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

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| Keywords: | Infant Feeding, Neurodisability, Nutrition, Occupational therapy |
Neuromuscular electrical stimulation for treatment of dysphagia in infants and young children with neurologic 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 neurologic impairment 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 neurologic impairment (NI) and severe dysphagia on videofluoroscopic swallow study who were referred to an occupational therapist (OT). Those with neurodegenerative conditions were excluded.

Intervention NMES treatments lasting 20-45 minutes twice weekly for the duration of 2-4 months. The NMES was administered during feeding therapy sessions by a trained Occupational Therapist.

Main outcome measures Improvement in swallowing function as measured by videofluoroscopic swallow study (VFFS) and the need for tube feeding, adverse events, and parental acceptability.

Results Seven of ten 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 VFFS. 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 vs. 0/7 at 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

Key words: dysphagia, pilot study, neurologic impairment, neuromuscular electrical stimulation

What is known about the subject

Children with neurologic 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 neurologic impairment to support its use in clinical practice.

What this study adds

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 randomized controlled trial on NMES for treatment of dysphagia in neurologically impaired infants.

Introduction

Children with neurologic 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 neurologic impairment (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. 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 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 7 observational studies and two subsequent randomized controlled trials (RCT) in adult stroke and head and neck cancer populations found modest improvement in swallowing function associated with NMES.

Pediatric 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. The study was approved by The Hospital for Sick Children’s research ethics board and 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 to 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 two to four month period of treatment with NMES provided by an OT with specialized training in the treatment technique. NMES treatment sessions were planned for twice a week for two months and then a further two months for those in whom improvement was not observed on VFFS at two 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. For this pilot study, the NMES treatment sessions lasted a median of 40 minutes (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 pediatric 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
3-16 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). Although thin liquids are ideal for infants, if
an infant was only able to safely swallow thickened liquids or purees safely as determined by
VFFS, 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. The primary outcome was improvement in swallowing function as determined by VFSS. The
VFSSs were 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 VFFS was performed at baseline, two months and four months. An
increase in the number of consistencies the child could safely swallow defined improvement. The
radiologist and OT completed a separate detailed structured reporting form adapted from
previous work 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 four 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.

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 analyzed 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 gastroesophageal reflux disease and died of pneumonia four weeks after the last NMES
treatment, and one withdrew from the study after two 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 hemorrhage, one had a pseudobulbar palsy associated with hypotonia, and three had hypotonia without an identified diagnosis after investigation. One required anti-epileptic drugs and all received medications for gastroesophageal reflux 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 1-5. 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 vs. 0/7 at 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 emphasized 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. The only published pediatric study on NMES for dysphagia 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 to 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 standardized 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.

There are a number of limitations to our study. First, the small sample size 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 (i.e. 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 4 or more consistencies on VFFS 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. It is important for well-designed RCTs of NMES treatment to be conducted in order to establish efficacy before its routine adoption in practice.

Abbreviations: Neuromuscular electrical stimulation (NMES); neurologic impairment (NI); randomized controlled trial (RCT); occupational therapist (OT); videofluoroscopic swallow study (VFSS); gastroesophageal reflux disease (GERD)

Funding Statement: This study was funded by a peer-reviewed grant from the Department of Paediatrics, The Hospital for Sick Children, Toronto. ALD was supported through the Norman Saunders Complex Care Initiative, Hospital for Sick Children. Sherna Marcus was supported through the Rehabilitation Department, Hospital for Sick Children.
Competing Interests: There is no real or perceived conflicts of interest to report. The funder of the study had no role in the study design, collection, analysis and interpretation of data, writing of the report, or the decision to submit the paper for publication.

Authors Contributions: Sherna Marcus conceptualized 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. Jeremy N Friedman conceptualized and designed the study; contributed to data analysis, interpretation of data; drafted and revised the article; and gave final approval of the manuscript as submitted. Ashley Lacombe-Duncan contributed to the design of the study, data collection and analysis, interpretation of data; drafted and revised the article; and gave final approval of the manuscript as submitted. Sanjay Mahant conceptualized and designed the study; contributed to data analysis, interpretation of data; drafted and revised the article; and gave final approval of the manuscript as submitted.

