# Glossopharyngeal neuralgia of tumor origin diagnosed in dental care. Case report

Neuralgia do glossofaríngeo de origem tumoral diagnosticada em atendimento odontológico. Relato de caso

Gisele Marchetti<sup>1</sup>, Daniel Bonotto<sup>1</sup>, Paulo Afonso Cunali<sup>1</sup>

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# **ABSTRACT**

**BACKGROUND AND OBJECTIVES**: The glossopharyngeal neuralgia is a neuropathy considered rare that manifests itself in the IX cranial nerve distribution characterized by an electric shock-like pain, often associated with hyperalgesia and allodynia. The etiology may be related to vascular changes, brain tumor, or even idiopathic. The aim of this study was to report a case of glossopharyngeal neuralgia secondary to a brain tumor diagnosed in a dental clinic, highlighting its clinical manifestations and discussing its nosological limit with other orofacial pain.

CASE REPORT: Female patient, 63 years old, sought care at a dental outpatient clinic of Orofacial Pain complaining about an intense electric shock and jumping pain of sudden onset on the lower right edge region and right tongue base. She reported that the events were triggered and exacerbated when chewing, opening the mouth, laughing and talking. The diagnostic hypothesis of glossopharyngeal neuralgia was tested by momentary depletion to the application of benzocaine 20% and pain remission with the administration of carbamazepine (400mg/day) for 20 days. The patient was referred to the Neurology service of the hospital, where the magnetic resonance imaging presented an expansive, solid, extra-axial lesion in the right prepontine cistern, suggesting meningioma.

**CONCLUSION:** The professional should be aware of the differential diagnosis of orofacial pains, especially in episodic neuropathies, to rule out the tumor etiology. In these cases, the quick referral to tertiary centers is fundamental for the good prognosis. **Keywords:** Neoplasia, Neuralgia, Orofacial pain.

1. Universidade Federal do Paraná, Curitiba, PR, Brasil.

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## Correspondence to:

Rua Engenheiro Ostoja Roguski, 631 – Jardim Botânico 80210-390 Curitiba, PR, Brasil. E-mail: marchettiodontologia@gmail.com

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# **RESUMO**

JUSTIFICATIVA E OBJETIVOS: A neuralgia do glossofaríngeo é uma neuropatia considerada rara que se manifesta na distribuição do IX par craniano com característica de dor em choque elétrico, muitas vezes associada à hiperalgesia e alodínea. A etiologia pode estar relacionada a alterações vasculares, a tumor intracraniano, ou ainda idiopática. O objetivo deste estudo foi relatar um caso de neuralgia do glossofaríngeo secundário a tumor intracraniano, diagnosticada em ambulatório odontológico, destacando suas manifestações clínicas e discutindo seu limite nosológico com outras dores orofaciais.

RELATO DO CASO: Paciente do sexo feminino, 63 anos procurou atendimento em ambulatório odontológico de Dor Orofacial com queixa de dor intensa em choque elétrico e pontadas de início súbito na região de rebordo inferior direito e base de língua direita. Relatou que os eventos eram deflagrados e exacerbados na mastigação, ao abrir a boca, rir e falar. A hipótese diagnóstica de neuralgia do glossofaríngeo foi testada pela depleção momentânea à aplicação de benzocaína a 20% e pela remissão da dor com administração de carbamazepina (400mg/dia) por 20 dias. A paciente foi referida para serviço hospitalar de neurologia, onde após ressonância magnética nuclear foi diagnosticada lesão expansiva sólida extra-axial na cisterna pré-pontina à direita, sugestiva de meningioma.

**CONCLUSÃO**: O profissional deve estar atento no diagnóstico diferencial das dores orofaciais, especialmente nas neuropatias episódicas, pois a etiologia tumoral deve ser descartada. Nesses casos, a rápida referência a centros terciários é fundamental para o bom prognóstico.

**Descritores**: Dor orofacial, Neoplasia, Neuralgia.

# INTRODUCTION

Orofacial pain is a factor of concern for the patient because many times it affects important physiological functions, such as chewing, swallowing, speak and laugh, also compromising the well-being and the quality of life (QoL) of the individual. The concept of QoL is marked by subjectivity, involving all the essential components of the human condition, whether physical, psychological, social or cultural. Pain, both facial and dental, is the most cited aspect of oral health indicators that impact the QoL, followed by sleep loss and masticatory problems<sup>1</sup>. However, in addition to these, a number of painful conditions affect the face, making it essential to establishing differential diagnosis<sup>2</sup>.

An example of such conditions is neuropathic pain that results in a primary injury or diseases of the somatosensory nervous system. It can be triggered by local trauma or systemic diseases, affecting peripheral or central nerve structures and can be classified into episodic or continuous<sup>3</sup>.

