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Neuromuscular electrical stimulation for treatment of dysphagia in infants and young children with neurological impairment: a prospective pilot study

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ABSTRACT

Objectives To describe the acceptability, safety and effectiveness of neuromuscular electrical stimulation (NMES) in infants and young children with neurological impairment (NI) who have severe dysphagia.

Design A prospective pilot study using a before and after study design.

Setting The Hospital for Sick Children, Toronto, Canada.

Patients Ten infants and young children (0–24 months) with NI and severe dysphagia on videofluoroscopic swallow study (VFSS) who were referred to an occupational therapist (OT). Those with neurodegenerative conditions were excluded.

Intervention NMES treatments lasting 20–45 min twice weekly for the duration of 2–4 months. The NMES was administered during feeding therapy sessions by a trained OT.

Main outcome measures Improvement in swallowing function as measured by VFSS and the need for tube feeding, adverse events and parental acceptability.

Results Seven of 10 enrolled subjects (median age, 8.9 months) completed biweekly NMES treatments (median number of treatments per subject, 18). All of the seven (100%) subjects who completed treatment showed an improvement in swallow function on VFSS. Of the five patients who were not safe to orally feed on any consistency of liquid or puree at baseline, three established full oral feeding and two established partial oral feeding. At baseline, 5/7 children were completely fed by tube versus 0/7 at the end of treatment. No adverse events occurred other than mild skin irritation at the site of electrode placement. Five of seven caregivers felt that feeding was improved and were satisfied with the intervention.

Conclusions Our prospective pilot study of NMES in seven neurologically impaired infants and young children with severe dysphagia suggests that NMES is safe, acceptable to parents and has potential efficacy. Trials are needed to determine if any treatment benefit exists.

Trial registration ClinicalTrials.gov NCT01723358.

What is already known on this topic?

- Children with neurological impairment may have dysphagia that necessitates long-term tube feeding.
- Neuromuscular electrical stimulation (NMES) is a proposed treatment for dysphagia that involves electrical stimulation of the swallowing muscles.
- There is little data on the effectiveness and safety of NMES for treatment of dysphagia in infants with neurological impairment to support its use in clinical practice.

What this study hopes to add?

- This prospective pilot study demonstrated that NMES is acceptable to parents, safe and has potential efficacy for treatment of dysphagia in neurologically impaired infants.
- This pilot study will provide data and inform the design of a randomised controlled trial on NMES for treatment of dysphagia in neurologically impaired infants.

INTRODUCTION

Children with neurological impairment (NI) often have major issues with feeding as a result of swallowing dysfunction.[1] Dysphagia may result in a reduced quality of life for the child and caregivers and necessitate long-term tube feeding. Compensatory strategies, such as positioning and thickening feeds, are used to increase swallow safety; however, these strategies do not improve the underlying swallowing impairment. There is no effective treatment for dysphagia in children with NI.

Neuromuscular electrical stimulation (NMES) is a proposed treatment for dysphagia that involves electrical stimulation of swallowing muscles to improve muscle strength, coordination and swallowing function.[2 3] Electrodes are placed on the skin over the anterior neck muscles and small amounts of electrical current are delivered in order to stimulate the muscles responsible for swallowing. In a series of treatment sessions, an occupational therapist (OT) or speech language pathologist with special training in the treatment technique

administers NMES while the patient is given foods or liquids to swallow. A meta-analysis of seven observational studies and two subsequent randomised controlled trials (RCT) in adult stroke and head and neck cancer populations found modest improvement in swallowing function associated with NMES.[3–5]

Paediatric specific data is needed to guide practice recommendations in children. However, there have been no prospective studies evaluating NMES for dysphagia in children with NI. Despite the lack of data, this therapy is currently being used to treat dysphagia in children in some centres. The aim of our pilot study was to describe the effectiveness, safety and acceptability of NMES in infants and young children with a static NI who have severe dysphagia. We also were interested in obtaining prospective data on the recruitment rate and the duration of the NMES treatment. This information will help plan future trials.

