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Research Article

Neurofunctional Speech Rehabilitation in Cerebral Palsy: A Study on the Combination of Transcranial Direct Current Stimulation and an Integrative Speech Therapy Program

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Abstract

Purpose: The aim of this study was to evaluate the effects of the combination of an integrative speech therapy program (ISTP) and transcranial direct current stimulation (tDCS) on language in cerebral palsy.

Method: Eight children with cerebral palsy and speech disorders were allocated to two groups (active *vs*. sham) and underwent ten sessions combining ISTP with tDCS over Broca's area. The participants were assessed before and after the intervention with regard to aspects of speech, oral motor skills and cognition.

Results: Significant differences were found between the pre and post intervention evaluations with regard to the number of phonemes (p=0.0172), the percentage of consonants correct-revised on the imitation and naming lists (p=0.0209, p=0.0202) and percentage of consonants correct on the naming list (p=0.0433).

Conclusion: The combination of ISTP and tDCS can lead to improvements in speech rehabilitation among children with cerebral palsy.

Keywords: Speech therapy; Brain stimulation; Cerebral palsy; Neurorehabilitation; Transcranial direct current stimulation

Introduction

Transcranial direct current stimulation (tDCS) is a non-invasive brain stimulation technique administered to patients with different neurological and psychiatric disorders. By reaching the brain cortex, the effect of a low electrical current can either facilitate or inhibit neuronal excitability, depending on electrode polarity [1,2].

Recent trials report that the neurophysiological effects of tDCS combined with behavioral training may enhance the rehabilitation process following a brain injury [3] Prominent neurorehabilitation studies have combined tDCS with motor training for children with cerebral palsy [4,5].

Cerebral palsy is an impairment of global motor development due to brain damage in the early stages of childhood, resulting in complex clinical symptoms, such as impaired gross motor function and communication [6]. Communication disorders related to oral motor impairment are secondary to deficits in sensory-motor brain control and are found in 40% of children with cerebral palsy [7,8]. The speech process is based on cortical linguistic and motor planning associated with Broca's area of the brain. As the motor area is affected in cerebral palsy, children with this disease can have difficulties in executing speech as well as planning of the associated motor acts. Thus, the aim of speech therapy for this population is to rehabilitate oral movements using peripheral exercises as well as enhance the planning of motor acts and the learning process through cortical stimulation [9,10].

Although there is evidence that the combination of anodal tDCS and speech therapy have promising effects [11], no previous study has been performed involving children with cerebral palsy. Thus, the aim of this pilot study was to compare the effects of anodal and sham tDCS combined with an integrative speech therapy program (ISTP) on speech skills in children with cerebral palsy. The hypothesis was that anodal tDCS combined with ISTP provides better results with regard to speech in children with cerebral palsy than the combination of sham tDCS and ISTP.

Methods

This randomized, sham-controlled, double-blind study received approval from the Human Research Ethics Committee of the Federal University of São Paulo (Brazil) under process number 25914514.6.0000.5505/2014 and was conducted in accordance with the ethical standards laid down in the Declaration of Helsinki [12]. All participants enrolled in the study provided written informed consent signed by a parent or guardian.

Participants

Children diagnosed with spastic cerebral palsy were recruited from the community as well as the Human Communication Disorders clinic of the university. The following were the inclusion criteria: a)

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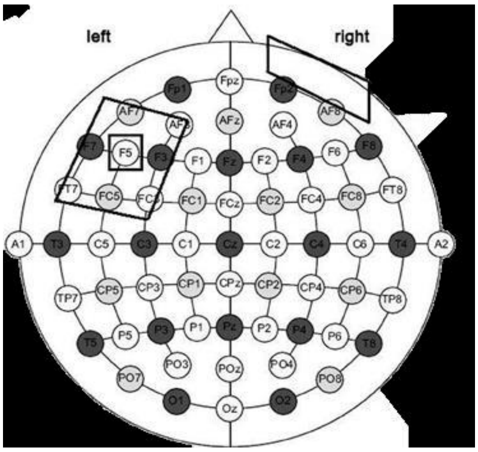

Figure 1: Parameters of the tDCS montage: anode over the Broca area and supra orbital contralateral cathode.

age five to eighteen years; b) speech disorders classified on levels II and III of the Viking Speech Scale [13] and levels II, III and IV of the Communication Function Classification System [14] and c) ability to repeat the oral patterns required for myofunctional orofacial therapy. The following were the exclusion criteria: a) hearing loss; b) visual impairment; c) history of seizures; d) metallic implant in the brain or hearing aids; and e) surgical procedure or neurolytic block in the 12 months prior to the beginning of the sessions.

