Critical Review:

The Effect of Expiratory Muscle Strength Training (EMST) on Dysphagia in individuals with Idiopathic Parkinson's disease.

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The current critical review investigates the impact of Expiratory Muscle Strength Training (EMST) on Dysphagia in individuals with idiopathic Parkinson's disease, as well as the clinical implications of the effects. The study designs evaluated in this review include two randomized, sham-controlled clinical trial designs, one non-randomized within groups pre-test/post-test design, and one single subject repeated measures design. Results of this critical appraisal yield promising evidence for the use of EMST as a viable treatment for dysphagia in individuals with Parkinson's disease. However due to the limitations of the research evaluated, further research is required before any affirmative clinical applications can be made.

Introduction

Parkinson's disease (PD) is a slow, progressive neurological disease that affects 50 per 100,000 individuals older than 50 years of age (Duffy, 2005). The signs and symptoms of the disease are thought to be due to the loss of nerve cells within the substantia nigra, in addition to the loss of dopamine content within the striatum. The most typical symptoms of the disease include, but aren't limited to; rest tremor, bradykinesia, rigidity, speech difficulties, and a loss of postural reflexes (Duffy, 2005). These symptoms are thought to be a result of an imbalance between acetylcholine and dopamine due to the depletion of dopamine within the striatum (Duffy, 2005).

Respiratory abnormality is another frequent symptom of the disease, and respiratory infection (i.e. pneumonia) is a common cause of death in this population. Abnormalities in respiration include reduced; vital capacity, chest wall movement, respiratory muscle strength and endurance, among others (Duffy, 2005). In addition, decreased voluntary and reflexive cough function has been noted in PD, which may reflect sensory changes in the upper aerodigestive tract (Ebihara, Saito, Kanda et. al., 2003). The reduction in voluntary reflexive cough is further impaired in individuals with PD who show a history of aspiration/penetration of material compared to individuals who do not (Pitts et al 2008).

Dysphagia is also a very common, but not universal, symptom in PD (Michoo and Hamdy, 2010). It has been estimated that throughout the course of the disease, 90% of individuals with PD will develop swallowing disturbances, or dysphagia (Ramig, Fox, & Sapir 2008). Dysphagia is described as any impairment in getting food from the upper aerodigestive tract into the stomach (Logemann, 1998). Individuals with PD can experience

a number of swallowing abnormalities at all three stages of deglutition (Logemann, 1998) such as increased oral transit time; poor bolus formation; residue; premature spillage; delayed triggering of the pharyngeal swallow; reduced laryngeal closure, and increased number of swallows (Bird, Woodward, Gibson, Phyland, & Fonda, 1994; Smith, Roddam, & Sheldrick, 2012). Many of these abnormalities can result in aspiration pneumonia, which is the most common cause of death in individuals with PD (Troche, et al., 2010), and can significantly impact a person's overall well being, health, and quality of life (Sapir, Ramig, & Fox, 2008).

Given the prevalence of respiratory difficulties and swallowing disturbances in this population and their effects on quality of life and morbidity, the proper investigation, treatment, and management of dysphagia in PD are of great importance. Recent literature has begun to investigate the use of Expiratory Muscle Strength Training as new treatment procedure for dysphagia and PD as an alternative to typical compensatory techniques. It has been suggested that using an EMST program may result in new motor unit recruitment and firing rates as well as a general increase in central nervous system activation (Saleem, Sapienza, & Okun's, 2005), thus counter balancing the progressive nature of the disease.

Expiratory Muscle Strength Training (EMST) is a device driven treatment program for Dysphagia consisting of a 4-week controlled protocol in which the muscles of expiration are strengthened. The strengthening of expiratory muscles is hypothesized to produce a functional improvement in cough and swallowing function in individuals with PD. EMST training aims to increase the amount of force an individual generates on expiration, which then can improve the systems ability to clear the airway during a cough and thereby increasing a person's airway

protection capability (Sapienza, Wheeler, 2006). This increase in muscle force generation has been associated with not only increased cough generation, but also increased hyolaryngeal elevation, and therefore increased swallowing safety (Troche et al., 2010). Of particular importance is the potential relationship between expiratory muscle function and dysphagia in individuals with PD.

