the intervention alone.

Insert Table 4 about here

Discussion

This study is the most comprehensive exploration of the speech breathing and phonatory function in dysarthria associated with cerebral palsy following intervention to date. Pre-therapy results confirm that participants had voice disorders and difficulties controlling their breathing for speech. Although some of the group measures were within normal limits in single words, there was wide variation between participants, with many falling within the pathological range. Voice impairment was clearly present in connected speech, suggesting that coupling of respiratory and phonatory systems is insufficiently controlled to accommodate the demands of the increased duration and complexity of connected speech, leading to leaking of air and reductions in the strength and stability of the vocal signal (Nip and Green, 2013, Weismer et al., 2001, Nip, 2017).

Pre-therapy measures of voice are similar to those currently available for children with cerebral palsy (Clarke and Hoops, 1980, Boliek and Fox, 2014) and adults with acquired neurological disorders such as multiple sclerosis and Parkinson's disease (Feijó et al., 2004, Rusz et al., 2011, Rahn Iii et al., 2007). Surprisingly, mean breath group duration was similar to non-disordered samples (Mitchell et al., 1996, Hoit et al., 1990). But, all participants' speech and articulation rates were much slower than observed for young people without speech disorders, meaning that they could produce fewer syllables per breath (Nip and Green, 2013). It is noted, however, that there was wide variation within the study sample, indicating mild to severe impairment in speech function. Further studies are required to explore patterns of impairment in dysarthria in cerebral palsy and to investigate associations with severity and type of motor disorder and underlying neuropathology (Morgan and Liegeois, 2010, Liegeois and Morgan, 2012).

Following therapy focusing on respiration, phonation, and speech rate small acoustic changes were noted in speech production. Unlike in previous research by Fox and Boliek (2012), these acoustic changes were maintained for six weeks without further therapy. We propose that the increase in the intensity of the vocal signal (RMS) and reductions in period-to-period variability in amplitude and frequency in single words (Shimmer APQ, Jitter PPQ, Jitter RAP) reflect improvements in respiratory and phonatory control (Glaze et al., 1990, Brockmann et al., 2008). Increased intensity relies on the generation of greater aero-dynamic energy, through increased breath supply, and greater periodicity suggests smoother air flow in more controlled expiration. However, it must be acknowledged that the changes in measures relating to phonation were small and the underlying impairment of vocal fold vibration appears largely unchanged by the therapy, as suggested by our earlier perceptual measures of the speech examined in the current study (Miller et al., 2013). Larger gains were made in loudness control and utterance duration. In connected speech participants were able to generate and control greater breath supply to create longer, louder utterances. They also increased the number of syllables per breath group, meaning that they could produce longer utterances and produce more speech within utterances. Similar findings have been reported for individuals with dysarthria after combined Lee Silverman Voice Therapy and breathing control intervention (Solomon et al., 2004, Solomon et al., 2001). Others have indicted a positive relationship of speech rate to utterance length (Huber and Darling, 2011) which would be compatible with findings here. Further research is needed to test the clinical significance of the longer, louder and faster utterances that children could produce. Unlike other acoustic measures, speech rate and utterance duration may be feasible as outcome measures in clinical practice as they can be calculated from audio recordings transferred to freely available software and take little time to calculate from transcriptions.

Mean fundamental frequency reduced across each of the four time points. This may be due to boys experiencing pubertal voice change. However, this explanation is unlikely as the rapid changes in pitch associated with breaking voices would be noted in concomitant wider F0 SD, which was not observed (Böhme and Stuchlik, 1995). Further research is needed to examine the intervention effects on fundamental frequency and should include longer follow-up and pre- or post-pubertal young people to reduce risks of confounding hormonal effects.

