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Neuromuscular electrical stimulation for children with dysphagia: a systematic review

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ABSTRACT

Objectives Dysphagia in childhood has important health impacts for the child and their family as well as the healthcare system. This systematic review aims to determine the effectiveness of neuromuscular electrical stimulation (NMES) for treatment of oropharyngeal dysphagia in children.

Methods A search was performed on November 2020 in MEDLINE (from 1946), EMBASE (from 1947), PsycINFO (from 1806), CINAHL (from 1937), CENTRAL (from 1996) and Scopus (from 1970) databases. Studies of children (≤18 years) diagnosed with oropharyngeal dysphagia using NMES in the throat/neck region were included. Screening, data extraction, and risk of bias assessment followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines. Risk of bias was assessed using the Cochrane Collaboration's tool for randomised controlled trials (RCTs) and a modified Newcastle-Ottawa assessment for observational studies. A meta-analysis was not conducted due to clinical heterogeneity in studies.

Results Ten studies were included (5 RCTs, 4 case series, 1 cohort study; including 393 children, mean or median age below 7 years, including children with neurologic impairments). In all studies, swallowing function improved after NMES treatment. The standardised mean difference (SMD) for improvement of swallowing dysfunction in treatment compared with control groups in the RCTs ranged from 0.18 (95% CI −0.7 to 1.06) to 1.49 (95% CI 0.57 to 2.41). Eight of 10 studies reported on the child's feeding ability, and, with one exception, there was improvement in feeding ability. Few studies reported on health status (N=2), impact on caregiver (N=1), adverse events and harms (N=2), and child's quality of life (N=1). In most studies, outcome follow-up was less than 6 months. The studies demonstrated moderate to high risk of bias.

Conclusions NMES treatment may be beneficial in improving swallowing function for children with dysphagia, however, given the quality of the studies, inadequate outcome reporting, and short follow-up duration, uncertainty remains. Well-designed RCTs are needed to establish its effectiveness before its adoption in clinical practice.

PROSPERO registration number CRD42019147353.

Strengths and limitations of this study

- ► First systematic review to assess effectiveness of neuromuscular electrical stimulation (NMES) in children with dysphagia.
- ► Synthesised observational and trial evidence suggests NMES improves swallowing function in children.
- ► A pooled intervention effect and meta-analysis were not conducted due to clinical heterogeneity.
- ► High-quality trials are needed before adopting NMES in routine clinical practice.
- ► Future randomised controlled trials should include comprehensive and valid outcome measurement and longer follow-up.

INTRODUCTION

Dysphagia is defined as difficult or abnormal swallowing.[1] A US national health survey conducted in 2012 found that among 61 million children about 1% suffered a swallowing problem lasting greater than 1 week in the previous year.[2] Dysphagia is prevalent in specific populations, including premature infants, and children with cerebral palsy, traumatic brain injury, craniofacial abnormalities, and children with medical complexity.[1] The prevalence of dysphagia ranges from 58% to 99% in children with cerebral palsy[3] and 10.5% in premature infants (<37 weeks).[1] Dysphagia has significant health impacts, including a range of nutritional and respiratory complications.[4 5] Dysphagia can result in poor oral intake and consequently can lead to malnutrition and failure to thrive. Aspiration and pneumonia are common respiratory complications.[1] Recurrent wheezing and chronic lung disease may also occur.[6] More broadly, dysphagia can impact daily activities and social interaction with peers.[7] It also has an impact on family and caregivers. For example, parents of children with dysphagia experience higher levels of anxiety and stress when compared with parents of children without dysphagia.[8 9] Furthermore, the care of children with dysphagia is costly,[10 11] in part due to increased hospitalisation rates, longer hospital lengths of stay, repeated emergency room visits, and increased costs of treatment, including tube feeding.[11] These frequent interactions with the healthcare system can be resource intensive[10 11] and may place burden and stress on those with dysphagia and their families.[12]

The current management for dysphagia in children includes compensatory strategies[4] and therapeutic interventions.[13] Specifically, compensatory strategies include modifying diet (eg, altering textures, thickening fluids, changing taste or temperature), postural changes, pacing, using various feeding tools (eg, different spoon size), and/or changing environments.[4 14] Therapeutic interventions include oral motor exercises, which involve lip control and tongue control and can be completed with the aid of a therapist and different equipment. For some children, despite compensatory strategies and therapeutic interventions, permanent tube feeding through a gastrostomy may be required to provide nutrition in a safe way, particularly in children with neurological disabilities.[4]

A proposed approach used to treat dysphagia is neuromuscular electrical stimulation (NMES).[15] NMES involves applying electrical current to muscles cutaneously using surface electrodes. Transcutaneous stimulation, administered by occupational therapists and/or speech language pathologists,[16] is then combined with traditional swallowing therapy (eg, compensatory strategies, therapeutic interventions, etc). NMES is usually applied to voluntarily contracting muscles in the neck region, where increased muscle contractions are stimulated through the recruitment of larger and more motor units.[15] Thus, it is hypothesised that NMES improves dysphagia by strengthening muscles involved in swallowing and/or by enhancing the sensory signals of the swallowing response.[15 17] NMES is usually conducted repeatedly over a period of time, for example, some protocols apply NMES up to 5 days a week for a period of 4 weeks, but variation exists.[18]

