



Referred Trigeminal Pain from Greater Occipital Nerve Entrapment: Importance of Accurate Greater Occipital Nerve Decompression

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OPEN ACCESS

Received: January 20, 2022

Revised: February 10, 2022

Accepted: February 11, 2022

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Trigemincervical complex (TCC) sensitization by chronic entrapment of the greater occipital nerve (GON) often causes difficult-to-understand referred trigeminal facial pain along with occipital neuralgia. This report emphasizes the importance of accurate diagnosis and GON decompression by describing a case of a 51-year-old female patient with referred trigeminal pain from occipital neuralgia misdiagnosed as chronic migraine. The patient underwent incomplete decompression surgery. She presented with a relentless stinging and stabbing pain in her left hemiface and temporo-occipital area for 8 years. The pain occurred suddenly without improvement after long-term medical and injection treatment. She was diagnosed with chronic migraine and underwent so-called "migraine surgery" three years ago, but to no avail. Incomplete decompression of the bilateral GONs within the trapezial tunnel was confirmed under a microscopic oblique-paramedian approach. After accurate decompression, the chronic stabbing pain in the left facial and temporo-occipital region disappeared. Only intermittent numbness was felt. It is important to recognize occipital neuralgia caused by chronic GON entrapment and referred facial pain caused by TCC sensitization. Referred pain still often seems to be misunderstood as various other diseases. Direct decompression of the trapezial tunnel using a neurosurgical microscopic approach based on an accurate assessment of anatomical considerations may be important for GON decompression.

Keywords: Headache; Neuralgia; Pain; referred; Trigeminal nerve

INTRODUCTION

Occipital neuralgia is a unilateral or bilateral paroxysmal, shooting, or stabbing pain in the posterior part of the scalp, in the distribution of the greater, lesser occipital nerves⁷. In

the absence of structural lesions and metabolic causes, occipital neuralgia has been mostly considered to have an idiopathic etiology²¹. However, greater occipital nerve (GON) entrapment in the trapezial canal has been found to be the main cause of occipital neuralgia by anatomical studies and

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decompression surgeries of the GON for occipital neuralgia and occipital-based headache^{2-4,14,15,20}.

Diagnosis of occipital neuralgia is not difficult if the typical pattern of neuralgic stabbing pain is present in the occipital area. However, if the pattern of pain radiating to the vertex and temporal area in occipital neuralgia, the terminal distribution of the GON, is not recognized, the involvement of the GON might not be recognized^{12,18}. In addition, the GON, major C2 afferent, communicates directly with the trigeminal cervical complex (TCC) located in the C2 dorsal horn¹. The TCC is where the convergence of nociceptive afferent input of the trigeminal system and high cervical (C1-C3) area occurs¹. Therefore, chronic GON entrapment can cause occipital neuralgia and headache. At the same time, referred pain in the facial trigeminal area can occur due to TCC sensitization. Failure to recognize referred facial trigeminal pain due to TCC sensitization is highly likely to lead to a diagnosis of craniofacial pain, which is difficult to understand as both occipital neuralgia and facial pain of unknown origin coexist^{9,10,16-19}. As a result, chronic GON entrapment patients may receive multiple diagnoses in different departments, resulting in long-term ineffective and unnecessary treatment¹¹.

We would like to introduce a case where typical occipital neuralgia accompanied by referred trigeminal pain was eventually diagnosed as chronic migraine and so-called migraine surgery was performed. In this case, there was no improvement in symptoms due to incomplete GON decompression by a proximal approach. The importance of accurate decompression through a microscopic neurosurgical approach in GON decompression is emphasized through this case report.

CASE REPORT

A 51-year-old female patient presented with a left occipital headache and facial pain that had persisted for eight years. Eight years earlier, dull pain in the back of the neck lasted about 10 days. Pain suddenly appeared in the left temporo-occipital area, around the ear, and on the left face (Fig. 1A). There were no specific antecedent causes including trauma. The pain was characteristically stinging and stabbing. An electrical tingling constantly occurred around the left eye, around the cheek, and under the chin. At the same time, she developed a burning sensation in her left auricle

and a stabbing pain within her left ear. Her left facial and occipital pain were accompanied by a continuous dull pain in the neck and left shoulder. The intensity of the pain was about 6-7/10 on the numerical rating scale (NRS-11). The pain persisted while she was awake.

