Effect of Expiratory Muscle Strength Training on Swallowing and Cough Functions in Patients With Neurological Diseases

A Meta-analysis

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Objective: The aim of this review was to evaluate and summarize the results of published studies exploring the effects of expiratory muscle strength training on swallowing and cough functions in patients with neurological diseases.

Data Sources: The study used Embase, PubMed, and the Cochrane Library as data sources.

Review Methods: Randomized controlled trials or pretest/posttest studies of adults with neurological diseases were included. The data included basic population characteristics, penetration-aspiration scores, peak expiratory flow rate, cough volume acceleration, and maximum expiratory pressure.

Results: Ten studies were included in this meta-analysis. Compared with the control groups, expiratory muscle strength training in patients with neurological diseases significantly reduced the penetration-aspiration scores (risk ratio = −0.94, 95% confidence interval = 1.27 to −0.61, $P < 0.01$) but did not increase the voluntary cough peak expiratory flow rate (risk ratio = 0.57, 95% confidence interval = 0.62 to 1.77, $P = 0.35$), cough volume acceleration (risk ratio = 33.87, 95% confidence interval = 57.11 to 124.85, $P = 0.47$), or maximum expiratory pressure (risk ratio = 14.78, 95% confidence interval = 16.98 to 46.54, $P = 0.36$).

Conclusions: Expiratory muscle strength training might improve swallowing function in patients with neurological diseases. However, conclusive evidence supporting the use of this approach in isolation for improving cough function is unavailable. Additional multicenter, randomized clinical trials performed using reliable and valid cough function outcome measures are required to explore the effects of expiratory muscle strength training on cough function.

What Is Known

- Neurological diseases are often associated with impaired swallowing and the coughing functions. Expiratory muscle strength training (EMST) is an emerging intervention option for individuals with swallowing or coughing impairments that may be related to expiratory muscle weakness.

What Is New

- To date, no meta-analysis has been published examining the effectiveness of EMST in patients with neurological diseases. This study summarized the results of published studies and found that EMST could improve swallowing function in patients with neurological diseases. Therefore, we suggest EMST as one way of achieving improved swallowing function and airway protection in patients with neurological diseases.

Key Words: Expiratory Muscle Strength Training, Breathing Exercises, Cough, Deglutition Disorders, Rehabilitation

(*Am J Phys Med Rehabil* 2019;98:1060–1066)

Expiratory muscle weakness is common in patients with neurological diseases and might be caused by peripheral and central nervous system conditions, such as stroke, Parkinson disease, and multiple sclerosis. The expiratory muscles expel air from the lungs and are of vital importance to swallowing and coughing. The reduced strength and discoordination of the expiratory and submental muscles may lead to swallowing dysfunction.[1] In addition, cough (voluntary or reflex) is the most immediate mechanism protecting against aspiration and aspiration pneumonia. Cough requires the coordinated activation of expiratory, inspiratory, and intrinsic laryngeal muscles.[2] The weakness of the expiratory muscles further reduces the expiratory pressure generation capacities necessary for an effective cough, which may also impair airway protection.[3]

Swallowing and the coughing use similar anatomical structures, and both serve as airway protective behaviors.[4] Impaired swallowing, coughing, and respiratory functions are common in patients with neurological diseases.[5] Stroke impairs respiratory muscle strength and cough flow by approximately 50% compared with individuals without stroke.[2] Dysphagia occurs in 45%–65% of individuals who experience acute stroke.[6] The prevalence of dysphagia in patients with multiple sclerosis is approximately 36%, according to subjective screening tests, and approximately 81%, according to objective tests of swallowing

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This study was funded by Postgraduate Research & Practice Innovation Program of Jiangsu Province, Integrated Care Model For Patients With Dysphagia After Stroke (Grant Number KYCX18_254), and National Key Research and Development Program of China (Grant Number 2017YFC0114302).

The authors read and approved the final manuscript.

Zhuo Wang, Lulu Zhang, and Xueyun Liu are in training.

Financial disclosure statements have been obtained, and no conflicts of interest have been reported by the authors or by any individuals in control of the content of this article.

Supplemental digital content is available for this article. Direct URL citations appear in the printed text and are provided in the HTML and PDF versions of this article on the journal's Web site (www.ajpmr.com).

