Breathing Dynamics for Non-speech and Speech Tasks Following Intensive Voice and Speech Treatment in Children with Motor Speech Disorders Secondary to Cerebral Palsy

by

Brianna J Mager

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Department of Communication Sciences and Disorders

University of Alberta

Abstract

Introduction. Cerebral palsy (CP) is the most frequently occurring cause of movement disorders in children. It has a prevalence of 2-2.5 cases per 1000 live births and is a chronic condition, requiring lifelong rehabilitation. The ability to communicate is a primary factor in positive functional outcomes for individuals with neuromuscular disorders such as CP. As many as 80% of these individuals have motor speech disorders due to neuromuscular control of the speech mechanism. Lee Silverman Voice Treatment (LSVT®LOUD) is an intervention program that uses established activity-dependent neuroplasticity principles to promote short- and long-term changes in voice and speech through an intensive voice intervention with a single target of increasing vocal loudness. Initial studies have shown that LSVT LOUD improves aspects of oral communication in children with dysarthria secondary to CP. Other features of motor speech function also have been shown to respond positively to LSVT LOUD, such as articulatory precision, pitch variability, and breath control. The present study examined the effects of LSVT LOUD on respiratory control for non-speech and speech tasks in children with motor speech disorders secondary to CP. **Method**. Nine children with CP and motor speech disorders received LSVT LOUD delivered by a registered speech-language pathologist, followed by a twelve-week maintenance period during which the children used a computer program or paper and pencil to guide and document their practice. Typically developing aged-matched children served as a control group. Assessments were conducted at three time points: pre-treatment, post-LSVT LOUD, and post-maintenance period. Both non-speech and speech tasks were designed to elicit a range of lung volumes and tracheal pressures, which were evaluated via chest wall kinematics, chest wall surface electromyography, and vocal loudness (sound pressure level). Tasks included vital capacity, maximum duration phonation, maximum duration phonation at

twice-conversational loudness, diadochokinetic sequential motion rates, and phrase repetition at conversational and twice-conversational loudness. Thirteen dependent variables were tested: lung volume events (initiations, terminations, excursions, percent rib cage contribution to lung volume excursion), intercostal muscle activation onsets/offsets, oblique onsets/offsets, intercostal relative activation, oblique relative activation, and intercostal-oblique intermuscular coherence. **Results.** The main findings of this study were that sixteen sessions of LSVT LOUD and a 12-week maintenance program, *i*) increased stability of speech breathing patterns including lung volume initiations, terminations, excursions and percent rib cage contributions to lung volume excursions; *ii*) increased intercostal muscular effort for the trained maximum duration phonation task; *iii*) decreased the intercostal muscular effort required to repeat untrained phrases using both conversational and perceived twice-conversational loudness; *iv*) increased intercostal-oblique intermuscular coherence in an untrained sequential motion task (/*pataka*/) requiring articulatory speed and precision. **Conclusion.** This preliminary investigation into the use of an intensive vocal loudness treatment (LSVT LOUD) for children with CP and a range of type and severity of motor speech disorders shows a promising short- and long-term treatment response. These results add to the voice and speech treatment literature and will help guide future stages of treatment efficacy research in paediatric motor speech disorders.

Preface

This thesis is an original work by Brianna Mager. The research project, of which this thesis is a part, received research ethics approval from the University of Alberta Research Ethics Board, Project Name: *Neural correlates of intensive voice treatment effects on children with cerebral palsy*; Pro00038076; 20 March 2013.

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Table of Contents

List of Tables

List of Figures

Introduction

Cerebral Palsy and Speech Disorders

Cerebral Palsy (CP) is the most frequently occurring cause of movement disorders in children. Over the past 40 years the prevalence of CP has risen to more than 2.5 cases per 1000 live births (Mutch, Alberman, Hagberg, Kodama, & Perat, 1992) and constitute the largest diagnostic group who are treated in pediatric rehabilitation (Odding, Roebroeck, & Stam, 2005). CP is characterized by a group of movement and postural disorders as a result of an *in utero* stroke, other brain infarct, or birth asphyxia. These disorders are permanent, stable, and often lead to challenges in educational and social participation (Donkervoort, Roebroeck, Weigerink, & Van der Heijden-Maessen, 2007). General characteristics of CP include hyperactive reflexes, muscle weakness, and reduced endurance. Characteristics might also include disturbances of sensation, perception, cognition, communication, and behaviour (Rosenbaum, Paneth, Leviton, Goldstein, & Bax, 2006).

Individuals with CP have a high incidence of respiratory dysfunction (Strauss, Shavelle, Rosenbloom, Brooks, 2008). Children with CP breathe in an uncoordinated fashion, relying on the abdominal muscles instead of the more biomechanically efficient rib cage muscles for expiration (Redstone, 2004). Breathing in this manner over a long period of time restricts chest wall movement and weakens the chest wall muscles. As a result, the ability to maximally inspire is impaired (Blumberg, 1955). Kinematic analysis of breathing revealed a chronic failure to expand the upper chest wall and lungs in these children. Wang, Chen, and Hsiao (2012) showed that, compared to children with typical development, children with CP have significantly lower respiratory muscle strength, as represented by Maximum Expiratory Pressure (MEP) and Maximum Inspiratory Pressure (MIP) values.

As many as 80% of individuals with CP have some form of speech disorder (Odding et al., 2005). Motor speech disorders in this population can involve both apraxia of speech and dysarthria. The most common type of speech disorder is dysarthria, which is a speech motor control deficit due to spasticity or muscle weakness. Dysarthrias characterized in this population can include spastic-type, flaccid-type, ataxic-type, or a combination of these (mixed-dysarthria) (Duffy, 2005). Dysarthria secondary to CP can involve all aspects of the speech mechanism (i.e., respiratory, laryngeal, velopharyngeal-nasal, and pharyngeal-oral).

The function of the respiratory subsystem is of particular interest in the present study. Seminal work by Wolfe (1950) showed that speech breathing problems were common in individuals with CP. In another piece of formative research, Blumberg (1955) studied general breathing and speech problems in 27 children with CP. He concluded that there was a direct relationship between the regularity of breathing rate, rhythm, and depth of respiration (inspiratory and expiratory volumes) and the quality of speech. Difficulty with speech breathing control unfavourably affects vocal loudness, utterance length, inspiratory timing, and voice quality.

Speech Breathing

Breathing for speech, or *speech breathing* refers to *the respiratory mechanics used to inhale before speaking and to generate and maintain subglottal air pressure during speech production* (R. Netsell, personal communication, as cited in Solomon & Charron, 1998, p. 1). Speech is generated with laryngeal, velopharyngeal, and pharyngeal-oral valving of the air stream. Both active muscle forces of the rib cage and abdomen as well as passive (recoil) forces of the chest wall and lung are used to generate air pressures needed to drive phonation (Hixon, Mead, & Goldman, 1976). The relative contributions of active and passive forces are lung

volume dependent. Aerodynamic measurements of tracheal pressures indicate that phonation requires pressures of approximately 5 to 8 cmH2O in typical adult speakers (Smitheran & Hixon, 1981). However, children often phonate using higher tracheal pressures of approximately 7 to 10 cmH2O (Stathopoulos & Sapienza, 1997). In adolescents and adults, speech is produced in the mid-range of the vital capacity (VC), using excursions of about 20% VC (Hixon, Goldman, & Mead, 1973; Hoit, Hixon, Watson, & Morgan, 1990). The rib cage contributes approximately 80% to lung volume excursions associated with speech. Healthy adults are able to phonate throughout most of their vital capacity by coordinating passive recoil forces with active force of the rib cage and abdominal muscles (Hixon et al., 1976). Children also typically begin speech in the mid-range of their VC, but often end their utterance at much lower lung volumes and use inconsistent patterns of rib cage and abdominal contributions to lung volume excursion (Boliek, Hixon, Watson, & Morgan, 1996; 1997; Hoit et al., 1990; Stathopoulos & Sapienza, 1993). Speaking at lower lung volumes (e.g., below end expiratory level) is less biomechanically efficient than ending breath groups at or above end expiratory level (Boliek et al., 1997; 2009). Generally, speech breathing is becoming adult-like between the ages of 10 to 12 years (Hoit et al., 1990; Stathopoulos & Sapienza, 1997).

Macefield and Gandevia (1991) found the presence of clear premotor cerebral neural firing when participants performed self-paced inspiratory or expiratory manoeuvres. Breathing manoeuvres like those used for speech require cortical drive to meet variable demands for generating appropriate airway pressures and lung volumes. Cortical control begins in the primary motor cortex and travels via the cortico-spinal tract to the peripheral nervous system. This entire pathway may be referred to as the *cortico-spinal-respiratory pathway.* Increasing the neural firing from the motor cortex, via the corticospinal tract, can modulate coordination of

speech breathing. Although Macefield and Gandevia (1991) found evidence for the conscious control of respiratory muscles from (in part) the primary motor cortex, cortical control during non-speech and speech tasks that require varying tracheal pressures and lung volumes is not well understood.

Breathing for Voluntary Respiratory Events

Four parameters are manipulated during breathing for speech: lung volume, tracheal pressure, expiratory airflow, and chest wall shape (Hixon, Weismer, & Hoit, 2008). Perceptual characteristics of lung volume, tracheal pressure, flow and shape are breath group length, vocal loudness, voice quality, and inspiratory duration, respectively. Patterns of recruitment of rib cage and abdominal muscles for voluntary breathing are dependent on the task requirements. Speech breathing requires slight increases in expiratory muscle recruitment compared to that needed during passive tidal (rest) breathing as shown in electromyographic (EMG) studies (Hoit, Plassman, Lansing, & Hixon, 1988). Compared to tidal breathing, speech and sustained phonation are characterized by shorter inspirations, longer expirations, larger lung volume excursions, and higher sustained expiratory pressures (Hixon et al., 1973, Hixon et al., 1976). During maximum duration phonations, individuals begin phonating at high lung volumes (top of VC) and employ inspiratory braking using the external intercostal muscles as a way to slow down the rate of airflow from the lungs while maintaining constant tracheal pressure. As lung volume decreases, the internal intercostal muscles activate to compress the lungs for continued expiration and maintenance of tracheal pressure. The abdominal muscles also begin to contract at lower lung volumes to maintain constant pressure (Hixon et al., 1976). Compared to phonation at conversational loudness, phonation produced at louder levels requires higher

tracheal pressure. Thus, earlier (higher percent VC) and greater chest wall expiratory muscle recruitment is needed (Hixon et al., 1976; Hixon & Weismer, 1995).

Analyzing speech breathing. Chest wall surface kinematic measures can be used to examine breathing events associated with non-speech and speech tasks. Measures of lung volume initiations (LVI), terminations (LVT), and excursions (LVE), along with percent rib cage contribution to lung volume excursions (PCTRC) can be assessed using variable inductance plethysmography (Respitrace, Ambulatory Monitoring Company, NY) (Hixon et al., 2008; use in children see: Boliek, Hixon, Watson, & Jones, 2009). This method involves placing wired elastic bands around the rib cage and abdomen. These are connected to a plethysmograph that records the average cross-sectional areas of the rib cage wall and abdominal wall during inspiratory and expiratory breathing events. In the present study, inductance plethysmography was selected to measure volume changes of the rib cage and abdomen because of its sensitivity to individual movements of both the rib cage wall and the abdominal wall. These measurements permit the calculation of overall lung volume change at the body's surface by summing the two cross-sectional areas (Hixon et al., 2008), and also leave the participant unencumbered.

Muscle Activity for Voluntary Respiratory Events

Chest wall muscle activation associated with various voluntary breathing tasks has been studied in healthy adult and child populations (Bailey & Hoit, 2002; Clair-Auger, Gan, Norton, & Boliek, under review; DeTroyer, Kirkwood, & Wilson, 2005; Hixon et al., 1976; Hoit et al., 1988; Tomczak, Greidanus, & Boliek, 2013) but rarely in a disordered population. Voluntary control of respiration for speech requires complex neural coordination of the respiratory system, which can be quantified by assessing *intermuscular coherence*. Intermuscular coherence is a measure that can be used to infer cortical control over peripheral muscles (Halliday et al., 2003;

Norton & Gorassini, 2006). Coherence values are assigned by determining the degree to which two signals in the same frequency are correlated with each other. Coherence values are quantitative descriptors reflecting the strength (relative correlation) of the oscillatory coupling between the two signals (Grosse, Cassidy, & Brown, 2002). The frequency at which coherence occurs can be used to infer the origins of the signals in the brain. In the present study, coherence was evaluated in three frequency bandwidths: low (2-12 Hz), medium (12-30 Hz), and high (30-60Hz). Signals in the low frequency bandwidth are thought to originate in sub-cortical areas of the brain, though recent studies have found evidence of sensorimotor involvement (Marsden et al., 2000). Signals occurring in the medium frequency bandwidth, or β-band, and high frequency bandwidth, or γ-band, are well established as having originated from the motor cortex (Grosse et al., 2002). However, research has indicated that the amount of coherence observed is influenced by more factors than originally assumed. Muscles in the chest wall for example, have been shown to have lower coherence during tasks that require greater lung volume excursion (Tomczak et al., 2013). Differences in coherence have even been shown in the same task between people with skilled (i.e., pianists) vs. strong (i.e., weight lifters) motor abilities (Semmler, Sale, Meyer, & Nordstrom, 2004). Smith and Denny (1990) found detectable levels of masseter muscle coherence during speech, and even higher values associated with a non-speech chewing task. Alternatively, research on neck intermuscular coherence has found no significant differences between conversational speech, reading, and a non-speech tongue retraction task (Stepp, Hillman, & Heaton, 2011).

Intermuscular coherence in various frequency bandwidths has been used to detect neural changes in cortical drive to muscles after a given treatment. For example, rehabilitation studies have shown that chronic interventions aimed at improving limb function following long-term

disuse are associated with neural adaptation as measured by an increase in intermuscular coherence (Norton & Gorassini, 2006). Other studies have also shown intermuscular coherence to be sensitive to training effects (Semmler & Nordstrom, 1998; Semmler et al, 2004; Ushiyama, Takahashi, & Ushiba, 2010). Based on previous research, intermuscular coherence of chest wall muscles associated with breathing during non-speech and speech tasks will be employed in the present study as an indication of neural adaptation following intensive voice treatment in children with CP and dysarthria.

Lee Silverman Voice Treatment (LSVT) and Neuroplasticity

Neuro-rehabilitation studies have recorded key elements of motor learning and principles of activity-dependent neuroplasticity that may be important considerations of successful treatment paradigms for children with neurological disease or impairments (Garvey, Giannetti, Alter, & Lum, 2007; Kleim & Jones, 2008; Kleim, Jones, & Schallert, 2003; Maas et al., 2008). In this context, activity-dependent neuroplasticity refers to changes in the central nervous system in response to physical activity. The key elements modulating neuroplasticity are: intensive treatment, repetitive practice, and sensory feedback associated with movement. Treatment research that translates principles of both motor learning and activity-dependent neuroplasticity into protocols for treatment of motor speech disorders in children and adults has become a growing movement (Boliek & Fox, 2014; Fox & Boliek, 2012; Levy, Ramig, & Camarata, 2012; Ludlow et al., 2008; Maas et al., 2008).

Previous studies in speech and physical therapy have documented that children with CP are capable of intensive treatment programs, with initial fatigue often decreasing with time (Bower, McLellan, Arney, & Campbell, 1996; Schindl, Forstner, Kern, & Hesse, 2000). The use of active, task-specific, repetitive practice, along with increased numbers of practice trials results

in marked improvements in gait (Damiano, Kelly, & Vaughan, 1995; Schindl et al., 2000), grip force production (Valvano & Newell, 1998), and anticipatory grip force precision (Gordon & Duff, 1999) in children with CP. Sensory input and active attention to sensory feedback during practice also may facilitate neuroplastic changes of cortical sensorimotor maps thought to foster internal representations for movement (Hadders-Algra, 2000; Hadders-Algra, van der Fits, Stremmelaar, & Touwen, 1999). Internal representations are thought to be important for maintenance of the new target behavior and carryover of skills from trained to untrained tasks.

Whereas current speech treatments for dysarthria secondary to CP are varied, most employ a systems approach to address respiration, phonation, articulation, and resonance (Pennington, Miller, Robson, & Steen, 2010). Recently reviewed observational studies of speech interventions for children with dysarthria indicated that teaching slow, loud speech may be associated with improvements in speech intelligibility, voice quality, and clarity (Levy et al., 2012; Pennington et al., 2010). LSVT LOUD reflects these ideas, although the program was developed for individuals with Parkinson's disease (PD) and has documented efficacy for that population (Ramig et al., 2001). The extent to which the effects of LSVT LOUD are specific to dysarthria associated with CP is not clear. However, positive outcomes in acoustic measures (e.g., vocal sound pressure level [SPL]) and listener perceptions (e.g., articulatory precision) have been reported post–LSVT LOUD from single-subject and case studies of adults with ataxic type dysarthria (Sapir et al., 2003), and in adults with flaccid type dysarthria and apraxia of speech secondary to stroke (Mahler & Ramig, 2012). Wenke, Theodoros, and Cornwell (2008) found positive changes in several acoustic and perceptual parameters following LSVT in ten patients with dysarthia post-stroke or TBI, six of whom displayed spastic-type characteristics. A study on an individual with Parkinson disease and compensatory supraglottic hyperfunction

found that LSVT increased vocal loudness, decreased supraglottic hyperadduction, and improved intonation and overall voice quality when compared with pre-treatment observations (Countryman, Hicks, Ramig, & Smith, 1997). A larger study on laryngostroboscopic variables in patients with Parkinson disease undergoing LSVT found less glottal incompetence without an increase in supraglottal hyperfunction after therapy, correlated with an increase in vocal intensity (Smith, Ramig, Dromey, Perez, & Samandari, 1995).

The training regimen of LSVT LOUD is consistent with the principles driving activity-dependent neuroplasticity and motor learning (Fox et al., 2006). Focusing on vocal loudness as the sole target for the treatment of dysarthria has been described as creating a single motor organizing theme. Increasing effort (as loudness) impacts other speech production systems (Duffy, 2005). The singular training target of healthy vocal loudness may be desirable for children with CP, who have disordered voice characteristics related to muscle weakness or incoordination such as found in spastic-type CP (Ansel & Kent, 1992; Workinger & Kent, 1991). A single focus on vocal loudness also limits the cognitive demands associated with treatment, which may be important for children with low-average to below-average cognitive functioning. Additionally, target elicitation is achieved through modeling behavior (e.g., "do what I do"), which minimizes explicit verbal instructions and may promote self-organization of the child's system in order to achieve the goal (Schmidt & Fitzpatrick, 1996). Further, LSVT LOUD delivers treatment in a standardized manner through a protocol of specific daily exercises for a finite period of treatment.

The fundamental purpose of this *Phase I* treatment study is to examine any therapeutic effects of LSVT LOUD on the respiratory behaviour of children with CP with varied types and severity of motor speech disorders, and to understand the underlying mechanism of change

associated with treatment (Robey & Schultz, 1998; Robey, 2004). In *Phase I*, researchers explore and specify the therapeutic effect as a dependent variable and the treatment protocol as an independent variable. The goals of a *Phase I* study are to detect a therapeutic effect if present, approximate population definitions, approximate treatment protocol, specify appropriate dose, and refine hypotheses. Previous single-subject multiple baseline research by Fox & Boliek in 2012 found a therapeutic effect in positive acoustic and perceptual changes in a relatively homogenous population of children with spastic CP and moderate dysarthria. A second study with a larger group of children having spastic CP and moderate to severe dysarthria and a matched control group (Boliek et al., in preparation) validated the results of the first study. The present study aims to examine the effects of LSVT LOUD on a larger group of children with various types and severity of CP and motor speech disorders. A limitation of this study is the potential to mask a treatment effect with a small sample size and numerous sources of variance. As is typical for a *Phase I* study, *Type I* error tolerance was liberal in order to identify the presence of a therapeutic effect if it exists.

