

Surgical Removal of Foramen Ovale Osteophyte Completely Relieved Atypical Trigeminal Neuralgia: A Case Report and Literature Review

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Abstract

Atypical trigeminal neuralgia (TN), usually caused by nonvascular compression, lacks a clearly localized trigger and complete remission periods. Although variations of foramen ovale may compress the mandibular nerve branch of the trigeminal nerve, resulting in atypical TN, only a few case reports are reported in the literature. The authors describe a case of a 50-year-old female diagnosed with atypical TN for two months. A high-resolution computed tomography imaging revealed an osteophyte of the left foramen ovale that may compress the mandibular nerve branch of the trigeminal nerve. The patient underwent osteophyte resection, and the pain disappeared completely and immediately after surgery without recurrence in the follow-up to six months. The numbness was also relieved slightly. This case provides a new perspective on the clinical diagnosis and treatment of patients with atypical TN.

Keywords: osteophyte, foramen ovale, atypical trigeminal neuralgia, resection, skull base

Introduction

Trigeminal neuralgia (TN) is a neuropathic disorder characterized by unilateral paroxysmal severe pain often described as electric shock-like, knife-like, stabbing, or burning-like in the distribution territory of the branches of the trigeminal nerve and triggered by innocuous stimuli.¹⁾ TN is commonly categorized as typical or atypical according to the nature of pain. Atypical TN is mainly characterized by a missing clearly localized trigger point and a complete remission period.²⁾ Most typical TN is caused by neurovascular conflicts in the cerebellopontine angle, while atypical TN may be associated with other etiologies, such as tumor, trauma, multiple sclerosis, etc.

A narrow foramen ovale was first suggested as a pathogenic mechanism in the development of atypical TN in the mandibular branch territory by Neto, and this hypothesis was supported by many anatomical and radiological studies.³⁾ However, surgical correction of a narrow foramen ovale for patients with atypical TN remains underexplored in the literature. The case presented here was elaborately

diagnosed by clinical and radiological analyses and was finally treated by surgically removing the foramen ovale osteophyte. To our knowledge, it is the first case of successful management of the aberrant foramen ovale to treat atypical TN.

Case Report

A 50-year-old female had uncomfotableness of the chin, mandible, and the anterior right side of the tongue due to affected trigeminal nerve for two months. This patient had no history of systemic diseases and calcium metabolism abnormalities. The symptom was manifested as persistent numbness (BNI (Barrow Neurological Institute) scored N-4) with intermittent tingling (BNI scored P-5) and a missing clearly localized trigger point. The previous treatment with oral carbamazepine (200 mg, bid, po.) was ineffective. Thus, atypical TN diagnosis within the 3rd branch territory was established clinically. Notably, the symptomatic region was confined within the sensory territory of the lingual and inferior alveolar nerves, the subdivisions of the 3rd

Received September 13, 2023; Accepted February 5, 2024

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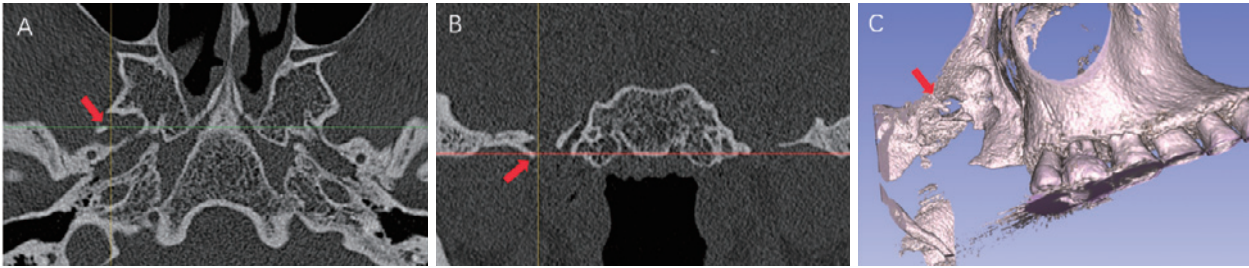


Fig. 1 HRCT and 3D reconstruction of the right foramen ovale region: (A) and (B) HRCT of the skull base showed an abnormality in the right foramen ovale region (red arrow). (C) 3D reconstruction demonstrated an osteophyte just underneath the posterolateral edge of the foramen ovale (red arrow).

