

Treadmill training combined with transcranial direct current stimulation over the primary motor cortex in a child with cerebral palsy and hydrocephalus: A case report.

Treino de marcha em esteira combinado com a estimulação transcraniana por corrente contínua sobre o córtex motor primário em uma criança com paralisia cerebral e hidrocefalia: relato de caso.

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Abstract

Introduction: Transcranial direct current stimulation (tDCS) is a promising technique that stimulates the cortex with a direct, low-intensity electric current and can potentiate motor learning. **Objective:** Describe the results of an intervention protocol involving anodal stimulation over the primary motor cortex combined with treadmill training in a child with cerebral palsy. **Method:** The intervention was comprised of ten sessions of anodal tDCS (1mA) over the primary motor cortex during the treadmill training. Stabilometric analysis was evaluated one week before and one week after the intervention. **Results:** A reduction in oscillations of the COP was found under both conditions (eyes opened and eyes closed). **Conclusion:** Our findings suggest that anodal tDCS over primary motor cortex can potentiate the results of treadmill training.

Keywords: Children, Balance, Transcranial Direct Current Stimulation.

Resumo

Introdução: Estimulação transcraniana por corrente contínua (ETCC) é uma técnica promissora que estimula o córtex com uma corrente elétrica direta, de baixa intensidade e pode potencializar a aprendizagem motora. **Objetivo:** Descrever os resultados de um protocolo de intervenção envolvendo estimulação anódica sobre o córtex motor primário combinado com treino em esteira em uma criança com paralisia cerebral. **Método:** A intervenção foi composta por dez sessões de anodal ETCC (1 mA) sobre o córtex motor primário durante o treino em esteira. Análise estabilométrica foi avaliada uma semana antes e uma semana após a intervenção. **Resultados:** Uma redução na oscilação do COP foi encontrado em ambas as condições (olhos abertos e olhos fechados). **Conclusão:** Nossos resultados sugerem que a ETCC anódica acima do córtex motor primário pode potencializar os resultados do treinamento em esteira.

Palavras chaves: Crianças, equilíbrio, estimulação transcraniana por corrente contínua.

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INTRODUCTION

Cerebral palsy (CP) refers to permanent, but modifiable motor development disorders stemming from a primary brain injury that causes secondary musculoskeletal abnormalities.⁽¹⁾ Hydrocephalus can be a cause of CP and is characterized by a disturbance in the circulation of cerebrospinal fluid, causing the intraventricular accumulation of fluid that result in progressive ventricular dilatation.⁽²⁾ Endoscopic third ventriculostomy provides direct communication between the third ventricle and subarachnoid spaces and is considered the treatment of choice for hydrocephalus when the child has a favorable neuroanatomy.⁽³⁻⁵⁾

Motor impairment secondary a brain injury generally results in abnormal biomechanics of the body.⁽⁶⁾ Balance regards postural control related to stability and body orientation, including control among body segments.⁽⁷⁾ Stability depends on the integration of the visual, vestibular, somatosensory and motor systems.⁽⁸⁾ Many patients with neurological dysfunction have difficulty maintaining an equilibrium among these systems, which leads to poor postural stability.⁽⁹⁾ Motor impairment in children with CP is related to poor postural control, which exerts an extensive negative impact on activities of daily living.^(7, 10)

Intensive physiotherapy is considered the gold standard for the treatment of children with delayed motor development and inadequate balance. Treadmill training is among the therapies currently studied for this population and evidence shows that such training is extremely promising for children less than six years of age,⁽¹¹⁾ resulting in improvements in gait pattern, gross motor function and balance.^(12, 13)