Purpose

Specific Aim

The specific aim of the present study was to investigate the acute and long-term effects of an intensive voice treatment (LSVT LOUD) on features of speech breathing as measured by chest wall kinematics and intermuscular coherence in children with varying types and severity of dysarthria secondary to CP.

Hypothesis

The overarching research hypothesis was that sixteen sessions of LSVT LOUD followed by a 12 week structured maintenance program would improve performance on non-speech and

speech tasks in the following ways: i) a shift in respiratory kinematics to represent more typical and stable breathing patterns, ii) a shift in muscle activation onsets and relative amplitudes to represent more typical and stable muscular effort, and iii) an increase in chest wall inter-muscular coherence.

Methods

Participants

Nine participants with CP (3 females) and eight age- and sex-matched typical children participated in the study. All children were between seven and sixteen years of age. Experimental participants had been previously diagnosed with cerebral palsy, with severities ranging from II to V (mild-moderate to severe) on the Gross Motor Function Classification System (GMFCS) expanded and revised scale (Palisano, Rosenbaum, Bartlett, & Livingston, 2008). Children with CP also had been previously diagnosed by a speech-language pathologist (SLP) with spastic (n=1), spastic-flaccid (n=7), or spastic-ataxic (n=1) dysarthria, ranging from mild to severe. Four of the nine CP participants were diagnosed with apraxia of speech (AOS) in addition to dysarthria. All participants were medically stable and seizure-free. All participants had basic oral communication skills and did not use alternative or augmentative communication devices. Control participants all had average or above average cognitive level as reported by their parent or guardian. Table 1 shows the complete description of each participant in the treatment group.

Participant	Sex	Age	Speech Diagnosis	GMFCS	Cognitive Level
LSVTF1	F	13	Spastic-Flaccid Dysarthria, Moderate	III	Above Average
LSVTM2	M	11	Spastic-Flaccid Dysarthria, Mild	II	Average
LSVTF3	F	12	Spastic-Flaccid Dysarthria, Mild; AOS, Mild-Moderate	II	Average
LSVTM4	M	12	Spastic-Flaccid Dysarthria, Mild	V	Below Average
LSVTM5	M	8	Spastic Dysarthria, Mild-Moderate	V	Average
LSVTM6	M	13	Spastic-Flaccid Dysarthria, Moderate-Severe; AOS, Moderate-Severe	IV	Below Average
LSVTF7	F	16	Spastic-Flaccid Dysarthria, Severe ; AOS, Severe	III	Average
LSVTM8	M	8	Spastic-Flaccid Dysarthria, Moderate-severe; Dysfluency, Mild	II	Below Average
LSVTM9	M	13	Spastic-Ataxic Dysarthria, Moderate-Severe; AOS, Mild	IV	Average

Table 1. Participant description including sex, age, speech diagnosis, rating on the Gross Motor Function Classification System (GMFCS), and cognitive level.

Experimental overview

All participants came to the testing laboratory on three separate occasions. The first (Time-1) and second (Time-2) visits occurred 4 weeks apart (immediately pre- and immediately post-LSVT LOUD), followed by the final session (Time-3) 12 weeks later (immediately following completion of the maintenance program). Only the CP group participated in LSVT LOUD and maintenance treatment. Figure 1 (below) outlines the testing and treatment procedure, including the tasks measured at each testing point and the dependent variables for

each task, described in detail below.

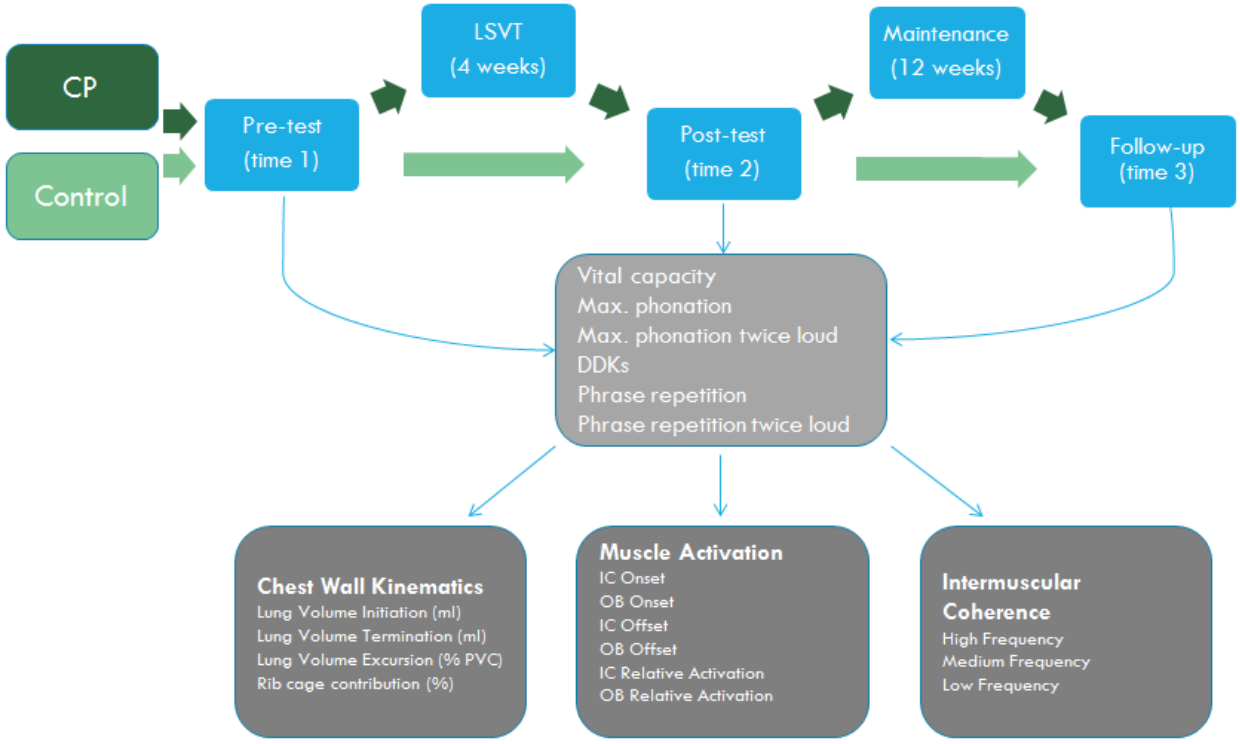


Figure 1. Overview of the experimental procedure, including tasks and dependant variables collected at each testing point.

Treatment consisted of 16 one-hour sessions with an LSVT LOUD trained SLP, 4 days a week for 4 consecutive weeks. This is the standard dosage for LSVT LOUD, described in more detail in Table 2. Treatment targeted vocal loudness through structured sessions focusing on high-intensity and repetition. After the four-week period, participants engaged in a 12-week maintenance program using either a computer program or paper and pencil to guide and monitor practice sessions. Table 2 provides a complete description of the therapy targets, dosage, and maintenance schedule.

Target	Treatment Sessions	Homework on treatment days (4 days/week)	Treatment on non-treated days (3 days/week)	Total Minimum Repetitions in one month	12 Week Maintenance Schedule
Long Ah	15 reps/day 16 days = 240	6 reps/day 16 days = 96	12 reps/day 14 days = 168	504	6 reps/day 84 days = 504
High Ah	15 reps/day 16 days = 240	6 reps/day 16 days = 96	12 reps/day 14 days = 168	504	6 reps/day 84 days = 504
Low Ah	15 reps/day 16 days = 240	6 reps/day 16 days = 96	12 reps/day 14 days = 168	504	6 reps/day 84 days = 504
Functional Phrases	10 phrases 5x/day 16 days = 800	10 phrases 2x/day 16 days = 320	10 phrases 4x/day 14 days = 560	1680	10 phrases 2x/day 84 days = 1680
Structured Reading	Week 1: 20 min 4 days = 80 min Week 2: 20 min 4 days = 80 min Week 3: 15 min 4 days = 40 min Week 4: 5 min 4 days = 20 min Total= 220 min	5 min/day 16 days = 80 min	10 min/day 14 days = 140 min	440 minutes structured reading/verbal practice with target voice	5 min/day 84 days = 420 min
Conversational Speech	Week 1: 5 min 4 days = 20 min Week 2: 5 min 4 days = 20 min Week 3: 10 min 4 days = 60 min Week 4: 20 min 4 days = 80 min Total= 180 min	5 min/day 16 days = 80 min	10 min/day 14 days = 140 min	440 minutes structured conversation with focus on target voice	N/A

Table 2. LSVT LOUD treatment schedule and repetition dose.

Following written and informed consent, the participants were oriented to the testing procedures and familiarized with the equipment. As previously described (Tomczak et al.,

2013), surface EMG recordings were made from the right side of the body over the 6th intercostal space and abdominal oblique regions (see Figure 2, below) using a standardized electrode surface placement protocol. Specifically, intercostal electrodes were placed ventrally at the parasagittal point that approximates the mid clavicle. Oblique muscle electrodes were placed midway between the anterior superior iliac spine and caudal border of the rib cage. For each location, two electrodes (Kendal Soft-E H69P, Tyco Healthcare Group, Mansfield, MA) were positioned 2 cm apart (center-to-center). It is possible that other muscles also may contribute to surface EMG signals within these parameters, such as the external intercostals and internal obliques. External intercostals are superficial to the internal intercostals, therefore EMG signals might reflect some inspiratory muscle activation (or inspiratory "braking") during portions of expiratory manoeuvers (Hixon et al., 1976). The electrode placement protocol optimized ventral-dorsal EMG location to avoid the inter-cartilaginous region. We confirmed the appropriate EMG positioning for the IC and OB muscles by observing EMG activity during inspiratory and expiratory manoeuvers and trunk rotation tasks at the beginning of the session (Tomczak et al., 2013). EMG signals were sampled at 10 kHz, high pass filtered at 100 Hz, and rectified.

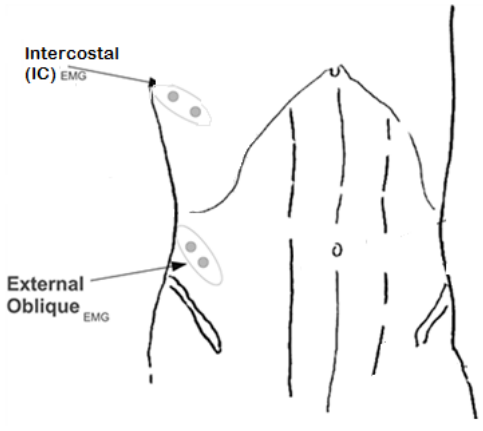


Figure 2. Placement of EMG surface electrodes for the intercostal (IC) and oblique (OB) muscles.

Respiratory data were collected via chest wall kinematics derived from inductance plethysmography (Respitrace, Ambulatory Monitoring Company, NY) using rib cage and abdominal cloth belts placed according to the standardized protocol reported in Boliek et al. (2009). Acoustic measures of speech were acquired using a small microphone (SHURE MX-185) placed on the mid-forehead, 10 cm above the mouth. Audio signals were amplified (M-Audiobuddy Pre-Amplifier) and digitally recorded at a sampling rate of 44.1 kHz on a laptop computer using TF32 Software (Milenkovic, 2001). Calibration of the audio signals involved the presentation of a 440 Hz tone presented at the mouth (KORG Orchestral Tuner, model OT-12) and a sound level meter (ExTech Sound level meter 407764) in line with the forehead-mounted microphone. Sound level in dB SPL was recorded for a 10 second sound sample and used in the calculation of vocal loudness in SPL for acoustic analyses. Digital video recordings were made during the testing to assist with eliminating data associated with extraneous movement and/or postural shifts. All kinematic, EMG, audio and video signals were simultaneously recorded using a digital recorder (A R Vetter Co, Rebersburg, PA) as well as Power Lab (ADInstruments, Colorado Springs, CO).

Participants were asked to maintain an upright posture while seated as much as possible, and supported to maintain this position as needed. Baseline data collection included 1) calibration of the inductance plethysmograph, 2) three trials of a vital capacity maneuver in accordance with the American Thoracic Society, 3) six trials of a maximum duration phonation task (three at conversational loudness and three at twice-conversational loudness), 4) six trials of repeated short phrases (three at conversational loudness and three at twice-conversational

loudness), and 5) three trials of a sequential motion task (pa-ta-ka). All children with CP were instructed by trained research assistants not associated with the treatment aspects of the study. Blinding was not possible due to the obvious communication and physical disorders associated with CP. Research assistants were trained to give consistent instructions with encouragement (i.e., "good job" or "nice work") but without using treatment cues in any form (e.g., "use your loud voice" or "go louder") with the exception of the cued tasks which were to phonate or speak at perceived twice-conversational loudness.

Calibration procedures for respiratory kinematics involved performing an isovolume maneuver (pulling the stomach in and letting it relax) while breath-holding. Participants were also asked to breathe (approximately 10 breaths) through a fitted mouth-piece (using nose clips) attached to a pneumotachometer and differential pressure transducer, amplified and integrated for volume. These two calibration tasks served to convert kinematic signals to volume signals. Maximum voluntary contraction tasks were performed to collect greatest muscle activity. A vital capacity task was used to elicit maximum IC activity, and trunk rotation against force (provided by the research assistant) was used to collect maximum OB activity.

Sample Size

Based on data from previous treatment studies with similar pediatric populations a sample size calculation for repeated measures with $\alpha = 0.05$ and $\beta = 0.50$ (*Phase I* standards) was used. This calculation produced a range from 4 to 8 participants needed to reach significant power. The present study used convenience sampling to achieve a sample size of 9 children with CP and a group of matched controls.

Data Analysis

A within-subjects design was used to detect differences pre-, post, and at 12 weeks follow up in the children with CP and from Time 1, 2 (4-weeks following Time 1), and 3 (12 weeks following Time 2) in the control group. Between-subjects factors were not analysed statistically, as significant differences between groups were expected. Comparisons between the CP group and the control group were visually inspected and descriptive in nature, used in the context of the treatment group values moving towards or away from control values. Data were normally distributed based on the Shapiro-Wilk test, therefore within–subjects repeated measures ANOVAs were used to test differences separately for each dependent variable. As is typical for *Phase I* treatment studies, differences were considered significant when $p < 0.05$, uncorrected. Trends were reported when $p < 0.09$, uncorrected. Follow-up paired *t*-tests were reported when univariate ANOVAs were significant. There was a significant degree of variability in the CP group, creating a risk of masking a therapeutic effect. Therefore, data were also examined individually in an effort to identify visual patterns of change (see supplemental materials).

Chest Wall Kinematic Analysis

Kinematic measures included lung volume initiation and termination in ml (LVI, LVT) relative to end expiratory level (EEL). Lung volume excursions were calculated as a percent of vital capacity (PVCLVE). The relative contribution of the rib cage to lung volume excursion was calculated as percent rib cage contribution to lung volume excursion (PCTRC).

Chest Wall EMG Analysis

The chest wall EMG protocol followed the standards outlined for children in Clair-Auger et al. (under review). Using a running average filter with a window size of 100 ms, onset of an EMG burst was detected when the smoothed signal amplitude rose above 2 SDs of the baseline

EMG (derived from the tidal breathing task) for a minimum duration of 500 ms. Termination of the burst was detected when the smoothed signal amplitude became less than 1 SD of the baseline EMG. The amount of muscular effort used was determined as a percentage of maximum muscular contraction (%MVC). IC and OB onsets, offsets, and muscular effort were calculated for each trial of each task and averaged across participants within each group.

Intermuscular Coherence Analysis

Intermuscular coherence between EMG intercostal – EMG oblique (IC-OB) coherence was calculated for each task at each time point. EMG recordings from the intercostal and oblique muscles that did not occur during expiration were removed from the data set, so as to analyze only EMG recordings occurring exclusively as a result of expiration during VC, maximum duration phonation, and speech productions. Data within a given task were concatenated for each participant prior to coherence calculations. Data were analyzed for coherence within three frequency bandwidths: low (2-15 Hz), medium (15-30 Hz), and high (30-60 Hz). Coherence was measured as the peak correlation above the 95% confidence interval. Coherence was deemed detected if a minimum of 3 sequential data points within a band (+/- 5 Hz) were above the 95% confidence limit. Amplitude of the signal indicating the strength of coherence was recorded and used for comparison. Intermuscular coherence was calculated as:

$$MSC\ |Cxy(w)|^2 = \frac{\left|\overline{Gxy(w)}\right|^2}{\overline{Gxx(w)} \cdot \overline{Gyy(w)}}$$

where MSC is magnitude squared coherence, Gxx(w) and Gyy(w) are the averaged power spectra of the muscles of interest (x and y) for a given frequency (w), and Gxy(w) is the averaged cross-power spectrum of signals x and y at frequency w (from Halliday, et al., 1995). Figure 3 shows exemplar EMG IC-OB intermuscular coherence from a participant with CP and his matched control across the three time points for the sequential motion (/*pataka*/) task.

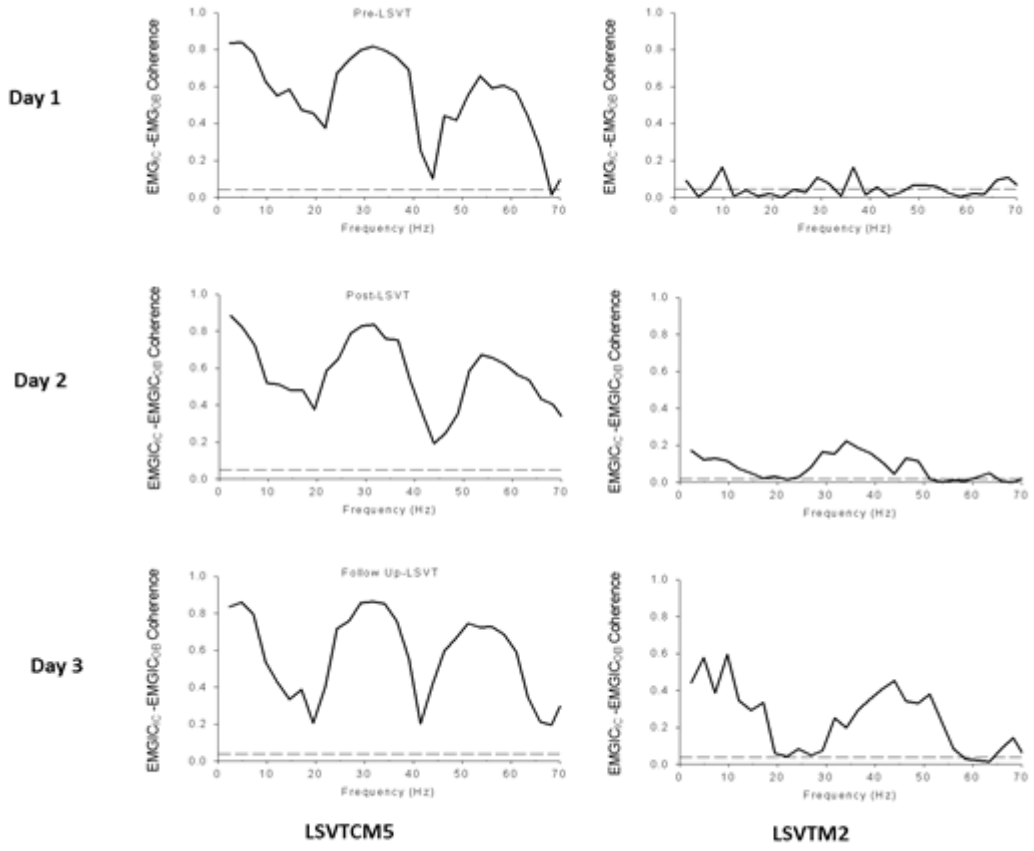


Figure 3. An exemplar of the Control (left) and CP (right) data for intermuscular coherence across three time points for the sequential motion task. Coherence is represented on the vertical axis, frequency on the horizontal axis, and the 95% confidence interval is represented by the horizontal dashed line.

