January 2012

Inspiratory Breathing Exercises for Vocal Tremor: A Preliminary Study

Jessica Tayseer Hilo

University of South Florida, hilo.jessica@gmail.com

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Inspiratory Breathing Exercises for Vocal Tremor:
A Preliminary Study

by

Jessica Tayseer Hilo

A thesis submitted in partial fulfillment of the requirements for the degree of
Master of Science in Speech-Language Pathology
Department of Communication Sciences and Disorders
College of Behavioral and Community Sciences
University of South Florida

Major Professor: Ruth Huntley Bahr, Ph.D.
Bonnie Smith, Ph.D.
Darla Freeman LeVay, M.A.

Date of Approval:
July 3, 2012

Keywords: essential tremor, neurologic voice disorders, spirometer, breathing exercises, laryngeal lowering

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Dedication

I would like to dedicate this thesis to my amazing and ever supportive family. Mama and Baba, I can never thank you enough for your love and the sacrifices you’ve made for me throughout my life. You have been my foundation and constant encouragement. Josh, Rammi, and Lizza, thank you for your love, support, and help over the past two years. I love you all and wouldn’t have made it without you guys.
Acknowledgements

I thank the following individuals for their assistance in the completion of this study. Dr. Ruth Bahr, my thesis advisor, for your constant support, encouragement, and motivation in completing this study. Your input of knowledge over the past two years has been invaluable to my understanding of the implications of this study and in the overall field of voice. Dr. Bonnie Smith, discussions with you and your constructive contributions to this study were very insightful and guided my outlook on the research. Ms. Freeman-LeVay, your confidence in me and input, not only in this study but also in my clinical graduate experience, has built my character and inspired me as a clinician and researcher. Dr. Marion Ridley for providing me opportunities to work with patients affected by EVT. Dr. Diehl for your assistance with gathering listener participants for this study. Ms. Kyna Betancourt for your constant encouragement, your wonderful heart, and your immense help with data programming. Ms. Vivian Maldonado for simply being an amazing support and encouragement throughout the thesis process. I am honored to have worked with you all.
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Abstract

Essential voice tremor (EVT) is a voice disorder that results from dyscoordination within the laryngeal musculature, which negatively impacts the symmetrical motion of the vocal folds. Several investigators have shown that individuals with EVT experience difficulty speaking and a reduced quality of life (QOL; Cohen, Dupont, & Courey, 2006; Verdonck-de Leeuw & Mahieu, 2004). While traditional voice therapy has been ineffective in lessening the severity of vocal tremor, a current approach (Barkmeier-Kraemer, Lato, & Wiley, 2011) designed to lessen the perception of vocal tremor has resulted in reported patient satisfaction with little actual change in voice quality. The present study focused on achieving positive voice changes by targeting the physiological aspects of voice production that may be altered through inspiratory breathing techniques, i.e., increased lung volume pressure and laryngeal lowering. The hypothesis was that such changes could result in reductions in vocal tremor and lead to perceived improvements in voice quality and concomitant increases in the participant’s QOL.

An ABAB (treatment reversal) single subject design was used to assess the effectiveness of inspiratory breathing exercises on reducing the severity of tremor in three women diagnosed with EVT. Pre-treatment measures were administered, and participant progress was determined after one week of treatment (post-treatment #1), a week of no treatment, and another week of treatment (post-treatment #2). The following measures were gathered from each participant to document treatment progress and effectiveness:
lung pressure volume levels, Voice Handicap Index (VHI) ratings, acoustic analyses of isolated vowels, and perceptual ratings on the Consensus Auditory Perceptual Evaluation of Voice (CAPE-V), as well as untrained listener ratings of vocal steadiness and pleasantness.

Physiological, QOL, acoustic, and perceptual data did not triangulate to demonstrate treatment effectiveness. However, individual treatment effects were found in increases in lung pressure volume for participant 1, decreases in CAPE-V scores for Participant 3, and decreases in VHI scores for Participants 1 and 3. Changes in voice acoustics and untrained listener perceptions were negligible. Thus, the results from this study indicate that inspiratory breathing exercises may show some promise in improving voice and QOL in certain tremor patients and that this technique warrants further research consideration.
Chapter 1

Introduction

Vocal Tremor Defined

Essential voice tremor (EVT) is a neurologically-based voice disorder “characterized by involuntary rhythmic modulations of pitch and loudness,” occurring at a frequency of 4-6 Hz (Barkmeier-Kraemer, Lato, & Wiley, 2011, p. 43; Sapienza & Ruddy, 2009). Others have further described the tremor as action-induced, “quavering” or irregular voice breaks occurring during speech production (Aronson & Bless, 2009; Barkmeier-Kraemer et al., 2011). Vocal tremor occurring specifically upon phonation may also occur due to dystonic tremor (Deuschl, Bain, Brin, & ad hoc Scientific Committee, 1998). Dystonic tremor is similar to essential tremor (ET); however, it involves more irregular, uncontrollable postures and writhing/twisting movements (Deuschl et al., 1998; Jedynak, Bonnet, & Agid, 1991). According to the consensus statement by the Movement Disorder Society (MDS), dystonic tremor may be differentiated from ET if the tremor ceases upon pitch alterations, singing, and emotional speech production (Deuschl et al., 1998). However, both ET and dystonic tremor share an unknown neurologic etiology and closely similar vocal tremor symptoms. Due to this similarity, the terms “EVT,” “vocal tremor,” and “voice tremor” will be used interchangeably throughout this discussion to indicate tremor of the voice upon phonation that may result from either essential or dystonic tremor-effected musculature.
Incidence and quality of life in persons with vocal tremor. Due to its neurogenic origin, EVT tends to affect aging populations most (Finnegan, Luschei, Barkmeier, & Hoffman, 2003) and may be present in isolation or in concert with other diseases, such as with Parkinson’s disease (PD), amyotrophic lateral sclerosis, spinal muscular dystrophy, spasmodic dysphonia, and other types of essential tremor, such as those affecting the limbs or face (Barkmeier-Kraemer et al., 2011; Deuschl et al., 1998). Although EVT has regularly been found to co-occur with these diseases, it is not believed to be caused by the same sources causing those diseases. In fact, different origins are more commonly noted in the research literature. For example, tremor, including vocal tremor, is one of the most apparent symptoms of PD (Solomon et al., 2000). However, treatments such as deep brain stimulation (DBS) and dopaminergic drugs, which help alleviate other symptoms of PD, have varying to no effects on vocal tremor (Gamboa et al., 1998; Putzer, Barry, & Moringlane, 2008). Furthermore, vocal tremor occurring upon phonation in individuals with PD is attributed to EVT and not to PD. This is true because PD is a resting tremor that occurs involuntarily while the act of phonation is a voluntary, intention-based action (Deuschl et al., 1998). Thus, vocal tremor that occurs during phonation is due to EVT because it is voluntary and intention-based (Deuschl et al., 1998).

Many individuals with voice tremor, regardless of underlying diagnosis, report: 1) significant difficulty initiating and maintaining phonation during conversation, 2) effortful speaking, 3) fewer communication attempts, 4) avoidance of social events, and 5) trouble sustaining relationships with others (Cohen, Dupont, & Courey, 2006; Verdonck-de Leeuw & Mahieu, 2004). In an investigation of quality of life issues related
to voice, Cohen et al. (2006) conducted a meta-analysis of 43 Voice Handicap Index (VHI) studies and 14 Short Form 36 (SF-36) studies involving patients with non-neoplastic voice disorders. Results from this analysis revealed that individuals with neurologically-based voice disorders, including EVT, had significantly lower QOL ratings and greater handicaps as compared to those without voice disorders and those with inflammatory or traumatic laryngeal disease (Cohen et al., 2006).

Some degree of vocal tremor has been reportedly associated with normal aging. Verdonck-de Leeuw and Mahieu (2004) conducted a longitudinal study following 11 healthy males (ages 50-81 years) for five years and revealed a decrease in vocal quality, noted most significantly in the acoustic signal and roughness of the voice over a five-year span. Their results showed that as the intensity of vocal instability (including tremor) increased with aging, many participants reported increased social avoidance behaviors. Hence, an increase in vocal instability, whether due to normal aging or a disorder, such as EVT, may have debilitating results and reduce the QOL of the individuals affected (Verdonck-de Leeuw & Mahieu, 2004; Cohen et al., 2006).

Etiology. As indicated earlier, little is known about the etiology of EVT; however, studies suggest that it may result from the tremor-affected musculature involved in speech production (Barkmeier-Kraemer et al., 2011; Deuschl et al., 1998). EVT has been found to co-occur with tremor of the head, face, tongue, jaw, larynx, and respiratory system (Barkmeier-Kraemer et al., 2011; Deuschl et al., 1998). Research regarding other types of tremor (i.e., hand and body) has indicated that damage to several neurological areas, such as the caudate nucleus, putamen, cerebellum, basal ganglia, thalamus, dentate nuclei and the triangular area which connects the dentate nuclei with the red nucleus and
inferior olive. Damage in these areas would disturb motor functions and coordination (Critchley, 1949; Deuschl, Raethjen, Lindemann, & Krack, 2001). However, it is not clear how this type of damage affects the voice/speech system. Some researchers suggest that the tremor only influences the larynx and respiratory systems (Farinella, Hixon, Hoit, Story, & Jones, 2006), while others feel the tremor affects the pharynx, larynx, and the respiratory system (Barkmeier-Kraemer & Story, 2010). Finnegan et al. (2003) have noted that uncontrolled activation of the thryarytenoid and cricothryoid muscles may be responsible for laryngeal EVT. Others have demonstrated that uncontrolled activation of the respiratory muscles results in changes in the desired subglottal pressure for voice production, contributing to a vocal tremor of respiratory origin (Farinella et al., 2006; Hachiniski, Thomson, & Buch 1975; Hixon, Mead, & Goldman, 1976; Tomada, Shibasaki, Kuroda, & Shin, 1987). Many individuals with EVT have reported an additional tremor in the diaphragm and expiratory musculature that may further exacerbate the tremor and instability of the vocal mechanism (Tomoda et al., 1987).

Barkmeier-Kraemer and Story (2010) have simulated EVT of a pharyngeal origin (which also encompasses the articulators) with a computer generated speech production model. In a set of preliminary experiments, they utilized this model to simulate vocal tremor stemming from different sources. They noted that since the ultimate perceived speech signal is produced at the level of the lips, tremor affecting any part of the vocal tract, such as the musculature of the pharynx and articulators, may also contribute to perceived EVT. Furthermore, the researchers utilized this same model to assess listener perception across the various sources of tremor.
While EVTs of laryngeal and respiratory origin result in similar modulation rates of pitch and frequency (Hixon, Klatt, & Mead, 1971), Barkmeier-Kraemer & Story (2010) found that tremor of laryngeal origin was more easily identified perceptually by naïve and experienced listeners than tremor of respiratory and pharyngeal origins. In fact, the naïve listeners in their study could not even detect tremor of respiratory origin, suggesting that this may be the most difficult tremor source to perceive.

**Perceptual and acoustic measures of voice tremor.** While research involving the Voice Handicap Index (VHI) shows that persons with voice disorders, such as EVT, can hear vocal deviations during their own phonation (Cohen et al., 2006), less is known about how listeners perceive a voice with tremor (Kreiman, Gabelman, & Gerratt, 2003). Farinella et al. (2006) simulated voice tremor of respiratory origin in five healthy individuals. A body plethysmograph was used to apply forced oscillatory pressure changes to the body surface, ranging from 0-4 cmH$_2$O in increments of .5 cmH$_2$O. This process induced vocal tremor in the participants when they phonated. Listeners were then asked to judge the lowest pressure point at which tremor could be detected (mean tremor detection threshold) from participant voice recordings in each pressure change condition. Their results indicated a mean tremor detection threshold of 1.37 cmH$_2$O for vowel productions and 2.16 cmH$_2$O for sentence productions. These findings suggest even slight pressure changes within the respiratory system may cause easily perceived tremulous voicing, especially in lengthened vowel productions.