Objectives

The primary objective of the current paper is to critically evaluate the existing literature related to the effect of Expiratory Muscle Strength Training (EMST) on dysphagia in individuals with PD.

Methods

Search Strategy

The sources for this paper were collected using the following computerized databases: Pubmed, Psych Info, Medline, and the Cumulative Index to Nursing and Allied Health Literature (CINAHL). The following search strategies were employed:

[Expiratory Muscle Strength Training], OR [muscle strength training], OR [EMST], OR [Respiratory Strength Training] AND [Parkinson's Disease], AND [dysphagia], OR [swallowing], OR [aspiration].

Selection Criteria

Inclusion criteria for the selection of research to be included in the current critical review was as follows; the articles had to be written and/or published in English, and they had to involve an investigation of the effects of Expiratory Muscle Strength Training on dysphagia or swallowing disturbances in Parkinson's patients. No limits of publication year, or geographical demographics were placed.

Data Collection and Analysis

The outcome of the literature search yielded 1 qualitative and 3 quantitative studies that matched the aforementioned search criteria, they are as follows; randomized, sham-controlled clinical trials (2), non-randomized, within groups, pre-test/post-test studies (1), and single subject repeated measure studies (1). The relative strength of the evidence presented by each study was determined through the analysis of their research designs according to Archibald (2013) (Adapted from: Oxford Center for Evidence-based Medicine Levels of Evidence (2009), and NHMRC additional levels of evidence and grades for recommendations for developers of guidelines (2009).

Results

Sapienza, Troche, Pitts, & Davenport, (2011) carried out a 4 week randomized clinical trial testing the effects of EMST on increased activation of expiratory muscles and how this increase in activation effected the maximum expiratory pressure (MEP) and pulmonary function in individuals with PD. Sixty participants with idiopathic PD completed EMST for 4 weeks, using a calibrated or a sham, handheld device. The treatment device was calibrated to 75% of the participants average MEP. An individual's average MEP was gathered through obtaining an indirect measure of expiratory muscle strength. Using a nose clip, patients were instructed to take a deep breath, place their lips around the tube-like device, and blow hard into the device. Three measures were taken until the participant's repeated performances were within 5% of each other. These three trials were averaged to obtain the final MEP value. The same techniques were used with the sham treatment group, but since their MEP values rarely changed, the clinician "reset" the device each week in order to maintain participant blindness. The treatment design involved pre-and post treatment measures of each individual's maximum expiratory pressure (MEP) and pulmonary function. All participants were randomly assigned to either the treatment group (n=30) or the sham group (n=30). Both the treatment and the sham device were identical to each other, the only difference between the two was that the resistance of the device in the sham group never increased and always remained at 75% of their original MEP value. All participants completed baseline measures of MEP and pulmonary function, and then completed the 4-week EMST program. Following completion of the program, all participants returned for post-treatment measures of MEP and pulmonary function.

No baseline differences were found between the two groups (p=0.901). Following 4-weeks of EMST, a significant time (pre-test vs post-test) by group (treatment vs sham) interaction was obtained (p < 0.01). In other words, after 4 weeks the active treatment group showed significantly greater change in maximum expiratory pressure (MEP) (p= <0.01) compared to the change in MEP for the Sham group (p= 0.154). No significant main effect of time or treatment group was obtained for the measure of pulmonary function. The authors argue that increased MEP reflects the increased voluntary control of the expiratory muscles, which may be positively related to airway defense mechanisms (Sapienza et. al., 2011). This increase in voluntary control of expiratory muscles is suggested to decrease the risk for aspiration-related pulmonary complications.

