The Effects of Intensive Expiratory Muscle Strength Training (EMST) on Respiratory and Speech Function in Adults with Cerebral Palsy: A Case Study

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Short Header: Effect of EMST on Respiratory and Speech Function

ABSTRACT

Expiratory Muscle Strength Training (EMST), or exhaling against high resistance at the airway opening, is a procedure that requires high chest wall muscular effort. High expiratory effort practiced over time is thought to strengthen the neural control of these chest wall muscles as measured by changes in intermuscular coherence. Previous research using EMST also has shown to positively impact speech, voice and swallowing function in adults with neurogenic communication disorders that includes Multiple Sclerosis and Parkinson Disease. The current project sought to explore the effects of EMST in an adult with cerebral palsy (CP). Changes to maximum expiratory pressures (MEPs), chest wall intermuscular coherence, chest wall kinematics, voice, speech, and quality of life following a two-week training period with the EMST device were investigated in one individual with severe CP. Findings showed that though the primary variable, MEP, did not change, intermuscular coherence indicated improvement in respiratory coordination. Positive changes were noted in other variables including vocal loudness and sustained phonation length. The findings suggested that coordination between speech subsystems (respiratory and laryngeal) may have improved as a result of the training procedure, leading to perceived improvements in the participant's functioning and quality of life related to speech, voice, and swallowing abilities.

Keywords: Expiratory Muscle Strength Training (EMST), Cerebral Palsy (CP), Maximum Expiratory Pressure (MEP), intermuscular coherence

INTRODUCTION

In utero stroke (resulting in cerebral palsy) is the most common cause of movement disorders in children, with a prevalence of 2-2.5 cases per 1000 live births (Mutch, Alberman, Hagberg, Kodama, & Perat, 1992). Cerebral palsy (CP) is a condition that requires life-long rehabilitation (Hilberink et al., 2007). Over 25% of individuals with CP have speech disorders that result in a reduction in social functioning and quality of life. When the respiratory subsystem is affected, individuals with CP have unfavourable vocal loudness, utterance length, inspiratory timing, and voice quality. A key contributor to speech breathing limitations is weakness in respiratory musculature, poor coordination of respiratory and laryngeal subsystems, or a combination of both. Individuals with CP also have a high risk of dysphagia across their lifespan (Baladin, Hemsley, Hanley & Sheppard, 2009). Few treatments have been proven effective for voice, speech, and swallowing disorders secondary to CP. However, research in other neurological populations, such as multiple sclerosis (MS) and Parkinson disease (PD), has shown that using respiratory therapies such as expiratory muscle strength training (EMST) can lead to improvements in speech and swallow functioning (Chiara, Martin & Sapienza, 2007; Pitts, Bolser, Rosenbek, Troche, Okun, & Sapienza, 2009). EMST is the training and strengthening of respiratory muscles, with the goal of improving maximal and submaximal expiratory pressure generation, or the amount of resistance one can exhale against. This is thought to drive the respiratory system and promote neuromuscular change.

Previous Research and EMST

Sapienza and colleagues (Sapienza, Troche, Pitts, & Davenport, 2011) have shown that maximal and submaximal expiratory pressure generation was favourably altered after EMST. Work by Tomczak, Pedersen, Esch and Boliek (in revision) found that EMST also may enhance common cortical drive (measured by intermuscular coherence as described in detail below) to the chest wall muscles used during speech production. Other related behaviours have been found to improve with respiratory muscle training. Vital capacity and maximum phonation, both measures of the largest volume of air one can voluntarily expire, increased subsequent to EMST (Sapienza et al., 2011). Finally, EMST has been utilized to facilitate change in cough and swallowing functions (Sapienza et al., 2011).

Chiara and colleagues (Chiara et al., 2007) study showed that eight weeks of EMST resulted in increased maximum pressure generation by participants with MS, correlating with changes in voice and speech production. Individuals were able to produce a longer phonation and an increase the number of words spoken per minute (Chiara et al., 2007). Moreover, a subgroup of participants had a significant change in the impact of dysarthria on quality of life, a positive indicator of the functional effectiveness of EMST in people with neurological disorders (Chiara et al., 2007).

EMST also has been shown to improve expiratory pressure generation in people with Parkinson disease (PD). Pitts et al. (2009) examined the effectiveness of EMST on improving cough and swallow function in a group of adults with PD who aspirated. Following four weeks of EMST, researchers measured an increase in maximum expiratory pressure (MEP). In addition, measures of effective cough and swallow function, or the timing of respiratory activity during swallowing, also improved after EMST (Pitts et al., 2009). Taken together, these changes led to a decrease in penetration/aspiration scores during a video fluoroscopy exam (Pitts et al., 2009). Improvements to swallowing might have been the result of increased expiratory lung volume, increased sensitivity of sensory receptors of the tongue and oropharynx, and/or increased hyolaryngeal displacement with increased expiratory force (Kim & Sapienza, 2005). EMST also has been found to result in an improvement in muscles used for laryngeal adduction and velopharyngeal closure (Pitts et al., 2009; Sapienza & Wheeler, 2006), which should have positive influences on swallowing as well as on voice quality and vocal loudness (Kim & Sapienza, 2005). Based on research using EMST in individuals with varying neurological deficits, it was anticipated that EMST would increase expiratory muscle strength, resulting in changes to voice, speech, swallowing and coughing in individuals with CP.

Intermuscular Coherence

Frequency analysis is used to analyze neuronal synchrony between two distinct measurement sites and is based on the correlation between the two separate signals (e.g., two muscles – intermuscular coherence, brain to muscle – corticomuscular coherence) in the time and frequency domain (Grosse, Cassidy & Brown, 2002). The main measure of correlation between two signals in the frequency domain is coherence (Grosse et al., 2002). Coherence is reported as a number between zero and one, where one indicates a perfect linear relationship at a set frequency and zero indicates the signals are not linearly related (Grosse et al., 2002). Therefore, the degree of dependence of respective signals in the frequency domain can be inferred through coherence analysis of EMG activity between synergistic muscles during a movement task (Tomczak et al., 2013). Previous research suggests that coherence in the 15 to 35 Hz frequency range is under primary motor cortex control (Farmer et al., 1993; Feurra et al., 2011; Grosse et al., 2002; Kilner et al., 1999 as cited by Tomczak et al., 2013). Previous work has shown that excitability in the motor cortex, in response to limb muscle training, can be inferred by a change in intermuscular coherence (Halliday et al., 2003, Norton and Gorssini 2006; Perez et al. 2012 as cited by Tomczak et al., 2013). Intermuscular coherence was used by Tomczak et al. (2013) to infer amount of common cortical drive to chest wall muscles following transcranial direct current stimulation across different speech and non-speech tasks. The authors found that the degree of intermuscular coherence, measured as coherence amplitude, was task dependent. Those findings suggest that greater common cortical drive to chest wall muscles was necessary for speech and conscious tidal breathing tasks in comparison to tasks that used breathing during maximum performance tasks.

Studies by Semmler and Nordstrom (1998) and Semmler, Sale, Meyer and Nordstrom (2004) investigated whether skill or generalized strength training increased of common cortical drive during weak abduction of the index finger. In their studies, the skill training groups were comprised of experienced musicians who played instruments that required independent finger movement of both hands such as the piano or flute. Participants who had engaged in several years of weight training, but had not engaged in specific hand strength training, were used for the strength condition. Results of both studies indicated that greater coherence was observed in the strength-trained individuals than the skill-trained individuals (Semmler & Nordstrom, 1998; Semmler et al., 2004). Those findings suggest that cortical adaptive control of movement may be strength, but not skill, dependent. Because the current study entails a muscle strength-training program, we expected to observe an increase in intermuscular coherence following EMST in our participant.

Chest Wall Kinematics

Abdominal and rib cage volumes can be measured using respiratory inductance plethysmography (RIP) (Respitrace; Ambulatory Monitoring, Ardsley, NY) (Hixon, Weismer & Hoit, 2008). This process involves placing respibands around the rib cage and abdomen that sense changes in the average cross-sectional areas of the rib cage wall and abdominal wall during breathing and speaking tasks. RIP was selected for this study to measure volume changes during maximum phonation and speaking tasks. RIP has been previously used to measure chest wall kinematics to determine volume-dependent differences across behavioural tasks in response to transcranial direct current stimulation (Tomczak, Greidanus & Boliek, 2013). Tomczak et al., (in revision) also have used inductance plethysmography to measure lung and abdomen volume changes secondary to acute EMST in healthy adults. These previous studies used simultaneous kinematic and chest wall surface EMG to measure lung volume and muscle activation events across tasks varying in lung volume, tracheal and oral pressures during maximum performance tasks (Tomczak et. al., 2013; Tomczak et al., in revision).