Research also has shown that the perception of EVT severity is dependent upon the acoustic variations (modulations) associated with pitch and loudness, respectively (Barkmeier-Kraemer et al., 2011; Farinella et al., 2006; Kreiman et al., 2003). In normal
voices, frequency and intensity naturally vary so as to produce slight instabilities in the voice (Aronson & Bless, 2009; Kreiman et al., 2003). However, in voice tremor, ther variations in these parameters increase significantly more than in normal voices (Kreiman et al., 2003). The rate of pitch and loudness modulations (indicating the severity of vocal tremor) has been found to be contingent upon the specific musculature affected (i.e., laryngeal or respiratory muscles) (Barkmeier-Kraemer et. al., 2011). Furthermore, the magnitude of these modulations has been found to be approximately 1.42 semitones, which is similar to a singer’s vibrato (Ramig & Shipp, 1987).

Researchers have further shown that multiple factors, such as tremor rate, regularity, and amplitude of frequency modulations, interact to affect expert listener perception of vocal tremor. Kreiman et al. (2003) defined three aspects of a voice that could be affected by EVT: tremor rate, regularity and amplitude. Tremor rate indicated how quickly the fundamental frequency (F0) varied, and was displayed as the number of tremulous waves seen within one second of voicing on a spectrogram. Tremor regularity was described as the form/shape and regularity of the voice displayed on a spectogram. Tremor amplitude was defined as the extent of F0 variations noted above and below the mean amplitude. Kreiman et al. (2003) found that as variations of any one of these parameters increased, the overall pattern of frequency modulation increased. However, they also found that listeners could not identify these specific parameters. Instead, they only perceived an overall increase in vocal tremor. Thus, they concluded that listener perception of tremor is dependent upon the total pattern of frequency modulations and not just variations in one parameter.
In another study, Dromey, Warrick, & Irish (2002) investigated the voices of ten individuals diagnosed with vocal tremor and found increased tremor rates as loudness varied in soft-voice conditions. This increase in tremor rates due to intensity may be dependent on the origin of the tremor. For example, in respiratory-induced voice tremor, tremor was noted to decrease with increases in loudness and increase in soft-voice conditions (Farinella et al., 2006). Furthermore, researchers have reported significantly higher jitter and shimmer values as EVT severity increased (Gamboa et al., 1998). Since jitter and shimmer are believed to represent fine changes in laryngeal function (Ramig & Ringel, 1983), changes in these values also may be apparent in the acoustic measurement of vocal tremor.

Dromey and colleagues (2002) also examined the effect of pitch changes on tremor severity in ten individuals diagnosed with EVT. Their results revealed that higher pitch phonation resulted in increased modulation rates for both amplitude and frequency and that low-pitch phonation resulted in decreased amplitude modulation rates. These researchers hypothesized that the increase in tremor due to high-pitch phonation may be a direct result of the increased muscle activation in the laryngeal system needed to raise pitch. While these investigators attribute the changes in amplitude and frequency modulation to changes in f0, it is possible that changes in laryngeal position are involved. For instance, in normal voicing, the larynx naturally rises or lowers during the production of high or low pitches, respectively. Thus, a raised larynx could contribute to increased muscular effort in the vocal system and consequently, may increase the severity of vocal tremor.
Medical and Surgical Management for EVT

Options for managing vocal tremor include pharmological drugs, alcohol, Botox, and deep brain stimulation (DBS). Pharmological drugs are the most frequently used treatments and include Beta-blockers, such as propranolol, and anticonvulsion/seizure drugs, such as primidone, clonazepam, and Zonisamide (Chen & Swope, 2007; Koller, Glatt, Biary, & Rubino, 1987). However, some researchers have found them effective in reducing tremor (Massey & Paulson, 1985; Tomoda, Shibasaki, Kuroda, & Shin, 1987) while others have found no change in tremor severity (Koller et al., 1987; Koller, Graner, & Mlcoch, 1985).

Many individuals with EVT and other forms of tremor have reported a temporary improvement of voice when using alcohol for short periods of time (Chen & Swope, 2007; Dromey et al., 2002; Koller et al., 1987; Massey & Paulson, 1985). The researchers suggest that tremor reduction occurs because alcohol acts as a central nervous system depressant (Pisoni & Martin, 1989) which reduces the muscle tension associated with increased tremor severity (Dromey et al., 2002). Several researchers have noted that vocal tremor increased as vocal stressors, such as a rise in F0 or vocal instability rose (Brown & Simonson, 1963; Dromey et al., 2002; Mendoza & Carballo, 1998; Tolkmitt & Scherer, 1986). So, the use of alcohol may reduce the vocal stress occurring during voice production in EVT. Given that the intake amount and possible side effects cannot be precisely determined in regards to tremor, alcohol is not recommended as a treatment for EVT (Chen & Swope, 2007; Dromey et al., 2002).

Some studies have shown that the injection of botulinum toxin type A (BoNT-A/BOTOX) unilaterally or bilaterally into the vocal folds temporarily decreases the severity
of EVT in some individuals. However, several side effects from Botox injections have been noted, including major effects of breathiness (sometimes lasting for 2-10 weeks) and dysphagia (Adler et al., 2004; Sulica & Louis, 2010; Warrick et al., 2000). Some individuals reported fatigue, hoarseness, increased phlegm, choking, coughing, and pneumonia after Botox injections (Adler et al., 2004; Sulica & Louis, 2010; Warrick et al., 2000). These negative side effects are likely due to reduced vocal fold adduction strength, which results in increased laryngeal airflow during phonation (breathiness) and reduced laryngeal airway protection (dysphagia, choking, coughing, and pneumonia) (Warrick et al., 2000).

While some individuals may be willing to tolerate these side effects, only a small number of individuals experienced objectively significant voice improvement as a result of unilateral or bilateral injections of Botox (Sulica & Louis, 2010; Warrick et al., 2000). Warrick et al. (2000) followed 10 individuals with diagnoses of EVT alone or EVT with adductor spasmodic dysphonia (ADSD) who were given unilateral and bilateral injections of Botox into their thyroarytenoid muscle. Reductions in tremor frequency were noted due to less vocal effort and laryngeal airway resistance. However, these effects were only noted in a small number of participants and seemed to be more beneficial for individuals with EVT and ADSD than individuals with EVT alone. They attributed this effect to the focused neurological dysfunction of ADSD to the thyroarytenoid muscle, where as EVT neurological dysfunction may disrupt multiple muscles involved in voicing and thus may have been less affected by the injection of BOTOX into the thyroarytenoid muscle (Warrick et al., 2000). Although this hypothesis is plausible, these results may simply
point to the effect of the Botox on the ADSD (Blitzer & Brin, 1994; Truong, Rontal, Rolnick, Aronson, & Mistura, 1991).

In addition to drug therapies, the use of deep brain stimulation (DBS) at the level of the thalamus has been studied to observe its effects on many types of tremor, including severe EVT (Moringlane, Pützer, & Barry, 2004; Putzer, Barry, & Moringlane, 2008; Sataloff, Heuer, Munz, Yoon, & Spiegel, 2002). Two studies found that the use of DBS was effective in decreasing voice tremor related to essential tremor, PD, and Holmes’ tremor; however these papers consisted of only single case studies (Sataloff et al., 2002; Moringlane, Pützer, & Barry, 2004). An additional study of nine individuals with phonatory issues related to PD found that the overall effectiveness of DBS on the phonatory system could not be determined because only some individuals benefitted from it (Putzer et al., 2008). Thus, due to these individual differences in effectiveness and conflicting research, the use of DBS must be discussed with great caution and might not be worth the potential surgical risks, unless the vocal tremor is severely debilitating.

**Voice Therapy for EVT**

Widely used, physiologic voice therapies, such as Lee Silverman Voice Treatment (LSVT), Vocal Function Exercises (VFEs), Lessac-Madsen Resonant Voice Therapy (LMRVT), and expiratory muscle strength training (EMST) have shown to be effective in treating a wide variety of voice disorders (Chen, Hsiao, Hsiao, Chung, & Chiang, 2006; Kim, Davenport, & Sapienza, 2009; Ramig, Countryman, Thompson, & Horii, 1995; Ramig, Sapienza, Brown, Martin, & Davenport, 1999; Sapir, Fox, & Countryman, 2001; Schulz, 2002; Silverman, et al., 2006; Titze, 2006; Yorkston, Spencer, & Duffy, 2003). However, previous research has regarded these approaches to be generally ineffective in
treating vocal tremor (Barkmeier-Kraemer et al., 2011; Boone & McFarlane, 1988; Chiara, Martin, & Sapienza, 2007; Sapir, 1989). Treatments, such as LSVT and EMST, have been extensively studied in patients with neurologic voice and speech disorders, specifically PD (Kim et al., 2009; Ramig et al., 1995; Ramig et al., 2001; Sapienza et al., 1999; Schulz, 2002; Silverman, et al., 2006; Yorkston et al., 2003). However, these treatments appear to be most beneficial in improving swallowing, speech intelligibility, and vocal loudness (Baumgartner, Sapir & Ramig, 2001; Countryman, Hicks, Ramig & Smith, 1997; Fox et al., 2006; Kim et al., 2009; Sapienza et al., 1999; Silverman et al., 2006). No changes were reported in voice tremor due to these treatments.

Barkmeier-Kraemer et al. (2011) described compensatory voice therapy strategies which involved laryngeal maneuvers (i.e., yawn-sigh, easy voice onset, anterior focus), relaxation, and breathing exercises. These activities were prescribed to reduce muscle tension in and around the head and neck region, which tends to co-occur with and exacerbates vocal tremor. In their single case study of a 55-year-old female, a nine-session therapy treatment focused on use of the following compensatory voice therapy strategies: yawn-sigh, easy voice onset, anterior focus, elevated pitch, relaxation and breathing exercises, and use of a more rapid articulation rate (Barkmeier-Kraemer et al., 2011). Although results from the study revealed patient self-satisfaction with the treatment, no significant differences could be demonstrated pre- or post-treatment in either listener perceptual ratings or acoustic outcomes, such as voicing durations, intensity, and fundamental frequency (Barkmeier-Kraemer et al., 2011). It is not clear why these specific techniques were selected as research has shown that the use of high-pitch phonation may actually increase vocal tremor (Dromey et al., 2002) and that use of
a faster speaking rate to shorten vowel length may be difficult to practically utilize in conversational speech.

**Development of a Novel Therapy Approach to Improving Voice in EVT**

The impetus for the present project stemmed from three observations: 1) the incidence of EVT in the aging population (Finnegan et al., 2003), 2) there is no evidence-based behavioral therapy shown to lessen vocal tremor in persons with EVT, and 3) the authors are aware of an anecdotal report of perceived voice improvement in an elderly woman with EVT following her use of respiratory therapy using an incentive spirometer after orthopedic surgery. The hypothesis for the last observation is that increases in lung volume resulted in laryngeal lowering which, in turn, resulted in improved laryngeal muscle forces/vocal fold vibration and decreases in vocal tremor.

**Relation of laryngeal position to EVT.** The relation of laryngeal position to vocal tremor may be a possible treatment focus. As mentioned earlier, Dromey et al. (2002) found that low-pitch phonation reduced the amplitude rate of vocal tremor. They concluded that a lowered larynx may have contributed to relaxation and stability in the vocal system, resulting in the reduction of tremor severity.

There is research support for the notion of laryngeal position and the reduction of tension. In a study of the inherent pitch of vowels, Sapir (1989) found that excess constriction in the thyroarytenoid and infrahyoid muscles of the larynx contributed to increased tension of the laryngeal system and to the development of vocal pathologies. He recommended treatment for laryngeal and phonatory dysfunction center on relaxation and reposturing of supralaryngeal articulatory mechanisms, as well as on limiting vertical laryngeal movements. Shipp (1987) also supports this idea with his finding that
maintenance of a lower laryngeal position results in improved vocal fold vibratory position, improved speech resonance, and lower vocal fold closure forces. All of these behaviors improve voice production.