This study by Sapienza and colleagues (2011) is judged to represent a level I research design according to Archibald's (2013) system for the evaluation of research designs. The Sapienza et al (2011) study yielded important clinical findings. Following 4-weeks of EMST, individuals with PD showed significantly increased MEP values, and thus voluntary control of expiratory muscles. There are several limitations to current study. First, only individuals with moderate clinical disability were included which is a fairly limited representation of this clinical population. Second, no assessment of swallowing function was performed. Finally, the study population had a very small proportion of females ((m: 47; f: 13) relative to the typical population. This study did however, have many strengths including; a large sample size, pre-post measurements that were obtained during stable medication states, strict inclusion criteria, exclusion of participants with cognitive impairments, and age equivalence across the treatment and sham groups. The results then, provide restrictive and limited support for the use of EMST for dysphagia in individuals with PD, but strong support for EMST on Maximum Expiratory Pressure changes in PD.

Saleem, Sapienza, and Okun's (2005) level I study involved a single subject repeated measures design that examined the effects of 4-weeks of EMST on the generation of maximum expiratory pressure. They hypothesized that 4-weeks of EMST would yield a strengthening of expiratory muscles in the same way that strength training targets weakness of limb muscles in the Parkinson population.

The participant in this study was a 54-year-old female with tremor-dominant idiopathic Parkinson's disease, and disease duration of 5 years. Two baseline measure sessions were carried out one week apart, followed by a 4-week EMST program. Post-treatment measures were completed in the seventh week. Due to self-perceived functional gains the participant requested that the program be extended. The program was extended for an additional 16 weeks of EMST, and post treatment data was collected again following a 4-week period of no training.

Results of the measures obtained at baseline, following training and detraining were plotted using a line graph and visually inspected. No statistical analyses were carried out on any of the measurements. Expiratory muscle strength was indirectly measured using values obtained for MEP. The patients MEP increased by 55% from baseline measures at the 4-week traditional endpoint. Post-treatment measures at the end of the 20-week program yielded a 153% increase in MEP, with a 16% decrease in MEP following 4-weeks of no training,

for a total of 104% increase in MEP from baseline to the 4 week follow-up. Motor improvements were determined through changes in the UPDRS scores. UPDRS scores improved with increased exposure to the program: following 20 weeks of EMST there was a 7-9 point decrease which relates to a 22% improvement. This wasn't observed in the typical 4-week treatment period or the 4-week follow-up period.

The strength of this study is that it allowed for further description of the effects of an extended EMST program on the MEP in an individual with Parkinson's disease. There are however, a number of limitations. The results of the study never underwent any statistical analyses, and the sample size was very restrictive, thereby significantly limiting the generalizability of the results. The findings suggest that EMST increases the strength of the expiration muscles thereby increasing MEP. No connections between MEP and functional cough or swallow outcomes can be made. The results then, provide restrictive and limited support for the use of EMST for dysphagia in individuals with PD, but strong support for EMST and increased cough function in PD.

The study by Pitts, Bolser, Rosenbek, Troche, Okun, & Sapienza, (2009) is judged to represent a level 2a research design according to Archiald's (2013) system for the evaluation of research designs. It used a nonrandomized within groups study investigating whether or not the incidence of penetration/aspiration would improve following a 4-week EMST program for individuals with PD and a history of penetration and aspiration (P/A). A total of 10 male individuals with PD between the ages of 60-82 were included in this study. Only those with mid-stage PD, and as Hoehn and Yahr scores between 2-3 were included. All participants demonstrated videofluroscopic evidence of P/A into laryngeal vestibule, and were screened for any comorbid cognitive impairments. Participants underwent 4-weeks of EMST training. Pitts et. al. (2009) measured pre and post values of airflow parameters produced during voluntary. Parameters measured included: inspiration phase duration (IPD), compression phase duration (CPD), expiratory phase peak flow (EPPF), expiratory phase rise time (EPRT), and cough volume acceleration (Cough VA (EPPF/EPRT)). The degree of P/A was used as the outcome measure for swallow. The authors hypothesized that voluntary cough pattern and P/A scores would improve following 4-weeks of EMST.

