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Safety and performance of oropharyngeal muscle strength training in the treatment of post-stroke dysphagia during oral feeding: protocol for a systematic review and meta-analysis

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ABSTRACT

Introduction Dysphagia is a common functional disorder after stroke. Most patients post-stroke are incapable of oral feeding, which often leads to complications such as malnutrition, aspiration pneumonia and dehydration that seriously affect the quality of life of patients. Oropharyngeal muscle strength training is a major method of swallowing training, and recent studies have focused on healthy adults, elderly persons, and patients with head and neck cancer or neurodegenerative diseases; but there have been few studies on such training in patients with post-stroke dysphagia. Our study aims to systematically review the safety and performance of oropharyngeal muscle strength training in the treatment of post-stroke dysphagia during oral feeding.

Methods and analysis The Cochrane Library, Web of Science, PubMed, Embase and ClinicalTrials.gov databases will be systematically searched, and all relevant articles in English from the establishment of the databases to January 2022 will be reviewed. The study will be conducted in accordance with the recommendations of the Cochrane Handbook for Systematic Reviews of Interventions and will be reported in accordance with Preferred Reporting Items for Systematic Reviews and Meta Analyses guidelines. The primary outcome measures include the Penetration–Aspiration Scale and the Functional Oral Intake Scale. Two authors will independently screen the articles, extract the data and assess the study quality. Any disagreements during this process will be resolved by discussion or by consultation with a third author. Next, quantitative or qualitative, subgroup and sensitivity analyses of the included literature data will be performed as appropriate.

Ethics and dissemination Ethical approval is not required for this systematic review as no primary data collection will be required. The results of the present study will be published in a peer-reviewed journal in the field of deglutition disorders.

PROSPERO registration number CRD42022302471.

STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ The study will be conducted in accordance with the recommendations of the Cochrane Handbook for Systematic Reviews of Interventions and will be reported in accordance with Preferred Reporting Items for Systematic Reviews and Meta Analyses guidelines.
⇒ Five key databases will be searched: the Cochrane Library, Web of Science, PubMed, Embase and ClinicalTrials.gov.
⇒ Two reviewers will independently complete the study screening, selection, data extraction and quality rating, and any disagreements will be resolved via discussions or consultations with a third author.
⇒ Different types, intensity, frequency and time of oropharyngeal muscle strength training may lead to a large degree of heterogeneity; subgroup analyses of the effects of the onset time, stage and type of the stroke on the training outcome of people post-stroke will also be carried out.
⇒ Only studies published in English will be included.

INTRODUCTION

Approximately 13.7 million people worldwide suffer from stroke annually,[1] and nearly half of these people experience varying degrees of functional impairment that seriously affect their quality of life.[2 3] Dysphagia is a notably common functional impairment in people post-stroke, with a reported incidence of up to 80%.[4] Patients with dysphagia are often incapable of oral feeding, resulting in complications such as malnutrition, dehydration, aspiration pneumonia and prolonged hospitalisation, in addition to increased hospital costs. Although most patients spontaneously recover swallowing

function, 11%–50% of patients still have dysphagia 6 months after stroke.[5]

The primary purpose of swallowing treatment is to reduce the incidence of aspiration and increase the level of oral feeding. Traditional dysphagia treatment includes oral facial massage,[6] thermal tactile stimulation[7] and various compensatory methods.[8 9] Although they can help patients begin to attempt oral feeding promptly, they do not facilitate the recovery of neural networks in regions of the cerebral cortex associated with swallowing, and training efficacy is short-lived.[10–12] Modalities for improving oropharyngeal muscle strength such as tongue-to-palate resistance training (TPRT),[13] Shaker exercise,[14] Iowa Oral Performance Instrument (IOPI),[15] tongue-strengthening exercises (TSEs)[16] and chin tuck against resistance (CTAR) exercise[17] use isometric or isokinetic muscle contractions to improve the strength and endurance of swallowing-related muscles. Studies involving healthy adults and older adults have shown that maximum tongue pressure[18] and suprahyoid muscle strength[19] are significantly improved after oropharyngeal muscle strength training, but there have been few studies on such training in patients with post-stroke dysphagia.

Thus, our study is a comprehensive systematic review that aims to address the following questions through evidence-based medicine: (1) Compared with conventional swallowing treatment, what is the effect of oropharyngeal muscle strength training on the swallowing process in patients with post-stroke dysphagia with respect to (a) safety of swallowing, evaluated with Penetration–Aspiration Scale (PAS), and (b) performance of swallowing, evaluated with Functional Oral Intake Scale (FOIS)?; (2) Does training efficacy differ depending on the type, time, frequency or intensity of training?; (3) Does the training efficacy depend on the onset time, stage and type of the stroke?