Evidence that supports NMES use in routine practice is uncertain. One of the most cited meta-analyses in adults with dysphagia demonstrated improvement with NMES and concluded that further research is needed.[19] Several more recent adult studies have also concluded that NMES improves swallowing dysfunction,[20–22] however, high-quality studies are still needed.[23] Findings from adult studies should not be extrapolated to children, given that the aetiology of dysphagia in adults is very different (ie, oropharyngeal malignancies, stroke, dementia) and that the goal of rehabilitation is different in adults (ie, adults are undergoing rehabilitation for lost skills and children are undergoing habilitation to develop skills). In addition, neuroplasticity changes with age. As well, outcomes that are important to children often differ from those that are important to adults. A recent Cochrane systematic review examined the evidence for various interventions to treat dysphagia in children with neurological impairment, but no studies on NMES were included.[24] Therefore, the objective of this systematic review is to assess the effectiveness of NMES for treatment of oropharyngeal dysphagia in children.

METHODS

The protocol for this systematic review was registered in PROSPERO. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines were followed in the development of the protocol and conducting the systematic review. Of note, several study authors who were involved in the systematic review (ShM, JF, SaM) have conducted research in this area and previously conducted a study on NMES. As this study was a systematic review, it did not require institutional ethics approval.

Search strategy and information sources

The search strategy included Medical Subject Headings (MeSH) headings and free-text terms related to 'dysphagia,' 'neuromuscular electrical stimulation', and 'children.' The MeSH and free text terms for 'children' used a validated filter.[25] MEDLINE (OVID from 1946), EMBASE (OVID from 1947), PsycINFO (OVID from 1806), CINAHL (EBCSO from 1937), Cochrane Central Register of Controlled Trials (OVID from 1996) and Scopus (from 1970) databases were searched from inception to 19 July 2019 and updated on 26 November 2020. The search strategy was developed and carried out by LR, a reference librarian with expertise in conducting systematic reviews. RP, ShM, PJG, SaM were also involved in the development of the search terms. Three of the five authors are clinicians involved in the care of children with dysphagia (ShM, SaM, PJG). The complete search strategy for all databases is in online supplemental appendix A. The MEDLINE search strategy was translated and adapted for the other databases.

The International Clinical Trials Registry Platform Search Portal and ClinicalTrials.gov were searched. Only completed trials were included in the review, ongoing trials were reported. As well, reference lists of included studies were reviewed. Citation searches were conducted on the included studies. Finally, we contacted experts in the field to identify studies. We used the web-based software platform Covidence to manage records.

Eligibility criteria

Studies of children (18 years and younger) diagnosed with oropharyngeal dysphagia were included. A diagnosis of dysphagia using clinical assessment and/or diagnostic testing (eg, videofluoroscopic swallow study (VFSS)) was required; it could not be solely by parental assessment. Studies that included both children and adults were only included if data for children and adults were reported separately and/or could be obtained separately from study authors. Studies of children with oesophageal dysphagia were excluded.

All studies using NMES in the throat and neck region were included. Studies using NMES in any other region were excluded. There were no restrictions based on the type of healthcare professional delivering the intervention, setting of the intervention or number of treatments. Studies using electrical stimulation intramuscularly were excluded.

Randomised controlled trials (RCTs), cohort studies, case–control studies, cross-sectional studies, and case series were included. Studies published as full-text, abstract only, and unpublished data were included. There were no restrictions by language of publication. For RCTs, studies using standard of care, an alternative intervention or a placebo treatment as the control group were included. Studies with no control group were also included in the review. Although evidence from RCTs is the gold standard for assessing effectiveness to establish an intervention for clinical practice, non-RCTs, which are at high risk of bias, were included given the paucity of evidence and to inform future trials.

Study selection

All potentially eligible records were independently screened by title and abstract by two review authors (RP and SaM). The full text of studies were retrieved for each included record, and two review authors (RP and SaM) independently screened studies for final inclusion. Reasons for excluding studies were recorded. Any disagreements were resolved through discussion and evaluated by a third review author (PJG) if necessary.

Primary and secondary outcomes

The primary outcome was improvement of swallowing dysfunction as measured by imaging studies, such as VFSS and fiberoptic endoscopic evaluation of swallowing (FEES). Secondary outcomes included: (1) swallowing function as reported by a clinician (eg, a clinical swallowing examination) or by the child and/or their caregiver (eg, patient-reported outcome measures), (2) child's feeding ability (eg, food type(s) the child can consume, ease of feeding, need for any form of enteral tube feeding, duration of time required for feeding, and child's overall experience of feeding), (3) child's health status (eg, anthropometric measurements, medical care, and home care), (4) social impact on child (eg, participation at mealtime), (5) impact on caregiver (eg, stress associated with feeding), (6) potential adverse events and harms (eg, aspiration pneumonia and/or recurrent chest infections, mortality), (7) child's quality of life as reported by the child and/or caregiver, and (8) caregiver's quality of life.

