Case Report

Altered somatosensory processing in secondary trigeminal neuralgia: A case report

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Abstract

Secondary trigeminal neuralgia might be very rarely preceded by trigeminal neuropathic pain. The patient, in this case, presented with paroxysmal pain in the left mandible and numbness of the lower lip and tongue. Sensory testing of these areas revealed cold and heat hyperalgesia and mechanical hyposensitivity in the mandibular region. Magnetic resonance imaging showed a mass in the left cerebellopontine angle. The patient was prescribed systemic mirogabalin (2.5 mg/day), which provided some relief until the tumor was removed. The histopathological diagnosis was an epidermoid tumor. This article discusses the clinical characteristics and sensory testing findings that distinguish secondary trigeminal neuralgia from trigeminal neuropathic pain based on the International Classification of Orofacial Pain.

Keywords: International Classification of Orofacial Pain, secondary trigeminal neuralgia, sensory testing

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INTRODUCTION

According to the International Classification of Orofacial Pain, space-occupying lesions can cause both secondary trigeminal neuralgia and trigeminal neuropathic pain.[1] The clinical characteristics can help distinguish between the two; however, the features of trigeminal neuropathic pain, such as hyperalgesia, allodynia, hypoesthesia, and hypoalgesia, might precede secondary trigeminal neuralgia.[2]

To our knowledge, the results of a quantitative sensory testing (QST) protocol for the assessment of trigeminal neuropathic pain due to a brain tumor have not been reported to date. Herein, we report the case of trigeminal neuropathic pain that might predict secondary trigeminal neuralgia.

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CASE REPORT

Patient presentation

A 48-year-old female presented with a chief complaint of paroxysmal pain in the left mandible and numbness of the lower lip and tongue and "wrenching" and "cutting" pain in the left tongue. The pain intensity was moderate with occasional exacerbations of severe headache from the left temporal to the mandibular region, lasting 1–2 min.

The initial onset of the patient's chief complaints was 1 year and 8 months ago, when she became aware of numbness on the left part of the tongue and lower lip. Six months

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later, she developed severe paroxysmal pain in the lower lip on the left side when drinking cold water; however, the pain resolved spontaneously. One month before presenting at our clinic, the patient developed paroxysmal pain in the entire lower jaw and temporal region on the left side, lasting 1–2 min, while washing the face, touching the cheeks, eating, and facing downwards. Numbness persisted since its initial onset but remained confined to the lower lip and tongue on the left side.

Examination of the temporomandibular joint revealed an active range of motion of >40 mm and tenderness in the left masseter muscle. Cranial nerve examination revealed slight sensory loss to light touch in the lower lip and tongue on the left side.

A panoramic radiograph showed radiolucent areas at the apices of the mandibular left second premolar and maxillary left second premolar and second molar [Figure 1]; however, provocation testing of these teeth did not reproduce the familiar pain.

Quantitative sensory testing

The QST protocol of the German Research Network on Neuropathic Pain was followed.[3] The values were abnormal for the cold pain threshold (CPT = 25.8°C), heat pain threshold (HPT = 40.3°C), and mechanical detection threshold (MDT = 1.0 mN). The MDT value indicated a loss of function, whereas the CPT and HPT values indicated a gain of function.[3]

Magnetic resonance imaging findings

Magnetic resonance imaging scans revealed a well-defined, intensely enhanced 23 mm × 10 mm mass in the left cerebellopontine angle [Figure 2a], compressing the left trigeminal nerve, which was equal in signal to the cerebrospinal fluid [Figure 2b-d]. The radiological diagnosis was an epidermoid tumor. Although the first line of treatment for trigeminal neuralgia is carbamazepine,[4] the patient was prescribed mirogabalin (Tarlige®) (2.5 mg/day) due to allergies. She experienced some relief 3 days later; however, she discontinued the treatment due to dizziness and drowsiness. The patient was referred to a neurosurgeon who excised the tumor under general anesthesia. The facial pain disappeared immediately thereafter; however, partial numbness in the tongue and lips persisted.