METHODS AND ANALYSIS
Study registration
This protocol was reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Protocols (PRISMA-P)[20] (see online supplemental appendix 1). The final study will be conducted in accordance with the recommendations of the Cochrane Handbook for Systematic Reviews of Interventions[21] and will be reported in accordance with PRISMA 2020 checklist.[22]

Inclusion criteria for studies
Studies will be included in this systematic review if they meet the following criteria:

Participants
The participants will be initial-onset stroke aged 18 years or above, with oropharyngeal dysphagia diagnosed after videofluoroscopic swallowing study (VFSS), flexible endoscopic evaluation of swallowing or clinical evaluation. Those with a history of oropharyngeal cancer or head and neck trauma or surgery prior to onset will be excluded. There will be no restrictions on race, nationality or sex of the participants.

Intervention
Interventions will include methods of oropharyngeal muscle strength training, including Shaker exercise, CTAR, IOPI, TPRT and TSE. Considered studies will include at least one of any of these methods, with no restrictions on time, frequency or intensity of training.

Comparison
Control groups in selected studies would have undergone conventional swallowing treatment, including orofacial muscle exercises, thermal tactile stimulation, and therapeutic or compensatory manoeuvres, or placebo treatment (defined as using the same equipment as the intervention, but without resistance).

Outcome measures
The primary outcome measures include (1) safety of swallowing evaluated with PAS[23] through VFSS to compare the changes in aspiration before and after intervention; and (2) performance of swallowing assessed with FOIS[24] to compare the changes in oral intake levels before and after intervention. The PAS is the primary tool to quantify swallowing safety. It is an 8-point scale used to characterise the depth and response to larynx passage and airway invasion during VFSS. A PAS score of 3 is frequently used as the cut-off for unsafe swallowing. The FOIS is a 7-point ordinal scale that documents the functional level of oral intake of food and liquid. It is a reliable and validated outcome parameter, to measure adequate oral intake.

Secondary outcome measures include (1) severity of dysphagia assessed through Functional Dysphagia Scale,[25] Videofluoroscopic Dysphagia Scale,[26] Dysphagia Outcome and Severity Scale[27] and Dysphagia Rating Scale[28]; (2) swallowing biomechanics with changes in maximal excursion of the hyoid, muscle strength of suprahyoid muscles and tongue pressure; (3) swallow-related quality of life assessed through Swallow Quality of Life Questionnaire.[29]

Study design and language
The systematic review will only include randomised controlled trials published in English. Case–control studies, cohort studies, case reports or other studies that do not provide data for analysis will be excluded.

Search strategy
The Cochrane Library, Web of Science, PubMed and Embase databases will be searched for relevant studies from inception until January 2022, in addition to ClinicalTrials.gov (www.clinicaltrials.gov). Key search terms include the following: "deglutition disorders," "dysphagia," "shaker exercise," "head lift exercise," "chin tuck against resistance," "Iowa oral performance instrument," "isometric lingual exercise," "tongue-to-palate resistance training," "tongue-strengthening exercises."

For a detailed search strategy, see online supplemental appendix 2.

Data collection
Study selection
EndNote (V.X9) software will be used to store and manage all articles retrieved from the databases. First, duplicate documents will be identified and eliminated through the duplication checking function of EndNote, and then an author will manually identify and eliminate them. After eliminating duplicates, two authors will independently screen the titles and abstracts of the articles; those that did not meet the inclusion criteria will be excluded. After preliminary screening, two authors will independently conduct a detailed full-text review of potentially eligible articles, exclude articles that do not meet the inclusion criteria and record the reasons for the exclusion, and finally determine the studies to be included in the systematic review. During this process, any disagreements between the two authors on whether an article should be included will be resolved by discussion or consultation with the third author.

Data extraction and management
Two authors will independently extract relevant study data from the included studies using a data extraction form (see online supplemental appendix 3). The extracted data will then be subjected to a final review by the third author, and any discrepancies will be resolved through discussion or consultation. The extracted data will include the basic information of the study, research methods, characteristics of the research participants (demographic information, inclusion and exclusion criteria, baseline characteristics, sample size, etc), interventional measures (type, time, frequency, and intensity of training in the experimental and control groups), outcome measurement (definitions, methods of measurement, different time points, etc) and results (primary outcome measures, secondary outcome measures, losses to follow-up, missing data, etc).

Risk of bias assessment
The risk of bias of the included studies will be independently assessed by two authors using the Cochrane risk-of-bias tool,[30] which covers seven domains of bias, including random sequence generation (selection bias), allocation concealment (selection bias), blinding of participants and personnel (performance bias), blinding of outcome assessment (detection bias), incomplete outcome data (attrition bias), selective reporting (reporting bias) and other biases. Each domain is judged as 'high risk', 'low risk' or 'unclear risk'. During this process, disagreements will be resolved through discussion or consultation with the third author.