Only increases in vocal intensity in single word speech and reduction in mean fundamental frequency in connected speech were associated with intelligibility change following intervention. Our regression analyses estimated that intensity increases of one Pascal would be associated with an increase in single word intelligibility of 37%. However, we observed mean increases in intensity of 0.17 Pascals. This mean change would translate to an increase of 6% if the effect of intensity increases could be considered in isolation. When the effect of intervention on intelligibility change in single words was adjusted for our observed change in intensity, the mean increase in intelligibility was reduced from 14.95% to 11.87%. Similarly, when the effect of the intervention on connected speech intelligibility was adjusted for the changes in mean fundamental frequency the mean increase in intelligibility was reduced from 16.99% to 14.69%. Our results therefore suggest that the changes in acoustic measures relating to respiration, phonation and speaking rate following the intervention had little impact on intelligibility change. The question of what (combination of) factors explain the change in intelligibility observed therefore remains. Lee, Hustad, and Weismer (2014) and Schölderle et al (2016) have demonstrated the impact of articulatory precision on intelligibility for children and adults who have dysarthria and cerebral palsy. The potential effect of the therapy on articulatory skill and ability to produce easily perceived speech contrasts therefore warrants further attention.

Lack of change in vocal parameters and/or their weak association with intelligibility may also be due to variability within and between participants. Small samples are known to have wider variation on normally distributed measures than larger samples. Limited resources and the labour-intensive nature of acoustic analysis meant that we were only able measure the acoustic parameters of nine words and four pause groups from each participant at each time point in the current study. The participant sample size is also quite small. Participants varied in both the severity of impairment in individual measures prior to therapy and in the amount of change made following intervention. Thus the study may lack power to detect true differences in the planned comparisons. It is also possible that participants' underlying motor disorder enabled them to change in one or more of the individual measures, but the specific measures on which they changed and the amount by which they changed was not consistent across the group, as previously observed by Fox and Boliek (Fox and Boliek, 2012). Furthermore, the therapy provided did not specify how participants should achieve their clearer voice or involve knowledge of performance. Participants could have made different physiological responses to the instructions, leading to varying impacts on individual vocal parameters measured here. Patterns in impairment across acoustic measures and response to therapy by individuals and groups of participants sharing similar vocal characteristics at the start of therapy should be investigated to provide indications on who may respond best to the therapy and the potential development of clinical subgroups.

This exploratory study has provided tentative evidence for changes in vocal intensity, utterance duration and speech rate following therapy that focusses on respiratory and phonatory control. However, the study has several limitations which reduce the strength of conclusions we can currently draw. As stated above, the participant sample size and the acoustic dataset were small. Participants varied considerably in most of the acoustic measures at the start of therapy, necessitating large changes in vocal parameters in relation to intelligibility to reach statistical significance. Our selection of four pause groups in connected speech always included the loudest pause group. Although this selection allowed for the consistent influence of loudest pause groups, the resulting measures are not true averages. Future research should examine the direct association between acoustic parameter and intelligibility of single words and pause groups. Larger studies may detect associations between acoustic parameters and intelligibility and will enable us to examine if patterns in acoustic parameters can define clinical subgroups of children with dysarthria.. The participants in this study acted as their own controls, with a baseline period in which no intervention was provided. Although change was only observed in fundamental frequency prior to therapy we cannot definitely conclude that other changes we observed were due to therapy that the participants received.

Conclusion

Young people with dysarthria associated with cerebral palsy can increase their speech intelligibility and vocal intensity, utterance duration and speech and articulation rate following therapy that focusses on respiratory and phonatory control and speech rate. Changes in acoustic measures relating to phonation observed in the study were small and suggest that underlying impairment of phonation is not addressed by the intervention. Furthermore, association between change in vocal parameters and gains in speech intelligibility has not been established. Further research is needed to understand the impact of impairments across the vocal tract in dysarthria associated with cerebral palsy and how therapy may bring about change in intelligibility for individuals.

