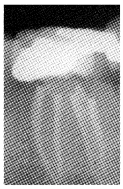
International Endodontic Journal

doi:10.1111/j.1365-2591.2009.01599.x



CASE REPORT

Toothache referred from auriculotemporal neuralgia: case report

R. A. Murayama[1], J. Stuginski-Barbosa[2], N. P. Moraes[1] & J. G. Speciali[2]

[1]Orofacial Pain Service, Oral Oncology Center, Dental School of Araçatuba, São Paulo State University (UNESP); and [2]Department of Neurology, Psychiatry and Medical Psychology, Faculty of Medicine of Ribeirão Preto (FMRP), University of São Paulo, São Paulo, Brazil

Abstract

Murayama RA, Stuginski-Barbosa J, Moraes NP, Speciali JG. Toothache referred from auriculotemporal neuralgia: case report. *International Endodontic Journal*, **42**, 845–851, 2009.

Aim To present a 52-year-old male patient who complained of intense pain of short duration in the region of the left external ear and in the ipsilateral maxillary second molar that was relieved by blockade of the auriculotemporal nerve in the infratemporal fossa.

Summary Extra- and intraoral physical examination revealed a trigger point that reproduced the symptoms upon finger pressure in the ipsilateral auriculotemporal nerve and in the outer auricular pavilion. The patient's medical history was unremarkable. The maxillary left second molar tooth was not responsive to pulp sensitivity testing and there was no pain upon percussion or palpation of the buccal sulcus. Periapical radiographs revealed a satisfactory root filling in the maxillary left second molar. On the basis of the clinical signs and symptoms, the auriculotemporal was blocked with 0.5 mL 2% lidocaine and 0.5 mL of a suspension containing dexamethasone acetate (8 mg mL^{-1}) and dexamethasone disodium sulfate (2 mg mL^{-1}), with full remission of pain 6 months later. The diagnosis was auriculotemporal neuralgia.

Key learning point

- Auriculotemporal neuralgia should be considered as a possible cause of nonodontogenic toothache and thus included in the differential diagnoses.
- The blockade of the auriculotemporal nerve in the infratemporal fossa is diagnostic and therapeutic. It can be achieved with a solution of lidocaine and dexamethasone.

Keywords: anaesthesia, auriculotemporal neuralgia, local, toothache.

Received 9 December 2008; accepted 20 April 2009

Introduction

Most dental and orofacial pain occurs as result of pulpal or periodontal diseases. Nonodontogenic pain may be referred from myofascial trigger points (Farella *et al.* 2000),

Correspondence: J. Stuginski-Barbosa, Rua Antônio Modenezi, 1239, Franca-SP 14406-310, Brazil (e-mail: juliana.dentista@gmail.com).

sinusopathies (Chen *et al.* 1997), angina pectoris (Durso *et al.* 2003), primary headaches (Gross 2006, Gaul *et al.* 2007), tumours (Uehara *et al.* 2007), and neuropathic pain such as trigeminal neuralgia (Spencer *et al.* 2008), deafferentation pain (Ram *et al.* 2009), and amputation neuromas (Vickers & Cousins 2000).

The diagnosis of orofacial pain is a challenge and becomes even more complicated when the signs and symptoms are due to rare conditions such auriculotemporal neuralgia (AN).

The auriculotemporal nerve is a branch of the mandibular nerve, the third division of the trigeminal nerve, which passes through the foramen ovale and enters the infratemporal fossa, where it divides into anterior and posterior branches. Some branches arise from the posterior division, one of them being the auriculotemporal branch, which crosses the neck of the mandibular head and splits into two other branches before entering the parotid gland (Ayesh *et al.* 2007).

AN is characterized by a strictly unilateral lancinating pain that may be perceived in the temporal region, temporomandibular joint (TMJ), the parotid, auricular and retro-orbital region (Damarjian 1970, Shankar & Brethauer 2007).

The most common disease due to involvement of the auriculotemporal nerve is Frey syndrome. This is characterized by hyperaesthesia, redness, increased temperature or sweating along the distribution of the auriculotemporal nerve and/or major auricular nerve, associated with the intake of foods that stimulate salivation (Benedittis 1990). Another rare disease affecting this nerve is AN (Speciali & Gonçalves 2005).

At the International Classification of Headaches (ICHD-II) (Headache Classification Commitee of the Internacional Headache Society 2004), AN is not cited as a sub item, but it fulfils the criteria for item 13.7 'Other terminal branch neuralgias' (Table 1). According to this classification, damage to and incarceration of the peripheral branch of the trigeminal nerve may trigger referred pain in the area innervated by the affected branch.

