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ORIGINAL ARTICLE

Effects of Neuromuscular Electrical Stimulation on Swallowing Functions in Children with Cerebral Palsy: A Pilot Randomised Controlled Trial

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Summary *Objective/Background:* Oral-motor and sensory dysfunctions are primary reasons for difficulties with swallowing in children with cerebral palsy (CP). Neuromuscular electrical stimulation (NMES) has been shown to provide positive effects on the swallowing function in adult populations with various neurological disorders. However, there is a lack of studies regarding the effects of NMES in children with dysphagia. The aim of the present study was to investigate the effects of NMES and oral sensorimotor treatment (OST) by occupational therapists in children with CP and dysphagia.
Methods: The present study was a two-group experimental design. Participants were randomly assigned to either the experimental group ($n = 10$) or the control group ($n = 10$). The NMES group received both NMES and OST, with NMES on the pharyngeal level for 20 minutes after OST, while the control group received OST and sham–NMES only. The treatment sessions occurred twice a week for 8 weeks.
Results: The experimental group demonstrated a significant improvement in: lip closure while swallowing, ability to swallow food without excess loss, ability to sip liquid, ability to swallow liquid without excess loss, and ability to swallow without cough ($p < .05$).
Conclusion: This study demonstrated that OST and NMES facilitated swallowing functions than OST and sham–NMES in children with CP and dysphagia. Future studies need to utilise

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video fluoroscopy swallowing study for outcome measurements in a large participant group.

Introduction

Cerebral palsy (CP) is a chronic movement disorder resulting from an abnormal development or injury in the immature brain. Children with CP demonstrate various movement disorders, including muscle weakness, abnormal postural responses, coordination failure, and slow muscle contraction, which cause functional problems, including abnormal gait (Shkedy Rabani et al., 2014), hand dysfunction (Klingels et al., 2012), and dysphagia (Arvedson, 2013; Scott, 2014). These functional dysfunctions frequently limit participation in daily activities. In children, eating is one of the most important activities of daily living, especially for development and quality of life (Kitazumi, 1998). Dysphagia causes malnutrition and negatively impacts development in patients with CP. Approximately 75% of children with CP have dysphagia, and that statistic increases to 86% in patients with quadriplegia (Morris & Klein, 1987). Children with CP have some difficulty with postural balance, postural control, and oral-motor control, which are all necessary for proper eating. Oral-motor and sensory dysfunctions are primary reasons for difficulties with swallowing in children with CP (Arvedson, 2013).

Oral sensory dysfunction causes dysphagia in children with CP (Arvedson, 2013; Arvedson, Rogers, Buck, Smart, & Msall, 1994). Children with CP have a difficulty recognizing oral sensation for localization of the input, because the sensory threshold is different from normal (Arvedson et al., 1994; Erasmus et al., 2009). Due to an abnormal oral sensory threshold, it is difficult to determine where the stimulation occurs within the oral area, including the lips, cheeks, tongue, and oral plates (Weindling, Cunningham, Glenn, Edwards, & Reeves, 2007). During the oral phase of swallowing, sensory information is essential for chewing and controlling the bolus of food. The touch and pressure receptors of the tongue and oral-cavity surfaces transmit sensory information to the brainstem and cerebral cortex to guide tongue shape and pharyngeal pressure according to bolus volume and viscosity (Ali, Cook, Laundl, Wallace, & de Carle, 1997). In a study of oral sensory dysfunction, an oral splint blocking sensory stimuli from the bolus significantly decreased pharyngeal pressure, and delayed the onset of hyoid motion and relaxation of upper oesophageal sphincter (Ali et al., 1997). The sensory stimuli thus modulate the swallowing process (Steele & Miller, 2010). Therefore, various sensory modalities have been used to treat dysphagia in CP (Arvedson, Clark, Lazarus, Schooling, & Frymark, 2010b).

