

VESTIBULAR SCHWANNOMA (ACOUSTIC NEUROMA) MIMICKING TEMPOROMANDIBULAR DISORDERS: A CASE REPORT

SCHWANNOMA VESTIBULAR (NEURINOMA DO ACÚSTICO) IMITANDO DESORDENS TEMPOROMANDIBULARES: UM RELATO DE CASO

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ABSTRACT

A pproximately 6 to 16% of patients with trigeminal neuralgia symptoms present intracranial tumors, the most common being the vestibular schwannoma (acoustic neuroma). Some symptoms reported by patients include hearing loss, tinnitus, headaches, vertigo and trigeminal disturbances. An increased muscle response in the surrounding head and neck musculature may also be observed, which mimics signs and symptoms of temporomandibular disorders. In these cases, magnetic resonance imaging (MRI) has proved to be a useful tool in tumor diagnosis. The differential diagnosis between myofascial and neuralgic pain is important, as both may present similar characteristics, while being of different origin, and demanding special treatment approaches. The purpose of this paper is to demonstrate the relationship among trigeminal neuralgia symptoms, intracranial tumors and temporomandibular dysfunction by presenting a clinical case.

Uniterms: Temporomandibular disorders; Temporomandibular joint; Chronic pain; Orofacial pain; Acoustic neuroma; Vestibular schwannoma; Schwannoma.

RESUMO

A proximadamente 6 a 16% dos pacientes com sintomas de neuralgia trigeminal apresentam tumores intracranianos, sendo mais comum o schwannoma vestibular (neurinoma do acústico). Alguns sintomas relatados pelos pacientes são perda da audição, zumbido, dores de cabeça, vertigens e distúrbios trigeminais. Uma resposta muscular aumentada na musculatura associada da cabeça e do pescoço também pode ser observada, o que pode mimetizar sinais e sintomas de desordens temporomandibulares. Nestes casos é de grande valia o uso de imagem de ressonância magnética (IRM) para detecção de tumores. É importante, também, a diferenciação de dores miofasciais e neurálgicas, pois ambas podem apresentar características semelhantes, mas com origens e tratamentos diferentes. O objetivo desse trabalho foi demonstrar através de relato de caso clínico a associação entre sintomas de neuralgia trigeminal, tumores intracranianos e disfunção temporomandibular.

Unitermos: Disfunção temporomandibular; Dor orofacial; Articulação temporomandibular; Dor crônica; Neurinoma do acústico; Schwannoma vestibular.

INTRODUCTION

The term acoustic neuroma defines a benign tumor of the Schwann cell neurilemma, which grows mostly in the lower vestibular nerve of the 8th cranial nerve⁷. From 1991 on, vestibular schwannoma (VS) became the most appropriate term, representing the real situation for the majority of cases. Vestibular schwannomas are among the most commonly diagnosed tumors and account for approximately 6% of all brain tumors³⁴.

Vestibular schwannomas are characterized by the slow progression of hearing loss, with or without balance loss. This happens because these tumors grow slowly, causing such a gradual decrease in labyrinth stimulation, that the central compensation mechanisms are able to reduce the impact of these symptoms²⁷. On the other hand, schwannomas that reach the trigeminal cranial nerve are characterized by pain and facial numbness. This pain frequently resembles trigeminal neuralgia^{26,34}. In schwannomas reaching the trigeminal nerve, although the facial nerve is anatomically impaired by the tumor growing adjacent to it, seldom does facial paralysis appear in clinical cases, which is a sign of the remarkable resistance to compression exhibited by the 7th nerve³⁵.

The afferent root of the trigeminal nerve, which links the Gasserian Ganglion and the pons, may be a site for schwannomas, with the tumor growing in the cerebellopontine angle^{7,34,35}. The majority of tumors growing in this region is comprised by vestibular schwannomas. At times, the trigeminal nerve can be partly affected by schwannomas, as the latter occurs adjacent to the former.