In addition, Boone and McFarlane (1993) demonstrated that an elevated larynx resulted in increased laryngeal muscle effort and tension. This makes voicing effortful and difficult. After using the yawn-sigh technique, their participants were noted to lower their larynx, which increased their vocal tract length and resulted in improved vocal resonance and decreased adductor force of the vocal folds. The implications of this study for EVT are noteworthy given that a lowered larynx in this study allowed for a more open pharynx and a slight opening of the glottis. These same behaviors may act to reduce the excessive irregular vocal fold movements found in EVT and the muscle tension, which often exacerbates the tremor (Iwarsson & Thomasson, 1998; Iwarsson, Thomasson, & Sundberg, 1998; Shipp, 1987).

**Lung volume.** Laryngeal lowering/vocal tract lengthening also can be achieved through increases in lung volume, which cause the diaphragm and the supporting respiratory musculature to stabilize and lower the larynx through a diaphragmatic-tracheal pull (Iwarsson & Sundberg, 1998; Iwarsson et al., 1998; Thomasson, 2003). Specifically, studies have shown that as lung volume increases, the length of the vocal tract also increases due to a diaphragmatic-tracheal pull which helps the infra- and suprathyroid musculature to lower the larynx (Iwarsson & Sundberg, 1998; Iwarsson et al., 1998; Thomasson, 2003). These findings further indicate that decreased laryngeal position, along with its reduced laryngeal muscle tension and effort, may, in turn, result
in reductions vocal fold closure forces. In persons with EVT, these changes might result in reductions in tremor severity as well as perceptions of vocal tremor.

Inspiratory exercises, like those assigned to surgical patients post-operatively, have been found to increase patient lung volume that may have been diminished due to lack of physical activity post surgery (Booker, 2005; Chumillas, Ponce, Delgado, Viciano, & Mateu, 1998). As stated above, such increases in lung volume may result in a lowered larynx position and associated vocal fold vibratory enhancement, which may result in a lessening of vocal tremor amplitude and/or frequency as well as a reduction in listener perceptions of tremor. This may, in turn, result in speaker (patient) perceptions of less impairment, as shown in QOL measures.

**Statement of the Problem**

EVT likely results from lack of coordination between vocal fold adduction and abduction (Barkmeier-Kraemer et al., 2011; Critchley, 1949; Deuschl et al., 2001). This dyscoordination within the laryngeal musculature negatively affects the vocal folds’ symmetrical motion and results in tremulous vocal output. Consequently, individuals with EVT experience difficulty speaking and reduced QOL (Cohen et al., 2006; Verdonck-de Leeuw & Mahieu, 2004).

Currently, there is no effective medical, surgical, or behavioral treatment for the disorder. The goal of existing physiologic voice therapies is to achieve a permanent or sustained voice improvement, something that may not be possible in the case of vocal tremor, given that its underlying cause is a continuous movement disorder of laryngeal and possibly pharyngeal and respiratory muscles. In addition, current compensatory strategies to minimize vocal tremor, such as those proposed by Barkmeier-Kraemer et al.
(2011; i.e., high-pitch phonation and faster speaking rate), may be impractical for persons with tremor to continuously implement in conversational speech given the demands of on-going speech as well as the absence of acoustic and perceptual research to support these strategies in terms of their reduction of tremor severity.

It is possible that a novel treatment strategy, which has a direct, positive effect on the larynx and laryngeal muscles, might lessen the severity of vocal tremor and thereby minimize its perception, as well as improve the QOL in persons with tremor. This finding is supported by previous research which suggests that high lung volume is associated with a lower larynx position and maintenance of a lower laryngeal position results in an improved vocal fold vibratory position (Iwarsson & Thomasson, 1998; Iwarsson et al., 1998), improved speech resonance, and lower vocal fold closure forces, all of which benefit/improve voice production (Shipp, 1987). Finally, this approach is supported by a recent anecdotal report by an older woman. She reported a reduction in vocal tremor and listener reports of improved voice production after completion of post-surgical breathing exercises using an incentive spirometer. Based on these observations, a simple exercise strategy, devoid of undesirable side effects and offering immediate tremor and possible vocal effort reduction for persons with EVT was proposed.

The specific purpose of this project is to test the hypothesis that inspiratory breathing exercises among patients with EVT can result in 1) reductions in acoustic measures of vocal tremor, 2) concomitant reductions in listener perceptions of vocal tremor, and 3) decreased vocal effort as well as improved QOL indices among patients with vocal tremor. The goal of our study is to answer the following questions:
1. Did the participant increase their lung volume as a result of inspiratory muscle training with an incentive spirometer?

2. Is there a significant gain in QOL based upon Voice Handicap Index (VHI) survey ratings pre- and post-treatment?

3. Are there any significant vocal benefits revealed through acoustical analysis pre- and post-treatment?

4. Is there a significant change shown in perceptual findings based on trained and untrained listener ratings (The Consensus Auditory Perceptual Evaluation of Voice (CAPE-V) and listener ratings) pre- and post-treatment?
Chapter 2

Method

Participants

The principal investigator (PI) provided flyers explaining the project to Tampa Bay area otolaryngologists, neurologists, and speech-language pathologists. These individuals were instructed to hand out the flyers to appropriate patients who would then contact the PI if they were interested in participating in the study. All interested and eligible individuals who contacted the PI were scheduled to come to the university to participate the study.

Tremor participants. Three adult women, ranging in age from 54-65 years volunteered. All had been clinically diagnosed with vocal tremor by an otolaryngologist or neurologist. They did not have any cognitive or intellectual impairment. Furthermore, they did not have any structural impairments/defects that impeded speech production. They were of good physical health and were able to successfully complete a home therapy program of breathing exercises. Each participant will be described in more detail below.

Participant 1 was a 58-year-old female who was diagnosed with EVT and ADSD about 6 years ago. She currently was being treated for her ADSD with Botox. Other prescribed medications included Nexium, Allegra, Mucinex, and Sertraline. According to the National Center for Voice and Speech (2011), use of these drugs may have a drying effect on the voice and increase vocal hoarseness.
Participant 2 was a 54-year-old female diagnosed with essential tremor of the voice and left side of the body, including the muscles of the head to the left hand. In addition, she had an Arnold-Chiari Malformation, and ADSD. She reported that her voice tremor began approximately 17 years ago and had become more severe over time, affecting the rest of her body. Initially, the patient noted that it was a strain to speak, as if “something was in [her] throat.” She also reported that as she continues to speak, she feels her “throat closes up”. She is receiving Botox treatment for the ADSD and is taking Topamax, an anti-seizure medication that has no known direct effects on the voice (National Center for Voice and Speech, 2011).

Participant 3 was a 65-year-old female diagnosed with severe essential tremor of the voice and hands. She was also diagnosed with Ankylosing Spondylitis (arthritis in the lower spine) and peripheral neuropathy, which she believes might be linked to a past surgery at 28 years of age in which half of her thyroid gland was removed. Her vocal and hand tremor began about 10 years ago and is believed to be familial, as relatives have been diagnosed with different types of bodily tremor. Her main vocal concerns were to be able to have a properly functioning voice and to decrease her social anxiety. In addition to allergy medications, fish oil, and multiple vitamin supplements, she was taking the following prescribed medications: Lexapro, Synthroid, aspirin, Lisinopril, Neurontin, Toprol XL, Lipitor, Indomethacin, Provigil, and Zantac or Prilosec, as needed. Some of these medications have been reported to have a drying effect on the voice and increase vocal hoarseness, and also increase the risk of vocal fold injuries (National Center for Voice and Speech, 2011).
Listeners. These participants were recruited by contacting undergraduate instructors at a local university and asking them to read an announcement to their class. A list of volunteers was collected and the examiner contacted them to set up a time for the listening experiment. The PI also spoke to second year speech-language pathology graduate students at that university asking them if they were interested in participating.

Twenty-one young college-age students (20 females and 1 male) volunteered to judge voice samples for the degree of vocal quality and tremor noted. One participant’s results were not included in the study as this participant reported a current sensorineural hearing loss. Thus, only 20 listener participants’ data was included and analyzed in this study. All of these 20 listeners had normal hearing, as determined by scoring 92% or better on a speech listening test (Griffiths, 1967) and had no history of neurological disease or language impairment. They were recruited from classes in the Communication Sciences and Disorders program at University of South Florida, Tampa, FL. Fourteen of the participants had no prior experience evaluating voice quality, while six of the participants were second year speech-language pathology graduate students, who had some familiarity with evaluating voice quality. However, all listeners were considered naive listeners in regards to evaluating voice quality.

Materials

Incentive spirometer. The participants used a simple incentive spirometer as the breathing trainer for this study. This type of spirometer is often given to patients post-surgery in order to improve the individual’s lung function. In this case, it was used to study the effects of improved lung function on reducing voice tremor. The breathing
exercises for this project were based on those modeled after the guidelines developed by the Cleveland Clinic Foundation (1995-2009) for patients post-surgery.

**Voice Handicap Index (VHI).** The *Voice Handicap Index (VHI)* (Jacobson et al., 1997) was administered to each participant three times over the span of the entire experiment. The VHI is a 30-item self-assessment questionnaire that measures the effects of a voice disorder in a person’s daily life. The items on the questionnaire represent the physical, functional, and emotional aspects of impairment associated with voice disorders. Each of these three subscales has ten statements, which are scored on a 5-point equal-appearing interval scale, with a possible total numerical score ranging from 0-40. The grand total of these subscales ranges from 0-120, with higher scores indicating a greater degree of (patient) perceived impairment or handicap. Significant or meaningful score changes are 8 or more for the subscales and 18 or more for the total VHI score (Jacobson et al., 1997).

The authors of the VHI assessed the validity of the instrument by administering it two separate times to 63 patients. Pearson product-moment correlation coefficient scores indicated strong test-retest reliability between subscale and total scores with reliability estimates ranging from $r = 0.84$ to $r = 0.92$. Furthermore, moderate-to-strong correlations between subscale scores were found, with the magnitude of the relationships between subscales ranging from $r = 0.70$ to $r = 0.79$ (Jacobson et al., 1997).

**Consensus Auditory Perceptual Evaluation of Voice (CAPE-V).** The *Consensus Auditory Perceptual Evaluation of Voice (CAPE-V; Kempster, Gerratt, Verdolini Abbott, Barkmeier-Kraemer, & Hillman, 2009)* is an instrument used to assess listener perceptions different elements of vocal quality. Only the PI used this instrument.
to judge the voices of the tremor participants across their overall productions of isolated vowels, the reading passage, and spontaneous speech tasks together. The PI was a second year Speech-Language Pathology graduate student who, although also considered a naïve listener, differed from the other listener participants in that she was trained to rate voices on the CAPE-V.

To better suit this experiment, the questions used to elicit the spontaneous speech portion on the CAPE-V were altered to reduce potential emotionality, which research shows could negatively affect voice output in patients with neurological disorders (Zraick et al., 2011). For example, the spontaneous speech probe “Tell me about your voice problem”, may induce emotions of sadness and grief, especially if the patient has felt some sort of reduced quality of life or loss secondary to vocal tremor. Thus, in order to avoid these effects on the study results, the following four topics were used in place of the CAPE-V questions provided: “Tell me about your last vacation,” “Tell me how to make your favorite dish,” “Tell me how to play your favorite sport or game,” and “Tell me about your favorite place to eat.” One topic was used for each of the four recording sessions in this project.

In terms of evaluating voice quality, The CAPE-V specifies six voice features for evaluation: overall severity (the overall impression of the level of voice deviance), roughness (perceived abnormality in voice quality), breathiness (the amount of audible air escaping during voicing), strain (perception of excessive vocal effort), pitch (the perceptual correlate of fundamental frequency), and loudness (the perceptual correlate of sound intensity). Each of the six voice quality features is measured on a visual analog scale measuring 100 mm. Ordinal ratings of “mild,” “moderate,” and “severe” are printed
below each scale and serve as supplemental severity indicators with terminology that is more familiar to clinicians than the discrete intervals alone. In addition to these six quality features, the CAPE-V allows for the addition of supplementary relevant perceptual features of an individual’s voice, such as the degree of nasality, spasm, tremor, intermittent aphonia, falsetto, glottal fry, weakness, or other aspects that may be important in the characterization of the individual features of a patient’s voice quality.