The development of new therapeutic techniques in physical rehabilitation programs is important to enhancing functional outcomes.⁽¹⁴⁾ Transcranial direct current stimulation (tDCS) is a promising, easily administered, well-tolerated technique involving inexpensive equipment and minimal adverse effects. This method stimulates the cortex with a direct, low-intensity (1 to 2 mA) electric current and two electrodes.⁽¹⁵⁾ Anodal and cathodal stimulation respectively enhance and inhibit cortex excitability. The neurophysiological effects of tDCS can potentiate motor learning.⁽¹⁶⁾ Recent studies have shown that anodal tDCS over the primary motor cortex during gait training has positive effects on postural control, gait and balance in children with cerebral palsy.^(14,17) However, these studies involved children with brain injuries resulting from deficiencies in cerebral blood flow and no found studies are found on this combination in children with motor impairment resulting from a brain injury caused by a congenital disorder, such as congenital hydrocephalus. Thus, the aim of this paper was to describe the results of an intervention protocol involving anodal tDCS over the primary motor cortex combined with tre-

admill training in a child with cerebral palsy and congenital hydrocephalus with consequent diparesis and postural control deficit.

Case report

This case involved a female child aged five years (height 106cm; body mass 16.5kg) with a diagnosis of cerebral palsy and congenital hydrocephalus with subsequent spastic diparesis, classified on level I of the Gross Motor Function Classification System.⁽¹⁸⁾ The child was born by cesarean birth due the prenatal diagnosis of congenital hydrocephalus. Following birth, hydrocephalus was treated using third endoscopic ventriculostomy.

The child had significant developmental delays. She began to control her head at six months of age, control the trunk and sitting posture at ten months, walk with support at 18 months and walk without support at 24 months. During the study period, the child walked independently with a discreet diparetic gait pattern and had difficulty walking in community settings. The main complaint of the parents was frequent falls, which interfered with the child's independence.

At the initial evaluation, the child exhibited grade 1 spasticity according to the Modified Ashworth Scale,⁽¹⁹⁾ full passive range of motion, grade three muscles strength in the flexors and grade four in the extensors of the lower limbs. After discussion with the medical staff, an evaluation of brain magnetic resonance imaging and electroencephalography, exclusion criteria for conducting the experimental intervention (tDCS combined with treadmill training) were discarded.

The intervention was comprised of ten 20-minute sessions of treadmill training without incline of the treadmill. Training speed was determined in each session and considered ideal when the child was able to walk comfortably with no signs and symptoms of fatigue and perform proper footing during the stance phase of gait. Stimulation involved a transcranial stimulation device (Soterix Medical Inc., USA) with two sponge (non-metallic) electrodes (5 x 5 cm) moistened with saline solution. The anodal electrode was positioned over the primary motor cortex of the non-dominant hemisphere following the 10-20 International Electroencephalogram System⁽²⁰⁾ and the cathode was positioned in the supra-orbital region on the contralateral side.

The child was evaluated one week before and one week after the intervention. Stabilometric analysis was performed for the evaluation of static balance. For such, a force plate (Kistler model 9286BA) was used, which allows the record of oscillations of the center of pressure (COP). The acquisition frequency was 50 Hz, captured by four piezoelectric sensors positioned at the extremities of the force plate, which measured 40 x 60 cm. The data were recorded and interpreted using the SWAY software program (BTS Engineering), integrated and syn-

chronized to the SMART-D 140® system. The child was instructed to remain in a standing position on the force plate, barefoot, arms alongside the body, with an unrestricted foot base, heels aligned and gazed fixed on a point marked at a distance of one meter at the height of the glabellum. Thirty-second readings were taken under two conditions: eyes open and eyes closed. Displacement of the COP was measured in the anteroposterior (x axis) and mediolateral (Y axis) directions under each visual condition.⁽¹⁷⁾

The child appeared for all intervention sessions and demonstrated excellent tolerance to the procedures. No adverse effects were reported or observed by the researchers or family. In all sessions, the child reported a tingling sensation from the current in the region below the anode electrode. The child needed no rest periods during the gait training. Table 1 shows the results of the evaluations before and after the intervention. A reduction in the oscillations of the COP was found under both conditions (eyes opened and eyes closed).

Figures 1A (Before) and 1B (After) show the stabilometric findings and illustrate the oscillations of the COP with eyes open before and after the intervention. The reduction in oscillations following the intervention demonstrates an improvement in static balance.