Results

Loudness

The goal of the "twice-conversational loudness" tasks was to increase the demands on tracheal pressure to above 8 cmH_20. The perceptual correlate for pressure is loudness; therefore the participants were encouraged to "go twice as LOUD as you typically talk, but do not shout" and were coached if shouting or unhealthy vocal qualities were observed. Table 3 and 4 show that all participants did increase loudness (dBSPL) when cued for maximum duration phonations

and sentence repetitions. For sentence repetitions, dBSPL was derived for each vowel and averaged for each phrase. Paired *t*-tests confirmed that conversational (Normal) and perceived twice-conversational loudness (Twice Loud) were significantly different for both groups and both tasks.

		Control	**CP**
Day 1	Normal	80.40	84.65
	Twice Loud	91.30	90.20
Day 2	Normal	81.11	87.49
	Twice Loud	89.51	95.35
Day 3	Normal	84.69	90.27
	Twice Loud	94.13	93.20
Paired *t*-test Normal vs. Loud		t=-13.2 (2) p<.01	t= -3.82(2) p<.06

Table 3. Average loudness (dBSPL) for the maximum duration phonation task at conversational loudness, microphone to mouth distance was 10 cm.

			Control	**CP**
Day 1	Normal	**Bobby**	75.17	79.75
		Blue	75.86	79.89
		Stew	76.66	79.63
	Twice Loud	**Bobby**	83.93	84.11
		Blue	83.40	83.62
		Stew	83.54	84.28
Day 2	Normal	**Bobby**	74.78	84.40
		Blue	75.71	84.45
		Stew	76.03	85.82
	Twice Loud	**Bobby**	82.67	89.31
		Blue	81.55	88.31
		Stew	83.16	88.65
Day 3	Normal	**Bobby**	79.04	82.58
		Blue	79.34	82.78
		Stew	79.59	82.49
	Twice Loud	**Bobby**	87.83	86.44
		Blue	87.41	85.89
		Stew	86.87	85.55
Paired *t*-test Normal vs. Loud			t= -24.3(8) p<.01	t= -15.7(8) p<.01

Table 4. Average loudness (dBSPL) for the maximum duration phonation task at twice-conversational loudness, microphone to mouth distance was 10 cm. Bobby = *Buy Bobby a Puppy*, Blue = *The blue spot is on the key*, Stew = *The potato stew is in the pot*.

Vital Capacity

Kinematics. Figure 4 shows group performance on the VC task, across the three time points, on measures of Lung Volume Initiation (LVI), Lung Volume Termination (LVT), Lung Volume Excursion in Percent of Vital Capacity (PVCLVE), and Percent Rib Cage Contribution (PCTRC) to lung volume excursion. Within subject repeated-measures ANOVAs (Table 5) were not significant in either the control or the children with CP ($p > 0.05$). Control children initiated VC manoeuvres between 1588 and 1822 ml above end expiratory level (EEL) and terminated these manoeuvres between 1110 and 1249 ml below EEL. In contrast, children with CP initiated their VC manoeuvres between 788 and 854 ml above EEL and terminated between 290 and 580 ml below EEL. Control children used 100 percent of their predicted VC when producing the VC manoeuvres (note: some children performed beyond their predicted VC values) whereas children with CP produced VC manoeuvres using excursions ranging from 44 to 57 %VC on average. Control children used between 86 to 88 PCTRC, whereas children with CP used between 66 to 73 PCTRC on average. A general decrease in variability towards stability was noted in the children with CP, immediately following treatment and at 12-weeks follow up when the range of individual performance (i.e., individual data points and box plots) was examined (see supplemental materials).

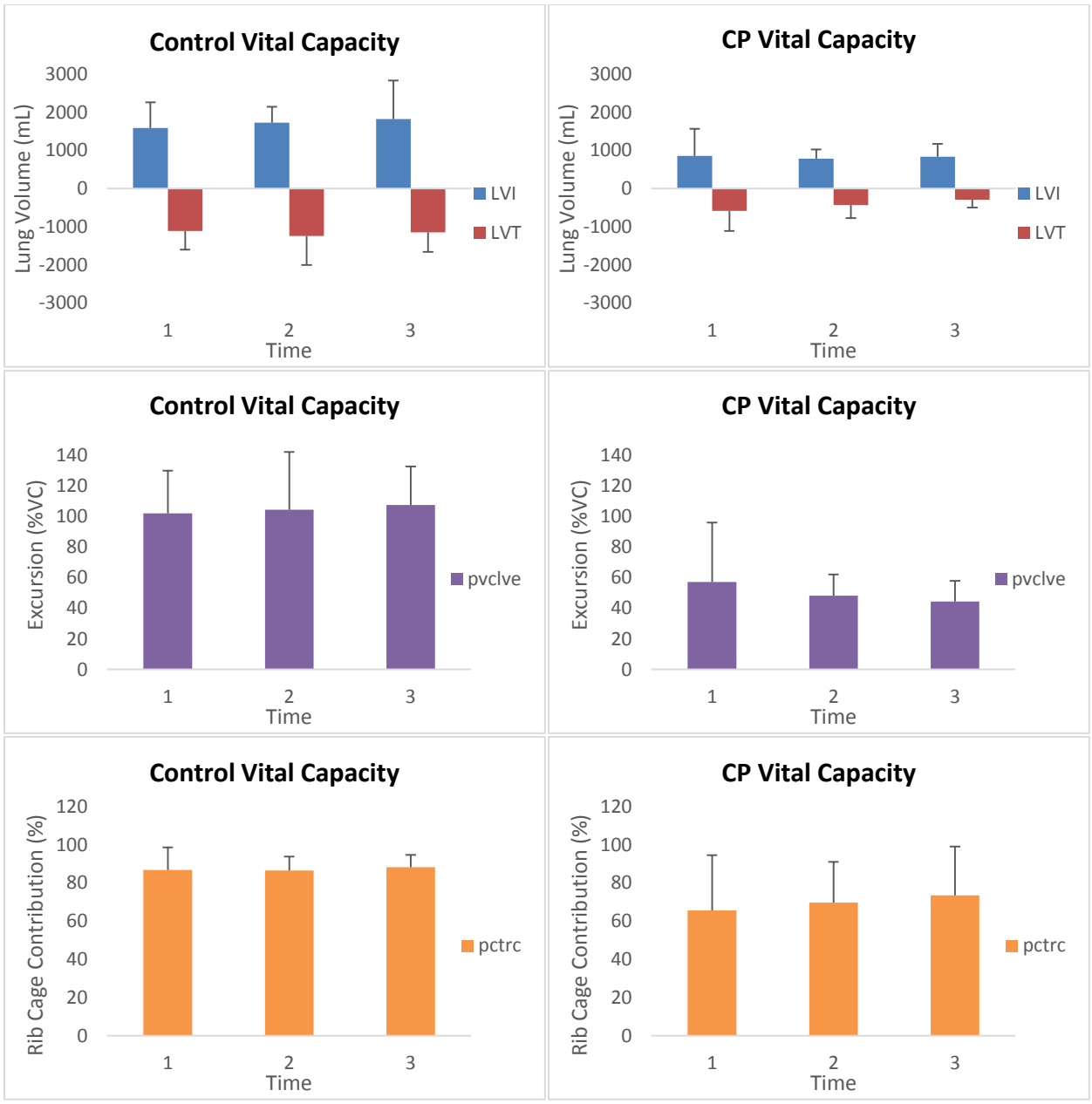


Figure 4. Average change in Lung Volume Initiation (LVI), Lung Volume Termination (LVT), Lung Volume Excursion in Percent of Vital Capacity (PVCLVE), and Rib Cage Contribution in Percent (PCTRC) over three time points during a vital capacity task.

Task	Control		CP	
	F	**Sig.**	**F**	**Sig.**
LVI	$1.09_{(2, 14)}$	.34[a]	$.06_{(2, 16)}$	.95
LVT	$1.08_{(2, 14)}$	.36	$2.68_{(2, 16)}$	.10
PCTRC	$.11_{(2, 14)}$	.90	$1.24_{(2, 16)}$	.30[a]
PVCLVE	$.98_{(2, 14)}$	.36	$.65_{(2, 16)}$	.54

[a]Reported Greenhouse-Geisser value

Table 5. Repeated-Measures ANOVA output for kinematics during a vital capacity task.

Muscle activation patterns. Figure 5 shows the activity of the IC and OB muscles during the VC task. The top two panels indicate at what percentage of predicted VC, muscle activity was detected (onset) and became undetectable (offset). In the control group, both IC and OB muscles were activated near the top of predicted VC (between 70 and 83 %VC) and terminated at or near the bottom of predicted VC (0 to 24 %VC). Children with CP exhibited onset of IC and OB muscular activation between 47 and 71% of predicted VC and offsets near the end of VC (0 to 21 %VC). Within subject repeated-measures ANOVA (Table 6) found no significant differences across time for either group. The bottom two panels in Figure 5 show the amount of muscular activation in percentage of maximum voluntary contraction (MVC) for each group across time points. Control children activated IC muscles from between 57to 59 %MVC and OB muscles from 32 to 53 %MVC. On average, children with CP activated IC muscles from between 42 to 46 % MVC and OB muscles from 27 to 42 %MVC.

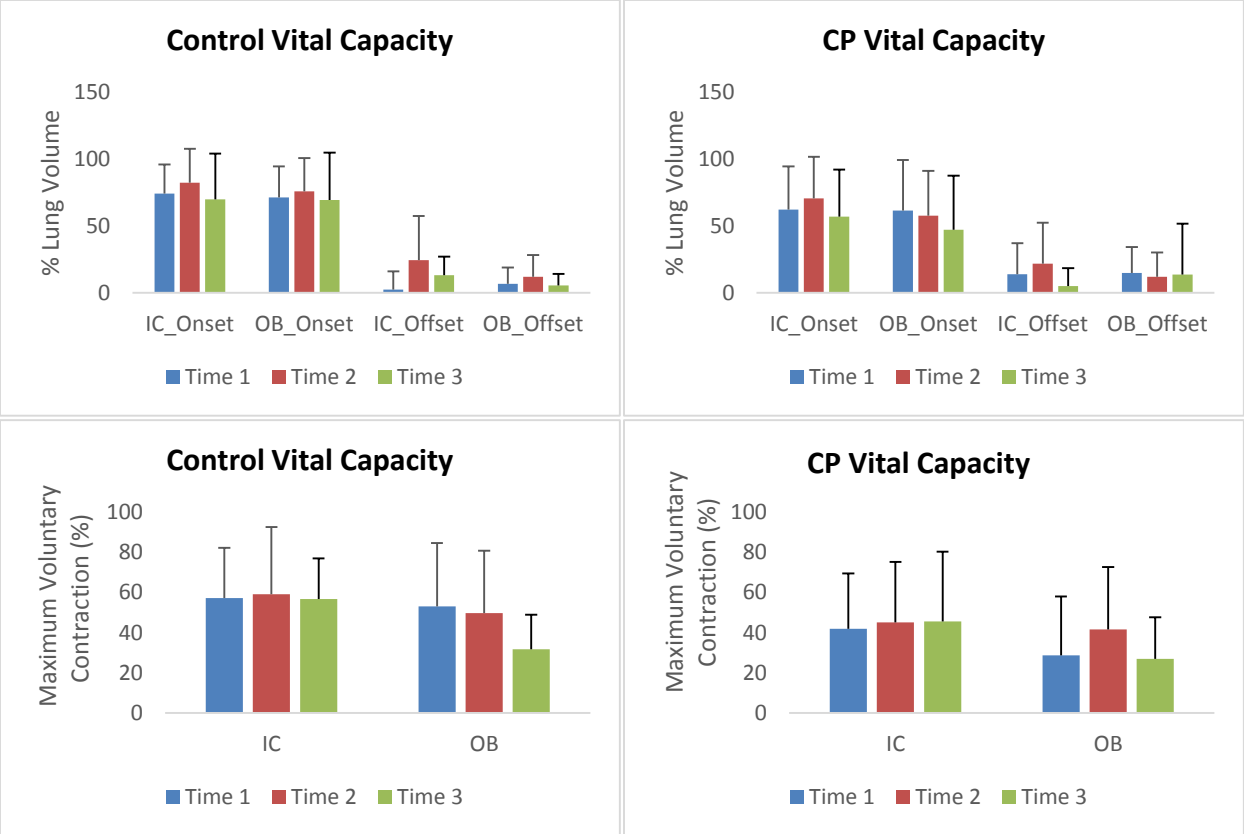


Figure 5. Effects of LSVT on intercostal (IC) and oblique (OB) onset, offset, and activation during a vital capacity task over three time points. The top two panels show the activation (onset) and deactivation (offset) of the muscles in percent predicted VC. The bottom two panels indicate activation of the muscle, as a percentage of maximum contraction.

Task	**Percent Lung Volume**				**Percent Maximum Voluntary Contraction**			
	Control		CP		Control		CP	
	F	**Sig.**	**F**	**Sig.**	**F**	**Sig.**	**F**	**Sig.**
IC_onset	$1.05_{(2, 14)}$	.38	$.73_{2, 16)}$	.50	$.03_{(2, 14)}$	.97	$.07_{(2, 16)}$	.93
OB_onset	$.15_{(2, 14)}$	.86	$.37_{(2, 16)}$	.70	$2.73^{(2, 14)}$	.1	$.81_{(2, 16)}$	.46
IC_offset	$1.98_{(2, 14)}$	.20[a]	$1.50_{(2, 16)}$	.25				
OC_offset	$.57_{(2, 14)}$	.58	$.03_{(2, 16)}$	.89[a]				

[a]Reported Greenhouse-Geisser value

Table 6. Repeated-measures ANOVA results for EMG onset, offset, and activation of the IC and OB muscles during a vital capacity task.

Intermuscular coherence. Figure 6 shows the inter-muscular coherence between IC and OB muscles. On average, peak coherence values for control children ranged from $r = 0.10$ to $r = 0.40$ across the three frequency bandwidth ranges. Similarly, children with CP exhibited

coherence ranges from $r = 0.16$ to $r = 0.39$ on average across the three frequency bandwidth ranges. Within subject repeated-measures ANOVAs (Table 7) found no significant change in coherence for the VC task, in any of the three frequency bandwidths across time points.

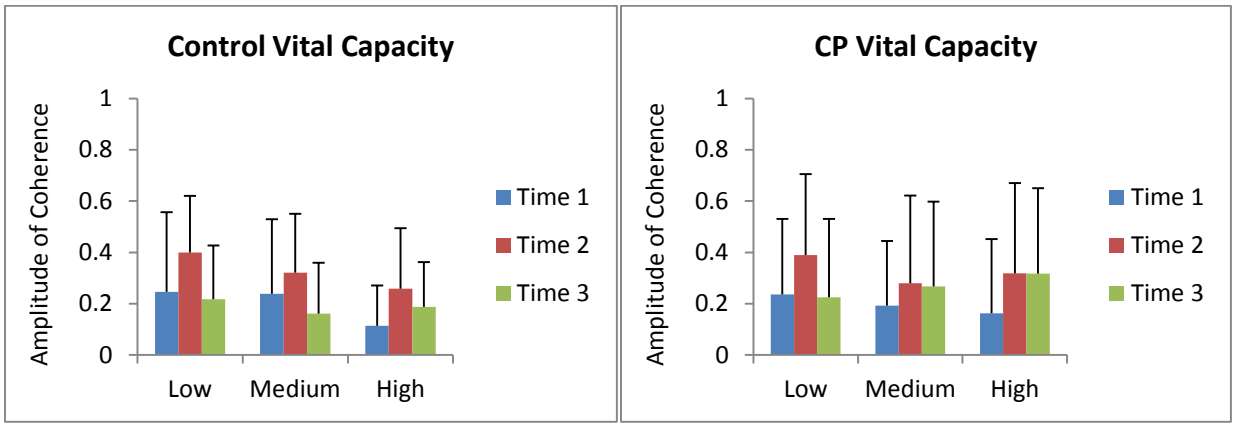


Figure 6. Effects of LSVT on chest wall peak amplitude of coherence in low, medium, and high frequencies during a vital capacity task over time.

Frequency	Control		CP	
	F	**Sig.**	**F**	**Sig.**
Low	$1.07_{(2, 14)}$	.37	$.79_{(2, 16)}$	.47
Medium	$1.31_{(2, 14)}$	.30	$.20_{(2, 16)}$	.82
High	$1.34_{(2, 14)}$	.29	$.75_{(2, 16)}$	.49

[a]**Reported Greenhouse-Geisser value**

Table 7. Repeated-measures ANOVA output for inter-muscular coherence of IC and OB muscles during a vital capacity task.

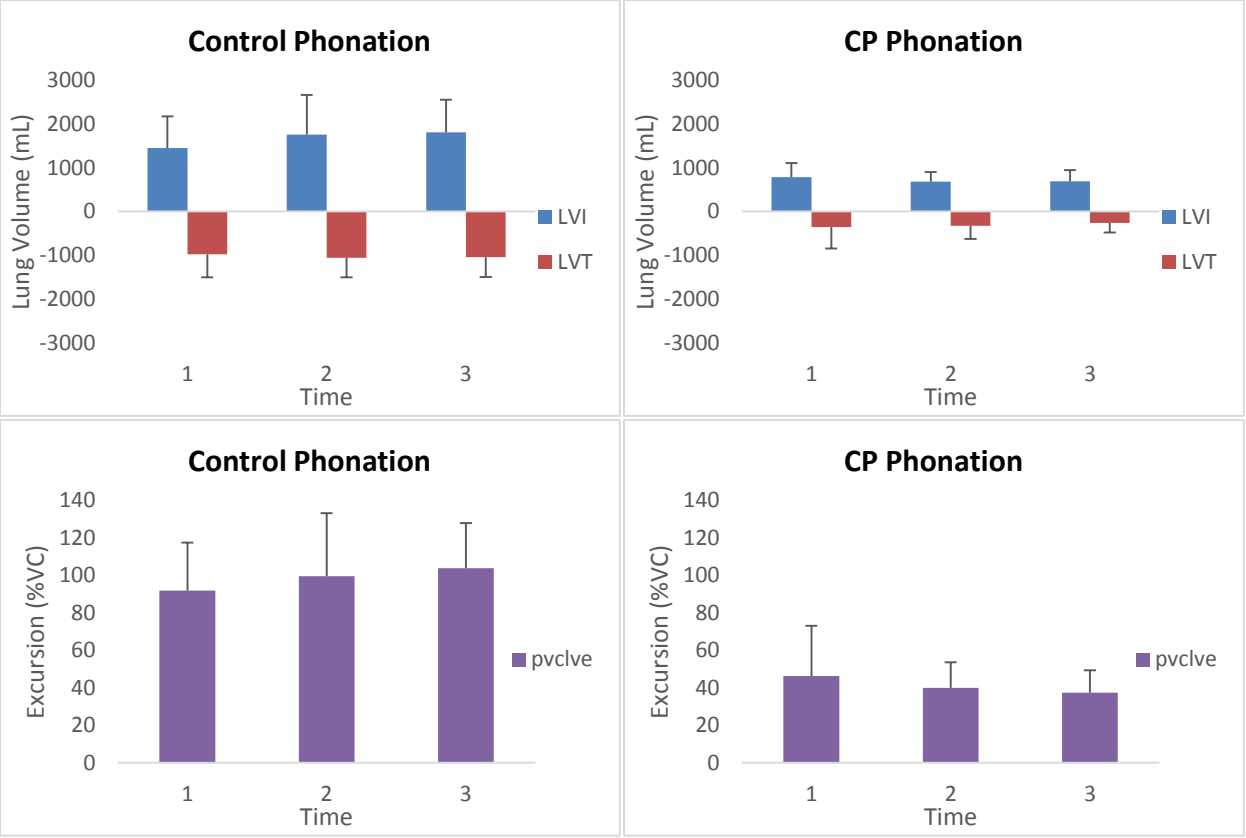
Summary of results for vital capacity. Statistically significant change was not present in the vital capacity task. A pattern of LVT occurring closer to EEL and increased PCTRC was noted across all three time points for the CP group, consistent with other maximum performance tasks as described below.