HYPOTHESES

Previous research suggests that EMST may lead to improvements in the respiratory related functions of speech and swallowing, and ultimately improved communication and swallowing aspects of quality of life. The intensive nature of the protocol used in the present study also complies with the principles of activity-dependent neural plasticity; thus any observed changes in voice, speech, and/or swallowing lend themselves to interpretation in this context. The researchers hypothesized that employing intensive EMST would improve maximal expiratory pressure, intermuscular coherence, respiratory kinematics, and acoustic measures of voice and speech, leading to improvements in respiratory and speech function. In addition, it was hypothesized that secondary benefits would be observed in communication and swallowing as assessed on standardized quality of life measures.

METHODS

Case Study Participant

One 23-year-old adult male with severe spastic quadriplegia CP was recruited for a case study through a contact at the *Cerebral Palsy Association of Alberta*. The participant was medically stable and seizure-free, was non-ambulatory, and had severe dysarthria with concomitant oral motor control difficulties that affected his ability to control his saliva. The participant also had Tourette's Syndrome, as reported by his mother. The participant's arms were generally restrained using bands attached to the table on his wheelchair. At the time of the study, the participant was living with a roommate and receiving in-home support services.

Procedures

The Health Research Ethics Board at the University of Alberta approved this study. Following written and informed consent, the participant completed a demographic questionnaire. The participant also subjectively rated swallowing and communication abilities using four quality of life scales. The four questionnaires addressed quality of life related to voice, speech, and swallowing abilities, and were completed with assistance from a family member. To assess speech- and voice-related quality of life, the participant completed the Speech Handicap Index (SHI) and Voice Handicap Index (VHI) respectively. The measures used to assess swallowing-related quality of life were the SWAL-QOL and the MD Anderson Dysphagia Inventory (MDADI). The participant was oriented to testing procedures and familiarized with the respiratory muscle trainer. Following the brief orientation, EMG electrodes and inductance plethysmography rib cage and abdominal bands were positioned on the chest wall and surface EMG electrodes were placed over intercostal and oblique muscles on the right side of the body in accordance with placement methods used by Tomczak et al. (2013). The participant was seated with his wheelchair back inclined to approximately 120 degrees.

Acoustic Measurement of Speech

Acoustic measures have been used in previous studies to assess the outcome of EMST in various populations. In their study using EMST with adults having MS, Chiara et al., 2007, included measures of maximum duration of a vowel sound ("ah") at conversational and perceived twice conversational loudness. Their results showed that maximum phonation at twice conversational loudness increased in duration following EMST. A review by Sapienza and Wheeler (2006) described the positive impact of EMST on the subglottal pressure necessary to control the duration of a sustained vowel. They suggested that the active involvement of expiratory muscles generated the positive airway pressure needed for tasks involving long durations of speech or loud speech. According to Putnam and Hixon (1991), increasing expiratory muscle strength can result in increased sound durations and greater sound pressure levels.

Instrumentation and Calibration Procedures

Surface EMG, kinematic, and acoustic signals were recorded simultaneously using two acquisition systems (Vetter FM data recorder and ADInstruments Power Lab) for later data analysis. Digital video recordings were made during the experiment to assist with later data analysis (i.e., elimination of data associated with extraneous movement and/or postural shifts).

Calibration procedures for respiratory kinematics following placement of the respibands involved the participant performing an isovolume manoeuvre (pulling the stomach in and letting it relax) while breath-holding, to establish the relationship between the rib cage and abdomen in a closed system (Konno & Mead, 1967 as reported in Tomczak et al., 2013). This manoeuvre was simulated by having the participant take a drink (breathing ceases during swallowing and the stomach reflexively tenses and relaxes). The participant was also asked to breathe through a fitted mouthpiece, which was attached to a bidirectional pneumotachometer (Hans Rudolph, Kansas City, MO) and differential pressure transducer (model DP45-14; Validyne Engineering, Northridge, CA) with his nasal openings obstructed with soft clips (methods described by Tomczak et al., 2013). These two calibration tasks served to convert kinematic signals to volume signals.

Acoustic measures of speech were acquired using a small lapel microphone, a boom mic and digital audio recorder. Sound pressure (dB SPL) was detected using a digital sound level meter placed at a standardized distance from the microphone (30 cm). MEP was calibrated in cmH2O against a digital pressure meter. MEP trials were conducted using a resistance device, modified to provide an infinite amount of resistance with the placement of a stopper at the level of the release valve. An additional modification of an air leak tube and pressure line connected to a transducer allowed the MEP to be measured. Maximum voluntary effort tasks were used to record the strongest muscle contraction for intercostal and oblique muscle groups.

Baseline data collection included: 1) three trials to assess MEP in accordance with the American Thoracic Society (Miller et al., 2005), 2) 60 seconds of resting tidal breathing, 3) three trials of a vital capacity manoeuvre in accordance with the American Thoracic Society (Miller et al., 2005), 4) three trials of a maximum phonation task at the participant’s perceived conversational loudness, 5) three trials of a maximum phonation task at the participant’s perceived twice conversational loudness level and 6) forced vital capacity as measured by a digital spirometer.

Following completion of baseline tasks, the participant was to take part in EMST spanning two weeks, using a calibrated expiratory resistive loading device. The initial session to learn the protocol for respiratory muscle consisted of the participant exhaling through a hand-held commercial resistive loading device (EMST 150, Aspire Products Inc.). Prior researchers involved in similar intensive respiratory muscle training protocols used resistances of 75-80% MEP. Inbar, Weiner, Azgad, Rotstein and Weinstein (2000) had healthy subjects perform inspiratory manoeuvres at upwards of 80% of maximal inspiratory pressure over a 30 minute session. Sapienza et al. (2011) used EMST devices set at 75% of participants' MEP in their study of people with PD. The current study also sought to employ an expiratory resistance that approximated 75% of MEP. However, due to the participant's inability to blow a minimum of 30 cmH2O of pressure that is required to open the valve of the device, the current study employed an EMST protocol that required the participant to perform one resistive expiratory manoeuvre approximating 100% of MEP (~30 cmH2O or less) for the duration of a complete expiratory breath, followed by a 30 to 60-second rest. This procedure was repeated five times. Following the fifth breathing manoeuvre, the participant rested for two minutes. This series of five repetitions with 30 to 60 seconds of rest between each manoeuvre was repeated five times, again with two minutes of rest between each set of five manoeuvres. The protocol required the participant to perform a total of 25 breaths and was practiced prior to the end of the baseline testing session. This protocol that was recommended by the manufacturer (EMST 150, Aspire Products Inc), has been used in previous studies (Kim, Davenport & Sapienza, 2009; Weiner, Magadle, Beckerman,Weiner & Berar-Yanay, 2003; Wheeler-Hegland, Rosenbek & Sapienza, 2008 Pitts et al., 2009), and is summarized below:

- Set 1: 1 expiratory manoeuvre at ~100% of MEP, 30 to 60-seconds rest (repeat 5 times)
- Rest: 2 minutes
- Set 2: 1 expiratory manoeuvre at ~100% of MEP, 30 to 60-seconds rest (repeat 5 times)
- Rest: 2 minutes
- Set 3: 1 expiratory manoeuvre at ~100% of MEP, 30 to 60-seconds rest (repeat 5 times)
- Rest: 2 minutes
- Set 4: 1 expiratory manoeuvre at ~100% of MEP, 30 to 60-seconds rest (repeat 5 times)
- Rest: 2 minutes
- Set 5: 1 expiratory manoeuvre at ~100% of MEP, 30 to 60-seconds rest (repeat 5 times)
- Rest: 10 minutes

All measurements were performed at baseline (pre-training) and at follow-up (post-training). "Training" was to occur twice a day over a two-week period, with the participant performing respiratory muscle exercises with expiratory resistive loading using a commercially available device manually held to the mouth. This training protocol was estimated to last approximately 30 minutes in duration, resulting in 60 minutes of training per day. This procedure was to be followed twice a day, with the target number of blows over the two-week training program being 500 blows total. During the training period, one of the researchers contacted the participant's care partner on a daily basis to monitor progress of the treatment and to ensure that the expiratory exercises were being completed. After the completion of the program, the participant returned to the lab so that post-training data could be collected. At this time, all measures (including subjective quality of life measures) from the baseline session were repeated.