Oral sensory stimulation by occupational therapists has been applied to improve the feeding ability in children with CP (Arvedson et al., 2010b). Vibration stimuli in the oral region normalise the abnormal muscle tone during feeding activities. Oral sensory stimulation, including thermal, tactile, and pressure stimuli, was also shown to reduce tongue thrust and the bite reflex disturbing the swallowing process, and to improve the chewing and swallowing functions in children with CP (Arvedson, Clark, Lazarus, Schooling, & Frymark, 2010a; Arvedson et al., 2010b; Erasmus et al., 2009; Scott, 2014). Oral-sensory-stimulation protocols have been widely used to treat dysphagia in children (Arvedson et al., 2010a). In addition, peripheral electrical stimuli have been applied as an intervention protocol to improve oropharyngeal swallowing in neurological disorders, including stroke (Carnaby-Mann & Crary, 2007), traumatic brain injury (Clark, Lazarus, Arvedson, Schooling, & Frymark, 2009), Parkinson's disease (Baijens et al., 2012), and CP (Christiaanse et al., 2011; Rice, 2012). In brain-imaging studies, electrical pharyngeal sensory stimulation induced the activation of areas in the cerebral cortex related to swallowing, such as the sensorimotor cortex (Fraser et al., 2002). Additionally, the amplitude of pharyngeal electromyography significantly increased after a 10-minute peripheral sensory stimulation with 10 Hz (Hamdy, Rothwell, Aziz, Singh, & Thompson, 1998).

In adults with neurological disorders, previous studies have demonstrated that neuromuscular electrical stimulation (NMES) improved swallowing functions. In stroke patients, NMES improved the ability to safely swallow, as measured by the functional oral intake scale (FOIS), and reduced feeding tube-dependent dysphagia in acute stroke (Kushner, Peters, Eroglu, Perless-Carroll, & Johnson-Greene, 2013). In Parkinson's disease with oropharyngeal dysphagia, NMES improved the quality of life in swallowing disorder (Heijnen, Speyer, Baijens, & Bogaardt, 2012). A few previous studies have suggested that NMES might provide positive effects on swallowing function in paediatric patients (Christiaanse et al., 2011; Rice, 2012). However, there remains a lack of studies investigating the effects of NMES on oral functions related to feeding in CP with dysphagia. Therefore, the aim of this study was to investigate the therapeutic effects of oral sensorimotor treatment (OST) and NMES on oral functions in children with CP and dysphagia.

Methods

Participants and study design

A total of 20 children participated voluntarily in this study. The participants were recruited in 50 outpatient settings of a university hospital by convenience sampling. All of them were diagnosed as having CP with dysphagia. The inclusion criteria were as follows: (a) diagnosed with CP by a rehabilitation doctor, (b) dysphagia confirmed by video fluoroscopy swallowing study (VFSS) or rehabilitation doctors, (c) no disorder in vision or hearing, (d) no seizure disorders, and (e) no pacemaker. All parents of the participants signed consent forms after an investigator explained the purpose and experimental process.

The design of the present study was two-group experimental. The participants were randomly assigned to either the experimental group ($n = 10$) or the control group ($n = 10$). Both groups received OST by occupational therapists in a rehabilitation department. The participants in the experimental group received an additional 20-minute treatment with NMES after OST, while the participants in the control group received OST and sham–NMES only, twice weekly for 8 weeks.

Intervention

Treatment sessions began with OST. OST included various sensory stimuli applied to both groups the cheeks, chin, lips, tongue, and oral palate using human fingers, a vibrator, and an ice stick. The upper and lower lips were pressed with four fingers to apply sensory stimuli. Also, these areas nearby the lips were provided with sensory stimuli using an ice stick and a vibrator. In the same manner, various sensory stimuli were applied to the cheeks, chin, tongue, and oral palate 10 times each. After OST for 10 minutes, NMES was provided for 20 minutes to the experimental group. The NMES device was attached with surface electrodes to each participant in both groups, but the device was not turned on in the control group. The participants were sitting in an upright position in a chair with a headrest during the treatment sessions. During NMES, the occupational therapist facilitated voluntary swallowing by having the participant swallow a thickened liquid. The therapist sat facing the participant during the treatment session. The therapy room was separated from the public to help each participant concentrate on the treatment sessions without disturbances. Evaluations were conducted immediately prior to and after the experimental or control treatments.