The objective of the present study was to report a case of nonodontogenic toothache in a maxillary molar which was relieved by blockade of the auriculotemporal nerve in the infratemporal fossa.

Case report

A 52-year-old male Caucasian patient complained of intense pain of 30 days duration in the region of the left outer auricular pavilion and in the ipsilateral maxillary second molar. The patient reported that his pain was paroxysmal, of high intensity (eight on the VAS scale)

Table 1 Classification of other neuralgias of the peripheral branches of the trigeminal nerve according to the International Headache Classification

13.7 Other terminal branch neuralgias
Description:
Injury or entrapment of peripheral branches of the trigeminal nerve other than the nasociliary and supraorbital nerves may give rise to pain referred to the area innervated by the branch affected. Examples are neuralgias of the infraorbital, lingual, alveolar and mental nerves.
Diagnostic criteria:
A. Pain in the distribution of a peripheral branch of the trigeminal nerve other than the nasociliary or supraorbital nerves
B. Tenderness over the affected nerve
C. Pain is abolished by local anaesthetic blockade or ablation of the nerve

Source: The International Classification of Headache Disorders (ICHD-II) (Headache Classification Subcommittee of the International Headache Society 2004).

and of short duration and that at times it could be triggered by touching his face and the area of the ear helix.

There were no other associated symptoms and the patient's medical history was unremarkable.

The patient believed that the pain originated in the maxillary left second molar. He reported that the composite resin restoration in this tooth had been removed previously without improvement of the pain by another dentist.

Extra- and intraoral physical examination revealed a trigger point in a helix region of the external ear that caused a shock-type pain referred to the maxillary left second molar and a burning pain in the ipsilateral temporal region. In addition, palpation of the region of the auriculotemporal nerve also elicited a burning pain irradiating towards the temporal region

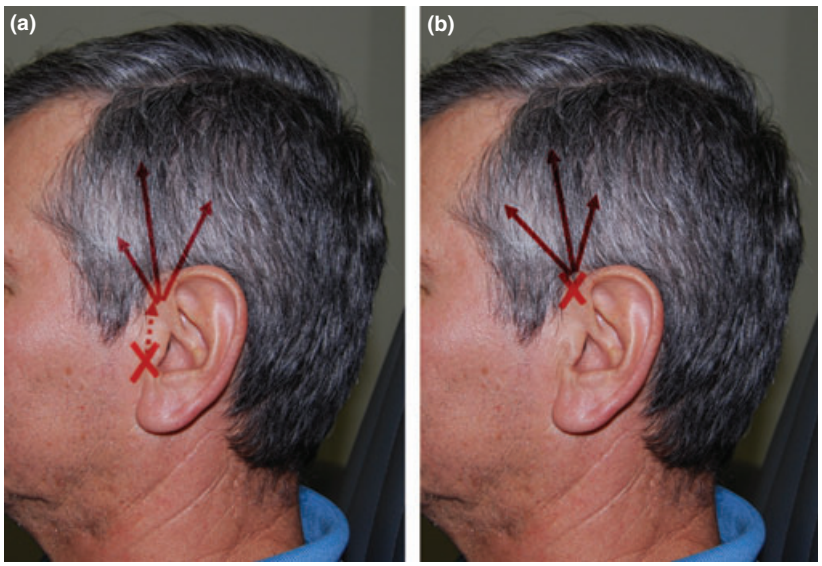


Figure 1 (a) Trigger point in a helix region of the outer ear that causes a shock-type pain referred to the left upper second molar and a burning pain in the ipsilateral temporal region. (b) Palpation of the region of the auriculotemporal nerve also elicited burning pain irradiating towards the temporal region.

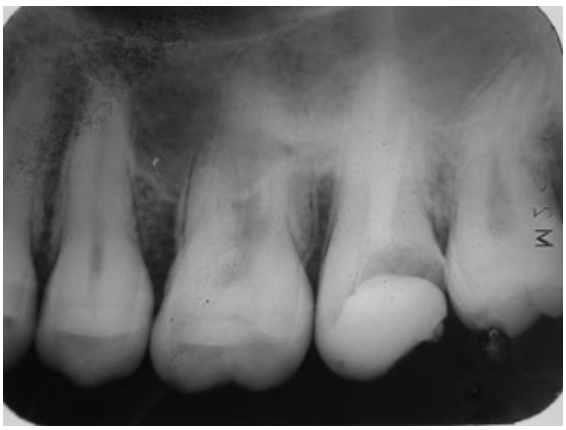


Figure 2 Periapical radiograph of maxillary left molar teeth.