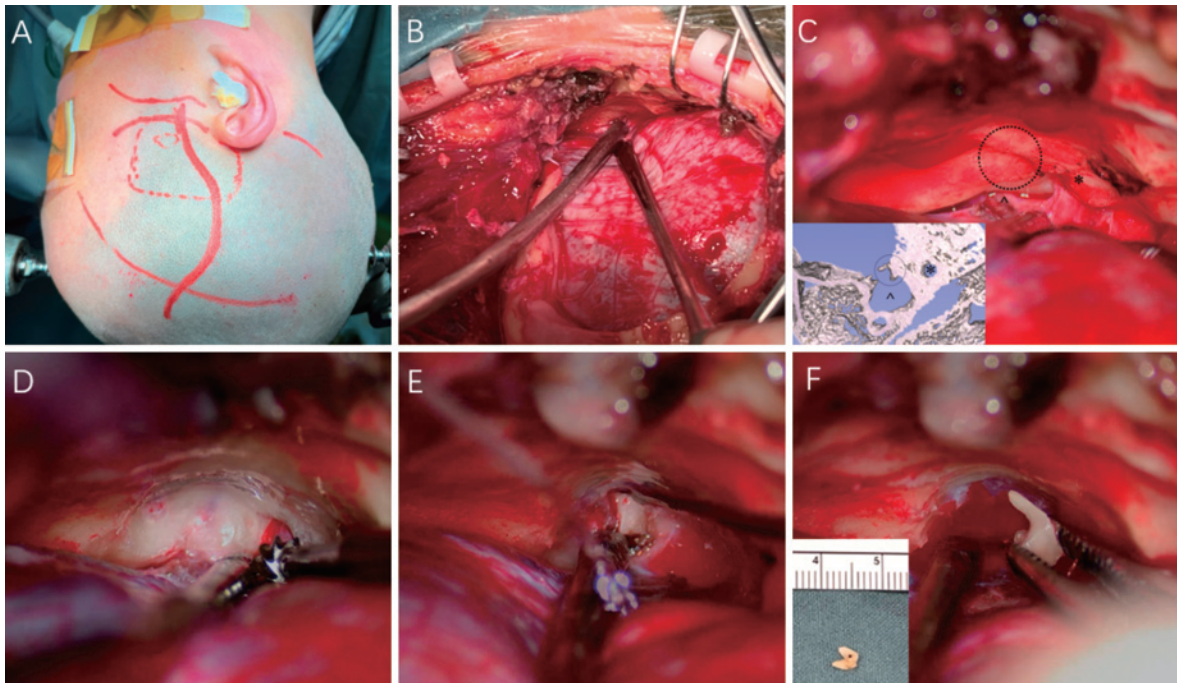


Fig. 2 Surgical procedure. (A) The skin incision for the middle fossa approach. (B) The level of the root of the zygomatic arch was exposed. (C) The foramen spinosum and the foramen ovale were exposed. (D) The osteophyte underneath the region of bone became loose. (E) The osteophyte from the surrounding membranous structure of the mandibular nerve branches was liberated. (F): The length of the osteophyte is about 5 mm, with a “shark tooth” appearance. (*, foramen spinosum; ^, foramen ovale; dotted circle, the root of osteophyte).

branch of the trigeminal nerve.

Magnetic resonance imaging (MRI) of the trigeminal nerve using thin-section axial fast imaging employing steady-state acquisition sequence and high-resolution computed tomography (HRCT) of the skull base were performed routinely. No obvious neurovascular conflict was detected in the MRI. However, an abnormality in the right foramen ovale region was observed in the axial and coronal HRCT images (Fig. 1A and B). Further three-dimensional (3D) reconstruction (3D slicer software, <http://www.slicer.org/>) of the skull base based on the HRCT data clearly demonstrated an osteophyte hanging on the extracranial surface of the greater wing of the sphenoid bone, just along the posterolateral edge of the foramen

ovale (Fig. 1C, arrows). It suggested that the lingual and inferior alveolar nerves might just be entrapped by this osteophyte as they are passing through the posterolateral aspect of the foramen ovale into the infratemporal fossa, thus resulting in the atypical and localized facial pain of this patient.

