

Persistent atypical odontalgia treated with transcranial magnetic stimulation. Case report*

Odontalgia atípica persistente tratada com estimulação magnética transcraniana. Relato de caso

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SUMMARY

BACKGROUND AND OBJECTIVES: Atypical odontalgia (AO) is a complex orofacial pain syndrome with difficult treatment. Transcranial magnetic stimulation (TMS) has shown good results for chronic pain patients. The objective of this study is to present a case of a patient with AO treated with TMS.

CASE REPORT: Female patient, 39 year-old patient, with a five year history of clinically refractory dental pain (#46) (VAS = 8). During that time, she had received dental and medical treatment without any improvement of her pain complaint. According the International Classification of Headache Disorders criteria she received the diagnosis of AO and was treated with several medications, with no improvement after six months; thus, she was referred to transcranial magnetic stimulation of the motor cortex. After TMS, she had complete improvement of pain which lasted two

months; after that, the pain restarted with low intensity (VAS = 3); complete follow-up was of 1 year.

CONCLUSION: Transcranial magnetic stimulation is new possibility for refractory cases of chronic facial pain.

Keywords: Atypical odontalgia, Neuropathic pain, Orofacial Pain, Transcutaneous magnetic stimulation.

RESUMO

JUSTIFICATIVA E OBJETIVOS: Odontalgia atípica (OA) é uma síndrome dolorosa orofacial de difícil tratamento. A estimulação magnética transcraniana (EMT) tem dado bons resultados em pacientes com dor crônica. O objetivo deste estudo foi apresentar um caso de paciente com OA tratada com EMT.

RELATO DO CASO: Paciente do sexo feminino, 39 anos com histórico de cinco anos de odontalgia clinicamente refratária (#46) (EAV = 8). Durante esse tempo ela recebeu tratamento dentário e médico sem nenhuma melhora de sua dor. De acordo com os critérios da Classificação Internacional de Distúrbios da Cefaleia, ela foi diagnosticada como sofrendo de OA e foi tratada com vários medicamentos, sem nenhuma melhora após seis meses; então, foi enviada para estimulação magnética transcraniana do córtex motor. Após a EMT ela apresentou melhora completa da dor que durou dois meses; após esse tempo a dor recomeçou, porém com baixa intensidade (EAV = 3); o acompanhamento completo foi de um ano.

CONCLUSÃO: A estimulação magnética transcraniana é uma nova possibilidade para casos refratários de dor crônica.

Descritores: Dor neuropática, Dor orofacial, Estimulação magnética transcraniana, Odontalgia atípica,

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INTRODUCTION

The variability of possible pain causes at the face and the complexity of the trigeminal system contribute to the inherent difficulties during the diagnosis and assessment of orofacial pain. Among them, "Atypical Odontalgia" (AO) is highlighted because of the controversies during diagnostic and treatment, and the potential neuropathic cause, at least in part of it¹. It is considered severe toothache without pathological signs², and currently is classified as idiopathic facial pain of central etiology³. An aggravator for this patients is the innumerous iatrogenic procedures that are performed because of toothache-like pain⁴, just as the same as what occurs with patients that have idiopathic trigeminal neuralgia⁵.

Among treatment options for AO, transcranial magnetic stimulation (TMS) had analgesic effect in patients with neuropathic pain, and the greater improvement occurred when pain was at the face, even if the stimulated cortical region was representative of the hand, probably because of the plasticity due to the activation of a close area to the pain at the somesthetic cortex⁶. Primary trigeminal neuropathies or secondary neuropathies after the injury of the central nervous system also improved with cortical stimulation after estereotathic introduction of the electrode⁷.

Thus, the purpose of this study was to report a case of a patient with refractory AO that was successfully treated with TMS of the motor cortex.

CASE REPORT

Female patient, 39 year-old, housewife, complained of very intense dental pain at the right first inferior molar (#46); intensity was 8 (Visual Analogic Scale - VAS), daily frequent, continuous with crisis lasting hours to days. She also complained of sleep and emotional disturbances. The onset of pain was about five years ago. During this time she received more than 10 different types of dental and medical treatments: occlusal adjustment, twice root canal therapy of tooth 46, root canal therapy of tooth 47, apicectomy of #46; extraction of #47, dental splints, non steroid anti-inflammatory drugs (NSAIDS), antidepressants and analgesics.

Patient assessment - the patient underwent a standardized protocol that consisted of an interview and systematic evaluation of cervical, cranial, facial, dental and other oral structures⁸. Aiming to exclude symptomatic diseases⁹, a hemogram and cranial computed tomography (CT) were solicited, both being normal. Physical exam

disclosed severe dental pain at #46 without occlusal interference or any dental or periodontal disease.

The diagnosis of Persistent Idiopathic Facial Pain (Atypical Odontalgia - AO) was made according to The International Classification of Headache Disorders criteria and can be observed in chart 1³.

Chart 1 - International Headache Society (IHS) diagnostic criteria for "persistent idiopathic facial pain"³

13.18.4 Persistent idiopathic facial pain	
Previously used term: atypical facial pain	
Comments: The term atypical odontalgia has been applied to a continuous pain in the teeth or in a tooth socket after extraction in the absence of any identifiable dental cause.	
Diagnostic criteria:	
A.	Pain in the face, present daily and persisting for all or most of the day, fulfilling criteria B and C
B.	Pain is confined at onset to a limited area on one side of the face ¹ , and is deep and poorly localized
C.	Pain is not associated with sensory loss or other physical signs
D.	Investigations including X-ray of face and jaws do not demonstrate any relevant abnormality.