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Table 1 Participant characteristics

Pt	Age	Sex	CP Type	GMFCS	Grade (GRBAS)	MLU in words	F0 Hz (mean recordings A and B)	Single word % intelligibility				Connected speech % intelligibility			
								T1	T2	T3	T4	T1	T2	T3	T4
A	15	F	Spastic and dyskinetic	4	2	6.9	288.55	18.67	13.33	22.00	40.00	7.10	7.90	8.63	20.33
B	15	M	Dyskinetic	2	1	6.4	140.83	48.00	40.67	56.67	51.33	37.80	43.77	85.83	74.13
C	14	F	Spastic and dyskinetic	5	2	8.4	286.37	50.67	43.33	66.67	68.00	17.20	54.83	46.80	57.10
D	15	M	Spastic	4	1	6.4	166.40	62.00	84.00	85.00	83.00	55.90	67.57	81.03	98.00

E	12	M	Spastic	4	1	5.8	237.79	56.67	62.00	50.00	90.67	61.03	70.47	34.40	98.73
F	12	M	Spastic	2	2	6.5	252.06	24.00	12.00	22.67	31.33	5.25	10.47	4.50	10.00
G	13	F	Spastic and dyskinetic	3	1	6.0	238.79	48.67	39.33	66.00	50.67	58.33	11.67	63.53	26.23
H	17	M	Spastic	3	1	5.7	142.30	42.67	32.00	34.00	50.67	6.40	2.60	15.25	20.93
I	18	F	Dyskinetic	2	2	7.2	336.92*	43.33	48.00	64.00	52.67	29.13	12.50	57.10	35.25
J	11	M	Spastic	4	2	6.8	337.35*	-	36.67	43.33	32.67	-	18.77	23.43	42.63
K	17	F	Spastic	5	2	6.3	260.83	47.33	45.33	56.00	52.67	15.07	16.60	9.95	10.43
L	13	F	Spastic	4	2	3.4	302.12	21.33	11.33	11.33	11.33	3.00	0.00	0.00	2.77

M	18	F	Spastic and dyskinetic	5	2	4.2	267.23	40.00	16.00	13.33	32.00	10.00	12.33	20.00	17.77
N	14	M	Spastic	4	1	5.5	281.02	50.00	22.67	69.00	77.33	35.17	1.43	80.00	54.33
P	13	F	Spastic	2	2	7.0	239.38	36.67	18.00	69.33	90.00	40.57	33.30	89.33	76.50
Q	16	F	Worster-Drought	1	2	7.1	260.50	23.33	30.67	36.67	38.00	46.77	32.10	25.00	52.00

Pt= Participant

T= Time point, 1= 6 weeks pre-therapy; 2= 1 week pre-therapy; 3 = 1 week post-therapy; 4 = 6 weeks post-therapy

- = missing data, child ill

* > 1 SD from typical F0 for age and gender of participant

Table 2 Acoustic measurements, definitions, reference values from the typically developing population, and inter-rater agreement in the current study

Acoustic measure	**Explanation of measure**	**Reference value from typically developing children**	**Inter-rater agreement**
Harmonics-to-noise ratio (HNR)	Amount of harmonic signal produced relative to the amount of noise	When the vocal folds vibrate irregularly sound waves produced lack a consistent structure; HNR of less than 20 is indicative of hoarseness (Boersma and Weenink, 2010).	SW: r = 0.80, p <0.001; CS: r = 0.99, p <0.001
Root mean square (RMS), measured in Pascals (Pa)	Overall acoustic intensity	Sound pressure level of one-to-one conversational speech in a quiet setting with speaker and listener at 1 metre apart would be expected to be around 60 dB or 0.02 Pa	SW: r = 0.99, p <0.001 CS: r = 0.99, p <.001
Shimmer: Amplitude Perturbation Quotient (Shimmer APQ)	Period-to-period variability in amplitude	Percentages greater than 3.8% are indicative of dysphonia (Boersma and Weenink, 2010)	SW: r = 0.57, p <0.001; CS: r = 0.99, p <0.001
Jitter: Relative Average Perturbation (Jitter RAP%)	Period-to-period variation in fundamental frequency averaged across three vocal fold vibration cycles	Jitter percentages < 0.6% are indicative of dysphonia (Boersma and Weenink, 2010).	SW: r = 0.497, p <0.001; CS: r = 0.98, p <0.001
Jitter: Period Perturbation Quotient (Jitter PPQ%)	Period-to-period variation in fundamental frequency averaged over five cycles	Jitter percentages < 0.6% are indicative of dysphonia (Boersma and Weenink, 2010).	SW: r = 0.52, p <0.001; CS: r = 0.99, p <0.001

