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A case of hypertrophic cranial pachymeningitis associated with invasive *Aspergillus* mastoiditis

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ABSTRACT

We report a rare case of hypertrophic cranial pachymeningitis (HCP) associated with invasive *Aspergillus* mastoiditis. A 63-year-old man with diabetes mellitus underwent mastoidectomy because of chronic discharge from his left ear. The mastoidectomy was unsuccessful in resolving purulent otorrhea; moreover, 7 months later, the patient developed left abducens nerve palsy. Magnetic resonance imaging revealed HCP at the left middle cranial fossa. Although the pathogen could not be identified, an *Aspergillus* infection was considered based on elevated serum β-D-glucan and a positive *Aspergillus* antigen test result. Voriconazole treatment resolved diplopia and left otorrhea and dramatically improved HCP.

1. Introduction

In recent years, the frequency of invasive fungal inflammation has increased with the corresponding increase in the use of steroids, incidence of diabetes mellitus, and the administration of antibacterial agents. However, the incidence of fungal mastoiditis is rare, even in immunocompromised patients. *Aspergillus* usually grows on the surface of the epithelium in anaerobic and wet environments and responds to simple conservative removal. When the ear's skin barrier is broken, the fungi might enter subcutaneous tissue and induce invasive fungal mastoiditis. This invasion might spread to the surroundings, resulting in osteomyelitis, meningitis, and encephalitis. As reported by Amonoo-Kuhofi [1], invasive temporal bone mycoses are difficult to diagnose and are often fatal.

Hypertrophic cranial pachymeningitis (HCP) is a rare inflammatory disease causing thickening and fibrosis of the dura mater. Its symptoms include headache, ataxia, and cranial nerve palsies. The etiology of HCP is diverse. Early reports of HCP showed an association with tuberculosis and syphilis. It was also found to occur idiopathically [2]. Recently, several other causes of HCP have been recognized, including infections, autoimmune disorders, and neoplasms [3]. However, the occurrence of HCP as a complication of insidious *Aspergillus* mastoiditis is very rare [4].

Here, we present a case of HCP resulting from invasive *Aspergillus* mastoiditis and discuss its diagnosis and treatment.

2. Case report

A 63-year-old man with a history of diabetes mellitus and a complaint of left otorrhea for the past 3 months was referred to our hospital. An otoscopic examination revealed that his left tympanic membrane (TM) was hyperemic and thickened; perforation of the TM was not recognized (Fig. 1a). The otorrhea sample was cultured, but it did not grow any bacterium or fungus. An audiometric analysis showed mild conductive hearing loss in the left ear and normal hearing in the right ear. Computed tomography (CT) revealed that the mastoid and tympanic cavities were diffusely clouded; however no intracranial pathological changes were observed (Fig. 2a).

Based on the diagnosis of chronic otitis media, we performed left mastoidectomy together with ventilation tube insertion in the left TM. The operation showed that the mastoid and tympanic cavities were filled with granulation tissue; histopathological study of the granulation revealed non-specific inflammation. Postoperative course was uneventful, except continual discharge from the left ear. The patient was discharged 10 days after the operation. Despite persistent postoperative care, otorrhea continued through the

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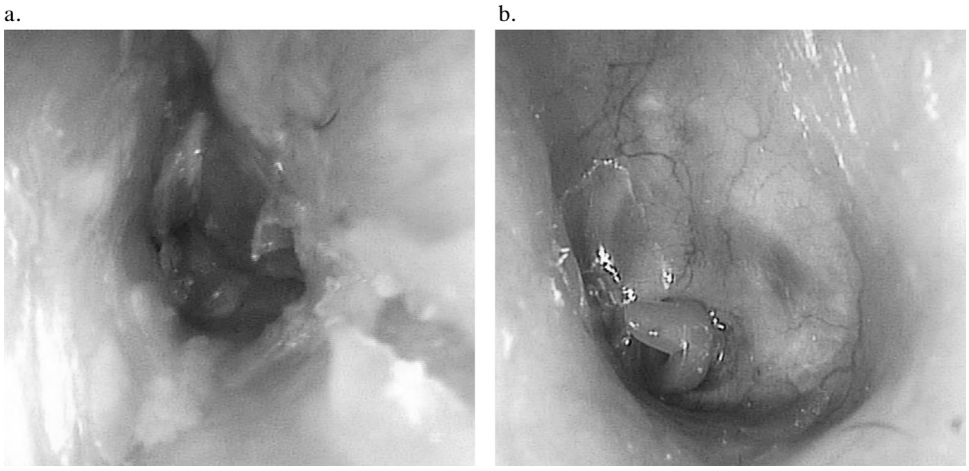

Fig. 1. (a) Left tympanic membrane (TM) before surgery. The TM is hyperemic and thickened without perforation. (b) Left TM after voriconazole therapy. The tympanic cavity is well aerated without otorrhea.

ventilation tube. The administration of various antibiotics, including cefazolin for 3 days, sitafloxacin for 1 week, and local instillation of ofloxacin for 1 month, was ineffective.

