Neurology Asia 2023; 28(2) : 449 – 453

Can exostoses of the external occipital protuberance and superior nuchal lines cause headache? A report of two cases

Sinan BAHADIR *MD*

Department of Neurosurgery, Amasya University Faculty of Medicine, Amasya, Turkey

Abstract

Compression of the occipital nerves can cause occipital headache. Several potential compression points have been identified along their course. One of these points is the hitherto undescribed instance of exostoses of anatomic structures of the occipital bone. A 34-year-old man with a prominent external occipital tuberance and superior nuchal lines complained of headache in the area of distribution of the greater occipital nerve with tenderness near the bony protrusion on palpation. A 44-year old woman experienced stabbing pain in the lateral orbital region with pressure on the ossified insertion of the ipsilateral trapezius muscle near the external occipital protuberance. Local nerve block achieved temporary relief of pain. Exostosis of the external occipital protuberance and superior nuchal lines may impinge on the greater and third occipital nerves. The characteristics of the pain in our cases differ in some aspects from those of occipital neuralgia.

Keywords: External occipital protuberance; greater occipital nerve; occipital headache; occipital neuralgia; third occipital nerve

INTRODUCTION

Compression, injury, or irritation of the greater and third occipital nerves (GON and TON, respectively) at vulnerable sites, such as the facet joints and muscle piercing points, have been described as causes of occipital neuralgia.[1,2]

Singh has suggested that due to the anatomical proximity of the GON and the TON to the external occipital protuberance (EOP) and superior nuchal lines, exostoses of these structures may cause occipital headache.[3]

In this study, we report two cases with headache associated with exostoses of these anatomical structures.

CASE REPORT

Patient 1

A 34-year-old man was admitted for throbbing headaches in his left occipital region from which he had suffered for almost a decade. The patient had been experiencing these headaches 3-5 times a year, each lasting 3-4 days. He was vague regarding whether the pain was intermittent or uninterrupted during these periods. The pain was to the left of the EOP and extended to the vertex on the same side. He had visited various health care professionals and had been given analgesics which provided mild relief. Previously performed cranial magnetic resonance imaging (MRI) was normal.

His physical examination revealed an overtly prominent EOP which was also visualized by plain radiographs. He had tenderness to the left of the EOP. Cranial computerized tomography (CT) revealed exostosis of the EOP and prominent superior nuchal lines extending to both sides (40 mm on the left, 25mm on the right) (Figure 1). Since compression of the GON was suspected, the patient was offered a local nerve block, which he rejected. He was prescribed oral gabapentin and advised follow-up, but he did not return again.

Patient 2

A 44-year-old woman with a 15-year history of migraine was admitted because of a different type of headache starting a year prior. She experienced

Address correspondence to: Dr. Sinan BAHADIR, Amasya Üniversitesi Tıp Fakültesi Sabuncuoğlu Şerefeddin Eğitim ve Araştırma Hastanesi, Beyin ve Sinir Cerrahisi Anabilim Dalı Amasya, Turkey. Tel: +90 532 646 65 72, E-mail: sinanbahadir@windowslive.com