Vestibular schwannomas may be classified as small, medium or large. Small neuromas are characterized by appearing only in the pons, where nerves responsible for auditory performance, equilibrium and movements of motor muscles as well as some vessels of the inner ear are found. The medium-sized neuromas stretch from the pons to the cranial cavity, yet without compressing any brain structure. On the other hand, larger neuromas stretch outside the internal canal towards the cranial cavity producing some pressure on the brain and thus altering important vital centers².

Trigeminal neuralgia is a symptom that may be present in vestibular schwannoma cases³⁸. It is described by the International Association for the Study of Pain (2004) as 'unilateral facial pain, resembling an electric discharge, limited to one or more branches of the trigeminal nerve path' ¹³. The pain is triggered by ordinary sensorial stimuli such as washing the face, shaving, applying make up, drinking water, speaking, brushing the teeth, among others²⁴. The pain lasts only a few seconds³, and the clinical diagnosis is based on the patient's history⁴⁰.

In a study using magnetic resonance imaging, Yang, et al.²⁸ observed that in 51 patients suffering from trigeminal neuralgia, 16% (8) presented vestibular schwannoma³⁸. Similarly, Samii and Matthies²⁸ reported that between approximately 1% and 3% of vestibular schwannoma patients presented trigeminal neuralgia symptoms.

Nevertheless, Selesnick, et al.³⁰ reported that in a group of 126 patients with vestibular schwannoma, none of them presented neuralgia.

Prevalence

Of every 100 temporal bones selected and submitted to post-mortem anatomy studies, one presents scwannoma on the vestibular nerve. The prevalence of this disorder, considering the current diagnosis conditions, is from 7 to 9 in 1,000,000 subjects³⁶. Schwannoma occurs slightly more often in women (59 to 62%)¹⁴. It happens independently of ethnicity and is more frequently diagnosed in men within the 50-60 year old age group (61%)⁷. It is estimated that between 2,000 and 3,000 new unilateral vestibular neuroma cases are diagnosed yearly (incidence) in the US, which characterizes a 1:100,000 occurrence¹. Research has shown that unilateral neuroma is not hereditary, and only one in 1,000 cases occurred in which mother and daughter suffered from the disorder^{7,14}.

Microscopic Findings

As a rule, two kinds of microscopic findings can be observed according to the description elaborated by Antoni in 1920. First, vestibular schwannoma of Type A is defined by a more closely packed tissue in which cells form a palisade pattern and in a bipolar, parallel cell arrangements disposed in interlacing bundles that alternate with enucleate fibrilar zones composed of cellular processes. Elongated nuclei occur typically^{4,7,16}. Second, type B occurs as a more loosely organized tissue, with a more noticeable reticulated appearance and interstitial edema⁷. Neoplastic cells having condensed nuclei and indistinguishable cytoplasm are observed, which may look like lymphocytes at first sight⁴. Using MRI (T2), type B corresponds to a hyposignal image due to its characteristic interstitial edema¹⁵.

Diagnosis and Prognosis

The vestibular signs, the effect on the trigeminal nerve, and the cerebellar and intracranial pressure signs may all become noticeable with tumor growth, thus enabling the delineation of a sequential and evolutionary pattern⁷. The clinical signs may be characterized as: a) progressive unilateral or asymmetrical sensorineural dysacusis, b) intermittent hearing loss, c) tinnitus and sensation of fullness in the ear, d) lasting positional vertigo, e) difficulties in walking, f) visual blurring, g) trigeminal neuralgia, and e) headaches^{7,10,19,20,40}. Hearing ability is normal in at least 8% of cases, and tumor is suspected in the case of unusual or bizarre complaints that are investigated by MRI⁷.

Matsuka, et al.²⁰ listed the following symptoms related to vestibular schwannoma: hearing deficits (60 to 97%), tinnitus (50 to 66%), vestibular disturbances (46 to 59%), numbness or tingling of the face (33%), headaches (19 to 29%), dizziness (23%), Bell's palsy (17%), and trigeminal disturbances – hyperesthesia, paresthesia, and neuralgia (12 to 45%).