Mean fundamental frequency of pause groups (F0 Hz Mean)	Average pitch	F0 ranges between 250-280 Hz for females aged 11-18 years and 220-250 Hz for pre-pubescent males, reducing to 140Hz after puberty (Kent, 1976);	CS: $r = 0.99$, $p < 0.001$
Standard deviation of mean fundamental frequency of pause groups (F0 Hz SD)	Pitch modulation as an indication of intonation and departure from monotone/monotonicity	25 Hz (Eguchi and Hirsh, 1969)	CS: $r = 0.92$, $p < 0.001$
Speech rate (Rate with pauses)	Calculated from the duration of the pause group (in seconds) divided by the total number of syllables in the pause group including pauses	Speech rate appears to increase throughout early childhood and stabilize at 4-4.5 syllables per second around age 11 years (Nip and Green, 2013).	CS: $r = 0.94$, $p < 0.001$.
Articulation rate (Rate without pauses)	Calculated as per speech rate but excluded pauses longer than 200 ms		CS: $r = 0.94$, $p < 0.001$
Duration (sec) with pauses	Entire duration of the pause group	Average breath group duration in spontaneous speech (including pauses) increases from 2.13 seconds in primary school age to 4.35 seconds in adults (Mitchell et al., 1996)	CS: $r = 0.999$, $p < 0.001$
Duration (sec) without pauses	Articulation time. Calculated as per duration with pauses but removing pauses longer than 200 ms.		CS: $r = 0.999$, $p < 0.001$

Table 3. Results of repeated measures multi-level analysis of variance showing main effects on individual acoustic measure of the impact of the intervention; the estimated mean change within pre and post periods; and the estimated mean difference between pairs of recordings taken at each of the four time points

Speech type	Acoustic measure	Mean (SD) acoustic measure score pre-intervention	Degrees of freedom for full repeated measures ANOVA	Post – Pre intervention			Change within pre and post periods			First vs second recording at each time point		
			df	***F***	***p***	**95%CI**	***F***	***p***	**95%CI**	***F***	***p***	**95%CI**
Single words	HNR	21.39 (6.41)	3,125	0.34	0.45	-0.54, 1.21	0.14	0.75	-0.74, 1.02	0.11	0.76	-0.74, 1.01
	RMS Pa	0.12 (0.13)	3,125	0.17	0.08	-0.02, 0.36	-0.04	0.45	0.45, 0.14	0.15	0.02	-0.08, 0.24
	Shimmer APQ %	3.50 (4.33)	3,125	-0.15	<0.001	-0.21, -0.09	-0.01	0.80	-0.07, 0.05	<0.01	0.92	-0.06, 0.06

	Jitter RAP %	0.47 (0.34)	3,125	-0.08	0.02	-0.14, -0.01	-0.01	0.86	-0.07, 0.06	-0.01	0.66	-0.08, 0.05
	Jitter PPQ %	0.51 (0.42)	3,111	-0.08	0.02	-0.15, -0.01	-0.01	0.82	-0.08, 0.06	-0.02	0.66	-0.08, 0.05
Connected speech	HNR	17.56 (5.30)	3,122	0.41	0.33	-0.41, 1.22	0.23	0.59	-0.59, 1.04	-0.07	0.87	-0.88, 0.75
	RMS Pa	0.06 (0.05)	3,122	0.03	<0.001	0.02, 0.04	<0.01	0.66	-0.01, 0.01	0.02	0.003	0.01, 0.03
	Shimmer APQ %	2.88 (1.80)	3,122	-0.18	0.16	-0.43, 0.07	-0.12	0.35	-0.37, 0.13	-0.08	0.52	-0.33, 0.17
	Jitter RAP %	0.87 (0.49)	3,122	-0.05	0.23	-0.12, 0.03	-0.04	0.33	-0.-1, 0.04	<0.01	0.49	-0.51, 0.01
	Jitter PPQ %	0.85 (0.47)	3,122	-0.05	0.24	-0.12, <0.01	-0.01	0.43	-0.11, 0.05	0.01	0.58	-0.06, 0.01
	F0 Hz Mean	252.40 (59.44)	3,122	-10.17	0.01	-18.16, -2.18	-11.85	0.004	-3.86, 0.45	0.45	0.91	-7.54, 8.44

