

Chronic Unilateral Temporo-Occipital Headache Attributed to Unilateral C3 Radiculopathy

Abstract

Although C1–C3 upper cervical radiculopathy can cause a headache, most case reports are occipital neuralgia (ON), not headache. Here, we report a unique case of chronic temporo-occipital headache due to C3 radiculopathy. A 62-year-old male presented with a chronic left-sided temporo-occipital headache with duration of 4 years. The headache was aching and pressure like in nature. It had a typical radiating pattern on every occasion. It started in the posterior temporal area above the ear. It then extended to retroauricular area, then suboccipital area, and lateral neck. No hypesthesia, allodynia, or limitation in neck motion was noted. Myelographic computed tomography revealed a left-sided C2/C3 foraminal stenosis. Subsequent foraminotomy and decompression of the left C3 completely alleviated the chronic left-sided temporo-occipital headache. Unilateral C3 radiculopathy can cause chronic temporo-occipital headache besides ON. The present case might be a typical example of “headache attributed to upper cervical radiculopathy” (A11.2.4) rather than cervicogenic headache according to the International Classification of Headache Disorders, 3rd edition (beta version).

Keywords: C3 radiculopathy, greater occipital nerve, headache, lesser occipital nerve, occipital neuralgia

Introduction

Compression of intradural C2 dorsal root,^[1] encroachment of C2 root at C1–C2 cervical foramen,^[2] and peripheral entrapment of the greater occipital nerve (GON) originating from the medial branch of the dorsal ramus of the C2 nerve root^[3] can result in typically paroxysmal and stabbing pain of occipital neuralgia (ON) in the distribution of GON. Entrapment of the lesser occipital nerve (LON) and third occipital nerve (TON) has also been recognized as the source for ON. LON is mainly derived from the ventral rami of the C2 root, although the third cervical nerve (or the loop between tow) sometimes contributes.^[3,4] Many clinical and anatomical studies have demonstrated that C1–C3 nerve roots and their peripheral nerves (GON, LON, and TON) are closely related to the pathophysiology of ON. However, chronic headache without paroxysmal lancinating pain in the distribution of LON owing to C3 radiculopathy from C2–C3 foraminal stenosis has not been reported yet.

Although the present case might be regarded as cervicogenic headache or

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occipital neuralgia at a glance, this could be a typical example of “headache attributed to upper cervical radiculopathy” (A11.2.4) according to the International Classification of Headache Disorders (ICHD), 3rd edition (beta version).^[5]

Case Report

A 62-year-old male with a 4-year history of chronic intermittent aching pain along the distribution of the left LON presented severe aggravation in frequency and intensity of left-sided headache with duration of 4 months. An aching pain over the left temporal area above the ear was associated with left mandibular angle areas during its initial development 4 years ago. He had been treated for dental caries. However, the treatment was not effective. He had experienced intermittent neck pain prior to the occurrence of left temporal headache. However, the pain was not severe enough to seek medical consultation. There was no precipitating event before the onset of headache. The pain was described as aching and pressure like in nature with moderate intensity (4–6 out of 10 in the numerical rating scale [NRS]). During the initial 2 years, the cluster of left temporal

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Hak-cheol Ko¹, Byung-chul Son^{1,2}

¹Department of Neurosurgery, Seoul St. Mary's Hospital, College of Medicine, The Catholic University of Korea, ²Catholic Neuroscience Institute, College of Medicine, The Catholic University of Korea, Seoul, Republic of Korea

Address for correspondence:

Prof. Byung-chul Son,
Department of Neurosurgery,
Seoul St. Mary's Hospital,
Catholic Neuroscience Institute,
College of Medicine, The
Catholic University of Korea,
222 Banpo-daero, Seocho-gu,
Seoul 06591, Republic of Korea.
E-mail: sbc@catholic.ac.kr

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headache with duration of 2–3 weeks occurred 3–4 times a year. The frequency of headache cluster was increased to 5–6 times a year and its intensity was gradually increased.