Data analysis and synthesis
Cochrane Review Manager software (V.5.3, The Cochrane Collaboration, London, UK) will be used in this review for the meta-analysis. If enough studies meet the inclusion criteria and have the same outcome, we will perform a meta-analysis of the primary and secondary outcome measures. If a given outcome measure has data from only one study, the results will be reported narratively.

Treatment efficacy will be measured using risk ratio and 95% CI for dichotomous data, mean difference or standardised mean difference and 95% CI for continuous data.

A low degree of overlap in the CI of the included studies will indicate heterogeneity. The χ^2 test and the I^2 statistic will be used for the analysis.[31] If $p > 0.1$ and $I^2 < 50\%$, a fixed-effects model will be used; if $p > 0.1$ and $I^2 \geq 50\%$, a random-effects model will be used. If $p \leq 0.1$, the heterogeneity is large and will be considered statistically significant, and subgroup analysis will be used to investigate the reasons for the heterogeneity. Subgroup analysis will investigate factors such as the type, intensity, frequency, and time of oropharyngeal muscle strength training, and the onset time, stage and type of stroke. If necessary, sensitivity analysis of the primary outcome measure will be used to investigate the effect of bias on the study results.

If the number of articles included in this study is sufficiently large (n>10), funnel plot analysis will be used to assess potential publication bias.

This study will assess five factors determining the quality of evidence (study limitations, inconsistency of results, indirectness of evidence, imprecision and reporting bias) in accordance with the Grading of Recommendations Assessment, Development and Evaluation system.[32] This system classifies the quality of evidence into four levels (very low, low, moderate or high). The evaluation process will be performed independently by two authors, and any disagreements will be resolved by discussion or consultation with the third author.

Patient and public involvement
Patients and the public will not be directly involved in the study. All data collected in this study will be derived from published data in databases or clinical trial registries.

ETHICS AND DISSEMINATION
Ethical approval is not required for this systematic review as no primary data collection is required. The results of the present study will be published in a peer-reviewed journal in the field of deglutition disorders.

Competing interests None declared.

Patient and public involvement Patients and/or the public were not involved in the design, or conduct, or reporting, or dissemination plans of this research.

Patient consent for publication Not required.

Provenance and peer review Not commissioned; externally peer reviewed.