Differential diagnosis between acoustic and trigeminal neuromas is fundamentally based on early impairment of auditory acuity ipsilateral to the lesion in patients with 8th nerve schwannoma^{7,34,35}. The use of imaging techniques is also necessary to diagnose the tumor. MRI is more advantageous as compared to CT scans and to any other imaging technique when identifying lesions in the trigeminal nerve path, and it is considered the technique of choice in the diagnosis of vestibular schwannomas¹². Although the schwannoma is benign because of growing in the vestibule, morbidity is high due to the compression of vital structures such as the cranial nerves and the brain¹.

Case report

A 59-year-old woman was referred to the Center for Post Graduation Studies on Pain and Temporomandibular Disorders (TMD) of the Brazilian Dental Association – State of Rio Grande do Sul (ABO-RS) with acute pain around the lower lip, jaw, and right temporomandibular joint (TMJ), which impaired mouth opening. The main complaint was the difficulty to open the mouth due to shooting pain attacks on the right side of the face. Pain happened along the trigeminal nerve path, yet not reaching beyond the median line. Pain attacks were typically sudden, acute, short and episodic. The patient's previous history, assessed by the HAD questionnaire (Hospital Anxiety and Depression Scale), revealed the clinical signs characteristic of depression and anxiety²². Pain prior to a car accident was also reported. The patient also described persistent and localized episodes of bilateral pain in the left and right masseter as well as in the temporalis muscles which increased with function, and the presence of painful hypertrophic bands. When palpating the right masseter, pain was felt in the insertion region and in the muscle itself and was graded as 3 by the patient on a four-point scale (0 = no pain, 1 = report of discomfort, 2 =report of pain, 3 = report of pain with withdrawal reflex). On the left side, pain happened in the masseter itself, and was graded as 1. As for right and left temporal muscles, the patient reported grade 1 pain in all muscle sheaths. Pterygoid muscles were also reported to be affected by the pain on both sides, graded as 3.

A visual scale was used to measure pain. The patient characterized facial pain episodes as grade 10, while muscle pain was graded as 7. This pain was graded 8 on average for the past 6 months. The patient's everyday chores as well as professional work was impaired by the pain, which also affected her social and family life. Furthermore, the patient exhibited clicking and popping sounds in both TMJs and reported displaced bite, a history of mandible locking, both at night and during the day, teeth grinding, and a certain degree of discomfort in the morning.

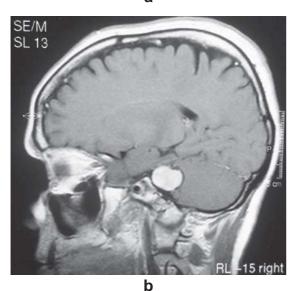
The Research Diagnostic Criteria for Temporomandibular Disorders (RDC/TMD) was used to assess the patient's physical characteristics. On the first occasion, this test was not completed, because of the very small mouth opening (26 mm). The patient was instructed to make use of moist heat on the affected muscle area for 20 min, 3 times a day, for fourteen days. A non-steroid anti-inflammatory drug selective cyclooxygenase 2 was prescribed, three times a day, for the same period.

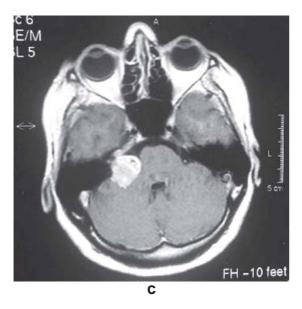
On a second assessment, the RDC/TMD questionnaire was again applied to the patient. Mouth opening was 36 mm, painless and with no need for intervention. With assistance, the patient was able to further open the mouth 44 mm with intense pain. An overbite of 3 mm was observed, along with a lateral centric slide of 2 mm to the left of the median line. The opening pattern was S-shaped, deviated to the right side, presenting a 4-mm maximum lateral excursive movement to the right and 12 mm to the left. Concerning articular sounds, a click was observed on the right side, at 23 mm opening, and on the left, at 25 mm. On closing, clicking was heard at 21 mm on the right and 25 mm on the left. Reciprocal sounds ceased in protrusive movement on both sides, but they were present in lateral movements.