Maximum Duration Phonation Produced at Conversational Loudness

Kinematics. Within subjects repeated measures ANOVAs (Table 8) were not significant in either the control or the experimental group ($p > 0.05$). A general decrease in variability towards stability was noted in the children with CP immediately following treatment and at 12-

weeks follow up when the range of individual performance (i.e., individual data points and box plots) was examined (see supplemental materials).

Figure 7 shows averages of the participants' performances on the maximum duration phonation task, across the three time points, on measures of LVI, LVT, PVCLVE, and PCTRC. Control children initiated the maximum duration phonations between 1450 and 1801 ml above EEL and terminated these manoeuvres between 981 and 1056 ml below EEL. In contrast, children with CP initiated their phonations between 781 to 785 ml above EEL and terminated between 264 and 359 ml below EEL. Control children used between 91 and 100 percent of their predicted VC when producing maximum duration phonations (note: some children performed beyond the predicted VC values) whereas children with CP produced phonations at excursions ranging from 37 to 46 % VC on average. Control children used between 81 to 86 PCTRC, whereas children with CP used between 66 and 72 PCTRC on average.



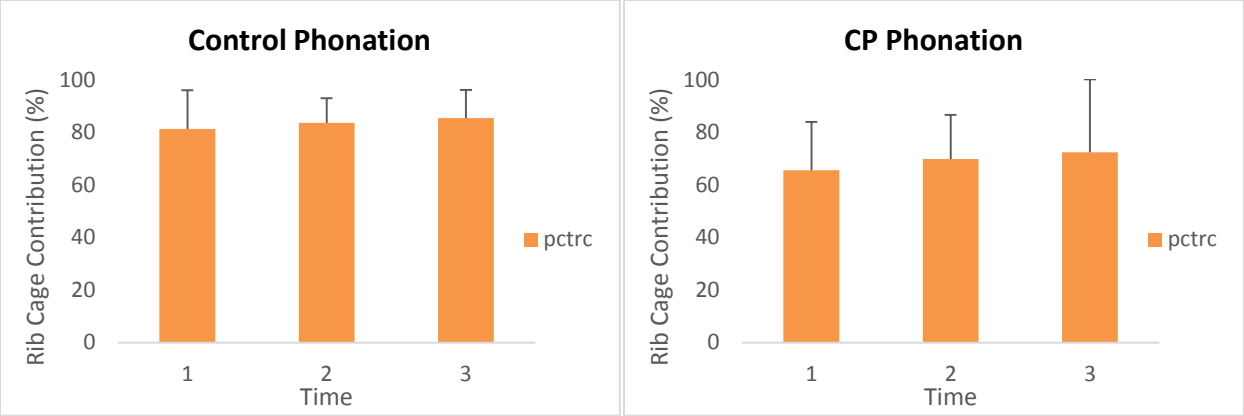


Figure 7. Group change in Lung Volume Initiation (LVI), Lung Volume Termination (LVT), Lung Volume Excursion in Percent of Vital Capacity (PCCLVE), and Rib Cage Contribution in Percent (PCTRC) over three time points, for a maximum duration phonation task at conversational loudness.

Task	Control		CP	
	F	**Sig.**	**F**	**Sig.**
LVI	2.17 (2, 14)	.15	.51 (2, 16)	.61
LVT	.25 (2, 14)	.78	.32 (2, 16)	.73
PCTRC	.30 (2, 14)	.75	2.08 (2, 16)	.18[a]
PVCLVE	1.47 (2, 14)	.26	.81 (2, 16)	.46

[a]**Reported Greenhouse-Geisser value**

Table 8. Repeated-Measures ANOVA output for kinematics during a maximum duration phonation task at conversational loudness.

Muscle activation patterns. Figure 8 shows the activity of the IC and OB muscles during the Maximum Duration Phonation task. The top two panels indicate at what percentage of predicted VC, muscle activity was detected (onset) and became undetectable (offset). In the control group, both IC and OB muscles were activated between 52 and 65 %VC. Intercostal activation terminated between 26 to 33 %VC whereas OB activity terminated near the bottom of predicted VC. Children with CP exhibited onset of IC between 40 and 61 %VC and OB muscular activation between 38 and 51 %VC. Both IC and OB offsets occurred near the end of

predicted VC. The bottom two panels in Figure 8 show the amount of muscular activation in %MVC for each group across time points. Control children activated IC and OB muscles between 21 and 39 %MVC. On average, children with CP activated IC muscles between 24 and 39 % MVC and OB muscles from 14 to 27 %MVC. A within subjects repeated measures ANOVA (Table 9) found no significant differences for IC or OB onsets or offsets over time for either group. A trend of $p = .09$ was noted for IC activation in %MVC in children with CP, as indicated at the bottom right of Figure 6. Paired *t*-tests found a significant difference between time 1 and time 2 ($t = -2.393$, df = 8, $p < .05$, two-tailed), indicating greater IC activation in %MVC at time 2.

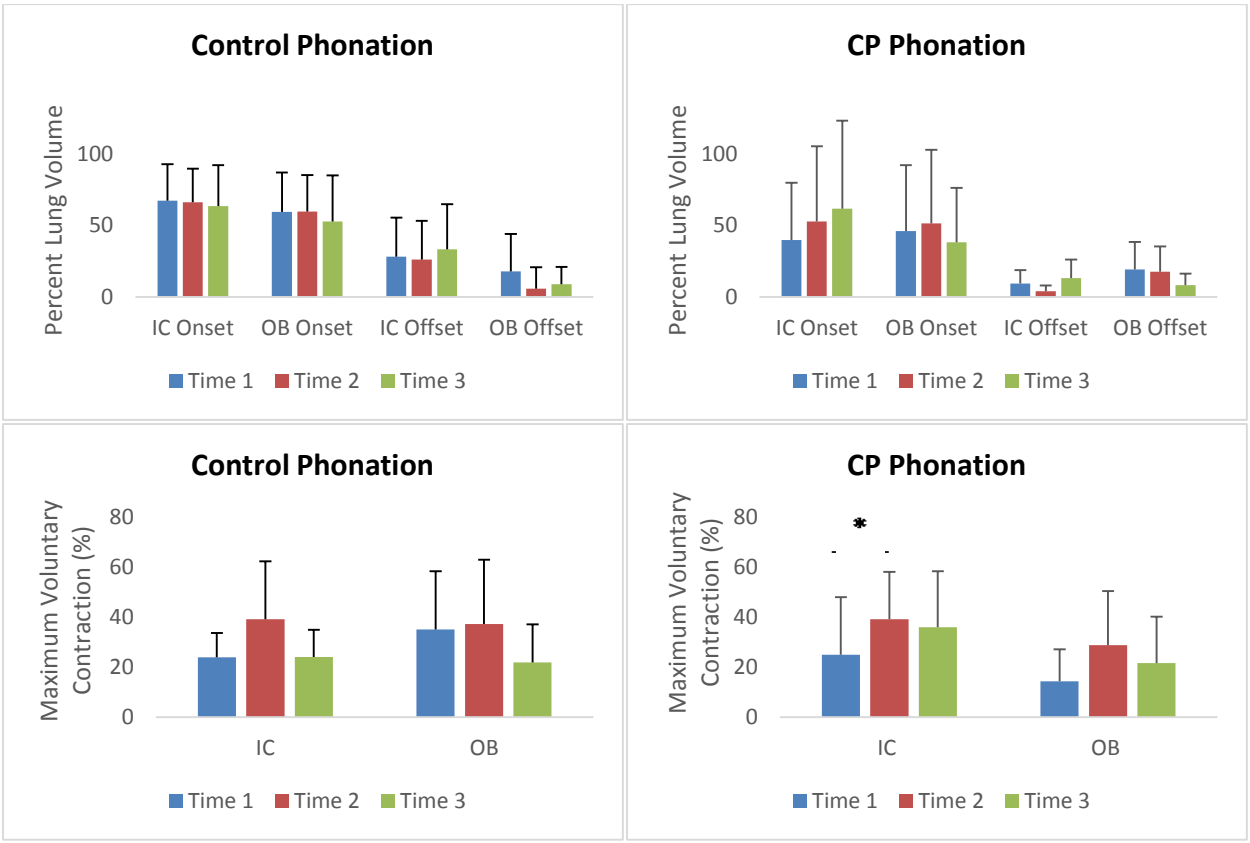


Figure 8. Effects of LSVT on intercostal (IC) and oblique (OB) onset, offset, and activation during a maximum duration phonation task at conversational loudness, over three time points. The top two panels show the activation (onset) and deactivation (offset) of the muscles in percent predicted VC. The bottom two panels indicate activation of the muscle, as a percentage of maximum contraction.

	Percent Lung Volume				Percent Maximum Voluntary Contraction			
	Control		CP		Control		CP	
	F	**Sig.**	**F**	**Sig.**	**F**	**Sig.**	**F**	**Sig.**
IC_onset	$.06_{(2, 14)}$	.86[a]	$.62_{(2, 16)}$	.55	$2.12_{(2, 14)}$	.16	$2.83_{(2, 16)}$	.09
OB_onset	$.14_{(2, 14)}$	.87	$.23_{(2, 16)}$	.79	$2.13_{(2, 14)}$	.16	$1.70_{(2, 16)}$	.22
IC_offset	$.29_{(2, 14)}$	.76	$.21_{(2, 16)}$	.81				
OB_offset	$.78_{(2, 14)}$	.48	$.15_{(2, 16)}$	.73[a]				

[a]Reported Greenhouse-Geisser value

Table 9. Repeated-measures ANOVA results for EMG onset, offset, and activation of the IC and OB muscles during a maximum duration phonation task at conversational loudness.

Intermuscular coherence. Figure 9 shows intermuscular coherence between the IC and OB muscles. On average, peak coherence values for control children ranged from $r = 0.50$ to $r = 0.63$ across the three frequency bandwidths. Similarly, children with CP exhibited ranges from $r = 0.34$ to $r = 0.56$ on average across the three frequency bandwidths. A within subjects repeated-measures ANOVA found no significant change in coherence in any of the three frequency bandwidths, as reported in Table 10.

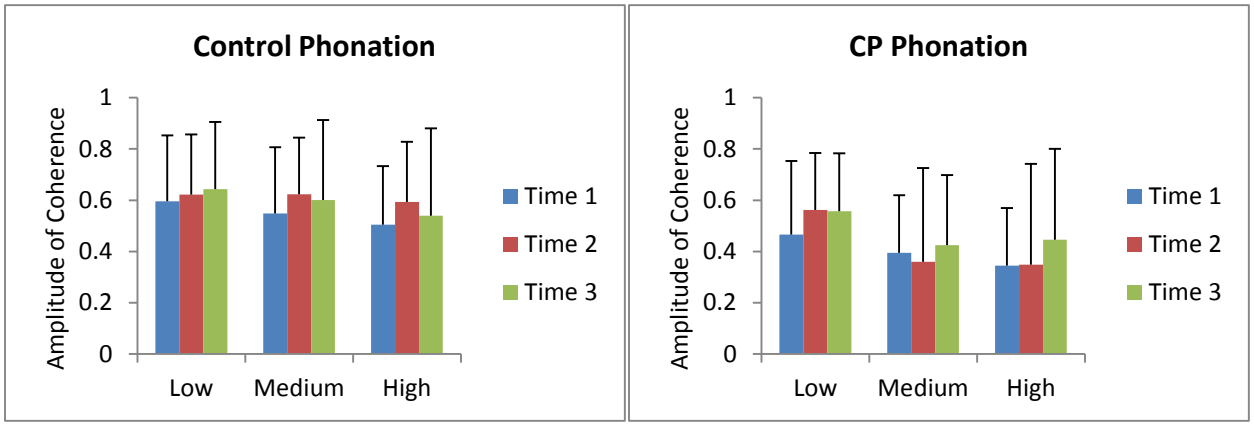


Figure 9. Effects of LSVT on chest wall peak amplitude of coherence in low, medium, and high frequencies during a maximum duration phonation task at conversational loudness.

Frequency	Control		CP	
	F	**Sig.**	**F**	**Sig.**

Low	1.21 (2, 14)	.33	.84 (2, 16)	.45
Medium	.48 (2, 14)	.63	1.50 (2, 16)	.25
High	.12 (2, 14)	.89	.52 (2, 16)	.61[a]

[a]Reported Greenhouse-Geisser value

Table 10. Repeated-measures ANOVA output for inter-muscular coherence of IC and OB muscles during a maximum duration phonation task at conversational loudness.

Summary of results for maximum duration phonation. A statistically significant change was present for an increase in IC relative activation immediately following LSVT LOUD for the trained maximum duration phonation task. A visual pattern was noted for LVT occurring near EEL. A visual trend of rising PCTRC was also noted across all three time points for the CP group, consistent with the vital capacity results described above.

Maximum Duration Phonation Produced at Twice-Conversational Loudness

Kinematics. Figure 10 shows group average performance on the maximum duration phonation task at twice-conversational loudness across the three time points, on measures of LVI, LVT, PVCLVE, and PCTRC. Control children initiated phonations between 1341 and 1926 ml above EEL and terminated these phonations between 886 and 1060 ml below EEL. In contrast, children with CP initiated their phonations between 633 and 892 ml above EEL and terminated between 265 and 340 ml below EEL. Control children used between 84 and 100 percent of their predicted VC when producing phonations (note: some children performed beyond the predicted VC values), whereas children with CP produced phonations at excursions ranging from 38 to 46% on average. Control children used between 79 and 87 PCTRC whereas children with CP used between 66 and 70 PCTRC on average.

Within subjects repeated-measures ANOVAs (Table 11) were not significant for the CP group ($p < .05$). The control group showed a significant change ($p < .05$) for LVI. Follow up *t*-tests found a difference between time 1 and 3 ($t = -2.40$, df = 7, $p < .05$, two-tailed). A general decrease in variability towards stability was noted for PCTRC in the children with CP

immediately following treatment and at 12-weeks follow up when the range of individual performance (i.e., individual data points and box plots) was examined (see supplemental materials).

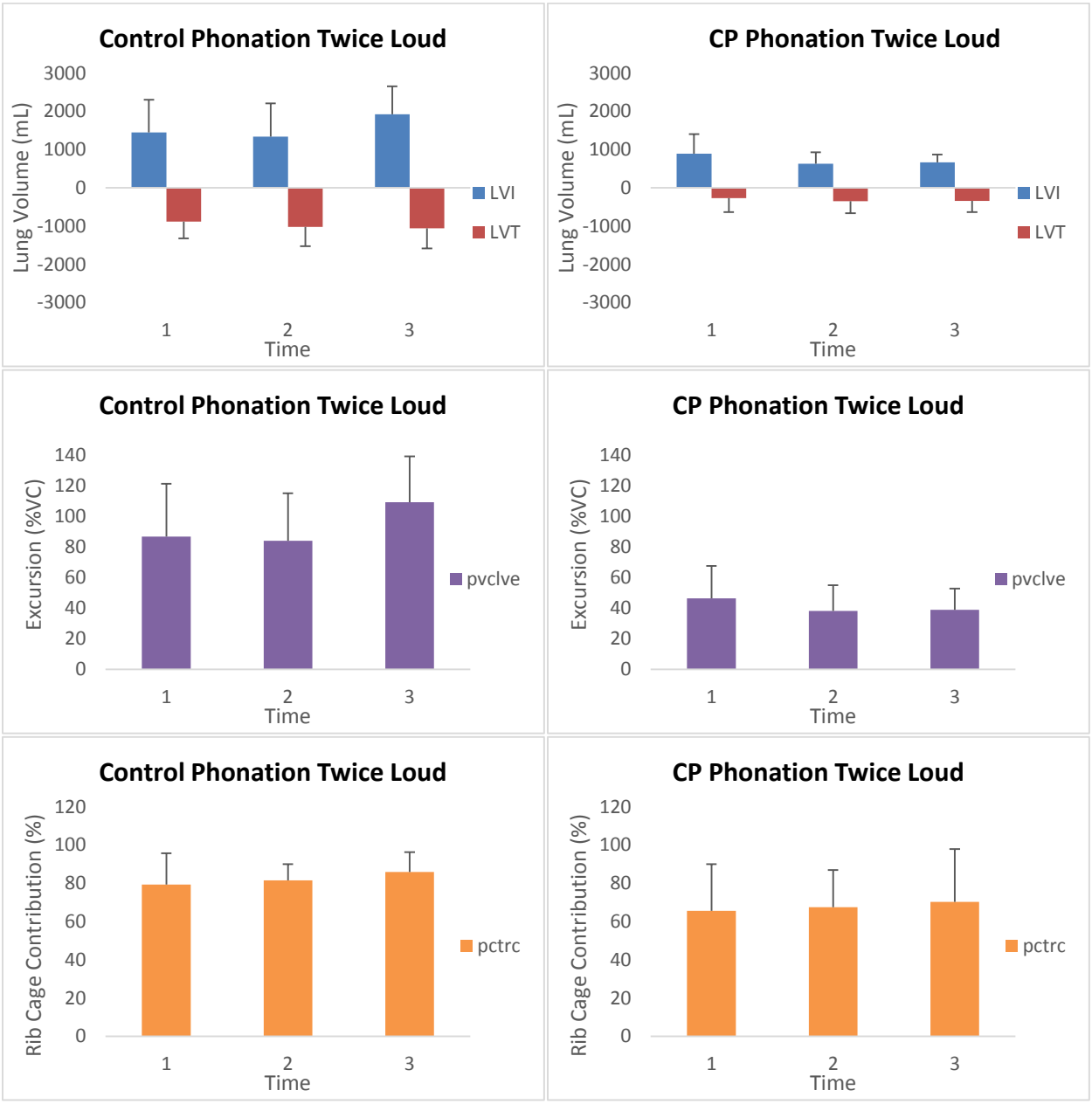


Figure 10. Group change in Lung Volume Initiation (LVI), Lung Volume Termination (LVT), Lung Volume Excursion in Percent of Vital Capacity (PVCLVE), and Rib Cage Contribution in Percent (PCTRC) over three time points, for a maximum duration phonation task at twice-conversational loudness.

Task	Control		CP	
	F	**Sig.**	**F**	**Sig.**
LVI	4.02 $_{(2, 14)}$	*.04	1.74$_{(2, 16)}$	.21
LVT	.46 $_{(2, 14)}$	.64	.32$_{(2, 16)}$	.73
PCTRC	.74 $_{(2, 14)}$	.50	.28$_{(2, 16)}$	.63
PVCLVE	2.30 $_{(2, 14)}$	.137	1.68$_{(2, 16)}$	.22

[a] Reported Greenhouse-Geisser value
***significance**

Table 11. Repeated-Measures ANOVA output for kinematics during a maximum duration phonation task at twice-conversational loudness.

Muscle activation patterns. Figure 11 shows the activity of the IC and OB muscles for maximum duration phonations produced at twice-conversational loudness. The top two panels indicate at what percentage of predicted VC muscle activity was detected (onset) and became undetectable (offset). In the control group, both IC and OB muscles were activated between 57 and 68 %VC. Intercostal activation terminated between 25 to 37 %VC whereas OB activity terminated near the bottom of predicted VC. Children with CP exhibited onset of IC between 48 and 54 %VC and OB muscular activation between 38 and 49 %VC. Both IC and OB offsets occurred near the bottom of predicted VC. The bottom two panels in Figure 9 show the amount of muscular activation in %MVC for each group across time points. Control children activated IC and OB muscles from between 20 and 38 %MVC. On average, children with CP activated IC muscles from between 34 to 37 % MVC and OB muscles from 22 to 31 %MVC. Within subjects repeated-measures ANOVA (Table 12) found no significant differences over time for either group for the maximum duration phonation task at twice-conversational loudness.

