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a case report

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耳痛で発症したくも膜下出血の1例

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Subarachnoid hemorrhage with the first complaint of otalgia : a case report

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Subarachnoid hemorrhage (SAH) is an emergency in neurology. As SAH sometimes attacks without significant clinical manifestations, it may be misdiagnosed as other diseases. In this paper, a case of SAH with the first complaint of otalgia was reported. Some related literatures were reviewed to discuss its clinical characteristics, early diagnosis and treatment.

Key words : subarachnoid hemorrhage, otalgia, intracranial aneurysm, early diagnosis

Introduction

Subarachnoid hemorrhage (SAH) is an emergency in neurology and it presents the proceeding symptoms of sudden severe headache, nausea, vomiting, and so on¹⁾. However, seldom does SAH present otalgia as the first complaint and symptom. In 1996, a patient of SAH complaining otalgia was diagnosed and treated in Miyoshi ENT Clinic. It was reported as follow, and some literatures were reviewed.

Case Report

A female patient of 53-year old, complained left side otalgia and radical headache around left ear for 3 days on September 4, 1996. No abnormal sign was found in tympanic membrane. The acoustic impedance test also showed no problem. When waiting for further examination, she fell into disturbance of consciousness suddenly. Suspected of apoplectic seizure, she was send to the Izumi Hospital for neurological

treatment. Skull computer tomography (CT) scanning suggested subarachnoid hemorrhage (Fig. 1), and dissecting aortic aneurysm (Fig. 2) was showed by digital subtraction angiography (DSA). Then the patient received the neurosurgical treatment there and recovered.

Discussion

SAH attacks in nearly every age according to different etiology. The epidemiological study showed that the incidence of SAH is



Figure 1 CT scanning showed subarachnoid hemorrhage.

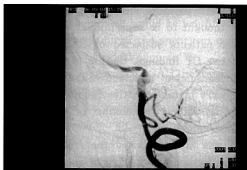


Figure 2 Dissecting aortic aneurysm was detected by DSA.

stable, at around 6 cases per 100,000 person-years¹⁾. According to an investigation on the incidence of SAH among 89,991 populations in Shimokita peninsula of Japan from 1989 to 1998²⁾, 198 cases were diagnosed as SAH, and the age-adjusted annual incidence of SAH was 21 per 100,000 person-years. The age and sex distribution of the cases indicated that the incidence became plateau after age 45 in men while increased after age 45 and reached the peak after 75 in women. Another study³⁾ showed that the mean age of cases was 57 years (ranged from 16 to 94 years), and 62% were female. Age-specific rates showed a continuous upward trend with age, although the shape and strength of this association differed between the sexes. Our case was a female of 53 years old, and was just around the age susceptible to aneurysmal SAH.

Generally, SAH occurs suddenly with the symptoms of severe headache, nausea and vomiting¹⁾. Sometimes, emotion and over-using strength are inducing reasons. Some of the patients have the proceeding symptoms of headache, and central nerve paralysis. Some fall into disturbance of consciousness, even coma. In this patient, otalgia and radical headache were the only signs of the initial attack of SAH. And then the patient

fell into a severe status of consciousness disturbance suddenly. This suggested that SAH sometimes came latently without any significant signs, which should be paid enough attention to.

SAH seldom attacks with the first complaint of otalgia. The possible mechanism may be that the blood and its decompositive products in subarachnoid cavity stimulate the superior cervical root, and the irritative symptoms of occipital and auricular nerve, as the branch of cervical root, are caused then.

SAH is mainly (85%) caused by the intracranial aortic aneurysm¹⁾. Deformation and atheromatous sclerosis of brain vessel are also the common etiology⁴⁾. In our case, SAH was caused by the dissecting aortic aneurysm.

Ruptured intracranial aortic aneurysm causes not only SAH, but also subdural hematoma (SDH) concurrently sometimes, with an unfavorable prognosis and high mortality in this occasion⁴⁾. SDH also happens subsequent to SAH usually after 7–10 days⁵⁾. Aortic atheromatous sclerosis, attack of transient ischemic, Hunt-Hess grade II and III and advanced age are risk factors.