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ISSN: 0894-9115

DOI: 10.1097/PHM.0000000000001242

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function reported in a meta-analysis.[7] Although a respiratory muscle impairment is often recognized in the later stages of multiple sclerosis, respiratory deficits also emerge in mild cases of multiple sclerosis without obvious respiratory symptoms.[8] Fontana et al.[9] and Ebihara et al.[10] reported a significant decrease in cough function in patients with Parkinson disease, whereas more than 80% of patients with Parkinson disease experience dysphagia during the course of their disease.[11]

Expiratory muscle strength training uses a calibrated, one-way, spring-loaded valve to mechanically overload the expiratory muscles. The valve cuts off the flow of air until a sufficient expiratory pressure is produced. The valve opens and air only begins to flow through the device if the target pressure is maintained.[4] The pressure threshold device is adjusted incrementally to progressively increase the overload.

Expiratory muscle strength training is an emerging intervention option for individuals with swallowing or coughing impairments that may be related to expiratory muscle weakness. Expiratory muscle strength training has been shown to increase active expiratory pressure by increasing the strength of the expiratory muscles for adequate ventilation and coughing. In addition, an increased and prolonged activation of the suprahyoid muscle group occurs during EMST.[12] The suprahyoid muscle leads to the anterior-superior movement of the hyoid bone during the swallowing process and opening of the cricopharyngeal muscle, which facilitates airway protection.[13] However, neurological diseases, including stroke and Parkinson disease, are often associated with the weakness of the suprahyoid muscles, leading to insufficient anterosuperior movement of the hyoid bone and causing problems such as aspiration.[14]

Recently, several studies have been conducted to explore the effect of EMST on swallowing and cough functions in patients with stroke, Parkinson disease, or multiple sclerosis. A prospective, randomized, blinded study of patients with Parkinson disease concluded that EMST improved swallow safety and hyolaryngeal function during swallowing, which is a potentially cost-effective and straightforward therapy for reducing penetration aspiration.[15] However, in a study by Hegland et al.,[16] voluntary cough and swallow measures were not significantly different after training in patients who have experienced a stroke. In addition, an earlier narrative review was performed to explore the benefits of EMST in patients with stroke or Parkinson disease.[17]

To date, no meta-analysis has been published in the literature examining the effectiveness of EMST for swallowing and cough functions in patients with neurological diseases. A synthesis of this information would provide evidence and help inform clinical practice in these populations. The objective of this review was to summarize and evaluate the results of published studies that explored the effects of EMST on swallowing and cough functions in patients with neurological diseases and provide guidance for the rehabilitation of swallowing and cough functions in clinical practice.

METHODS

Literature Search

An electronic search of Embase, PubMed, and the Cochrane Library (Cochrane Database of Systematic Reviews, Cochrane Central Register for Controlled Trials) was completed up to September 2018. MeSH terms combined with free terms were used to search for potentially relevant articles across databases, including "deglutition disorders," "cough," "respiration disorders," "respiratory muscles," "resistance training," and "exhalation." These search strategies are presented in Appendix S1 (Supplemental Digital Content 1, http://links.lww.com/PHM/A815). The references of the identified studies were screened for additional relevant studies. This study conforms to all PRISMA guidelines and reports the required information accordingly (see the Supplemental Checklist, Supplemental Digital Content 2, http://links.lww.com/PHM/A816).

Inclusion and Exclusion Criteria

The studies were screened using the PICOS criteria as follows[18]: (*a*) Participants (P): all of the patients were diagnosed with neurological diseases, such as stroke, Parkinson disease, and multiple sclerosis; (*b*) Intervention (I) and Control (C): EMST was considered the intervention, and treatment without EMST was considered the control; (*c*) Outcomes (O): at least one outcome measure was related to swallowing or cough function; and (*d*) Study design (S): considering the limited number of studies, we reviewed pretest/posttest studies and randomized controlled trials.

Selection of Reviews

The lead author screened retrieved titles and abstracts to identify potentially relevant reviews. The full texts of these studies were then assessed by independent researchers for eligibility. Differences obtained during the selection process were resolved through a discussion.

