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Transcranial direct current stimulation treatment in chronic after-stroke dysphagia: A clinical case

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Abstract

Background: Transcranial direct current stimulation (tDCS) is a non-invasive neuromodulation technique that has shown positive effects in a variety of motor diseases including dysphagia. However, its clinical application and underlying microstructural effects are still being researched. We tested whether tDCS applied together with swallowing training could benefit a treatment-resistent patient with dysphagia after a stroke. **Method:** The clinical case was a 64 year old male who suffered from a left medial cerebellum stroke lesion leading to severe dysphagia. The treatment consisted of a traditional swallowing rehabilitation treatment, which served as a baseline measure, followed by an intervention adding tDCS (anodal/left M1/16 sessions/20 min/1mA). The variables measured were (1) clinical symptoms of dysphagia, (2) quality of life and (3) microstructural changes with magnetic resonance imaging (MRI) and diffusion tensor imaging (DTI). **Results:** Results showed (1) a slight reduction of the clinical symptoms, (2) an improvement in the quality of life domains of communication, fatigue and sleep and (3) an enhancement of the connectivity and increase of the number of fibers of the injured left medium cerebellum peduncle. **Conclusions:** These results support the use of tDCS as a coadjuvant neurorehabilitation tool in cases which are resistant to traditional motor rehabilitation.

Keywords: Transcranial direct current stimulation, dysphagia, stroke, diffusion tensor imaging, neurorehabilitation.

Resumen

Tratamiento con estimulación transcraneal de corriente directa en disfagia crónica post ictus: caso clínico. **Antecedentes:** la estimulación transcraneal de corriente directa (tDCS) es una técnica de neuromodulación no invasiva que ha mostrado efectos positivos sobre varias patologías motoras, incluyendo disfagia. Empero, su aplicación clínica y efectos microestructurales subyacentes están aún bajo investigación. Consecuentemente, en este estudio comprobamos si la tDCS aplicada junto a terapia de deglución podría beneficiar a un paciente con disfagia debida a ictus resistente a tratamiento. **Método:** el paciente fue un hombre de 64 años con disfagia severa, consecuencia de un ictus en el cerebelo medial izquierdo. El tratamiento consistió en una rehabilitación de deglución, empleada como línea base, seguida de una intervención añadiendo tDCS (anodal/M1 izquierdo/16 sesiones/1 mA). Las variables medidas fueron; (1) síntomas clínicos de disfagia, (2) calidad de vida, y (3) cambios microestructurales medidos con resonancia magnética (MRI) y tensor de difusión (DTI). **Resultados:** los resultados mostraron: (1) una leve reducción de síntomas clínicos, (2) una mejora en calidad de vida en las áreas de comunicación, fatiga y sueño, y (3) una mejora en la conectividad y aumento de las fibras del pedúnculo cerebeloso izquierdo. **Conclusiones:** estos resultados apoyan el uso de tDCS como herramienta de neurorehabilitación coadyuvante para casos resistentes a tratamiento de rehabilitación motora tradicional.

Palabras clave: estimulación transcraneal de corriente directa, disfagia, ictus, imagen por tensor de difusión, neurorehabilitación.

According to the World Health Organisation, nearly 15 million people suffer from strokes every year and 60% of those patients survive (World Health Organization, 2008, 2011). Dysphagia is developed by 50-60% of all stroke-patients, and it is characterized by swallowing difficulties that can range from mild to severe, leading in the most extreme cases to the necessity of feeding gastrostomy tubes (FGT) (Martino et al., 2005). For its high disruptiveness in nutrition, respiration and daily life activities, dysphagia has been related to further complications such as aspiration pneumonia, dependency, depression, malnutrition and mortality (Namasivayam & Steele, 2015), producing a negative impact on the quality of life.