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REFERENCES

 1 Mendelson SJ, Prabhakaran S. Diagnosis and management of transient ischemic attack and acute ischemic stroke: a review. *JAMA* 2021;325:1088–98.
 2 EFFECTS Trial Collaboration. Safety and efficacy of fluoxetine on functional recovery after acute stroke (effects): a randomised, double-blind, placebo-controlled trial. *Lancet Neurol* 2020;19:661–9.
 3 Yang P, Zhang Y, Zhang L, *et al*. Endovascular thrombectomy with or without intravenous alteplase in acute stroke. *N Engl J Med* 2020;382:1981–93.
 4 Dziewas R, Stellato R, van der Tweel I, *et al*. Pharyngeal electrical stimulation for early decannulation in tracheotomised patients with neurogenic dysphagia after stroke (PHAST-TRAC): a prospective, single-blinded, randomised trial. *Lancet Neurol* 2018;17:849–59.
 5 Cohen DL, Roffe C, Beavan J, *et al*. Post-Stroke dysphagia: a review and design considerations for future trials. *Int J Stroke* 2016;11:399–411.
 6 Zhong L, Rao J, Wang J, *et al*. Repetitive transcranial magnetic stimulation at different sites for dysphagia after stroke: a randomized, Observer-Blind clinical trial. *Front Neurol* 2021;12:625–83.
 7 Cheng I, Sasegbon A, Hamdy S. A systematic review and meta-analysis of the effects of intraoral treatments for neurogenic oropharyngeal dysphagia. *J Oral Rehabil* 2022;49:92–102.
 8 Benjapornlert P, Kagaya H, Inamoto Y, *et al*. The effect of reclining position on swallowing function in stroke patients with dysphagia. *J Oral Rehabil* 2020;47:1120–8.
 9 Rodd BG, Tas AA, Taylor KDA. Dysphagia, texture modification, the elderly and micronutrient deficiency: a review. *Crit Rev Food Sci Nutr* 2021:1–17.
10 Nakao Y, Yamashita T, Honda K, *et al*. Association among age-related tongue muscle abnormality, tongue pressure, and Presbyphagia: a 3D MRI study. *Dysphagia* 2021;36:483–91.
11 Chang K-V, Wu W-T, Chen L-R, *et al*. Suboptimal tongue pressure is associated with risk of malnutrition in community-dwelling older individuals. *Nutrients* 2021;13. doi:10.3390/nu13061821. [Epub ahead of print: 27 May 2021].
12 Minagi Y, Ono T, Hori K, *et al*. Relationships between dysphagia and tongue pressure during swallowing in Parkinson's disease patients. *J Oral Rehabil* 2018;45:459–66.
13 Lin C-H, Chung S-Y, Lin C-T, *et al*. Effect of tongue-to-palate resistance training on tongue strength in healthy adults. *Auris Nasus Larynx* 2021;48:116–23.
14 Kagaya H, Inamoto Y. Possible rehabilitation procedures to treat sarcopenic dysphagia. *Nutrients* 2022;14. doi:10.3390/nu14040778. [Epub ahead of print: 12 Feb 2022].
15 Oh J-C, . Effects of effortful swallowing exercise with progressive anterior tongue press using Iowa oral performance instrument (IOPI) on the strength of swallowing-related muscles in the elderly: a preliminary study. *Dysphagia* 2022;37:158–67.
16 Fukuoka T, Ono T, Hori K, *et al*. Effects of Tongue-Strengthening exercise on tongue strength and effortful swallowing pressure in young healthy adults: a pilot study. *J Speech Lang Hear Res* 2022;65:1686–96.
17 Chang MC, Park S, Cho JY, *et al*. Comparison of three different types of exercises for selective contractions of supra- and infrahyoid muscles. *Sci Rep* 2021;11:7131.
18 McKenna VS, Zhang B, Haines MB, *et al*. A systematic review of isometric lingual Strength-Training programs in adults with and without dysphagia. *Am J Speech Lang Pathol* 2017;26:524–39.
19 Park J-S, Hwang N-K. Chin tuck against resistance exercise for dysphagia rehabilitation: a systematic review. *J Oral Rehabil* 2021;48:968–77.
20 Shamseer L, Moher D, Clarke M, *et al*. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. *BMJ* 2015;350:g7647.
21 Cumpston M, Li T, Page MJ, *et al*. Updated guidance for trusted systematic reviews: a new edition of the Cochrane Handbook for systematic reviews of interventions. *Cochrane Database Syst Rev* 2019;10:Ed000142.
22 Page MJ, Moher D, Bossuyt PM, *et al*. PRISMA 2020 explanation and elaboration: updated guidance and exemplars for reporting systematic reviews. *BMJ* 2021;372:n160.
23 Galovic M, Stauber AJ, Leisi N, *et al*. Development and validation of a prognostic model of swallowing recovery and enteral tube feeding after ischemic stroke. *JAMA Neurol* 2019;76:561–70.
24 Hongo T, Yamamoto R, Liu K, *et al*. Association between timing of speech and language therapy initiation and outcomes among post-extubation dysphagia patients: a multicenter retrospective cohort study. *Crit Care* 2022;26:98.
25 Mao H, Lyu Y, Li Y, *et al*. Clinical study on swallowing function of brainstem stroke by tDCS. *Neurol Sci* 2022;43:477–84.
26 Seo K-H, Jang J, Jang EG, *et al*. Clinical effectiveness of the sequential 4-channel NMES compared with that of the conventional 2-channel NMES for the treatment of dysphagia in a prospective double-blind randomized controlled study. *J Neuroeng Rehabil* 2021;18:90.
27 Bascuñana-Ambrós H, Renom-Guiteras M, Nadal-Castells MJ, *et al*. Swallowing muscle training for oropharyngeal dysphagia: a non-inferiority study of online versus face-to-face therapy. *J Telemed Telecare* 2021;1357633:x211035033.
28 Hong JT, Oh JS, Lee DH, *et al*. Association between the severity of dysphagia and various parameters of the cervical spine; videofluoroscopic analysis in neutral and retraction position of the normal volunteers. *Spine* 2020;45:103–8.
29 Herrmann C, Schradt F, Lindner-Pfleghar B, *et al*. Pharyngeal electrical stimulation in amyotrophic lateral sclerosis: a pilot study. *Ther Adv Neurol Disord* 2022;15:17562864211068394.
30 Sterne JAC, Savović J, Page MJ, *et al*. Rob 2: a revised tool for assessing risk of bias in randomised trials. *BMJ* 2019;2:l4898.
31 Higgins JPT*et al*. Measuring inconsistency in meta-analyses. *BMJ* 2003;327:557–60.
32 Guyatt GH, Oxman AD, Vist GE, *et al*. Grade: an emerging consensus on rating quality of evidence and strength of recommendations. *BMJ* 2008;336:924–6.