Quality Assessment and Data Abstraction

All included trials were appraised on the reporting of the following seven key methodological criteria: random sequence generation, allocation concealment, blinding of participants and personnel, blinding of outcome assessment, incomplete outcome data, selective reporting, and other bias. The quality assessment was conducted independently by two researchers after the final selection of the article.[19]

Data were independently extracted by two researchers. Basic information, such as the first author's surname, publication year, country, study design, patient characteristics, and intervention details, was extracted and analyzed. The penetration-aspiration score (PAS), peak expiratory flow rate (PEFR), cough volume acceleration (CVA), and maximum expiratory pressure (P_EMax) were extracted to compare the clinical effects of EMST and the control intervention on swallowing and cough functions. The PAS is a standard method used by both researchers and clinicians to assess penetration, aspiration, and swallowing safety on a 3-oz bolus.[20] The PAS is divided into eight different levels, and a higher level indicates a greater degree of swallowing dysfunction. The PEFR and CVA, which indicate cough strength, were assessed using an oral pneumotachograph connected to a spirometry mouthpiece filter; the measures were obtained when the participant was seated and instructed to "cough hard like something is stuck in your throat."[21]

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Statistical Analysis

Review Manager Software (Version 5.3; Cochrane Collaboration) was used to conduct all analyses. Combined data for the binary outcomes extracted from the original studies were pooled to estimate the risk ratios (RRs) and corresponding 95% confidence intervals (CIs), whereas continuous outcomes were pooled to estimate overall weighted mean differences and corresponding 95% CIs. Heterogeneity was tentatively divided into a lack of and obvious heterogeneity. The I^2 test and χ^2 test were performed to assess heterogeneity. $P < 0.1$ or $I^2 > 50\%$ indicated significant heterogeneity. If significant heterogeneity was observed, a random-effects model was used for the analysis. Otherwise, a fixed-effect model was used. Sensitivity analyses were performed by sequentially excluding each study. Publication bias was analyzed to avoid exaggerating the effect of the overall estimates.

RESULTS

Characteristics of the Included Studies

Five hundred eighty-seven articles were initially obtained from Embase, PubMed, and the Cochrane Library. The PRISMA flow diagram presented in Figure 1 shows the literature selection process. Ten studies were eventually selected for this meta-analysis.[4,15,16,21–27] Seven of the included studies were randomized control trials, and three studies used a single-group pretest/posttest study design. The ten studies included the following 334 participants: 167 patients with stroke, 70 patients with Parkinson disease, 36 patients with multiple sclerosis, and 61 patients with amyotrophic lateral sclerosis. The duration of treatment ranged from 4 to 8 wks between studies. The EMST device was set to various pressures, ranging from 50% to 60% P_EMax or from 50% to 75% maximum expiratory pressure (MEP). In most of the control groups, the placebo device was identical to the EMST device, with no spring loading. Additional relevant characteristics of the included studies are shown in Table 1.

Review Quality

The outcomes of the quality assessment of the studies were input into Review Manager Software (Version 5.3; Cochrane Collaboration) according to the quality assessment judgment criteria (Fig. 2). The risks of bias are assessed as low, high, and unclear risks. Most of the studies had a low risk of bias for random sequence generation, allocation concealment, blinding of participants and personnel, and blinding of outcome data. However, three studies using a single-group pretest/posttest study design had a high risk of bias for random sequence generation, allocation concealment, and blinding of participants and personnel. All studies had a low risk of bias for incomplete outcome data, selective reporting, and other sources of bias.

Sensitivity Analysis and Publication Bias

A sensitivity analysis was performed by omitting each study to ensure the accuracy of the results. The results showed a stable effect on the PAS. No obvious publication bias was observed in a visual examination of the funnel plots. However, because of limitations of the Review Manager Software, this visual assessment of publication bias lacked reliability.

Effect of EMST on Swallowing Function

Five studies reported the PAS as an outcome. Compared with the control groups, patients in the EMST groups showed