Seven months after the operation, the patient reported diplopia and headache without fever. A physical examination revealed paralysis of the left abducens nerve. No spontaneous nystagmus was observed, and the function of the facial nerve was normal. Magnetic resonance image (MRI) revealed HCP at the left middle cranial fossa extending to the cavernous sinus (Fig. 3a–c). The CT scan did not show a bony deficit in the mastoid (Fig. 2b). Repeated culture of the otorrhea samples did not reveal any pathogen. Serological examinations revealed elevation of the C-reactive protein (0.58 mg/dL, normal range 0.00–0.20 mg/dL), β-D-glucan (15 pg/mL, normal range <11 pg/mL), and HbA1c levels (6.3%, normal range 4.6–6.2%). The *Aspergillus* antigen test yielded positive results; the *Cryptococcus neoformans* antigen test was negative. A serum sample was negative for rheumatoid factor (RF), antinuclear antibody (ANA), proteinase 3 anti-neutrophil cytoplasmic antibody (PR3-ANCA), myeloperoxidase anti-neutrophil cytoplasmic antibody (MPO-ANCA), and tuberculosis. Cerebrospinal fluid (CSF) examination also revealed an elevated cell count of 742 cells/μL (normal range 0–5/μL), although protein and glucose levels were well within the normal range. Culture analysis of the CSF did not reveal any pathogen. Based on the suspicion

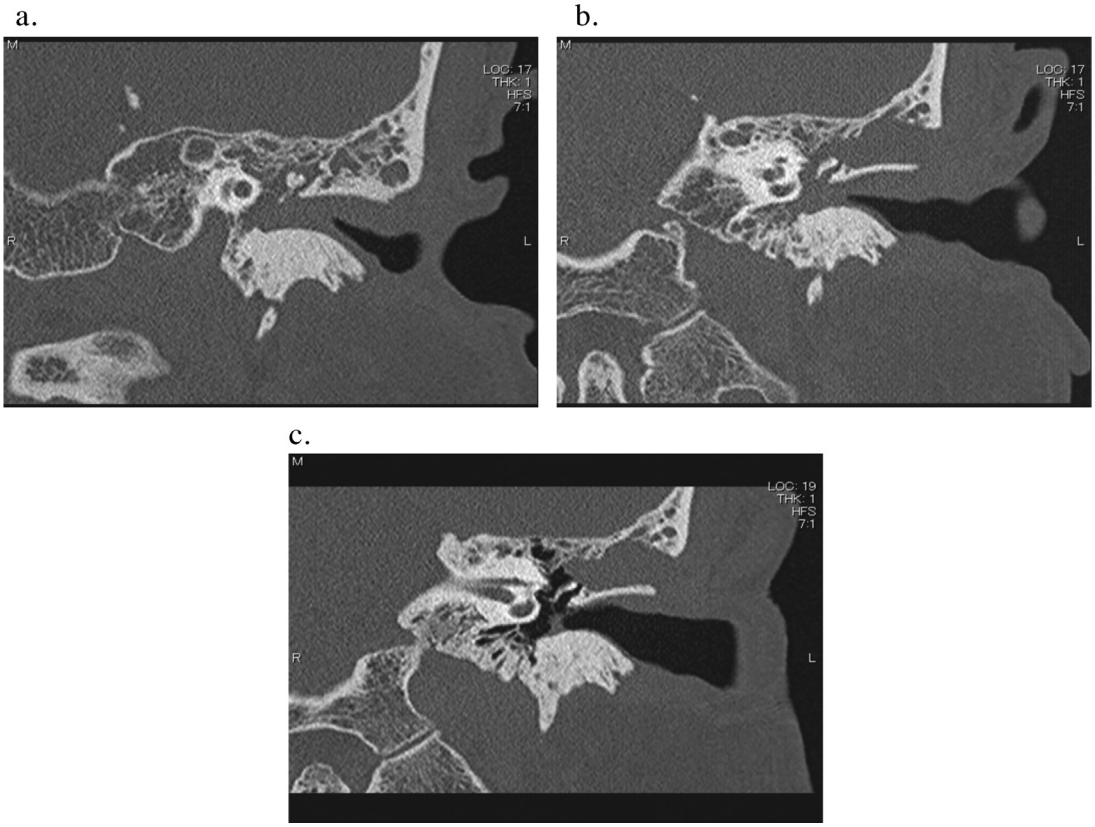

Fig. 2. (a) CT scan before mastoidectomy: the mastoid and tympanic cavities are diffusely clouded. (b) CT scan performed when paralysis of the left abducens nerve was observed. The mastoid and tympanic cavities are fully clouded without any bony deficit. A mastoidectomy was performed. (c) CT scan after voriconazole therapy showing improved aeration of the tympanic cavity.