Despite its apparent simplicity, swallowing is a complex motor behaviour that requires the coordination of more than 25 pairs of oropharyngeal muscles and multiple brain regions that include the motor cortex, the brainstem area and the cerebellum (Hamdy et al., 1996; Rangarathnam, Kamarunas, & McCullough, 2014).

One of the main challenges after a stroke is therefore the loss of the functional motor abilities, as Kwakkel et al. (Kwakkel, Kollen, van der Grond, & Prevo, 2003) showed that only 11,6% of the stroke patients accomplish a complete motor rehabilitation by 6 months after the stroke. Consequently, there is a need to find new potential therapeutic tools that maximize the effect of post-stroke motor rehabilitation.

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Stroke leads to a dysregulation of cortical excitability (Carter et al., 2010), and given the neural repair mechanisms that are likely to be involved in the recovery process, there has been increasing interest in the role of neuromodulation techniques to treat swallowing problems (Simons & Hamdy, 2017). Transcranial Direct Current Stimulation (tDCS) is a non-invasive, safe (Bikson et al., 2016) brain stimulation technique that delivers mild electrical current, changing the excitability of the underlying neurons (Galea, Jayaram, Ajagbe, & Celnik, 2009).

Many studies have already shown the positive benefits of repetitive tDCS sessions on the enhancement of the human motor function (Sánchez-Kuhn, Pérez-Fernández, Cánovas, Flores, & Sánchez-Santed, 2017).

Besides the damaged cerebellum has been targeted in some tDCS studies before, studies have not shown that a modification of the cerebellar excitability with tDCS can improve post-stroke motor function (Lefebvre & Liew, 2017). Nevertheless, the stimulation of the motor cortex with anodal tDCS (a-tDCS) has shown to improve swallowing (Kumar et al., 2011) and specifically in dysphagic population (Yang et al., 2012). However, the functional and microstructural changes that explain and underlie these long-lasting behavioral improvements are still not defined.

Hence, the aim of the present study was to know the impact of repetitive tDCS over the undamaged left motor cortex in a severe dysphagic chronic cerebellar stroke patient on the swallowing mechanism, tracking the clinical symptoms of dysphagia, the functional brain connectivity and the self-perceived quality of life.

Method

Participants

The participant was a man, 64 years old and diabetic. The patient presented a chronic severe dysphagia resistant to swallowing therapy caused by a cerebellar stroke occurred 24 months ago. The stroke caused a hypotrophy in the post inferior area of the left cerebellum with an extension the medulla oblongata leading to the impossibility to swallow, receiving FGT feeding. In addition, the participant presented hypersensibility on the right body side.

Instruments

a) Transcranial Direct Current Stimulation (tDCS). tDCS was administered with Magstim DC-Stimulator Plus from neuroConn (Ilmenau, Germany). The stimulation protocol (Figure 1) was comprised within the safety limits for tDCS (Bikson et al., 2016) and possible discomforts or side-effects were registered.

b) Magnetic Resonance Imaging (MRI) and Diffusion Tensor Imaging (DTI). The patient underwent a diffusion tensor imaging (DTI) and MRI using a 3T scanner (Achieva XR 3.0 T; Philips Medical System, Eindhoven, The Netherlands). The DTI images were processed in Fiber Tracking software. These neuroimaging sessions were undertaken in the Medical Technology Clinic (Almería, Spain).

c) Swallowing Quality of Life (SWAL-QoL) questionnaire. We applied an adapted version of the Spanish version of the SWAL-QoL questionnaire (Zaldibar-Barinaga & Pinedo-Otaola, 2013) which was designed for the assessment of the self perceived quality of life of patients with dysphagia. The domains of feeding time, food selection and the items 9, 10, 16, 17, 18, 19, 20 and 21 from the domain "frequency of symptoms" were eliminated due to the severity of the present case.

Procedure

As seen in Figure 1, during the first two weeks of baseline, the patient received 8 sessions of swallowing therapy by Therapist 1 and Therapist 2 evaluated the clinical symptoms.

