

Primary auriculotemporal neuralgia. Case report

Neuralgia do auriculotemporal primária. Relato de caso

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ABSTRACT

BACKGROUND AND OBJECTIVES: Auriculotemporal neuralgia is an uncommon condition. Symptoms are brief severe pain attacks, especially in the temporal region. Although many cranial neuralgias are related to nervous compression, they may be present without identifiable etiologic factors. This study aimed at describing a case of primary auriculotemporal neuralgia and respective therapeutic approach.

CASE REPORT: Male patient, 72 years old, presented for assistance reporting severe pain in left temporal region, described as shock and of very short duration. After clinical evaluation and imaging exams, no significant changes were detected. Patient has satisfactorily responded to carbamazepine.

CONCLUSION: This case shows that auriculotemporal neuralgia has clinical presentation similar to other neuralgias. Diagnosis is primarily obtained by pain characteristics evaluation and exclusion of possible secondary causes.

Keywords: Auriculotemporal nerve, Auriculotemporal neuralgia, Neuropathic pain.

RESUMO

JUSTIFICATIVA E OBJETIVOS: A neuralgia do auriculotemporal é uma condição rara. Os sintomas consistem em ataques de dor intensa, de curta duração, sobretudo na região temporal. Embora muitos casos de neuralgias cranianas sejam relacionados à compressão nervosa, podem ocorrer sem fatores etiológicos identificáveis. O objetivo deste estudo foi descrever um caso de neuralgia auriculotemporal primária e respectiva conduta terapêutica.

RELATO DO CASO: Paciente do gênero masculino, 72 anos, compareceu para atendimento relatando dor intensa em região temporal esquerda, descrita como choque, de curtíssima duração. Após avaliação clínica e exames de imagens, não foram detectadas alterações significativas. O paciente respondeu de forma satisfatória à carbamazepina.

CONCLUSÃO: O caso descrito mostra que a auriculotemporal possui apresentação clínica semelhante àquela de outras neuralgias. O diagnóstico é obtido principalmente pela análise das características da dor e exclusão de possíveis causas secundárias.

Descritores: Dor neuropática, Nervo auriculotemporal, Neuralgia do auriculotemporal.

INTRODUCTION

Neuropathic pain results from a primary nervous system injury or disorder, triggered by local trauma or systemic diseases and affecting peripheral or central nervous structures. Considering the symptom, orofacial neuropathic pain may be classified in two categories: episodic or continuous. Episodic neuropathic pain is characterized by pain paroxysms described as electric shock or twinge, similar to what happens with trigeminal neuralgia (TN)¹. Pain paroxysms are in general followed by remission periods, known as refractory periods. Pain may be classified in primary (classic) or secondary (symptomatic) and the difference between them is important for adequate diagnosis and management. While the vast majority of patients have brief, severe and paroxysmal pain, continuous pain may also be present^{2,3}. Most common neuralgias are trigeminal, post-herpetic, glossopharyngeal and occipital. Less frequent are supraorbital and intermediate nerve neuralgia. These disorders may underlie infectious and/or inflammatory neurologic diseases, in addition to other primary headaches³.

The auriculotemporal nerve (n.AT) is a sensory branch originating from mandibular nerve posterior trunk. Due to its pathway, there is major risk for compression and irritation. Its nervous roots form a short trunk, which supplies several branches and innervates temporomandibular joint (TMJ), temporal region, pinna and external acoustic meatus. It conducts sympathetic fibers to the scalp and parasympathetic fibers to the parotid gland⁴. Anatomic relationship between n.AT and masticatory muscles, TMJ and surrounding vessels in the infra temporal fossa region, creates favorable conditions for compressive syndromes^{5,6}.

Auriculotemporal neuralgia (NAT) is an uncommon condition. In a tertiary center, reported frequency was just 0.4%⁷. It seems to be more prevalent in middle-aged females. Symptoms are excruciating pain attacks, especially in the temporal region. Pain in TMJ, parotid and ear, with irradiation to temporal region is also described and may be relieved by auriculotemporal nerve anesthetic block^{8,9}. Although not having its own classification, the name epicranial neuralgias has been suggested, including neuralgias of other peripheral branch-

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es^{10,11}, such as supraorbital, supratrochlear, nasal, greater and lesser occipital nerves¹².

Although many cranial neuralgias are primary, other etiologic factors may be involved. Refractory cases or with paresthesia may indicate the presence of expansive injuries. Although mechanisms of such conditions are not totally explained, non-odontogenic toothache⁷, refractory facial pain⁸, as well as NAT secondary to synovial cyst in the condyle¹³ are reported by the literature.