The patient underwent osteophyte removal through the middle fossa approach in a left lateral decubitus position (Fig. 2A). A lazy-S incision was made from 1 cm in front of the right tragus to the superior temporal line, and the temporalis muscle was peeled and retracted. A subtemporal craniotomy was performed down to the level of the root of the zygomatic arch (Fig. 2B). The temporal lobe was elevated extradurally until the level of the foramen

spinosum and ovale (Fig. 2C). The middle meningeal artery was coagulated and cut for further exposure of the posterolateral aspect of the foramen ovale, the root of the osteophyte to be removed. This bone region was drilled using a 3-mm burr until the osteophyte underneath became loose (Fig. 2D). A dissector was applied to free the osteophyte from the surrounding membranous structure of the mandibular nerve branches, and a 5-mm-long “shark tooth” was removed (Fig. 2E and F). As a result, the foramen ovale had also been widened posterolaterally. The dura was intact, and the bone flap was restored.

The pain disappeared completely and immediately after surgery without recurrence in the six-month follow-up period. The numbness was also mostly relieved (BNI scored N-4).

Discussion

Since Jannetta initially proposed the theory that TN was induced by the root entry zone compression of the nerve by the neighboring offending artery, the microvascular decompression (MVD) procedure has greatly contributed to TN⁴⁾ treatment, with a long-term rate of complete pain relief exceeding 80%.⁵⁾ However, a proportion of patients (about 28%-50%)⁶⁻⁸⁾ still present with pain attack intervals followed by continuous or near-continuous background and numbness (i.e., atypical TN), which would be associated with a high probability of no response to MVD, indicating various reasons of nonvascular factors. A study hypothesizes that persistent compression within a narrow foramen ovale could result in localized demyelination of the mandibular division, consequently increasing a REZ (root entry zone)-like region in the foramen ovale. Vessels traversing the foramen ovale may closely interact with the mandibular division within this constrained pathway.⁹⁾ It is suggested that the variation of foramen ovale may also cause TN by compressing the peripheral trigeminal nerve. Therefore, the etiology of TN cases where vascular compression is excluded by preoperative imaging evaluations must be explored.

Some studies have suggested that a narrow foramen ovale heavily influences the pathogenic mechanism of atypical TN based on the differences in the size of the foramen ovale between the painful and painless sides using anatomical and radiological data.^{3,10,11)} Besides the simple natural size, some bony structural variations can also narrow the foramen ovale. It has been well documented that the ossification of pterygospinous and pterygoalar ligaments underneath the foramen ovale may entrap the mandibular nerve branches and potentially result in atypical TN. Specifically, the pterygoalar ligament extends from the root of the lateral plate of the pterygoid process to the inferior surface of the greater wing, running beneath the foramen ovale to divide it into two parts.¹²⁾ Iwanaga et al.¹³⁾ further classified this ligament/bony bar into Type I, run-

ning between the buccal nerve and main trunk, and Type II, running medial to the inferior alveolar nerve and lateral to the lingual nerve. According to their description, the osteophyte in our case would probably be the posterior segment of a partially ossified pterygoalar ligament of Type I, attaching to the inferior surface of the greater wing, hanging along the posterolateral rim of the foramen ovale, and thus compressing the lingual nerve and the inferior alveolar nerve just medial to it, resulting in the atypical but confined symptoms of this patient. This speculation was further supported by the visualized radiological reconstruction and postoperative efficient outcome.

Transcutaneous procedures, such as percutaneous rhizotomy and balloon compression, are alternative surgical choices for TN treatment.^{14,15)} During these procedures, a puncture needle must pass through the foramen ovale. Thus, the bony variations around the foramen ovale, like the osteophyte in our case, could present an obstacle in these operations. Therefore, this case shows that patients with TN scheduled for transcutaneous procedures must undergo an exhaustive preoperative radiological evaluation to exclude any foramen ovale abnormalities.

In conclusion, we described a case of atypical TN caused by an osteophyte of foramen ovale in a middle-aged female patient whose symptom was completely relieved after removing this bony abnormality through the middle fossa approach. This case provides a new perspective for clinical diagnosis and treatment of patients with atypical TN without signs of neurovascular conflict in preoperative MRI. Lastly, the morphometric characteristics of the foramen ovale using HRCT with reconstructive technique must be recognized.

Haopeng Wang and Xuhui Wang have contributed equally.

Acknowledgments

We gratefully acknowledge the foundation of the National Natural Science Foundation of China (No. 82171360 and No. 81974186) and the Science and Technology Commission of Shanghai Municipality Project (No. 21Y21900500).

Ethics Statement

Informed written consent was obtained from the patient for publication of this case report and the accompanying images.

Conflicts of Interest Disclosure

There are no financial disclosures or conflicts of interest to report. The authors alone are responsible for the content and writing of this paper.

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