DISCUSSION

The aim the present study was to investigate whether ten sessions of anodal tDCS over primary motor cortex combined with gait training could potentiate the effects of treadmill training on static balance in a child with cerebral palsy stemming from hydrocephalus. The findings revealed a promising improvement in balance, as demonstrated by the reduction in the oscillations of the COP with and without visual restriction.

Studies involving the combined use of tDCS and gait training in children with cerebral palsy have been published in the last year and report the positive effects of this combination, especially with regard to spatiotemporal gait variables and a reduction in the oscillations of the COP.^(14, 17) The experimental intervention in the present case report was based on these studies and involved the same protocol. Thus, the authors expected to find promising results. A previous study found that a single session of anodal tDCS in combination with gait and balance training resulted in an improvement in body control and gait velocity.^(21, 22) In a study involving patients with hemiparesis stemming from a stroke, three sessions of anodal stimulation were performed over the affected motor cortex combined with a specific motor training for the paretic ankle, resulting in an improve-

Table 1. Results before and after transcranial direct current stimulation in combination with treadmill training in a child with hydrocephalus.

Center of pressure oscillations	Before	After	Effect
Anteroposterior with eyes open(mm)	38.2	12.2	-26.0 (68.0%)
Mediolateral with eyes open(mm)	76.2	25.5	-50.7(66.5%)
Anteroposterior with eyes closed(mm)	54.7	26.4	-28.3(51.7%)
Mediolateral with eyes closed(mm)	65.1	38.4	-26.7(41.0%)

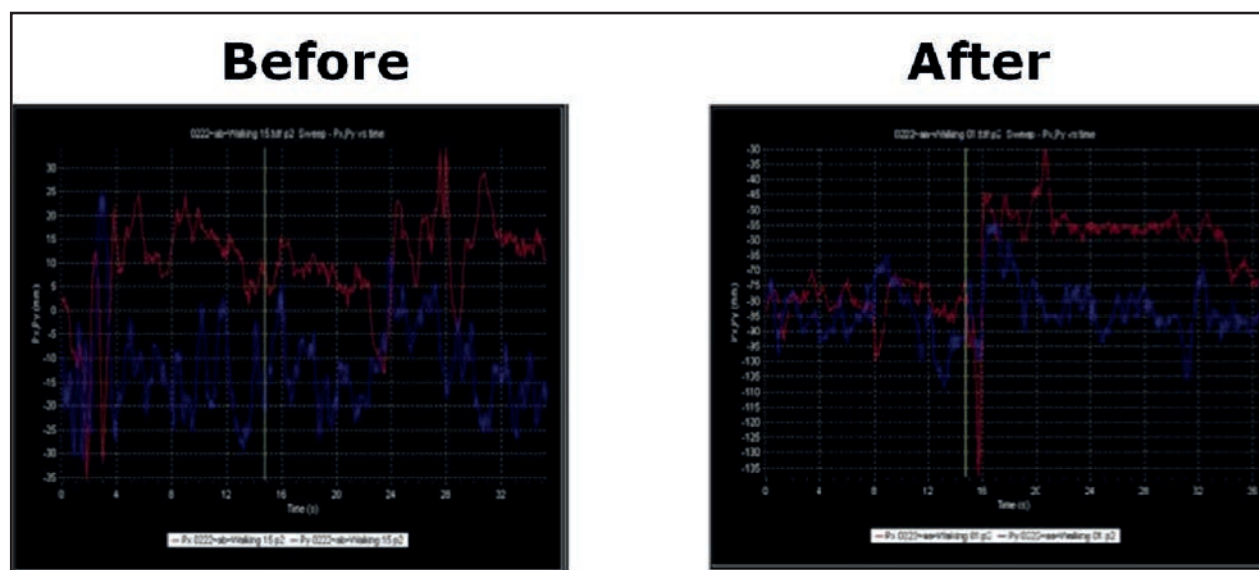


Figure 1. Oscillations of the center of pressure with eyes open before and after anodic transcranial direct current stimulation and treadmill training.

ment in dorsiflexion and plantar flexion movements.⁽²³⁾ The present findings are in agreement with the study cited, as ankle movements constitute a major mechanism in balance and postural control.