During the 4-week program, participants were required to used the device at home 5 days a week, performing 5 sets of 5 breaths through the device for a total of 25 breaths per day. Verbal and written instructions were provided. The resistance on the device was set at 75% of the patient's PEmax (average of 3 measurements of

maximum air pressure generated; an indirect measure of expiratory muscle strength). Results showed that P/A scores decreased significantly following EMST (p = 0.01). Results also yielded significant increases in PEmax scores following training (p=0.005). Changes in the participant's cough parameters were also noted: significant reduction in CPD (p=0.005), and EPRT (p=0.01). As a result of the decrease in EPRT, significant increase in Cough VA (p=0.01) was observed. The significance of cough parameter findings indicates that overall effectiveness of the participants' cough increased post treatment (the ability of the cough to create enough force to expel material from the airway) (Pitts et. al., 2009).

This study by Pitts and colleagues (2009) indicates that following 4-weeks of EMST, individuals with PD showed a significant increase in cough parameters and a significant decrease in P/A scores. The strengths of the current study include strict inclusion criteria, systematic procedure guidelines (written and verbal) for participants to follow at home, and good intrarater reliability (a trained SLP carried out all VFSS assessments). The limitations of the study include a restricted sample size (n=10, all male) and limited diversity (only those with mid-stage PD included) that is not very representative of the disordered population thus hindering generalizability. Limitations aside, the results of this study provide support for the use of EMST for dysphagia in individuals with PD.

The study by Troche, et al., (2010) is judged to represent a level I research design according to Archibald's (2013) system for the evaluation of research designs. It used a randomized, blinded, sham-controlled clinical trial to investigate the outcome of a 4-week device driven EMST program on swallow safety in patients as measured Parkinson's penetration/aspiration (P/A) scale. The researchers hypothesized that by increasing submental pressure; there would be an increase in swallowing safety related to decreases in the instances of P/A. The study involved 60 patients with idiopathic Parkinson's disease. All participants participated in 4-weeks of EMST for 20 min per day, 5 days a week using a calibrated or sham handheld device. Pre and post measures of P/A scores swallow timing, and hyoid movement was obtained using videofluroscopic evidence. Participants were randomly assigned to either the active (n=30) or sham treatment group (n=30). Like other EMST programs, this study used a calibrated, one way, spring-loaded valve to overload submental and expiratory muscles mechanically (Troche et. al., 2010). The sham device was identical to the EMST device except that the spring was removed, thus rendering the pressure release valve nonfunctional. Given that devices looked the same, clinicians and participants were both blinded to treatment group. All participants completed baseline swallowing assessments, 4 weeks of EMST (active or sham), followed by post-treatment assessments.

No differences were obtained at baseline between sham or active treatment groups (p =0.881). Using a repeated measures analysis of variance, a significant interaction between time (pre vs post) and group (active vs sham) was obtained (p= 0.001). Following 4-weeks of EMST, the active treatment group had significantly improved P/A scores. This was not observed in the sham group. No significant effect of age (p= 0.426) and severity level (p = 0.894) on treatment outcome was observed. No statistically significant changes were observed in hyoid elevation following EMST. However, post treatment, hyoid elevation times decreased substantially in the sham-group. The authors argue that this decrease in hyoid elevation in the sham-group may be related to decreased swallowing safety as there exists a minimum time requisite for the coordination of airway protection and bolus propulsion (Troche et. al., 2010). There was however, a significant time (pre vs post) by group (active vs sham) interaction (no values given) for hyoid displacement at different stages of the swallow: UES opening and closure, bolus transit, and laryngeal opening and closure. In other words, displacement of the above increased for individuals in the active treatment group, and decreased in the sham-treatment group. Measures of swallowing-quality of life also showed improvement independent of treatment group (p=0.007).