After screening, the participants were randomly allocated to two groups: a control group submitted to ISTP combined with sham tDCS and an experimental group submitted to ISTP combined with active anodal tDCS. Simple randomization was performed with the allocation duly sealed in sequentially numbered opaque envelopes. A staff member not involved in the recruitment or development of the trial performed the randomization process.

Assessments

Participants were evaluated before and after the intervention protocol on two non-consecutive days, by a blinded speech therapist regarding their allocation group. The assessments covered three areas: a) quality of speech, communication and language; b) oral movements; c) cognitive aspects.

Quality of speech, communication and language: We evaluated using standardized quantitative scales. The Viking Speech Scale [13] classifies the speech production of children aged four years and older on four levels. Level I represent good communication quality and level IV represents severe communication impairment characterized by incomprehensible speech. The Communication Function Classification System [14] is used to assess communication functionality (sending and receiving) according to parents and unusual listeners. This scale has five classification levels. Level I corresponds the execution of the proper rules of communication between partners in any environment and level V identifies children with ineffective communication who are not understood by family members or unusual listeners and do not understanding others.

The phonological section of the ABFW Child Language test [15] was employed for the assessment of speech skills. This test is used to assess phonological organization through the naming of a list of figures and repeating a list of words. Both lists are phonetically balanced for Brazilian Portuguese. The percentage of consonants correct (PCC) and percentage of consonants correct-revised (PCCR) indices [16] were used for the samples of speech. The PCC and PCCR indices are validated for speech analysis and respectively demonstrate the number of correct consonants with and without distortion.

Oral motor skills: We assessed using the myofunctional orofacial assessment protocol (AMIOFE) with scales proposed by de Felício & Ferreira [17].This protocol allows the analysis of aspects of the stomatognathic system, such as jaw position, mobility and functions, for the characterization of muscle and functional conditions on the basis of scores determined by the presence/absence of a myofunctional disorder as well as the degree of disorder.

Cognitive aspects: Peabody vocabulary test [18] was used to asses cognitive skills. It assess receptive auditory language among children aged from two years and six months to [18] years. On this test, the subjects indicate the figure corresponding to the word they have just heard. The results are presented in scores that classify cognitive aspects according to age. This evaluation served as a safety measure to ensure the absence of cognitive impairment during the stimulation process.

The first outcome was measured by the phonological section of the ABFW Child Language test [15]. Secondary outcomes were measured by the AMIOFE [17] protocol.

Intervention

The therapeutic interventions were conducted by speech therapist. The protocol was performed in five weekly 30-minute sessions for two weeks (total: ten sessions). Each session was divided into two phases: motor global exercises for the first ten minutes and an integrative speech therapy program (ISTP) combined with anodal or sham tDCS during the remaining twenty minutes. Stimulation was administered over Broca's area, which are the part of the brain responsible for symbolic and linguistic aspects as well as the motor planning of speech.

The ISTP is a method that employs global and oral motor exercises to train and improve speech skills by integrating phonological and cognitive therapies. Global motor exercises were performed in the first ten minutes of therapy. For such the homolateral Padovan method was employed, which is based on neuro-evolutive development [9]. This method uses physical exercises with simultaneous auditory and linguistic stimulation. Myofunctional orofacial and phonological therapy was performed during the last twenty minutes. This part of the therapy was focused on the stimulation of the sensory-motor skills of speech. Basic speech functions were trained using the following exercises: 1) pneumophonic coordination – with oral and nasal breathing as well as the emission of vowels; 2) Suction stimulation – the oral intake of liquids through a thin catheter and an orthodontic pacifier, with the movements monitored and corrected by an experienced speech therapist; 3) Chewing – the child was encouraged to perform unilateral and bilateral chewing using a rubber tourniquet strap, with a focus on strength and function; 4) Swallowing – with liquid and a toothpick stimulus to position the tongue; 5) Language and Speech – stimulation of phonemes through sensory integration and phonology. The therapist provided motor, auditory, visual and tactile stimulation for the correct execution using figures, context and phonological oppositions [19].