As SAH sometimes attacks without significant symptoms, it may be misdiagnosed as common cold, vertigo, vessel-nerval headache, transient ischemic attack (TIA), cerebular infarction, cerebral hemorrhage, epidemic encephalitis, nerval pain of occipitalis and sciatic nerve, and so on. It was also reported that 14.8% patients with aneurysms were diagnosed with a significant delay varying from 2 days to a few months⁶⁾, before definitive treatment. The delay in diagnosis of aneurysmal subarachnoid hemorrhage resulted in poor clinical grade and influenced neurological outcome significantly.

The clinical symptoms differ a little between younger and elder. A group of 59

cases of SAH were delivered into elder group (over 60-year old) and younger group (under 60-year old) so as to compare the clinical characteristics as summed up in Table 1⁷⁾. As a result, it was suggested that elder patients tended to present consciousness disturbance, with lower headache and meningeal irritation signs on the contrary, and was easier to be misdiagnosed, with more concurrent complication, recurrent tendency and higher mortality. The present case seemed to match the characteristic to some degree.

Early diagnosis helps to get favorable prognosis. The diagnosis of atypical SAH usually relates to age, amount of bleeding, occasion of CT scanning and spinal puncture⁸⁾. CT could act as scanning test in acute stage of SAH, while in subacute and chronic stage, magnetic resonance imaging (MRI) is helpful, which is less clear than magnetic resonance angiography (MRA) and DSA. Spiral CT angiography was useful in the detection of cerebral aneurysms in patients with SAH when angiography revealed no diagnostic findings. Anterior communicating artery aneurysms are generally well hidden in these types of SAH cases. A repeated angiography session was warranted in patients with nonperimesencephalic SAH and in those who initial angiography revealed no

diagnostic findings, although a third session was thought to be superfluous⁹⁾.

For patients suspected of SAH but with negative CT findings, the use of fluoroscopy-guided lumbar puncture could reduce the frequency of false-positive diagnoses of acute SAH as well as the number of unnecessary angiograms¹⁰⁾.

It was also suggested that teaching programs focused on local physicians had a profound impact on outcome at low cost, as misdiagnosed warning episodes caused greater loss of lives and higher morbidity on a population basis than delayed ischemic complications from vasospasm in aneurysmal SAH did¹¹⁾.

A clinical study by Nakase *et al*¹²⁾ has demonstrated two risk factors for intracranial aneurysm recurrence, namely, young age and internal carotid (IC)-posterior communicating artery (PC) aneurysms with a primitive-type PC. Furthermore, direct operation for recurrent aneurysm is often embarrassing due to adhesion to the surrounding tissue as a sequela of the previous operation, hence understanding of the anatomical correlation between the old clip and the recurrent portion is important.

Treatment of residual or recurrent aneurysms after surgical clipping is a challenge and most surgeons prefer to avoid a second

Table 1 Comparison of clinical characteristics in patients with SAH between younger and elder⁷⁾

Items	Younger group (<60yr)	Elder group (≥60yr)
Chronic occurrence	13.3%	30.5%
Proceeding disease	37.2%	64.1%
Headache	47.8%	13.6%
Consciousness disturbance	30.7%	83.0%
Complication	15.0%	39.0%
Misdiagnosis	15.9%	44.0%
Mortality	12.4%	36.0%

surgical attempt. However, sometimes the endovascular approach would be helpful when complete obliteration of the aneurysm cannot be achieved by surgical clipping¹³⁾.

Active management of SAH indicated favorable prognosis. Outcome prediction is usually based on patient characteristics and clinical and radiological findings. Current clinical grading scales are imprecise, with low interobserver reproducibility. Therefore, outcome prediction remains inconsistent and difficult, especially for patients with poor clinical grade. Recently, serum levels of soluble intercellular adhesion molecule-1 (sICAM-1) and apolipoprotein E genotype were found related to poor outcome in patients with aneurysmal SAH^{14), 15)}.

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今回われわれは耳痛で発症したくも膜下出血の1例を経験した。症例は53歳女性、左耳痛とその周辺への放散痛を主訴として来院した。CTおよび血管造影(DSA)にて、くも膜下出血と解離性動脈瘤が判明した。本症例においてはくも膜下腔に貯留している血液とその分解成分による神経根への刺激により耳痛が生じたものと推察された。時にくも膜下出血は典型的な症状を伴わず、耳痛、めまい、風邪様の症状にて発症することがある。単なる耳痛を訴えて耳鼻科を受診した症例に対してもくも膜下出血の可能性を念頭に置く必要がある。