NMES was provided with a dual-channel device (Simplus DP 200; Cybermedic Corp., Iksan, South Korea). The parameters of electrical stimuli were 80 Hz of 300 milliseconds with 1-second interval. For channel 1, two sets of electrodes were placed horizontally over the throat between the jaw and hyoid, approximating the belly of the digastric muscles. For channel 2, another two sets of electrodes were placed horizontally between the hyoid and thyroid notch, approximating the infrahyoid muscles. The current intensity was determined by palpation for muscle contraction. Muscle contraction was experienced without pain. The typical current level ranged from 3 mA to 5 mA.

Outcome measures

Behavioural Assessment Scale of Oral Functions in Feeding

The Behavioural Assessment Scale of Oral Functions in Feeding (BASOFF) was developed to measure variations of nine behaviours related to feeding, including jaw closure, lip closure over a spoon, tongue control, lip closure while swallowing, swallowing food without excess loss, chewing food (tongue/jaw control), sipping liquids, swallowing liquids without excess loss, and swallowing food without coughing (Ottenbacher, Dauck, Gevelinger, Grahn, & Hassett, 1985). The rating scale for each behaviour is from 0 to 5. A score of 0 represents a completely passive response to the feeding process, while a score of 5 indicates a functionally normal response. The interrater reliability coefficients for BASOFF were between .72 and .76. The test–retest reliabilities were .68 and .79 in the previous study on BASOFF. In this study, the interclass correlation coefficient (ICC) was .93 ($p < .05$). For ICC, 20% of the participants ($n = 4$) were evaluated independently by two occupational therapists.

American Speech–Language–Hearing Association's National Outcomes Measurement System swallowing scale

The American Speech–Language–Hearing Association (ASHA)'s National Outcomes Measurement System (NOMS) swallowing scale was used to determine the severity of dysphagia (Mullen, 2004; Mullen & Schooling, 2010; Schooling, 2003). The ASHA NOMS is a multidimensional tool developed to assess supervision and diet level, and represents functional status, which is rated on a scale from 1 to 7. Level 7 means that a participant has the ability to eat independently without limitation of swallowing function, while level 1 indicates that a participant does not have ability to swallow anything safely by mouth. To estimate the interrater reliability in the present study, ICC was computed from a set of swallowing level data collected independently by two occupational therapists using the ASHA NOMS swallowing scale. Twenty percent of the participants ($n = 4$) were evaluated to calculate ICC, which was .801 in this study ($p < .05$).

Data analysis

The Mann–Whitney *U* independent sample test and chi-square test were used to identify the potential baseline differences between groups. The Wilcoxon signed-rank test was used to evaluate changes between pre- and post-intervention groups. The differences in functional changes between the two groups were analysed by the Mann–Whitney *U* independent sample test. The level of statistical significance was set at $p < .05$. All data were analysed using Predictive Analytics Software 18 (IBM SPSS Software).

Results

The results of the chi-square test indicated that there was no significant difference in the distribution of sexes or types of CP between the groups ($p > .05$), which was confirmed by the chi-square test. The Mann–Whitney *U* independent sample test showed that there was no significant difference in age between the groups ($p > .05$). The demographics of the participants are summarised in Table 1.

The results are summarised in Table 2. There was no significant difference in oral function, including BASOFF and ASHA NOMS (control: 5.00 ± .94, NMES: 4.60 ± 1.07), between the groups in the pretest ($p > .05$). The total BASOFF score increased significantly in both groups ($p < .05$). The NMES group demonstrated a significant improvement in all subcategories, while the control group

Table 1 Demographic Characteristics of Participants (n = 20).

	Control (n = 10)	NMES (n = 10)
Age (y)	6.00 ± 2.40	6.20 ± 2.78
Sex		
Male	6	7
Female	4	3
Type of CP		
Hemiplegia	4	2
Diplegia	3	5
Quadriplegia	2	3
Flaccid	1	0

Note. CP = cerebral palsy; NMES = neuromuscular electrical stimulation; y = year.

showed no significant improvement in three subcategories, including jaw closure, sipping liquid, and swallowing liquid without excess loss ($p < .05$). In a comparison of functional changes between the groups, the NMES group demonstrated a significantly greater improvement than the control group in lip closure while swallowing, swallowing food without excess loss, sipping liquid, swallowing liquid without excess loss, swallowing without cough, and total score ($p < .05$). In the ASHA NOMS swallowing scale, there was no significant change between or within the groups.