Transcranial Direct Current Stimulation*:* anodal tDCS was performed for twenty minutes in each session during the ISTP. Stimulation was applied with a current of 1 mA over Broca's area to enhance behavioral learning through the development of a favorable neural environment. Stimulation was applied using a tDCS device (Soterix, Soterix Medical, USA) with two sponge surface electrodes (5 x 5 cm^2) soaked in saline solution (Figure 1). As stated previously, children were randomly assigned to one of two treatments: anodal tDCS and sham stimulation. The anodal electrode was placed over the left Broca's area (Brodmann area 44/45) following the International [10-20] System for electroencephalography (F7) and the cathode was placed over the contralateral supraorbital region [20]. For sham stimulation, the electrode placement procedures were the same, but the stimulator was switched on only for the first thirty seconds, giving the children the initial sensation of stimulation to ensure blinding and no electrical current was administered throughout the rest of the session. This is a valid control method with no neuromodulatory effects [4].

Statistical analysis

Continuous variables were expressed as mean and standard deviation values and categorical variables were expressed as frequency values. Descriptive statistics were employed for the clinical and demographic data. As the Shapiro-Wilk test demonstrated that the data had non- parametric distribution, the results of the combination of ISTP and tDCS (active *vs.* sham) were analyzed using the two-sample Wilcoxon-Mann-Whitney test. The paired-sample Wilcoxon signed rank test was used to compare result within groups. For the primary and secondary outcomes, changes in each score were calculated as the difference in pre-intervention and post- intervention means between groups (tDCS *vs.* sham) using the two-sample Wilcoxon-Mann- Whitney test. The effect size was determined using a nonparametric technique that divides the squared Mann-Whitney U value by group size [21]. A p-value <0.05 indicated a statistically significant result. The data were organized and analyzed with the aid of the Statistical Package for Social Sciences (SPSS, version 19.0, SPSS, Inc., Chicago, IL, USA).

Results

Fifteen children were screened for the present pilot study. Eight were eligible and randomly allocated to either the experimental (anodal tDCS + ISTP) or control (sham stimulation + ISTP) group. Of the seven subjects that were not eligible for the clinical trial, three had no speech impairment and four did not have the cognitive ability to repeat the required motor patterns. All patients completed the intervention protocols and assessments.

Table 1 displays the demographic and clinical characteristics of the groups. No significant differences were found with regard to patient characteristics at baseline (p≥0.05). No severe adverse events occurred during the study, although four children who received active stimulation and two in the sham group reported a tingling sensation under the electrodes. The Peabody cognitive test revealed no significant differences before and after the intervention in the experimental group (p=0.68) (Table 2).

Table 1: Anthropometric character ISTPics and speech classification of children studied.

Experimental group (N=4)	Control group (N=4)	
atDCS + speech therapy	Sham tDCS + speech therapy	
Age (ys)*	8.3(3.9)	9.3(4.1)
Gender (F/M)**	2/2	1/3
VSS (I/II/III)**	1/3	2/2
CFCS (I/II/III)**	3/1	2/2
Peabody	77.0(75.5)	96.7(57.3)

Table 2: Peabody test results before and after the intervention.

Experimental Group		Control Group		
Before I	77	(75,51)	96	(57,3)
After I	78	(74,71)	98	(56,5)

Table 3A: Comparison of differences between pre- and post-intervention values of the number of phonemes, PPC-Nom, PCCR-Nom, PCCR-Im.

Control Group			Experimental Group		
					p
Phonemes	0,50	(1,00)	4,25	(0,95)	0,017*
PCC-Nom	3,39	(3,00)	10,03	(5,28)	0,057
PCC-Im	4,02	(3,30)	11,02	(9,32)	0,200
PCCR-Nom	4,42	(2,14)	17,01	(5,70)	0,029*
PCCR-Im	4,01	(3,04)	18,61	(6,07)	0,028*

Values are reported as mean ± standard deviation; PCC- Non: Percentage of Consonants Correct - Nomination; PCCR -Nom: Percentage of Correct Consonants Revised - Nomination; PCCR-Im: Percentage of Correct Consonants Revised - Imitation; (1) Wilcoxon rank-sum test (Mann-Whitney test) comparing the U difference (pre-post) between groups; *P <0.05.

Table 3B: Comparison intra-group (CG): before and after intervention.