The main reason for reporting this case regards the cause of motor impairment, as brain damage was likely secondary to congenital hydrocephalus. The pathophysiology of hydrocephalus may involve compressive brain lesions, with a more global impairment of brain structures. As the brain damage had a non-specific location, it is not possible to affirm that the child had a lesion specifically in the primary motor cortex.

The definition of the area to be stimulated was based on clinical symptoms. In general, spastic diparesis is caused by lesions in the pyramidal system. Thus, the decision was made to perform anodal tDCS over the primary motor cortex, which is an important region that can facilitate the reorganization of the brain. Previous studies report that anodal stimulation of this area can enhance functional outcomes and motor learning due to the potentiation of neuroplastic changes.⁽²⁴⁾

An important fact regarding tDCS is the prolonged effect of gait training. In clinical practice, the effects of physical therapy are minimized or completely lost following long periods without therapy. Kashi et al.⁽²⁵⁾ evaluated 30 healthy volunteers who received five 15-minute sessions of anodal stimulation (2 mA, active and placebo) over the prefrontal cortex during locomotion training. The active group showed improved postural control and increased gait velocity in comparison to the pla-

cebo group, demonstrating that anodal tDCS is capable of causing changes in cortex excitability, thereby favoring motor control.

The more important limitation this study is a description of a single case. Conclusions regarding the efficacy of combined treatment are limited due to the case report design. In this case, the tDCS protocol combined with treadmill training demonstrated promising effect in this child with diparesis and hydrocephalus. Spontaneous improvement is possible in this case report, but the magnitude of the changes in static balance suggest that the combination of treadmill training and anodic tDCS made an important contribution.

CONCLUSION

In the present case, an improvement was found in static balance after ten sessions of stimulation, with a reduction in the oscillations of the center of pressure in the anteroposterior and mediolateral directions, suggesting that anodal tDCS over primary motor cortex can potentiate the results of treadmill training.

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REFERENCES

1. Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Damiano D, et al. A report: the definition and classification of cerebral palsy April 2006. *Dev Med Child Neurol Suppl.* 2007;109(suppl 109):8-14.
2. Jucá CEB, Lins Neto A, Oliveira RSd, Machado HR. Tratamento de hidrocefalia com derivação ventrículo-peritoneal: análise de 150 casos consecutivos no Hospital das Clínicas de Ribeirão Preto. *Acta cir bras.* 2002;17(supl. 3):59-63.
3. Warf BC. Hydrocephalus in Uganda: the predominance of infectious origin and primary management with endoscopic third ventriculostomy. *Journal of Neurosurgery: Pediatrics.* 2005;102(1):1-15.
4. Warf BC, Mugamba J, Kulkarni AV. Endoscopic third ventriculostomy in the treatment of childhood hydrocephalus in Uganda: report of a scoring system that predicts success: Clinical article. *Journal of Neurosurgery: Pediatrics.* 2010;5(2):143-8.
5. Kulkarni AV, Drake JM, Mallucci CL, Sgouros S, Roth J, Constantini S. Endoscopic third ventriculostomy in the treatment of childhood hydrocephalus. *The Journal of pediatrics.* 2009;155(2):254-9. e1.
6. Kavčič A, Vodušek D. A historical perspective on cerebral palsy as a concept and a diagnosis. *European journal of neurology.* 2005;12(8):582-7.
7. Rose J, Wolff DR, Jones VK, Bloch DA, Oehlert JW, Gamble JG. Postural balance in children with cerebral palsy. *Developmental Medicine & Child Neurology.* 2002;44(01):58-63.
8. Berg K. Measuring balance in the elderly: preliminary development of an instrument. *Physiotherapy Canada.* 1989;41(6):304-11.
9. Roque AH, Kanashiro MG, Kazon S, Grecco LAC, Salgado ASI, Oliveira CSd. Analysis of static balance in children with cerebral palsy spastic diparetic type with and without the use of orthoses. *Fisioterapia em Movimento.* 2012;25(2):311-6.