During the first month of the pain, her pain was very severe. It did not get better despite taking multiple medications from several hospitals. After a month of excruciating pain, the intensity of the pain gradually improved. However, the pain lasting 2 to 3 months continued to recur at 3 to 4 times a year. The stabbing pain lasted as little as 30 min to as long as 4 to 5 hr. If her pain persisted very badly, the muscles of her left eyelid and cheeks trembled and she felt a tingling sensation in her left tongue. The pain in the left temporo-occipital

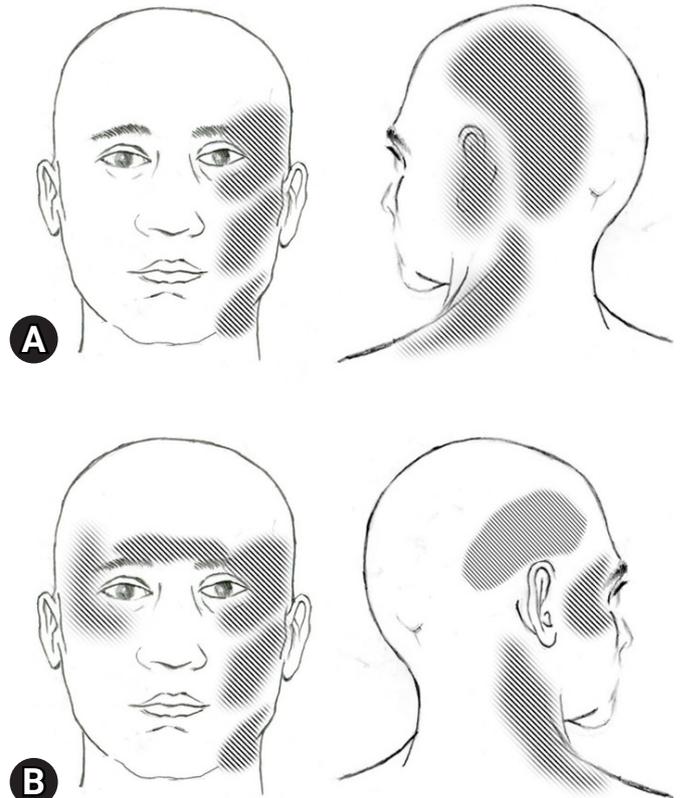


Fig. 1. Distribution of referred trigeminal pain from occipital neuralgia. (A) Drawing showing the distribution of stabbing pain that occurred suddenly 8 years ago (hatched area). It occurred not only on the left hemiface, including around the orbit, cheeks, under the chin, and around the ears, but also in the left temporal and occipital region. (B) A drawing showing the distribution of newly developed stabbing pain (hatched area) in the right temporal region and around the eye.

area persisted even in the absence of facial pain attacks. Daily activities, neck posture, and movement were not associated with pain intensity. She was treated by several doctors from different departments, including neurology, dentistry, ophthalmology, otolaryngology, and pain clinics. However, the pain was not controlled. At one year after the occurrence of severe occipital and facial pain, stinging pains appeared in the right temporal region, vertex, and around the right eye as if on the left (Fig. 1B). Pain intensity on her right side was less than on her left side. Over the past two months, her left occipital and facial pain attacks were so severe that it made her daily activities impossible. Thus, she visited the author's outpatient clinic.

Neurological examination revealed no objective abnormal findings of the cranial nerve. There was no apparent sensory abnormality in the left trigeminal area or no limitation of neck movement. No tenderness or allodynia was observed in the occipital area. Magnetic resonance imaging of the brain and cervical spine taken at other hospitals did not show any abnormal findings. No abnormal findings were found in laboratory examination. A physical examination revealed a vertical midline incision scar of about 5 cm in the suboccipital area. She said that she had been diagnosed with chronic migraine at a plastic surgery clinic three years ago. Thus, she had undergone a so-called migraine surgery (Fig. 2A). Although the pain in the left facial and occipital area improved for two months after the migraine surgery, existing symptoms recurred with the same intensity at two months after the surgery. After that, there was no other way to control her symptoms. She said that she was getting botulinum toxin injections with the diagnosis of chronic migraine every three months. She said that botulinum toxin injections had no effect on the frequency or intensity of her pain. She said that the stabbing pain in the left occipital area continued to occur even during the interview at the outpatient clinic.

Considering that her left temporo-occipital pain coexisted on the left hemiface and ear and the occipital pain had a stabbing nature, occipital neuralgia caused by entrapment of the GON was thought to be the cause of a referred trigeminal facial pain. Block of the left GON (ONB) with 2 mL of 2% lidocaine significantly improved her left occipital and facial pain for about 3 hr. The same temporary pain reduction was confirmed through two ONBs. GON decompression was explained to her.