Data extraction and management

One review author (RP) extracted the data and a second review author (SaM) verified the extracted data. Data regarding the methods, participants, interventions, outcomes, results were extracted into an a priori developed data extraction form. Any disagreements were resolved through discussion, and if necessary, by involving a third review author (PJG). Missing data were obtained by contacting study authors by email, up to a maximum of three email attempts.

Risk of bias

Risk of bias for all included studies was assessed independently by two review authors (RP and SaM). Any disagreements were resolved through discussion, and if necessary, by involving a third review author (PJG). For RCT studies, risk of bias was assessed using the Cochrane Collaboration's tool,[26] according to the following domains: random sequence generation, allocation concealment, blinding of participants and researchers, blinding of outcome assessment, incomplete outcome data, selective reporting, and other bias. Each domain was judged as low, high, or unclear risk of bias.[26] For observational studies, risk of bias was assessed using a modified version of the Newcastle-Ottawa assessment tool,[27] according to the following domains: selection, comparability, and outcome. Each domain was then rated using the star system following the coding manual instructions.[27] The scale ranged from 0, indicating the highest risk of bias (ie, none of the criteria fulfilled) to 7, indicating the lowest risk of bias. The overall risk of bias was summarised across the studies included in the review. The risk of bias for each study was considered when evaluating treatment effects.

Statistical analysis

RCTs and observational studies were considered separately. A summary statistic was calculated for the outcomes in each study and grouped according to the predefined primary and secondary outcomes. A risk ratio was calculated for dichotomous outcomes. A standardised mean difference was calculated for continuous outcomes, where the numerator was the difference in change scores between the groups and the denominator was the pooled SD of the combined pre and postoutcome scores across groups.[28] Median (range) was converted to mean (SD) when necessary. Forest plots of effect estimates were constructed for each study and an overall estimate. Given the heterogeneity between the included studies, a meta-analysis was not conducted, and a pooled intervention effect was not calculated. Review Manager (RevMan) was used to conduct the analyses.

Patient and public involvement

There was no patient or public involvement in this review.

RESULTS

Of the 1015 unique records screened by title/abstract, 28 full-text records were assessed for eligibility, and 17 records met eligibility criteria for the systematic review. An additional three articles were included from reference lists and grey literature. Of the 20 records that met eligibility criteria for systematic review, 10 full-text articles were included in systematic review (figure 1) and 10