In regards to the validity and sensitivity of the CAPE-V, researchers have found that the measure allows for consistency across clinical assessments with its specific protocol instructions for the tasks, procedures, and scaling routine. It was also found that the CAPE-V has strong inter- and intra-rater concurrent reliability. Pearson’s coefficient analysis between subscales revealed overall intra-rater reliability to range from $r = 0.35$ (for strain) to $r = 0.82$ (for breathiness). Intra-class correlation (ICC) coefficient analysis revealed inter-rater reliability to range from coefficients of 0.28 (for pitch) to 0.76 (for overall severity) (Zraick et al., 2011).

**Speech listening test.** Prior to rating tremor participant voices, each naïve listener participant was given an abbreviated version of a speech listening task (Griffiths, 1967) to determine that they had normal speech perception abilities. The task consists of 25 sets of minimal pairs heard through headphones and presented within the ECoS Version 2™: Experiment Generator and Controller program (Avaaz Innovations, Inc., 2002). Each word was presented in the carrier phrase, “The first (second, third, etc.) word is ____.” While the word was being spoken, five words were presented on a computer screen, the target word and four minimal pairs based on the target word. The participant was asked to click on the word they heard or thought they heard. This task was scored within the ECoS
program (Version 2™; Avazz Inc., 2002) and each participant was required to obtain 92% accuracy to continue participation in this experiment. If the listener did not meet this criterion, his/her data were not included in the statistical analyses.

**Procedures**

The procedures in which the tremor participants took part in consisted of an ABAB (treatment reversal) single subject design. This will be described below.

**Initial visit.** Vocal tremor participants were instructed about the project and informed that they could withdraw from the study at any time, without penalty. They were also told that there were no known risks associated with participation in the treatment program and they were asked if they were willing to participate. All participants agreed, the informed consent form was signed, and the study began. (This study received Institutional Review Board approval from University of South Florida, Tampa, FL, IRB# Pro00000310.)

After agreeing to participate, the tremor participant received a hard copy of the Voice Handicap Index (VHI; Jacobson et al., 1997) and was asked to rate herself on this voice quality of life measure. Once this form was completed, the clinician provided the participant with a hard copy of the Rainbow Passage and gave her time to review it. Once she felt ready, the participant was seated in front of a Dell computer and a MicroMic Series II, AKG C 420 condenser headset microphone was placed on her head. The microphone to mouth distance was adjusted to approximately 6 cm to the right side of her mouth. The microphone was connected to the computer and the voice was recorded in Praat (Boersma & Weenink, 2009) at a 22,050 Hz digitization rate.
The PI then gave the tremor participants verbal instructions and in some cases examiner models during the voice recording procedure (see Appendix A for specific information regarding tremor participant instructions). During the initial recordings, a conversational sample was gathered by asking the participant to “Tell me about your last vacation.” The tremor participant was also instructed to sustain the isolated vowels /i/ and /a/ twice for at least 10 seconds and then to read the Rainbow Passage (Fairbanks, 1960). These tasks were presented randomly across participants and recording sessions (see Table 2.1 below). For example, if tremor participant 1 produced the Rainbow Passage first in the Pre-Treatment condition, she would then follow it with production of isolated vowels and produce the conversational speech sample last. Then, participant 1 would produce the conversational speech sample first in the Post-Treatment #1 condition, followed by producing the Rainbow Passage and finally the sustained vowels, and so on (see Table 2.1).

Table 2.1: Randomization order of speaking tasks produced by tremor participants by treatment session.

<table>
<thead>
<tr>
<th></th>
<th>Tremor Participant 1</th>
<th>Tremor Participant 2</th>
<th>Tremor Participant 3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pre-Tx</td>
<td>Rainbow Passage</td>
<td>Isolated Vowels</td>
<td>Conversational Sample</td>
</tr>
<tr>
<td>Post-Tx #1</td>
<td>Conversational Sample</td>
<td>Rainbow Passage</td>
<td>Isolated Vowels</td>
</tr>
<tr>
<td>No Tx</td>
<td>Isolated Vowels</td>
<td>Conversational Sample</td>
<td>Rainbow Passage</td>
</tr>
<tr>
<td>Post-Tx #2</td>
<td>Rainbow Passage</td>
<td>Isolated Vowels</td>
<td>Conversational Sample</td>
</tr>
</tbody>
</table>

After the voice recordings were made, the PI verbally instructed the tremor participants in the daily use of the incentive spirometer (modeled after the guidelines
developed by the Cleveland Clinic Foundation, 1995-2009). Tremor participants were also given these instructions in writing. The instructions were as follows:

1. Hold the spirometer in an upright position.
2. Place the mouthpiece of the spirometer in your mouth and seal your lips tightly around it.
3. Breathe in slowly and as deeply as possible as the yellow piston rises toward the top of the column.
4. Hold each breath as long as possible, but at least for five seconds.

* Repeat the above steps at least 10 times in the morning and 10 times in the evening for one week with a higher yellow indicator on the spirometer used as a goal to work toward during each repetition. Once the targeted indicator is reached, the next highest indicator becomes the next goal for the participant to reach (see Appendix B).

In order to track progress, the patient was instructed to write down the number of repetitions completed and to circle the initial and highest pressure level for each set of ten breathing repetitions on a self-check form. Training began the morning after this initial visit with the PI. Tremor participants were asked to complete the breathing protocol for one week, discontinue the treatment protocol for one week, and reinstate it for another week.

At the conclusion of the initial visit, each tremor participant was provided with copies of the following documents: two copies of the Voice Handicap Index (VHI), the self-check schedule form, and the Rainbow Passage. She was instructed to fill out the VHI survey after the first and second week of training and mail them back to the clinic.
Finally, she was instructed to read the Rainbow Passage during all post-treatment recordings. To increase the possibility that the participant would complete three additional recording sessions, follow-up visits were recorded during a weekly phone call to the patient. These post-treatment telephone appointments were scheduled with each participant during the initial visit. Participant 3 chose to come into the laboratory for all of her subsequent visits, while participants 1 and 2 elected the telephone option.

For all telephone recordings, an AT&T-SN landline telephone acted as the microphone, which was connected to a Re-Tell Telephone Recording Connector (landline telephone adaptor) and a Studio V3 ART preamplifier inserted into the microphone input on the computer. All samples were then recorded through the computer software program Pratt (Boersma & Weenink, 2009) With the exception of mode of recording (telephone or direct microphone), all other procedures for subsequent appointments were the same for all the participants. The PI rated all voice samples on the CAPE-V after the initial visit and after the two treatment sessions.

**Post-treatment week (#1).** At the end of week one, the PI spoke with each tremor participant in order to check on her progress and to record her voice. The examiner asked her to rate her voice on the VHI and had the phone participants mail it back to the clinic. Participant 3 filled it out and returned it to the PI in person. Then, the examiner recorded the three speech samples, the conversational sample with the prompt, “Tell me how to make your favorite dish”, the isolated vowels /i/ and /a/, and the Rainbow Passage (Fairbanks, 1960) in the randomized orders as shown in Table 2.1. Finally, the PI rated the participant’s voice with the CAPE-V (Kempster, et al., 2009). After the completion of
the recordings, the clinician instructed the patient to discontinue the breathing exercise protocol for the following week.

**No treatment week.** At the end of the no treatment week, the clinician again met with the participants to make voice recordings. The same procedures were followed as described above. The conversational prompt for this session was “Tell me how to play your favorite sport or game.” After the completion of the recordings, the clinician instructed each patient to reinstate the breathing exercise protocol for the following week.

**Post-treatment week (#2).** At the end of the third week (after the second week of the breathing treatment), the clinician met with the participants in order to check on their progress and record their voices. The same procedures were followed. The VHI was completed and returned to the PI, along with the treatment self-check forms. The conversational sample was gathered with the following prompt, “Tell me about your favorite place to eat.” Finally, the examiner asked each participant about how she felt about the breathing protocol.

**Acoustic measures.** One-second vowel samples taken from the middle third of the better vowel production obtained during each recording session were analyzed for each speaker in order to note any change in vocal tremor or voice quality over the course of treatment. To analyze the vowels, the following vocal parameters were obtained using the Voice Report within Praat (Boersma & Weenink, 2009): fundamental frequency (F0) mean and standard deviation (F0 SD), percentage of jitter (local), percentage of shimmer (local), and harmonics-to-noise ratio (HNR, dB).

In the Praat manual, Boersma and Weenink (2009) provided definitions and standards for the measures of perturbation used in this study. Jitter (local) was defined as
the “average absolute difference between consecutive periods, divided by the average period” and any values above 1.040% are considered pathologic. Shimmer (local) was defined as the “average absolute difference between the amplitudes of consecutive periods, divided by the average amplitude” and values above 3.810% are considered pathologic (Boersma & Weenink, 2009). Harmonics to Noise Ratio (HNR) was defined as the “degree of acoustic periodicity” and any values below 20 dB are considered to be associated with abnormally hoarse vocal quality.

These acoustic measures were chosen because similar measures were used in previous studies evaluating vocal tremor (Dromey et al., 2002; Gamboa, et al., 1998; Ramig & Shipp, 1987). The frequency rates of tremor were analyzed by isolating one second of a vowel production on the computer screen and counting the sinusoidal waves noted by comparing the F0 (blue) and amplitude (yellow) lines.

Figure 2.1: A spectrogram of a one-second vowel sample used to determine tremor frequency rate. The tremor rate here is 3 Hz.
**Tape preparation for evaluation by naïve listeners.** Each recorded sample from the tremor participants was opened in Praat (Boersma & Weenink, 2009). Spectographic views of the data were edited into the different types of speech samples for each participant for use in the listening experiment. Vowel sample duration was measured and middle third of the sample was saved. The second sentence and half of the third sentence of the *Rainbow Passage* (Fairbanks, 1960), “The rainbow is a division of white light into many beautiful colors. These take the shape of a long, round arch with its path high above...”, was extracted from the reading passage. As for the conversational speech samples, three separate 5-second samples were taken from the middle third of the recording and saved as voice exemplars.

All recorded speech samples then were band-pass filtered from 250-3000 Hz and intensity was adjusted to an average of 70 dB via Praat software (Boersma & Weenink, 2009). This was done in order to equalize samples for sample clarity and loudness across recording conditions, lab microphone or landline telephone.

Using the ECoS Version 2™: Experiment Generator and Controller program (Avaaz Innovations, Inc., 2002), the listening experiment was created. The speech listening task (the Griffiths test, 1967) was first, followed by recorded instructions and a practice condition. The experimental condition then was presented as three separate blocks: Block 1 (Reading Sentence Block), Block 2 (Conversational Sentence Block), and Block 3 (Vowel Block). For determination of listener reliability, twenty percent of the stimuli in each block were repeated in the same order at the end of the listening block. In this way, the consistency of listener response could be established.
The practice condition consisted of nine trials. These samples were taken from portions of the tremor participants’ voice recordings that were not used in the experimental condition. There were three sentences randomly chosen from the participants’ readings of the fourth sentence of the Rainbow Passage (“There is, according to legend, a boiling pot of gold at one end.”), three sentences randomly chosen from the participants’ conversational speech tasks, and three random vowel samples. All tremor participants were represented in each speech sample selected for the practice condition. For the experimental portion, Blocks 1 and 2 included sentences taken from the Rainbow Passage and their conversational speech sample. This resulted in 12 voice samples (four from each of the three patients) from each speaking condition. Five of these sentences (20%) were repeated for consistency of rating determinations, for a total of 29 trials. Block 3 included 16 isolated vowel samples (two samples for each vowel taken from the four treatment conditions) from each speaker. This resulted in 48 vowel samples, with ten additional vowel samples (20%) repeated for reliability. The total number of trials for this block was 58 trials. Speech samples were quasi-randomly presented within each block, so that the same voice did not appear within two samples of itself. A short break was created after each block to give the listener a break and to instruct them for the next block.