GON decompression was performed bilaterally using an

oblique paramedian approach under general anesthesia (Fig. 2A)^{12,18}. As expected, decompression of the GON within the trapezial canal through the so-called migraine surgery performed three years ago was insufficient according to the authors' surgical findings (Fig. 2B). The left GON was not decompressed at all. The right GON was decompressed only in the proximal path before its entrance to the trapezial canal, while the portion of the GON within the trapezial canal was not touched (Fig. 2B). Severe GON entrapment by the aponeurotic edge of the trapezius muscle was confirmed bilaterally. Circumferential decompression of the GON was performed in the trapezial canal and its proximal course to the semispinalis capitis muscle under microscopic vision (Fig. 2C). The postoperative course was uneventful. She was discharged on the second day of surgery.

Three weeks after the surgery, she reported that existing left temporo-occipital, periorbital, and hemifacial stabbing pain still occurred but decreased to 30% of the preoperative intensity. Her right temporal and periorbital pain were no longer felt. The slight numbness in bilateral occipital area after surgery disappeared one month after surgery. Two months after the surgery, she assessed that the stinging pain in the left occipital, periorbital, and face was mild. It was no longer severe. She reported that a stabbing pain within the left ear canal was felt intermittently. Her periodic attacks of severe left-sided headaches and facial pain no longer occurred. At the 6-month follow-up visit, she assessed that the existing pain had improved by 80% by GON decompression. She reported no restrictions in daily life due to her pain. She no longer had the stabbing pain. However, she wanted to continue her medication (gabapentin 300 mg a day) because of intermittent numbness in her left face. Nine months after the surgery, she reported that there were no more pain attacks in the left occipital, periorbital, or face. She was taking the pills when she felt a mild tingling in her face that lasted about 30 min, occurring once or twice a week. She rated her pain intensity as 2 on the NRS-11.

DISCUSSION

Simultaneous occurrence of hemifacial pain and occipital pain: Is this chronic migraine?

The pain in the left temporo-occipital and hemiface shown in the present case was diagnosed with various diseases during eight years. She was diagnosed with trigeminal