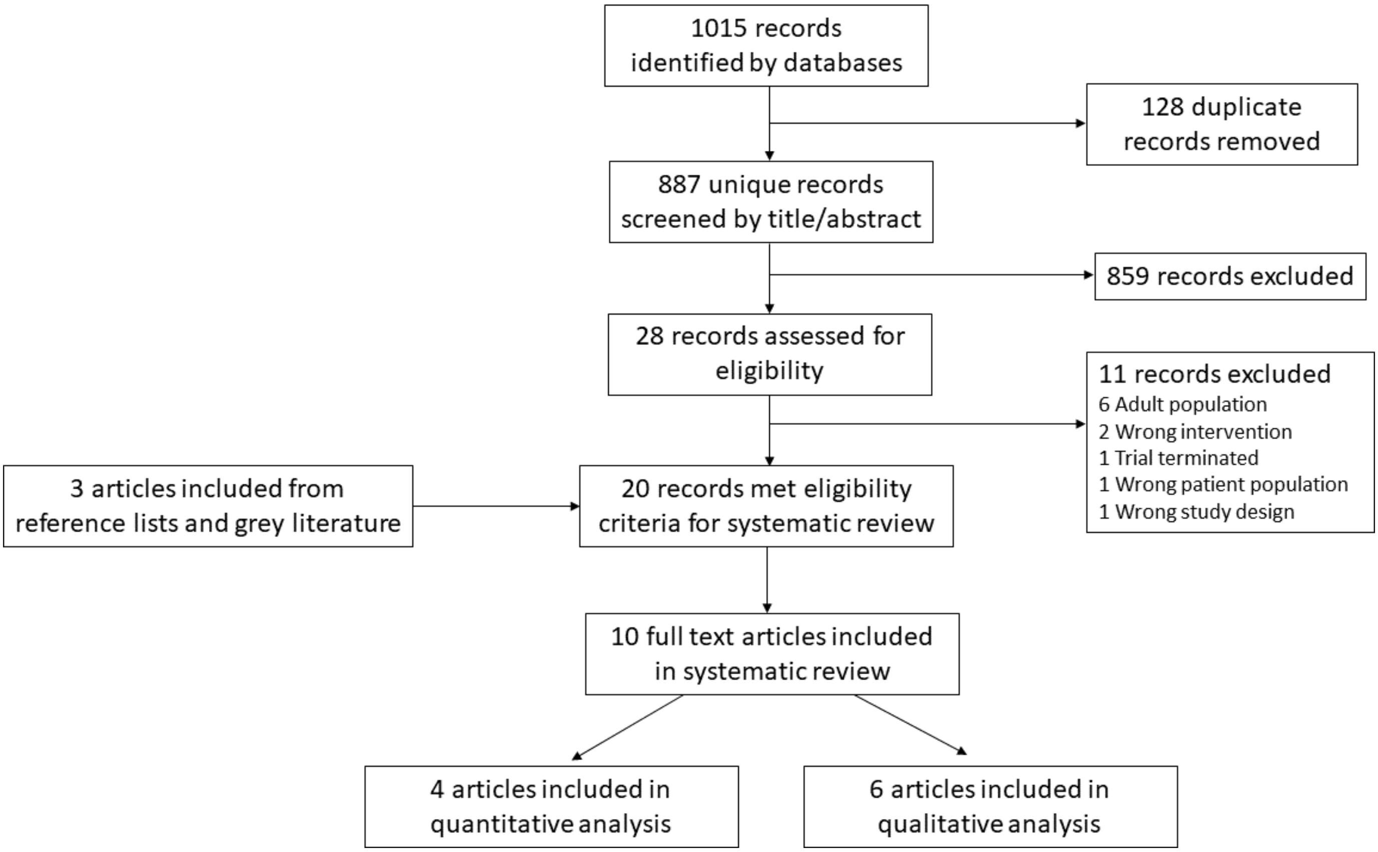

Figure 1 PRISMA flowchart. PRISMA, Preferred Reporting Items for Systematic Reviews and Meta-Analyses.

abstracts were reported in online supplemental appendix B.

Study characteristics

A total of five RCT,[9 29–32] four prospective case series (ie, before and after study with no control group)[33–36], and one retrospective cohort study (with a control group)[37] were included in the review (table 1). One RCT (Gao *et al*)[30] included NMES in both treatment and control groups as well as a cointervention in the treatment group, and, therefore, was only the control group was analysed. In addition, we were unable to obtain the required data to analyse the study by Christiaanse *et al* quantitatively,[37] and, therefore, this study was analysed qualitatively. Thus, four studies (four RCTs) were analysed quantitatively[9 31 32 38] and six studies were analysed qualitatively.[30 33–37]

The studies were from six countries: Egypt,[32] China,[29 30] Canada,[34] Turkey,[9] South Korea[31 33] and USA.[35–37] The mean age of the included participants was less than 4 years in eight studies[9 29 30 32 34–37] and the mean age was 6 years in one study.[31] One study did not specify the age of the participants.[9] A total of six studies included participants exclusively with neurological impairment (eg, cerebral palsy)[29–34] and four studies included some participants with neurological impairment.[9 35–37] The follow-up period ranged from a minimum of 4 weeks in two studies[9 33] to a maximum of 8 months in one study.[35] The sample size of the studies ranged from five participants[33] to 62 participants per group (control and treatment groups)[30] (table 1). Most studies used VFSS before and after the intervention to diagnose dysphagia and evaluate response to treatment. Study funding sources are provided in online supplemental appendix C.

Intervention characteristics

Most studies placed the electrodes around the hyoid bone and thyroid notch,[29 30 32 34–37] and in one study, electrode placement was further adjusted on an individual basis.[34] One study placed electrodes on the sublingual muscles,[30] and one study placed electrodes on the submental region.[33] The frequency of sessions ranged from 5 days per week[9 29 30 33] to 1–2 times per week.[31 32 34–37] The session duration ranged from 20 min[29–31] to 1 hour,[9 35] and treatment duration ranged from 4 weeks[933] to 6 months.[36] In most studies, the voltage applied was 80 Hz and current intensity was adjusted until a therapeutic level was reached.[9 31 33–37] A range of interventions were employed for the control groups, but they all included some form of muscle stimulation (eg, oral motor exercises)[9 29–32 36 37] and two studies included a placebo/sham-NMES treatment[31 32] (table 2).

Outcomes

Swallowing function

All studies reported on swallowing dysfunction (table 3 summarises outcomes reported across studies), and in all cases, swallowing function improved over the course of NMES treatment. However, only five used imaging to

