

A Case of Vestibular Schwannoma with Oral Burning Sensation: Surgical Complication or Burning Mouth Syndrome?

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Case report

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Abstract

Background: Vestibular schwannoma (VS) or acoustic neuroma is a benign brain tumor. Despite improvements in surgical technique, the removal of VS is related to some complications of which the recovery is difficult and sometimes lead to permanent deficits. About 10.7% of complication of vestibular schwannoma surgery is reported to affect facial or tongue area, including numbness of pain, which is similar to a typical symptom of burning mouth syndrome (BMS). However, up to our knowledge, there is no documented case of BMS either comorbid with vestibular schwannoma or occur due to surgery complications.

Clinical Presentation: A 46-year-old woman complained about left-sided facial pain, a burning sensation on the left side of her tongue and maxilla, and a spontaneous bitter taste. Four years before her first visit to our clinic, left-sided facial pain and burning sensation on the left side of the tongue suddenly became severe. She visited a neurosurgeon and VS was found in the left cerebellopontine angle. After the tumor was removed, facial pain, burning sensation of the tongue, and bitter taste improved simultaneously but quickly recurred after a while. The patient had a panic attack because of severe pain and started visiting a psychiatrist. The psychiatrist diagnosed her as having somatic symptom disorder and depression, prescribing sertraline 100 mg, zolpidem 5 mg, and lorazepam 1.0 mg and referred her to our clinic. At the first visit, no abnormal intraoral nor extraoral findings were detected. Based on the characteristics of the pain, we made a diagnose of BMS. Amitriptyline was initiated at 10 mg/day and increased to 30 m. Within 1.5 months, the pain and burning sensation of the tongue and maxilla almost remitted completely, whereas bitter taste showed moderate improvement.

Conclusions: Our case suggests there might be an exceptional case in which BMS and VS can occur simultaneously. Specifically, an oral burning sensation and dysgeusia after VS surgery are not always a case of surgical complication or untreatable.

Background And Importance

Vestibular schwannoma (VS) or acoustic neuroma is a benign brain tumor that accounts for approximately 9% of intracranial tumors and 80% of tumors in the cerebellopontine angle [1]. Despite improvements in surgical technique, the removal of VS is related to some complications such as headaches, hearing loss, facial weakness, eye problems, and other issues depending on the patient's sex, age, and tumor size [2, 3]. Wiegand et al. reported the surgical complications of VS and its treatment; suggested the recovery from complications is often difficult and complications sometimes lead to permanent deficits [4, 5].

BMS presents uncomfortable sensation or oral pain in the mouth without clear clinical causes and pathophysiology [6]. BMS mainly affects middle-aged and elderly women. About 10.7% of complication of vestibular schwannoma surgery is reported to affect facial or tongue area, including numbness of pain, which is similar to a typical symptom of BMS. However, up to our knowledge, there is no documented case of BMS either comorbid with vestibular schwannoma or occur due to surgery complications. Here, we report a case of which VS occurs simultaneously with burning mouth syndrome (BMS).

Clinical Presentation

A 46-year-old woman complained about left-sided facial pain, a burning sensation on the left side of her tongue and maxilla, and a spontaneous bitter taste. The symptoms were limited to the left side of the face and showed daily fluctuation. Four years before the patient's first visit to our clinic, left-sided facial pain and burning sensation on the left side of the tongue suddenly became severe. She visited a neurosurgeon and VS was found in the left cerebellopontine angle (Fig. 1 (a)). After the tumor was removed nearly totally at a university hospital, facial pain, burning sensation of the tongue, and bitter taste improved simultaneously but quickly recurred after a while.

During the last four years, the pain and bitter taste often fluctuated. Two months before the initial visit to our clinic, the pain became more severe than ever before. The patient had a panic attack, started visiting a psychiatrist, and then quit her job as a nurse. The psychiatrist diagnosed her as having somatic symptom disorder and depression, prescribing sertraline 100 mg, zolpidem 5 mg, and lorazepam 1.0 mg and referred her to our clinic. The patient has no other significant medical history. At the first visit, no abnormal intraoral nor extraoral findings were detected. The Saxon test's result was 2.03 g/ml; which was in a normal range. Semmes-Weinstein monofilament testing also revealed no sensory abnormality. There was no ulcer or redness and swelling of the oral mucosa. The visual analog scale for pain, pain catastrophizing scale, and Zung self-rating depression scale scores were 48, 20, and 64, respectively [7]. Eating food eased the symptoms. While taste perception was within the normal limit on the right side, the filter-paper disk test revealed severe loss of taste on the left side of the tongue, in which the patient could not sense sweet and bitter tastants and barely sensed salty and sour tastants. The left trigeminal nerve was not depicted clearly on Magnetic resonance imaging (Fig. 1 (b)). The oral burning and dysgeusia were partially explained by VS [8]. However, based on the characteristics of the pain, such as its daily fluctuation and attenuation with eating, the symptom seemed not to fit well with typical neuropathic pain.

