

PEER REVIEW HISTORY

BMJ Paediatrics Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

ARTICLE DETAILS

TITLE (PROVISIONAL)	Neuromuscular electrical stimulation for treatment of dysphagia in infants and young children with neurologic impairment: a prospective pilot study
AUTHORS	Marcus, Sherna; Friedman, Jeremy; Lacombe Duncan, Ashley; Mahant, Sanjay

VERSION 1 – REVIEW

REVIEWER	Reviewer name: Francesco Mozzanica Institution and Country: Department of Clinical Sciences and Community Health, University of Milan, Italy Competing interests: None
REVIEW RETURNED	29-Oct-2018

GENERAL COMMENTS	<p>Thank you for the opportunity to review this interesting article.</p> <p>The authors studied the effect of NMES in the treatment of dysphagia in children with neurological impairment. The study design is decent, the materials and methods section is clear and the results are interesting.</p> <p>I have only two minor concerns: The first one is related to the small number of enrolled subjects and to the variability of the neurologic impairment that finally lead to dysphagia. In addition, a control group is not present. The authors correctly highlighted this limitation in the discussion section. Consequently, even if this paper has enough quality, the results here reported should be considered preliminary. Second, the authors should add some information regarding additional application of NMES in children. For example, there are numerous studies assessing the utility of this treatment in children with cerebral palsy. Moreover, additional information regarding the utility of NMES in adults with dysphagia should be added. For example, in the study of Scarponi et al. (Folia phoniatrica et logopedica 2015) a positive evolution of dysphagia after NMES was demonstrated.</p>
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VERSION 1 – AUTHOR RESPONSE

Reviewer 1. Comment 1.

“The first one is related to the small number of enrolled subjects and to the variability of the neurologic impairment that finally lead to dysphagia. In addition, a control group is not present. The authors correctly highlighted this limitation in the discussion section. Consequently, even if this paper has enough quality, the results here reported should be considered preliminary.”

Response. We have made the following changes in the manuscript:

Discussion, paragraph 5. We have added the point about the variability of the neurologic impairment. "There are a number of limitations to our study. First, the small sample size, the variability in the underlying nature of neurologic impairment and lack of a control group clearly limits this to a 'pilot' study and does not allow for conclusions to be drawn about the effectiveness of the treatment."

Discussion, last paragraph. We have highlighted the fact that the results reported are preliminary. "As our data on efficacy is only preliminary, it is important for well-designed RCTs of NMES treatment to be conducted in order to establish efficacy before its routine adoption in practice."

Reviewer 1. Comment 2

"Second, the authors should add some information regarding additional application of NMES in children. For example, there are numerous studies assessing the utility of this treatment in children with cerebral palsy."

Response. We have added a paragraph in the discussion to address the reviewer's suggestion with additional supporting references.

Discussion, paragraph 4. "In children, NMES has been studied for indications other than dysphagia such as for improving strength and motor function in children with cerebral palsy. For example, NMES has been applied to lower limb muscle group(s) during exercise or walking at repeated sessions over time with the goal of improving gait. Results of these studies have shown none to modest benefits on muscle strength, motor performance and gait, and because of limitations in the quality of evidence (i.e. non-randomized and/or small sample size) NMES has not been recommended or cautiously recommended for lower extremity muscle rehabilitation.^{13,14,15} In the cerebral palsy population, NMES has been used for treatment of other muscle groups including the abdominal muscles to improve sitting, and upper extremity muscles to improve function, but again the evidence for its effectiveness is limited.^{13,14}"

13. Electrical stimulation in cerebral palsy: A review of effects on strength and motor function. *Dev Med Child Neurol.* 46 (3):205-213.

14. Wright PA, Durham S, Ewins DJ, Swain ID. Neuromuscular electrical stimulation for children with cerebral palsy: a review. *Arch Dis Child.* 2012. 97(4):364-371.

15. Novak I, McIntyre S, Morgan C et al. A systematic review of interventions for children with cerebral palsy: state of the evidence. *Dev Med Child Neurol.* 2013. 55 (10):885-910.

Reviewer 1. Comment 3.

"Moreover, additional information regarding the utility of NMES in adults with dysphagia should be added."

Response. We have added a paragraph in the discussion to address the reviewer's suggestion with accompanying references, included a recent Cochrane review.

Discussion, paragraph 2. "Similar to our study, larger adult reports of the safety of NMES have observed no serious adverse events and only occasional skin irritation related to electrode placement.¹⁰ In adults with dysphagia due to various causes including stroke and head and neck cancer, non-randomized studies have shown a small statistically significant improvement in clinical swallowing performance before and after the intervention.³ A 2018 Cochrane review identified six randomized controlled trials in 312 adults with stroke comparing NMES with traditional dysphagia therapy.

11 The meta-analysis found that NMES was effective in reducing pharyngeal transit time, but not in reducing the proportion of patients with dysphagia or penetration aspiration score, and did not improve swallowing ability. The review authors concluded that there was insufficient trial evidence to guide clinical practice around the use of NMES.”

11. Bath PM, Lee HS, Everton LF. Swallowing therapy for dysphagia in acute and subacute stroke. Cochrane Database of Systematic Reviews. 2018. Issue 10. Art. No: CD000323. DOI: 10.1002/14651858.CD000323.pub3.