Before			After		
					p
Nº Fonemas	14,50	9,71	15,00	8,71	0,317
PCCNom	63,01	42,02	66,40	44,30	0,109
PCCIm	62,04	41,47	66,07	44,12	0,109
PCCRNom	64,58	43,06	69,00	41,85	0,066▪
PCCRIm	65,39	43,61	69,41	40,95	0,068▪

*significance level 5%; ▪significance level 10%.
Values are reported as mean ± standard deviation; PCC- Non: Percentage of Consonants Correct - Nomination; PCCR -Nom: Percentage of Correct Consonants Revised - Nomination; PCCR-Im: Percentage of Correct Consonants Revised - Imitation; (1) Wilcoxon ran.

Primary outcomes

Quality of speech assessed using the ABFW language test was the primary outcome. The experimental group performed better than the control group with regard to the difference between after and before intervention in PCC-R on the imitation and naming lists (U=-2.31; p=0.02) and PCC on the naming list (U=-2.02; p=0.04) (Table 3A,B). The results were also significant for the number of phonemes executed (U=-2.38; p=.02) (Table 3C).

Secondary outcomes

Table 4,5 displays the results of the myofunctional orofacial assessment. No statistically significant differences were found

Table 3C: Comparison intra-group (EG): before and after intervention.

	Before			After	
		p			
Nº Fonemas	4,25	5,31	8,5	5,802	0,065▪
PCCNom	14,80	25,72	24,89	30,21	0,69
PCCIm	14,70	26,04	25,77	34,91	0,68
PCCRNom	24,73	30,15	41,55	33,04	0,067▪
PCCRIm	27,95	33,04	46,57	37,51	0,06

k-sum test significance level 5%; ▪significance level10%.
Values are reported as mean ± standard deviation; PCC- Non: Percentage of Consonants Correct - Nomination; PCCR -Nom: Percentage of Correct Consonants Revised - Nomination; PCCR-Im: Percentage of Correct Consonants Revise- Imitation; (1) Wilcoxon rank-sum test.

Table 4: Effect obtained in AMIOFE protocol regarding posture, mobility and function in the control group.

AMIOFE Items		Before			After	
Max Score	Mean	SD		Mean	SD	p
			Posture			
Lips	3	1,75	0,957	1,75	0,957	1,00
Jaw	3	0,75	0,957	0,75	0,957	1,00
Cheeks	3	1	1,414	1	1,414	0,670
Facial Simetria	3	0,5	0,577	0,5	0,577	0,537
Tongue	3	0,75	0,957	0,75	0,957	0,670
Hard Palate	3	0,5	0,557	0,5	0,577	0,537
			Mobility			
Lips	12	5,5	4,123	5,75	3,774	0,336
Tongue	18	5,5	3,109	6,25	3,304	0,502
Jaw	15	6,0	3,366	6,0	3,366	0,106
Cheeks	12	4,75	3,593	4,75	3,593	0,283
			Function			
Breath	3	0,5	0,577	0,5	0,577	0,537
Swallow	15	3,75	3,304	3,75	3,304	0,172
Chew	10	6,5	5,507	6,5	5,507	0,399

Wilcoxon Test; *p <0.05. Values are reported as mean ± standard deviation. Higher scores indicate positive performance.

between groups (p≥0.05). In the analysis of the phonemic framework, phonemes were considered a secondary outcome.

Discussion

This study provides important preliminary results regarding the effects of anodal tDCS administered over Broca's area in children with cerebral palsy. No previous studies were found exploring the potential of anodal stimulation to optimize the effects of speech therapy in this population. Despite the small sample size, the results suggest that anodal tDCS can contribute significantly to improving speech in the naming and imitation domains.

Speech disorders in children with cerebral palsy can affect neurodevelopment. Most learning and communication skills in such patients are compromised by speech and language disorders. Thus, rehabilitation processes are essential to developing the functions affected by the brain damage and contributing to both independence and improved quality of life [8].

Table 5: Effect obtained in AMIOFE protocol regarding posture, mobility and function in the experimental group.