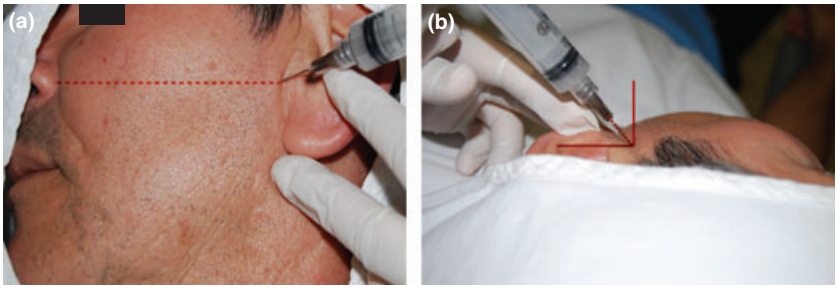


Figure 3 Auriculotemporal blockade. (a) The needle is inserted below the TMJ, in the posterior margin of the head of the mandible immediately in front of the tragus. (b) Depth of 1–1.2 cm, at a horizontal 45° angle in the direction of the nose.

(Fig. 1). No pain occurred upon palpation of the TMJ and masticatory muscles. The mandibular movements were normal.

The maxillary left second molar tooth was not responsive to pulp sensitivity testing and there was no pain upon percussion or palpation of the sulcus area in the region. A periapical radiograph revealed a satisfactory root filling in the maxillary left second molar (Fig. 2).

The auriculotemporal nerve was blocked with 0.5 mL 2% lidocaine and 0.5 mL of a suspension containing dexamethasone disodium sulphate (2 mg mL^{-1}) and dexamethasone acetate (8 mg mL^{-1}), according to the technique of Speciali & Gonçalves (2005), as follows: the needle was inserted below the TMJ, in the posterior margin of the head of the mandible immediately in front of the tragus, to a depth of 1–1.2 cm, at a horizontal 45º angle in the direction of the nose, with care taken to first perform aspiration in order to avoid intravascular injection (Fig. 3). There was full remission of pain that has lasted for 6 months.

Discussion

The clinical manifestations of cranial neuralgias cited in the International Classification of Headache Disorders (2004) are diversified, including paroxysmal pain in classical trigeminal neuralgia and paroxysmal or constant pain in supraorbital neuralgia. The sensitive extracranial terminal branches of the trigeminal nerve can cause different clinical phenotypes depending on the type, physiopathology and localization of the involvement of these nerve endings. The specific clinical signs and symptoms of each nerve ending have been described in the literature (Pareja *et al.* 2003, Headache Classification Subcommittee of the International Headache Society 2004, Speciali & Gonçalves 2005).

The major causes of neuralgia of the extracranial branches of the trigeminal nerve are believed to be mechanical irritation due to compression (incarceration), traction or friction. This occurs more frequently when the nerve goes through anatomical structures that might compromise it, such as muscles (Johansson *et al.* 1990).

One of the probable sites of incarceration of the auriculotemporal nerve is the infratemporal fossa (Loughner *et al.* 1990), where the nerve is at risk of compression by myospasm of the superior lateral pterygoid muscle, generating irritation (Schmidt *et al.* 1998). Another situation that may cause AN is the presence of medial dislocation of the disk of the TMJ, with consequent compression of the auriculotemporal nerve against the mandibular fossa during the mandibular movement to the contralateral side (Johansson *et al.* 1990).

In some cases, the auriculotemporal nerve emerges together with the temporal artery (Torres *et al.* 2004), a fact leading to the assumption that the pulsation of the temporal artery could be the cause of AN.

In AN the pain may affect the region of the external auricular pavilion, the auricular and preauricular region, the parotid gland, the capsule of the TMJ, the temporal region, and the maxillary region. Speciali & Gonçalves (2005) reported six cases of AN, in two of which the pain was referred to the maxillary region, as in the case reported here.

The main clinical characteristics of AN are pain of moderate to severe intensity, often continuous and associated with exacerbations perceived as stabbing pains. Involvement is strictly unilateral, with no changes in side and the pain may worsen or be triggered by pressure on the periauricular region at the level of the tragus (Speciali & Gonçalves 2005). The anaesthetic blockade of the affected branch eliminates the pain (IHS-II 2004).