One year prior to presentation, the pattern of occurrence and spreading of the headache became typical on every occasion. An aching pain gradually developed in his left upper temporal area over the ear and spread to posterior temporal and retroauricular occipital areas [Figure 1]. It then extended to the suboccipital area and lateral neck. Duration lasted up to 1 h. The headache even developed during sleep. He could not fall asleep again after awakening. He has been using three pillows on bed for more than a year to prevent the occurrence of headache. Four months prior to admission, the headache was continuously present. It occurred more than five times a day. He had been treated with various kinds of medication including triptans, propranolol, indomethacin, tramadol, and antidepressants. They were not effective at all. Repeated blocks of the GON were partially effective about 2 h. Only injection of diclofenac sodium was effective for 3 h. Magnetic resonance imaging (MRI) findings of the brain and cervical spine were nonspecific. He was finally referred for the management of refractory headache.

The aching and deep pressure-like headache was severe in intensity (6–7 out of 10 on NRS) at presentation. It was not aggravated with neck motion including extension. Daily activity did not influence the occurrence or frequency of headache. His medical history was unremarkable, including diabetes and gout. He denied any history of trauma to the neck. The headache was not associated with tinnitus, photophobia, lacrimation, nausea, or vomiting. Upon examination, there was no hypesthesia, allodynia, or tenderness in his left head or neck. No limitation of neck motion was observed. Neurologic examination including cranial nerve was normal. Careful reevaluation of cervical

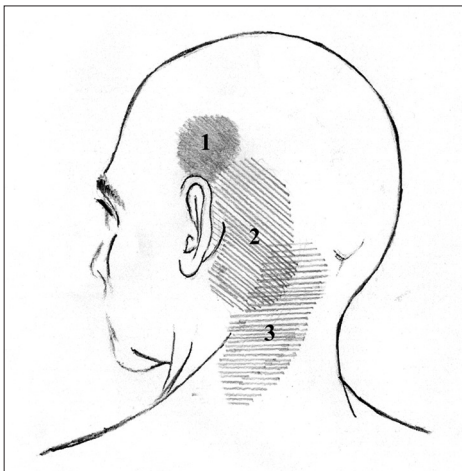


Figure 1: A schematic diagram demonstrating the distribution and spreading of the temporo-occipital headache owing to C3 radiculopathy. An aching pain gradually developed in his left posterior temporal area over the ear (1) and spread to retroauricular occipital area (2). It then extended to suboccipital area and lateral neck (3)

spinal MRI raised a suspicion of foraminal stenosis at the left C2/3. Therefore, myelographic computed tomography (CT) of the cervical spine was requested. A sagittal oblique three-dimensional reconstruction of myelographic CT revealed significant narrowing of the left C2/3 foramen and small osteophyte of the uncovertebral joint [Figure 2]. Under impression of the left C3 radiculopathy owing to foraminal stenosis, three times of selective C3 root blocks were performed with 2 ml of 1% lidocaine. The pain was completely relieved for 2 h.

Considering chronicity and severity of the headache, decompressive foraminotomy of the left-sided C2/3 spine was performed. The lateral edge of dural sac and proximal nerve root sheath of the left C3 were decompressed under microscopic vision [Figures 3 and 4]. Immediately after awakening from anesthesia, the pain completely disappeared. No hypesthesia or paresthesia was reported. The patient refused any medication against headache. There was no recurrence of headache or sensory deficit in his left temporal area at 1-year postoperative follow-up.

Discussion

Cephalic pain due to C3 radiculopathy

Cephalic pain from cervical disease is mediated by C1, C2, or C3 nerve.^[6] Nociceptive inputs from C1, C2, and C3 nerves can cause pain in the head, but not in the neck.^[7] Although the pattern of pain referral in greater or lesser ON is well known, pain distribution in C3 radiculopathy is uncommon because isolated C3 radiculopathy is rare. C3 pain dermatome consists of a craniofacial area including scalp above and behind the ear, the medial and lateral surfaces of the pinna, the lateral cheek over the angle of the jaw, the submental region, and the lateral and anterior aspects of the upper neck.^[6] In the present case, the pain attack showed a typically stereotyped spreading pattern which initially



