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Trigeminal Neuralgia from Acute Sphenoid Sinusitis: Consideration of Anatomical Sphenoid Sinus Variation – A Case Report

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Keywords
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Abstract
Although the etiology of classical trigeminal neuralgia is clearly understood to be neurovascular compression, the exact etiology of trigeminal neuralgia with continuous pain is often unknown. Mild sphenoid sinusitis is not usually considered to induce trigeminal neuralgia, especially when limited to the maxillary nerve. We report a rare case of trigeminal neuralgia of the maxillary nerve caused only by mild sphenoid sinusitis and discuss the significance of the anatomical structure and diagnostic procedures. A 45-year-old woman noticed a sudden onset of temporal pain followed by numbness on her right cheek. Her right gingiva also experienced sensory disturbance. The symptoms gradually subsided after the initial onset, but they persisted. She visited our hospital for further examinations and had no febrile episodes throughout the course. A tingling sensation and sensory disturbance were only identified in the maxillary nerve. No other neurological symptoms were noted. Magnetic resonance imaging revealed mild sphenoid sinusitis on the right side. The absence of the bony boundary between

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the sphenoid sinus and maxillary nerve was revealed using thin-sliced computed tomography (CT). The patient's symptoms were diagnosed as maxillary neuropathy caused by mild sinusitis. The bony defect around the maxillary nerve was considered to have affected development of the pathological process. Even mild sphenoid sinusitis can cause inflammation to spread to the maxillary nerve if no bony boundary exists between it and the sphenoid sinus. A coronal CT study is highly beneficial for clarifying the pathophysiological mechanism of trigeminal neuralgia limited to the maxillary nerve.

Introduction

Trigeminal neuralgia is categorized, based on the cause, into classical trigeminal neuralgia, secondary trigeminal neuralgia, and idiopathic trigeminal neuralgia by the International Classification of Headache Disorders [1]. To understand the pathophysiological mechanisms of trigeminal neuralgia and select an appropriate treatment, identifying the etiology is crucial. Classical trigeminal neuralgia is characterized by brief, recurrent, unilateral, electric shock-like pains limited to the distribution of one or more divisions of the trigeminal nerve. It is easily diagnosed and well-known to be induced by neurovascular compression of the trigeminal nerve. However, unilateral trigeminal neuralgia with concomitant continuous pain, formerly called atypical trigeminal neuralgia, is difficult to categorize and the cause of it is difficult to identify.

Here, the authors report a patient who presented with trigeminal neuralgia with concomitant continuous pain of unknown cause. To detect the etiology, we examined the patient using a coronal computed tomography (CT) scan of the paranasal sinuses. In the following sections, we discuss the significance of the treatment strategy in consideration of anatomical variation.

Case Report

A 45-year-old woman was on a flight and suffered from a sudden onset of right temporal pain for 30 min, followed by a tingling sensation on the right cheek. After she arrived at her destination, she experienced a throat blockage for approximately 20 min; therefore, she visited an ear, nose, and throat (ENT) doctor as well as a neurologist at another hospital. Neurological examination only revealed a tingling sensation in the area of the second branch of the right trigeminal nerve (the maxillary nerve). Otolaryngological examinations were unremarkable, except for percussion pain from the upper right central incisor to the first molar and gingival sensory disturbance in those regions (Fig. 1). The patient was examined using head magnetic resonance imaging (MRI), which detected inflammatory findings in the right ethmoid sinus and slight fluid retention in the right sphenoid sinus (Fig. 2). No pathological findings in or around the brainstem and cisternal portion of the trigeminal nerve were noticed. Under a diagnosis of idiopathic trigeminal neuralgia, the woman received conservative treatment. Because her symptoms persisted, the patient revisited the neurologist 1 month after the onset, and an additional MRI scan revealed chronic sinusitis in the right sphenoid sinus (Fig. 3).