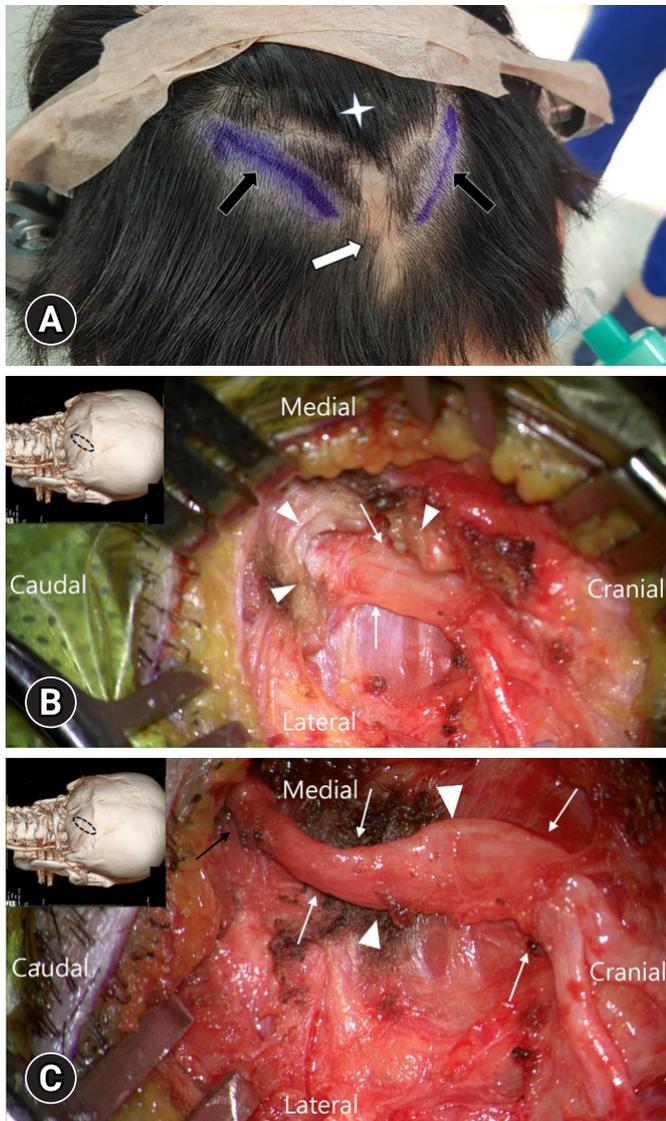


Fig. 2. Intraoperative findings during decompression of the greater occipital nerve (GON). (A) A photograph showing the paramedian oblique incisions (black arrows) of the distal approach to the trapezial tunnel and the existing midline incision located in the suboccipital area (white arrow). White stars indicate the positions of the inion. (B) Intraoperative photograph showing entrapment of the right GON (white arrows) via an aponeurotic fibrous attachment of the trapezius (white arrowheads) at the superior nuchal line. The aponeurotic edge of the trapezial canal was partially divided. (C) Intraoperative photograph showing complete decompression of the right GON (white arrows) by dividing the trapezial tunnel. The most heavily compressed portion of the right GON pathway (white arrowheads) showed reddish discoloration and post-stenotic dilation. The proximal decompression of the right GON was made at the point where the GON pierced the semispinalis capitis muscle (black arrow). Its proximal and distal courses were completely addressed.

neuralgia due to pain in the unilateral face. The fact that intermittent stabbing pain was not evoked by facial movements indicated that her pain did not meet the diagnostic criteria for trigeminal neuralgia⁷⁾. Moreover, trigeminal neuralgia cannot account for the pain in her ear canal and temporo-occipital area. Finally, after years of treatment with chronic migraine, she underwent a so-called migraine surgery. Temporary improvement of symptoms in the first two months after the first operation indicated the possibility of chronic migraine.

Chronic migraine is defined as a headache occurring 15 or more days/month for three months. It should have features of migraine headache at least 8 days/month⁷⁾. In the present case, unilaterality, intensity (moderate to severe), the duration, and frequency of headache meet the criteria for chronic migraine⁷⁾. However, it did not have a pulsating quality. Instead, it was stabbing in nature. It was not induced by routine physical activities. It was not accompanied by symptoms such as photophobia or nausea. Thus, it did not meet the criteria for migraine headache⁷⁾. In addition, the ipsilateral facial pain accompanying the headache could not be explained by migraine. The possibility of trigeminal autonomic cephalalgia can be excluded due to the absence of autonomic symptoms.

In fact, the intermittent stinging and stabbing pain in the temporo-occipital area seen in the current case is a typical feature of occipital neuralgia¹⁾. Occipital neuralgia is defined as a unilateral or bilateral paroxysmal, shooting, or stabbing pain in the posterior part of the scalp, in the distribution of the greater, lesser occipital nerves⁷⁾. If stabbing pain persists, the possibility of occipital neuralgia is very high. In addition, if the location of the pain coincides with the area of distribution of the GON, it is not difficult to diagnose occipital neuralgia. The distribution of the GON is not confined to the occipital area^{12,18)}. Pain associated with GON entrapment can radiate not only to the occipital area but also to the vertex, temporal, retroauricular and subauricular areas, angle of the jaw, the lateral and posterior neck^{12,18)}. If the typical pain distribution of the GON is not recognized, it could be simply judged that the headache is located in the temporal or occipital area. In the current classification of headaches, headaches including migraines are not classified according to the location of the pain⁷⁾. If the pain distribution of the GON entrapment is recognized, it is easy to know that the stabbing pain of typical occipital neuralgia is located in the

area of the GON.

Interestingly, this pain in the present case did not improve for many years despite multiple nerve blocks. It was not diagnosed with occipital neuralgia. Typical occipital neuralgia is characterized by a response to ONB that is included in the diagnostic criteria⁷. On the other hand, in the two ONBs performed by the authors of the present report, temporary improvement of occipital pain including the left hemiface was confirmed easily. In fact, she remembered that whenever she had a nerve block, she always had injections in various areas around her neck, including the back of her head. It seems that it is difficult to confirm the existence of occipital neuralgia and GON entrapment only by repeatedly blocking multiple sites for temporary symptom relief without analyzing the pain pattern of the occipital-based GON distribution. Diagnostic nerve block should be performed selectively with the target nerve and specific disease.

Referred trigeminal pain from GON entrapment

We reported improvement of pain in the left hemiface and temporo-occipital area lasting eight years through GON decompression in the present case. Although referral of the pain of occipital neuralgia to the fronto-orbital (V1) area through sensitization of the trigeminocervical complex (TCC) in the trigeminal spinal nucleus located in the C2 dorsal horn is well known, there are few reports about its clinical presentations^{9-11,16,17,19}. Referred facial and occipital pain is caused by convergence of nociceptive inputs of trigeminal and occipital regions in the TCC¹. This pain referral to the facial trigeminal distribution can occur not only in the V1 region, but also in V2 and V3 regions. It can even cause hemifacial sensory changes^{9-11,16,17,19}. In addition, referred trigeminal pain may occur much earlier than the onset of occipital neuralgia¹⁶. Considering the simultaneous presence of occipital and unexplained facial pain and the possibility of delayed occurrence of occipital pain than facial pain, it is thought that the referred trigeminal pain due to sensitization of TCC exhibits various clinical manifestations^{9-12,16-19}.