Baseline					**Treatment**				
x 2 weeks					**x 4 weeks**				
Day 1	Day 2	Day 3	Day 4	SWAL-QoL Pre-test	Day 1	Day 2	Day 3	Day 4	SWAL-QoL Post-test
					⚡	⚡	⚡	⚡	
ST	ST	ST	ST	MRI+DTI Pre-test	ST	ST	ST	ST	MRI+DTI Post-test
EV		EV			EV		EV		

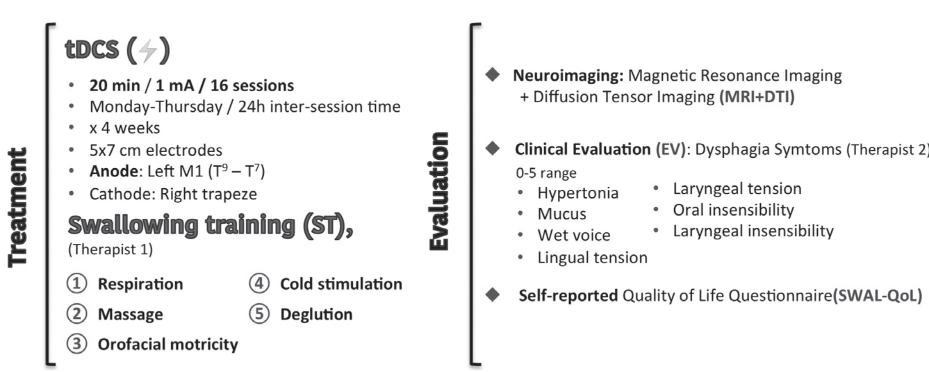

Figure 1. Experimental procedure

SWAL-QoL and MRI+DTI were tested before (Pre-Test) and after the 16 treatment sessions (Post-test). The 16 treatment sessions consisted in tDCS stimulation followed by swallowing therapy.

The experimental procedure was approved by the Committee on Bioethics in Human Research (CIH) of the University of Almeria, Spain. The patient gave its written informed consent to participate in the study, undertaken in accordance with the ethical standards of the World Medical Assembly (WMA) Declaration of Helsinki. The personal information was treated under the Spanish personal data protection law of the 13th December 15/1999.

Data analysis

Data was analysed by assessing the reduction or augmentation reported in (1) the clinical symptoms of dysphagia, (2) the self reported quality of life and (3) the microstructural brain connectivity. The potential clinical changes were assessed comparing the 2 × weeks baseline period vs. the 4 × weeks treatment period; and the quality of life and microstructural changes were assessed by comparing the Pre-test and the Post-test results.

Results

Magnetic Resonance Imaging (MRI) and Diffusion Tensor Imaging (DTI)

MRI with DTI results (see Figure 2.a) displayed significant reduction of fibbers shown by an asymmetry of the left cerebellar peduncle (CP) with respect to the right (green), with clear reduction of the quantitative values of anisotropy factor (AF) and an increased apparent diffusion coefficient (ADC) (yellow) (CP right, AF = 0.67 and CDA = 0.76; CP left, AF = 0.53 and ADC = 1.15).

After the treatment, MRI with DTI results (see Figure 2.b) displayed an increased connectivity and number of fibbers in the left cerebellar peduncle with respect to the right (green), with similar quantitative values of the AF. The increased ADC matched persisted (yellow) (CP right, AF = 0.64 and ADC = 0.66, CP left, AF = 0.59 and ADC = 1.78).

Clinical symptoms

As seen in Table 1, all the evaluated clinical symptoms showed a slight general reduction after the treatment from the Baseline measures (BL) to the Stimulation measures (STIM). The highest reduction was seen in mucus (-1.00 points), followed by lingual tension (-0.50 points), hypertonia, laryngeal tension, oral insensibility, laryngeal insensibility (-0.38 points), and wet voice (-0.25 points).