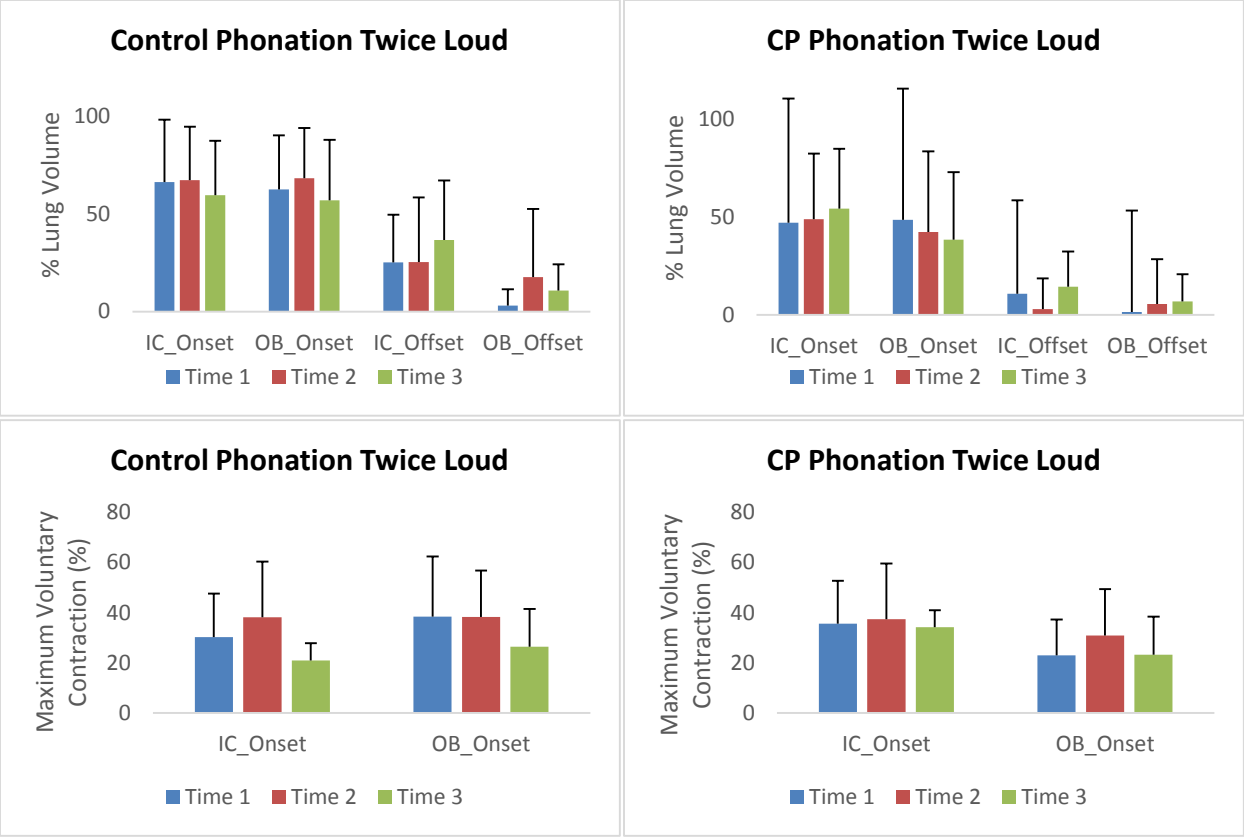


Figure 11. Effects of LSVT on intercostal (IC) and oblique (OB) onset, offset, and activation during a maximum duration phonation task at twice-conversational loudness over three time points. The top two panels show the activation (onset) and deactivation (offset) of the muscles in percent predicted VC. The bottom two panels indicate activation of the muscle, as a percentage of maximum contraction.

Task	**Percent Lung Volume**				**Percent Maximum Voluntary Contraction**			
	Control		CP		Control		CP	
	F	**Sig.**	**F**	**Sig.**	**F**	**Sig.**	**F**	**Sig.**
IC_onset	$.26_{(2, 14)}$	.77	$.07_{(2, 16)}$	.93	$3.02_{(2, 14)}$	.08	$.10_{(2, 16)}$	.91
OB_onset	$.33_{(2, 14)}$	.73	$.09_{(2, 16)}$	.92	$1.46_{(2, 14)}$	.27	$.36_{(2, 16)}$	.71
IC_offset	$.70_{(2, 14)}$	.51	$.31_{(2, 16)}$	.64[a]				
OB_offset	$.82_{(2, 14)}$	.46	$.06_{(2, 16)}$	.94				

[a]Reported Greenhouse-Geisser value

Table 12. Repeated-measures ANOVA results for EMG onset, offset, and activation of the IC and OB muscles during a maximum duration phonation task at twice-conversational loudness.

Intermuscular coherence. Figure 12 shows intermuscular coherence between the IC and OB muscles. In general, peak coherence values for control children ranged from $r = 0.4$ to r

= 0.6 on average across the three frequency bandwidths. Children with CP exhibited ranges from $r = 0.2$ to $r = 0.59$ on average across the three frequency bandwidths. There appears to be a visual trend for an increase in peak coherence immediately following treatment and 12-weeks post treatment across all frequency bandwidths in the children with CP. However, a within subject repeated-measures ANOVA found no significant change in coherence in any of the three frequency bands, as reported in Table 13.

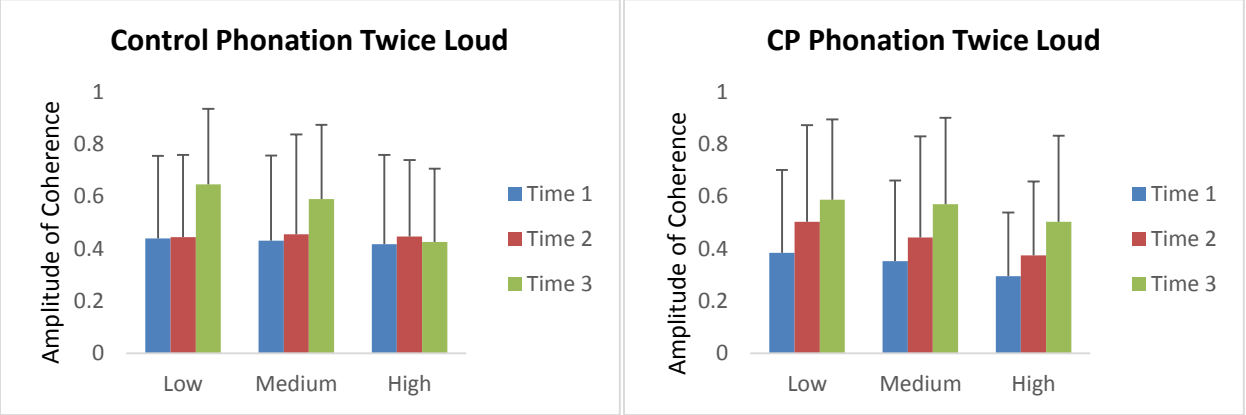


Figure 12. Effects of LSVT on chest wall peak amplitude of coherence in low, medium, and high frequencies during a maximum phonation duration task at twice-conversational loudness.

Frequency	Control		CP	
	F	**Sig.**	**F**	**Sig.**
Low	2.495 $_{(2, 14)}$	0.118	2.145$_{(2, 16)}$	0.149
Medium	1.921 $_{(2, 14)}$	0.183	1.502 $_{(2, 16)}$	0.252 [a]
High	0.108 $_{(2, 14)}$	0.899	1.748$_{(2, 16)}$	0.206

[a]**Reported Greenhouse-Geisser value**

Table 13. Repeated-measures ANOVA output for inter-muscular coherence of IC and OB muscles during a maximum phonation duration task at twice-conversational loudness.

Summary of results for maximum duration phonation at twice-conversational loudness. Statistically significant change was not present for the trained maximum duration phonation at twice-conversational loudness task. A visual pattern was noted for LVT occurring closer to EEL and an increase PCTRC was noted across all three time points for the CP group, consistent with the maximum performance results described above. A pattern of increasing levels

of intermuscular coherence across all three time points and all three frequency bandwidths is also noted in Figure 12 for the CP group.

Sequential Motion

Kinematics. Figure 13 shows group average performance on a sequential motion task (/*pataka*/) across the three time points, on measures of LVI, LVT, PVCLVE, and PCTRC. Control children initiated the phonations between 1238 and 1585 ml above end expiratory level (EEL) and terminated these productions between 738 and 812 ml below EEL. In contrast, children with CP initiated their phonations between 444 and 550 ml above EEL and terminated between 172 and 375 ml below EEL. Control children used between 77 and 87%VC for producing sequential movements, whereas children with CP produced sequential motions using excursions ranging from 27 to 34% VC on average. Control children used between 77 to 83 PCTRC to lung volume excursion, whereas children with CP used between 65 to 77 PCTRC on average. Within groups repeated-measures ANOVAs (Table 14) were not significant in either the control or the experimental group.

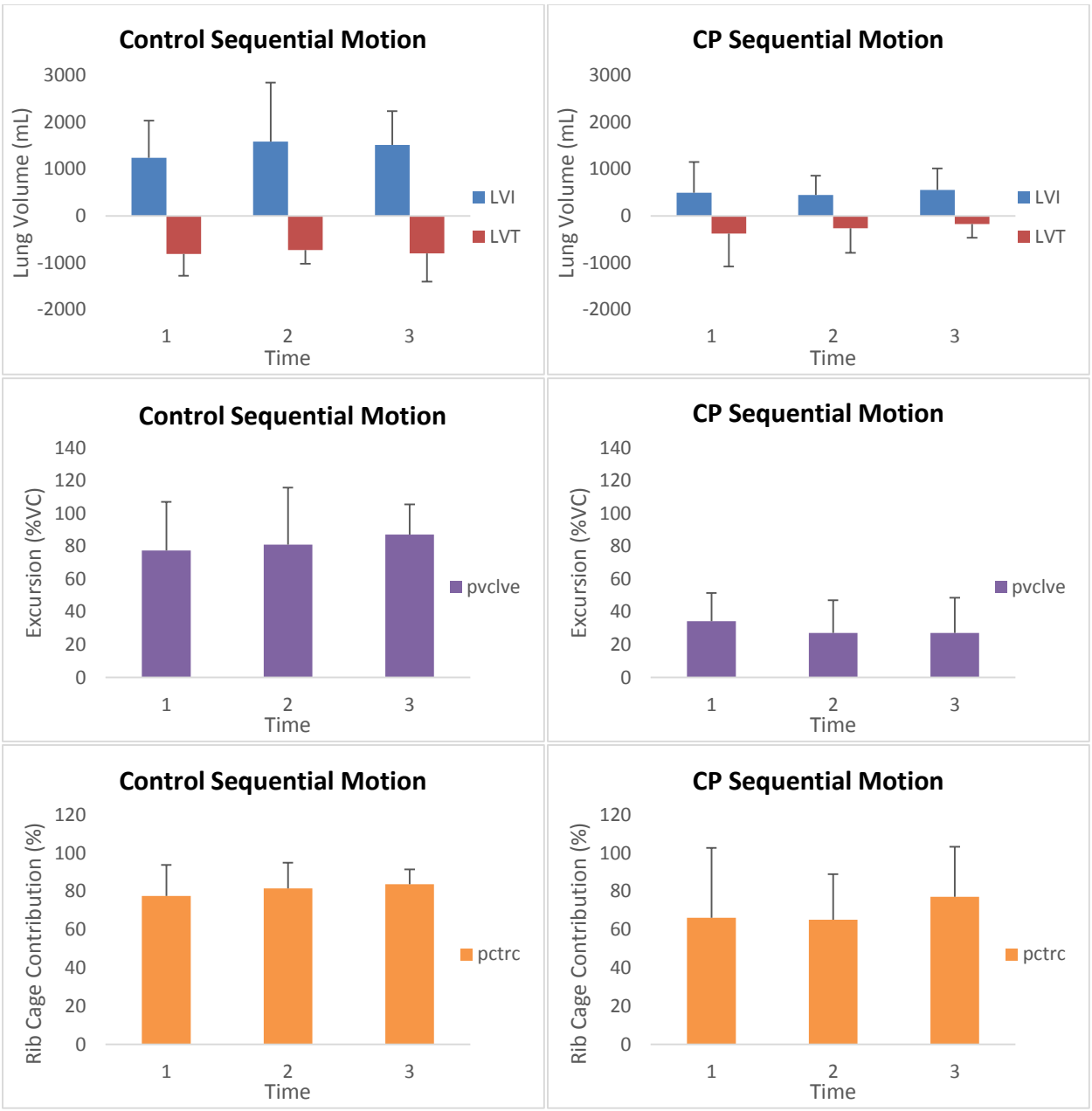


Figure 13. Group change in Lung Volume Initiation (LVI), Lung Volume Termination (LVT), Lung Volume Excursion in Percent of Vital Capacity (PVCLVE), and Rib Cage Contribution in Percent (PCTRC) over three time points, for a sequential motion task at conversational loudness.

	Control	**CP**

Task	F	Sig.	F	Sig.
LVI	.95 $_{(2, 14)}$	.41	.11 $_{(2, 16)}$	.90
LVT	.12 $_{(2, 14)}$	.89	.50$_{(2, 16)}$	.61
PCTRC	.73 $_{(2, 14)}$	.50	.59$_{(2, 16)}$	.57
PVCLVE	.53 $_{(2, 14)}$	.60	.96$_{(2, 16)}$	.40

[a] Reported Greenhouse-Geisser value

Table 14. Repeated-Measures ANOVA output for kinematics during a sequential motion task.

Muscle activation patterns. Figure 14 shows the activity of the IC and OB muscles during productions of a maximum duration phonation with articulation (/*pataka*/). The top two panels indicate at what percentage of predicted VC, muscle activity was detected (onset) and became undetectable (offset). In the control group, IC and OB muscles were activated between 50 and 64 %VC. Intercostal activation terminated between 16 to 25 %VC whereas OB activity terminated near the bottom of predicted VC. Children with CP exhibited onset of IC between 37 and 51 %VC and OB muscular activation between 28 and 45 %VC. Both IC and OB offsets occurred near the bottom of predicted VC. The bottom two panels in Figure 14 show the amount of muscular activation in %MVC for each group across time points. Control children activated IC and OB muscles from approximately 18 to 31 %MVC. On average, children with CP activated IC muscles from approximately 27 to 37 % MVC and OB muscles from 13 to 26 %MVC. Within subjects repeated-measures ANOVA (Table 15) found no significant differences over time for either group for the sequential motion productions.

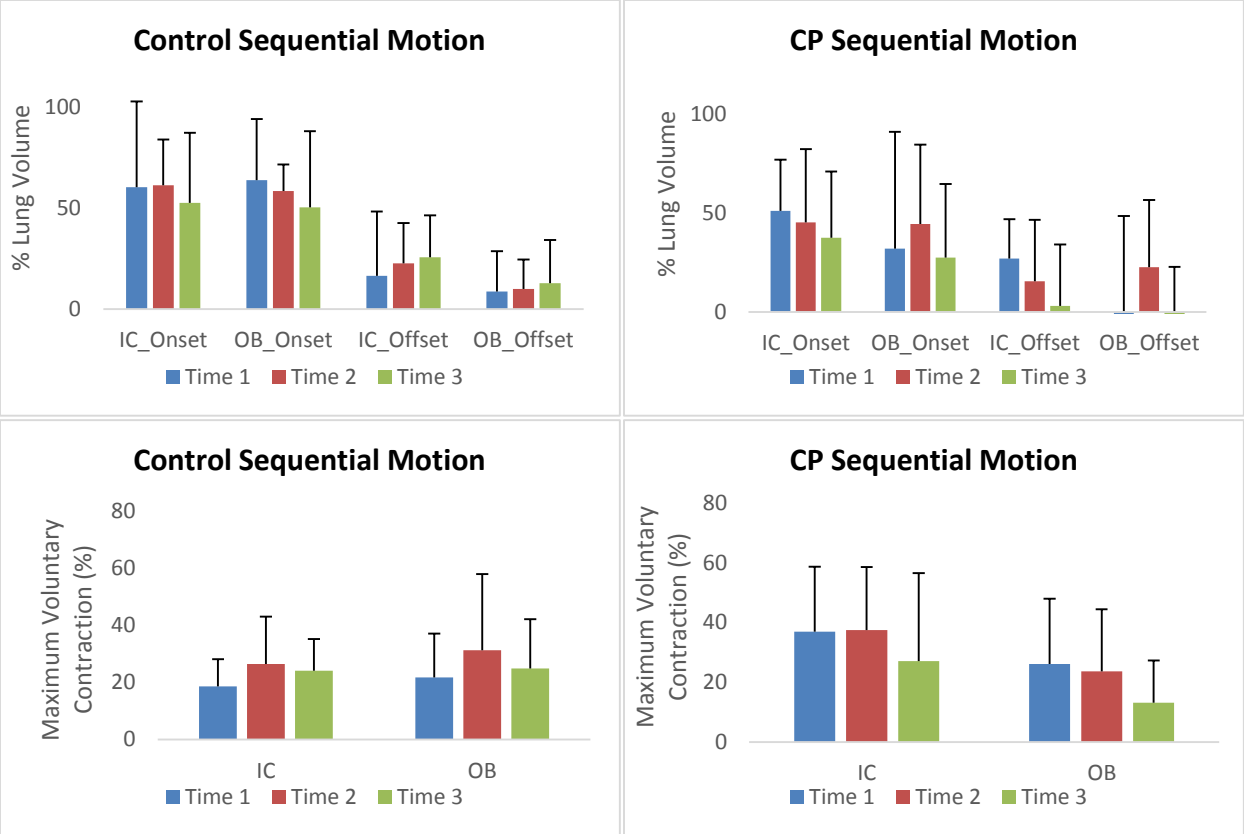


Figure 14. Effects of LSVT on intercostal (IC) and oblique (OB) onset, offset, and activation during a sequential motion task over three time points. The top two panels show the activation (onset) and deactivation (offset) of the muscles in percent predicted VC. The bottom two panels indicate activation of the muscle, as a percentage of maximum contraction.

Task	**Percent Lung Volume**				**Percent Maximum Voluntary Contraction**			
	Control		CP		Control		CP	
	F	**Sig.**	**F**	**Sig.**	**F**	**Sig.**	**F**	**Sig.**
IC_onset	$.20_{(2, 14)}$	.82	$.44_{(2, 16)}$	.65	$.68_{(2, 14)}$	.52	$1.06_{(2, 16)}$	.37
OB_onset	$.49_{(2, 14)}$	.62	$.35_{(2, 16)}$	.61[a]	$1.02_{(2, 14)}$	.39	$1.39_{(2, 16)}$	.28[a]
IC_offset	$.31_{(2, 14)}$	.74	$2.17_{(2, 16)}$	.15				
OC_offset	$.17_{(2, 14)}$	.84	$1.50_{(2, 16)}$	.25				

[a]Reported Greenhouse-Geisser value

Table 15. Repeated-measures ANOVA results for EMG onset, offset, and activation of the IC and OB muscles during a sequential motion task.

Intermuscular coherence. Figure 15 shows inter-muscular coherence between the IC and OB muscles. In general, peak coherence values for control children ranged from $r = 0.45$ to $r = 0.66$ on average across the three frequency bandwidths. Children with CP exhibited ranges

from $r = 0.25$ to $r = 0.6$ on average across the three frequency bandwidths. Within subjects repeated-measures ANOVAs found one significant change in coherence ($p < .01$) in the high frequency bandwidth for children with CP, as reported in Table 16. Paired *t*-test of time 2 and time 3 found significant change ($t = -5.063$, df = 8, $p < 0.01$), indicating an increase of coherence at time 3. No other effects were considered significant at $p < .05$, although a graphically observable trend to increased coherence in the medium frequency bandwidth for the children with CP was noted. The control group remained stable throughout the three time points.