Discussion

The present study was a pilot randomised trial investigating the therapeutic effect of NMES in CP with dysphagia. This study demonstrated that OST and NMES facilitated swallowing functions than OST and sham—NMES in children with CP and dysphagia. Thus, this study indicated that NMES might be an effective tool for dysphagia in the paediatric population. Some previous studies have demonstrated similar therapeutic effects on adults with dysphagia.

NMES demonstrated a significant improvement for swallowing function in the adult population. In a preliminary meta-analysis, NMES demonstrated positive overall effects on swallowing in dysphagia due to various aetiologies, including stroke, cancer, head trauma, and respiratory failure (Carnaby-Mann & Crary, 2007). The seven analysed studies, including two controlled trials and five pre—post trials, included a total of 255 patients. The absence of publication bias was revealed by a funnel plot. A significant summary effect size of NMES for swallowing was reported (Hedges g, .66; 95% confidence interval, .47—.85; $p < .001$). The average improvement in swallowing function across all seven studies was 20% after the NMES treatment. This preliminary meta-analysis concluded that NMES may be an effective treatment in the rehabilitation of dysphagia for an adult population. However, in paediatric populations with dysphagia, the effects of NMES are still controversial, and there are not sufficient studies (Christiaanse et al., 2011; Rice, 2012).

A retrospective study reported that NMES did not demonstrate a significant difference from oral-motor therapy in a paediatric population with dysphagia (Christiaanse et al., 2011). In this study, the treatment group included 47 participants, and the control group included 46 participants. The participants did not have homogeneous diagnoses, but various diagnoses such as Down syndrome, CNS anomalies, pulmonary disease, congenital heart disease, and preterm birth. The experimental group received weekly 30—45-minute NMES sessions of oral-motor stimulation or food ingestion, while the control group received weekly 45—60-minute sessions of oral-motor therapy. The results demonstrated that both groups significantly improved on the FOIS, but the amount of change between groups was not significant. Although this study was the report on the evaluation of NMES in a large

Table 2 Comparisons of Swallowing Functions Between and Within Groups (n = 20).

Outcome measure	Control (n = 10)		NMES (n = 10)		Functional changes	
	Pre	Post	Pre	Post	Control	NMES
BASOFF						
Jaw closure	3.60 ± 1.50	3.80 ± 1.39	3.60 ± 1.07	4.20 ± .78*	.20 ± .42	.60 ± .51
Lip closure over a spoon	2.90 ± 1.37	3.50 ± .85*	2.80 ± .91	3.80 ± .63*	.60 ± .96	1.00 ± .66
Tongue control	2.80 ± 1.31	3.20 ± 1.22*	2.90 ± .99	3.40 ± .69*	.40 ± .51	.50 ± .52
Lip closure while swallowing	2.70 ± 1.05	3.10 ± .99*	2.60 ± 1.07	3.50 ± 1.17*	.40 ± .51	.90 ± .56*
Swallowing food without excess loss	2.70 ± 1.05	3.10 ± .99*	2.60 ± 1.07	3.50 ± 1.17*	.40 ± .51	.90 ± 1.17*
Chewing food	2.50 ± 1.17	3.00 ± 1.15*	2.80 ± .78	3.40 ± .84*	.50 ± .52	.60 ± .51
Sipping liquid	2.80 ± 1.39	3.00 ± 1.63	2.60 ± .84	3.50 ± 1.17*	.20 ± .42	.90 ± .84*
Swallowing liquid without excess loss	2.80 ± 1.39	3.00 ± 1.63	2.60 ± .84	3.50 ± 1.17*	.20 ± .42	.90 ± .56*
Swallowing without cough	2.70 ± 1.05	3.10 ± .99*	2.60 ± 1.07	3.50 ± 1.17*	.40 ± .51	.90 ± .56*
Total score	25.6 ± 11.29	28.70 ± 10.84*	25.10 ± 8.64	32.30 ± 8.79*	3.30 ± 2.94	7.20 ± 3.22*
ASHA NOMS	5.00 ± .94	5.10 ± 1.10	4.60 ± 1.07	4.90 ± 1.10	.10 ± .70	.30 ± .48