This study aimed at reporting a case of NAT where underlying secondary factors were not found, and which has favorably responded to low dose of carbamazepine.

CASE REPORT

Male patient, 72 years old, who came for assistance complaining of severe left temporal region pain, described as shock. First crisis had been 5 years ago with spontaneous remission. Two months ago pain reappeared, with very short duration, with several episodes varying from 1 to 2 minutes. These episodes were repeated three to four times a day and did not wake up patient at night. At physical evaluation, no pain trigger-zone was found. Patient had controlled diabetes and referred having been submitted to prostatectomy due to cancer. Brain magnetic resonance was normal. Panoramic X-rays of jaws and computerized tomography of temporomandibular joints had not shown significant changes (Figures 1, 2 and 3). Diagnostic hypothesis was NAT. Carbamazepine (200mg) was prescribed during the first two days, continuing with 400mg for the following 15 days. In the first week using the drug, patient was reevaluated and reported lower frequency of shocks (2 to 3 per day), which he defined as “pinching”. In the following week, patient referred symptoms remission. Maintenance dose of 200mg was then kept and 4 months later there has been total symptoms control.



Figure 1. Panoramic X-rays approximate left side view
There were no significant changes.

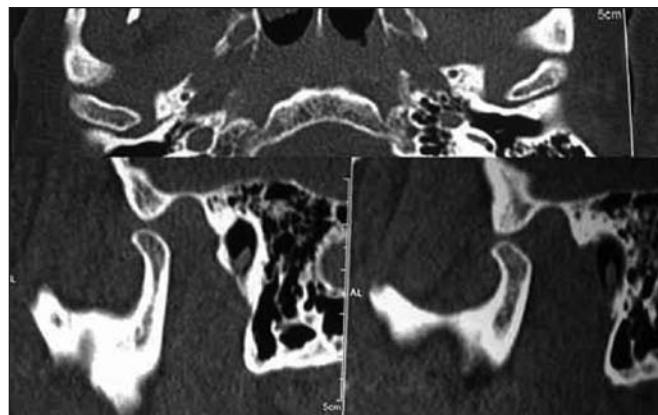


Figure 2. Computerized tomography of temporomandibular joint (open mouth)

There were no temporomandibular joints expansive injuries.



Figure 3. Ultrasound of left temporal region

DISCUSSION

Due to its rarity, the International Classification of Headache Disorders (ICHD-II) does not have a specific category for NAT, being it classified as “*neuralgia of other terminal branches*”. Diagnostic criteria are: 1) Pain in the distribution of a trigeminal nerve peripheral branch, except nasociliary and supraorbital nerves; 2) Sensitivity on affected nerve; 3) Pain is eliminated with anesthetic block or nerve ablation¹¹. In the new ICHD-III classification, however, this category was not included¹⁰. Differential NAT diagnosis should be obtained to rule out other disorders, including temporomandibular disorders, toothache, continuous migraine, earache, trigeminal neuralgia, temporal arteritis, myofascial pain and atypical facial pain²⁻⁷.

Primary diagnosis of our patient was obtained by means of pain characteristics, such as location, quality, intensity, frequency and duration. Imaging exams (panoramic X-rays, TMJ tomography and temporal region ultrasound) were asked to exclude possible secondary causes of the disease. Masticatory muscles palpation has not shown relevant changes. For being a well located pain in temporal region without irradiation patterns, in shock, we have considered initial diagnosis of NAT.

In the period between 2004 and 2005, a total of 1500 patients were evaluated in a pain control tertiary center. Six cases of NAT were documented. All patients were females aged

between 23 and 65 years. The interval between beginning of symptoms and diagnosis has varied from 1 month to 20 years. Pain was moderate to severe, located around the ear, and irradiating to condyle and temporal region in all patients. Pain was continuous in 5 cases and associated with stabbing-type exacerbation in three. One patient had pre-auricular trigger-point.

One patient has reported this symptom triggered by gustatory stimulation causing diet limitations. Pain was unilateral, not involving the opposite side. For all patients, pain was triggered or worsened by pressure in the pre-auricular region or slightly above it². In our case, patient has referred beginning of symptoms two months ago, had severe, spontaneous and brief pain episodes which were repeated at few minutes intervals.

n.AT trunk has close anatomic relationship with condyle and TMJ capsule region, which may contribute to sensory disorders in this site. In several studied cases, nerve was in direct contact with the medial aspect of the capsule or of the neck of the condyle. Studies have not observed anatomic correspondences between right and left sides, so just one side can be affected. n.AT may also be in direct contact with lateral pterygoid muscle¹⁴. Findings support the hypothesis that the anatomic relationship between n.AT and condyle, articular fossa and lateral pterygoid muscle might be associated to its compression and irritation, thus contributing to neuropathic pain in its innervated regions¹⁵.