Seven weeks after onset, the patient's symptoms continued, such as a tingling sensation from the right side of her nose to around her upper lip, and, therefore, she visited our hospital

for a further workup. During the entire period, she noticed no febrile episodes. Neurological examinations revealed only sensory disturbance on the second branch area of the right trigeminal nerve. Other cranial nerves were intact, and a blood examination revealed no abnormal findings. The CT examination indicated sphenoid sinusitis with a small amount of fluid retention. And the coronal thin-sliced bone CT revealed the absence of the bony boundary between the sizeable sphenoid sinus and maxillary nerve (Fig. 4).

According to these results, it was conceivable that her symptoms were caused by inflammation from sphenoid sinusitis that had spread to the maxillary nerve. The bone defects surrounding the maxillary nerve contributed to the spread of inflammation to the nerve. Because the symptoms were improving, the patient was conservatively followed up without any medication, and she was entirely symptom free 5 months after onset.

Discussion/Conclusion

In summary, the patient noticed right facial pain while on a flight, which was restricted to the maxillary nerve of the right trigeminal nerve. The symptoms persisted and were treated as idiopathic trigeminal neuralgia. In consideration of the pathophysiological mechanisms of the symptomatic progression of this patient, the following two mechanisms were postulated: barosinusitis and the anatomical variation of the sphenoid sinus. These are discussed in the following paragraphs.

Barosinusitis

Barosinusitis is an acute form of paranasal sinusitis. It develops in a short period because of changes in air pressure, such as during skydiving or boarding an airplane [2]. Changes in air pressure cause disturbances of the pressure balance in paranasal sinuses, resulting in sinus mucosal damage. Acute sinusitis, including barosinusitis, is usually limited to the sinus mucosa without bony destruction of the paranasal sinuses [3].

Anatomical Variation of the Sphenoid Sinus

The symptomatic significance of this patient was that her facial pain was restricted to the maxillary nerve, but was not extended to any other branch of the trigeminal nerve. To clarify this mechanism, we examined her CT scans. In the coronal bone images from the thin-slice study, no bony boundary between the maxillary nerve and sphenoid sinus was observed. A precise examination of the CT images revealed no other chronic inflammatory changes, such as mucosal thickening or any other bony changes or destruction. It has been reported that inflammation associated with acute sinusitis is usually limited to the mucosa and does not destroy the bony structure [3]. The symptoms of this patient had an acute onset, and no chronic changes were observed in the CT scans. According to this evidence, the pathological process of this patient was caused by acute sinusitis. The absence of bone between the maxillary nerve and sphenoid sinus was considered to be a long-lasting result, such as a congenital defect, but not to have been caused by bony destruction from a chronic inflammatory process.

Nerves without surrounding bone tissue are vulnerable to inflammation occurring in nearby tissues. In these situations, congenital bone defects surrounding the maxillary nerve can cause neurological symptoms immediately after sphenoid sinusitis.

The structure around the maxillary nerve varies according to individual sphenoid sinus development. Over 50% of sphenoid sinuses develop along the greater wing of the sphenoid

bone, beyond the foramen rotundum [4]. As the sphenoid sinus develops widely along the greater wing, the bone covering the nerve becomes thinner, and the maxillary nerve becomes more exposed in the sphenoid sinus. In extreme cases, a part of the maxillary nerve is covered exclusively by the mucosa in the sphenoid sinus [5].

The coronal thin-sliced CT scan is exceptionally beneficial for examining the bony structure around the maxillary nerve as well as the relationship between the nerve and sinus. Notably, images of the peripheral trigeminal nerve around the sinus can be obtained using this method.

Several reports have been published regarding maxillary nerve neuropathy caused by severe chronic fungal infections [6–8]. Fungal infection in the paranasal sinuses is a long-standing pathological process, which causes bony destruction in those sinuses, resulting in neuropathy around them. However, no reports have been published on maxillary nerve neuropathy in association with congenital bony absence caused by acute sinusitis, such as barosinusitis. To the best of our knowledge, this is the first report to describe such a pathology in consideration of the anatomical importance of the paranasal sinuses.