Neuropathies vary according to with the involved nerve<sup>4</sup>. The glossopharyngeal nerve (IX) provides a general somatic sensation to touch, pain, and temperature of the posterior third of the tongue, pharynx, middle ear and the area near the external acoustic meatus<sup>3</sup>. The glossopharyngeal neuralgia (GPN) has a paroxysmal character in the innervation area. The prevalence is very low when compared with other neuropathies, estimated between 0.2 and 0.7 cases for every 100,000 people per year<sup>5</sup>. The accepted description for GPN is a painful disorder characterized by unilateral pain usually brief, electric shock type, with abrupt start and end, affecting the ear, the base of the tongue, the tonsillar fossa or beneath the angle of the jaw<sup>6</sup>.

GPN is divided into two clinical types based on pain distribution: eardrum (affecting the ear) and oropharyngeal (affecting the oropharynx area)<sup>7</sup>. It is often difficult for patients with GPN to identify what are the areas of origin of pain since these structures are in deep regions of the mouth, pharynx and the ear<sup>8</sup>.

GPN etiology seems to be related to the demyelination of axons or degeneration of the IX and X cranial nerves, and it can be associated with the compression of IX nerve by vascular structures, intracranial injuries or tumors. The compression causes constant mechanical irritation of the nerve and can significantly decrease the excitability threshold and promote an increase in the action potential<sup>9</sup>.

Thus, the differential diagnosis of orofacial pain is extremely important considering its huge negative impact on QoL of the patient, symptoms and possible tumor causes.

The objective of this study was to discuss the semiological techniques and the approach of GPN with tumor etiology for a patient who came to a dental clinic in search for treatment.

## **CASE REPORT**

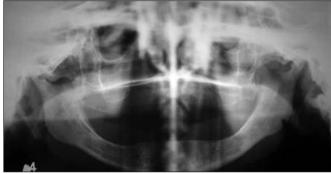
Female patient, 63 years old, with a prosthesis (dentures) in the upper and lower dental arches for eight years, sought care at the Orofacial Pain dental outpatient clinic. The major complaint was severe pain which started eight years ago, in the region of the lower edge and the right side of the base of the tongue, referring 10 on the visual analog scale (VAS). She described the pain with characteristics of an electric shock and sudden stinging.

During the history taking, she told that the pain events were triggered and exacerbated when chewing, opening the mouth, laughing, speaking and wearing the total inferior prosthesis. The episodes of pain were described as brief and very frequent and were also associated with vertigo and dizziness. She said she usually took paracetamol and dipyrone, but with no significant effect on pain relief. Furthermore, she reported headache events once to twice a week, with no nausea or changes in vision. As for the medical history, the patient reported being on pharmacological control for hypertension and hypothyroidism. The family had a history of stroke, cardiac alterations, and cancer. Regarding the psychosocial situation, she reported having little hope in the

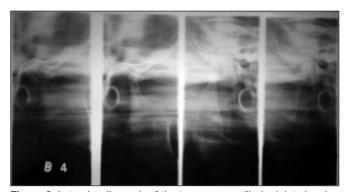
future, irritation, lack of energy, tiredness and social withdrawal since the pain started. She added that the pain interferes with her sleep and disrupts her daily activities.

In the physical examination, the cranial nerves were normal, except for the glossopharyngeal, due to the motor limitation of the posterior region of the tongue, that was attributed to the algesic condition. Even a light touch on the mucosa of the lower-right edge and the base of the tongue generated a painful response. The application of topical benzocaine at 20% in this region led to a momentary pain depletion, confirming the GPN diagnostic hypothesis.

As an initial approach, with the purpose of confirming the diagnostic hypothesis, we prescribed carbamazepine (200mg/day) during the first seven days, and then 400mg/day. It was further requested a panoramic radiograph (Figure 1) and lateral X-Rays of the temporomandibular joint (TMJ) (Figure 2), to rule out somatic causes linked to the masticatory system.



**Figure 1.** Panoramic radiograph within the normality standards for the age and dental condition of the patient



**Figure 2**. Lateral radiograph of the temporomandibular joint showing absence of external interference at the site of pain

When the patient returned 20 days later, she reported a significant improvement in pain (VAS=5). Pain remission with the use of carbamazepine strengthened the diagnostic hypothesis of neuropathic pain. The dose was adjusted to 200 mg/day because of complaints of drowsiness with the use of the drug. With the diagnostic hypothesis established, we perceived the need for multidisciplinary care. Therefore, the patient was referred to the neurology service of a local hospital, where the magnetic resonance imaging (MRI) presented an expansive, solid, extra axial lesion in the right prepontine cistern, suggesting meningioma (Figure 3).

