Case Report

Transcutaneous electrical nerve stimulation (tens) in brain stem paroxysms: case report

Estimulação elétrica neural transcutânea (tens) na paroxismia do tronco cerebral: relato de caso

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ABSTRACT | INTRODUCTION: Brainstem paroxysmia has been described in patients with multiple sclerosis, characterized by rapid episodes of nystagmus and vertigo evoked by cephalic movements. Neuromodulation has shown therapeutic success for several dysfunctions of multiple sclerosis, but it has not yet been used in paroxysmia. **OBJECTIVE:** To assess the additive effect of neuromodulation by transcutaneous electrical neural stimulation (TENS) on the periocular musculature in a patient with multiple sclerosis and brain stem paroxysmia that is not responsive to classical rehabilitation associated with medication. **MATERIAL AND METHODS:** Nystagmus evaluation with video nystagmography, facial skin sensitivity analysis with esthesiometer, video recording of blepharoclonus, and pain sensation subjectively quantification (by a personal description of the patient); execution of 10 eyes consecutive exercises sessions associated with TENS in the right lower oblique and lateral rectus muscles. **RESULT:** Improvement in pain, blepharoclonus, and right eye nystagmus. **CONCLUSION:** Neuromodulation with TENS seems to be a valid complementary therapy for patients with brainstem paroxysmia unresponsive to other clinical treatments, but more studies are needed to confirm this finding.

KEYWORDS: Multiple sclerosis. Nystagmus. Neuromodulation.

RESUMO | INTRODUÇÃO: A paroxismia de tronco encefálico, descrita em pacientes com esclerose múltipla (EM), caracteriza-se por episódios rápidos de vertigem e nistagmos evocados por movimentos cefálicos. A neuromodulação tem apresentado sucesso terapêutico em várias disfunções da EM, mas ainda não foi utilizada na paroxismia. **OBJETIVO:** Avaliar o efeito aditivo da neuromodulação por estimulação elétrica neural transcutânea (TENS) na musculatura periocular em paciente com EM e paroxismia de tronco encefálico não responsiva à reabilitação clássica associada à medicação. **MATERIAL E MÉTODOS:** Avaliação do nistagmo com videonistagmógrafo, da sensibilidade cutânea facial com estesiômetro, do blefaroclônus por gravação em vídeo e da sensação dolorosa de forma subjetiva (descrição pessoal da paciente); execução de 10 sessões consecutivas semanais de exercícios oculares associados à TENS na musculatura do oblíquo inferior e reto lateral direitos. **RESULTADO:** Melhora da algia, do blefaroclônus, da vertigem e do nistagmo ocular direito. **CONCLUSÃO:** A neuromodulação com TENS parece ser uma terapia complementar válida para pacientes com EM e paroxismia de tronco não responsivos aos demais tratamentos clínicos, porém mais estudos são necessários para confirmar esse achado.

PALAVRAS-CHAVE: Esclerose múltipla. Nistagmo. Neuromodulação.

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Introduction

Multiple sclerosis (MS) is a chronic inflammatory and demyelinating disease of autoimmune origin. Multifocal and temporary lesions characterize it in the central nervous system (CNS) with axonal damage. It mainly affects young female adults and causes great psycho-social impact.[1] Demyelination frequently occurs in the brain and cerebellum; therefore, abnormal vestibular sensations, such as vertigo and imbalance, are common in its clinical course.[2] About 78% of patients have balance abnormalities, 18-63% have nystagmus, and true vertigo is found in more than 17%.[3] Seizures, hearing loss, and paralysis are rare.[4] Brainstem paroxysm was described in 1925 by Carmichael and Critchley as "facial nystagmus" triggered in the lateralization of the gaze in patients with ME.[5]

The management of MS symptoms includes non-pharmacological methods such as rehabilitation and psychological support, medications, and surgical procedures.[4]

Vestibular rehabilitation is a supportive therapy for patients with balance disorders. It has an excellent response in peripheral vestibular disorders. In patients with central disorders, such as those with MS, the results seem less expressive.[6]

TENS has been used for pain relief; it sends additional somatosensory afferents, decreasing pain perception. Its use in pain has been evaluated both in animal and human model experiments. In high frequency and low intensity, it generates a non-painful tingling sensation, and in low frequency and high intensity, it activates the small-diameter nociceptive afferents fibers, A and C, taunting a painful but tolerable sensation. These afferents are capable of stimulating structures at the level of the CNS, neuromodulating them, and generating analgesia.[7,8]

Neuromodulation has expanded into several neurological subspecialties. Movement disorders and painful syndromes after strokes are among the most common neurological indications. In MS, the intrathecal baclofen pump has been the main neuromodulation technique used by many patients to improve the management of spasticity.[9]

There is no description of neuromodulation use for symptoms of brainstem paroxysms in patients with MS, which is why we developed an interest in reporting this case.