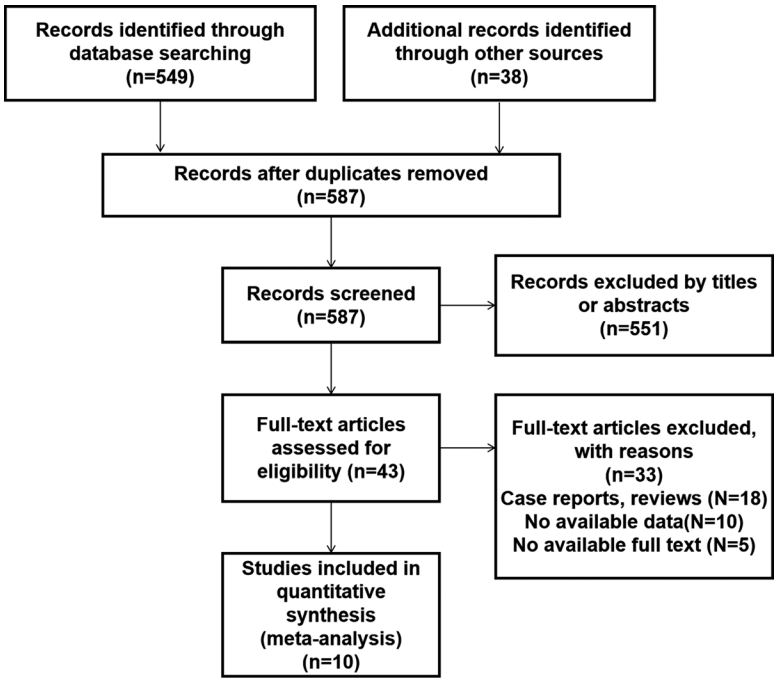


FIGURE 1. The PRISMA flow diagram of the review process.

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TABLE 1. Characteristics of the studies included in the review

Author	Study Design	Patient Characteristics	Intervention (Description, Frequency and Duration, Resistance/Load)	Control (Description, Resistance/ Load [%MEP])	Outcomes
Pitts et al.[4] (2009)	Before-after study	10 patients Parkinson disease	EMST 5 sets × 5, breaths = 25 per day 5 d/wk for 4 wks 75% MEP		PAS, P_EMax, EPRT, EPPF, CVA
Troche et al.[15] (2010)	RCT	60 patients Parkinson disease	EMST 5 sets × 5, breaths = 25 per day 5 times a week for 4 wks 75% MEP	Sham-EMST	PAS, hyoid movement
Kulnik et al.[26] (2015)	RCT	82 patients Stroke	Expiratory training 5 sets of 10 breaths 4 wks of daily training 50% P_EMax	Sham training 10% P_EMax	P_EMax, PECF, Pneumonia
Park et al.[22] (2016)	RCT	27 patients Stroke	EMST 5 sets × 5, breaths = 25 per day 5 d/wk for 4 wks 75% MEP	Sham-EMST device No spring loading	Activity of sEMG, PAS, FOIS
Hegland et al.[16] (2016)	Before-after study	14 patients Stroke	EMST 5 sets × 5, breaths = 25 per day 5 d/wk for 4 wks 60% P_EMax		MBSImP, PAS, PEMax, FVC, FEV1, PEFRT, PEFR, Cough expired volume, CVA, UtC
Silverman et al.[23] (2016)	RCT	36 patients Multiple sclerosis	EMST 5 sets × 5, breaths = 25 per day 5 d/wk for 4 wks 75% MEP	Sham-EMST device Without an internal pressure threshold spring	MEP, PAS, SWAL-QOL
Plowman et al.[24] (2016)	Before-after study	15 patients Amyotrophic lateral sclerosis	EMST 5 sets × 5, breaths = 25 per day 5 d/wk for 5 wks 50% MEP		MEP, maximum hyoid displacement, ALSFRS-R
Moon et al.[25] (2017)	RCT	18 patients Acute stroke	EMST Seven trainings per session 5 times a week for 4 wks 70% MEP	Sham-EMST	FDS, PAS, VR, PR
Eom et al.[27] (2017)	RCT	26 patients Stroke	EMST 5 sets × 5, breaths = 25 per day 5 d/wk for 4 wks 70% MEP	Sham-EMST No loading device	VDS, PAS
Plowman et al.[21] (2018)	RCT	46 patients Amyotrophic lateral sclerosis	EMST 5 sets × 5, breaths = 25 per day 5 d/wk for 5 wks 50% MEP	Sham-EMST No physiologic load	MEP, VFSS, DIGEST, PAS, FOIS, EAT-10, FVC, ALSFRS-R