	F0 Hz SD	64.19 (27.58)	3,122	-3.94	0.22	-10.28, 2.39	1.25	0.70	-5.09, 7.58	-1.88	0.56	-8.22, 4.46
	Speech rate	0.63 (0.22)	3,122	0.06	0.04	<0.01, 0.12	0.04	0.24	-0.03, 0.10	0.01	0.65	-0.05, 0.08
	Articulation rate	0.51 (0.22)	3,122	0.07	0.006	0.02, 0.13	0.02	0.44	-0.03, 0.07	0.01	0.62	-0.03, 0.07
	Duration (sec) with pauses	5.40 (2.94)	3,122	1.11	0.004	0.37, 1.86	<0.01	0.99	-0.76, 0.75	-0.46	0.23	-1.22, 0.29
	Duration (sec) without pauses	4.59 (2.88)	3,122	1.13	0.002	0.40, 1.85	<0.01	0.99	-0.73, 0.72	-0.37	0.32	-1.09, 0.36

Table 4. Results of regression models showing the impact of speech and language therapy on speech intelligibility controlling for acoustic measure

	Acoustic measure	**Effect of acoustic measure†**					**Effect of SLT adjusted for acoustic measure**				
		F	**df**	***p***	**95% CI**		***F***	**df**	***p***	**95% CI**	
Single words							14.95	1, 125	<0.001	11.01	18.89
	HNR	-0.16	2, 125	0.66	-0.88	0.56	15.01	2, 125	<0.001	11.06	18.95
	RMS Pa	37.46**	2, 125	0.007	10.13	64.79	11.87	2, 125	<0.001	7.46	16.28
	Shimmer APQ %	-65.60	2, 125	0.98	-257.22	125.91	14.66	2, 125	<0.001	10.67	18.79
	Jitter RAP %	-817.35	2, 125	0.07	-1692.06	57.35	13.83	2, 125	<0.001	9.77	17.88
	Jitter PPQ %	-658.95	2, 111	0.053	-1326.96	9.05	13.94	2, 111	<0.001	9.58	18.30
Connected speech							16.99	1, 127	<0.001	12.08	21.92
	HNR	-0.69	2, 127	0.18	-1.687	0.31	16.09	2, 127	<0.001	11.21	20.98
	RMS Pa	15.34	2, 127	0.70	-61.54	92.22	15.39	2, 127	<0.001	10.05	20.72
	Shimmer APQ %	58.10	2, 127	0.93	-264.20	380.50	15.93	2, 127	<0.001	11.02	20.81
	Jitter RAP %	-133.49	2, 127	0.81	-1236.03	969.05	15.75	2, 127	<0.001	10.83	20.67
	Jitter PPQ %	188.98	2, 127	0.73	-901.87	1279.83	15.90	2, 127	<0.001	10.97	20.82

	F0 Hz Mean	-0.11*	2, 127	0.02	-0.21	-0.02	14.69	2, 127	<0.001	9.78	19.59
	F0 Hz SD	-0.01	2, 127	0.90	-0.15	0.13	15.78	2, 127	<0.001	10.85	20.71
	Speech rate	-1.23	2, 127	0.87	-16.14	13.68	15.90	2, 127	<0.001	10.90	20.87
	Articulation rate	0.78	2, 127	0.40	-1.02	2.59	15.99	2, 127	<0.001	11.09	20.89
	Duration with pauses	0.73	2, 127	0.22	-0.43	1.88	14.99	2, 127	<0.001	9.94	20.05
	Duration without pauses	0.73	2, 127	0.24	-0.48	1.94	14.99	2, 127	<0.001	9.92	20.07

† The change in intelligibility corresponding to a unit increase in each acoustic measure

The first rows for single word and connected speech results show the effect of the intervention on speech intelligibility without the additional effect of acoustic measures

* p<0.05

**p<0.01