In conclusion, the pathophysiological mechanisms of trigeminal neuralgia without neurovascular compression findings in the trigeminal nerve have been overlooked and treated as idiopathic trigeminal neuralgia. However, understanding such mechanisms by clarifying the exact causes is critical because it can lead to the selection of appropriate treatments. We applied a coronal thin-sliced CT study to our patient with trigeminal neuralgia, and it identified a bony absence between the maxillary nerve and sphenoid sinus. Such an anatomical variation significantly contributes to the understanding of the pathological process. Even in mild sphenoid sinusitis, inflammation can quickly spread to the maxillary nerve and induce solitary neuropathy.

Statement of Ethics

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

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Conceptualization, K.N.; acquisition of data, K.N., R.Y., and I.O.; analysis and/or interpretation of data, K.N., R.Y., and I.O.; drafting the manuscript, K.N. and R.H.; revising the

manuscript critically for important intellectual content, S.N. and R.H.; supervision, R.H. and S.N.; project administration, K.N.

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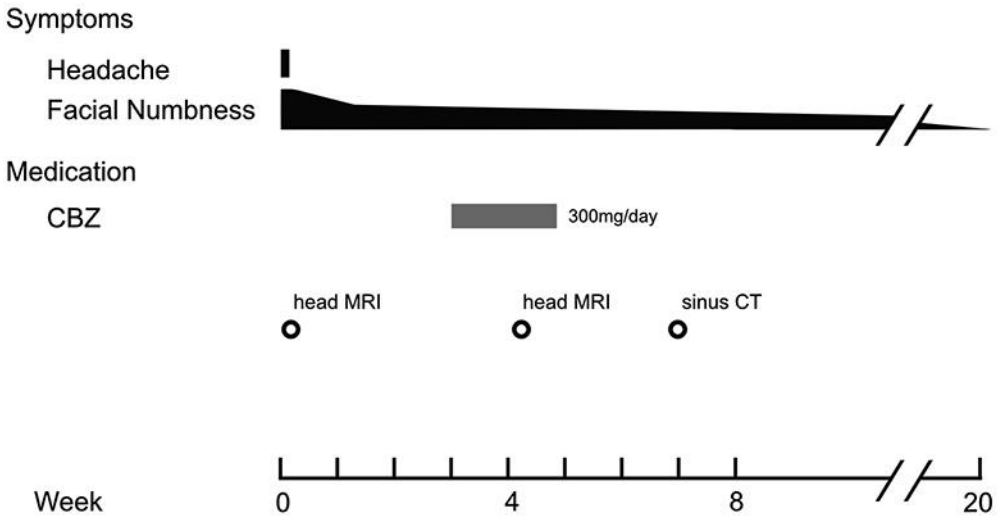

Fig. 1. Timeline. This timeline shows the patient's symptoms, imaging studies, and treatment.

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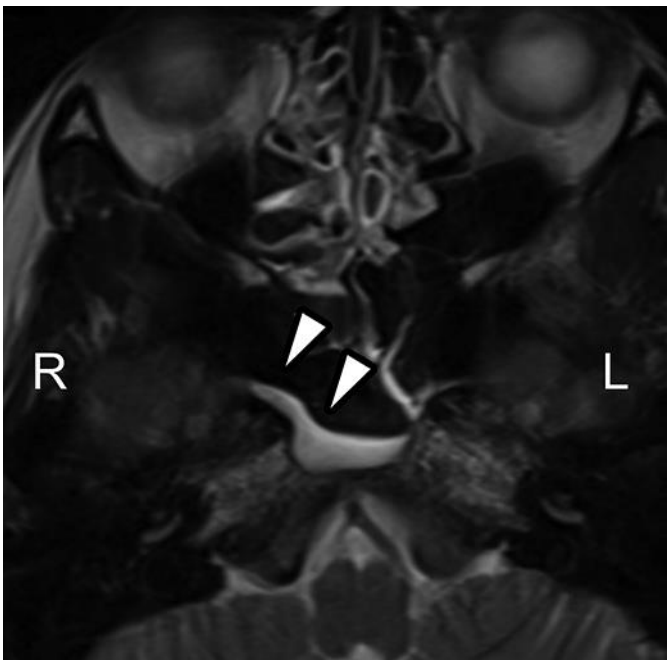

Fig. 2. Axial T2-weighted head MR image on the day following initial onset. This MR image reveals acute sphenoid sinusitis, including fluid (arrowheads).