Table 1 Study characteristics

Identification			Number of participants		Characteristics of participants				
					Treatment		Control		
Author	Year	Country	Treatment	Control	Age, years mean (SD)	Neurological impairment, N (%)	Age, years mean (SD)	Neurological impairment, N (%)	Follow-up time length
Randomised controlled trials									
El-Sheikh *et al*[32]	2020	Egypt	20	20	3.26 (1.16)	20 (100%)	3.96 (0.98)	20 (100%)	2 months
Lv *et al*[29]	2019	China	45*	15	3.93 (0.73)	45 (100%)	3.92 (0.85)	15 (100%)	3 months
Gao *et al*†[30]	2018	China	62	62	3.01 (1.12)	62 (100%)	3.02 (1.02)	62 (100%)	3 months
Serel Arslan *et al*[9]	2017	Turkey	12	12	1.01 (0.63)	1 (8.3%)	1.28 (0.93)	1 (8.3%)	1 month
Song *et al*[31]	2015	South Korea	10	10	6.2 (2.78)	10 (100%)	6.00 (2.40)	10 (100%)	2 months
Prospective case series									
Andreoli *et al*[36]	2019	USA	15	-	2.51 (3.20)	11 (73%)	-	-	6 months
Ma and Choi[33]	2019	South Korea	5	-	Not reported	5 (100%)	-	-	1 month
Marcus *et al*[34]	2019	Canada	7	-	0.66 (0.37)	7 (100%)	-	-	2–4 months
Rice[35]	2012	USA	5	-	1.45 (1.05)	2 (40%)	-	-	3–8 months
Retrospective cohort studies									
Christiaanse *et al*[37]	2011	USA	47	46	2.58 (1.66)	32 (68%)	0.83 (0.68)	16 (35%)	6 months

*Three treatment groups; 15 participants in each group.
†Included NMES in both treatment and control groups, and therefore, was analysed as a prospective case series.
NMES, neuromuscular electrical stimulation.

define swallowing function, and, therefore, we grouped swallowing dysfunction as measured by imaging or clinical examination for this outcome. The standardised mean difference for improvement of swallowing dysfunction in treatment compared with control groups in the RCTs ranged from 0.18 (95% CI −0.7 to 1.06)[31] to 1.49 (95% CI 0.57 to 2.41)[9] (figure 2). The study by Lv *et al*[29] (RCT) exhibited a graded improvement in swallowing function with increasing current intensity in the different treatment groups (figure 2). In two small prospective case series studies, all participants improved in swallowing function (online supplemental table 1). In the study by Ma *et al*[33] (prospective case series study) and Gao *et al*[30] (RCT treated as cohort study), the scales used to grade swallowing dysfunction showed an overall improvement (online supplemental table 1). Specifically, in the Gao *et al* study, 58% of participants improved their swallowing effectiveness. Improvement in the swallowing activities ranged from 3% improvement on the chewing movement subscale to 11% on the free eating subscale.[30] In the Ma *et al* study, there was a 45% improvement in swallowing semisolids and 33% improvement in swallowing liquids as measured by the Penetration Aspiration Scale.[33] In the Andreoli *et al* study, there was a statistically significant improvement in mean swallowing function as measured by the Functional Oral Intake Scale (FOIS) (from 3.07 (1.94) to 4.47 (2.26)). In the Christiaanse *et al* study, there was no significant difference observed between the treatment and control groups for change in swallowing function measured by FOIS (online supplemental table 1).[36]

Child's feeding ability

Eight of 10 studies reported on child's feeding ability.[9 29–31 34–37] In all but one study, there was improvement in feeding ability over the course of the NMES treatment (figure 2, panel C). Similarly, the study by Lv *et al* exhibited a graded improvement in feeding ability with increasing current intensity in different treatment groups (figure 2).[29] Marcus *et al* reported that all participants improved in their ability to swallow different consistencies safely and 5 of 5 participants progressed from tube feeding to full or partial oral feeding.[34] Similarly, all participants in the Rice *et al* study improved their ability to swallow different consistencies.[35] There was an improvement in the feeding ability of 69% of participants in the Gao *et al*'s study.[30] In the Andreoli *et al*'s 2019 study, improvement was noted in seven of eight patients with gastrostomy dependence; for patients without gastrostomy dependence, full oral feeds without restriction was achieved in six of seven patients (online supplemental table 1).[36] The standardised mean difference for improvement in feeding behaviours compared with the control group in the study by Song *et al*[31] (RCT) was 0.39 (95% CI −0.49 to 1.28). The exception was the study by Christiaanse *et al*[37] (retrospective cohort study), which found that the