Date of Submission: 12 May 2022; Date of Acceptance: 20 February 2023

https://doi.org/10.54029/2023jwm

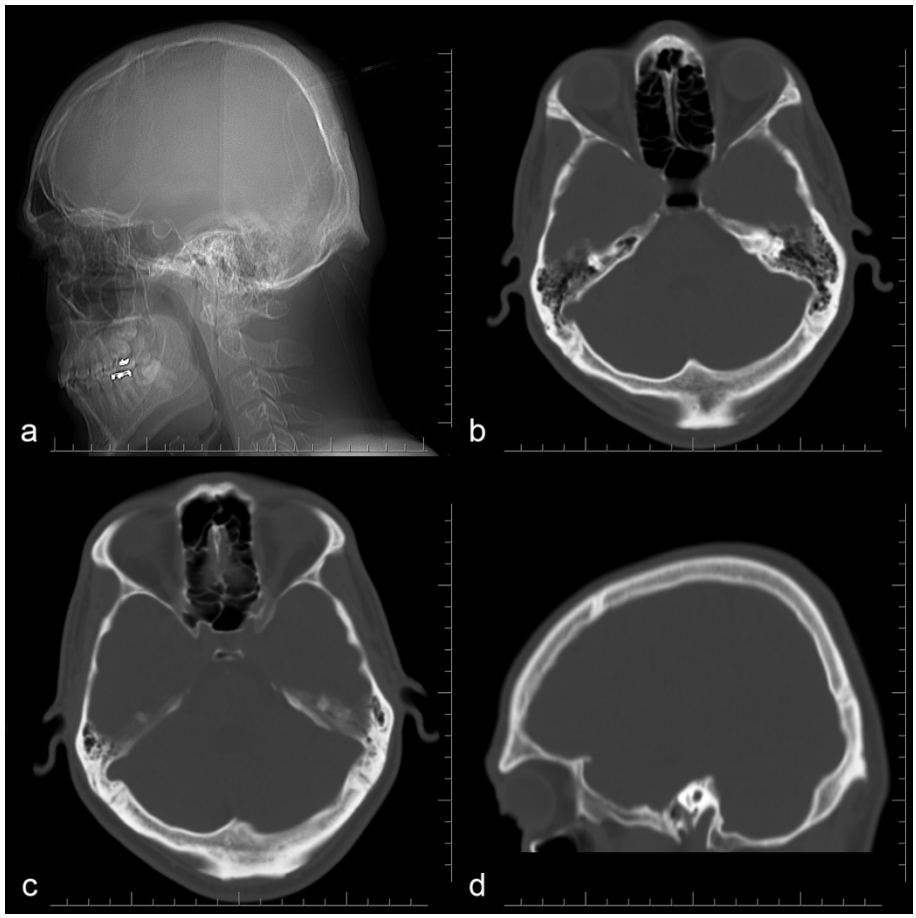

Figure 1. a) Lateral CT scenogram shows the exostosis of EOP. b) Axial CT scan shows EOP protrusion of 15 mm. c) Axial CT scan demonstrates the prominent superior nuchal lines on both sides. d) Sagittal reconstruction of the CT scan shows the left superior nuchal line still prominent 35 mm from the midline before diminishing in size at 40mm.

intermittent sharp pain at the lateral aspect of her left eye on leaning the back of her head against something. She then noticed a protrusion at the back of her head and discovered that she was able to replicate the pain by pressing on the protrusion. She had a past history of diabetes mellitus and no record of any head trauma.

Physical examination found tenderness on the left side of the EOP, and mechanical compression reproduced the symptoms. MRI ruled out an intracranial pathology. CT scan revealed a calcification continuous with the EOP, with a thin hypodense zone in between, suggesting calcification of the trapezius muscle aponeurosis (Figure 2). An anesthetic injection resulted in relief for a week. She was then referred to a neurology outpatient clinic for further medical treatment.

DISCUSSION

The International Headache Society (IHS) describes over 180 types of headaches in their International Classification of Headache Disorders, Third Edition (ICHD-3).[1] The headache syndromes attributed to disorders of the cranium are classified under Section 11, and those that involve cranial nerves and upper cervical roots through occipital nerves are classified under Section 13.[1]

The cases we present had headaches attributable to anatomical variants in the EOP, superior nuchal lines, and adjacent neurovascular structures as supported by local tenderness (both patients), reproduction of the symptoms by pressure (Patient 2), and temporary relief by a local anesthetic agent (Patient 2). The lack of cervical-related signs and symptoms as well as the presence of tenderness around the EOP rules out