To investigate n.AT compression mechanism, Komarkitki et al.⁵ have studied the infratemporal fossa in 16 specimens and have found that the number of n.AT roots varied between 1 and 5. They have observed that, in variations where n.AT had 4 to 5 roots, there were complex connections. In five cases, lowest n.AT roots started in the inferior alveolar nerve instead of in the mandibular nerve. This study findings point to two major factors which may be involved with auriculotemporal nerve compression in its pathway inside the infratemporal fossa. The first is anatomic variation while the second is the presence of masticatory muscles disorder, especially lateral pterygoid muscle. According to the authors, even minor structural or functional changes may lead to painful syndromes. In fact, our patient complained of shock and unilateral pain in temporal region. However, there were no trigger-points related to jaw functional demands in this case. Pharmacological treatment is still the first line to control several neuralgias. It may be obtained with the association of classic analgesics and anticonvulsant or antidepressant drugs. In patients with classic trigeminal neuralgia, carbamazepine is considered the first choice drug. However, several adverse effects are associated to its use, among them skin erythema, negative effect on bone density, liver changes and pharmacological interaction. Oxcarbazepine may be used in patients nor responding to carbamazepine. Second line drugs are baclofen, lamotrigine, pregabalin and gabapentin¹⁶.

Anesthetic blocks, surgery and other procedures may be necessary for refractory cases³. Literature states that n.AT anesthetic block is diagnostic and therapeutic. It is performed

with 0.5mL 2% lidocaine without vasoconstrictor and dexamethasone (2.5mg/0,5 mL), being described successful reports in symptoms control^{2,7,8}. Anesthetic block induces immediate pain relief in patients with different types of headaches. In addition, its analgesic effect is longer as compared to anesthesia, so that patients have pain relief for weeks or even months. This prolonged anesthetic effect after peripheral block may be related to effect on central pain modulation. Anesthetic blocks are indicated for some primary headaches, secondary headaches and cranial neuralgias. One must take care with certain populations of patients, including pregnant women, elderly, those allergic to substances used, with history of previous vasovagal episodes and in use of anticoagulants. Described peripheral anesthetic blocks involve injections in greater and lesser occipital nerves, supratrochlear, supraorbital and auriculotemporal nerves¹⁷. Reported complications include pain, bleeding, infection an intravascular injection³. To investigate analgesic efficacy and tolerability to pharmacological treatment, 200 patients with classic trigeminal neuralgia (TN) were selected. Carbamazepine (CBZ) was prescribed for 100 individuals and oxcarbazepine (OXC) was prescribed for the other 100. Response to CBZ was found in 98%, with mean dose of 600mg (200-1200), and response to OXC has been 94%, with mean dose of 1200mg (600-1800). In a period of approximately 8.6 months, 27% of patients responding to CBZ had major adverse effects, being necessary to discontinue the drug or to decrease it to unsatisfactory levels. After 13 months, the same has happened with 18% of respondents to OXC. Among patients with good initial response, only 3 individuals using CBZ and 2 using OXC have developed late resistance. During the course of disease, paroxysms have worsened in 3% of patients and their duration has increased 2%. In a 7-year period, no clinical sensory deficit manifestations were observed for all patients. Based on available evidences, CBZ or OXC are considered first therapeutic option^{18,19}.

In a retrospective study with 72 classic TN patients, CBZ was highly effective in 60.8% of cases in the long term with maintenance dose. Other drugs were used in more refractory cases, including gabapentin, which has relieved symptoms with mean duration of 18 months. Peripheral neurectomy was needed for 23% of patients²⁰.

Considering our case report, where there were no secondary underlying etiologic factors, we decided for CBZ as first therapeutic option. In addition, although literature states that anesthetic block may be considered initial clinical decision, factors such as pain characteristics, patient's age and the possibility of response to conservative therapy were taken into consideration

CONCLUSION

This case shows that NAT has clinical presentation similar to that of other neuralgias. The diagnosis of this uncommon condition is primarily obtained by evaluating pain characteristics and by excluding possible secondary causes.

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