By looking at the evaluations within each week, an observable stability was seen among the scores during the Baseline period, while scores become unstable during the stimulation, showing the greatest general reduction of the symptoms in the first and last weeks of the stimulation.

SWAL-QoL questionnaire

As shown in Table 2, there was an enhancement in the self-perceived quality of life of the domains of fatigue (+20.00 points), sleep (+20.00 points), communication (+10.00 points) and symptom frequency (+3.33 points), while we found a decrease in swallowing as a burden (-10.00 points), social (-20.00 points), eating fear (-20.00 points), desire to feed (-26.67 points) and mental health (-39.33 points).

Side effects

The patient reported mild itching sensations during the stimulation sessions, especially at the reference electrode, presumably due to the hypersensitivity on the right side of the body. No further discomfort was reported.

Discussion

To the best of our knowledge, the present study is the first attempt to assess the potential connectivity changes produced by the treatment with tDCS and swallowing training on a post-stroke dysphagia patient. Those changes were observed in the damaged area, and consisted in an increased connectivity and number of

2. a) MRI with DTI - Pre-treatment

2. b) MRI with DTI - Post-treatment

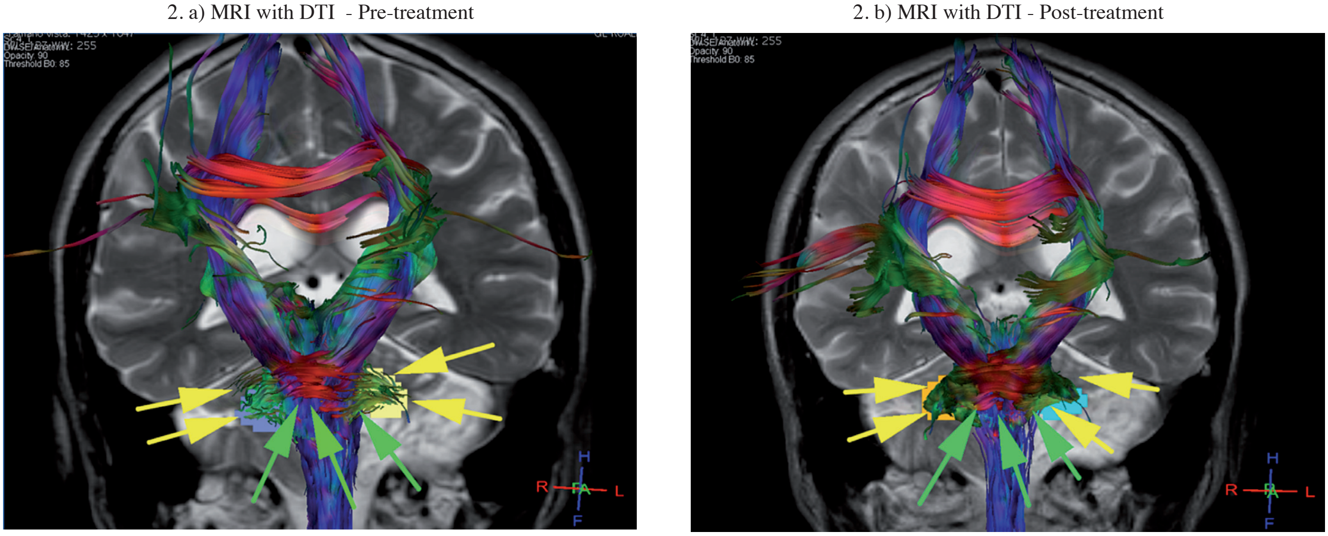

Figure 2. Shows Pre and Post -treatment MRI and DTI results

Table 1
Shows the results of the evaluations of the clinical symptoms of dysphagia: Mean (Standard Deviation); score range 0-5 (were 5 is the highest presence of the symptom) in the first (BL1) and second (BL2) weeks of Baseline, during the four weeks of stimulation (S1, S2, S3 and S4) as well as for the complete Baseline (BL) and Stimulation (STIM) periods for each clinical symptom including HYP: Hypertonia; MUC: Mucus; WV: Wet voice; LIN_TEN: Lingual tension; LAR_TEN: Laryngeal Tension; OR_INS: Oral insensibility and LAR_INS: Laryngeal insensibility