In this particular case, there were signs and symptoms indicative of neuralgic pain of the fifth branch (trigeminal division), probably related to brain tumor. A physician, under the recommendation of the dentists supervising the case, requested a MRI evaluation, which was performed forty days after the clinical evaluation. MRI revealed a slight disk displacement, with a decrease on the right side and an extensive tumoral lesion in the right acoustic channel stretching to cerebellopontine angle (Figure 1a), suggesting a Vestibular Schwannoma.

Initially, the treatment proposed was to reduce localized muscle pain using anti-inflammatory drugs, muscle relaxation drugs, and moist heat in the affected muscular region, for four days. Concomitantly, a Michigan-type bite plane was manufactured to stabilize occlusion and to rule out dental influence as a trigger to central pain, to decrease abnormal muscle activity, and to diminish muscle pain sensitivity. In the follow-up period, the splint underwent adjustments, and from that moment on, the patient started using the splint both at night and during the day, removing it only to eat and clean the teeth. With sequential adjustments and frequent controls, the patient reached a favorable response to the use of the splint, improving the pain prospects. The splint was indicated due to the presence of muscle and skeletal pain indicative of TMD in conjunction with stabilizing agents. There was an improvement not only in the muscle and skeletal pain, including maximum mouth opening, but also in the neuralgic pain. The patient was referred to a psychiatrist for a more comprehensive evaluation of her psychological condition, which revealed symptoms that were suggestive of anxiety and depression. The patient was advised to also see a brain surgeon to evaluate the tumoral lesion (Figures 1b and 1c) and to investigate dizziness, vertigo and neuralgia. After medical assessment, the patient began to take sertraline, carbamazepine and clonazepam, with subsequent decrease in pain crises. The tumor was surgically removed. In this particular case, the facial nerve was damaged causing hearing loss and Bell's palsy on the affected side. However, the pain subsided and no longer affected the patient.







FIGURES 1a, 1b, 1c- Frontal, sagittal and horizontal view (arrows) of an vestibular schwannoma (vestibular schwannoma) of the eight cranial nerve in the internal auditory canal

DISCUSSION

As a rule, vestibular schwannomas grow slowly²⁰. In the present study, MRI showed that there was a 0.5-cm growth in 8 months. Due to this slow growth and to the neurological adaptations undergone by patients, clinical symptoms are concealed and at times only superficially observed and investigated^{8,18,30}.

Facial pain caused by tumors is often related to neurological abnormalities such as: a) sensory changes, b) loss of reflexes, and c) constant pain. Nevertheless, these neurological symptoms may be misdiagnosed or wrongly interpreted in some cases²⁰. Patients place little value on these symptoms, because they are not very intense and non-debilitating and because they ignore the need for more specific treatment. According to Matsuka, et al.²⁰, patients take between 0.6 and 5 years to look for medical care. Similarly, the case study presented here revealed that the patient had felt myofascial pain as early as at 8 years of age.

Regarding differential diagnosis, some of the symptoms described in the literature, which are associated to the internal auditory canal, are otovestibular dysfunctions, such as hearing loss, tinnitus, vertigo and dizziness⁶. In the present case report, such symptoms were occasionally experienced. According to Fricton, et al. 11 (1985), patients with temporomandibular disorders may also describe similar otologic and neurological symptoms. For example, 42.1% of 164 TMD patients presented tinnitus, 41.5% perceived ear pain, 23.1% vertigo, and 17.7% perceived hearing loss. In addition, 27.4% of the subjects presented tingling, 26.2% numbness, 14% blurred vision, 12.2% twitching, 7.9% tremors, and 7.3% lacrimation. These symptoms might be explained by peripheral and central sensitization due to ongoing inflammatory process in the masticatory muscles and/or temporomandibular joint or may be in fact medical conditions with associated and reflex muscular and skeletal symptoms, similar to TMD³¹. At the present time, medical imaging, particularly MRI, as well as a multidisciplinary assessment, are probably the greatest tools in differentiating the two possible sources of pain as well as otologic and neurological symptoms. The MRI is indicated when trigeminal neuralgia is suspected, considering that 10% of trigeminal neuralgias are caused by intracranial tumors^{28,30,38}.