METHODS

This prospective pilot study used a before and after study design. Although a RCT would be the optimal study design to determine effectiveness, the lack of data on NMES in children precluded the feasibility and ethical approval of a RCT. Informed consent to participate in the study was obtained from parents. The study was registered at clinicaltrials.gov (NCT01723358). An independent data safety monitoring board met to review safety data during the study.

The study population included infants and young children, ages 0–24 months, who were referred to an OT for dysphagia at the Hospital for Sick Children, Toronto, Canada. Subjects eligible for the intervention were those with NI and severe dysphagia on videofluoroscopic swallow study (VFSS) as defined by aspiration with swallow of at least two of five consistencies of liquid or puree (see table 1). Infants and young children with neurodegenerative conditions were excluded.

The intervention consisted of a 2-month to 4-month period of treatment with NMES provided by an OT with specialised training in the treatment technique. NMES treatment sessions were planned for twice a week for 2 months and then a further 2 months for those in whom improvement was not observed on VFSS at 2 months. The frequency and duration of NMES treatment was based on a review of the literature on NMES used in other studies and the feasibility for parents return for treatment.[2] For this pilot study, the NMES treatment sessions lasted a median of 40 min (range 20–45) twice weekly for a median of 18 treatments (range 10–48). Each session involved feeding the child to elicit swallowing while the NMES was being delivered. The consistencies of the liquid and/or puree given, the position of the child and the equipment used was dependent on the child's level of function and swallowing issue. The electrodes were specifically designed for NMES for dysphagia and were a paediatric size (1.7 cm). Electrode placement was determined based on the child's swallowing impairment as determined by VFSS. Placement of two electrodes was either horizontally on the skin just above the hyoid or horizontally on the skin around the thyroid notch. Stimulation intensity ranged from 3 to 16 mA and was based on the response of the child with optimal intensity being the intensity at which the child appeared to swallow the best while remaining calm.

Liquid and puree consistencies were based on the National Dysphagia Diet Task Force terminology, which was modified with additional consistencies to reflect a typical infant diet (see table 1 for descriptions of the five consistencies).[6 7] Although thin liquids are ideal for infants, if an infant was only able to safely swallow thickened liquids or purees as determined by VFSS, then feeding these consistencies were recommended over no oral feeding at all. The thickening agents that were used were infant cereal or cornstarch-based thickener. The recipes for the different consistencies were determined using the line spread test, a tool that measures viscosity.[8]

The primary outcome was improvement in swallowing function as determined by VFSS. The VFSS was performed by a radiologist and OT. The swallowing function was determined by evaluating the number of consistencies the infant or child could safely swallow without aspiration on the VFSS. The VFSS was performed at baseline, 2 months and 4 months. An increase in the number of consistencies the child could safely swallow defined improvement. The radiologist and OT completed a separate detailed

Table 1 Consistencies of liquids and purees

Consistency*	Description*	Examples of consistency
Thin liquid	Flows quickly through prongs of a fork, leaving little or no residue	Milk, infant formula
Thick liquid (nectar-like)	Flows through prongs of a fork, leaving slight residue	Tomato juice
Thin puree (honey-like)	Coats the prongs of a fork and slowly sinks through. Flows in a continuous narrow stream when poured.	Room temperature honey
Medium puree	Flows in a continuous wide stream when poured	Pancake mix Baby fruit puree
Thick puree (spoon-thick)	Does not pour. Drops off the spoon in a soft bolus. Can be eaten with a spoon but not a fork. Does not hold its shape	Apple sauce

*Terminology adapted from National Dysphagia Diet Task Force and Marcus and Breton.[6 7].