Notes: Data are presented as mean ± SD. ASHA NOMS = American Speech—Language—Hearing Association's National Outcomes Measurement System swallowing scale; BASOFF = Behavioral Assessment Scale of Oral Functions in Feeding; NMES = neuromuscular electrical stimulation.
*$p < .05$.

paediatric population, the results indicated that NMES was equally effective with oral-motor interventions in the paediatric population. However, an additional analysis demonstrated that the FOIS level improved significantly more in children with acquired dysphagia than in the control group. This result may support the potential therapeutic effects of NMES on dysphagia in a paediatric population with acquired dysphagia.

A previous case series with five participants reported that NMES with oral-motor stimulation improved the swallowing function in children with pharyngeal dysphagia (Rice, 2012). The average age was 17 months (3—32 months). All participants were diagnosed with pharyngeal dysphagia due to multiple aetiologies, such as genetic defect, perinatal asphyxia, bronchopulmonary dysplasia, and failure to thrive. The 1-hour NMES sessions were performed twice a week. The number of treatment sessions varied across children from 11 to 63. The current intensity (0—25 mA) was adjusted for each participant to the maximum tolerated level. Between the NMES sessions, parents or caregivers were asked to provide oral-motor stimulation using z-vibe, nuk brushes, and flavoured chewy tubes. Modified barium swallow studies were performed to evaluate the swallowing function after the treatment sessions. In all five participants, NMES improved the swallowing function. Although there are some limitations, including varied treatment times for each child and multiple aetiologies, this case series provides support that NMES has positive effects on the swallowing function in children with pharyngeal dysphagia.

The present study showed that NMES had positive effects on the swallowing function related to the oral and pharyngeal phases. Swallowing is a complex motor performance, including voluntary and involuntary motor components through multiple neural networks from the oral phase through the oesophageal phase (Steele & Miller, 2010). Since appropriate sensory inputs affect both voluntary and involuntary motor outputs throughout the swallowing process, the therapist in a previous study guided voluntary swallowing by providing various sensory stimulations in paediatric patients (Arvedson et al., 2010b). Peripheral sensory stimulation, including NMES, influences multiple pathways, including both cortical and brainstem pathways, to trigger swallowing, alter motor output, and activate ascending pathways (Fraser et al., 2002; Hamdy et al., 1998; Steele & Miller, 2010). This neuroscientific evidence may be an explanation for the positive effects of NMES on swallowing function. Although NMES is more related to the pharyngeal phase of swallowing than to the oral phase, the present study demonstrated that NMES significantly improved function related to the oral phase, including lip closure while swallowing. The swallowing function is the integrated process of oral and pharyngeal phases; improved functions related to the pharyngeal phase might lead to an improvement in the oral phase, particularly the jaw closure. Clinically, NMES is a very practical intervention tool for dysphagia in the paediatric population. The development of NMES provides a therapeutic tool for occupational therapists to provide sensory stimulation on the pharyngeal area.

One of the limitations of this study was the small number of participants. Also, the distribution of CP type was different between the groups. And, we did not consider that the NMES score can increase due to maturation of participants before comparing the effects of NMES in two groups. In addition, the pharyngeal swallowing function was measured by an indirect method of observing the behaviour rather than a direct method, such as VFSS. Despite these limitations, the present study was the pilot study to demonstrate the positive therapeutic effects of NMES in CP. Future studies need to utilise VFSS for outcome measurements in a large participant group. Furthermore, various NMES protocols need to be investigated in paediatric populations with dysphagia.

Conclusion

We investigated the therapeutic effects of OST and NMES on the swallowing function in children with CP with dysphagia. The results demonstrated that NMES facilitated oral function related to swallowing, including lip closure while swallowing, ability to swallow food without excess loss, ability to sip liquid, ability to swallow liquid without excess loss, and ability to swallow without cough. Thus, this study suggested that NMES might be an effective therapeutic tool to facilitate OST for dysphagia on children with CP and dysphagia.

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