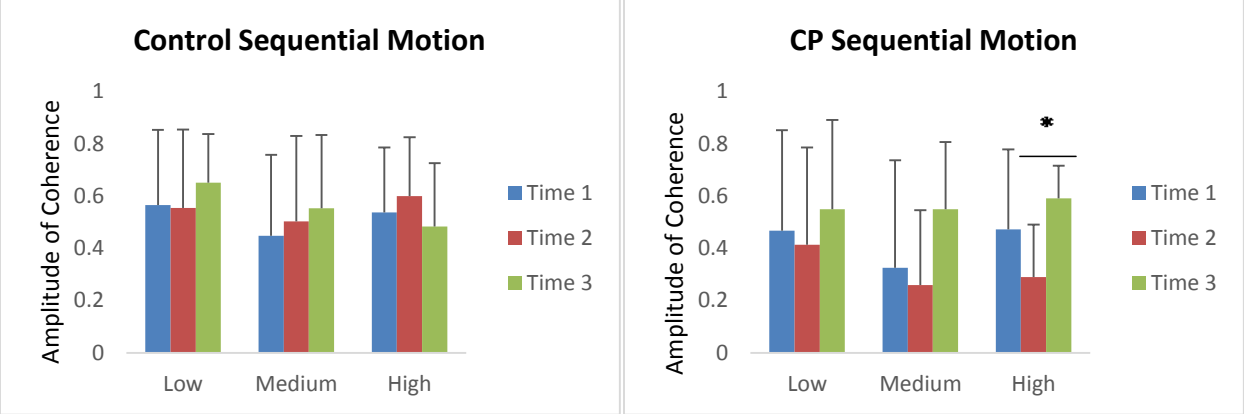


Figure 15. Effects of LSVT on chest wall peak amplitude of coherence in low, medium, and high frequencies during a sequential motion task.

Frequency	Control		CP	
	F	**Sig.**	**F**	**Sig.**
Low	$.60_{(2, 14)}$	.56	$.27_{(2, 16)}$	.77
Medium	$.29_{(2, 14)}$	.75	$1.52_{(2, 16)}$	.25
High	$.59_{(2, 14)}$	.57	$6.58_{(2, 16)}$	.008**

[a]Reported Greenhouse-Geisser value
****Highly significant p<.01**

Table 16. Repeated-measures ANOVA output for inter-muscular coherence of IC and OB muscles during a sequential motion task.

Summary of results for sequential motion. One statistically significant increase in intermuscular coherence in the high frequency bandwidth was present for the untrained sequential motion task at time 3 in the CP group (Figure 15). A visual pattern of LVT occurring

near EEL and an increase in PCTRC was noted across all three time points for the CP group, consistent with the maximum performance results described above.

Untrained Phrases Produced at Conversational Loudness

Kinematics. Figure 16 shows group average performance on a phrase repetition task across the three time points, on measures of LVI, LVT, PVCLVE, and PCTRC. Control children initiated speech between 354 and 394 ml above end expiratory level (EEL) and terminated these productions between 91 and 155 ml below EEL. In contrast, children with CP initiated speech between 393 and 432 ml above EEL and terminated near EEL. Control children used between 17 and 20 % VC for producing speech, whereas children with CP produced speech, using excursions 16 %VC on average. Control children used between 71to 79 PCTRC to lung volume excursion, whereas children with CP used between 68 to 70 PCTRC on average. Within groups repeated-measures ANOVAs (Table 17) were not significant in either the control children or the children with CP ($p > 0.05$).

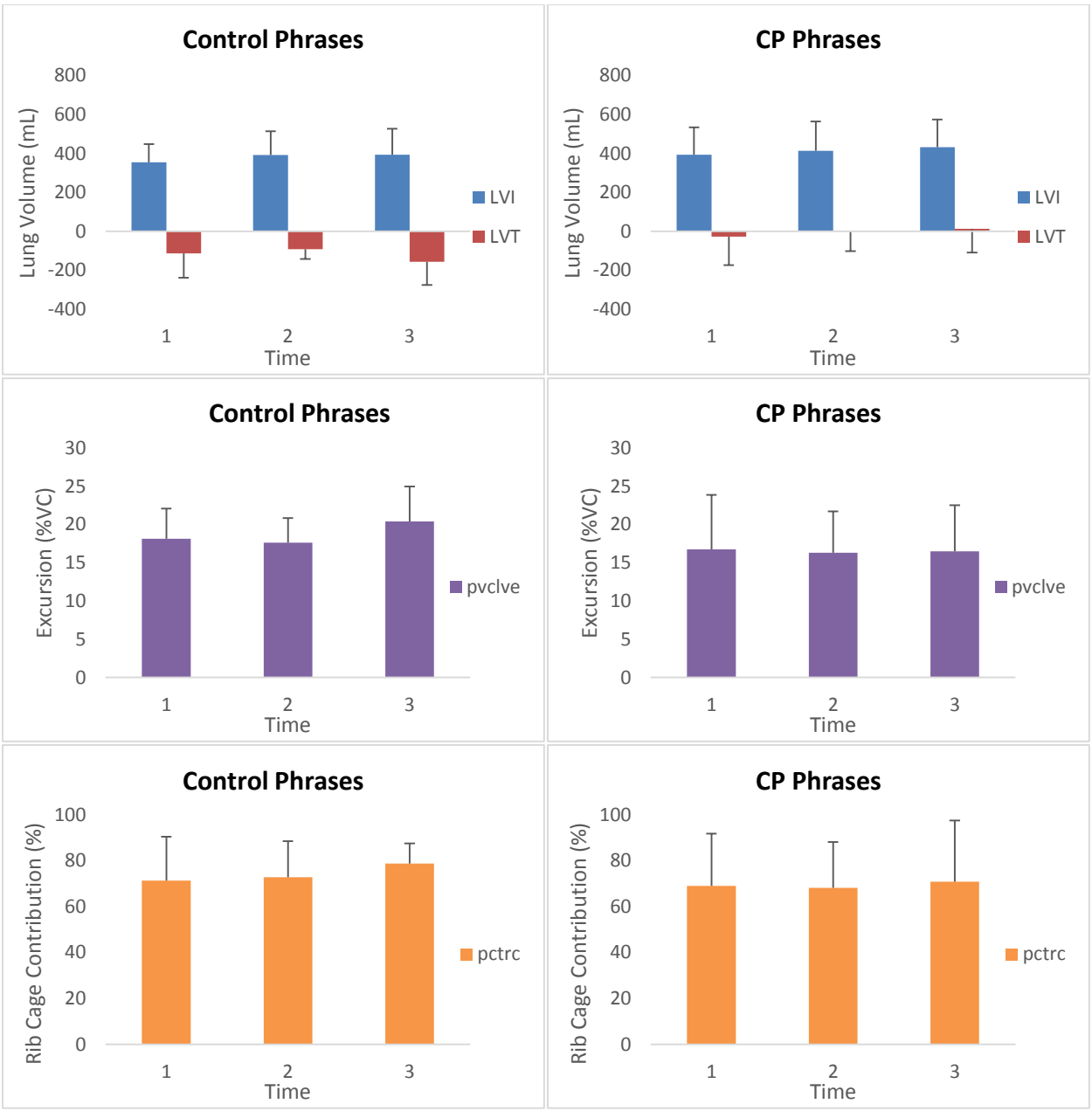


Figure 16. Group change in Lung Volume Initiation (LVI), Lung Volume Termination (LVT), Lung Volume Excursion in Percent of Vital Capacity (PVCLVE), and Rib Cage Contribution in Percent (PCTRC) over three time points, for untrained phrase repetition at conversational loudness.

Task	Control		CP	
	F	**Sig.**	**F**	**Sig.**
LVI	$.40_{(2, 14)}$	.68	$.30_{(2, 16)}$	.75
LVT	$1.19_{(2, 14)}$	.33	$.55_{(2, 16)}$	.59
PCTRC	$.90_{(2, 14)}$	.43	$.08_{(2, 16)}$	.93
PVCLVE	$2.18_{(2, 14)}$	.15	$.02_{(2, 16)}$	.98

[a] Reported Greenhouse-Geisser value

Table 17. Repeated-Measures ANOVA output for kinematics during a phrase repetition task at conversational loudness.

Muscle activation patterns. Figure 17 shows the activity of the IC and OB muscles for speech. The top two panels indicate at what percentage of predicted VC, muscle activity was detected (onset) and became undetectable (offset). In the control group, IC and OB muscles were activated between 24 and 42 %VC. Intercostal and OB activation terminated between 18 and 37 %VC. Children with CP exhibited onset of IC and OB activation between 29 and 47 %VC. Both IC and OB offsets occurred between 13 and 38 % VC. The bottom two panels in Figure 15 show the amount of muscular activation in %MVC for each group across time points. Control children activated IC and OB muscles from approximately 5 to 8 %MVC. On average, children with CP activated IC muscles from approximately 16 to 24 %MVC and OB muscles from 13 to 17 %MVC. Within subjects repeated-measures ANOVA (Table 18) showed a trend of $p = .08$ indicating a decrease in intercostal muscle activation at time 3 in the children with CP. Paired-sample *t*-tests indicated a significant change between time 2 and time 3 ($t = 2.841$, df = 8, $p < .05$).

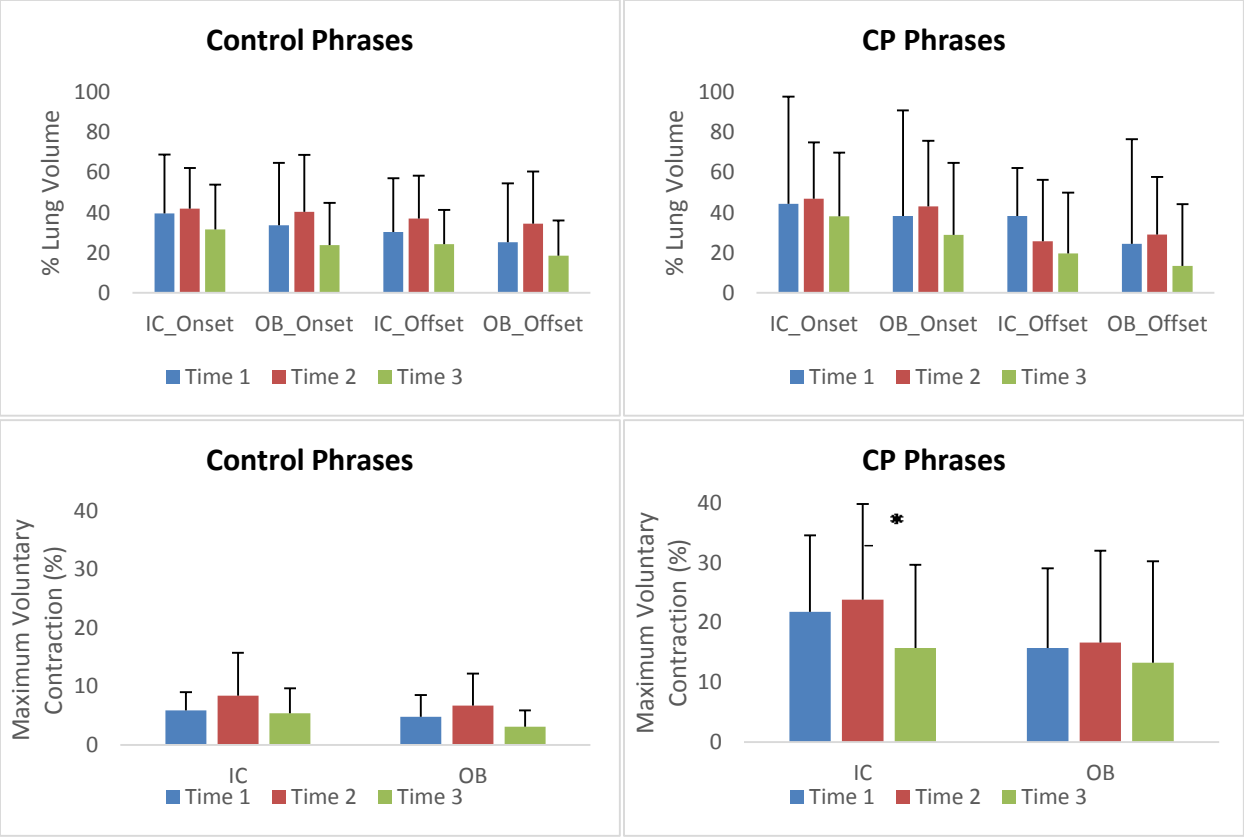


Figure 17. Effects of LSVT on intercostal (IC) and oblique (OB) onset, offset, and activation while repeating untrained phrases over three time points. The top two panels show the activation (onset) and deactivation (offset) of the muscles in percent predicted VC. The bottom two panels indicate activation of the muscle, as a percentage of maximum contraction.

Task	**Percent Lung Volume**				**Percent Maximum Voluntary Contraction**			
	Control		CP		Control		CP	
	F	**Sig.**	**F**	**Sig.**	**F**	**Sig.**	**F**	**Sig.**
IC_onset	$.50_{(2, 14)}$	.62	$.13_{(2, 16)}$	.88	$.82_{(2, 14)}$	.41[a]	$2.9_{(2, 16)}$	.08
OB_onset	$1.00_{(2, 14)}$	.39	$.32_{(2, 16)}$	.73	$2.73_{(2, 14)}$	.10	$.11_{(2, 16)}$	.90
IC_offset	$.79_{(2, 14)}$	.47	$1.25_{(2, 16)}$	.31				
OC_offset	$1.17_{(2, 14)}$	.34	$.53_{(2, 16)}$	.56				

[a]Reported Greenhouse-Geisser value

Table 18. Repeated-measures ANOVA results for EMG onset, offset, and activation of the IC and OB muscles during a phrase repetition task at conversational loudness.

Intermuscular coherence. Figure 18 shows inter-muscular coherence between the IC and OB muscles. In general, peak coherence values for control children ranged from $r = 0.59$ to $r = 0.79$ on average across the three frequency bandwidths. Children with CP exhibited ranges

from $r = 0.38$ to $r = 0.58$ on average across the three frequency bandwidths. Within subjects repeated-measures ANOVAs found no significant effects ($p < .05$) as shown in Table 19. The experimental and control groups remained stable throughout the three time points for all three frequency bandwidths.

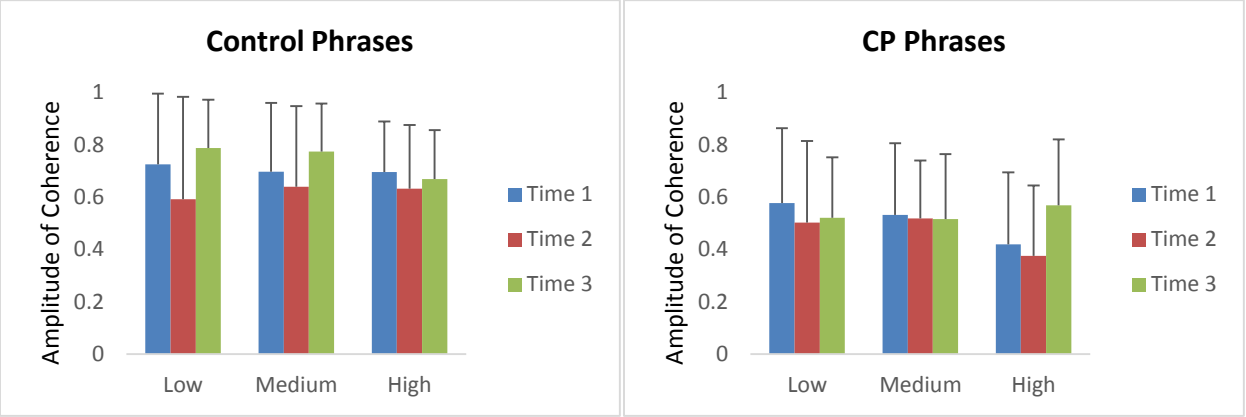


Figure 18. Effects of LSVT on chest wall peak amplitude of coherence in low, medium, and high frequencies during a phrase repetition task at conversational loudness.

Frequency	Control		CP	
	F	**Sig.**	**F**	**Sig.**
Low	$1.18_{(2, 14)}$	.34	$.19_{(2, 16)}$	.71[a]
Medium	$.83_{(2, 14)}$	.46	$.01_{(2, 16)}$	.99
High	$.36_{(2, 14)}$	.71	$1.53_{(2, 16)}$	.25

[a]**Reported Greenhouse-Geisser value**

Table 19. Repeated-measures ANOVA output for inter-muscular coherence of IC and OB muscles during a phrase repetition task at conversational loudness.

Summary of results for untrained phrases produced at conversational loudness.

One statistically significant decrease in IC relative activation was present for the untrained phrase repetition task at time 3 in the CP group (Figure 17). A visual pattern of LVT occurring near EEL and an increase in PCTRC was noted across all three time points for the CP group, consistent with the previous task results described above. In general, both kinematic and muscle activation patterns were similar between the two groups for the production of untrained phrases.

Untrained Phrases Produced at Twice-conversational Loudness

Kinematics. Figure 19 shows group average performance on a speech produced at twice-conversational loudness across the three time points, on measures of LVI, LVT, PVCLVE, and PCTRC. Control children initiated speech between 452 and 543 ml above EEL and terminated these productions between 105 and 146 ml below EEL. In contrast, children with CP initiated their phonations between 380 and 442 ml above EEL and terminated near EEL. Control children used between 21 and 24 %VC for producing speech at twice typical loudness, whereas children with CP produced loud speech, using excursions between 17 and 20 %VC on average. Control children used between 71 and 79 PCTRC to lung volume excursion, whereas children with CP used between 65 and 72 PCTRC on average. Within groups repeated-measures ANOVAs (Table 20) were not significant in either the control children or the children with CP.

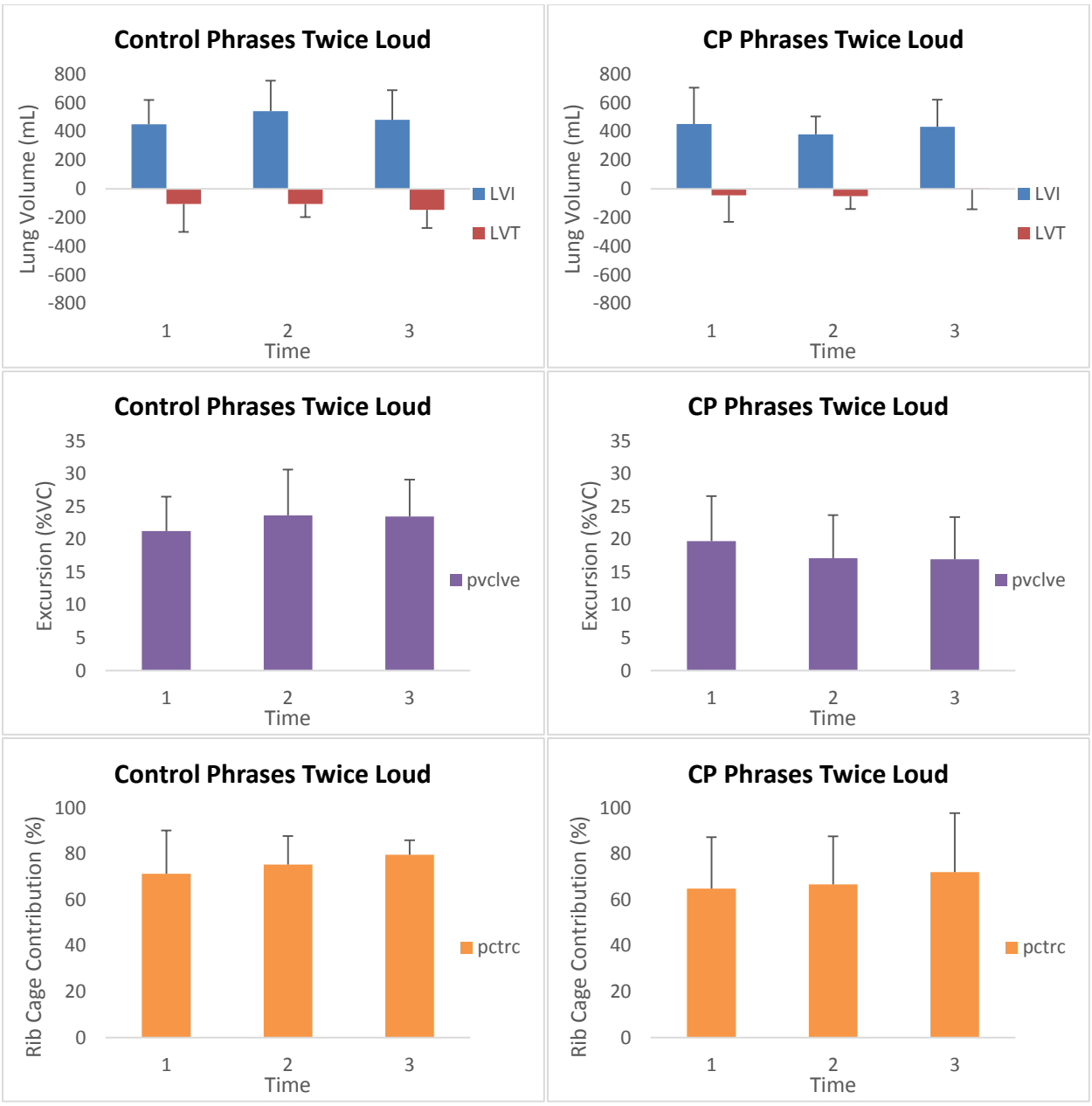


Figure 19. Group change in Lung Volume Initiation (LVI), Lung Volume Termination (LVT), Lung Volume Excursion in Percent of Vital Capacity (PVCLVE), and Rib Cage Contribution in Percent (PCTRC) over three time points, for a phrase repetition task at twice-conversational loudness.