Objective

Report a case of vestibular rehabilitation associated with neuromodulation with TENS in a patient with MS and brainstem paroxysms.

Material and methods

Protocol number 32886620.4.0000.0066 approved on the Brazil Platform.

The patient signed a consent form and an authorization term for the use of images.

Nystagmus was analyzed and quantified by Videonystagmography (VisualEyes525, InteracousticsR) (VNG) before neuromodulation and after it ended a month later.

Facial sensitivity was analyzed by esthesiometry before starting neuromodulation and after it ended, one month later.

TENS (TENS 3000, portable, USA) was configured with a frequency of 70Hz, pulse duration of 120μs, N-Pulse mode, which means constant stimulation. The mode shows the programming to which the device displays the current. The pulse rate is automatically varied in a cyclic pattern for nominally 10 seconds (150Hz maximum). The pulse rate decreases linearly for 4 seconds after setting the setpoint to a value that is 40% lower. The lowest pulse rate will continue for 1 second. It will increase linearly for 4 seconds, returning to its original value. The original pulse rate will continue for 1 second. The cycle is then repeated. The waveform is asymmetric, biphasic with a square pulse. Continuous stimulation in the muscles of the lateral rectum and lower oblique of the right eye for 40-50 minutes. The electrodes were placed on the face, extraocular musculature, close to the belly of the target musculature (figure 1).

Figure 1. Electrodes position

The device intensity was regulated according to the patient's perception and generally corresponded to a sensation of vibration in the extraocular muscles accompanied by a slight contraction. The first part of the therapy was initiated only with the TENS stimulus without exercise association. In the last 10 to 15 minutes, classic vestibule-ocular reflex exercises were associated, according to vestibular rehabilitation protocols (Cawthorne and Cooksey, and Herdman). The same exercises were prescribed at home, 2 to 3 times a day, in addition to exercises for training the vestibule-ocular reflex on the computer, both without association with TENS. It started with horizontal exercises, progressing to vertical exercises.

Case report

SFO, a female, 25 years old, seven years ago (2012/05), was diagnosed with MS outbreak-remission form. She presented recurrent episodes of paralysis of right hemidimidium, right hemiface, left hemidimidium, aphasia, diplopia, vertigo, nausea, and vomiting; weight loss (11 kg in 5 months), and sudden deafness in the right ear. Skull resonance with detection of demyelinating lesions affecting the periventricular region, cerebellum, middle peduncles, pons, medulla, corpus callosum, spinal cord segments from C2 to C6 (figures 2 and 3) and CSF analysis with a result compatible with MS. Brainstem audiometry on 13/02 with thresholds of 60 dB in the left ear and 105 dB in the right ear, with an increase in the absolute latencies of waves III and V and interpeak latencies I-V and III-V bilaterally. In 17/01, after a bilateral outbreak, she lost strength in all four limbs and decreased hearing in the right side; she started to experience sudden, rapid vertigo and vertigo and tinnitus induced by the rotation of the head to the right or by the lateralization of the gaze to the right, pain, and blepharoclonus in the right eye. Audiometry on 18/01 showed left hearing normal and moderate to severe sensorineural hearing loss in the right ear. Brainstem audiometry on 18/04 detected a threshold of 105 dB in the right ear and 25 dB in the left ear. Resonance of the internal auditory canal was normal (18/03). She was diagnosed with brainstem paroxysms and was treated with carbamazepine at 100 mg twice a day, but the patient only tolerated 100 mg at night, starting on 18/05. There was a considerable improvement in the symptoms of paroxysms with medication, but the persistence of blepharoclonus and right eye pain. Upon examination, she presented spontaneous torsional nystagmus, counterclockwise, to the right, with open eyes, which increased in intensity upon the lateralization of the gaze to the right. There was a decrease in the tactile sensitivity of the right hemiface and a slight decrease in the strength of the left lower limb without functional impairment.