ALSFRS-R, Amyotrophic Lateral Sclerosis Rating Scale-Revised; DIGEST, The validated Dynamic Imaging Grade of Swallowing Toxicity; EAT-10, the Eating Assessment Tool 10; EPPF, expiratory phase peak flow; EPRT, expiratory phase rise time; FDS, functional dysphagia scale; FEV1, forced expiratory volume in the first second; FOIS, Functional Oral Intake Scale; FVC, forced vital capacity; MBSImP, Modified Barium Swallow Impairment Profile; PECF, peak expiratory cough flow of voluntary cough; PEFRT, peak expiratory flow rise time; PR, pyriform sinuses residue; sEMG, suprahyoid muscles; SWAL-QOL, Swallowing Quality of Life Questionnaire; UtC, urge to cough; VDS, Videofluoroscopic Dysphagia Scale; VFSS, videofluoroscopic swallowing study; VR, vallecular residue.

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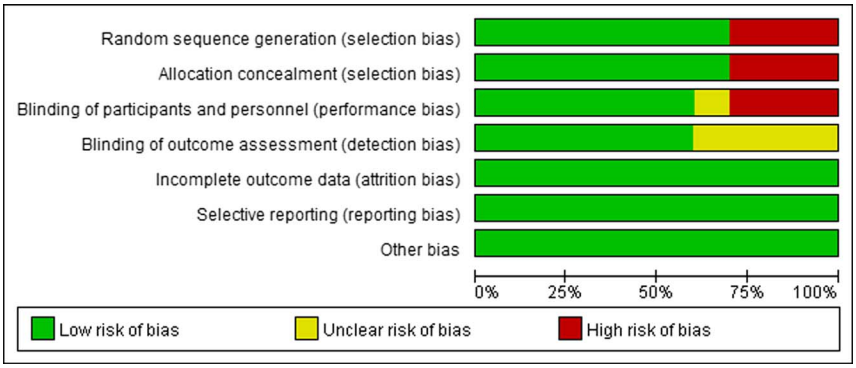


FIGURE 2. Risk of bias graph: review author's judgements about each domain presented as percentages across the included studies.

a significant reduction in the PAS (RR = −0.94, 95% CI = −1.27 to −0.61, $P < 0.01$, Fig. 3). Two studies reported the number of patients with unsafe swallowing according to the PAS. However, a significant difference was not observed between the control and intervention groups (RR = 0.61, 95% CI = 0.19–1.94, $P = 0.40$, Fig. 3).

In addition, Silverman et al.[23] did not observe a difference in overall swallow-related quality of life, but the EMST group experienced a significantly greater increase in scores for the burden and pharyngeal swallow domains than the control group. Another study of patients with stroke observed significant improvements in scores on the functional dysphagia scale and vallecular residue in the intervention group.[25] Eom et al.[27] also discovered that the intervention group showed greater improvements in the pharyngeal phase of the Videofluoroscopic Dysphagia Scale. However, Park et al.[22] did not observe statistically significant differences in scores on the Functional Oral Intake Scale between groups. Another prospective double-blind, randomized, placebo-controlled clinical trial of patients with amyotrophic lateral sclerosis did not identify group differences in the Dynamic Imaging Grade of Swallowing Toxicity scale or in the patient-reported swallowing outcome (Eating Assessment Tool 10).[21] Therefore, because of the use of diverse outcome measures, we were unable to perform a meta-analysis.

Effect of EMST on Cough Function

Four studies were included in the analysis of cough function, with two examining the voluntary cough PEFR and two examining the cough CVA. The meta-analysis of these studies failed to observe any impact on the voluntary cough PEFR or CVA (RR = 0.57, 95% CI = 0.62–1.77, $P = 0.35$; RR = 33.87, 95% CI = −57.11 to 124.85, $P = 0.47$, respectively). However, high heterogeneity was present between these trials ($P_{\text{heterogeneity}} = 0.11$, $I^2 = 60\%$; $P_{\text{heterogeneity}} < 0.01$, $I^2 = 90\%$, respectively). See Figure 4 for additional details.

Kulnik et al.[26] did not observe a significant effect of the intervention on peak expiratory cough flow between the placebo and intervention groups. Another prospective pre-post intervention trial revealed that cough urgency and cough effectiveness both increased for reflex cough but did not increase for voluntary cough.[16] A significant effect of training on expiratory phase peak flow was not observed in patients with Parkinson disease.[4] We were unable to perform a meta-analysis because of the limited number of studies and use of diverse outcome measures.