Data Analysis

The participant's lung volume excursion (initiation, termination) was expressed in percentage of predicted vital capacity (Tomczak et al., 2013). Relative rib cage contribution was calculated by dividing rib cage excursion by lung volume excursion in accordance to methods used by Tomczak et al. (2013).

Maximal Expiratory Pressure. MEP was measured was measured as the average of the greatest point in a 1-s window on 3 waveforms that were within 5% of each other.

Intermuscular Coherence. Intermuscular coherence analysis has previously been described by Tomczak et al. (in review). Briefly, EMG activity was analyzed for expiration with the EMG window selected from the onset of expiration to the end of expiration. These data were then passed through a Tukey window to reduce erroneous high-frequency signals at the borders of adjoining breath trials, concatenated, and rectified. The formula used for spectral analysis is reported in Tomczak, Greidanus, and Boliek (2013). Segments of length 2048 with no overlap, were derived from a frequency resolution of 2.44 Hz and calculated offline in the MATLAB environment (MathWorks, Natick, MA) using open access scripts (www.neurospec.org).

Kinematic Analysis. After calibration, vital capacity, maximum phonation, and speech tasks were identified and the expiratory limb for each trial was digitized relative to a stable end expiratory level. Each breath group for speech was analyzed separately. Measures derived included: lung volume initiation (in mLs), lung volume termination (in mLs), lung volume excursion (in percent vital capacity), percent rib cage contribution to lung volume excursion, expiratory duration (in sec), and average expiratory flow (mLs/sec).

Quality of Life Questionnaires

Four questionnaires were completed by the participant with familial assistance pre- and post-EMST. The MD Anderson Dysphagia Inventory (MDADI) and SWAL-QOL were subjective questionnaires related to swallowing and quality of life filled out by the participant. The MDADI is a measure of swallowing-related quality of life that includes physical, emotional, functional, and global subscales regarding the impact of dysphagia (Chen et al., 2001). Like the MDADI, the SWAL-QOL looks at the patient perspective regarding quality of life and quality of care (McHorney et al., 2002). The Speech Handicap Index (SHI) (Dwivedi et al., 2010) has been used to evaluate changes in speech and the effects of speech on participants' daily activities. The VHI was created to measure the impact of disordered voice on quality of life and daily functioning (Jacobson et al., 1997).

RESULTS

EMST Protocol Adherence

The participant did not strictly adhere to the EMST protocol. Reasons for infidelity are discussed below. The inconsistency of practice ultimately lead to the protocol being less intensive than was originally designed: to reach the criteria of 500 blows, the training period was extended from 2 weeks to 4 weeks as shown in the following graphs.

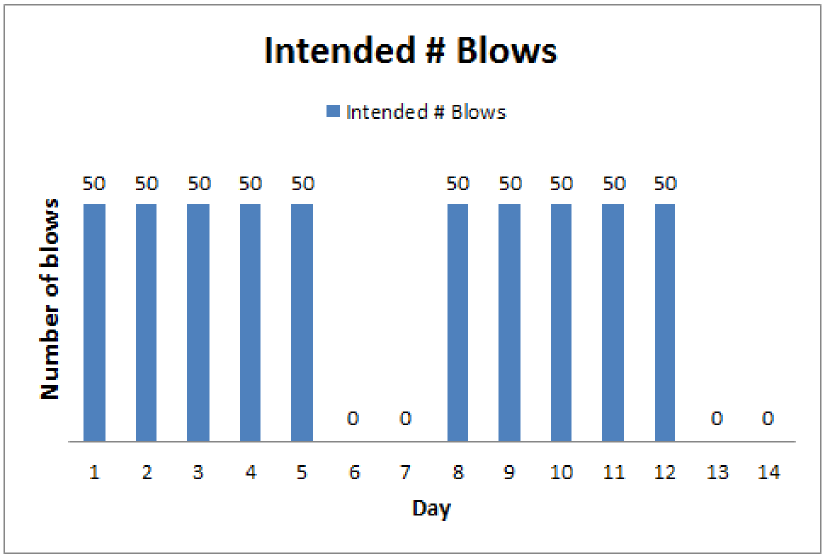

Figure 1. Intended Number of Blows during 14-Day Training Period

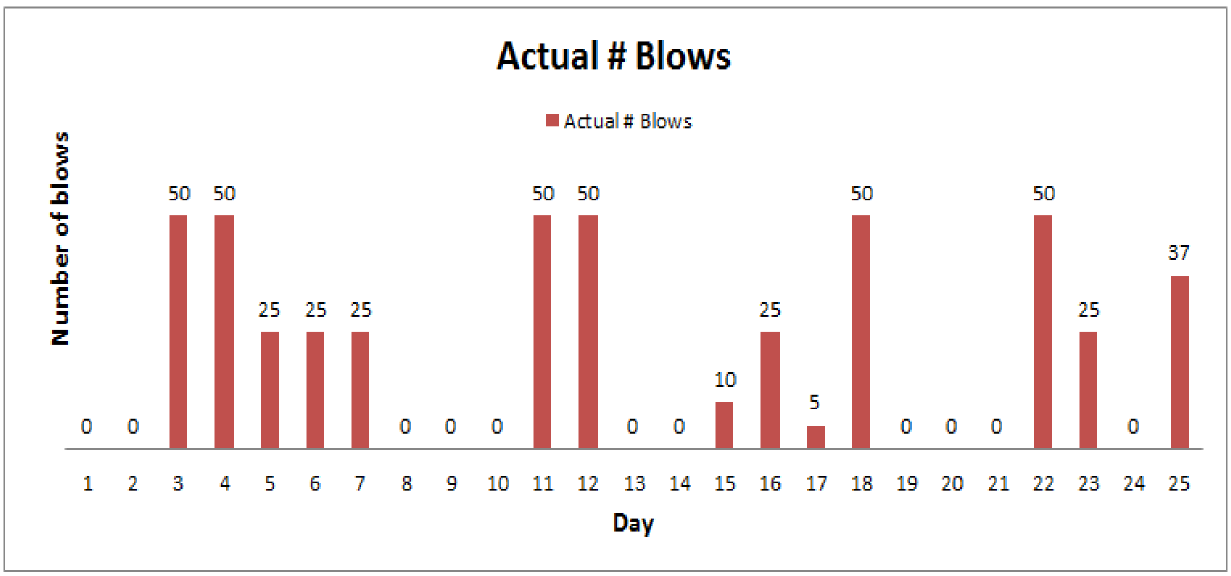

Figure 2. Actual Number of Blows Completed During Extended 25-Day Training Period

Maximal Expiratory Pressure

Maximal expiratory pressure was measured in cmH2O. *Figure 3* outlines the changes in MEP following EMST.

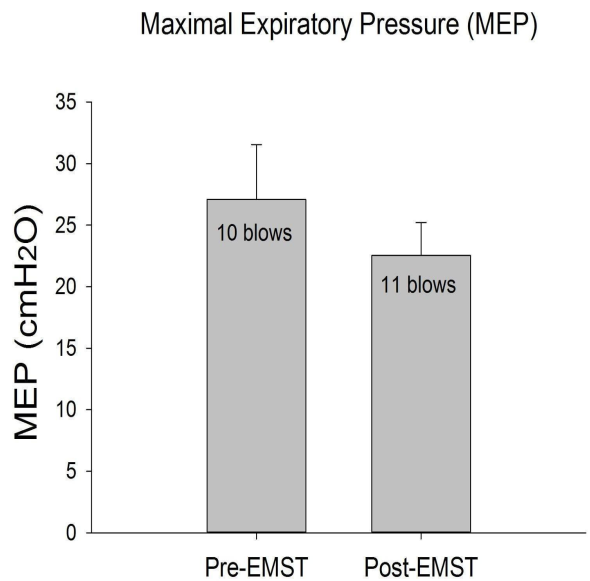

Figure 3. Maximal Expiratory Pressure (MEP), Pre and Post-EMST

MEP decreased slightly from an average of 26 cmH2O prior to training to an average of 23 cmH2O after training *(Figure 3)*.