VNG (July 2018) with central findings (spontaneous torsional nystagmus counterclockwise, with horizontal component to the right 5º/s and vertical to down also 5º/s); calibration, pendular nystagmus, saccades and optokinetic with alterations; spontaneous nystagmus intensifications in bilateral Rose and bilateral torsion positions; directional preponderance of nystagmus for the right side on air caloric test. In July 2018, she started vestibular rehabilitation with exercises to optimize the horizontal and vertical vestibule-ocular reflexes. In the same month, we proposed her an experimental therapy with TENS neuromodulation, aimed at relieving blepharoclonus and periocular pain and reinforcing nystagmus control. In September 2018, she had severe pain in the right malar region, radiating to the right ear. When the pain subsided, she noticed a return of hearing in that ear. New audiometry was performed, which confirmed the normal level of hearing in the right ear. She did ten consecutive sessions of classic vestibular rehabilitation associated with TENS with weekly intervals to optimize the horizontal vestibule-ocular reflex. After those treatments, she showed improvement of nystagmus (the horizontal component reduced to 1º/s), the continued vertical 5º/s, normalization of the tactile sensitivity of the face, and cessation of blepharoclonus and periocular pain. Reduction in 80% of the horizontal nystagmus component. She was instructed to perform home exercises to improve the vertical vestibule-ocular reflex, keeping the horizontal exercises. One month after the last session, she continued well, with the vertical component even smaller (2º/s) and the horizontal component with the same speed (1º/s).

Figures 2 and 3. Demyelinating lesions in periventricular region, cerebellum, middle peduncle, pons, and spinal cord

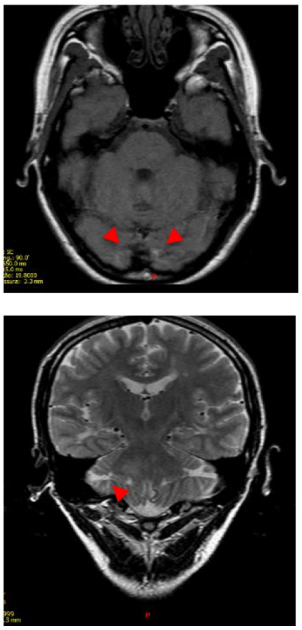

Table 1. Patient clinical evaluation

Sign/Symptom	Blepharoclonus	Angular Speed of Nystagmus	Face Esthesiometry	Periocular Pain	Hearing Loss
Pre-treatment	Moderate	5º/s horizontal 5º/s vertical	Right Hemiface: Tactile perception: Red filament (4 g) Left Hemiface: Tactile perception: Blue filament (0,02 g)	Intense	Moderate to severe hearing loss in the right ear. Normal hearing in left
After treatment	Ceased	1º/s horizontal 2º/s vertical	Fluctuated during treatment, had moments with hyperesthesia on the right, but at the end, maintaining a similar pattern at the beginning.	Ceased	Normal hearing in both ears

Discussion

Nystagmus is seen in many CNS diseases, particularly those affecting the brainstem and cerebellum.

Keane, in 1978, described two patients with demyelinating disease who exhibited painful, repeated facial contractures precipitated by the eccentric look, mainly involving the orbicularis musculature of the eyes. These eyelid problems fluctuated in intensity but persisted for years and did not respond to phenytoin or a short course of treatment with carbamazepine.[5]

Quiles et al. in 1980 reported three cases of MS that presented paroxysmal attacks during their course, also called brainstem seizures.[10]

It is common for medicated nystagmus`s treatment that potassium blockers channels drugs in downbeat nystagmus, baclofen in alternating nystagmus, gabapentin, and memantine in acquired pendular nystagmus, and carbamazepine in upper oblique myokymia.[11] Treatments with these drugs have shown a good response concerning nystagmus control, with few side effects, although their recommendation use only has an evidence C level.[12]