In the five-month follow-up, the episodes of pain during the day were described as rare and of less intensity. The inferior prosthe-

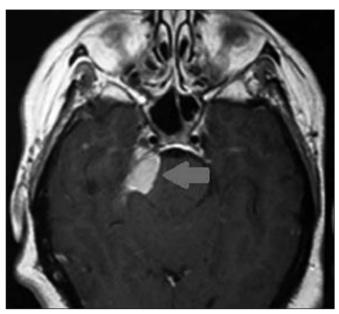


Figure 3. Magnetic resonance imaging axial view showed a lesion suggestive of meningioma indicated by the arrow

sis was adjusted aiming at the elimination of possible compression in the sublingual trigger areas. The dose of carbamazepine was adjusted from 200 to 400mg/day.

She returned six months later reporting the absence of pain events. Currently, the patient has quarterly consultations with a neurologist and is waiting for availability for neurosurgery.

# DISCUSSION

The nosological limit of orofacial neuropathic pain can be difficult to measure due to the absence of clinically diagnosable injury, the disproportion between the painful stimulus and the response, and the overlapping of structures with distinct innervation. Although it is not possible yet to define a universal consensus on the diagnostic criteria, several clinical characteristics are suggestive of these diseases, such as paroxysmal pain along the pathway of the affected nerve, allodynia, hyperalgesia and trigger areas able to trigger short-term pain events, with the characteristic of electric shock<sup>10</sup>. In this clinical case, the patient sought help at a dental center with the main complaint of intense pain in the lower edge region and on the right side of the base of the tongue, suggesting a possible problem of the prosthesis. However, the observation of the characteristics of the pain and the physical examination conducted led to the hypothesis of GPN, a possibility reinforced by the fact that the patient was over 50 years, the age group where painful facial neuropathies become more common<sup>9,11</sup>. MRI was performed to rule out secondary neuralgia to intracranial injuries. This procedure is justified by several authors<sup>11,12</sup>.

We also noticed that the pain reported by the patient was unilateral, a factor supported by the literature, that demonstrates that neuropathic pain is, for the most part, established on only one side, being bilateral in rare cases<sup>6</sup>.

In GPN, painful events can occur spontaneously, but they are generally associated with a specific trigger stimulus. Some stimuli

are chewing, swallowing, coughing, yawning, sneezing, blowing the nose, touching the ear, talking, laughing, ingesting acid, sweet, cold or hot food, or even turning the head to one side<sup>1,13</sup>. Such stimuli were reported by the patient, reinforcing the diagnosis for GPN.

In these cases, when daily and common activities become pain triggers, the QoL of the patient can be seriously compromised, with a heavy impact on its physical and emotional well-being<sup>1</sup>. In this sense, the patient of this case study reported that pain disrupted her sleep and daily activities, causing irritation, lack of energy, tiredness, and social withdrawal.

To relieve pain, the patient took paracetamol and dipyrone, with no satisfactory results. Superficial somatic pain, such as the ulceration of the oral mucosa that often affects patients wearing a prosthesis, usually improves with this kind of drug. However, neuropathic pain such as GPN does not respond to common pain medications<sup>10</sup>.

Pain assessment using VAS throughout the reassessment period showed a general reduction in the painful condition. VAS is considered a useful tool with good reproducibility to measure pain<sup>13</sup> because it turns the subjectivity of a painful experience into objective for clinical evaluation.

During the physical examination, the momentary depletion of pain by the application of benzocaine at 20% in the posterior region of the tongue, region innervated by the glossopharyngeal nerve, confirmed the diagnostic hypothesis of GPN. This response to the topical anesthetic on the trigger area in the mucosa is considered a useful clinical test for differential diagnosis<sup>6</sup>. Studies have shown that pain can be proven by the stimulation of specific points in the area of superficial distribution of the glossopharyngeal nerve<sup>14</sup>. Therefore, the adjustments made on the base of the inferior prosthesis aiming at the reduction of the compression in these trigger areas are justified.

Pharmacotherapy is the first line of treatment for GPN. The drugs of choice are carbamazepine, gabapentin, and pregabalin<sup>15</sup>. The selection of carbamazepine is based on several studies in which this drug proved effective for the treatment of paroxysmal nerve pain<sup>16,17</sup>. However, many patients are sensitive to carbamazepine and develop adverse reactions, especially drowsiness, dizziness, vomiting, diarrhea, rashes or even bradycardia<sup>17</sup>. Such effects can be minimized when the dose is gradually increased<sup>16</sup>. Therefore, it is crucial that the dental surgeon know the clinical characteristics of neuralgias that involve the face and be prepared to perform the differential diagnosis. In the diagnostic hypothesis of paroxysmal neuropathic pain, ruling out secondary neuralgia is a priority, and the referral to a neurological evaluation becomes of key importance.

## CONCLUSION

The dental surgeon should be skilled to make the differential diagnosis of orofacial pain, especially of paroxysmal neuralgia. Recognize the clinical characteristics and properly refer the patient to a neurological center is fundamental for a good prognosis, especially in cases of neuralgia of tumor etiology.

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