Intermuscular Coherence

Intermuscular coherence between intercostal and oblique muscles were measured pre- and post-training. *Figure 4* illustrates coherence changes found between pre- and post- measures on each task.

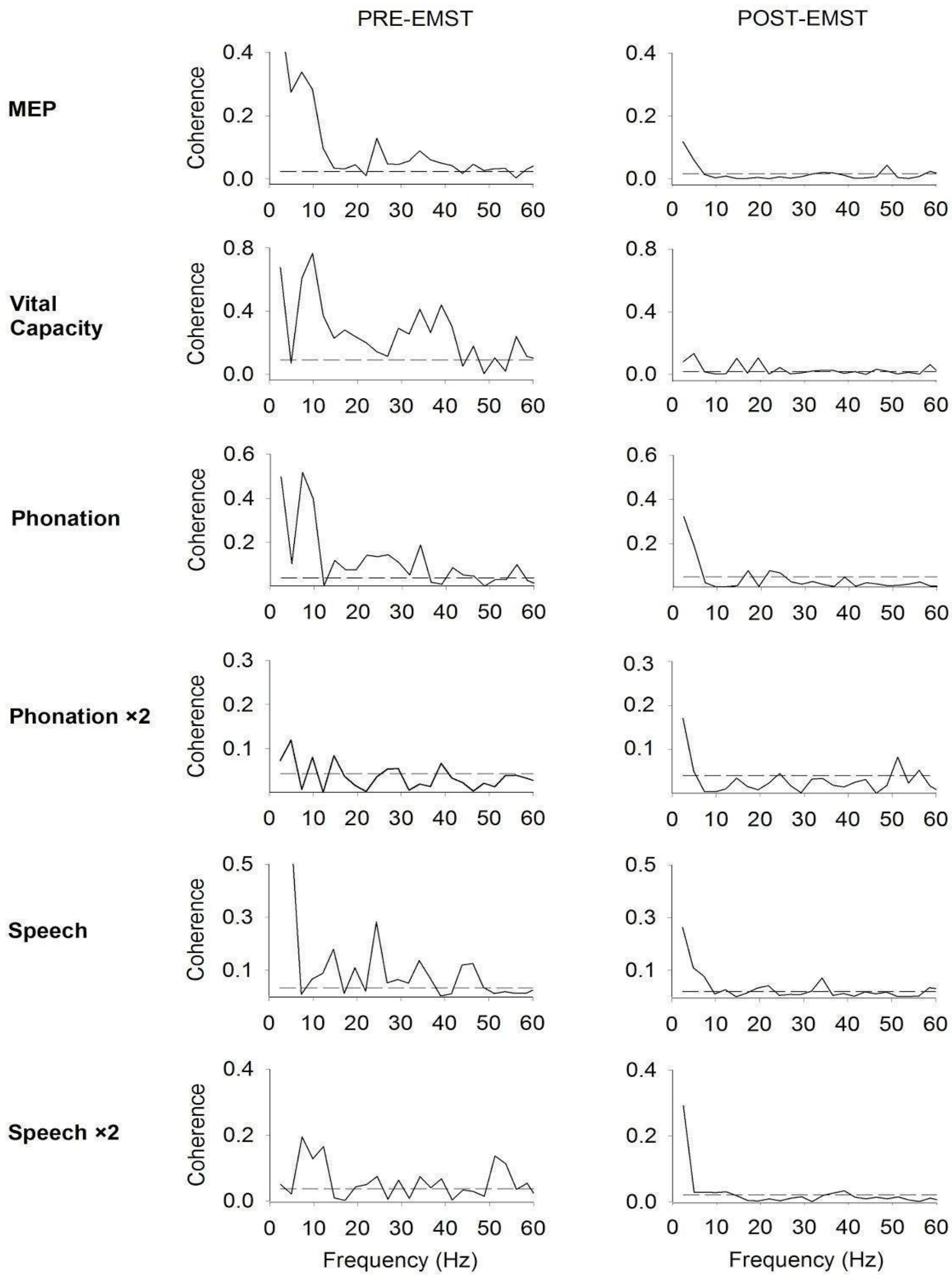

Figure 4. Intermuscular Coherence Across Speech Tasks, Pre and Post EMST

Coherence decreased overall for all tasks post-training. During pre-measures, peaks occur in the beta range (20-40Hz) indicating significant coherence in this range. Significant coherence peaks also occurred in the alpha range (0-15Hz) pre-training. For all tasks, significant peaks were not observed and coherence was minimal post EMST.

Phonation at Conversational Loudness and Phonation at Twice Conversational Loudness

Kinematic measures included 1) expiratory durations for all tasks (maximal expiratory pressure and maximal phonation tasks), 2) average expiratory flow, 3) lung volume initiation, termination, and excursion, and 4) percent rib cage contribution to lung volume excursion. The following figures outline kinematic changes found between pre and post measures. A description of results is provided after each set of figures.

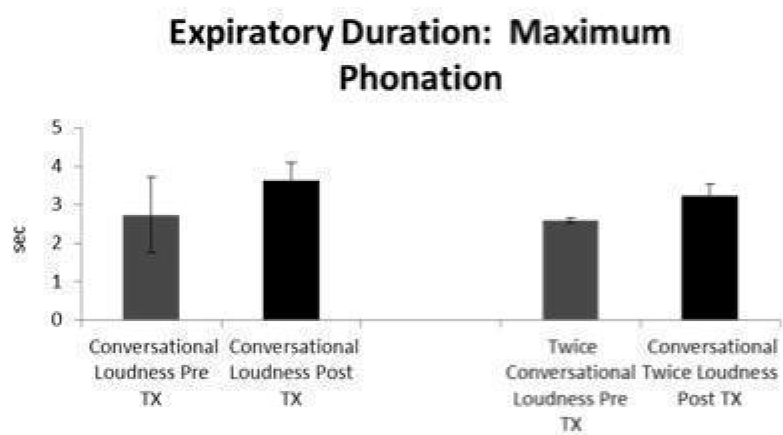

Figure 5. Expiratory Duration: Maximum Phonation

Expiratory Duration. Average expiratory duration of maximum phonation at conversational loudness increased slightly from 2.7 seconds pre-training to 3.6 seconds post training (*Figure 5).* Average duration of maximum phonation at twice conversational loudness increased slightly from 2.6 seconds to 3.2 seconds post training.

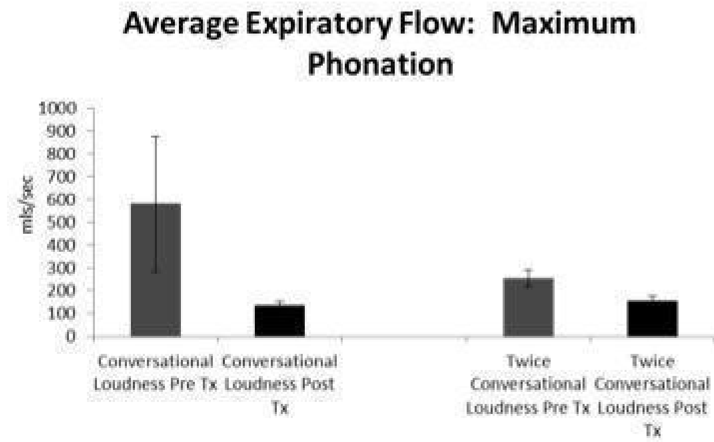

Figure 6. Average Expiratory Flow: Maximum Phonation

Average Expiratory Flow. The average measures for expiratory flow (mL/sec) decreased from 581 mL/sec pre-training to 134 mL/sec post-training for maximum phonation at conversational loudness and decreased from 212 mL/sec to 152 mL/sec for maximum phonation at twice conversational loudness *(Figure 6).*