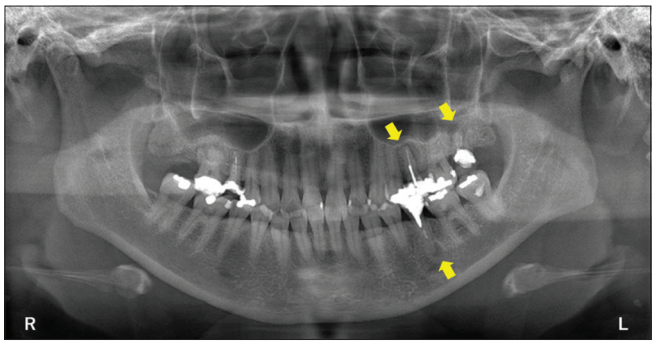

Figure 1: Panoramic radiograph shows radiolucent areas at the apices of the mandibular left second premolar and maxillary left second premolar and second molar

Histopathological report

The specimen was a cystic lesion backed by a nonthinned flat epithelium, and the content was mainly a stalk. The histopathological diagnosis was an epidermoid tumor.

DISCUSSION

Secondary trigeminal neuralgia is diagnosed when there are the symptoms of classical trigeminal neuralgia, including paroxysmal pain, caused by underlying diseases, such as space-occupying lesions.[5] Based on the anatomical findings, Kitahara *et al*. classified the relationships between tumors (acoustic nerve tumor, meningioma, and epithelioid tumor) and the trigeminal nerve and cerebral blood vessels into three groups.[2,6] In the Type 1 relationship, the tumor compresses a blood vessel that compresses the root entry zone of the trigeminal nerve, causing trigeminal neuralgia. In the Type 2 relationship, the tumor compresses the trigeminal nerve that is compressed by the artery opposite to the tumor (commonly the superior cerebellar artery), causing trigeminal neuralgia. The Type 3 relationship has three subgroups, in which the tumor directly compresses the trigeminal nerve, causing trigeminal neuralgia.

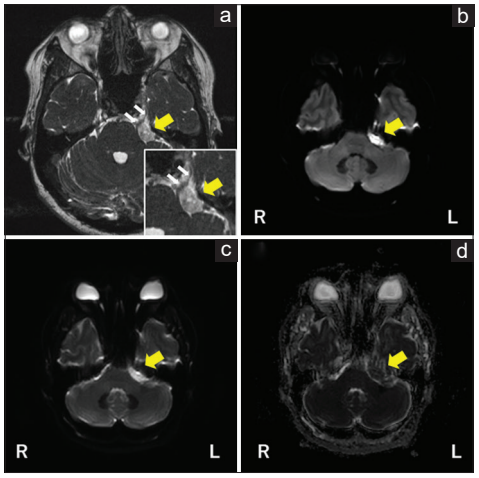

Figure 2: Magnetic resonance imaging revealed a well-defined, intensely enhanced mass in the left cerebellopontine angle (a), compressing the left trigeminal nerve (b and c). The pons was slightly compressed by the mass (d), which extended from near the centre of the pontine tank on the rostral side to the left side of the medulla oblongata on the caudal side

In this case, the anatomical relationship was Type 3, with the tumor directly compressing the trigeminal nerve root. Cerebrovascular vessels, such as the superior cerebellar artery and the anterior inferior cerebellar artery, were not involved in the compression of the trigeminal nerve root. Our patient exhibited cold and heat hyperalgesia and mechanical hyposensitivity in the mandibular region on the affected side. The cold threshold is mediated by Aδ and C-fibers, whereas the heat threshold is mediated by C-fibers. The patient complained of severe recent pain when drinking cold water, which was likely a clinical manifestation of cold hyperalgesia.

Svensson *et al*. developed a simple neurosensory testing that can be used by the general dentist.[7] It can be performed as follows: A metal spatula is cooled in a refrigerator until it is 5°C or heated in a water bath until it is 40°C and then applied to the affected area and contralateral unaffected area to test for the differences in the temperature sensitivity. A cotton swab is applied to the affected area and the contralateral unaffected area to assess for differences in the detection of light pressure.

Temporomandibular disorders (TMDs) are the most common type of nonodontogenic pain. The diagnosis is often elusive, as it requires a comprehensive assessment.[8] In this case, the entire left mandible was affected during severe pain, which required distinguishing it from TMD.

In cases where dentists have not arrived at a definitive diagnosis, irreversible treatment should be avoided and various diagnostic techniques should be explored.[8-10]

CONCLUSION

Trigeminal neuropathic pain might precede secondary trigeminal neuralgia. When patients with sensory impairment visit a general practitioner, such as a dentist, the clinical features should be identified and simple neurosensory testing should be performed to differentiate between the two.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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