Table 2 NMES and control group intervention details

Author	Year	Additional intervention components	Electrode placements	Frequency of sessions	Duration of NMES session	Duration of treatment	Voltage applied	Current intensity	Control intervention
Randomised controlled trials									
El-Sheikh *et al*[32]	2020	Oral motor exercises	1. Over the neck between hyoid and jaw 2. Side of face	Twice weekly	20 min	2 months	60 Hz	7 mA to 15 mA	Oral motor exercises; placebo NMES
Lv *et al*[29]	2019	-	1. Above hyoid bone, 2. Above the thyroid upper notch	5 days a week	20 min	3 months	80 Hz	10 mA, 15 mA, 20 mA (three treatment groups)	Routine rehabilitation training (oral massage and acupuncture)
Gao *et al**[30]	2018	Oral motor exercises, Acupuncture	Superficial muscles and sublingual muscles	5 days a week	20 min	3 months	20–30 Hz	3–7 mA	Oral motor exercises, NMES
Serel Arslan *et al*[9]	2017	Thermal tactile application, hyolaryngeal mobilisation	1. Below the jaw 2. Above the thyroid notch	5 days a week	30 min	1 month	80 Hz	Until a therapeutic level was reached	Non-nutritive stimulations
Song *et al*[31]	2015	Oral sensorimotor treatment	1. Over the throat between jaw and hyoid 2. Between hyoid and thyroid notch	Twice weekly	20 min	2 months	80 Hz	Typical level ranged from 3mA to 5 mA (determined by palpation for muscle contraction)	Oral sensorimotor treatments and sham-NMES
Prospective case series studies									
Andreoli *et al*[36]	2019	Comprehensive feeding therapy programme	1. Superior aspect of the thyroid cartilage, 2. Along tongue base superior to hyoid bone	Weekly	Up to 50 min or as tolerated	6 months	80 Hz	7.5 (2.7) mAmp (maximal mean (SD))	No control group
Ma and Choi[33]	2019	---	Submental region	5 days a week	30 min per day	1 month	80 Hz	Until the patients felt a minimal stimulation level	No control group
Marcus *et al*[34]	2019	Feeding to elicit swallowing during NMES session	1. Above the hyoid, or 2. Around the thyroid notch Location determined based on the child's swallowing impairment	Twice a week for 2 months; Further 2 months for those in whom improvement was not observed at 2 months	40 min (median; range 20–45)	2–4 months	80 Hz	Range from 3 to 16 mA, based on the response of the child	No control group

Continued

Table 2 Continued

Author	Year	Additional intervention components	Electrode placements	Frequency of sessions	Duration of NMES session	Duration of treatment	Voltage applied	Current intensity	Control intervention
Rice[35]	2012	Oral motor stimulation techniques between sessions	1. Above the thyroid notch, or 2. Around the thyroid notch	1–2 times a week	1 hour	Varied	80 Hz	0 to 25 mA (adjusted for each child to their highest tolerated level)	No control group
Retrospective cohort studies									
Christiaanse *et al*[37]	2011	Non-nutritive oral motor stimulation or a meal (during NMES); Oral motor exercises between sessions	1. Below the jaw, 2. Above the thyroid notch In general, infants would only have a pair of electrodes overlying the anterior belly of digastric muscles	2. 9 times a week (mean, range 0.7–4.6)	30–45 min depending on patient age	2.2 months (mean, range 0.5–6.2 months)	80 Hz	Adjusted until a therapeutic level was reached	Usual diet manipulation, oral motor therapy

*Included NMES in both treatment and control groups, as well as a co-intervention in the treatment group, and therefore, only the control group was analysed.
NMES, neuromuscular electrical stimulation.

intervention was not associated with an improvement in feeding ability compared with the control group (online supplemental table 1).

Child's health status

Lv *et al*[29] reported on nutritional status, which improved over the course of treatment with standardised mean differences ranging from 0.59 (95% CI −0.15 to 1.32) to 1.33 (95% CI 0.53 to 2.13) for the different treatment groups compared with the control group using increasing voltage of NMES. El-Sheikah *et al*[32] found that weight increased over the course of treatment (standardised mean difference of 0.04 (95% CI −0.58 to 0.66) (figure 2).

Potential adverse events and harms

Marcus *et al*[34] reported on adverse events and harms, which included mild skin irritation at the placement of electrode and resolved without treatment (N=6). Andreoli *et al*[36] reported that the NMES was well tolerated and there were no treatment complications. Adverse events were not reported in the other studies.

Child's quality of life as reported by the child

Gao *et al*[30] reported on child's quality of life using the Gessell scale and there was an overall improvement over the course of treatment. More specifically, there was a mean improvement of 7% for the adaptabilities subscale, 8% for the language behaviour subscale, and 6% for the personal social development subscale (online supplemental table 1).

Impact on caregiver

Marcus *et al*[34] reported on the impact on the caregiver, specifically five of seven caregivers felt that their child's feeding improved and were satisfied with the intervention. However, a common reported negative aspect was the frequent need to travel to the hospital for treatments.

No studies reported on the social impact on child or caregiver's quality of life (table 3).

Quality of studies

The risk of bias for RCTs was assessed using the Cochrane collaboration tool.[26] All studies were rated low for attrition bias and reported bias, most studies had a high level of bias for areas related to blinding, most studies were low for random sequence generation, and all studies were unclear for allocation concealment (online supplemental table 3). Furthermore, for all the RCTs, there was no reported primary outcome or sample size estimation. The observational studies were assessed for risk of bias using a modified version of the Newcastle-Ottawa scale:[27] three of five studies scored 4 (out of seven),[30 34 35] one study scored 1 (out of seven),[33] one study scored 5 (out of seven),[36] and one study scored 6 (out of eight)[37] (online supplemental table 2).