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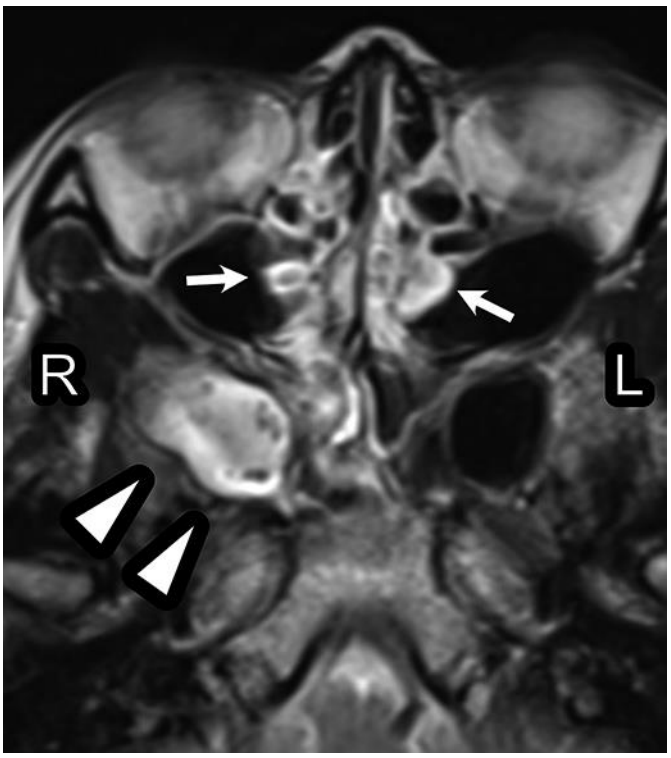

Fig. 3. Axial T2-weighted follow-up MR image 1 month after onset. This MR image reveals chronic sinusitis of the right greater wing, which is filled with mucosa and fluid (arrowheads), and the ethmoid sinus, which has thick mucosa (arrows).

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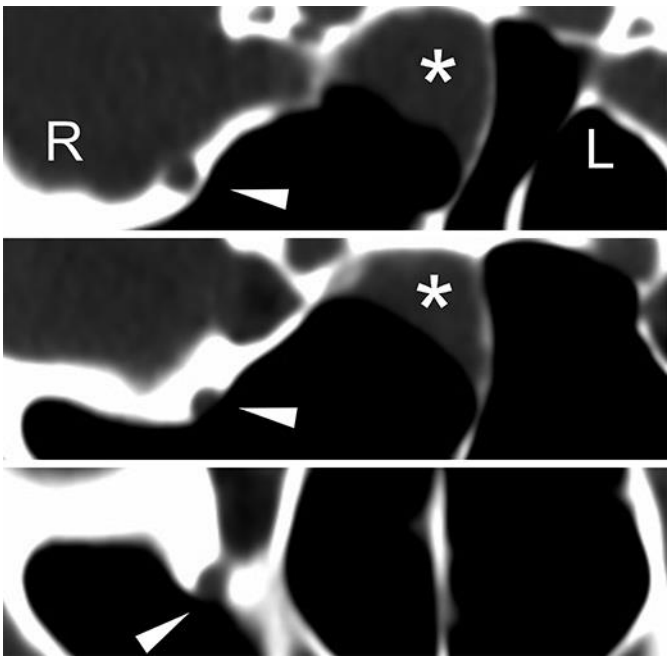

Fig. 4. Coronal cross-sectional CT images 7 weeks after onset. These coronal sinus CT images line the dorsal to the ventral side toward the bottom. They reveal a bone defect around the maxillary nerve facing the sphenoid sinus (arrowhead) as well as sphenoid sinusitis with fluid retention (asterisk).