**Listening experiment.** Upon entering the lab, each listener participant was greeted and thanked for their participation. They were given a brief explanation of the procedures for the rating task and the opportunity to ask any questions. Then, the PI read through the informed consent with them and requested that they sign once they felt comfortable doing so. They were also asked if they had any history of speech, language,
or hearing impairment and were asked to detail their specific exposure to rating voices, if any.

Each listener participant was seated at a computer and provided with written and oral instructions on how to complete the speech listening task (for complete listener instructions, see Appendix C). The listener was asked to wear Numark PHX USB+ analog DJ Headphones to complete the experiment. The headphones were cleaned between listening sessions using alcohol-free antiseptic wipes. To determine if they had adequate listening skills to perform the desired task, the speech listening task (Griffiths, 1967) was administered first. After passing the speech perception task with at least 92% accuracy, the practice condition began, followed by the experimental tasks.

For the practice and experimental tasks, listeners heard a voice sample and were asked to rate that sample on two different 7-point equal-appearing interval scales presented on the computer screen. The first 7-point scale included was marked as “Steadiness” and the second 7-point scale was marked as “Pleasantness.” Listeners were instructed that a rating of 1 indicated great vocal steadiness or pleasantness and that a measure of 7 indicated poor vocal steadiness or pleasantness. The listeners clicked the mouse cursor onto the number they believed most represented the degree to which the speaker’s voice was steady or pleasant. The scales looked as follows:

<table>
<thead>
<tr>
<th>Steadiness</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pleasantness</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>6</td>
<td>7</td>
</tr>
</tbody>
</table>

*Figure 2.2: An example rating scale used in the listening experiment.*
Listeners used these scales to rate all tremor participant voice samples (sustained vowels, conversational speech, and Rainbow Passage sentences). Once the listeners completed the voice ratings, they clicked on the button labeled “Accept,” after which the next voice sample appeared. No feedback was provided during the practice condition to allow the listener to develop their own strategy for rating the voice sample. Listeners were allowed to repeat the sample one time by clicking on the button labeled “Replay Stimulus.”

**Rating Consistency**

After all voice recordings were made, the PI completed CAPE-V ratings (Kempster et al., 2009) for each tremor participant across all treatment conditions. To determine rating consistency, the PI re-rated all voice samples a month after their initial recordings. These two rating scores remained similar to each other numerically such that overall severity levels did not change for any of the patients’ voices; thus both initial and secondary scores were averaged and these means were reported in the patient data.

Listener ratings were individually compared for the repeated trials within each block. Those that differed by more than 1 rating interval were noted. In other words, if the initial rating was a 4, then the listener could judge that stimuli to be a 3, 4, or 5 the next time it was heard. Only ratings from listeners who achieved 80% consistency or greater in matching their original judgment were statistically analyzed. Only one of the 20 listener participants was dropped for not meeting this standard. Percentage of consistency was then averaged across all listeners, with a 92% mean consistency rating achieved across both scales.
Data Analysis

Lung volume data were gathered through the daily schedule patients kept as they participated in the breathing treatment protocol. Initial and highest lung pressure levels gathered from the daily schedule and were transferred from the written schedule forms to Excel data sheets. The data were then placed in graphic format and comparisons were made across morning and evening sessions and across treatment sessions.

In addition to lung volume data, three additional sets of data were used to evaluate treatment effectiveness. The first data set was evaluated in terms of changes in tremor participant scores on the VHI. The second set was evaluated in terms of acoustic changes in isolated vowel samples. The third set was evaluated in terms of changes in the trained PI perceptual ratings on the CAPE-V and the untrained listener participant perceptual ratings of steadiness and pleasantness. For the untrained listener participant perceptual ratings of steadiness and pleasantness, two-way ANOVAs were used to determine differences across sample type (vowel, /i/ vs. /a/ or connected speech: conversation vs. reading) and recording condition (pre-test and the 3 post-test conditions). These analyses were run for the dependent variables of Steadiness and Pleasantness individually, for a total of four separate ANOVAs. T-tests with a Bonferroni correction ($p = 0.05$) were used to determine significant effects and effect sizes.
Chapter 3

Results

An ABAB (treatment reversal) single subject design was used to assess the effectiveness of inspiratory breathing exercises on reducing the severity of EVT. For this project, pre-treatment measures were administered, then participant progress was measured after one week of treatment (post-treatment #1), a week of no treatment, and another week of treatment (post-treatment #2). The following measures were gathered from each participant to document treatment completion and effectiveness: lung pressure volume levels, VHI ratings, acoustic analyses of isolated vowels, and trained PI perceptual ratings on the CAPE-V, as well as untrained listener perceptual ratings of steadiness and pleasantness. These data will be presented individually and described for each participant in figures or tables, as appropriate.

Note for Listener Participant Ratings

As previously mentioned, untrained listener participant responses were analyzed with eight separate two-way ANOVAs and T-tests with a Bonferroni correction were used for post-hoc testing. Results revealed statistically significant results for multiple sets of listener ratings. However, actual changes in listener mean scores predominantly fell within +/- 1-point of one another for all participants. This range was also the cut off used for listener reliability measures, indicating that no sizeable differences in vocal tremor or in overall vocal quality were perceived by the listeners when rating pre-treatment and post-treatment recordings (see Appendix D). These results will be described for each
participant in conjunction with the other measures of treatment effectiveness gathered for this study.

Presentation of Results by Tremor Participant

Case 1. Tremor participant #1 was a 58-year-old female diagnosed with mild EVT and mild ADSD about 6 years ago. She underwent treatment for the ADSD with a Botox injection one month prior to the initiation of treatment. She reported that the Botox typically lasted for approximately three months. Data will be described in the following order: lung function data, VHI ratings, acoustic analyses, CAPE-V data, and listener ratings of tremor.

Verification of treatment completion: Lung function data. Lung volume data were gathered through the daily schedule patients kept as they participated in the breathing treatment protocol. Initial and highest lung pressure levels, taken from the daily schedule, were graphed and comparisons were made across morning and evening sessions and across the two separate treatment sessions (see Figures 3.1 and 3.2 below). Overall, the results for Participant 1 revealed that the initial and highest lung pressure volumes remained relatively close regardless of time of day or treatment week. These results indicate that no remarkable increases for lung pressure volumes were found for this participant.
Figure 3.1: Initial and highest morning lung pressure volumes of treatment week #1 (1-8) and treatment week #2 (9-14) for Participant 1.

Figure 3.2: Initial and highest evening lung pressure volumes of treatment week #1 (1-8) and treatment week #2 (9-14) for Participant 1.
**VHI ratings.** For Participant 1, there were meaningful decreases in VHI total scores from baseline to post-treatment and in the emotional subtest scores (see Figures 3.3 and 3.4). Functional subscale scores also evidenced reductions from baseline to post-treatment (#2), however these differences did not reach significance. These findings would suggest that the participant did perceive some benefit from treatment, especially in terms of how she felt about her voice difficulties. There did not appear to be a perceptible change in the physical realm (i.e., in the amount of effect necessary to produce voice).

![Graph showing VHI scores across treatments for Participant 1](image)

*Figure 3.3: VHI total scores across treatments for Participant 1.*
**Acoustic analyses.** Few differences were noted in the acoustic realm as a result of treatment (see Tables 3.1 and 3.2). Tremor rate slightly increased for /a/ and evidenced a sizeable jump after Treatment #1 for /i/. Neither change is noteworthy in that all tremor rates remained within the range expected for EVT. It is noteworthy that treatment did not appear to result in a consistent decline in tremor rate.

As all post-treatment recording were made over the phone, increases in F0 for all post-treatment conditions occurred as the phone acts as a band-pass filter of frequencies and passes frequencies approximately between 250-2500 Hz. Pre-treatment measures of F0 for the vowel /i/ reveal a much higher F0 frequency than those noted in pre-treatment measurements of the vowel /a/. This difference is most likely due to the fact that /i/ is a high vowel and has a higher intrinsic pitch than /a/ (Whalen & Levitt, 1995).
Despite overall increases in jitter and shimmer rates and decreases in HNR values, all values remained within the normal range (1.040%, 3.810%, and near 20 dB respectively) for the vowel /a/ indicating no effect of treatment. These values differed from those for the vowel /i/, as increases in shimmer rates and decreases in HNR values were outside the normal range, indicating subtle increases in the perturbation of the amplitude of vocal fold vibration over the course of treatment. Other than possibly due to the differences in the intrinsic pitches of /a/ and /i/ (Whalen & Levitt, 1995), it is not clear why these values were different across vowel productions.

**Table 3.1: Acoustic measurements for /a/ across treatments for Participant 1.**

<table>
<thead>
<tr>
<th>Treatment Condition</th>
<th>F0 Mean (Hz) &amp; SD</th>
<th>Tremor Rate (Hz)</th>
<th>% Jitter</th>
<th>% Shimmer</th>
<th>HNR (dB)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pre-Tx</td>
<td>211 (2.6)</td>
<td>2.75</td>
<td>0.34</td>
<td>1.71</td>
<td>25.83</td>
</tr>
<tr>
<td>Post-Tx (#1)</td>
<td>272 (2.4)</td>
<td>3.25</td>
<td>0.44</td>
<td>3.09</td>
<td>20.97</td>
</tr>
<tr>
<td>No Tx</td>
<td>261 (1.6)</td>
<td>4</td>
<td>0.71</td>
<td>3.43</td>
<td>18.45</td>
</tr>
<tr>
<td>Post-Tx (#2)</td>
<td>271 (3.3)</td>
<td>5.5</td>
<td>0.32</td>
<td>3.26</td>
<td>21.63</td>
</tr>
</tbody>
</table>

Table 3.2: Acoustic measurements for /i/ across treatments for Participant 1.

<table>
<thead>
<tr>
<th>Condition</th>
<th>F0 Mean (Hz) &amp; SD</th>
<th>Tremor Rate (Hz)</th>
<th>% Jitter</th>
<th>% Shimmer</th>
<th>HNR (dB)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pre-Tx</td>
<td>244 (1.5)</td>
<td>3</td>
<td>0.18</td>
<td>1.56</td>
<td>30.35</td>
</tr>
<tr>
<td>Post-Tx (#1)</td>
<td>276 (1.2)</td>
<td>8</td>
<td>0.36</td>
<td>6.5</td>
<td>15.01</td>
</tr>
<tr>
<td>Post-Tx (No Tx)</td>
<td>268 (1.6)</td>
<td>3.5</td>
<td>0.37</td>
<td>10.15</td>
<td>9.71</td>
</tr>
<tr>
<td>Post-Tx (#2)</td>
<td>272 (2.1)</td>
<td>4</td>
<td>0.4</td>
<td>9.48</td>
<td>10.6</td>
</tr>
</tbody>
</table>

**CAPE-V ratings.** Participant 1 presents with a mild voice disorder that is largely characterized by the perception of vocal tremor (see Figure 3.5). The most obvious perceptual change as a result of treatment was in roughness, which was very mild to
begin with and seemed to decrease with treatment. These results appear to contrast with acoustic data which showed slight increases in shimmer and HNR, for the /i/ vowel, which could be perceived as roughness.

Tremor seemed to decrease after the first round of treatment, but not the second. There were no significant changes in strain or overall severity across conditions. The lack of change in perceptual ratings across conditions might be explained by the observation that this participant initially presented with a mild voice disorder, so there was less room for change.

![Figure 3.5: CAPE-V mean subscale scores across treatments for Participant 1.](image)

**Listener participant ratings of tremor speech samples.** In order to further assess the perceptual benefits of treatment, the current study had unfamiliar listeners rate taped voice samples of the tremor participants on two different seven-point scales. Ratings of
one indicated great vocal steadiness or pleasantness and ratings of seven indicated poor vocal steadiness or pleasantness. Analysis of listener ratings for this tremor participant revealed a subtle effect attributed to speech sample type with the ratings being slightly lower for the /i/ vowel. Furthermore, rating scores rose after the second treatment condition for /a/ both connected speech samples on both rating scales, suggesting a small decline in voice quality for these utterances (see Appendix D).