Effect of EMST on P_EMax

A meta-analysis of 80 patients from two studies did not reveal a significant increase in P_EMax between the control and

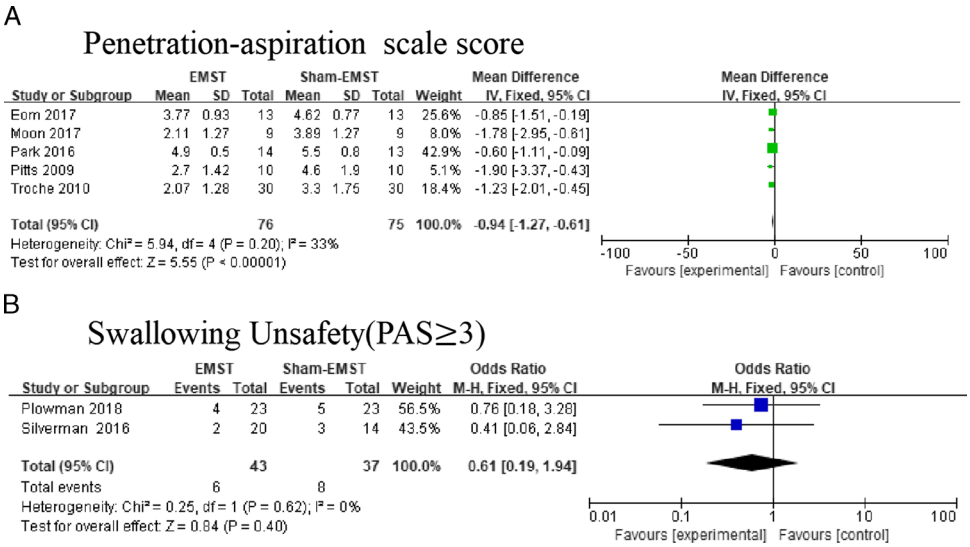


FIGURE 3. Effect of EMST on swallowing function.

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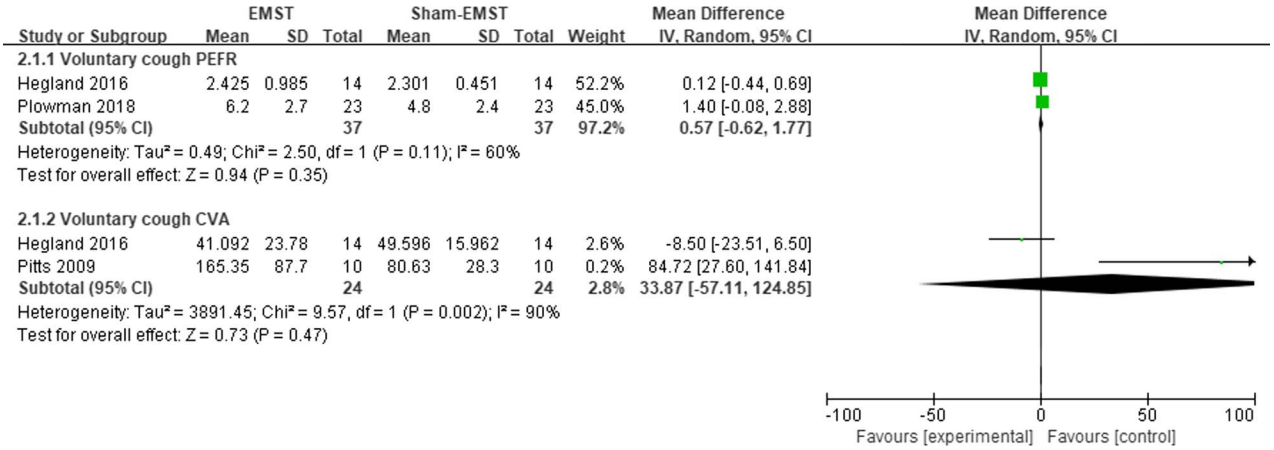


FIGURE 4. Effects of EMST on voluntary cough PEFR and CVA.

intervention groups (RR = 14.78, 95% CI = −16.98 to 46.54, $P = 0.36$). Figure 5 shows a forest plot comparing the control groups and intervention groups.