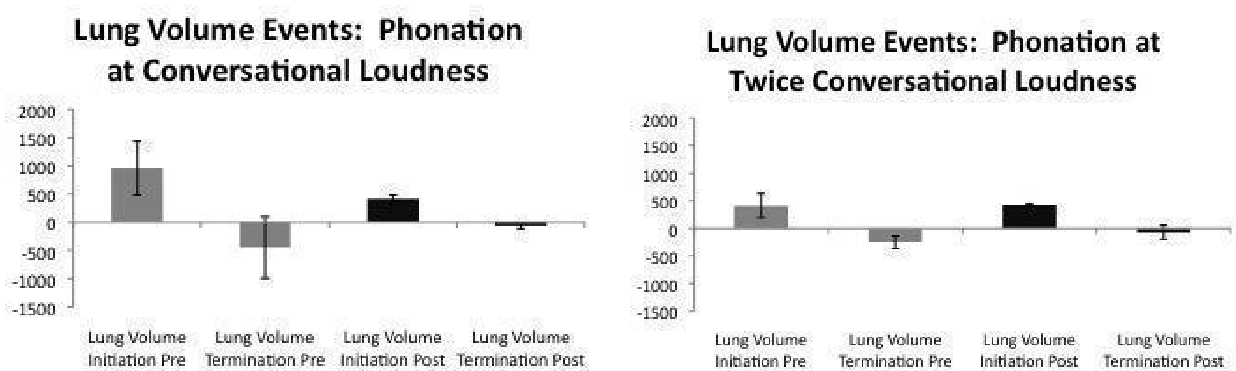

Figure 7. Lung Volume Events: Phonation Tasks

Lung Volume Events. Phonation at conversational loudness showed decreased lung volume excursion post EMST *(Figure 7).* Phonation at twice conversational loudness showed decreased lung volume termination post EMST, however the overall change in lung volume excursion post EMST was small *(Figure 7).*

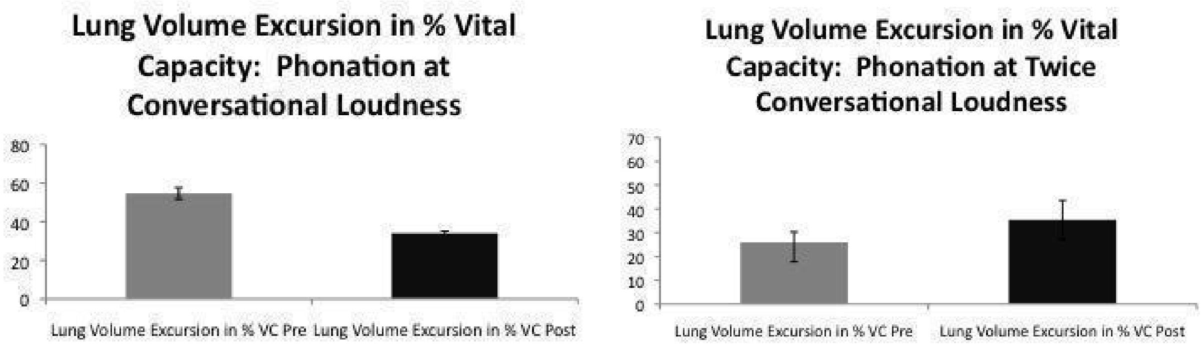

Figure 8. Lung Volume Excursion in % Vital Capacity: Phonation Tasks

Lung Volume Excursion. Overall lung volume excursion (percent vital capacity) decreased post-training for the phonation at conversational loudness task *(Figure 8),* meaning that the participant never used his full vital capacity, causing his expiratory lung volume to be smaller after training than it was before training. The participant was unable to use his full vital capacity during any observed maximal phonation trials. For the phonation at twice conversational loudness tasks, lung volume excursion increased by approximately 10% *(Figure 8)*, a shift towards what would be expected for a healthy adult.

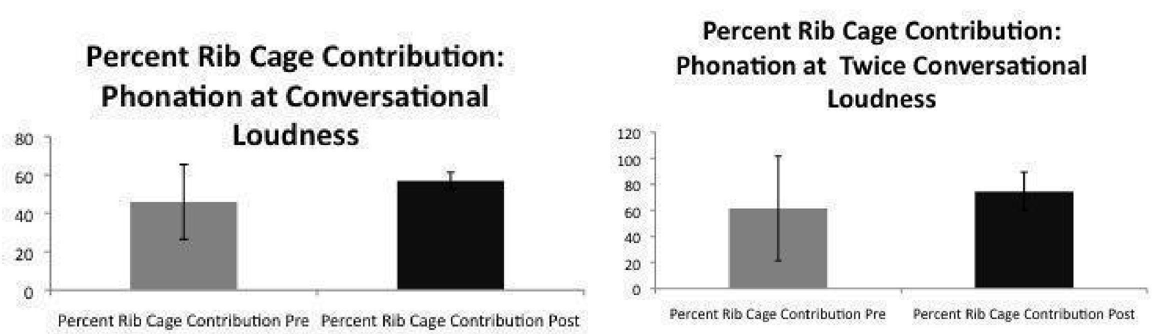

Figure 9. Percent Rib Cage Contribution: Phonation Tasks

Percent Rib Cage Contribution. The percent rib cage contribution for phonation at conversational loudness and at twice conversational loudness *(Figure 9)* increased after EMST. Before EMST, rib cage contribution to lung volume excursion was variable and lower than that typically found in healthy adults (80%) for both conversational loudness and twice conversational loudness phonation tasks. However after EMST, percent rib contributions to lung volume excursion increased and were less variable. For phonation at conversational loudness, rib cage contribution was approximately 65% post EMST. For phonation at twice conversational loudness, rib cage contribution was approximately 80% post EMST.

Speech at Conversational Loudness and Speech at Twice Conversational Loudness

Kinematic measures included 1) expiratory durations for speech tasks (speech breath groups), 2) average expiratory flow, 3) lung volume initiation, termination, and excursion, and 4) percent rib cage contribution to lung volume excursion. The following figures outline kinematic changes found between pre and post measures. A description of results is provided after each set of figures.

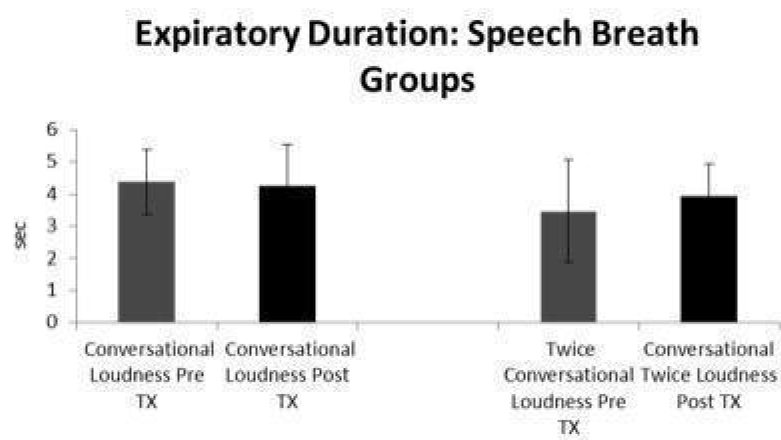

Figure 10. Expiratory Duration: Speech Breath Groups

Expiratory Duration. The participant's expiratory duration of speech at conversational loudness did not change significantly (4.4 sec to 4.3 sec) from pre to post-training *(Figure 10)*. Expiratory duration at twice conversational loudness increased slightly from 3.5 seconds to 4.0 seconds after EMST *(Figure 10).*

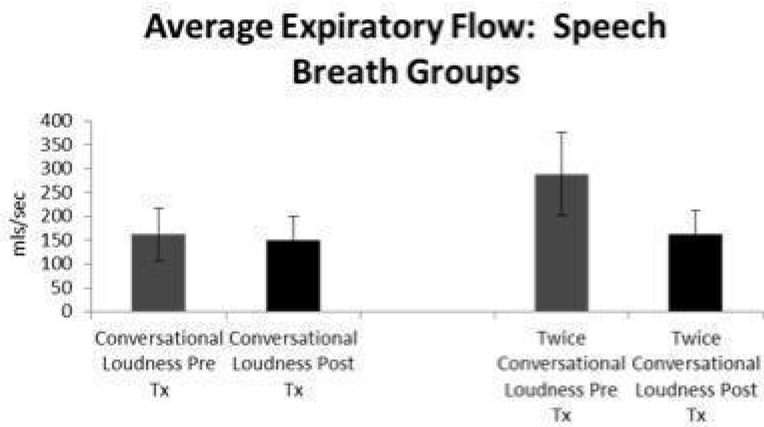

Figure 11. Average Expiratory Flow: Speech Breath Groups

Average Expiratory Flow. For speech at conversational loudness a decrease in average expiratory flow from 162 mL/second to 147 mL/second after training was observed *(Figure 11)*. For speech at twice conversational loudness, a substantial decrease in expiratory flow from an average of 289 mL/second to 155 mL/second after EMST training was observed *(Figure 11)*.