**Summary of results for Participant 1.** Taken together, the results suggest unremarkable increases in lung pressure volumes during the inspiratory breathing treatment. The participant’s QOL as measured by VHI data revealed a significant decrease in total scores and in emotional subscale scores from baseline to post-treatment conditions. Acoustic data was within the normal range, except for the /i/ vowel, which evidenced a slight increase in shimmer and HNR values across conditions. Trained listener CAPE-V rating scores across all treatment conditions indicated that roughness experienced the most obvious reduction. These ratings on CAPE-V parameters also suggested increases in voice quality severity ratings (e.g., increases in strain and tremor ratings) for this participant; however, these changes were minimal. Finally, untrained listener ratings showed little change in vocal steadiness and pleasantness for this participant over time. Overall, the participant’s speech evidenced minimal changes in acoustic and perceptual measures with breathing treatments, but this participant perceived a reduction in vocal handicap with the treatment conditions.

**Case 2.** Participant 2 was a 54-year-old female diagnosed with EVT and moderately-severe ADSD. She was receiving Botox treatments for the ADSD, with her last Botox injection occurring seven weeks prior to breathing treatment initiation. Given
that Botox typically reduces the symptoms of ADSD for approximately three months, she would have been due for an injection shortly after the completion of this treatment protocol.

*Verification of treatment completion: Lung function data.* Lung function data were graphed for morning and afternoon sessions during treatment (see Figures 3.6 and 3.7). Results indicated that lung pressure values tended to be higher in the evening. It was also noted that lung pressure levels increased more during the first week of the treatment protocol and were basically maintained at the higher levels during the second week of treatment. This finding suggests that the breathing treatment was effective in increasing lung pressure.

*Figure 3.6: Initial and highest morning lung pressure volumes of treatment week #1 (1-8) and treatment week #2 (9-14) for Participant 2.*
Figure 3.7: Initial and highest evening lung pressure volumes of treatment week #1 (1-8) and treatment week #2 (9-14) for Participant 2. * Note: Lung pressure volume values are missing as a result of the participant having not circled pressure values for that session on the treatment schedule form.

**VHI ratings.** Total VHI scores, as well as the functional and emotional subscale scores, significantly increased from baseline across both post-treatment conditions. Results in the physical subscale items showed less change. These results indicate that the participant’s perception of their voice disorder actually worsened after treatment. The subscale scores suggest that the participant may have been experiencing increases in self-perceived voice severity related to functional and emotional factors and less to physical impacts. The latter finding could suggest that treatment did influence perceived vocal effort (i.e., a physical parameter). However, this patient was approaching the need for another Botox injection for control of her SD. Therefore, the beneficial effects of her last injection might have been wearing off, contributing to some of these voice-related difficulties.
Figure 3.8: VHI total scores across treatments for Participant 2.

Figure 3.9: VHI subscale scores across treatments for Participant 2.
Acoustic analyses. Due to the participant’s ADSD, she was unable to sustain vowels without glottal fry and exhibited frequent breaks in phonation. As a result, acoustic measurements of these vowels were not possible.

CAPE-V ratings. Mean CAPE-V rating scores across all treatment conditions are provided in Figure 3.10. These high ratings suggest a significant voice disorder, with the perception of vocal tremor playing a more minor role. The only notable change in the ratings was a decline in strain that was noted after the first course of treatment. Unfortunately, this decline in strain was not evidenced at the end of the second treatment. These results are not surprising in that this patient did present with a significant history of SD and the presence of this vocal disorder could be affecting the PI’s voice ratings.

Figure 3.10: CAPE-V mean subscale scores across treatments for Participant 2.
Listener participant ratings of tremor speech samples. Overall, little change was noted in listener ratings for this tremor participant over time (see Appendix D). Ratings were slightly lower for readings of the Rainbow Passage and for conversational speech than for vowel samples. This finding would suggest that the participant’s tremor was less obvious in connected speech, as reflected by improved steadiness and pleasantness ratings for this condition. Ratings for vowels revealed that pleasantness ratings tended to be higher than steadiness ratings, but vowel steadiness ratings seemed to increase as treatment progressed. However, differences across all conditions were within 1.5 increments, suggesting perceptual changes related to conditions were small.

Summary of results for Participant 2. Overall results indicated that the treatment was effective in increasing lung pressure for this participant. However, this participant did not report any benefit in terms of decreased self-perception of vocal handicap, as evidenced by significant increases in total VHI data from baseline to both post-treatment sessions. Acoustic measurements were not possible since the participant could not sustain a vowel. Trained listener CAPE-V ratings suggested a significant voice disorder for this participant, with the perception of vocal tremor playing a more minor role and with inconsistent declines in vocal strain. Finally, listener ratings showed little change in vocal steadiness and pleasantness for this participant over time. Overall, these negative results may have been influenced by the timing of this participant’s upcoming Botox injection, indicating that her ADSD symptoms were increasing. It is also possible that this patient was not an ideal candidate for this treatment given her significant, concomitant spasmodic dysphonia.
**Case 3.** Participant 3 was a 65-year-old female diagnosed with moderately-severe familial tremor of the voice. At the present time, she was not undergoing any voice treatment. She described her voice as being characterized by frequent voice breaks and increased vocal effort and strain in addition to moderately-severe vocal tremor.

*Verification of treatment completion: Lung function data.* Lung function data were graphed for morning and afternoon sessions during treatment (see Figures 3.11 and 3.12). Results revealed that evening lung pressure values were consistently higher than morning lung pressure values throughout the breathing treatment, starting around 1500 ml with performance plateauing between 2000-2500 ml. This patient reported that the breathing exercises were beneficial to her voice and overall quality of life. She planned to continue with them after the experimental period.

![Graph of lung pressure volumes](image)

*Figure 3.11: Initial and highest morning lung pressure volumes of treatment week #1 (1-8) and treatment week #2 (9-14) for Participant 3.*
Figure 3.12: Initial and highest evening lung pressure volumes of treatment week #1 (1-8) and treatment week #2 (9-14) for Participant 3. *Note: Lung pressure volume values are missing as a result of the participant having not circled pressure values for that session on the treatment schedule form.

VHI ratings. Analysis of the VHI data revealed a small decrease in scores from baseline to both post-treatment sessions (see Figures 3.13 and 3.14). There was also little variability on the individual subtest scores. These scores would suggest little change in voice QOL as a result of treatment. It is interesting that this participant’s satisfaction with the breathing treatments was not as notably reflected in changes in her VHI scores, especially in the physical subscale scores where one might expect the most notable, positive change (i.e., a reduction in vocal effort) for a person with vocal tremor.
Figure 3.13: VHI total scores across treatments for Participant 3.

Figure 3.14: VHI subscale scores across treatments for Participant 3.
Acoustic analyses. This participant was unable to sustain vowels due to frequent voice breaks. Thus, acoustic measures for these vowels are not reported.

CAPE-V ratings. Mean CAPE-V ratings indicate that overall severity and tremor were the most obvious aspects of this participant’s vocal disorder (see Figure 3.15). These results, except for vocal roughness, also show improvement that appears to be related to treatment. That is, trained listener ratings of overall severity, strain and tremor were lower (indicating voice improvement) after each week of treatment, with the decline most apparent in the parameter of vocal strain. These ratings substantiate the participant’s perception of vocal improvement after the exercises. It should be noted that the roughness domain displayed a similar pattern as the others; however, the initial roughness score was the lowest.

![Figure 3.15: CAPE-V mean subscale scores across treatments for Participant 3.](image-url)
**Listener participant ratings of tremor speech samples.** Listener ratings of pleasantness and steadiness showed little change over the treatment conditions for this participant. Vowel pleasantness ratings showed a small treatment effect as listener ratings decreased slightly after treatment. In terms of connected speech, steadiness and pleasantness ratings changed very little over treatment time, while steadiness and pleasantness ratings increased only in the no treatment condition for the reading passage, suggesting improvement after treatment #1.

**Summary of results for Participant 3.** An increase was noted in evening lung pressure values for this participant throughout the breathing treatment. Lung volume levels increased from 500-1000 ml over treatment. However, the participant did not experience a statistically significant change in total VHI scores as a result of treatment. As previously stated, acoustic measures could not be obtained for this participant given the severity of her voice disorder. Trained listener CAPE-V ratings were lower for severity, strain and tremor each time after treatment. Untrained listener ratings revealed slightly improved steadiness and pleasantness ratings while reading aloud over the course of treatment. When these results are taken together, they show mixed results with respect to success of the breathing treatment on perceived voice and QOL indicators. However, the patient felt the treatment was beneficial enough to continue after the study ended.

**Summary of Results**

Several different outcome measures were used to determine treatment effectiveness. Physiological, QOL, acoustic, and perceptual data did not triangulate to demonstrate treatment completion and effectiveness. However, individual measures showed some treatment benefit from the breathing exercises. Specifically, physiological
changes in breathing were most evident for Participant 3. While changes in voice QOL were not noted for this participant, significant QOL changes were evidenced by Participant 1. Acoustic measures for Participant 1 revealed a stable F0 across all post-treatment measures. This finding would suggest that there was not a substantial change in laryngeal position after treatment. Only Participant 3 evidenced notable changes in CAPE-V ratings; however the presence of spasmodic dysphonia in Participants 1 and 2 may have been a factor here. Finally, listener ratings revealed little to no change in voice quality for Participants 1 and 2 and only minimal changes in connected speech were noticeable after treatment for participant 3. Nevertheless, the actual difference in the means for the listener ratings across treatment conditions for all participants was so small that they were not deemed to be practically significant. Hence, unfamiliar listeners perceived no dramatic changes in either vocal tremor or in overall vocal quality for any of the participants across treatment conditions. While the data do not strongly support the use of this treatment technique, it should be noted that Participants 1 (in her VHI score) and 3 (by self-report) experienced benefit from this approach.
Chapter 4

Discussion

The purpose of this project was to determine whether the completion of inspiratory breathing exercises in patients with EVT could result in: 1) increased lung volume levels, 2) improved voice QOL as demonstrated by reductions in VHI scores among patients with vocal tremor, 3) acoustic improvements in voice as indications of improvements in vocal fold vibration, and 4) concomitant reductions in trained and untrained listener perceptions of vocal tremor and/or improvements in perceived voice quality. Three women diagnosed with vocal tremor participated in this ABAB single subject design. These women provided baseline measures of vocal performance, completed a basic breathing protocol using a spirometer twice daily for a week. Treatment was discontinued for one week and then reinstituted for a second week. Data were gathered to assess if there were any significant vocal benefits gained by the participants.

Increases in Lung Volume

The patients were compliant in the completion of the inspiratory breathing exercises, as evidenced by their daily schedules of practice. All participants showed larger lung volumes in the evening when compared to the morning values. Participants 2 and 3 showed at least 500-1000 ml improvements in lung volume over the course of treatment. Participant 3 reported that she experienced enough of a benefit from the breathing treatments that she planned to continue with them after the experiment.
Quality of Life and Vocal Tremor

Previous research has indicated that individuals with neurologically-based voice disorders, including EVT, often experience reduced QOL (Cohen et al., 2006; Verdonck-de Leeuw & Mahieu, 2004). These decreases in QOL have been related to significant difficulty initiating and maintaining phonation during conversation, effortful speaking, fewer communication attempts, avoidance of social events, and trouble sustaining relationships with others (Cohen et al., 2006; Verdonck-de Leeuw & Mahieu, 2004). During the pre-treatment session of the study, participants in this study also reported difficulty maintaining phonation, effortful speaking, and avoidance of social situations. It was hypothesized that tremor participant QOL would improve if voice improved.

To answer the second research question, VHI scores pre- and post-treatment (#1 and #2) were compared to note an improvement in voice QOL as a result of participating in the breathing exercises. Results revealed significantly lower VHI scores (i.e., a positive change in voice QOL) for Participant 1 and reported voice and QOL improvements for Participant 3 that were not noticed in the VHI scores. Participant 2 experienced increased VHI ratings after treatment, which would suggest a decreased voice QOL. It should be noted that this participant presented with severe ADSD in addition to EVT, therefore it is difficult to rule out which disorder is contributing most to her reduced QOL or if both disorders are contributors.