DISCUSSION

To our knowledge, no meta-analysis has been conducted to determine the effectiveness of EMST on swallowing and cough functions in patients with neurological diseases. Based on the results from this review, EMST may be effective at improving swallowing function, which potentially has important implications for the management of dysphagia in patients with neurological diseases. Because EMST is an indirect measurement method that does not use the direct use of food, it is useful for assessing swallowing function in patients with neurological diseases. In addition, EMST is relatively inexpensive, portable, and easy to use, requiring no assistance from specialists.

Expiratory muscle strength training decreased the PAS in patients with neurological diseases, which indicates an improvement in swallowing function. Similar results were obtained in previous studies. Several factors, including muscular and neural effects, potentially explain this result. First, EMST promotes the activation of suprahyoid muscles, which are important in swallowing to elevate the hyolaryngeal complex and open the upper esophageal sphincter.[28] A recent study of patients with subacute stroke showed markedly greater activity of the suprahyoid muscle group in the EMST group than in the control group.[22] Second, EMST also facilitates improvements in the submental, palatal, lingual, velopharyngeal, and pharyngeal musculature, the coordination of which is important for bolus transport during swallowing, the maintenance of swallowing safety and efficiency, or functional improvements.[28] According to Yanagisawa et al.,[29] expiratory resistive loading due to EMST activates the orofacial muscles, such as the orbicularis oris muscle. Third, afferent stimulation of sensory receptors of the tongue and oropharynx may increase the activity of the swallowing center in the medulla oblongata of the brainstem.[30] Therefore, EMST is involved in both neural and muscle group adaptations, contributing to improvements in the swallowing function.[31]

This meta-analysis failed to identify any effects on the voluntary cough PEFR or CVA. In addition, a meta-analysis of reflex cough or other cough function measures was unable to be performed because of the limited number of studies. Pitts et al.[4] postulated that the inclusion of 80% of participants with Parkinson disease presenting with restrictive lung disease in their study contributed to the result that EMST did not improve voluntary cough PEFR or CVA. The positive results for the PAS also indicated improved cough function, whereas strong voluntary coughing reduces penetration and aspiration by removing foreign materials, even when food flows into the airway.[32] This finding might be attributed to the nature of EMST, which requires a short duration and isometric contraction of expiratory muscles to generate the maximum pressure. Therefore, the effect of EMST on cough function remains unclear, and conclusive evidence supporting the use of this approach in isolation for improving cough function is unavailable. Further research is needed to identify which populations benefit from EMST.

Limitations

This review included both pretest/posttest studies and randomized controlled studies, limiting our ability to determine a causal relationship. The small sample sizes of the included studies may also have contributed to the insufficient statistical power, increasing the risk of type II errors. Additional randomized trials with larger sample sizes are needed to minimize the risk of type II errors in statistical analyses. Moreover, this systematic review included only peer-reviewed, published research written

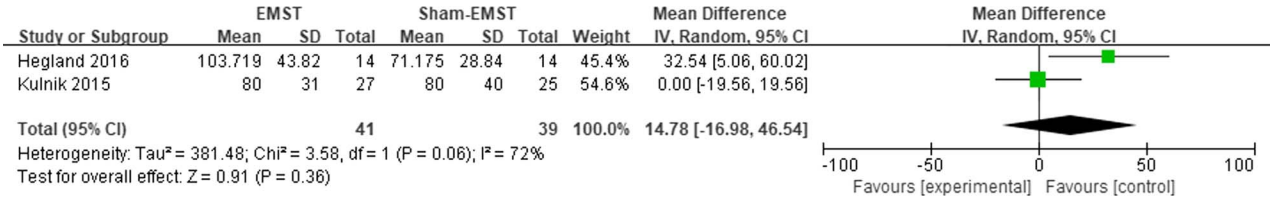


FIGURE 5. Effect of EMST on P_EMax.

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in English, although a sensitivity analysis was performed to minimize publication bias. Studies with inferior accuracy and quality may lead to a greater risk of publication bias than thoroughly peer-reviewed publications. Furthermore, a meta-analysis of cough function, such as peak expiratory cough flow and reflex cough, was not performed because of the use of different outcome measures in the included studies. Overall, a paucity of studies have investigated the effects of EMST on patients with neurological diseases. If the limitations of this study are addressed in the future, the effects of EMST may be more clearly ascertained.

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