References


Neuromuscular electrical stimulation is no more effective than usual care for the treatment of primary dysphagia in children. Pediatr Pulmonol. 2011;46:559-65
Table 1. Consistencies of Liquids and Purees

<table>
<thead>
<tr>
<th>Consistency*</th>
<th>Description*</th>
<th>Examples of Consistency</th>
</tr>
</thead>
<tbody>
<tr>
<td>Thin liquid</td>
<td>Flows quickly through prongs of a fork, leaving little or no residue</td>
<td>Milk, infant formula</td>
</tr>
<tr>
<td>Thick liquid (nectar-like)</td>
<td>Flows through prongs of a fork, leaving slight residue</td>
<td>Tomato juice</td>
</tr>
<tr>
<td>Thin puree (honey-like)</td>
<td>Coats the prongs of a fork and slowly sinks through. Flows in a continuous narrow stream when poured.</td>
<td>Room temperature honey</td>
</tr>
<tr>
<td>Medium puree</td>
<td>Flows in a continuous wide stream when poured</td>
<td>Pancake mix</td>
</tr>
<tr>
<td>Thick puree (spoon-thick)</td>
<td>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</td>
<td>Apple sauce</td>
</tr>
</tbody>
</table>

* Terminology adapted from references 6 and 7.
Table 2. Number of consistencies of liquids and purees swallowed safely on videofluoroscopic swallow study (VFSS) and oral feeding ability level by participant at baseline, 2 months and 4 months after NMES intervention

<table>
<thead>
<tr>
<th>Demographics at Baseline</th>
<th>Baseline</th>
<th>2 months</th>
<th>4 months</th>
<th>Improvement Score</th>
<th>Oral Feeding Change</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Tube feeding</td>
<td># of Safe Consistencies on VFSS</td>
<td>Oral Feeding Ability Level</td>
<td># of Safe Consistencies on VFSS</td>
<td>Oral Feeding Ability Level</td>
</tr>
<tr>
<td>Gestational Age, Age and Weight at baseline</td>
<td>Neurologic Impairment etiology or description</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Term, 4.2 mo, 5.3 kg</td>
<td>Hypotonia</td>
<td>+</td>
<td>0</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Term, 13.0 mo, 6.2 kg</td>
<td>Genetic/Syndromic</td>
<td>-</td>
<td>3</td>
<td>4</td>
<td>4</td>
</tr>
<tr>
<td>Term, 1.8 mo, 3.9 kg</td>
<td>Hypotonia</td>
<td>+</td>
<td>0</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>35 weeks, 12.5 mo, 8.8 kg</td>
<td>Hypotonia</td>
<td>+</td>
<td>0</td>
<td>1</td>
<td>5</td>
</tr>
<tr>
<td>24 weeks, 10.6 mo, 7.4 kg</td>
<td>Preterm, IVH</td>
<td>-</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>Term, 8.9 mo, 6.9 kg</td>
<td>Pseudobulbar Palsy</td>
<td>+</td>
<td>0</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Term, 4.5 mo, 5.2 kg</td>
<td>Genetic/Syndromic</td>
<td>+</td>
<td>0</td>
<td>1</td>
<td>5</td>
</tr>
</tbody>
</table>

a 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.

b The increase in the number of consistencies the infant could safely swallow from baseline to last measurement on VFSS

cProportion of intake by mouth at baseline and at last measurement
Table 3. Number of consistencies of liquid and puree swallowed safely on videofluoroscopic swallow study (VFSS) and oral feeding ability level\(^a\) at baseline and end of treatment for study group

<table>
<thead>
<tr>
<th>No. of safe consistencies on VFSS</th>
<th>Baseline (n=7)</th>
<th>End of Treatment (n=7)</th>
</tr>
</thead>
<tbody>
<tr>
<td>0 safe consistencies, n (%)</td>
<td>5 (71%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>1 safe consistency, n (%)</td>
<td>0 (0%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>2 safe consistencies, n (%)</td>
<td>0 (0%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>3 safe consistencies, n (%)</td>
<td>2 (29%)</td>
<td>2 (29%)</td>
</tr>
<tr>
<td>4 safe consistencies, n (%)</td>
<td>0 (0%)</td>
<td>2 (29%)</td>
</tr>
<tr>
<td>5 safe consistencies, n (%)</td>
<td>0 (0%)</td>
<td>3 (43%)</td>
</tr>
</tbody>
</table>