This study by Troche and colleagues (2010) indicates that participants in the EMST active treatment group yielded significantly reduced P/A scores following EMST. Further they showed that EMST extends the duration of UES opening and closure, and laryngeal closure and opening. Thus, enabling the UES to open wider, and longer, the bolus thus has a better chance of clearing the pyriform sinus, thereby reducing the amount of residue and potential for aspiration post swallow. The strengths of the current study include; a large sample size, double blind (clinician, and participants) design, and good inter-rater reliability. There remain some limitations of the study; restricted generalizability since all participants had mild-moderate dysphagia severity, and did not control for disease severity. The results of this study provide support for the use of EMST for dysphagia in individuals with PD.

Discussion

Based on the results of this critical appraisal, there appears to be moderately-strong evidence that 4 weeks of Expiratory Muscle Strength Training can help to

reduce symptoms of Dysphagia in individuals with Idiopathic PD. Caution must be used however in the interpretation and generalization of the results presented for the following reasons. First, all of the research currently in this area are being or have been carried out by the same group of researchers, therefor yielding very one sided perspectives on the training program. As such new perspectives from various sources are needed. Second, all of the studies presented had internal imitations that restrict generalizability. For instance, the studies carried out by Sapienza, Troche, Pitts, & Davenport, (2011) and Saleem, Sapienza, and Okun's (2005) didn't determine the presence of dysphagia in any of their participants, nor measured the effect of their EMST program on the swallow function of their participants. Rather, they claimed that an increase in the MEP of individuals with PD would result in improved cough and swallow function, but provided no evidence of this. Further, while the research design adopted by Troche et al (2010) was excellent, they failed to control for the stage of disease amongst their participants. While this doesn't negate the effect of an EMST program on dysphagia in PD, it would have been a clinically relevant piece to consider. It would be interesting clinically to know whether EMST would have better/worse effects depending on which stage of the disease a patient is in. Since the stage of disease progression and dysphagia severity were so restrictive in most studies, results are not very representative of the disordered population as a whole, thereby hindering generalizability.

Clinical Implications

Given the limitations outlined above, caution should be taken when applying these findings in our clinical practice until further research is carried out.

Although the results of this critical appraisal yield many limitations, and thus impact the support of using EMST programs on dysphagia in PD, there remain positive clinical considerations. For instance, EMST may represent a functional option for patients in rural areas that is relatively low cost and cuts down on direct therapy time and costs. Specifically, since it is typically used as a home based program it significantly cuts down on need of clinical resources, travel time, and cost for rural families.

Conclusion

There is moderately strong evidence that EMST can improve dysphagia severity in individuals with Parkinson's disease. Further research is still needed in order to make this evidence more clinically sound. In order to provide more compelling evidence for the use

of EMST for treatment of dysphagia in PD, future research should explore the following:

- (1) The relationship between Penetration/Aspiration scores and timing events (e.g. laryngeal elevation) following 4-weeks of EMST. More information is needed on the level at which EMST may be manifesting system changes.
- (2) Examining carry-over following EMST in order to establish that there isn't simply practice effects of the training. It would be important to know whether the program result in a system wide change, or merely a practice effect.
- (3) Exploring the effects of EMST on various degrees of clinical impairment (e.g. various stages of PD and dysphagia severity). It would be clinically relevant to know whether there were different effects of the training depending on a patients severity and stage of disease.
- (4) Exploring the indirect effects of Voice therapy on swallow function in PD. It is clinically important to understand if the outcomes found are really representative of the intervention approach or is it just a function of therapy.
- (5) Implementing Quality of Life measures to research designs testing EMST. Understanding this component is a significant clinical implication that should be explored.
- (6) The use of a standardized evaluation tool for measuring degree of dysphagia or penetration/aspiration (e.g. P/A scores or VFSS data) baseline and outcome measures in individuals with PD.

Until such research has been carried out, caution should be used when applying these findings clinically.

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