According to the World Health Organization’s (WHO; 2001) International Classification of Functioning, Disability, and Health (ICF) levels, these difficulties in speaking could result in a loss in performing important activities (i.e., difficulty speaking
in conversation) and in social losses (i.e., social avoidance behaviors). These behaviors fall within the ICF levels of disability-activity and handicap-participation, respectively. As these behaviors are associated with the above stated disabilities and handicaps, the increases in QOL noted for Participants 1 and 3 suggest that treatment was beneficial, at least for everyday communication. Therefore, the personal benefit that the participants experienced from this treatment suggests that it may be helpful to others with EVT; however, more research is needed.

**Acoustic Results after Inspiratory Breathing Treatment**

The third research question focused on acoustical improvements in voice production that could be related to the treatment. Acoustic measures for two of the participants could not be analyzed, as these participants were not able to sustain vowels at a modal pitch and with frequent voice breaks. Thus, only the acoustic measures from the first participant will be discussed.

Previous research has suggested that the use of high-pitch phonation increases vocal tremor, while low-pitch phonation, possibly associated with a lowered and relaxed larynx, results in stability of the vocal system, and a reduction in the severity of vocal tremor (Dromey et al., 2002). However, post-treatment measures of F0 in Participant 1 appeared to remain stable, suggesting the larynx probably did not lower and that vocal fold movement did not significantly change as a result of treatment. Researchers have also reported significantly higher jitter and shimmer measures and lower harmonics-to-noise ratios with increases in EVT severity (Gamboa et al., 1998). These acoustic measures also were assessed in this study given that they are regarded as associated with the fine changes in laryngeal function (Ramig & Ringel, 1983) that may be apparent after
treatment. However, these measures showed minimal change after treatment in tremor participant 1.

**Perceptual Changes after Inspiratory Breathing Treatment**

The fourth research question focused on the perceptual changes noted by trained and untrained listeners following inspiratory breathing treatment. Untrained listeners noted small differences in steadiness and pleasantness when rating vowels and connected speech samples. However, the actual differences in listener rating means across treatment conditions were so small that they were not deemed to be perceptually meaningful. Therefore, on this perceptual task, untrained listeners did not perceive major differences in the speech of the tremor participants as a result of the breathing treatment.

Several factors may have influenced untrained listener ratings. One factor may have been the nature of the listening task itself. It seems that listeners may have been uncertain what to listen for and as a result their scores, resulting in scores that regressed to the mean. As a score of 1 indicated great vocal steadiness or pleasantness and a measure of 7 indicated poor vocal steadiness or pleasantness, listeners may have been uncomfortable rating either one of those extremes and may have decided to rate the voices toward the middle (a rating of 3 or 4). Thus, listeners may have further benefitted from additional training that included several examples of voices ranging from great to poor vocal steadiness and pleasantness to provide listeners with a reference point that would aid in their decision making on the experimental task.

Another factor that may have affected listener ratings was age related voice changes. Research has shown that as individuals age, their voices may weaken and they experience increases in unsteadiness and in pitch alterations (Hodge, Colton, & Kelley,
2001; Linville & Fisher, 1985; Ramig & Ringel, 1983; Stoicheff, 1981; Verdonck-de Leeuw & Mahieu, 2004). It is also known that changes in the perceptual parameters of pitch, loudness, vocal quality, and voice tremor, can contribute to one’s perception of the aging voice in women (Hollien, 1995; Verdonck-de Leeuw & Mahieu, 2004). So, when the listeners heard the vocal tremor, they may have responded with an aging stereotype (Huntley, 1995). Since naïve listeners can correctly estimate a speaker’s age within ± 5 years of their chronological age (Huntley, Hollien, & Shipp, 1987; Shipp & Hollien, 1969), it is possible that they thought that the voices were from elderly women and that tremor was to be expected in that population.

In contrast to group untrained listener ratings of steadiness and pleasantness, trained listener CAPE-V ratings, conducted by the PI, showed reductions in the areas of overall severity, strain, and tremor for the one participant diagnosed with vocal tremor alone (Participant 3). In this case, the most improvement was noted in the reduction of strain. These ratings were in agreement with overall improvement in voice quality on the CAPE-V ratings, as well as in this participant’s own perception of voice improvement following the exercises. It is interesting to note that for Participant 3, post-treatment lung pressure levels were much higher than all pre-treatment pressure levels throughout the breathing treatment. This suggests that inspiratory breathing treatment may have indeed had a positive effect on voice for this participant. Inspiratory exercises, like those used in this study, have been found to increase patient lung volume (Booker, 2005; Chumillas et al., 1998) through increases in lung pressure volumes. Although larynx position was not directly measured in this study, it is possible that increases in lung (pressure) volume as it
related to increased lung volume may have resulted in a lowered larynx position and, therefore, associated vocal fold vibratory enhancement.

A possible relationship between lung pressure levels and CAPE-V ratings could also be seen for Participant 1 given this participant’s overall lung pressure levels indicated no remarkable increases for lung pressure volumes and CAPE-V ratings changed minimally after treatment for this participant. In contrast, lung pressure volumes for Participant 2 increased during both weeks of treatment although this participant’s CAPE-V scores showed no improvement. For both Participants 1 and 2, having concomitant ADSD may have impacted voice results, preventing notable voice improvement as a result of breathing treatments and/or affecting listeners’ perceptions of voice changes.

The apparent disagreement in listener perceptions between the finding of voice improvements in CAPE-V ratings but not in the untrained listener task of steadiness and pleasantness for participant 3 may have been associated with lack of specific training for the listening task or lack of listener exposure to disordered voices, specifically vocal tremor. For example, Farinella et al (2006) demonstrated that clinically trained listeners could perceive tremulous voicing when slight pressure changes occurred within the respiratory system (Farinella et al., 2006). It is possible that additional training of listeners might have altered their ability to perceive small voice differences across treatment conditions in this study.

**Clinical Implications**

Current voice treatments for EVT focus on compensatory strategies that change speech/voice production in such a way as to lessen perceived vocal tremor (Barkmeier-
Kraemer et al., 2011). These compensatory strategies involve increasing the participant’s articulation rate, decreasing the length of voicing segments, utilizing a breathier, softer voice, and speaking in a slightly elevated pitch to reduce the participant’s tendency to produce lower pitch inflections at the end of utterances. By contrast, the present study focused on achieving positive voice changes through improved physiological changes obtained directly through inspiratory breathing techniques. The hypothesis was that such changes would result in reductions in vocal tremor that would result in perceived improvements in voice and concomitant improvements in participants’ QOL.

The most important similarity between the Barkmeier-Kramer et al. (2011) results and the current study was the patient satisfaction after treatment. However, Barkmeier-Kraemer et al. could not prove significant gains for the participant on their perceptual outcome measures, while the current study did document some improvements in voice quality using CAPE-V ratings for tremor participant 3 (the only one diagnosed with EVT alone). These findings may indicate that the physiologic focus of the present study holds some promise for improved voice QOL in patients with EVT.

**Study Limitations**

A major limitation of this study was its small number of participants. Despite frequent contact with physicians and neurologists, only three individuals were referred to this study. Of the three participants, two were additionally diagnosed with ADSD and were currently undergoing Botox treatment. It is possible that the ADSD and its treatment may have impacted the effects of this breathing treatment, as well as listener ratings of their voices. This difficulty in recruiting participants may be due to the fact an EVT
diagnosis infrequently occurs in isolation and as a result, otolaryngologists and neurologists may have needed more specification on the type of patients requested.

Furthermore, the invalidity of the acoustic measures for Participants 2 and 3 also greatly impacted the study’s results. Since these participants were unable to sustain a vowel due to frequent voice breaks, valid acoustic measures could not be obtained and analyzed for treatment and tremor frequency rates were difficult to count.

The use of a 7-point rating scale also may have impacted study results. It is believed that the listeners were reluctant to use the ends of the rating scale due to their unfamiliarity with rating vocal tremor. This belief was further substantiated by the tendency for listeners to regress to the middle of the scale. This avoidance of the ends of the rating scale may be eliminated by decreasing the 7-point scale to a numerically smaller scale (i.e., a 5-point scale; Kreiman & Garrett, 2000). It is also possible that providing listeners with training on the rating of steadiness and pleasantness may have increased their confidence. Finally, the use of a paired-comparison rating task, where a voice sample from one treatment condition is directly compared to another voice sample, may have simplified the listening task by only requiring a same-different judgment.

An additional limitation of this study was the use of a trained naïve listener (the PI) to complete CAPE-V ratings. Despite being trained to rate voices using the CAPE-V, the PI may not have been able to appropriately rate participant voice quality due to her lack of exposure and diagnostic experience with voice disorders and voice tremor in particular. Furthermore, the possibility of listener bias cannot be dismissed, as the PI was an author in the study and was aware of the session number within the treatment conditions when conducting CAPE-V ratings.
The length of treatment time used in this study (one week) may have also limited the results of this study. A longer treatment application may indicate whether or not the treatment could provide long-term effects and result in stronger and larger treatment effects for QOL and perceptual ratings. Conversely, sampling time may have negated a demonstration of treatment effects. That is, the most voice improvement might occur in the interval immediately following a treatment. Therefore, sampling of the voice and related parameters during this time period may more clearly demonstrate positive treatment effects of this approach.

**Directions for Future Research**

Results of this study do show limited voice improvements for vocal tremor, as seen in Participant 3, the one with tremor only. A future study enrolling patients with EVT only may demonstrate a clearer impact of this approach. One such study may seek to include extended background information on tremor participants, such as previous vocal diagnostic assessments and video laryngostroboscopy studies (VLS) from otolaryngologists. In addition, selecting different parameters, which might more clearly reflect voice improvement and are easier to measure in this patient population, might better demonstrate treatment effectiveness. For example, the number of voice breaks during sustained vowels and patient perceptions of vocal effort level during speech, among others, might show more direct effects of this approach on voice production. As research suggests differing areas along the vocal tract from which the tremor might be originating (Barkmeier-Kraemer & Story, 2010; Farinella et al., 2006; Finnegan et al., 2003; Hachinski et al., 1975; Hixon et al., 1976; Tomada et al., 1987), it would be of great interest to note whether inspiratory breathing exercises may be more specifically
beneficial for those having EVT of pharyngeal, laryngeal, or respiratory origin. Future research tackling this subject is first and foremost limited by the lack of observable and measureable cues by which to identify differences between these proposed origins of vocal tremor. However, research has currently begun to study ways in which these origins may be objectively measured and identified (Barkmeier-Kraemer & Story, 2010).

Future studies should also investigate the possible treatment effects between the use of inspiratory versus expiratory breathing exercises in relation to desired vocal outcomes. Furthermore, the length of time that breathing treatment effects last should also be investigated. These studies may also want to consider the use of another possible measure of QOL than the VHI. It is possible that the participants had completed this measure of voice QOL before, which may bias their self-ratings. Thus, a subjective rating, such as the amount of vocal effort exerted during phonation pre-and post-breathing exercises may be a possible option.

Finally, developing improved ways for listeners to assess voice changes among tremor patients (for example, a paired comparison paradigm, as noted above), as well as selection of listeners (trained versus untrained; naïve versus experienced) may also improve future research in this area. For example, greater effects may be found for CAPE-V ratings if experienced unfamiliar listeners who have had extensive exposure to and experience evaluating disordered voices were used. Listener rating bias may also be reduced if listeners were blinded to the time of the recording session. Finally, future studies may also consider separating the differing types of voice samples (vowel, reading, and conversational samples) while using the CAPE-V, as the current study used ratings that were based upon rating these samples. It would also be helpful to consider additional
aspects of the CAPE-V protocol, such as vocal parameter, consistency versus inconsistency.