**Oral Feeding Ability Level**

<table>
<thead>
<tr>
<th>Level, n (%)</th>
<th>Baseline (n=7)</th>
<th>End of Treatment (n=7)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Level One, n (%)</td>
<td>5 (71%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>Level Two, n (%)</td>
<td>0 (0%)</td>
<td>2 (29%)</td>
</tr>
<tr>
<td>Level Three, n (%)</td>
<td>0 (0%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>Level Four, n (%)</td>
<td>2 (29%)</td>
<td>5 (71%)</td>
</tr>
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\(^a\) 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.
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**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. The study was approved by The Hospital for Sick Children’s research ethics board and 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 to 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 two to four month period of treatment with NMES provided by an OT with specialized training in the treatment technique. NMES treatment sessions were planned for twice a week for two months and then a further two months for those in whom improvement was not observed on VFFS at two 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. For this pilot study, the NMES treatment sessions lasted a median of 40 minutes (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 pediatric 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 3-16 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). Although thin liquids are ideal for infants, if an infant was only able to safely swallow thickened liquids or purees safely as determined by VFFS, 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.

The primary outcome was improvement in swallowing function as determined by VFSS. The VFSSs were 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 VFFS was performed at baseline, two months and four months. An increase in the number of consistencies the child could safely swallow defined improvement. The radiologist and OT completed a separate detailed structured reporting form adapted from
previous work 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 four 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 (i.e. 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 analyzed 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 gastroesophageal reflux disease and died of pneumonia four weeks after the last NMES treatment, and one withdrew from the study after two 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 hemorrhage, one had a pseudobulbar palsy associated with hypotonia, and three had hypotonia without an identified diagnosis after investigation. One required anti-epileptic drugs and all received medications for gastroesophageal reflux 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 1-5. 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 vs. 0/7 at 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 emphasized 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. 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. A 2018 Cochrane review identified six randomized controlled trials in 312 adults with stroke comparing NMES with traditional dysphagia therapy. 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 pediatric study on NMES for dysphagia\textsuperscript{11} 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 to 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 standardized 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 (i.e. non-randomized and/or small sample size) NMES has not been recommended or cautiously recommended for lower extremity muscle rehabilitation.\textsuperscript{13,14,15} 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.\textsuperscript{13,14}

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. 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 (i.e. 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 4 or more consistencies on VFFS 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.

**Abbreviations:** Neuromuscular electrical stimulation (NMES); neurologic impairment (NI);
randomized controlled trial (RCT); occupational therapist (OT); videofluoroscopic swallow
study (VFSS); gastroesophageal reflux disease (GERD)

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**Competing Interests:** There is no real or perceived conflicts of interest to report. The funder of the study had no role in the study design, collection, analysis and interpretation of data, writing of the report, or the decision to submit the paper for publication.

**Authors Contributions:** Sherna Marcus conceptualized 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. Jeremy N Friedman conceptualized and designed the study; contributed to data analysis, interpretation of data; drafted and revised the article; and gave final approval of the manuscript as submitted. Ashley Lacombe-Duncan contributed to the design of the study, data collection and analysis, interpretation of data; drafted and revised the article; and gave final approval of the manuscript as submitted. Sanjay Mahant conceptualized and designed the study; contributed to data analysis, interpretation of data; drafted and revised the article; and gave final approval of the manuscript as submitted.