AMIOFE Items		Before			After	
Max Score	Mean	SD		Mean	SD	p
			Posture			
Lips	3	1,75	0,957	2	0,816	0,704
Jaw	3	0,75	0,957	1	0,816	0,704
Cheeks	3	1,5	1,732	1,5	1,732	0,670
Facial Simetria	3	0,75	0,5	0,75	0,5	0,537
Tongue	3	0,5	0,577	0,5	0,577	0,670
Hard Palate	3	0,25	0,5	0,25	0,5	0,537
			Mobility			
Lips	12	3,25	1,258	4,5	1,914	0,576
Tongue	18	4	2,828	7,5	3,415	0,617
Jaw	15	2,25	2,061	2,25	2,061	0,106
Cheeks	12	2,5	1.290	3,25	2,061	0,496
			Function			
Breath	3	0,25	0,5	0,25	0,5	0,818
Swallow	15	1	0,816	1,5	1,290	0,251
Chew	10	4	0,042	4	0,042	0,399

Wilcoxon Test; *p <0.05. Values are reported as mean ± standard deviation. Higher scores indicate positive performance.

Recent studies have described advances in the use of neuromodulatory techniques regarding motor and cognitive rehabilitation for patients with brain damage. Although few trials have been conducted with the pediatric population, which limits a more detailed discussion, studies involving adults with post-stroke aphasia have shown the positive effects of tDCS over Broca's area, which may be associated with improved language skills [22,23].

Broca's area is responsible for symbolic speech motor planning [24]. This cortical area is activated when it is necessary to evoke an object or idea and turn it into movement of the speech organs to produce a verbal expression. Previous studies relate Broca's area to nomination and its excitability or inhibition is associated with changes in the speech process [11]. The present findings are in agreement with these data, as neuromodulation over Broca's area on the left side combined with integrative speech therapy led to significant improvements in motor speech activity and phonologic organization [25], as demonstrated by the improvement in the naming activity as well as tongue and lip movements related to speech (Table 4,5).

Speech disorders in children with cerebral palsy may be related to motor changes resulting from brain damage. A lack of muscle coordination can affect speech accuracy and verbal fluency. These changes occur in early childhood and often result in deficiencies regarding the acquisition of phonemes, which is essential to communication through speech. Therefore, the aim of the present study was to analyze the effects of a technique that combines neurofunctional therapy and phoneme deployment through multisensory stimuli [9,19]. This technique incorporated neuro-evolutive movements to provide a standard of motor learning.

Similarly, the resumption of the early movements of sucking, chewing and swallowing is recommended to improve oral motor skills [9]. Concurrently to the training of motor patterns, phonemes were trained through multisensory (tactile, visual, auditory and proprioceptive) stimuli. To achieve an adequate motor pattern, the perception of movement is trained through sensory feedback, which assists the learning process on the cortical level. The a priori belief the use of therapy focused on motor and phonologic skills would contribute positively to language rehabilitation was confirmed by the within-group results, as both groups showed improvements at the end of the ten intervention sessions.

The phonetic framework was analyzed before and after the intervention using the ABFW test and spontaneous conversation. The difference between groups was significant, indicating an improvement in the acquisition of new phonemes by the children submitted to active tDCS. The bilabial-plosive (/ p /, / b /) and alveolar-plosives (/ t /, / d /) phonemes were acquired more, which are easier to recognize through tactile and visual perceptions. The findings suggest that the results of the ISTP were potentiated by cortical anodal tDCS over Broca's area [26].

Despite the lack of a statistically significant difference, a clinical improvement was found in tongue mobility in the active tDCS group, which contributed to the acquisition of alveolar- plosives phonemes (Table 5). It should be stressed that ten sessions performed over two consecutive weeks can be considered a relatively short period to observe significant changes in patients with brain damage and therefore may have been too short a time to modulate speech function. Further studies are needed with a larger population as well as longer intervention protocols and follow-up assessments.

The major limitation of the present study was the small sample size. This occurred due to the difficulty recruiting children with cerebral palsy and speech disorders. However, the main importance is the originality of the topic. No published articles were found that combine motor speech therapy with tDCS in children with cerebral palsy. Another limitation was the difference in the communication level and age of the subjects. However, both groups were balanced, which validates the interpretation of the results of this pilot study. Moreover, the present findings can give direction to phase [2] studies seeking to standardize a neuromodulatory rehabilitation protocol in the field of speech disorders for children with brain damage.

Conclusion

Based on the present results, the combination of anodal tDCS over Broca's area and an integrative speech therapy program has a significant effect on the rehabilitation of speech in children with cerebral palsy, as demonstrated by the increase in the number of acquired phonemes and the percentage of consonants correct-revised. These findings can serve as a basis for subsequent randomized, controlled, clinical trials with the goal of extending the protocol and its features.

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