Table 3 Summary of outcomes reported

Outcome	Study El-Sheikh *et al*[32]	Andreoli *et al*[36]	Lv *et al*[29]	Gao *et al*[30]	Serel arslan *et al*[9]	Song *et al*[31]	Ma and Choi[33]	Marcus *et al*[34]	Rice[35]	Christiaanse *et al*[37]
Swallowing function										
Measured by imaging studies	–	–	–	–	+	–	+	+	+	+
Reported by clinician	–	+	+	+	–	+	–	–	–	–
Reported by child and/or their caregiver	+	–	–	–	–	–	–	–	–	–
Child's feeding ability	–	+	+	+	+	+	–	+	+	+
Child's health status	+	–	+	–	–	–	–	–	–	–
Social impact on child	–	–	–	–	–	–	–	–	–	–
Impact on caregiver	–	–	–	–	–	–	–	+	–	–
Potential adverse events and harms	–	–	–	–	–	–	–	+	–	–
Child's quality of life	–	–	–	+	–	–	–	–	–	–
Caregiver's quality of life	–	–	–	–	–	–	–	–	–	–

'+' indicates that the outcome was reported in the study and '–' indicates that the outcome was not reported in the study.

DISCUSSION

Key findings

Dysphagia is a serious, often chronic health problem with significant impacts for the child, family, and the healthcare system.[4 5 8 9 39] It can lead to poor oral intake and malnutrition, aspiration and pneumonia as well as impact daily activities and social interaction with peers.[4 5 39] Parents and caregivers of children with dysphagia experience increased anxiety and stress,[8 9] and there are increased costs and utilisation of the healthcare system.[10 11] NMES is a proposed intervention for the treatment of dysphagia. In this review, we analysed 10 studies,

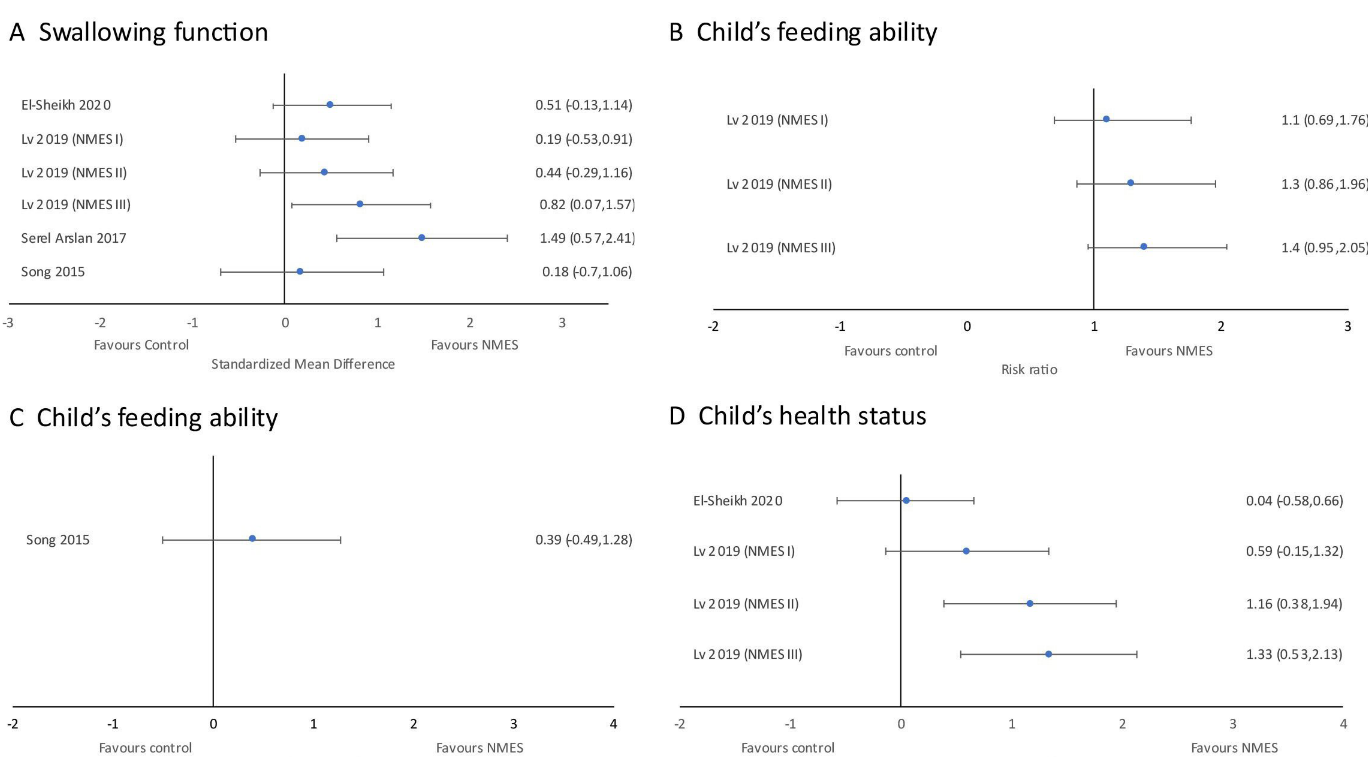

Figure 2 Outcomes in RCTs.[11] Studies did not report all outcomes; for standardised mean difference in Panel A, C, D, point estimate of 0=no effect, and for risk ratio in Panel B, point estimate of 1=no effect. RCT, randomised controlled trial; NMES, neuromuscular electrical stimulation.