Table 2 Number of consistencies of liquids and purees swallowed safely on VFSS and oral feeding ability level by participant at baseline, 2 months and 4 months after NMES intervention

	Demographics at baseline			Baseline		2 months		4 months			
	Gestational age, age and weight at baseline	Neurological impairment aetiology or description	Tube feeding	Safe consistencies on VFSS (n)	Oral feeding ability level*	Safe consistencies on VFSS (n)	Oral feeding ability level	Safe consistencies on VFSS (n)	Oral feeding ability level	Improvement score†	Oral feedingchange‡
1	Term, 4.2 months, 5.3 kg	Hypotonia	+	0	1	0	1	3	2	+3	0% to <50%
2	Term, 13.0 months, 6.2 kg	Genetic/syndromic	–	3	4	4	4			+1	100%–100%
3	Term, 1.8 months, 3.9 kg	Hypotonia	+	0	1	0	2	5	4	+5	0%–100%
4	35 weeks, 12.5 months, 8.8 kg	Hypotonia	+	0	1	5	4			+5	0%–100%
5	24 weeks, 10.6 months, 7.4 kg	Preterm, IVH	–	3	4	5	4	4	4	+1	100%–100%
7	Term, 8.9 months, 6.9 kg	Pseudobulbar palsy	+	0	1	0	1	3	2	+3	0% to <50%
9	Term, 4.5 months, 5.2 kg	Genetic/syndromic	+	0	1	5	4			+5	0%–100%

*Oral feeding ability and the need for tube feeding was assessed using a structured scale based on parental report of dietary intake that included: level 1: nothing by mouth, all nutrition by tube feeds; level 2: <50% intake by mouth, supplemental tube feeding required; level 3: >50% intake by mouth, supplemental tube feeding required; level 4: all by mouth, no tube feeding.
†The increase in the number of consistencies the infant could safely swallow from baseline to last measurement on VFSS.
‡Proportion of intake by mouth at baseline and at last measurement.
IVH, intraventricular haemorrhage; NMES, neuromuscular electrical stimulation; VFSS, videofluoroscopic swallow study.

structured reporting form adapted from previous work[9] to classify results from the VFSS. The secondary outcomes included another measure of effectiveness, the need for tube feeding at end of treatment, adverse events and parental acceptability of the intervention. The need for tube feeding was assessed using a structured questionnaire administered to parents at baseline, 2 months and 4 months. Adverse events were assessed at every treatment session. A structured questionnaire assessed local complications (skin irritation) and systemic complications including respiratory distress after the treatment, seizures after the procedure and admission to hospital after treatment. At the end of treatment, parents were asked to rate their satisfaction with the intervention, their perception of feeding improvement and any positive and negative aspects to the intervention.

Patient and public involvement

Parents of patients were first involved in this pilot study in the outcomes measurement stage. Parents of patients were asked to assess their satisfaction with the intervention and the burden of the intervention as an outcome of the study as described above. Patients and the public were not directly involved in other aspects of the study (ie, research question and outcomes development, study design, recruitment and conduct of the study and dissemination of the study results).

Statistical analysis

The study sample consisted of a convenience sample of 10 infants and young children aged 0–24 months; seven completed the treatments and were included in the analysis. The primary outcome, improvement in swallowing function on VFSS, was presented as the proportion of children who demonstrated an improvement in swallowing function from baseline to 4 months. Secondary outcomes were analysed using descriptive statistics.

RESULTS

Eleven eligible subjects were approached and parents of 10 infants and young children with NI agreed to be enrolled in the study from February 2011 to January 2012. Three subjects did not complete treatment: one died due to renal failure, one discontinued treatment due to severe gastro-oesophageal reflux disease and died of pneumonia 4 weeks after the last NMES treatment and one withdrew from the study after 2 months of treatment due to difficulty attending sessions. Of the seven infants and young children who completed the NMES treatment and were included in the analysis the median age was 8.9 months (range 1.8–13.0) and three were female. Two had a genetic or syndromic diagnosis, one had preterm brain injury with intraventricular haemorrhage, one had a pseudobulbar palsy associated with hypotonia and three had hypotonia without an identified diagnosis after investigation. One required antiepileptic drugs and all received medications for gastro-oesophageal reflux

Table 3 Number of consistencies of liquid and puree swallowed safely on videofluoroscopic swallow study (VFSS) and oral feeding ability level* at baseline and end of treatment for study group

