—Case Reports—

A Case of Headache Attributed to Otitis Media Chronica Cholesteatomatica with Cerebral Sigmoid Sinus Thrombosis

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Abstract

Otitis media chronica cholesteatomatica with cerebral sigmoid sinus thrombosis is an important differential diagnosis in the evaluation of headache. We describe a 31-year-old Filipino man with chief complaints of headache, otalgia, vomiting, and vertigo, and no significant past medical history. Two years before admission he stuffed tissues into the right external auditory canal because of a noise in the street on the night of the New Year's festival and sometimes had right ear discharge. One month before admission he had a right occipital headache with right otalgia and fever. One day before admission he vomited. Vertigo developed on the day of admission. On physical examination at admission, the patient was somnolent and had a body temperature of 36.9℃, and meningeal signs were obvious. Magnetic resonance of the brain revealed right otitis media chronica cholesteatomatica and right cerebral sigmoid sinus thrombosis. Computed tomography of the skull base revealed that the tympanic cavity and mastoid air cells were filled with a mass of soft-tissue density invading toward the sigmoid sinus. Cerebrospinal fluid examination showed a cell count of 32/3 mm^3. The patient's level of consciousness and symptoms improved after administration of ampicillin and ceftriaxone sodium. A diagnosis of "headache attributed to disorder of ears," with *The International Classification of Headache Disorders, 2nd edition (ICHD-II)* code 11.4, was made on the basis of symptoms and imaging findings. Otitis media chronica cholesteatomatica had invaded toward the sigmoid sinus and was thought to be the cause of cerebral sigmoid sinus thrombosis and meningitis. Six weeks after admission the patient underwent mastoidectomy and tympanoplasty to cure the cholesteatoma and prevent recurrence of inflammation. The postoperative progress was satisfactory. In cases of headache with otalgia, "headache attributed to disorder of ears" should be considered.

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Key words: headache, otalgia, otitis media chronica cholesteatomatica, cerebral sigmoid sinus thrombosis, meningitis

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Table 1 Summary of laboratory findings

Laboratory tests	
Complete blood cell count	leukocytosis (14,120/mm^3)
Blood chemistry	hyperbillirubinemia (1.3 mg/dL)
Glucose	normal value (134 mg/dL)
C-reactive protein	elevated C-reactive protein (4.0 mg/dL)

Introduction

According to *The International Classification of Headache Disorders, 2nd edition (ICHD-II)*[1], headaches are classified into three categories, namely, "the primary headaches", "the secondary headaches", and "cranial neuralgias, central and primary facial pain, and other headaches". The representative primary headaches are migraine, tension-type headache, and cluster headache, but the secondary headache should be paid attention to and ruled out when headache is diagnosed.

Otitis media chronica cholesteatomatica with cerebral sigmoid sinus thrombosis is an important cause of "headache attributed disorder of ears" (*The ICHD-II* code 11.4) in terms of the secondary headache. We describe a case of headache in a 31-year-old man which shows the importance of the ear disease as a cause of headache.

Case Report

The patient was a 31-year-old Filipino man with chief complaints of headache, otalgia, vomiting, and vertigo, and no significant past history. Two years earlier he had stuffed tissues into the right external auditory canal because of a noise in the street on the night of the New Year's festival and sometimes had right ear discharge. One month before admission he had right occipital headache with right otalgia and fever. One day before admission he vomited. Vertigo developed on the day of the admission.

On physical examination at admission, the patient was somnolent. The temperature was 36.9℃. Blood pressure was 112/68 mmHg. Meningeal signs were obvious. Laboratory studies showed leukocytosis (14,120 /mm^3), slight hyperbilirubinemia (1.3 mg/dL), and elevated level of C-reactive protein (4.0 mg/dL). When the postprandial hematological examination was considered, blood glucose was within normal limits. The slightly increased serum level of bilirubin normalized soon after admission (**Table 1**). Chest X-ray films showed that the lungs were clear, and films of the abdomen showed a normal pattern of bowel gas. Results of electrocardiography were normal. Magnetic resonance (MR) of the brain revealed right otitis media chronica cholesteatomatica and right cerebral sigmoid sinus thrombosis (**Fig. 1**). A computed tomography (CT) scan of skull base revealed that tympanic cavity and mastoid air cells were filled with a mass of soft-tissue density that had destroyed the skull base and invaded toward the sigmoid sinus (**Fig. 2**). Cerebrospinal fluid examination showed elevated pressure (420 mm H_2O), cell count (32/3 mm^3), and protein level (74 mg/dL) (**Table 2**).