Task	Control		CP	
	F	**Sig.**	**F**	**Sig.**
LVI	$.84_{(2, 14)}$	.45	$.31_{(2, 16)}$	.74
LVT	$.34_{(2, 14)}$	.72	$.46_{(2, 16)}$	.64
PCTRC	$.88_{(2, 14)}$	.44	$.42_{(2, 16)}$	.57[a]
PVCLVE	$.55_{(2, 14)}$	.59	$.65_{(2, 16)}$	.54

[a] **Reported Greenhouse-Geisser value**

Table 20. Repeated-Measures ANOVA output for kinematics during a phrase repetition task at twice-conversational loudness.

Muscle activation patterns. Figure 20 shows the activity of the IC and OB muscles for speech produced at twice typical loudness. The top two panels indicate at what percentage of predicted VC, muscle activity was detected (onset) and became undetectable (offset). In the control group, IC and OB muscles were activated between 27 and 51 %VC. Intercostal and OB activation terminated between 18 and 40 %VC. Children with CP exhibited the onset of IC and OB activation between 31 and 44 %VC. Both IC and OB offsets occurred between 19 and 25 % VC. The bottom two panels in Figure 20 show the amount of muscular activation in %MVC for each group across time points. Control children activated IC and OB muscles from approximately 10 to 12 %MVC. On average, children with CP activated IC and OB muscles from approximately 15 to 26 % MVC. Within subjects repeated-measures ANOVA (Table 21) showed no significant differences in onset and offset of the IC and OB muscles for either group. A significant effect of $p < .05$ was found for IC activation (%MVC) in the children with CP. A paired *t*-test showed that the difference between time 2 and 3 was significant ($t = 3.051$, df = 8, $p < .05$, two-tailed). However, significance also was found in the control group for OB activation (%MVC). Paired *t*-tests found a significant difference from time 1 to time 3 ($t = 3.662$, df = 7, $p < .01$, two-tailed). A trend of $p = .08$ was also noted in the control group for OB onset, but a paired t-test did not confirm a significant change from time 1 to time 3 ($t = 1.933$, df = 7, $p < .1$, two-tailed).

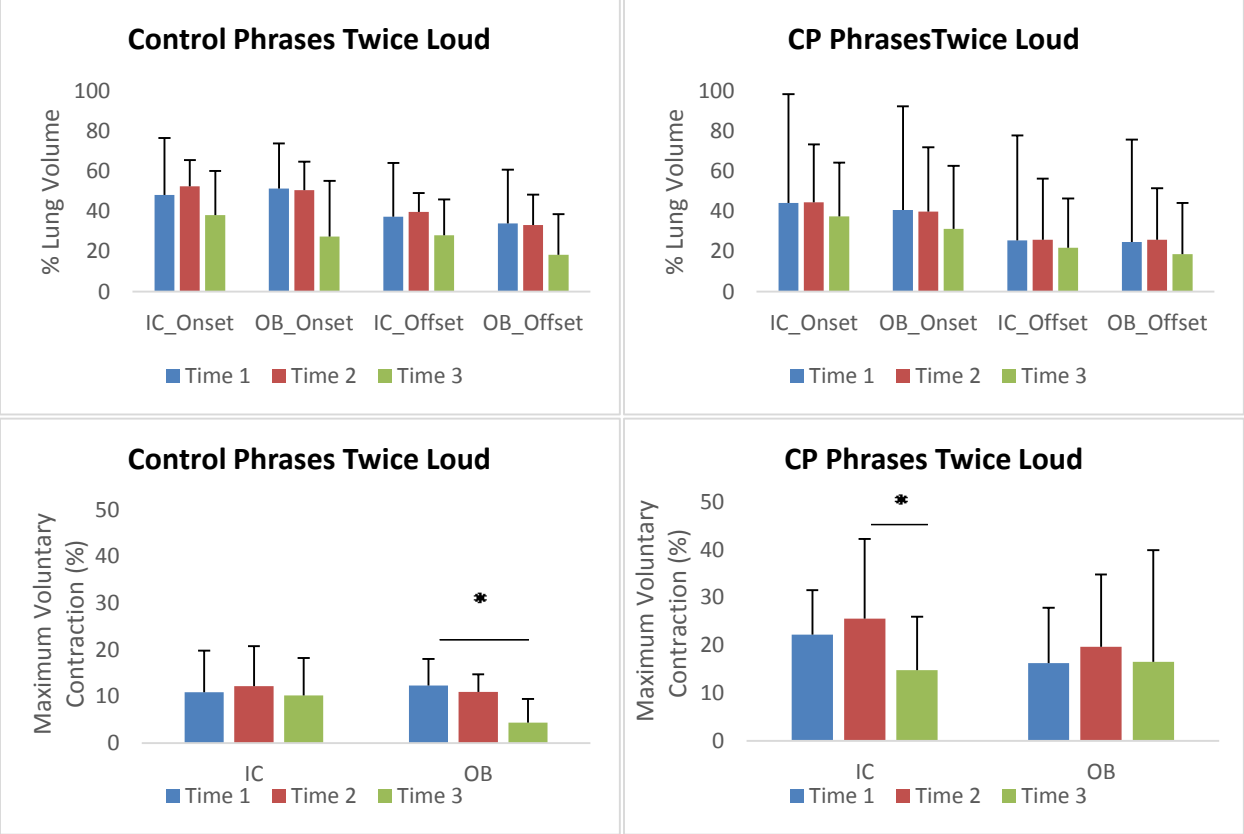


Figure 20. Effects of LSVT on intercostal (IC) and oblique (OB) onset, offset, and activation while repeating untrained phrases in a loud voice over three time points. The top two panels show the activation (onset) and deactivation (offset) of the muscles in percent predicted VC. The bottom two panels indicate activation of the muscle, as a percentage of maximum contraction.

Task	**Percent Lung Volume**				**Percent Maximum Voluntary Contraction**			
	Control		CP		Control		CP	
	F	**Sig.**	**F**	**Sig.**	**F**	**Sig.**	**F**	**Sig.**
IC_onset	$.97_{(2, 14)}$	.40	$.11_{(2, 16)}$	.89	$.22_{(2, 14)}$	.81	$3.62_{(2, 16)}$	.05*
OB_onset	$3.11_{(2, 14)}$	.08	$.18_{(2, 16)}$	.84	$4.97_{(2, 14)}$	.02*	$.10_{(2, 16)}$	.91
IC_offset	$.79_{(2, 14)}$	.47	$.04_{(2, 16)}$	.96				
OB_offset	$1.43_{(2, 14)}$	.27	$.13_{(2, 16)}$	.88				

[a]Reported Greenhouse-Geisser value
***Significance**

Table 21. Repeated-measures ANOVA results for EMG onset, offset, and activation of the IC and OB muscles during a phrase repetition task at twice-conversational loudness.

Intermuscular coherence. Figure 21 shows the intermuscular coherence between the IC and OB muscles. In general, peak coherence values for control children ranged from $r = 0.53$ to

$r = 0.73$ on average across the three frequency bandwidths. Children with CP exhibited ranges from $r = 0.38$ to $r = 0.56$ on average across the three frequency bandwidths. Within subjects repeated-measures ANOVAs found no significant effects ($p < .05$) as shown in Table 22. The experimental and control groups remained stable throughout the three time points for all three frequency bandwidths.

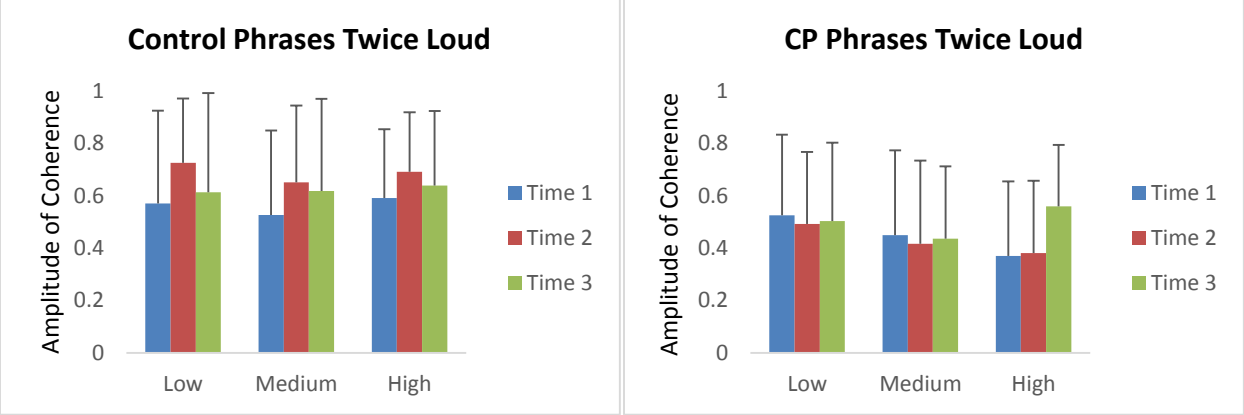


Figure 21. Effects of LSVT on chest wall peak amplitude of coherence in low, medium, and high frequencies during a phrase repetition task at twice-conversational loudness.

Frequency	Control		CP	
	F	**Sig.**	**F**	**Sig.**
Low	$.79_{(2, 14)}$	.47	$.04_{(2, 16)}$	.96
Medium	$.62_{(2, 14)}$	.48[a]	$.04_{(2, 16)}$	.96
High	$.41_{(2, 14)}$	.67	$1.59_{(2, 16)}$	.23

[a]**Reported Greenhouse-Geisser value**

Table 22. Repeated-measures ANOVA output for intermuscular coherence of IC and OB muscles during a phrase repetition task at twice-conversational loudness.

Summary of results for untrained phrases produced at twice-conversational loudness. One statistically significant decrease in IC relative activation was present for the untrained phrase repetition task at time 3 in the CP group (Figure 20), consistent with the change observed in the conversational loudness task. A significant change also was noted in the control group for a decrease in OB relative activation from time 1 to time 3. A visual pattern of LVT

occurring closer to EEL and an increase in PCTRC was noted across all three time points for the CP group, consistent with the previous task results described above.

Discussion

The purpose of this exploratory study was to investigate the acute and long-term effects of an intensive voice treatment (LSVT LOUD) on features of speech breathing control as measured by chest wall kinematics, intermuscular coherence, and muscle activation in children with varying types and severity of dysarthria secondary to CP. The results suggest that a single course of intensive treatment targeting vocal loudness may promote measurable physiological changes in the control of speech breathing in children with dysarthria and CP.

The main findings of this study were that sixteen sessions of LSVT LOUD and a 12-week maintenance program *i*) increased stability of speech breathing patterns including lung volume initiations, terminations, excursions and percent rib cage contributions to lung volume excursions; *ii*) increased IC muscular effort for the trained maximum duration phonation task; *iii*) decreased the IC muscular effort required to repeat untrained phrases using both conversational and perceived twice-conversational loudness; and *iv)* increased IC-OB intermuscular coherence in an untrained sequential motion task (DDK: /*pataka*/) requiring articulatory speed and precision. Moreover, following treatment, some speech breathing values observed in the children with CP moved towards their control counterparts. For example, following treatment, children with CP tended to terminate lung volumes closer to EEL and increased the PCTRC for lung volume excursions, both of which suggest that treatment drives the system towards increased biomechanical efficiency. Finally, each child showed positive change on at least two kinematic and three muscular activation behaviours on tasks that were

trained. Children with CP also showed changes on at least one kinematic and one muscular activation behaviour on tasks that were untrained, suggesting motor learning has occurred (see summary tables in supplementary materials).

The tasks used in this study were designed to manipulate various tracheal pressures and lung volume targets in treated and untreated contexts. Specifically, the VC manoeuvre involved a large inspiration and expiratory excursion with little to no resistance or needed maintenance of pressure in the airway. Maximum duration phonation tasks required similar lung volume excursions to the VC task, but required the maintenance of constant tracheal pressures across the VC range (Hixon et al., 1976). In contrast, speech is typically produced in the mid-range of the VC, requiring about 20 percent of the VC, and the maintenance of a constant tracheal pressure while speaking. Phonations and speech produced at perceived twice-conversational loudness required increases in tracheal pressure from approximately 5-7 cmH2O-conversational loudness to 15-20 cmH2O. Maximum duration phonations were targeted in LSVT LOUD whereas the phrases produced and measured at each of the three time points served as untrained activities. The outcomes for each of these tasks will be discussed in the context of speech breathing dynamics (i.e., kinematic and muscular events) and possible treatment effects.

Vital Capacity

The VC task was considered a maximum performance non-speech task and it was not trained during LSVT LOUD. The control group was able to start the manoeuvre near the top of their predicted VC and terminate well below EEL indicating the ability to use 100% predicted VC for this manoeuvre. Control children used predominantly rib cage contribution to lung volume excursion. Taken together, these lung volume events were in line with previous observations in adults (Hixon et al., 1973). In contrast, children with CP consistently initiated

the VC task from lower lung volumes and terminated at a higher lung volumes than the controls, using approximately 50% of their predicted vital capacity overall. This is consistent with previous research which showed that children with CP have difficulty fully inspiring to the top of their inspiratory capacity (Blumberg, 1955; Stamer, 2000). This would have an effect on the amount of air available to expire in a VC manoeuvre. However, Park et al. (2006) found much smaller differences between 6-12 year-old children with spastic quadriplegic CP and typical controls than observed in the present study. This discrepancy may reflect the younger age group average in Park et al., as GMFCS levels were similar. Whereas LVI remained relatively stable at all three time points, participants with CP were able to terminate their VC at lower lung volumes following treatment therefore increasing the amount of air expired (44% PVCLVE to 57% PVCLVE). Additionally, PCTRC increased from 66-73%, moving towards the more biomechanically efficient breathing pattern displayed by the control group (85%). The present findings suggest that LSVT LOUD may have modulated the relative contribution of the rib cage and abdomen towards a more biomechanically efficient action (Hixon et al., 1973; 1976; Hoit et al., 1990). Overall, the CP group's respiratory kinematics became less variable after intensive treatment and maintenance. This may suggest that LSVT LOUD enhanced some motor stability even on a non-targeted, non-speech task.

In the present study, control children showed that IC and OB muscles were activated near the top of predicted VC (between 70 and 83 %VC) and terminated at or near the bottom of predicted VC (0 to 24 %VC). Muscle activations patterns were generally between 57 to 59 %MVC for IC muscles and from 32 to 53 %MVC for OB muscles, indicating that control children efficiently use expiratory muscular pressures to expire to the bottom of VC. Muscular effort was employed for this task in the context of greater chest wall compliance, static lung

compliance, airway resistance, and elastic recoil pressures of the lungs at all levels of VC compared to adults (Zapletal et al., 1987). Moreover, the observations in the present study were similar to those reported by Clair-Auger et al. (under review).

Children with CP exhibited onset of IC and OB muscular activation between 47 and 71% of their predicted VC and offsets near the end of VC (0 to 21 %VC). However, it is important to recall that these children did not exhibit the same inspiratory capacity as their control counterparts nor did they demonstrate the same degree of expiratory control. On average, children with CP activated IC muscles from between 42 to 46 % MVC and OB muscles from 27 to 42 %MVC indicating they were capable of supplementing the natural recoil pressures of the lung and chest wall to enhance expiratory drive (Hixon et al. 1976; Hoit et al, 1988; McFarland & Smith, 1989).

On average, peak coherence values for control children and for participants with CP, ranged from $r = 0.10$ to $r = 0.40$ across the three frequency bandwidths. In the present study, coherence values were higher than those reported for adults (Tomczak et al., 2013), but showed a similar pattern of being lower for VC tasks than speech and other breathing tasks occurring in the mid-range of the VC. Coherence values in the present study also are consistent with Smith and Denny's bilateral diaphragm EMG recordings for deep breathing (1990). However, their study found increases in coherence for deep breathing and other non-speech activities compared to speech or speech like tasks (Smith & Denny, 1990; Stepp et al., 2011). Whereas LSVT LOUD may have had an effect on lung volume and muscular events, cortical drive to the muscles for VC manoeuvres appeared unaffected by treatment. This is contrary to studies in typical adults and adults with incomplete spinal cord injury, which found that motor-unit coherence is influenced by training (Semmler et al., 2004; Norton & Gorassini, 2006).

Maximum Duration Phonation Produced at Conversational Loudness

Maximum duration phonation tasks required similar lung volume excursions to the VC task, but in addition, required the maintenance of constant tracheal pressures across the VC range. Producing maximum duration phonations was a task targeted in LSVT LOUD. The control group was able to start the manoeuvre near the top of their inspiratory capacity and terminate phonation well below EEL. They used 91-100% of their predicted VC, with some participants exceeding the predicted value as in the VC manoeuvre. This slight decrease in LVE reflects the aeromechanics associated with maintaining constant tracheal pressure throughout the entire lung volume (Hixon et al., 1976). Controls produced biomechanically efficient excursions using primarily rib cage contribution (81-86%RC) indicating that the addition of a pressure target did not affect the relative contribution of rib cage and abdomen in these typical children. The lung volumes associated with maximum duration phonations in this study are similar to those observed in adults (Hixon et al., 1976) and in children (Clair-Auger et al., under review). Moreover, the present findings are in line with those reported by Hixon and colleagues (1976) and reviewed by Hixon and Weismer (1995) in healthy adults. Due to the anatomical configuration of the chest wall, the rib cage will reach its maximum inward movement before the abdomen, as the abdominal muscles can continue to compress against the soft organs that make up the abdominal contents. Similar observations have been observed in adults and children when phonating or speaking at lower lung volumes (Hixon & Weismer, 1995; Stathopoulous & Sapienza, 1997; Boliek et al., 2009).

In contrast, children with CP consistently initiated the maximum duration phonation task at lower and terminated at higher lung volumes than the controls, using up to 46% of their predicted VC. This is consistent with the research discussed previously that describes children

with CP as having a limited inspiratory capacity (Blumberg, 1955; Stamer, 2000). LVI and LVT remained relatively stable at all three time points. PVCLVE decreased slightly (46-37%) but excursions appeared to increase in overall stability. Children with CP showed an increase in PCTRC from 66-73%, thus, moving towards the more biomechanically efficient breathing pattern (Hixon et al., 1973). Overall, the CP group's respiratory kinematics became less variable after intensive treatment and maintenance.