	Baseline (n=7)	End of treatment (n=7)
Safe consistencies on VFSS (n)		
0 safe consistencies, n (%)	5 (71%)	0 (0%)
One safe consistency, n (%)	0 (0%)	0 (0%)
Two safe consistencies, n (%)	0 (0%)	0 (0%)
Three safe consistencies, n (%)	2 (29%)	2 (29%)
Four safe consistencies, n (%)	0 (0%)	2 (29%)
Five safe consistencies, n (%)	0 (0%)	3 (43%)
Oral feeding ability level		
Level 1, n (%)	5 (71%)	0 (0%)
Level 2, n (%)	0 (0%)	2 (29%)
Level 3, n (%)	0 (0%)	0 (0%)
Level 4, n (%)	2 (29%)	5 (71%)

*Oral feeding ability and the need for tube feeding was assessed using a structured scale based on parental report of dietary intake that included: level 1: nothing by mouth, all nutrition by tube feeds; level 2: <50% intake by mouth, supplemental tube feeding requirement; level 3: >50% intake by mouth, supplemental tube feeding requirement; level 4: all by mouth, no tube feeding.

disease. At baseline, five subjects were not safe to feed on any consistency of liquid or puree and were completely tube fed and the other two were feeding orally.

All of the seven (100%) subjects who completed treatment showed an improvement in swallow function (see tables 2 and 3). The increase in the number of consistencies swallowed safely ranged from 1 to 5. Of the five patients who were not safe to orally feed on any consistencies at baseline, three established full oral feeding and two established partial oral feeding. At baseline, 5/7 children were completely fed by tube versus 0/7 at the end of treatment. No adverse events occurred other than mild skin irritation at the site of electrode placement (n=6) which resolved with no treatment. Five of seven caregivers felt that feeding was improved and were satisfied with the intervention. A common negative aspect reported by parents was the frequent need to travel to the hospital for treatments.

DISCUSSION

In this small prospective pilot study of NMES treatment in infants and young children with a static NI, we observed an improvement in swallowing function over time and no major adverse events. The treatment was acceptable to parental caregivers and they perceived an improvement in feeding associated with the treatment. Feasibility data from this pilot study showed a high recruitment rate and that the treatment duration at each session was not excessively long. However, as one parent discontinued the treatments due to difficulty in attending the sessions, the burden of the repeat hospital visits for the intervention needs to be emphasised in any future trial.

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-randomised studies have shown a small statistically significant improvement in clinical swallowing performance before and after the intervention.[3] A 2018 Cochrane review identified six randomised 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.

The only published paediatric study on NMES for dysphagia[12] is a retrospective study that did not find a treatment benefit in children with primary dysphagia. They did observe a benefit, however, in children with acquired dysphagia. The NMES treated group in this retrospective study had an average age of 31 months and was compared with a control group. However, the two groups were dissimilar in age, underlying diagnoses and initial degree of swallowing dysfunction. Due to the limitations of a retrospective study based on chart review and the absence of standardised timing of assessment of oral intake and swallowing function, it is difficult to make inferences about the effectiveness of NMES based on this study or compare the findings from this study to ours.

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 (ie, non-randomised and/or small sample size) NMES has not been recommended or cautiously recommended for lower extremity muscle rehabilitation[13–15] In the cerebral palsy population, NMES has also 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]

There are a number of limitations to our study. First, the small sample size, the variability in the underlying nature of NI 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. Rather,

the main goal of this pilot study was to obtain data on outcomes and feasibility which will be helpful for planning a future trial. Based on the outcomes we observed, a trial with a sample size of 40 in each group (ie, NMES treatment and control group), will provide an 80% power (assuming alpha of 0.05, two-sided test) to detect a 30% difference between groups in those who are safe on more than four or more consistencies on VFSS and a 30% difference between groups in those who are completely fed by mouth with no tube feeding (oral feeding ability level 4). Second, neither the research assistant nor the OT providing treatment was blinded to the participant treatment allocation. Third, parent self-reported data, which may be prone to social desirability bias, were used for assessment of secondary outcomes including oral feeding ability and acceptability of NMES.

Despite these limitations, our prospective pilot study suggests that NMES is safe, acceptable to parents and has potential efficacy in the population studied. 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.

Contributors All authors conceptualised and designed the study; contributed to data collection and analysis, interpretation of data; drafted and revised the article and gave final approval of the manuscript as submitted.

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Competing interests None declared.

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