including five RCTs,[9 29–32] four prospective case series studies without a control group[33–36] and one retrospective cohort study[37] where NMES was used to treat dysphagia in children. Overall, both the RCTs and observational studies reported an improvement in swallowing dysfunction following NMES intervention, which demonstrates that NMES treatment may be beneficial for children with dysphagia. However, given the limitations in the primary studies, well-designed trials are needed before adopting this intervention in clinical practice.

Findings in the context of previous research

This is the first systematic review to investigate the use of NMES for children with dysphagia. Several systematic reviews and meta-analysis have been conducted in different populations of adults with dysphagia (eg, stroke, head and neck cancer), with some reported benefits on swallowing dysfunction,[20–22] however, these results need to be interpreted with caution in children. Two recent reviews examined the management and interventions for children with dysphagia,[1 24] but no studies on NMES were included. In addition, the population of children with dysphagia is heterogenous, including neonates and infants with a developing muscular system, children with a disordered neuromuscular system (such as children with cerebral palsy), and children with a developed neuromuscular system (such as children with acquired dysphagia).[40]

To date, the studies in this area are limited in number, and many demonstrated moderate to high risk of bias. For example, most RCTs did not blind participants, personnel, and outcome assessment. Though studies randomised patients to treatment and control groups, there was insufficient information provided about allocation concealment across all studies. In the Song *et al* study,[31] there was a significant difference between treatment and control groups at baseline. The observational studies had similar issues: the representativeness of the sample was unclear in two studies,[33 37] there was a significant difference between treatment and control groups in one study,[37] and one study provided insufficient data on participants.[33] Finally, in 7 of 10 studies, the follow-up period was less than 6 months.[9 29–34] A longer follow-up period (eg, 1 year) is important to determine whether any benefit derived from NMES is temporary or sustained. Together, these factors contribute to moderate to high risk of bias in the included studies.

Limitations

This review has several limitations. First, due to clinical heterogeneity, we did not calculate a pooled intervention effect and conduct a meta-analysis. Second, we included a wide range of studies, several of which did not have a control group. In these cases, it cannot be determined that the observed effect was due to NMES. Some of these studies had additional intervention components, such as oral motor exercises, and this may have contributed to the observed effect. In addition, there is a natural maturational development that occurs with time, which may have also contributed to the observed effect. However, when there is limited evidence, it is important to survey the literature more broadly, and, therefore, we included these studies in our review. There were 10 abstracts and unpublished studies that were not included in the analysis, however, there was insufficient data reported to include these reports in our review. Third, the populations of children with dysphagia in the included studies are clinically heterogenous—neonates and infants, children with neuromuscular disorders, and children with acquired dysphagia. The populations different physiologically, which has implications for use of NMES. Finally, some studies may have been missed despite a comprehensive systematic search of the literature.

Implications for future research

We have several recommendations for future trials. We recommend the use of imaging studies to objectively evaluate swallowing function, such as VFSS and FEES, which are established imaging techniques used to evaluate swallowing function.[41 42] Furthermore, it is imperative to use validated outcome measures for children with dysphagia that can be applied across different paediatric populations. In terms of validated patient and/or parent-reported outcomes, there are several potential measures,[43–46] however, there is no established standard that would allow for systematic reporting and comparison across studies. Second, only several of the a priori primary and secondary outcomes were reported across the studies. Dysphagia has impacts across biological, social, and psychological realms, and, therefore, it is important for future trials to assess a broad range of outcomes to develop a more complete clinical picture. For example, none of the studies in this review reported on social impact on the child, impact on the caregiver, and the caregiver's quality of life. In order to ensure outcomes important to patients and their families are assessed, we recommend patients and their families be included as partners when designing future studies. In addition, the effects of the intervention should be assessed in different patient populations (eg, young infants and older children) and aetiologies (eg, premature infants, neurologic impairment). It would be important to assess outcomes for a sufficiently long duration of follow-up to determine whether the effect of NMES persists over time. However, in most of these studies, the follow-up period was less than 6 months. It is also important to establish standards for the implementation of NMES regarding probe placement, duration, frequency, voltage, and current intensity. Finally, all trials should be reported according to the Consolidated Standards of Reporting Trials guidelines, including sample size justification as well as blinding participants and personnel, which were notably missing in many of these studies.

Conclusions

Dysphagia in childhood has important health impacts for the child and their family as well as the healthcare system. Evidence from small trials with moderate to high risk of bias and observational studies suggest that NMES improves swallowing function in children. There is limited evidence on other important outcomes. Further high-quality trials are needed before NMES is adopted in routine clinical practice.

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