

안면부 통증을 보인 비전형적 Gradenigo 증후군 1예

전북대학교 의과대학 신경과학교실

임의성 오선영 서만욱 우성룡 정석영 양태호

Atypical Gradenigo's Syndrome Showing Isolated Facial Pain

Eui-Seong Lim, M.D., Sun-Young Oh, M.D., Man-Wook Seo, M.D., Seong-Ryong Woo, M.D.,
Seok-Young Jeong, M.D., Tae-Ho Yang, M.D.

Department of Neurology, Chonbuk National University Medical School, Jeonju, Korea

J Korean Neurol Assoc 23(3):434-435, 2005

Key Words: Gradenigo's syndrome, Facial pain, Petrositis

Gradenigo's syndrome consists of the triad of otitis media, mastoiditis, pain in the region innervated by the first and second divisions of the trigeminal nerve and ipsilateral abducens nerve palsy.¹ In 1907, Gradenigo described a symptom complex consisting of abducens nerve palsy, severe pain in the distribution of the trigeminal nerve, and acute suppurative otitis media. In the antibiotic era, the incidence of Gradenigo's syndrome decreased markedly and only a few cases have been reported. Gradenigo's syndrome is often recognized late due to the subtlety of its signs and symptoms.² We report a patient with unilateral facial pain. He was diagnosed as having atypical Gradenigo's syndrome on the basis of clinical and radiologic findings.

CASE REPORT

A 61-year-old man visited the department of

Neurology, Chonbuk National University Hospital in May 2002. He complained about right-sided, long lasting facial pain and headache. A dentist had treated him for the last six months because of right preauricular pain, without any symptoms of otitis media and externa.

Physical examination showed that facial pain could be localized in the second branch of the right trigeminal nerve. The pain occurred intermittently and was exacerbated by chewing or speaking. However, no other cranial nerve dysfunction was noted, including facial and abducens nerves. The patient was alert and showed intact higher cortical function. And there was neither fever nor meningeal irritation signs. Medications for a relief of pain, including carbamazepine 400 mg and indomethacin 50 mg, were given. Then, the facial pain was slowly improved and the patient did not come to our clinic.

In August 2003, the patient was suffered from tinnitus in the right ear and ipsilateral facial pain. Brain MRI demonstrated a chronic inflammation of the right petrous apex, showing low-signal intensity on T1-weighted images and high-signal intensity on T2-weighted images (Fig. 1). In October 2003, an otolaryngological examination revealed no active infection or aural discharge in the right ear. On pure tone audiometry, there was no

Received August 9, 2004 Accepted November 18, 2004

* Man-Wook Seo, M.D.

Department of Neurology, Chonbuk National University Hospital
634-18 Geumam 2-dong, Deokjin-gu, Jeonju-si, 561-712, Korea
Tel: +82-63-250-1895 Fax: +82-63-251-9363
E-mail: smw@chonbuk.ac.kr

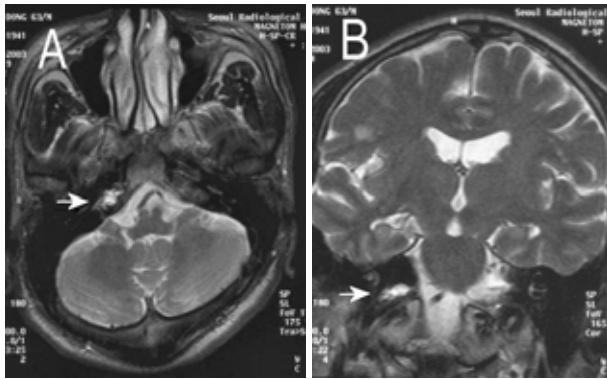


Figure 1. Brain MRI. This shows high signal intensity (arrow) in right petrous apex on T2 weighted axial (A) and coronal images (B).

sound matched with his tinnitus. Temporal bone CT demonstrated the loss of the pneumatization of the right petrous apex. With these findings, no antibiotics or surgical intervention was performed. Two weeks later, he visited our neurology clinic. Physical examination showed that the right facial pain was steady in duration, lancinating in nature, and worsened at night. Bone SPECT, however, did not show any increased uptake in the right temporal bone. So, he was treated with combinations of medicine including carbamazepine 600 mg, indomethacin 50 mg, gabapentin 900 mg, nortryptiline 12.5 mg, of daily amounts. And then, the facial pain was improved and did not recur.

DISCUSSION

We presented an unusual cause of facial pain, Gradenigo's syndrome. Even the classic triad was not observed in the present case, the combination of petrositis that was confirmed by MR imaging and the ipsilateral facial pain made us to consider the possibility of the diagnosis.

The most common symptom in the Gradenigo's syndrome is deep pain in the ear, face, and retrobulbar region, as in the present case.³ The deep pain of petrositis has been explained by four possible mechanisms; referred pain from the ear, dural irritation in the tegmen area, inflammation of the gasserian

ganglion, or localized meningitis.^{3,4} So, the Gradenigo's syndrome might be considered as a diagnostic possibility in the presence of unilateral facial pain.

Actually, many patients diagnosed as having the Gradenigo's syndrome do not present with all the three components. In Gradenigo's description of 57 cases of petrous apicitis, 24 cases had the pure triad, 29 cases had additional cranial nerve deficits, and 4 cases had meningitis resulting in death.³ In Chole and Donald's description of 8 cases of petrous apicitis, only 1 case had the pure triad and the remainder had no triad.³

The widespread use of antibiotics to treat otitis media and externa has virtually eliminated an infectious cause of the Gradenigo's syndrome. In the modern era, patients with these findings should be evaluated for neoplasm such as meningioma, chordoma, trigeminal neuroma, gasserian ganglion tumors, or metastasis; cholesteatoma, hematoma; aneurysm of the intrapetrous internal carotid artery; or carcinomatous meningitis.^{1,4} The petrous portion of the temporal bone is a pyramidal structure located in the vicinity of the fifth and sixth cranial nerves. Infections in the petrous apex may be life-threatening because of their propensity to spread medially to involve the meninges, cavernous sinus, and brain.⁵

Physicians must be alert to the underlying cause of facial pain and the Gradenigo's syndrome should be considered in the patients presenting with hemifacial pain.

REFERENCES

1. de Graaf J, Cats H, de Jager AE. Gradenigo's syndrome: a rare complication of otitis media. *Clin Neurol Neurosurg* 1988;90:237-239.
2. Tutuncuoglu S, Uran N, Kavas I, Ozgur T. Gradenigo syndrome: a case report. *Pediatr Radiol* 1993;23:556.
3. Chole RA, Donald PJ. Petrous apicitis. Clinical considerations. *Ann Otol Rhinol Laryngol* 1983;92:544-551.
4. Hardjasudarma M, Edwards RL, Ganley JP, Aarstad RF. Magnetic resonance imaging features of Gradenigo's syndrome. *Am J Otolaryngol* 1995;16:247-250.
5. Finkelstein Y, Marcus N, Mosseri R, Bar-Sever Z, Garty BZ. Streptococcus acidominimus infection in a child causing Gradenigo syndrome. *Int J Pediatr Otorhinolaryngol* 2003;67:815-817.