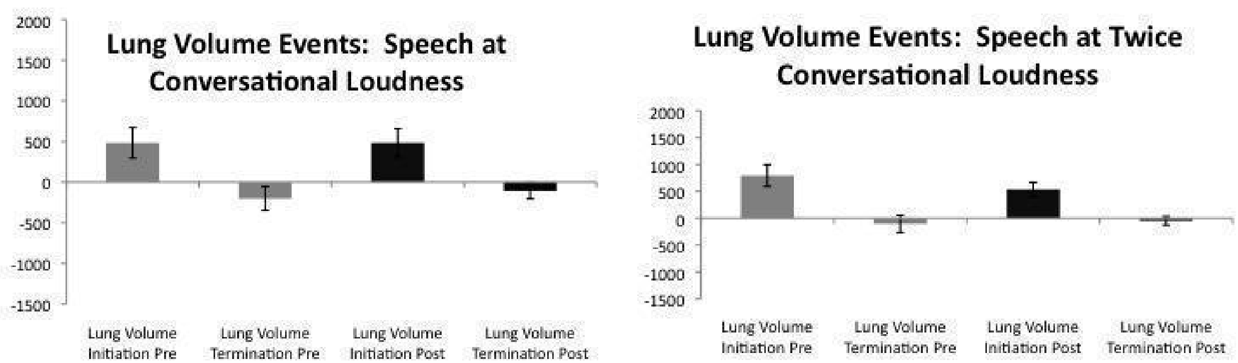

Figure 12. Lung Volume Events: Speech Tasks

Lung Volume Events. At both conversational and twice conversational loudness levels, there was no significant change in lung volume initiation or termination after training *(Figure 12)*.

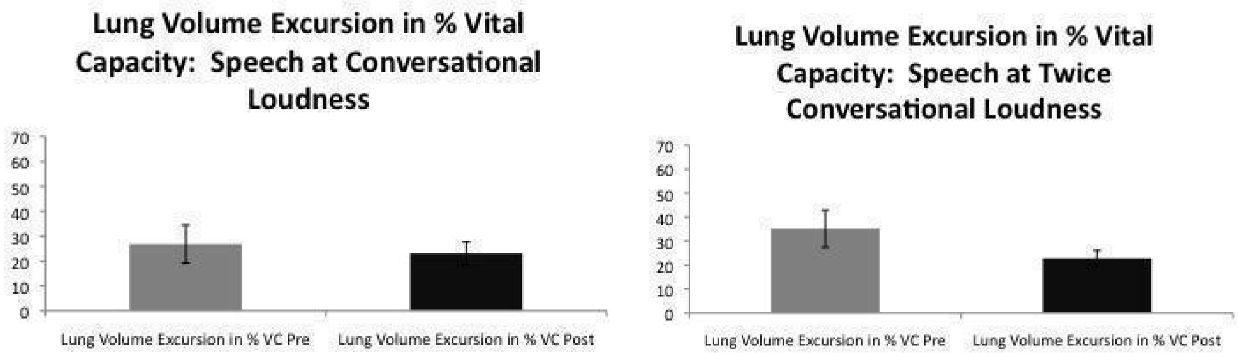

Figure 13. Lung Volume Excursion in % Vital Capacity: Speech Tasks

Lung Volume Excursion. Lung volume excursion decreased from 25% vital capacity pre-training, to 23% vital capacity post-training for speech at conversational loudness (*Figure 13)*. This was not a significant change, however, his lung volume excursion was comparable to a healthy adult's both pre and post-training. At twice conversational loudness, lung volume excursion decreased from 35% vital capacity pre-training, to 20% vital capacity post-training *(Figure 13)*. The amount of variability in lung volume excursion decreased on speech tasks at both conversational and twice conversational loudness, post-training.

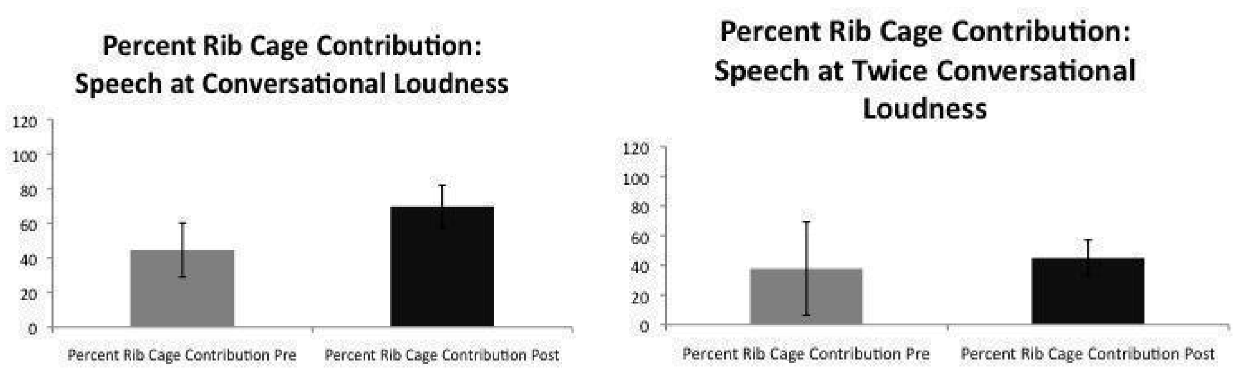

Figure 14. Percent Rib Cage Contribution: Speech Tasks

Percent Rib Cage Contribution. The participant showed an increase in rib cage contribution, from 45% pre-training, to 70% after completion of the training protocol at conversational loudness *(Figure 14)*. On the speech at twice conversational loudness level task, there was no significant change in the participant's percent rib cage contribution post- EMST training (*Figure 14*). Overall, the amount of variability for percent rib cage contribution decreased following EMST, at both conversational and twice conversational loudness.

Acoustics

Acoustic measures during phonation (i.e. sustained "ah") and speech tasks were recorded in dB SPL. Table 1 outlines changes found between pre and post measures of phonation and speech tasks.

Table 1
Maximum and Average Loudness Levels Across Speech Tasks, Pre- and Post- EMST Training

	PRE	**PRE**	**POST**	**POST**
	Avg. dB SPL	**Max. dB SPL**	**Avg. dB SPL**	**Max. dB SPL**
Phonation x1	83.1	91	88.1	93.7
Phonation x2	90.4	103	97.9	99.3
Speech x1 (phrases)	64.3	75	75	88
Speech x2 (phrases)	74	72	79.6	103

Table 1 *Loudness Levels Across Speech Tasks, Pre and Post-EMST*

For phonation at conversational loudness level, the maximum dB SPL increased after EMST. A decrease in maximum dB SPL was observed during phonation at twice conversational loudness. Maximum dB SPL for speech tasks at both loudness levels increased following EMST. Finally, average dB SPL for both phonation tasks and spoken phrases at conversation and twice conversation loudness levels increased following EMST.

Quality of Life

Changes in quality of life related to, voice, speech and swallowing abilities were indicated by the results of the Voice Handicap Index (VHI), Speech Handicap Index (SHI), MD Anderson Dysphagia Inventory (MDADI), and the SWAL-QOL. Due to the fact that some questions were left unanswered prior to treatment, percentages were used to detect clinically significant changes.

The questions in the VHI are representative of three aspects of voice disorders: physical, emotional, and functional. Physical items represent self-perceptions of voice production; emotional items represent affective responses to voice difficulties; and functional items represent the impact that voice difficulties may have on completing daily activities. Based on the structure of the test, higher scores indicate that specific aspects of voice have more negative impacts on quality of life. The following table outlines changes found between pre and post measures of voice-related quality of life.