According to Pareja *et al.* (2003), AN may be a form of epicrania, a term proposed to define the group of headaches apparently originating from epicranial tissues; These superficial pains tend to be of short duration, but can be continuous and chronic, with clinical fluctuations over time. Local signs and symptoms of sensory dysfunction, as well as sensitivity to palpation of this area and also at the sites of emergence or along the path of the nerves may be present. Autonomic symptoms similar to those of trigeminal-autonomic headaches are not commonly associated.

In the case reported here the pain was triggered by pressure on palpation of the region of auriculotemporal innervation and by palpation on the helix of the external auricular pavilion; the pain was referred to the temporal region and to the maxillary left second molar. In the differential diagnosis, other diseases that might provoke similar symptoms were excluded, such as cervicogenic headache, temporomandibular dysfunction, migraine, continuous hemicrania, otitis, trigeminal neuralgia, atypical facial pain, 'red ear syndrome', infection of the parotid gland, temporal arteritis, and odontogenic pain.

The pain provoked by touch or pressure in these regions may be due to the generation of ectopic impulses at the site of probable incarceration (Campero *et al.* 1998, Tal *et al.* 1999).

A possible complication of the anaesthesia blockage is the occurrence of transitory hemifacial paralysis due to anaesthesia of the facial nerve in cases in which the needle is inserted more superficially than desired, or to diffusion of the anaesthetic solution through the parotid gland reaching the nerve.

In the present case there was full improvement and resolution of painful symptoms after a single blockade, with no recurrence after a 6-month follow-up.

When describing AN, Damarjian (1970) reported full remission of symptoms with anaesthetic blockade of the auriculotemporal nerve, although symptoms recurred after 1–2 years in 30% of cases. New blockades were performed in these cases, 3–4 per patient on average, with one patient receiving 14 blockades. The administration of oral carbamazepine after successful blockade of the auriculotemporal nerve seems to reduce the recurrence of pain. Pareja *et al.* (2003) reported that blockade with, or injection of, corticosteroids was indicated for the control of epicrania symptoms, usually providing complete relief of transitory or long-lasting duration.

Speciali & Gonçalves (2005) reported that the exclusive use of commonly administered oral medications for neuropathic pain do not produce significant relief of patient symptoms.

An alternative procedure for cases of unsuccessful blockade would be ultrasound-guided injection (Shankar & Brethauer 2007). This technique may be a good alternative in view of a greater precision in identification of the auriculotemporal nerve, whose pathway may have anatomical variations.

It is interesting to note that pain relief exceeds the effect of local anaesthetic solution in blockades performed not only in AN, but also in other epicranial neuralgias. The use of

lidocaine in these cases appears to promote reversible inhibition of sodium channels, inhibiting nerve conduction in sensory fibre and thus providing longer analgesia (Ashkenazi & Levin 2007).

It has been suggested that chronic changes in nociceptive processing and neuroplasticity may occur in blockades of the occipital nerves, which also has a prolonged effect (Afridi *et al.* 2006). However, the mechanisms involved in the prolonged effect of peripheral blockade have not been fully elucidated.

Although the effects of locally applied corticosteroids have not been determined, it has been suggested that this treatment may suppress the afferent ectopic firings at the site of nerve injury by an action of C fibres (nociceptive), reducing the release of neuropeptides (Johansson & Bennett 1997). The use of prolonged-action corticosteroids in combination with local anaesthetics may cause a sufficient prolongation of the time of analgesia to inhibit already established central sensitizations, with consequent permanent or greatly prolonged analgesia, as was the case for the present patient.

Conclusion

The present clinical case report suggests that AN may be the cause of pain referred to the teeth characterizing nonodontogenic toothache and therefore should be considered in the differential diagnosis.

Disclaimer

Whilst this article has been subjected to Editorial review, the opinions expressed, unless specifically indicated, are those of the author. The views expressed do not necessarily represent best practice, or the views of the IEJ Editorial Board, or of its affiliated Specialist Societies.

References

Afridi SK, Shields KG, Bhola R, Goasby PJ (2006) Greater occipital nerve injection in primary headache syndromes – prolonged effects from a single injection. *Pain* **122**, 126–9.

Ashkenazi A, Levin M (2007) Greater occipital nerve block for migraine and other headaches: is it useful? *Current Pain Headache Report* **11**, 231–5.