**References**


Table 1. Consistencies of Liquids and Purees

<table>
<thead>
<tr>
<th>Consistency*</th>
<th>Description*</th>
<th>Examples of Consistency</th>
</tr>
</thead>
<tbody>
<tr>
<td>Thin liquid</td>
<td>Flows quickly through prongs of a fork, leaving little or no residue</td>
<td>Milk, infant formula</td>
</tr>
<tr>
<td>Thick liquid (nectar-like)</td>
<td>Flows through prongs of a fork, leaving slight residue</td>
<td>Tomato juice</td>
</tr>
<tr>
<td>Thin puree (honey-like)</td>
<td>Coats the prongs of a fork and slowly sinks through. Flows in a continuous narrow stream when poured.</td>
<td>Room temperature honey</td>
</tr>
<tr>
<td>Medium puree</td>
<td>Flows in a continuous wide stream when poured</td>
<td>Pancake mix Baby fruit puree</td>
</tr>
<tr>
<td>Thick puree (spoon-thick)</td>
<td>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</td>
<td>Apple sauce</td>
</tr>
</tbody>
</table>

* Terminology adapted from references 6 and 7.
Table 2. Number of consistencies of liquids and purées swallowed safely on videofluoroscopic swallow study (VFSS) and oral feeding ability level by participant at baseline, 2 months and 4 months after NMES intervention

<table>
<thead>
<tr>
<th>Demographics at Baseline</th>
<th>Neurologic Impairment etiology or description</th>
<th>Tube feeding</th>
<th>Baseline</th>
<th>2 months</th>
<th>4 months</th>
<th>Improveme</th>
<th>Oral Feeding Change</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gestational Age, Age and Weight at baseline</td>
<td></td>
<td># of Safe Consistencies on VFSS</td>
<td>Oral Feeding Ability Level</td>
<td># of Safe Consistencies on VFSS</td>
<td>Oral Feeding Ability Level</td>
<td># of Safe Consistencies on VFSS</td>
<td>Oral Feeding Ability Level</td>
</tr>
<tr>
<td>Term, 4.2 mo, 5.3 kg</td>
<td>Hypotonia</td>
<td>+</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td>Term, 13.0 mo, 6.2 kg</td>
<td>Genetic/Syndromic</td>
<td>-</td>
<td>3</td>
<td>4</td>
<td>4</td>
<td>4</td>
<td>4</td>
</tr>
<tr>
<td>Term, 1.8 mo, 3.9 kg</td>
<td>Hypotonia</td>
<td>+</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>2</td>
<td>5</td>
</tr>
<tr>
<td>35 weeks, 12.5 mo, 8.8 kg</td>
<td>Hypotonia</td>
<td>+</td>
<td>0</td>
<td>1</td>
<td>5</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>24 weeks, 10.6 mo, 7.4 kg</td>
<td>Preterm, IVH</td>
<td>-</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>4</td>
<td>4</td>
</tr>
<tr>
<td>Term, 8.9 mo, 6.9 kg</td>
<td>Pseudobulbar Palsy</td>
<td>+</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td>Term, 4.5 mo, 5.2 kg</td>
<td>Genetic/Syndromic</td>
<td>+</td>
<td>0</td>
<td>1</td>
<td>5</td>
<td>4</td>
<td>5</td>
</tr>
</tbody>
</table>

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

<table>
<thead>
<tr>
<th>No. of safe consistencies on VFSS</th>
<th>Baseline (n=7)</th>
<th>End of Treatment (n=7)</th>
</tr>
</thead>
<tbody>
<tr>
<td>0 safe consistencies, n (%)</td>
<td>5 (71%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>1 safe consistency, n (%)</td>
<td>0 (0%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>2 safe consistencies, n (%)</td>
<td>0 (0%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>3 safe consistencies, n (%)</td>
<td>2 (29%)</td>
<td>2 (29%)</td>
</tr>
<tr>
<td>4 safe consistencies, n (%)</td>
<td>0 (0%)</td>
<td>2 (29%)</td>
</tr>
<tr>
<td>5 safe consistencies, n (%)</td>
<td>0 (0%)</td>
<td>3 (43%)</td>
</tr>
</tbody>
</table>

**Oral Feeding Ability Level**

<table>
<thead>
<tr>
<th>Oral Feeding Ability Level</th>
<th>Baseline (n=7)</th>
<th>End of Treatment (n=7)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Level One, n (%)</td>
<td>5 (71%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>Level Two, n (%)</td>
<td>0 (0%)</td>
<td>2 (29%)</td>
</tr>
<tr>
<td>Level Three, n (%)</td>
<td>0 (0%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>Level Four, n (%)</td>
<td>2 (29%)</td>
<td>5 (71%)</td>
</tr>
</tbody>
</table>

\(a\) 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.