Thus, we made a diagnose of BMS. Due to partial remission owing to sertraline, amitriptyline was initiated at 10 mg/day and increased to 30 mg with careful observation of effectiveness and side effects. Within 1.5 months, the pain and burning sensation of the tongue and maxilla almost remitted completely, whereas bitter taste showed moderate improvement. She stated that she could return to her job and even enjoy a family trip, which was impossible before. In the subsequent 1.5 years, the patient has continued to take amitriptyline 30 mg/day without any side effects, her facial pain and burning sensation of the tongue disappeared completely.

Discussion

Regarding complications of VS surgery, pain in the trigeminal nerve area is reported [3]. However, in this case, tongue pain preceded the detection of the VS and did not disappear after surgery, therefore it might not represent an example of a surgery complication. The oral burning pain could come from BMS, while the VS could happen as coincidental comorbidity.

According to the International Association of Pain and International Headache Society, the criteria for BMS diagnosis excludes organic abnormalities, such as brain tumors [9]. Our case suggests there might be an exceptional case in which BMS and VS can occur simultaneously. Specifically, an oral burning sensation and dysgeusia after VS surgery are not always a case of surgical complication or untreatable. Hence, careful consideration when applying strictly the present diagnostic criteria is essential for clinicians.

Abbreviations

VS
Vestibular schwannoma
BMS
Burning mouth syndrome

Declarations

CONSENT TO PUBLISH

Written informed consent for publication was obtained from all patients. A printed copy (in Japanese) is available if requested.

AVAILABILITY OF DATA AND MATERIAL

Not applicable

COMPETING INTERESTS

The authors declare that they have no competing interests.

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AUTHOR CONTRIBUTIONS

TS and AT treated the patient in the psychosomatic dental clinic. TS, TTHT and MT drafted the manuscript. TTHT, MT, CH, CT, MW, TA critically revised the manuscript. All authors have read and approved the final manuscript.

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Figures

(A)



(B)

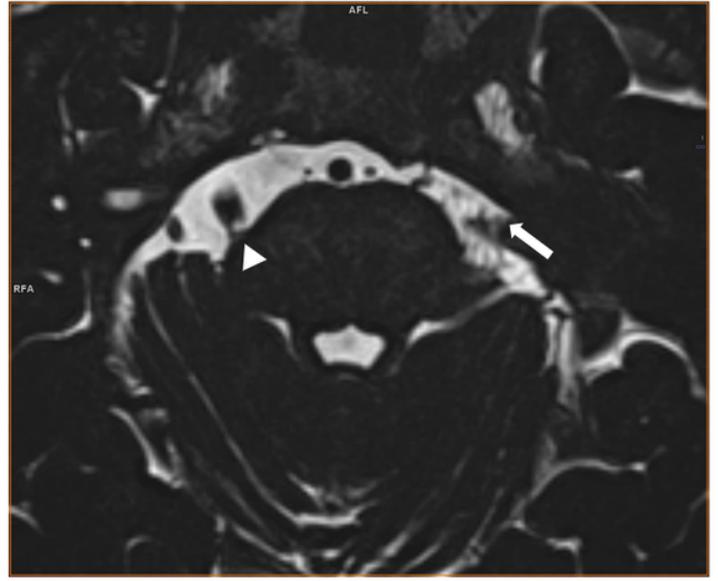


Figure 1

(a) Magnetic resonance imaging scans from 5 years before the patient's first visit to our clinic (well-defined mass involving the left cerebellopontine angle) (white arrow: vestibular schwannoma affecting left trigeminal nerve, arrow head: right trigeminal nerve) (b) Magnetic resonance imaging scans at the first visit to our clinic (white arrow: left trigeminal nerve, arrow head: right trigeminal nerve)