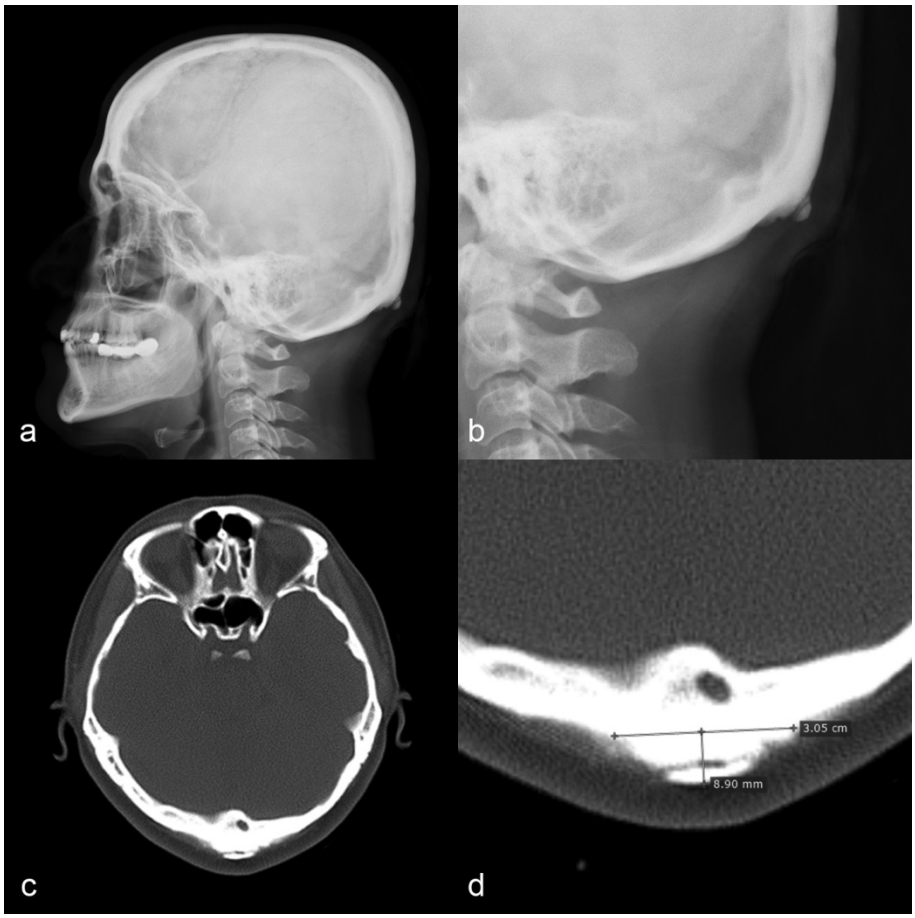

Figure 2. a) Lateral plain radiograph shows the calcification at the region of the EOP. b) An enlarged image shows the continuous contours of the EOP-calcification complex separated by a thin hypodense zone. c) Axial CT section at the level of EOP shows the bony tubercle. d) The thin hypodense zone and the dimensions of the extrusion can be seen in the enlarged image.

cervicogenic headache. The two clinically relevant structures in close proximity to the EOP and superior nuchal lines are the GON and the TON.

The GON originates from the C2 dorsal ramus. It can be compressed in-between the atlas and axis, the inferior capitis oblique and semispinalis capitis muscles, and in the course of its passage through the tendinous aponeurosis of the trapezius muscles.[2,4-8] Compression of the GON by the occipital artery has also been reported.[8,9] The GON is located at a mean distance of 4 cm (range 3.5-6.5 cm) lateral to the EOP.[8]

The TON originates from the third occipital nerve and curves around the C2-3 facet joint.[2] It is found 0-4 mm (mean 3 mm) lateral to the EOP and sends a small communicating branch to the GON just below the EOP.[8] The most heavily implicated compression point for TON is the C2-3 facet joint.[10]

The EOP and superior nuchal lines act as origin points for the trapezius muscle. Recently, Singh reported a case with a prominent exostosis arising from the EOP and superior nuchal lines.[3] He proposed that calcification/ossification of the muscle might have occurred due to microtrauma, activation of underlying osteocytes following the elevation of periosteum, deposition of calcium due to a metabolic disorder, or local inflammation of the bone from past injury to nearby tissues.[3] He suggested that the GON might be irritated due to inflammation and spasm of the trapezius muscle by impingement of the bony tubercle.[3] He also suggested that the TON might be compressed by the tubercle due to its close proximity to the EOP.[3] Another study by Shahar *et al*. found the prevalence of exostosis of EOP in 33% of cases, showing the widespread prevalence of this entity.[11] Though painful exostosis of EOP has been documented in young adults, these cases presented with local pain rather than neuralgia-like headache.[12]

Table 1: Diagnostic criteria of occipital neuralgia (ICHD-3) and respective characteristics of headache in the presented cases

Occipital Neuralgia diagnostic criteria	Patient 1	Patient 2
A. Unilateral or bilateral pain in the distribution(s) of the greater, lesser and/or third occipital nerves and fulfilling criteria B–D	Does not fulfill criteria C and D	Does not fulfill criteria C
B. Pain has at least two of the following three characteristics:	+	+
1. recurring in paroxysmal attacks lasting from a few seconds to minutes	+	with pressure
2. severe in intensity	+	+
3. shooting, stabbing or sharp in quality	throbbing	+
C. Pain is associated with both of the following:	-	-
1. dysaesthesia and/or allodynia apparent during innocuous stimulation of the scalp and/or hair	-	-
2. either or both of the following:	+	+
a) tenderness over the affected nerve branches	+	+
b) trigger points at the emergence of the greater occipital nerve or in the distribution of C2	-	-
D. Pain is eased temporarily by local anaesthetic block of the affected nerve(s)	not tested	+
E. Not better accounted for by another ICHD-3 diagnosis	+	+