**Conclusion**

The purpose of this study was to investigate the effects of inspiratory breathing exercises on voice production, specifically whether these exercises had a beneficial effect on the reduction of vocal tremor as shown in an improvement in lung pressure levels, voice QOL, improved acoustic measures of voice, and listener perceptions of vocal improvement. The primary treatment effects were found in increases in lung pressure volume for Participant 1, decreases in CAPE-V scores for Participant 3, and decreases in VHI scores in Participants 1 and 3. Thus, the results from this study indicate that inspiratory breathing exercises may show some promise in improving voice and QOL in certain tremor patients and warrant further research consideration.
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Appendices
Appendix A: Complete Participant Instructions

Introduction & informed consent
Thank you for coming today and your interest in this study. Here is a copy of the study consent form, which also serves as an abstract for the study. It will give a little bit of background before we get into the study itself.

(Hand participant Consent Form)
A participant report showed that the use of a spirometer after surgery (unrelated to tremor) resulted in reduced severity of their vocal tremor. We went ahead and did some research and found that a breathing treatment may help the larynx lower and reduce muscles tension which may increase vocal dysfunction (i.e. tremor). We want to find out some more about this and see if it really does make a difference. We are unsure if the breathing exercises will make a change in the function of the larynx or if it will make a change in the respiratory system or both. So, in this study we are going to have participants try the breathing exercises as a possible voice tremor treatment for about three weeks. We will take surveys and make recordings of each participant’s voice before, during, and after the breathing treatment in order to see if any changes occurred.
Let’s read through the consent form to give you some more specifics about this experiment on voice tremor.

(Hand participant Informed Consent and read through it orally with participant)
Do you have any questions? (Take time to answer any of the participant’s questions)
(After reading through consent and answering questions, let participant keep copy of consent form and give participant pen & pencil to sign the experimenter’s copy of the form)

VHI
Before we begin with the recordings, we would like to get some insight into how you feel about your voice presently. (Hand VHI and pen & pencil to participant to fill out) This 30-item survey is a list of statements that many people have used to describe their voices and the effects of their voices on their lives. You’ll place a check mark in the response box that indicates how frequently you have the same experience. Please go ahead and fill that now.
(Allow participant 3-5 minutes to complete VHI. Prepare for recordings during this time)
Appendix A (Continued)

Recordings

Now, we are going to begin with the voice recordings. Please come sit in this black chair over here by the computer.

(Wait till participant has come and sat down)
Now I’m going to set you up with this microphone here. (Show headphones to participant and use a wipe to clean it. Point to ear part) This part of the microphone will sit around your ears (Point to microphone part) and this part is the microphone itself. Using a microphone like this allows us to keep the microphone a consistent distance from the mouth which helps with recording. Please go ahead and put your hair behind your ears for me. (Place headphones onto participant from the back)

Rainbow Passage Reading

I would first like to record your voice while reading. Please review this paragraph and when you are ready, let me know and I will have you read it aloud.

(Wait till participant is ready then begin recording)

Conversational Sample

Now, I would like to record your voice during a conversational monologue. I’m going to ask you a question and I would like you talk for about 1 minute. I will nod my head to indicate that I understand what you are saying, but I would like to get at least one minute of you talking alone. Tell me about your last vacation. All right go ahead.

(Wait till participant is ready then begin recording)

Isolated Vowels

Now, I would like for you to sustain some vowel sounds for me. Please take a deep breath and then sustain the vowel sound /i/ for as long as you can. Tell me when you’re ready.

(Wait till participant is ready then begin recording.)
Go ahead and repeat that same vowel sound for me again and tell me when you’re ready. Don’t forget to take a deep breath before you start.

(Wait till participant is ready then begin recording. If either recording time is too short, ask participant to repeat it)
Now please take a deep breath and sustain the vowel sound /a/ for as long as you can. Tell me when you’re ready. Repeat

(Follow same protocol as for /i/)

(Remove headphones, wipe again, and set them down on computer table) All right, that’s it for the recordings. Let’s go back and sit at the table and I’ll explain the breathing exercise treatment. (Return back to table with participant)
Appendix A (Continued)

Breathing Exercise Protocol & Schedule

(Point to spirometer) This is the device we’ll be using for the breathing exercises. It’s called a spirometer. If you’re not familiar with how a spirometer works, you simply breathe in through the mouth piece here (Point to mouthpiece) and this blue cylinder inside the tube here (Point to blue cylinder inside clear tubing) will automatically go up to the pressure level that you generate. The breathing pressure is measured in milliliters, which is abbreviated “ml”. The blue arrow indicators (Point to blue arrow indicators) here on the side of the tube indicate the range where the blue cylinder should reach during normal breathing. The blue indicator here (Point to moveable blue marker) can be manually raised and lowered. As we progress through the treatment, you will adjust this marker upward to make the breathing exercises a little more difficult. Do you have any questions? (Take time to answer any of the participant’s questions)

Now that you’re familiar with the spirometer, let’s read and practice how we’re going to use it in this study. (Hand participant “Cleveland Clinic Foundation Guidelines for Incentive Spirometer Use” directions and read while having participant do each step using the spirometer) Do you have any questions? (Take time to answer any of the participant’s questions)

In order to keep track of your breathing exercise and the pressure levels that you reach, we have a schedule for you to use. (Hand participant Self-Check Schedule Form and read through steps written under the “Directions” portion) Do you have any questions? (Take time to answer any of the participant’s questions)

You will complete the breathing exercises twice a day for 7 days. After 7 days, I will call you to make the voice recordings. Then, you will discontinue the treatment for 1 week, after which I will again call you and make voice recordings. At the completion of this week, you will reinstate the breathing exercises for a final week following the same protocol as the first week. I will again call you and make the final voice recordings and you will mail the materials back to me.

We will make the voice recordings over the telephone (for participants 1 and 2 only). You will not need any special equipment to do this. I will call you and assist you with the voice recordings.

Also, we have a package for you to take home that includes forms that you’ll either need during the remaining recordings or will send back to us.

(Pull out documents from package and go through each item with participant)

Included in the package is 1 copy of the reading passage you read earlier that we’ll need you to read again for us whenever we call you to record your voice again. Also included are 2 copies of the voice survey you filled out earlier. We’ll need you to fill these out again during and after you’ve completed the breathing treatment. But don’t worry, we will be calling you in order to remind you when to do it. The surveys and the schedule form are the only documents that we will need you to send back to us. These surveys are located in smaller envelopes that we have already stamped and self-addressed. So all you simply need to do is fill out the surveys and put them back in their
Appendix A (Continued)

envelope and place it in the mail. However, just be sure to include the schedule form in the last envelope that you send back to us. Do you have any questions? (*Take time to answer any of the participant’s questions*)

**Conclusion**

Before we end, I would like to schedule the first phone call recording with you. We’ll need this day to be during the week of __________. Which day and time would work best for you in order for us to do this recording? (*Have participant decide on a date and time. Write down the appointment on two sheets of paper. Give one copy to the participant and keep one for self*)

Decided Date & Time: __________________________________________

Ok great! Thank you for being willing to participate in our study. We look forward to seeing the results from this study and we will stay in touch with you.
Appendix B:
Cleveland Clinic Foundation Guidelines for Incentive Spirometer Use

The following directions provided below regarding the daily use of the incentive spirometer were modeled after the guidelines developed by the Cleveland Clinic Foundation, 1995-2009.

Directions:

1. Sit on the edge of your bed or chair.
2. Hold the spirometer in an upright position.
3. Place the mouthpiece in your mouth and seal your lips tightly around it.
4. Breathe in slowly and as deeply as possible, raising the blue piston toward the top of the column. The blue coach indicator should be in the blue outlined area.
5. Hold your breath as long as possible (for at least five seconds). Allow the piston to fall to the bottom of the column.
6. Rest for a few seconds and repeat steps one to five at least 10 times in the morning and once in the evening.
7. Position the blue indicator on the left side of the spirometer to show your best effort. Use the indicator as a goal to work toward during each repetition.
   • Repeat steps one through seven everyday for the next week (Week 1).
   Discontinue the exercises for the following week (Week 2) then reinstitute the exercises everyday for following week (Week 3).
Appendix C:
Complete Listener Instructions

Introduction & informed consent
Thank you for coming today and your interest in this study. The purpose of this study is to determine whether specific breathing exercises completed by individuals with vocal tremor will result in a lessening/decrease in their vocal tremor, that is, an improvement in their voice. So in this part of the study, we are having listeners judge the voice quality of audio-recorded voices that consist of the different voices producing short sentences and sustained vowels.

Before we begin, I would like to ask you a few quick questions. What is your age? Have you had any history of speech, language, or hearing impairment? Have you had any experience rating voices? (Record the participant’s answers one after each question. If patient has any history of impairments have them explain and record. Have participant continue but decide later if results will be included in study.)

Ok great! Here is a copy of the study consent form for you to keep that also serves as an abstract about the study that will give a little bit of background before we get into the study itself.

(Hand participant Informed Consent Form and read through it)
Do you have any questions? (Take time to answer any of the participant’s questions)

(After reading through consent and answering questions, give patient pen & pencil to sign experimenter’s copy of informed consent form)

Griffith’s Test
Now, we are going to go ahead and begin with the experiment. Please come sit in this black chair over here by the computer.

(Wait till listener has come and sat down)
Before you listen to the voice recordings, I will first have you do a speech listening task. During this task, you will listen to words heard though the headphones and click on the word that matched the one you heard or thought you heard.

Do you have any questions (Take time to answer any of the listener’s questions)?

Ok, I’m going to set up with these headphones here. (Show headphones to listener and use a wipe to clean it. Place headphones on listener).

Now, let’s get started. (Begin the Griffith’s Test) (One the listener has finished, check the listener’s score and save their results as a text file. Whether he/she passes or fails, allow them to participate in rating the voice recordings, however do not include their results in the study results)
Appendix C (Continued)

Voice Ratings

Practice Task
Ok great! Now, we are going to go ahead and begin with the voice recordings.
As I mentioned before, you will be listening to short sentences and sustained
vowels one at a time. Just rate the voices as you normally would if you heard this voice
in your every day life. If you need to repeat the recording, you are allowed to repeat it
only one time by clicking on the button labeled “Replay Stimulus.”
When you are ready to rate the voice, two 7-point equal-appearing interval scales
will appear. The first 7-point scale will include a from marker labeled “Steadiness” and
the second 7-point scale will include a front marker labeled “Pleasantness.” You will use
the computer mouse to click the number you believe most represents the degree to which
the speaker’s voice was deviant.
Please be sure to rate the voice on both parameters before you go on to the next
voice. Please focus on the voice and not any differences in the quality of the recordings.
Do you have any questions? (Take time to answer any questions)
Ok, let’s first start by having you practice with some sample voices. (Open ECoS
Practice Task. Have listener begin and wait until he/she has completed the practice test)
So do you have any questions about this? (Take time to answer any questions)

Experimental Task
Ok, let’s get started on the experimental part. Again, please focus on the voice
and not any differences in the quality of the recordings. (Open ECoS Experimental Task.
Have listener begin and wait until he/she has completed the entire task. One the listener
has finished, save their results as a text file)

Conclusion
Ok great! We’re all done. Thank you for being willing to participate in our study!
Appendix D:
Graphs of Listener Rating Means

**Figure D1:** Listener-rating means for vowel steadiness across treatments for Participant 1.

**Figure D2:** Listener-rating means for vowel pleasantness across treatments for Participant 1.
Figure D3: Listener-rating means for steadiness of connected speech across treatments for Participant 1.

Figure D4: Listener-rating means for pleasantness of connected speech across treatments for Participant 1.
Appendix D (Continued)

Figure D5: Listener-rating means for vowel steadiness across treatments for Participant 2.

Figure D6: Listener-rating means for vowel pleasantness across treatments for Participant 2.
Appendix D (Continued)

Figure D7: Listener-rating means for steadiness of connected speech across treatments for Participant 2.

Figure D8: Listener-rating means for pleasantness of connected speech across treatments for Participant 2.
Appendix D (Continued)

Figure D9: Listener-rating means for vowel steadiness across treatments for Participant 3.

Figure D10: Listener-rating means for vowel pleasantness across treatments for Participant 3.
Appendix D (Continued)

Figure D11: Listener-rating means for steadiness of connected speech across treatments for Participant 3.

Figure D12: Listener-rating means for pleasantness of connected speech across treatments for Participant 3.