Clinical symptom	BL1	BL2	S1	S2	S3	S4	BL	STIM
HYP	2.00(0.00)	2.00(0.00)	1.33(0.71)	2.00(0.00)	1.00(0.00)	2.00(0.00)	2.00(0.00)	1.62(0.52)
MUC	3.43(0.71)	3.43(0.71)	2.00(0.00)	3.00(0.00)	2.00(0.00)	3.00(0.00)	3.50(0.58)	2.50(0.53)
WV	2.40(0.71)	2.40(0.71)	2.00(0.00)	3.00(0.00)	2.00(0.00)	2.00(0.00)	2.50(0.58)	2.25(0.46)
LIN_TEN	2.40(0.71)	2.40(0.71)	1.00(0.00)	3.00(0.00)	2.00(0.00)	2.00(0.00)	2.50(0.58)	2.00(0.76)
LAR_TEN	3.00(0.00)	3.00(0.00)	2.00(0.00)	3.00(0.00)	3.00(0.00)	2.40(0.71)	3.00(0.00)	2.62(0.52)
OR_INS	3.00(0.00)	3.00(0.00)	2.00(0.00)	3.00(0.00)	3.00(0.00)	2.40(0.71)	3.00(0.00)	2.62(0.52)
LAR_INS	3.00(0.00)	3.00(0.00)	2.00(0.00)	3.00(0.00)	3.00(0.00)	2.40(0.71)	3.00(0.00)	2.62(0.52)

Table 2
Shows the scores obtained in each of the nine measured domains of the SWAL-QoL obtained in the questionnaire in the Pre-test and in the Post-test

SWAL-QoL questionnaire score		
Domain	**Pre-test**	**Post-test**
Fatigue	20.00	40.00
Sleep	20.00	40.00
Communication	30.00	40.00
Symptom frequency	26.67	30.00
Swallowing as a burden	40.00	30.00
Social	40.00	20.00
Eating fear	40.00	20.00
Desire to feed	46.67	20.00
Mental health	100	64.00

fibbers in the left cerebellar peduncle. A modest reduction of the clinical symptoms was registered and the self-perceived quality of life resulted enhanced after the treatment in those domains related to the pathology, while deteriorated in those related to mental health.

The augmentation of the myelinisation in the damaged cerebellar area by the stimulation of the motor cortex area confirms the ipsilateral connections between the left sensorimotor cortex and the cerebellum (Lemon, 2008). These specific area and task-related changes might be due to the effect of combining the stimulation with the rehabilitation training (Page et al., 2015).

There was only a slight decrease of the symptoms, which can be explained do the severity of the case, as previous literature reported positive effects of tDCS over the clinical symptoms of dysphagia in less severe cases (Pisegna, Kaneoka, Pearson, Kumar, & Langmore, 2015).

Regarding the quality of life, the scores of the domains related to the pathology itself resulted enhanced, although the self-perceived quality of life in those related to mental health (swallowing as a burden, social, eating fear, desire to feed and mental health) resulted deteriorated. For the interpretation of these outcomes, it is important to consider the slight enhancement in the clinical symptoms, which was not high enough to result perceptible. In order to work specifically over the mental health of dysphagia patients, other kind of tDCS interventions shifting the target brain area could be beneficial, such as the stimulation of the left dorsolateral prefrontal cortex (DLPFC) (Tae-Gyu, Soo-Han, & Ko-Un, 2017).

In conclusion, the present study highlights the potential modulatory capacity of tDCS, producing microstructural changes in the damaged area by the administration of a repetitive stimulation intervention.

Conflict of interests

The authors declare that they have no competing interests.

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