In addition to nystagmus pharmacological treatment, other therapeutic approaches have been used by different medical fields. In ophthalmology, orthoptic procedures and surgeries are options. Ciuffreda KJ et al. performed auditory biofeedback training to control nystagmus in five patients with congenital nystagmus and obtained an average reduction of 82%, 86%, and 34% in amplitude, the peak of low-speed component and frequency of nystagmus, respectively.[13]

Brainstem paroxysm, an event secondary to MS demyelinating injury or stroke, enters the differential diagnosis of vestibular paroxysms. Vestibular paroxysm is a disorder that is characterized by rapid attacks of vertigo, nystagmus, and sometimes tinnitus, triggered by cephalic rotation, which are repeated several times throughout the day; their pathophysiological mechanism is effective neural discharges originated from vascular compression at the proximal portion of the eighth cranial nerve; it has an excellent response to treatment with carbamazepine / oxcarbazepine.[14]

In MS, carbamazepine is used to treat certain types of seizures and neuralgia, such as acute facial pain, in shock, caused by trigeminal neuralgia.[15]

Another treatment modality in MS is the use of neuromodulation.[9] Neuronal activity regulates oligodendrocytes, and axonal myelination can quickly increase the axonal diameter, promote the reentry of oligodendrocyte progenitor cells into the cell cycle, or direct them to differentiate into oligodendrocytes.[16]

Several authors describe cases of sudden deafness in patients with MS with good evolution concerning hearing recovery.[17]

Our patient presented an unusual form of MS manifestations, with sudden deafness and progression to episodes of the paroxysmal brainstem. She was subjected to unusual treatment, which developed our interest in reporting her case. She underwent an experimental treatment with neuromodulation associated with vestibular rehabilitation.

Considering the effectiveness of carbamazepine in improving some neurological symptoms of patients with ME[15] and its excellent response to vestibular paroxysms (a dysfunction also resulting from an effective discharge[14]), we opted for nystagmus treatment with this drug. However, the patient only tolerated a low dose and did not show complete improvement of nystagmus, blepharoclonus, and right periocular pain.

As it is a disease with CNS involvement, where the response to classical rehabilitation is not as effective as in peripheral dysfunctions[6] and due to periocular pain and blepharoclonus, we chose to associate neuromodulation with classical treatment.

Therefore, the experimental therapy with TENS was designed to fulfill two purposes: the first, pain relief, and the second, to reduce nystagmus and try to help compensate.

The theory of optimization of neuronal recovery with the acceleration of myelination induced by stimulation of neuronal pathways provides a physiological basis for the proposal of neuromodulation in patients with MS.[16]

The patient presented an excellent clinical response to the treatment. However, we cannot say what or which of the treatments (medication, classical vestibular rehabilitation, neuromodulation) are or were responsible for the therapeutic success or if the improvement resulted from spontaneous remyelination.

Her good audiological recovery is in line with other cases already described.[17]

The rapid improvement (after ten weeks) with the resolution of periocular pain, blepharoclonus, and decreased nystagmus, symptoms that have already been present for a year and a half, lead us to believe that neuromodulation may have played a role in this process.

Conclusion

Report of an unusual case of MS, with a large number of symptoms (brainstem paroxysms, sudden deafness, hypoesthesia of the right hemiface) and its good response to treatment with carbamazepine associated with classical vestibular rehabilitation and peripheral neuromodulation with TENS. Right periocular pain and right blepharoclonus disappeared; normalization of right hearing; and reduction in the angular velocity (horizontal component in 80% and vertical in 60%) of torsional nystagmus present in the right eye. The case evolution is suggestive that neuromodulation had an effect that was at least additive to other therapeutic modalities. Similar articles are needed to validate these findings.

Author contributions

Lins EMDS made the diagnosis and clinical treatment, conceived the alternative neuromodulation for the patient, participated in the bibliographic review and making of the text. Boffino CC conceived the alternative neuromodulation for the patient, performed the classic vestibular rehabilitation program associated with neuromodulation, participated in the bibliographic review and making of the text. Mantovani DCP and Pereira FPS assisted in capturing the complementary exams and in the bibliographic review. Silveira EAAS assisted in the clinical diagnosis and the literature review.

Competing interests

No financial, legal, or political conflicts involving third parties (government, companies and private foundations, etc.) have been declared for any aspect of the submitted work (including, but not limited to, grants and funding, participation in advisory council, study design, preparation manuscript, statistical analysis, etc.).

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