Regarding treatment, the differential diagnosis described above and the multidisciplinary team approach for chronic pain management are of utmost importance. If TMD is suspected to be the major etiology after negative confirmation of neoplasia (particularly vestibular schwannoma), an interocclusal Michigan bite appliance may be sufficient to decrease all symptoms. In the clinical case presented here, the patient did improve after the use of a Michigan-type bite splint, but this treatment was aimed towards the secondary muscle pain (TMD type) caused by the vestibular schwannoma. The slight improvement in the neuralgic symptoms might be due to placebo effect. In other words, in this case, the TMD signs and symptoms were a consequence of the tumor (referred pain and muscle splinting) rather than its cause. For a neuralgic pain

secondary to a brain tumor, anticonvulsants are the drugs used at the beginning of treatment to prevent and treat dizziness as well as in migraine prophylaxis. This class of drugs is also used in the management of chronic pain (neuropathies and neuralgias)²⁵. For trigeminal neuralgia, carbamazepine is the drug of first choice, controlling pain in 70-98% of cases. Some patients may present drug intolerance^{29,40}.

Bullit, et al.³ reported that carbamazepine is an efficient drug in the treatment of trigeminal neuralgia associated to intracranial tumors. Other studies have also proved the efficiency of other drugs as gabapentine in trigeminal neuralgia therapy^{5,33}. The action of gabapentine has not been fully established and may involve the modulation of gama-aminobutiric acid and glutamate, or even have an effect on calcium channels²⁵. With the prescription of such drugs, the patient reported a decrease in the occurrence and duration of crises. However, Yang, et al.³⁸ reported an increase in the number of patients suffering from trigeminal neuralgia associated with intracranial tumors which are refractory to these drugs.

Other drugs may be useful in the control of neuralgic pain, such as baclofen, lamotrigine and tricyclic antidepressants. In the case presented here, the pain attacks were controlled, but crises still persisted due to the delay of surgery. Tumors that involve the trigeminal nerve are uncommon, but are an important cause of trigeminal neuralgia and myofascial pain³. Because of the similar characteristics, the concomitant occurrence of myofascial and neuropathic pain makes the diagnosis difficult. The differential diagnosis between these types of pain is necessary for their correct management. Localized myofascial pain increase in intensity when the affected region is palpated, and that one which occurs with hypersensitive bands and trigger points were all symptoms reported by the patient reported here. Neuropathic pain is generated in the central nervous system and may simulate a superficial somatic pain, similar to the myofascial pain²³.

The treatment must be appropriate and specific for the different types of pain. The use of anti-inflammatory and central muscle relaxation drugs together with moist heat becomes effective in the control of somatic myofascial pain. The neuralgic pain must always be investigated in detail, as it yields debilitation and also because such pain may at times come from intracranial tumors³. The use of imaging in such cases becomes indispensable. The MRI is the most effective technique to detect intracranial structural lesions. It has advantages over CT and any other type of imaging in the identification of soft tissues lesions, nerve paths and presence of tumors, as mentioned above 15,17. The improved resolution for soft tissues and the capacity to visualize multilayer sections contribute to a better assessment of intracranial anatomic segments of the trigeminal nerve.

Some authors have reported that conservative treatment may be a choice for patients with few symptoms, small lesions, and who are not able to undergo surgery, due to the slow tumor growth. Surgical procedures are the best choice for patients who did not respond favorably to conservative treatment³⁹. The prognosis of the vestibular schwannoma surgery is good, although all surgical procedures are under a certain morbidity risk^{16,21}. Hearing impairment in the affected side is a likely outcome as well as a lesion in the trigeminal nerve. Samii and Matthies²⁸ observed that after complete removal of 979 vestibular schwannomas, in 93% of cases, there was preservation of the facial nerve. These findings were also corroborated by Yamakami³⁷. However, these two authors found major neurological side effects which included paresthesia, hematomas, brain and spinal fistulas, hydrocephalus, bacterial meningitis.

The literature reveals that there might be a need for a clinical assessment when an intracranial tumor, such as the vestibular schwannoma, is diagnosed and when the patient initially reports unilateral hearing loss, apart from tinnitus. A clinical diagnosis must be carried out, together with an investigation of the patient's history and imaging techniques such as MRI.

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