The patient's level of consciousness and symptoms improved after administration of ampicillin and ceftriaxone sodium. A diagnosis of "headache attributed to disorder of ears" with *The ICHD-II* code 11.4 was made on the basis of symptoms, and MR and CT findings. The cerebrospinal fluid findings of elevated pressure, cell count, and protein with normal culture finding suggested that there was also nonbacterial meningitis. Otitis media chronica cholesteatomatica had destroyed the skull base and made contact with the sigmoid sinus and was thought to be the cause of cerebral sigmoid sinus thrombosis and meningitis. Both cerebral sinus thrombosis and meningitis have otogenic intracranial consequences[2,3], but the precise mechanisms of cerebral sinus thrombosis and meningitis were obscure in the present case. Six weeks after admission, mastoidectomy and tympanoplasty were performed (**Fig. 3**) to cure the cholesteatoma and prevent recurrence of inflammation.

The postoperative progress was satisfactory. In

Table 2 Cerebrospinal fluid examination

Cerebrospinal fluid tests	
Pressure	420 mmH_2O
pH	7.0
White blood cells	32/3 mm^3 (with 88% lymphocytes)
Total protein	74 mg/dL
Glucose	72 mg/dL
Cl	123 mEq/L
Bacteriologic culture, fungus, and tuberculosis	—

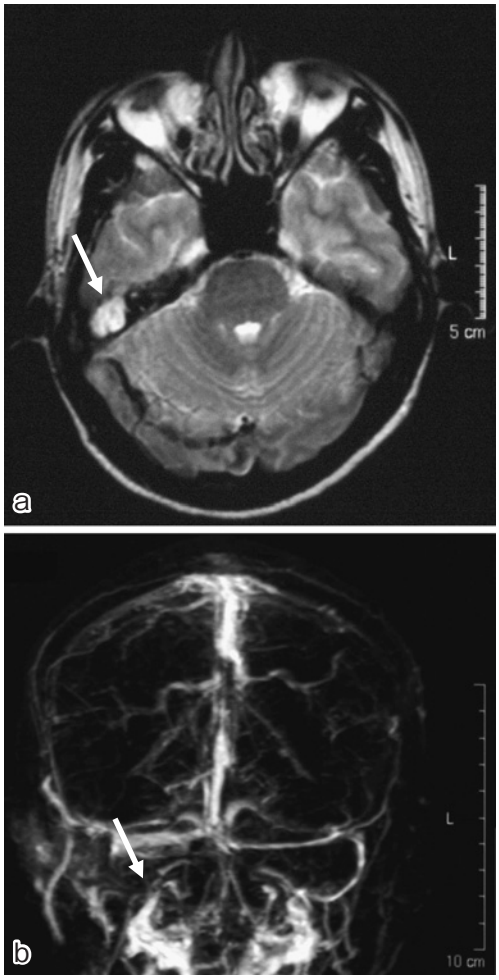

Fig. 1 Brain magnetic resonance (MR) imaging. Abbreviation. L, left.
a: T2-weighted MR imaging of the brain revealed right otitis media chronica cholesteatomatica, measuring 1×2 cm, with high-intensity areas **(white arrow)**.
b: MR venography showed a defect of the right cerebral sigmoid sinus, namely, right cerebral sigmoid sinus thrombosis **(white arrow)**.

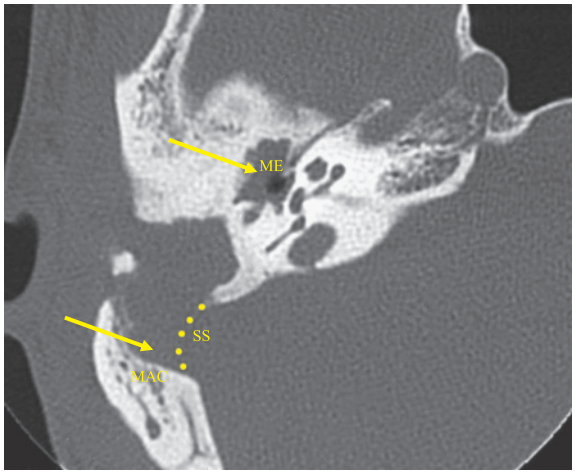

Fig. 2 CT scan of base of the skull. Abbreviation. ME, middle ear. MAC, mastoid air cells. SS, sigmoid sinus.
CT scan of skull base revealed that tympanic cavity and mastoid air cells were filled with a mass of soft-tissue density **(yellow arrows)** destroying the skull base **(yellow dotted line)** and invading toward the sigmoid sinus.