The GON originates from the medial branch of the dorsal rami of the second cervical nerve root²⁰. It ascends through the semispinalis muscle and runs rostrally before emerging into the scalp by piercing the aponeurotic fibrous sling between the trapezius and the sternocleidomastoidus near their occipital attachment to the superior nuchal

line²⁰. This aperture (trapezial tunnel) is a common site of entrapment of the GON^{2-4,12,14,15,18,20}. After piercing the aponeurotic sling of the trapezial tunnel, the GON widens toward its course to the periphery². This finding (widening of the nerve) makes it susceptible to entrapment especially in a firm trapezial aponeurosis^{2,15,20}. The GON constitutes the main sensory afferents through the C2 root¹. This afferent input is transmitted directly to the C2 dorsal horn^{12,14,18}. The chronic, continuous, and noxious afferent input of GON entrapment appears to be associated with sensitization and hypersensitivity of second-order neurons in the TCC¹. If insight into the pattern of referred trigeminal pain in the face due to TCC sensitization caused by chronic GON entrapment is lacking, patients are mostly misdiagnosed as cervicogenic headache, tension headache, chronic migraine, atypical facial pain, and even trigeminal neuralgia as in the present case¹¹.

Excluding structural and metabolic causes, occipital neuralgia has been considered idiopathic in etiology²¹. However, several anatomical studies have suggested that GON entrapment is the main cause of occipital neuralgia^{2-4,12,14,15,18,20}.

Decompression of the GON at the trapezial tunnel can result in excellent pain relief in idiopathic occipital neuralgia^{8,13}. It was effective not only for occipital neuralgia but also for the referred trigeminal pain from occipital neuralgia and GON entrapment^{3,4,9-12,14,16-19}.

Accurate decompression of the GON

The approach to GON decompression can be categorized into a proximal type to first address the proximal course of the GON within the semispinalis capitis muscle¹⁸ and a distal type to address the distal GON course within the trapezial canal, which the authors conducted. In the present case, the left trapezial tunnel was not found to be decompressed^{9-11,16,17,19}. Only the proximal portion of the main trapezial tunnel was decompressed on the right side prior to surgery. Complete relapse of the patient's symptoms following the so-called migraine surgery and almost complete improvement after a distal approach indicate that complete decompression of the GON in the trapezial tunnel is important. However, the authors' distal approach cannot be evaluated as superior because there has been no systematic study comparing proximal and distal approaches.

In the first few cases of GON decompression, we used the proximal approach suggested by some plastic surgeons in

the United States^{3-6,14}). However, it was inconvenient for us to extend the incision distally to decompress the GON in the trapezial tunnel¹⁸). Therefore, using an incision directly above the trapezial tunnel, we traced branches of the GON to decompress it in the tunnel and followed it down to further decompress the underlying semispinalis capitis muscle^{9-12,16-19}). Since the GON exhibits a superolateral course from the midline, oblique paramedian incision can be performed^{2-4,12,14,15,20}). Because most entrapment is caused by the trapezial aponeurotic edge in the trapezial tunnel, the distal approach provides direct access to the main entrapment point¹⁸). Using a microscopic field of view, damage to GON branches can be prevented and the proximal course of the decompressed GON at the trapezial tunnel can be traced to decompress the portion that penetrates the semispinalis capitis muscle¹⁸). Therefore, we recommend a direct decompression of the trapezial tunnel through an oblique paramedian approach using microscopy for neurosurgeons.

CONCLUSION

Except for structural abnormalities of the craniovertebral junction and metabolic causes, most cases of occipital neuralgia are caused by chronic entrapment of the GON. The GON is entrapped in the trapezial tunnel, where the tendinous aponeurotic edge of the trapezius muscle attaches to the superior nuchal line. With temporal and vertex regions of the GON distribution in mind, if there is a stabbing pain, occipital neuralgia should be considered first. If pain in occipital neuralgia is also present on the face, referred trigeminal pain should be considered. A direct approach to the trapezial tunnel using a neurosurgical microscope is recommended for an accurate decompression of the GON.

CONFLICTS OF INTEREST

No potential conflict of interest relevant to this article was reported.

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