In the control group, both IC and OB muscles were activated between 52 and 65 %VC. IC activation terminated between 26 to 33 %VC whereas OB activity terminated near the bottom of predicted VC. Muscle activations were generally between 21 and 39 %MVC for IC and OB muscles, indicating that control children used approximately one-third of their maximum muscular effort phonate through most of the VC. These data indicate that typically developing children used additional muscular force to maintain a constant tracheal pressure against a continuous shrinking lung volume as has been reported for adults (Hixon et al., 1976; Hoshiko, 1960; McFarland & Smith, 1989). In contrast, muscle onset and overall effort was lower in the CP group, relative to the controls. Children with CP exhibited onset of IC between 40 and 61 %VC and OB muscular activation between 38 and 51 %VC. Both IC and OB offsets occurred near the end of predicted VC. These results emphasize the difference in abilities between children with CP and their typical peers, with CP muscle onset occurring at a lower %VC. Children with CP used 24 - 39 % MVC of IC muscles, and from 14 to 27 %MVC of OB muscles. Children with CP did not use more muscular effort to sustain phonation, although they functioned lower in their predicted VC where outward recoil pressures of the chest wall are greater (Hixon et al., 1976). It may be that children with CP in the present study made additional laryngeal adjustments to maintain phonation at these lower lung volumes, which is a strategy

observed previously in older children and adults (Russell & Stathopoulos, 1988; Stathopoulos & Sapienza, 1997). There was, however, a trend in the present data indicating that children with CP increased chest wall muscular effort immediately following treatment. Raising chest wall muscular effort may be a better method for sustaining phonation than relying on adjustments of the larynx, as it does not increase strain on the laryngeal musculature (Smith, Ramig, Dromey, Perez, & Samandari, 1995; Countryman, Hicks, Ramig, & Smith, 1997).

On average, peak coherence values for control children ranged from $r = 0.50$ to $r = 0.63$ across the three frequency bandwidths, and were slightly stronger than that observed in the children with CP ($r = 0.34$ to $r = 0.56$). Coherence values for both groups were similar to those reported with adults for the same task (Tomczak et al., 2013). The present coherence findings suggest that whereas increases in muscular contraction associated with maintaining phonation against a decreasing lung volume occurred, increased effort might not have heightened the neural coupling between the intercostal and oblique muscles. Moreover, children with CP used more rib cage contributions to lung volume excursions following treatment; however, the neuromuscular control appeared to stay the same.

Maximum Duration Phonation Produced at Twice-Conversational Loudness

This is a maximum performance task requiring an increase in tracheal pressure from approximately 5-7 cmH2O-conversational loudness to 15-20 cmH2O. Phonation produced at twice-conversational loudness would be considered a *cued loudness* training task in LSVT LOUD. Control children initiated phonations well above EEL and terminated significantly below EEL, using between 84 and 100 percent of their predicted VC with some children performing beyond the predicted VC values. Control children used between 79 and 87 PCTRC, maintaining the biomechanically efficient pattern shown in the literature (Hixon et al., 1976). These values

were not affected by increasing effort to meet a higher pressure target. The lung volumes associated with this task are similar to those described in Hixon et al. (1976) for adults, and Clair-Auger et al. (under review) for children.

In contrast to their typically developing peers, children with CP initiated phonations lower and terminated higher in their predicted VC. Children with CP also produced phonations at excursions ranging from 38 to 46% on average, only half of the size of the control group's LVEs. LVI lowered and LVT rose slightly relative to EEL over the three time points. PVCLVE decreased marginally (46-37%) as well. Overall, the respiratory kinematics of the CP group were unaffected by the increased demand on tracheal pressure for this task. A small trend was noted in PCTRC moving from 66-70%, towards a more biomechanically efficient breathing pattern (Hixon et al., 1973).

In the control group, both IC and OB muscles were activated between 57 and 68 %VC. Intercostal activation terminated between 25 to 37 %VC whereas OB activity terminated near the bottom of predicted VC. Control children activated IC and OB muscles between 20 and 38 %MVC. Comparatively, children with CP exhibited onset of IC between 48 and 54 %VC and OB muscular activation between 38 and 49 %VC. Both IC and OB offsets occurred near the bottom of predicted VC. On average, children with CP activated IC muscles from between 34 to 37 % MVC and OB muscles from 22 to 31 %MVC. Pre-test muscle activation was higher than for the same task at conversational loudness, suggesting that the children with CP were already increasing muscular effort before intervention. No significant change in muscle activation was noted post-LSVT LOUD. These results show that typical children and children with CP both needed IC and OB muscular force to maintain a constant tracheal pressure whilst lung volume decreased, and were able to raise muscular force to accommodate increasing tracheal pressures

above 8 cmH_20. They may have also made laryngeal adjustments in order to maintain higher tracheal pressures, similar to adults and older children (7-10 years) as described in previous studies (Stathopoulous & Sapienza, 1997; Dromey & Ramig, 1998; Clair-Auger et al., under review).

Peak coherence values for control children ranged from $r = 0.4$ to $r = 0.6$ on average across the three frequency bandwidths. These values are consistent with control group performance on other tasks within this study, as well as that reported in the literature on adults (Smith & Denny, 1990; Stepp et al., 2010; Tomczak et al., 2013). Children with CP exhibited ranges from $r = 0.2$ to $r = 0.59$, which were similar to controls. However the children with CP were more variable than their control counterparts. There appears to be a visual trend for an increase in peak coherence immediately following treatment and 12-weeks post treatment across all frequency bandwidths in the children with CP. These values suggest that LSVT LOUD may have had some influence on the neural coupling between the intercostal and oblique muscles for this trained task. Previous research has also shown a training effect for intermuscular coherence of leg muscles (Ushiyama et al., 2010) and hand muscles (Semmler & Nordstrom, 1998; Semmler et al., 2004). These studies found that participants who were trained in a skilled task recorded consistently lower coherence values than those who trained in a strength task. This may suggest that LSVT LOUD training is strength-based, and that it is sufficient to affect the neural control of the chest wall muscles for this task.

Sequential Motion

The 'pataka' maximum performance task required maintenance of constant tracheal pressure across the entire lung volume, plus quick and accurate articulation. This is an untrained task in LSVT LOUD. Control children initiated the phonations high above end expiratory level

(EEL) and terminated well below EEL. Control children used between 77 and 87 %VC for producing sequential movements, in line with those reported for maximum duration phonations by Hixon and colleagues (1976) and reviewed by Hixon and Weismer (1995) in healthy adults. The control children were not able to phonate through their entire VC due to inability to generate enough tracheal pressure at low lung volumes, similar to previous observations of children and adults when phonating or speaking at lower lung volumes (Hixon & Weismer, 1995; Stathopoulous & Sapienza, 1997; Boliek et al., 2009). Control children used between 77 to 83 PCTRC to lung volume excursion, maintaining the relative contribution of rib cage and abdomen used by typical older children and adults (Hixon et al., 1976; Clair-Auger et al., under review). In contrast, children with CP did not initiate DDKs at as high of lung volumes as observed in the controls and terminated slightly below EEL, using excursions ranging from 27 to 34% VC on average. This is consistent with the values from previous maximum performance tasks in the present study, as well as in previous research on the limited inspiratory capacity of children with CP (Blumberg, 1955; Stamer, 2000). LVI remained stable across all three time points, while LVT rose slightly towards EEL at times 2 and 3. Terminating at higher lung volumes may indicate a positive therapeutic effect of LSVT LOUD, as children with CP often terminate too low in their VC (Boliek & Fox, 2014; Boliek et al., in preparation). PVCLVE decreased slightly post-treatment, and maintained this change for 12 weeks. Children with CP had an average of 65 PCTRC at the pre-test, which rose to 77 PCTRC at time 3. This trend may reflect the ability of LSVT LOUD to promote chest wall configurations that are more biomechanically efficient (Hixon et al., 1976), even on untrained tasks.

In the control group, IC and OB muscles were activated between 50 and 64 %VC. These values are similar to their performance on the maximum duration phonation task at

conversational loudness, indicating that control children did not modify the onset of the chest wall muscles in order to accommodate for the addition of an articulatory target. However, while OB activity remained the same, IC activation terminated 10%VC lower for the sequential motion task than for maximum duration phonation. Control children also activated IC and OB muscles slightly less than for maximum duration phonation, suggesting that typical children are able to modify the timing of muscular activity without necessarily increasing effort. Children with CP also exhibited onset of IC and OB 5-10% lower in their VC than in the maximum duration phonation tasks, and maintained IC and OB offsets near the bottom of their predicted VC. The children with CP used similar muscular effort to that of the maximum duration phonation task, perhaps suggesting that they did not require additional muscular force to complete the task. Another possible conclusion is that the children with CP were unable to increase muscular effort, even if it may have been beneficial for the task.

In general, peak coherence values for control children ranged from $r = 0.45$ to $r = 0.66$ on average across the three frequency bandwidths. The control group remained stable throughout the three time points. Children with CP exhibited similar values, from $r = 0.25$ to $r = 0.6$ on average across the three frequency bandwidths. One statistically significant change was noted in the high frequency bandwidth. The high frequency bandwidth is associated with cortical signalling, indicating a potential increase in cortical control of speech breathing for the sequential motion task. Also an increased coherence trend was observed in the medium frequency bandwidth for the children with CP from pre-treatment to immediately after LSVT LOUD and 12-weeks of maintenance. It is possible that loudness training heightened the neural coupling between the intercostal and oblique muscles for this task. In combination with the change in PCTRC and LVT, this result may suggest that the neuromuscular control of speech

breathing was affected by LSVT LOUD by strengthening the common cortical drive to the chest wall. These results are similar to the increase in coherence after leg training in participants with incomplete spinal cord injury and moderate leg function (Norton and Gorassini, 2006). Our results may also concur with the literature on skill versus strength training (Ushiyama et al., 2010; Semmler & Nordstrom, 1998; Semmler et al., 2004). One piece of LSVT LOUD is repetition of several target phrases that are functional in the trainee's life. If this is considered skilled training (rather than strength), it is possible that LSVT may have promoted increased coherence for a similarly skilled sequential motion task.

Untrained Phrases Produced at Conversational Loudness

The phrases used in testing were not those trained in LSVT LOUD. Control children initiated speech approximately 375 ml above EEL and terminated 125 ml below, using between 17 and 20 % VC. These results agree with previous literature on speech breathing which found that speech occurs in the midrange of the lung volume and uses approximately 20% of VC in adults (Hixon et al., 1973) and in children (Hoit et al., 1990; Boliek et al., 2009). Children with CP initiated speech at slightly higher lung volumes and terminated near EEL, using excursions of 16 %VC. These values were also similar to those of typical children in the same age range Hoit et al. (1990). LVI and LVT rose slightly over the three time points for children with CP, while PVCLVE remained stable. This may reflect another positive treatment effect, as speaking at lower lung volumes (e.g., below end expiratory level) is less biomechanically efficient than ending breath groups at or above end expiratory level (Boliek et al., 1997; 2009). In terms of rib cage contribution to breathing, control children used between 71to 79 PCTRC, similar to the 80% described in Hixon et al. (1976) for adults and larger than the 51% observed for 6 year-olds, as expected (Boliek, et al., 2009). This is consistent with previous research on children from age

7 to 16 years (Hoit et al., 1990) and 4 to 14 years (Stathopoulos & Sapienza, 1997) showing that speech breathing becomes adult-like between the ages of 10 to 12 years. The CP group also used higher rib cage contributions than typical younger children, but were 10% lower than the control group on average. This is consistent with the findings of Redstone et al. (2002) that children with CP use non-optimal rib cage vs. abdominal contributions for speech breathing.

In the control group, IC and OB muscles were activated between 24 and 42 %VC and terminated between 18 and 37 %VC. Similarly, children with CP exhibited onset of IC and OB activation between 29 and 47 %VC and offsets between 13 and 38 % VC. Control children used IC and OB muscular effort of approximately 5 to 8 %MVC, indicating that minimal chest wall muscular effort was required for typical children to complete a phrase repetition task at conversational loudness. These values are consistent with previous literature (Hixon et al., 1973, Hixon et al., 1976; Hoit et al., 1990). On average, children with CP activated IC muscles from approximately 16 to 24 % MVC and OB muscles from 13 to 17 %MVC. These values indicated that children with CP needed to use significantly higher muscular effort for a phrase repetition task than their typically developing peers. A trend was noted in decreasing intercostal muscle activation at time 3, potentially indicating that less muscular effort was necessary to repeat phrases at conversational loudness post-LSVT LOUD. This effect also was shown in the phrase repetition task at twice-conversational loudness (below), and therefore may reflect a positive therapeutic effect of LSVT LOUD or spreading effects to untrained speech tasks. This spreading effect also has been reported elsewhere (Fox & Boliek, 2012; Boliek & Fox, 2014).

On average, peak coherence values for control children ranged from $r = 0.59$ to $r = 0.79$ across the three frequency bandwidths. Children with CP displayed lower coherence values ($r = 0.38$ to $r = 0.58$). Both control and CP groups remained stable throughout the three time points

for all three frequency bandwidths. These values agree with previous literature, which also reported consistent respiratory intermuscular coherence during speech (Smith & Denny, 1990; Stepp et al., 2010; Tomczak et al., 2013). However, the maximum value for coherence from these studies of typical adults was 0.40, lower than our findings. This may be explained by the more limited range of frequency bandwidths examined in the previous studies. For example, Smith and Denny used 20 – 60 Hz, and our study ranged from 2 – 60 Hz. Also, the speech task in Smith and Denny's study involved a reading passage constructed to contain long phrases and sentences, which might have increased task complexity in comparison to the complexity needed for the simple sentence repetition task used in the present study. The lower coherence observed in children with CP compared to controls may reflect that speaking is more difficult and may require the engagement of a distributed neuromotor control network. This interpretation is supported by Stepp et al. (2010), who found that adults exhibited lower coherence on tasks requiring divided attention (saying digits backwards), which they considered to be more difficult than typical speaking tasks.

Untrained Phrases Produced at Twice-Conversational Loudness

Speech is typically produced in the mid-range of the VC, requires about 20% of the VC, and the maintenance of a constant tracheal pressure while speaking. Speech produced at perceived twice-conversational loudness requires increases in tracheal pressure from approximately 5-7 cmH2O-conversational loudness to 15-20 cmH2O. Control children initiated speech approximately 500 ml above EEL and terminated 125 ml below EEL, using between 21 and 24 %VC. Cueing the children to speak louder therefore raised the LVI, but did not affect LVT. In contrast, children with CP did not raise their LVI, initiating speech at 410 ml above EEL and terminating slightly below EEL. They produced loud speech using excursions between

17 and 20 %VC on average. LVI remained stable over all three time points for children with CP, but LVT rose slightly towards EEL at time 3 and therefore PVCLVE decreased slightly. These changes were maintained at 12 weeks post-treatment. Values for PCTRC were similar to the phrases at conversational loudness for both groups, and reflect values found in the literature (Hixon et al., 1973; Hoit et al., 1990; Boliek et al., 2009). PCTRC also increased from 65% at time 1 to 72% at time 3 for children with CP, consistent with the pattern shown in the previous tasks.

In the control group, IC and OB muscles were activated between 27 and 51 %VC and terminated between 18 and 40 %VC. Control children activated IC and OB muscles from approximately 10 to 12 %MVC. Significance was found in the control group for a decrease in OB activation (%MVC) from time 1 to time 3. This may point to performance variability from day-to-day in typical children at this age and is not likely due to maturation. Children with CP exhibited the onset of IC and OB activation between 31 and 44 %VC and offsets between 19 and 25 % VC. On average, children with CP used 15 to 26 % MVC of IC and OB muscles, indicating that they needed more muscular force to perform this task than the control group. Both groups increased their muscular effort in order to increase tracheal pressures associated with changes in vocal loudness (Hixon et al., 1976). A significant effect was found for IC activation (%MVC) in the children with CP between times 2 and 3, indicating a decrease in muscular effort. This effect was also shown in the phrase repetition task at conversational loudness, and therefore may reflect a positive therapeutic effect of LSVT LOUD on the untrained speech task. Specifically, increases in strength and endurance may have played a role in the relative effort required for speaking following treatment (Ramig & Dromey, 1996).

Intermuscular coherence values were not significantly affected by the higher pressure target in this task. Peak coherence values for control children ranged from $r = 0.53$ to $r = 0.73$ across the three frequency bandwidths. Children with CP exhibited slightly lower values than the control group, from $r = 0.38$ to $r = 0.56$. Both groups remained stable throughout the three time points for all three frequency bandwidths, showing no treatment effect of LSVT LOUD on coherence for this task.

Conclusions

This preliminary study on the use of an intensive vocal loudness treatment (LSVT LOUD) for children with CP and dysarthria shows a promising response from a population with extreme individual variability. The results build on our understanding of treatment at the *Phase I* level (Robey & Schultz, 1998; Robey, 2004). The main findings of this study were that LSVT LOUD had an effect on the respiratory biomechanics of speech and non-speech tasks in children with CP. Changes were also evident in the muscular effort used for speaking, and in the cortical drive for untrained maximum performance tasks requiring articulatory speed and precision. In some areas, values associated with speech breathing became more similar to their typically developing peers.

Limitations

The sample size of this preliminary *Phase I* treatment study was small. Convenience sampling could have led to a selection bias in that the children and families who volunteer for an intensive treatment study have more time and interest in this type of therapy than the average through the entire population. Also, compliance during the maintenance program was tracked but not formally enforced, which may have introduced some variability. Control participants did not undergo any intervention during the study, but may have been involved in other strength

training activities outside of the study may have affected respiratory control. Control and CP participants were not matched for height and weight, variables which affect their predicted vital capacities. Control children were also not matched for cognitive level to their CP counterparts, and this may have affected their performance on the test tasks. Although visual analysis identified several important treatment effects of LSVT LOUD on the speech breathing of children with CP, the behavioural and neural variability across participants resulted in difficulties detecting statistical significance. The CP group was heterogeneous in age, type of motor speech disorder, severity of motor speech disorder, level on the GMFCS, and cognitive level. A possible method of reducing this variability would have been to limit all selection criteria but one (e.g. age, speech diagnosis, severity). The lack of group findings may have been due to the unique responses of individuals to the treatment. However, some variability is present even in the control group reflecting an age range that was inclusive of the entire refinement stage of speech breathing development (Boliek et al., 2009; Hoit et al., 1990).

Future directions

The purpose of this exploratory study was to investigate the acute and long-term effects of an intensive voice treatment (LSVT LOUD) on features of speech breathing control as measured by chest wall kinematics, intermuscular coherence, and muscle activation in children with varying degrees and types of dysarthria secondary to CP. The results suggest that a single course of intensive treatment targeting vocal loudness may promote measurable physiological changes in the speech breathing patterns of children with dysarthria and CP. The results from this study could be combined with additional data on resting breathing and conversational speech to provide a more complete description of respiratory behaviour in typical children and children with CP. Also, these data could be interpreted along with data on breath group length, rate, and

airflow. Further analysis of variability in performance within each individual across the three time points may provide more insight in to the stability phenomena observed across the CP group. It may also be valuable to examine the performance of the participants with differing speech diagnoses against each other (e.g. those participants with and without apraxia of speech). Age also may have been a significant factor in the individual response to treatment, as younger children display higher levels of lung and chest wall compliance and may respond differently to this type of treatment than older children (Blumberg, 1955; Stathopoulous & Sapienza, 1997; Solomon & Charron, 1998). Between-groups analysis of participants with mild and severe speech diagnoses may yield important information about response and non-response to treatment, as in Norton and Gorassini (2006). The individual summary tables provided in the supplementary materials will allow for these types of analyses. Future directions should include larger sample sizes and the examination of the specific effects of LSVT LOUD on participants with different types and severity levels of CP and dysarthria.

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