	PRE			POST		
	Score	Maximum	%	Score	Maximum	%
Physical	17	36*	47.2	19	40	47.5
Emotional	14	40	35.0	8	40	20
Functional	23	40	57.5	13	40	32.5
TOTAL	54	116	46.6	40	120	33.3

Table 2 *VHI Scores, Pre- and Post-EMST Treatment*

*A question in the Physical component (P18) was not completed in the pre-treatment measure.

VHI Scores. The decrease in scores suggested that the participant's voice-related quality of life improved over four weeks of treatment. The greatest change was found in the functional domain.

Like the VHI, the SHI is structured so that higher scores imply a greater difficulty or negative impact on speech-related quality of life. The following table outlines changes found between pre- and post- measures.

	PRE			POST		
	Score	Maximum	%	Score	Maximum	%
TOTAL	83	112*	74.1	50	120	41.6

Table 3 *SHI Scores, Pre- and Post-EMST*

*Two questions (#18 and #23) were not completed in the pre-treatment measure.

SHI Scores. The decrease in the participant's score indicated an improvement in speech-related quality of life, overall.

MDADI Scores. The results for the MDADI are subdivided into physical, emotional, and functional components, with an additional question addressing global perception of swallowing abilities. The MDADI is structured so that higher scores indicate higher functioning. The following table outlines changes found between pre and post measures of swallowing-related quality of life.

	PRE			POST		
	Score	Maximum	%	Score	Maximum	%
Physical	24	40	60.0	30	40	75.0
Emotional	16	25*	64.0	18	30	60.0
Functional	16	25	64.0	19	25	76.0
Global	4	5	80.0	2	5	40.0

TOTAL	60	95	63.2	69	100	69.0

Table 4 *MDADI Scores, Pre- and Post-EMST*

*A question in the Emotional category (E3) was not completed in the pre-treatment measure.

The participant's results indicated an increase in the physical and functional components of swallowing-related quality of life. The global rating, however, decreased significantly.

SWAL-QOL Scores. Like the MDADI, higher scores on the SWAL-QOL also suggest greater swallowing-related quality of life. The following table outlines changes found between pre and post measures.

	PRE			POST		
	Score	Maximum	%	Score	Maximum	%
TOTAL	115	160*	71.9	182	225	80.9

Table 5 *SWAL-QOL Scores, Pre- and Post-EMST*

*Questions 2 and 4, as well as components of questions 3 and 9, were not answered in the pre-treatment measure.

The participant's results suggested an overall increase in swallowing-related quality of life.

DISCUSSION

The main finding from the present case study was that EMST did not alter intermuscular coherence in the direction hypothesised. The lack of detectable intermuscular coherence might infer that a more distributed drive to the respiratory muscles resulted from training. Evidence from other outcomes indicated that perhaps training had a positive effect on the coordination between the respiratory and laryngeal subsystems. Other variables such as respiratory kinematics and quality of life were favourably altered in the post training assessment. As a general observation, variability in performance appeared to be decreased post-training across many of the tasks. This suggests that EMST may have led to an increase in more consistent coordinative skill between the respiratory and laryngeal systems, not simply their strength.

The primary variable expected to change, MEP, did not respond as expected to the training. We anticipated MEP to increase following EMST, but instead observed no change in post EMST MEP values. The slight decrease in MEP is not likely to be clinically meaningful given the small change. Notably, the MEP findings do not suggest a favourable improvement in chest wall pressure generation ability occurred. In terms of baseline and post-EMST testing, we believe that the subject did perform his "best effort" trials with the EMST. Therefore, it is unlikely that a lack of change in MEP was due to measurement or technique error. Indeed, testing protocols in our laboratory were optimized, practiced, and repeated to ensure "best effort" trials were included in the analyses. From a measurement error perspective, we performed a calibration on our pressure transducer for determining MEP. As well, the participant had a more severe diagnosis than that which was originally intended to be tested using this protocol. He did not meet the original exclusionary criteria of mild or moderate motor impairment, but was selected due to convenience of sample. Because the participant had a more severe CP, the physical demands of the task may not have been appropriate for his abilities. As noted above, a minimum pressure of 30cmH2O was required to open the release valve on the EMST device. To generate this pressure, one must have sufficient respiratory strength and coordination. The participant did not have the strength required to consistently generate this pressure; at the lowest possible setting, he was only able to open the EMST device valve once. This may have affected training as participants are intended to practice with the device set to open at 75% of the maximum pressure they are able to generate. If a participant is unable to open the valve at its lowest setting, then the participant will blow at an effective 100% MEP. In this respect, the pressure generated by the participant, and the required exertion or force to generate that pressure, may have changed from one practice session to the next, as there was nothing to signal to the participant that he had reached his target.

Prior to training, the participant displayed intermuscular coherence in the alpha (0-15 Hz) and beta (20-40 Hz) ranges during both the sustained phonation and speech at conversational loudness tasks *(Figure 4)*. Following EMST training, no significant coherence was detected. This may be due to the skilled nature of respiratory and laryngeal functioning. Semmler and Nordstrom (1998) found intermuscular coherence in strength trained individuals, but none for skill-trained individuals, on a finger abduction task. The results of their study indicated that skill-training may employ a more distributed activation pattern of cortical pathways, whereas strength-training may activate more specific pathways via common cortical drive, resulting in coherence (Semmler & Nordstrom, 1998). According to Semmler et al. (2004), highly skilled tasks require less coherence than strength-based tasks. A decrease in coherence therefore can be due to an increase in coordinative skill in the speech system, namely the laryngeal adjustment resulting in increased loudness and increases in percent rib cage contribution during phonation and speech tasks (Semmler et al., 2004).

One interesting result was that the participant was able to generate an overall louder speech signal in the post-training session, contrary to the fact that intermuscular coherence decreased. This was unexpected for two reasons. Firstly, one would anticipate that with greater practice, coherence of firing between muscle groups would increase. Secondly, it appears contradictory to have vocal loudness increase as coherence between muscles decreases. However, this may be explained by considering the overall efficiency and coordination of the speech subsystems including changes in rib cage contribution and possibly laryngeal adjustments, which in combination may represent a healthy biomechanical adjustment.

The percent rib cage contribution for phonation at conversational loudness and twice conversational loudness *(Figure 9)* increased post-training. Before EMST, rib cage contribution to lung volume excursion was variable and lower than that typically found in healthy adults (80%). After EMST, percent rib cage contributions to lung volume excursion increased and were less variable. However, average and maximum loudness (dB SPL) both increased (Table 1). These results indicate an increase in coordination and stability of the respiratory system for sustained phonation. According to Hixon et al. (2008), healthy adults use approximately 20% of their vital capacity during speech. The participant showed a significant increase in rib cage contribution, from 45% pre-training, to 70% after completion of the training protocol *(Figure 9)*. This shows an increase toward 80%, which is the percent rib cage contribution expected for a healthy adult (Hixon et al., 2008). Therefore, biomechanically, the participant was using a more efficient mechanism for speech breathing post-training than pre-training.

On the speech at twice conversational loudness level task, there was no significant change in the participant's percent rib cage contribution post- EMST training (*Figure 20*). The researchers observed a substantial decrease in expiratory flow during speech tasks at both loudness levels. At twice conversational loudness, the participant's average expiratory duration increased from 3.5 seconds to 4.0 seconds *(Figure 10)*. These findings suggest that the participant was not losing air as quickly during the task and therefore was able to produce speech for a longer duration.

Because the participant did not increase the size of breath he took before phonating (no change in lung volume initiation or termination) from pre-training to post-training, these findings appear to lend support to the hypothesis that he made a laryngeal adjustment, such as better vocal fold closure, in order to increase his loudness. As the coordination between speech subsystems (respiratory and laryngeal) improves and efficiency increases, less effort is required to generate a stronger speech signal. Therefore, an increase in loudness may be observed even with decreased intermuscular coherence, because the respiratory and laryngeal subsystems are operating with better coordination and efficiency.