Treatment – after diagnosis, she was treated with tricyclic antidepressants (amitriptyline, nortriptyline), gabapentine (900 mg), with no improvement within 6 months. She was then referred to TMS, and one session was performed. The parameters of stimulation can be observed in table 1.

Table 1 – Parameters of the transcranial magnetic stimulation (TMS).

Frequency	10 Hz
Intensity of stimulation	120% of the motor threshold
Number of series	25
Duration of series	5 seconds
Intervals	25 seconds
Bobbin	Bobbin in eight
Bobbin angle	Perpendicular to the cortex to be stimulated
Local of stimulation	Left prefrontal dorsolateral cortex

After 3 days since the stimulation was performed, we had partial improvement of pain; after 6 days, she was completely pain free, and it lasted 2 months, when pain returned with a low intensity (VAS = 3). Complete follow-up was of 1 year.

DISCUSSION

To our knowledge, this is the first report of treatment of AO

with TMS. AO is, at least in part, neuropathic of central origin¹, with complex treatment and need of correct diagnosis, including investigation of comorbidities¹⁰. As in other idiopathic neurological pains of the trigeminal system, it is common to find iatrogenic procedures^{4,5} in the history of these patients, and it happened in this case. It is difficult to determine if dental pain occurred prior to AO, or was just a symptom already of the neuropathic condition.

Since the introduction of TMS, the use of this technique in clinical neurophysiology, neurology, neurosciences and psychiatry has rapidly being diffused in countries as USA, Canada, Japan and Europe, which scientific and clinical objectives^{11,12}. TMS is one of the safer noninvasive and innocuous methods developed to study and treat the central and peripheral nervous systems¹³. Its precise mechanism still remains uncertain; TMS produces an electrical current parallel to the cortex, activating preferentially horizontally oriented neurons, at the surface. Inhibitory and facilitator neurons can be preferentially stimulated with this technique, according to the frequency of stimulation. Frequencies higher than 1 Hz produce stimulation and lower than 1 Hz produce inhibition. The collateral effects that can occur are convulsive crises, but they are extremely rare¹⁴. The intervals among the series that are smaller than 1 second seem to have also an important role in the triggering of convulsive crises, but intervals higher than 20 seconds seem to be very safe¹⁴. Although the great importance of TMS in the treatment of neuropathic pain, and the evidences that at the face the effects are higher^{6,7}, there is still a lack of studies investigating complex neuropathic pain disorders at the trigeminal system, which often fail in their current treatments. This is the case of the patient in this study, with had failure of current treatment, and the TMS was used as an option, as a non invasive technique, with great results. The possible reasons for the effectiveness of TMS in neuropathic pain probably involve the activation of interneuron's between the motor and somatosensory cortexes, and the reasons for the higher efficacy for facial pain are possibly because the areas of cortex representation of the face are very large, like the representing areas of the hands, in comparison to other body parts^{6,7}.

In conclusion, we could observe great improvement in the reported case of atypical odontalgia, which persisted during one year of follow-up. Although the result is remarkable, considering it is a refractory pain to other treatments, other studies are necessary to confirm its efficacy. TMS can be a good alternative for unpleasant neuropathic pain.

REFERENCES

1. Benoliel R, Heir GM, Eliav E. Neuropathic facial pain. In: Sharav Y, Benoliel R, (editors). Orofacial pain & headache. Mosby: Elsevier; 2008. p. 255-94.
2. Merskey H, Bogduk N. Classification of chronic pain. Seattle (WA): IASP Press; 1994. p. 222.
3. The International Classification of Headache Disorders: 2nd ed. Cephalalgia 2004;24 (Suppl 1):9-160.
4. Merrill RL. Intraoral neuropathy. Curr Pain Headache Rep 2004;8(5):341-6.
5. de Siqueira SR, Nóbrega JC, Valle LB, et al. Idiopathic trigeminal neuralgia: clinical aspects and dental procedures. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2004;98(3):311-5.
6. Lefaucheur JP, Drouot X, Menard-Lefaucheur I, et al. Neurogenic pain relief by repetitive transcranial magnetic cortical stimulation depends on the origin and the site of pain. J Neurol Neurosurg Psychiatry 2004;75(4):612-6.
7. Nguyen JP, Keravel Y, Feve A, et al. Treatment of deafferentation pain by chronic stimulation of the motor cortex: report of a series of 20 cases. Acta Neurochir Suppl 1997;68:54-60.
8. Siqueira JT, Lin HC, Nasri C, et al. Clinical study of patients with persistent orofacial pain. Arq Neuropsiquiatr 2004;62(4):988-96.
9. Zakrzewska JM. Facial pain: neurological and non-neurological. J Neurol Neurosurg Psychiatry 2002;72(Suppl 2):ii27-ii32.
10. Dworkin RH, O'Connor AB, Backonja M, et al. Pharmacologic management of neuropathic pain: evidence-based recommendations. Pain 2007;132(3):237-51.
11. George MS, Belmaker RH. Transcranial magnetic stimulation in neuropsychiatry. Washington (DC): American Psychiatric Press; 2000. p.298.
12. Walsh V, Pascual-Leone A. Neurochronometrics of Mind: TMS in Cognitive Science. Cambridge (MA): MIT Press; 2003. p 379.
13. Chokroverty S, Hening W, Wright D, et al. Magnetic brain stimulation: safety studies. Electroencephalogr Clin Neurophysiol 1995;97(1):36-42.
14. Wassermann EM. Risk and safety of repetitive transcranial magnetic stimulation: report and suggested guidelines from the International Workshop on the Safety of Repetitive Transcranial Magnetic Stimulation. June 5-7. 1996. Electroencephalogr Clin Neurophysiol 1998;108(1):1-16.

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