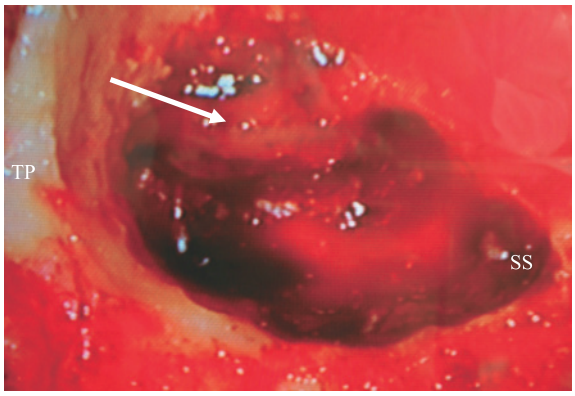

Fig. 3 View of mastoidectomy. Abbreviation. SS, sigmoid sinus. TP, tegmen plate.
The goals of the operation were to remove the cholesteatoma **(white arrow)**, which was the focus of the meningitis, and to reconstruct the tympanic membrane.

the near future, reconstruction of the ossicular chain of this patient is scheduled to be performed.

According to *The ICHD-II*, diagnostic criteria of "headache attributed to disorder of ears" consists of A. headache accompanied by otalgia and fulfilling criteria C and D; B. structural lesion of the ear diagnosed by appropriate investigations; C. headache and otalgia develop in close temporal relation to the structural lesion; and D. headache and otalgia resolve simultaneously with remission or successful treatment of the structural lesion. The headache of the present patient fulfilled these diagnostic criteria.

Discussion

This case of headache in a 31-year-old man was attributed to otitis media chronica cholesteatomatica with cerebral sigmoid sinus thrombosis and showed the importance of ear disease in the differential diagnosis of headache.

The otogenic intracranial consequences include cerebral abscess, cerebellar abscess, cerebellar venous infarction, meningitis, cerebral sinus thrombosis, otitic hydrocephalus, and cortical thrombophlebitis[2,3]. The prevailing cause among ear diseases is otitis media chronica, especially otitis media chronica cholesteatomatica. Diagnosis of the intracranial consequences is made on the basis of clinical features, neurological signs, and investigative findings. Otogenic intracranial consequences should be treated together with the otitis media as a primary lesion. The therapy includes administration of appropriate antibiotics and operations, such as mastoidectomy and removing cholesteatoma. It has been reported that 9% of patients who undergo mastoidectomy have endocranial complications[4]. When headache, prolonged fever, consciousness disturbance, menigeal sign, and seizure are observed in the course of otitis media chronica, "headache attributed disorder of ears" (*The ICHD-II* code 11.4) and the intracranial consequences must be paid attention to. The secondary headache including "headache attributed disorder of ears" should be considered and ruled out when headache is evaluated.

However, generally speaking, after the introduction of antibiotics, intracranial complications in otitis media became less common, and otitis media chronica cholesteatomatica seldom becomes severe or leads to the complication of thrombosis of the deep cerebral sinuses. Barbara, et al[5]. have reported a case of noncoalescent mastoiditis in a young child in whom a genetic thrombotic disorder (prothrombin G20210A allele mutation) was identified as the predisposing factor for the unusual complication of thrombosis of deep cerebral sinuses. In the present case, the patient was Filipino. Therefore, assessment of coagulation factors is necessary in the population at risk, and genetically induced deep venous thrombosis should be considered. We could not investigate this matter in detail. On the other hand, the otitis media in our patient might not have been treated appropriately in the early stage.

In conclusion, in a case of headache with otalgia, "headache attributed to disorder of ears" should be considered.

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