Changes in quality of life related to swallowing abilities, voice, and speech were indicated by the results of the four quality of life questionnaires completed by the participant (with assistance from family members). The greatest perceived improvement was in speech-related quality of life, as indicated by a significant decrease in total score on the Speech Handicap Index. Notable perceptual changes related to the participant's speech included: *never* running out of air when speaking (Pre: Almost always), *never* feeling incompetent due to speech (Pre: Almost always), *never* feeling ashamed of a speech problem (Pre: Almost always), and only *sometimes* being asked to repeat what was said (Pre: Always). These changes may be related to the hypothesized improvement in coordination between the respiratory and laryngeal subsystems. As suggested by increased loudness and decreased intermuscular coherence post-training, EMST may have led to less effortful (e.g., no shortness of breath) speech production. As a result, perceived intelligibility and functionality of the participant's speech increased. Overall, the participant's responses post-treatment reflected a significant improvement in quality of life related to speech, particularly regarding avoidance behaviours, and feelings of embarrassment or shame.

The results of the VHI revealed a positive change in voice-related quality of life based on a total score decrease of approximately 13.3%. Although minimal improvement occurred in the physical domain of voice perception, dramatic improvements (i.e., 15%) occurred in the functional and emotional domains. Significant changes in the questionnaire included: people *never* experiencing difficulty understanding the participant in a noisy room (Pre: Almost always), *never* experiencing tension when speaking with others (Pre: Almost always), and people *almost never* experiencing difficulty hearing the participant (Pre: Almost always). Changes demonstrated during phonation tasks reflected changes in the functional domain of voice-related quality of life stated above. Specifically, an increase of laryngeal use resulting in increased subglottal pressure led to significant increases in loudness during conversational and maximal loudness tasks. The physical improvements likely generalized to daily functional tasks (such as people being able to hear the participant speak). Overall, the participant's responses on the VHI post-treatment reflected a significant improvement in quality of life related to emotional and functional aspects of voice.

The questionnaires related to swallowing-related quality of life (i.e., MDADI and SWAL-QOL) both demonstrated a moderate improvement in the participant's perceived swallowing function. The results of the MDADI indicated an improvement of 15% and 12% in the physical and functional domains, respectively. However, the emotional domain experienced a slight decrease of approximately 4%. Additionally, the participant's response to the statement related to global perception of swallowing function (i.e., "My swallowing ability limits my day-to-day activities") was "Disagree" (score of 4) pre-treatment, and "Agree" (score of 2) post-treatment. The inconsistencies in the results suggested an increased awareness of swallowing abilities and difficulties that occurred during the research process. Increased awareness may have resulted in feelings of embarrassment. Emotion-related responses that reflected this included: “I do not feel self-conscious when I eat” (Pre: Agree; Post: Disagree), and “Other people are irritated by my eating problem” (Pre: No response; Post: Agree). Despite this, improvements in functional and physical domains suggested that the EMST treatment protocol positively impacted the participants quality of life related to swallowing.

Like the MDADI, the results of the SWAL-QOL demonstrated a moderate improvement in overall swallow-related quality of life with a score increase of approximately 10.8%. Notable changes in responses post-EMST included: *never* being discouraged by swallowing problem (Pre: Sometimes), *always* being frustrated with swallowing problem (Pre: Sometimes), *strongly disagreeing* that swallowing problems cause difficulties during social gatherings (Pre: Disagree), *hardly ever* choking on liquids (Pre: Sometimes), and only *sometimes* gagging (Pre: Often). The responses indicated that although the participant had perceived moderate improvements in swallowing abilities, emotional components (i.e., discouragement or frustration) were negatively impacted. As previously stated, this may be related to increased awareness of swallowing difficulties. In contrast, physical components and functional components related to social gatherings and leisure activities showed an overall improvement. Perceived reduction in choking and gagging may be related to increased efficiency of speech subsystems discussed above. Increased laryngeal adduction may have resulted in a more effective protective mechanism during swallowing. EMST training may have also improved velopharyngeal closure and hyolaryngeal displacement as found in previous studies (Pitts et al., 2011; Kim & Sapienza, 2005). Therefore, both questionnaires revealed that the participant’s quality of life related to swallowing improved in regards to physical and functional domains, and reduced slightly in the emotional domain.

Despite positive changes regarding the participant's quality of life related to speech, voice, and swallowing abilities, there were potential confounds that may have impacted the answers to the questionnaires. Firstly, the participant's mother assisted with completing all four questionnaires pre-treatment, and only the MDADI post-treatment. The participant's father assisted with completing the VHI, SHI, and SWAL-QOL post-treatment. It is important to note, however, that all tests saw a minimal total change of 10%, including the MDADI (completed with the mother's assistance pre- and post-EMST). Both swallowing questionnaires resulted in similar subjective and objective changes post-treatment, suggesting that the participant's responses were not significantly impacted by the person assisting with test completion. A second factor was timing of questionnaire completion; specifically, the pre-treatment questionnaires were completed prior to the first lab visit, whereas the post-treatment questionnaires were completed following the final lab visit. Changes in the participant's demeanour may have caused unrepresentative changes in quality of life scores. For instance, feelings of relief or fatigue following the second lab visit may have impacted the participant's responses. Thirdly, not all questions were answered in the pre-treatment questionnaires, resulting in each question having a greater impact on overall percentage. Therefore, precautions must be taken when interpreting the data.

Research of the effects of intensive EMST training in populations with speech and swallowing disorders is understudied. The present study is the first to apply an EMST protocol to an adult with CP, with fairly robust positive effects. However, there is much still to be learned and discovered. Future studies of the effects of EMST in the population of CP are warranted, especially studies to gather data of the effects of EMST on individuals with a less severe presentation of cerebral palsy. Such studies should include measures of corticomuscular coherence, strive to recruit greater numbers of participants, investigate the effects of EMST in persons with less severe CP (mild-moderate), and increase the intensity with which the protocol is performed. Future studies should include the measure of corticomuscular coherence so that changes in the neuroplasticity, or neural control over targeted muscles, can be measured. With a greater number of participants, researchers can be more confident in generalizing the results to the wider population of people with CP. As this was a case study, very little can be said about how this protocol may alter kinematic, coherence, or speech measures in other individuals with CP. One of the challenges of this study was that the participant did not have the respiratory strength required to open the valve on the EMST device. This led to uncertainty about how much pressure the participant generated during at home training sessions. Participants with a mild-moderate diagnosis of cerebral palsy would be better able to generate the pressure required to open the valve, and would therefore have objective measures of their success in obtaining that level of pressure as they train. Alternatively, employing an EMST device that operates at a lower pressure range (below 30 cmH2O) should be considered. In additionally, participants with a milder variant of CP are more likely to be able to perform training sessions independently, leading to fewer challenges in having care aids that are familiar and comfortable with the device and procedure. This, in turn, may lead to a more regular training schedule, ideally reaching an intensity of two practice sessions per day, resulting in 500 blows over the course of two weeks. This increase in intensity is important to facilitate greater neuroplasticity. With these changes implemented, future research can increase our understanding of the benefits of EMST training within this population.

Whereas there was no detectible change in MEP, the effects of training were observed in intermuscular coherence, respiratory kinematics, measures of vocal dB SPL, and quality of life outcomes. These results indicate the robustness of the training even when training was not carried out exactly to the prescribed protocol as described below.

Contributors to inconsistency of practice included: (1) the participant becoming ill after the protocol had commenced, resulting in several days of rest; (2) change in the participant's schedule on evenings and weekends; (3) participant's inability to perform training sessions independently, relying on others to assist in holding the device to his mouth, as well as holding his lips to form a proper seal; and (4) change in caregiver aids from shift to shift, and their differing familiarity with the device and procedure.

CONCLUSION

The current case study explored the impacts of EMST on an individual with severe CP. The results suggested that the participant's intermuscular coherence did not increase, as was hypothesized. However, hypotheses regarding improvements in speech as indicated by acoustic and respiratory kinematic measures were confirmed via increased loudness levels. Researchers speculated that increased loudness combined with decreased expiratory flow might have been due to increased efficiency of speech subsystems, particularly related to laryngeal adduction. Subjective improvements in quality of life related to voice, speech, and swallowing abilities were also found in the areas of daily functioning and participation. Overall, although the EMST protocol had variable results for this participant, there was evidence of improvement relative to pre-training, and that additional research with adults with CP is warranted. The findings from this case study contribute to evidence dictated by academic literature, further demonstrating the potential use of EMST as a tool to improve speech, voice, and swallowing function.

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