Figure 2: A sagittal oblique image of myelographic computed tomography of the cervical spine showing narrowing of the left C2/3 foraminal stenosis and small osteophyte causing chronic C3 radiculopathy

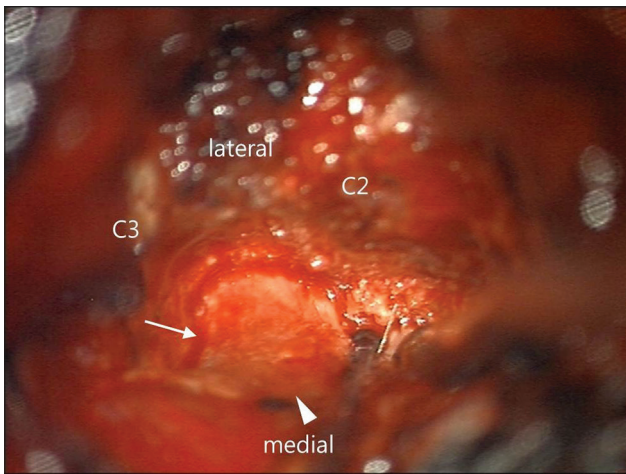


Figure 3: An intraoperative photograph showing the decompressed C3 nerve root (arrow) with foraminotomy. An arrowhead indicates the cord dura

started in the temporal area above the ear, and then spread to the posterior temporal area behind the ear, suboccipital area, and the upper posterior neck in every attack. In the suboccipital area, C3 radicular pain involved the area supplied by the TON. TON arises from the medial branch of the dorsal ramus of the C3 nerve.^[8] The medial terminal branch of TON supplies the skin over the rostral end of the neck and the occiput near the external occipital protuberance. More lateral branches of TON are directed toward the mastoid process.^[8]

Headache, not occipital neuralgia, from C3 radiculopathy

The present case did not show any feature of ON such as pain paroxysm, pain quality of shooting, stabbing, or sharp, dysesthesia, allodynia, tenderness, or trigger points suggested in the ICHD, 3rd edition.^[5] Rather, it was mostly aching and constant during the attack which lasted >30 min. As it showed side-locked unilateral distribution without the feature of trigeminal autonomic cephalalgias, the present case has been treated under the impression of migraine and cervicogenic headache. The present case might be thought as cervicogenic headache, a disorder of the cervical spine and its component bony, disc, and/or soft-tissue elements, usually but not invariably accompanied by neck pain.^[5] However, cervicogenic headache is generally referred to an occipital referral of pain arising from the atlantoaxial or upper zygapophyseal joints or from tender trigger points in neck muscles or their insertions.^[5] It is now well understood that convergence between upper cervical and trigeminal nociception is a logical cause of headache.^[4] Therefore, headache, not ON, caused by upper cervical radiculopathy is classified in Appendix as A11.2.4, headache attributed to upper cervical radiculopathy.^[5] However, the headache in the present case was caused by pure C3 radiculopathy due to unilateral foraminal stenosis, not by any referral to the trigeminal distribution with involvement of the trigeminocervical complex.^[9]



Figure 4: A postoperative, sagittal oblique image of computed tomography of the cervical spine showing widening of the left C2/C3 foramen (arrow)

Conclusion

Chronic refractory side-locked temporal headache radiating to retroauricular and suboccipital areas owing to C3 radiculopathy from foraminal stenosis is presented in this case report. Until now, all case reports regarding chronic pain from upper cervical radiculopathy of C1–C3 have been described as ON. The present case is regarded as typical example of “headache attributed to upper cervical radiculopathy” (A11.2.4) according to the ICHD, 3rd edition (beta version).

Institutional review board approval

All procedures performed in the current study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This study was approved by our university’s institutional review board. The ethical reference number is KC17ZESE0251.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given his consent for images and other clinical information to be reported in the journal. The guardian understands that names and initials will not be published and due efforts will be made to conceal patient identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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