Ayesh EE, Ernberg M, Svensson P (2007) Effects of local anesthetics on somatosensory function in the temporomandibular joint area. *Experimental Brain Research* **180**, 715–25.

Benedittis G (1990) Auriculotemporal syndrome (Frey's Syndrome) presenting as tic doloureux. *Journal of Neurosurgery* **72**, 955–8.

Campero M, Serra J, Marchettini P, Ochoa JL (1998) Ectopic impulse generation and autoexcitation in single myelinated afferent fibers in patients with peripheral neuropathy and positive sensory symptoms. *Muscle and Nerve* **21**, 1661–7.

Chen YH, Tseng CC, Chao WY, Harn WM, Chung SF (1997) Toothache with a multifactorial etiology: a case report. *Endodontics & Dental Traumatology* **13**, 245–7.

Damarjian E (1970) Auriculo-temporal neuralgia – an original diagnostic and therapeutic approach. *The Rhode Island Medical Journal* **53**, 100–1.

Durso BC, Israel MS, Janini ME, Cardoso AS (2003) Orofacial pain of cardiac origin: a case report. *Cranio* **21**, 152–3.

Farella M, Michelotti A, Gargano A, Cimino R, Ramaglia L (2000) Myofascial pain syndrome misdiagnosed as odontogenic pain: a case report. *Cranio* **20**, 307–11.

Gaul C, Sándor PS, Galli U, Palla S, Ettlin DA (2007) Orofacial migraine. *Cephalalgia* **27**, 950–2.

Gross SG (2006) Dental presentations of cluster headache. *Current Pain and Headache Reports* **10**, 126–9.

Headache Classification Commitee of the Internacional Headache Society (2004) Classification and diagnostic criteria for headache disorders, cranio neuralgia and facial pain. *Cephalalgia* **24**, 1–160.

Johansson A, Bennett GJ (1997) Effect of local methylprednisolone on pain in a nerve injury model: a pilot study. *Regional Anesthesia* **22**, 59–65.

Johansson A-S, Isberg A, Isacsson G (1990) A radiographic and histologic study of the topographic relations in the temporomandibular joint region: implications for a nerve entrapment mechanism. *Journal of Oral Maxillofacial Surgery* **48**, 953–61.

Loughner BA, Larkin LH, Mahan PE (1990) Nerve entrapment in the lateral pterygoid muscle. *Oral Surgery Oral Medicine Oral Pathology* **69**, 299–306.

Pareja JA, Pareja J, Yangüela J (2003) Nummular headache, trochleitis, supraorbital neuralgia, and other epicranial headaches and neuralgias: the epicranias. *Journal of Headache Pain* **4**, 125–31.

Ram S, Teruel A, Kumar SK, Clark G (2009) Clinical characteristics and diagnosis of atypical odontalgia: implications for dentists. *Journal of the American Dental Association* **140**, 223–8.

Schmidt BL, Pogrel MA, Necoecha M, Kearns G (1998) The distribution of the auriculotemporal nerve around the temporomandibular joint. *Oral Surgery Oral Medicine Oral Pathology* **86**, 165–8.

Shankar H, Brethauer J (2007) Ultrasound guided steroid injection for auriculo-temporal neuralgia. *Anesthesiology* **107**, A903.

Speciali JG, Gonçalves DA (2005) Auriculotemporal neuralgia. *Current Pain Headache Report* **9**, 277–80.

Spencer CJ, Neubert JK, Gremillion H, Zakrzewska JM, Ohrbach R (2008) Toothache or Trigeminal Neuralgia: Treatment Dilemmas. *The Journal of Pain* **9**, 767–70.

Tal M, Wall PD, Devor M (1999) Myelinated afferent fiber types that become spontaneously active and mechanosensitive following nerve transection in the rat. *Brain Research* **824**, 218–23.

Torres TS, Lucena Neto B, Silva GO *et al.* (2004) Anatomia quirúrgica del nervio auriculotemporal em el acesso pre-auricular. *International Journal of Morphology* **22**, 327–30.

Uehara M, Tobita T, Inokuchi T (2007) A case report: toothache caused by epidermoid cyst manifested in cerebellopontine angle. *Journal of Oral and Maxillofacial Surgery* **65**, 560–1.

Vickers ER, Cousins MJ (2000) Neuropathic orofacial pain part 1 – prevalence and pathophysiology. *Australian Endodontic Journal* **26**, 19–26.