10. de AC Duarte N, Grecco LAC, Franco RC, Zanon N, Oliveira CS. Correlation between Pediatric Balance Scale and functional test in children with cerebral palsy. *Journal of Physical Therapy Science*. 2014;26(6):849.
11. Valentin-Gudiol M, Bagur-Calafat C, Girabent-Farrés M, Hadders-Algra M, Mattern-Baxter K, Angulo-Barroso R. Treadmill interventions with partial body weight support in children under six years of age at risk of neuromotor delay: a report of a Cochrane systematic review and meta-analysis. *European journal of physical and rehabilitation medicine*. 2013;49(1):67-91.
12. Grecco LAC, Zanon N, Sampaio LMM, Oliveira CS. A comparison of treadmill training and overground walking in ambulant children with cerebral palsy: randomized controlled clinical trial. *Clinical rehabilitation*. 2013;0269215513476721.
13. Grecco LAC, Tomita SM, Christovão TC, Pasini H, Sampaio LM, Oliveira CS. Effect of treadmill gait training on static and functional balance in children with cerebral palsy: a randomized controlled trial. *Brazilian journal of physical therapy*. 2013;17(1):17-23.
14. Grecco LAC, Duarte NdAC, Mendonça ME, Cimolin V, Galli M, Fregni F, et al. Transcranial direct current stimulation during treadmill training in children with cerebral palsy: A randomized controlled double-blind clinical trial. *Research in developmental disabilities*. 2014;35(11):2840-8.
15. Brunoni AR, Nitsche MA, Bolognini N, Bikson M, Wagner T, Merabet L, et al. Clinical research with transcranial direct current stimulation (tDCS): challenges and future directions. *Brain stimulation*. 2012;5(3):175-95.
16. Nitsche MA, Schauenburg A, Lang N, Liebetanz D, Exner C, Paulus W, et al. Facilitation of implicit motor learning by weak transcranial direct current stimulation of the primary motor cortex in the human. *Journal of Cognitive Neuroscience*. 2003;15(4):619-26.
17. Duarte NdAC, Grecco LAC, Galli M, Fregni F, Oliveira CS. Effect of transcranial direct-current stimulation combined with treadmill training on balance and functional performance in children with cerebral palsy: a double-blind randomized controlled trial. *PloS one*. 2014;9(8):e105777.
18. Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Developmental Medicine & Child Neurology*. 1997;39(4):214-23.
19. Yam WKL, Leung MSM. Interrater reliability of Modified Ashworth Scale and Modified Tardieu Scale in children with spastic cerebral palsy. *Journal of child neurology*. 2006;21(12):1031-5.
20. Homan RW, Herman J, Purdy P. Cerebral location of international 10–20 system electrode placement. *Electroencephalography and clinical neurophysiology*. 1987;66(4):376-82.
21. Grecco LA, Duarte NA, Zanon N, Galli M, Fregni F, Oliveira CS. Effect of a single session of transcranial direct-current stimulation on balance and spatiotemporal gait variables in children with cerebral palsy: A randomized sham-controlled study. *Brazilian journal of physical therapy*. 2014(AHEAD):00-.
22. Kaski D, Dominguez RO, Allum JH, Bronstein AM. Improving Gait and Balance in Patients With Leukoaraiosis Using Transcranial Direct Current Stimulation and Physical Training An Exploratory Study. *Neurorehabilitation and neural repair*. 2013;27(9):864-71.
23. Madhavan S, Weber II KA, Stinear JW. Non-invasive brain stimulation enhances fine motor control of the hemiparetic ankle: implications for rehabilitation. *Experimental brain research*. 2011;209(1):9-17.
24. Stagg CJ, Bachtiar V, O'Shea J, Allman C, Bosnell RA, Kischka U, et al. Cortical activation changes underlying stimulation-induced behavioural gains in chronic stroke. *Brain*. 2011:awr313.
25. Kaski D, Quadir S, Patel M, Yousif N, Bronstein AM. Enhanced locomotor adaptation aftereffect in the "broken escalator" phenomenon using anodal tDCS. *Journal of neurophysiology*. 2012;107(9):2493-505.