There are some limitations of this study. Though we suspect the GON and TON were the affected nerves, the clinical picture does not fully comply with the occipital neuralgia diagnostic criteria as described by the IHS (Table 1).[1] The headache in the first case was throbbing in nature, rather than shooting or stabbing. And the headache in the second case was not paroxysmal but occurred with pressure on the bony structures. None of the cases experienced dysaesthesia or allodynia with innocuous stimulations of the scalp. However, since the GON and TON are the clinically relevant nerves, these cases can be considered special types of occipital neuralgia. Second, since a local anesthetic block was not performed in the first case, migraine might be the diagnosis. While some aspects of the case resembled migraine without aura (throbbing unilateral pain), the absence of nausea, vomitting, phonophobia, or photophobia meant that the ICHD-3 diagnostic criteria for migraine were not met.[1] Migraine with aura was excluded since no aura occurred prior to the onset of pain. Additionally, the patient's symptoms and signs did not fulfill other subtypes of migraine defined in ICHD-3.[1] Moreover, despite several visits to neurologists in the past, he was not diagnosed with migraine at any time. For the second case, it can be argued that the exostosis can only affect the TON, but not the GON; and that pain in the orbital region cannot be related to the TON. However, it is known that the TON and GON have several communicating branches and that TON neuralgia can mimic GON neuralgia.[10,13] Furthermore, occipital neuralgia may involve the fronto-orbital region through trigeminocervical interneuronal connections in the trigeminal spinal nuclei.[1,14] As a final limitation, no surgery was demonstrated to verify the compression of aforementioned nerves.

In conclusion, exostoses of the EOP and superior nuchal lines are potential causes of neuralgic pains arising from the GON and TON. We hope that our cases will lead to increased recognition and better characterisation of these headache syndromes in other studies.

DISCLOSURE

Financial support: None

Conflict of interest: None

REFERENCES

1. International Headache Society. Headache Classification Committee of the International Headache Society (IHS) The International Classification of Headache Disorders, 3rd edition. *Cephalalgia*. 2018;38:1-211. doi: 10.1177/0333102417738202
2. Cesmebasi A, Muhleman MA, Hulsberg P, Gielecki J, Matusz P, Tubbs RS, et al. Occipital neuralgia: anatomic considerations. *Clin Anat* 2015;28:101-8. doi: 10.1002/ca.22468

3. Singh R. Bony tubercle at external occipital protuberance and prominent ridges. *J Craniofac Surg*. 2012;23:1873-4. doi: 10.1097/SCS.0b013e31826c7d48
4. Hoppenfeld JD. Cervical facet arthropathy and occipital neuralgia: headache culprits. *Curr Pain Headache Rep*. 2010;14:418-23. doi: 10.1007/s11916-010-0151-5
5. Loukas M, El-Sedfy A, Tubbs RS, *et al*. Identification of greater occipital nerve landmarks for the treatment of occipital neuralgia. *Folia Morphol (Warsz)* 2006;65:337-42.
6. Bogduk N. The anatomy of occipital neuralgia. *Clin Exp Neurol* 1981;17:167-84.
7. Bogduk N. Local anesthetic blocks of the second cervical ganglion: a technique with application in occipital headache. *Cephalalgia* 1981;1:41-50. doi: 10.1111/j.1468-2982.1981.tb00007.x
8. Tubbs RS, Salter EG, Wellons JC, Blount JP, Oakes WJ. Landmarks for the identification of the cutaneous nerves of the occiput and nuchal regions. *Clin Anat* 2007;20:235-8. doi: 10.1002/ca.20297
9. Cornely C, Fischer M, Ingianni G, Isenmann S. Greater occipital nerve neuralgia caused by pathological arterial contact: treatment by surgical decompression. *Headache* 2011;51:609-12. doi: 10.1111/j.1526-4610.2010.01802.x
10. Bogduk N, Marsland A. On the concept of third occipital headache. *J Neurol Neurosurg Psychiatry* 1986;49:775-80. doi: 10.1136/jnnp.49.7.775
11. Shahar D, Sayers MGL. Prominent exostosis projecting from the occipital squama more substantial and prevalent in young adult than older age groups. *Sci Rep* 2018;8:3354. doi: 10.1038/s41598-018-21625-1
12. Marshall RC, Abela C, Eccles S. Painful exostosis of the external occipital protuberance. *J Plast Reconstr Aesthet Surg* 2015;68:e174-6. doi: 10.1016/j.bjps.2015.06.013
13. Tubbs RS, Mortazavi MM, Loukas M, *et al*. Anatomical study of the third occipital nerve and its potential role in occipital headache/neck pain following midline dissections of the craniocervical junction. *J Neurosurg Spine* 2011;15:71-5. doi: 10.3171/2011.3.SPINE10854
14. Mason JO, 3rd, Katz B, Greene HH. Severe ocular pain secondary to occipital neuralgia following vitrectomy surgery. *Retina* 2004;24:458-9. doi: 10.1097/00006982-200406000-00021