



Pyogenic Ventriculitis Caused by Cholesteatoma

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The case report highlights the potential intracranial complications associated with otitis media, particularly cholesteatoma and mastoiditis, including meningitis, brain abscesses, epi- and subdural empyema, and sigmoid sinus thrombophlebitis. Pyogenic ventriculitis (PV), an inflammation of the ependymal lining of cerebral ventricles, is a rare but serious complication often secondary to factors such as meningitis, cerebral abscesses, or neurosurgery. The case presented involves a 52-year-old male with a history of head trauma who developed PV secondary to cholesteatoma. Despite initial challenges in diagnosis, timely antibiotic therapy and surgical intervention resulted in successful treatment. The study emphasizes the importance of considering ear diseases as potential sources of intracranial infection, particularly in cases of delayed symptom presentation or history of head trauma.

Keywords: cholesteatoma; intracranial complication; mastoidectomy; pyogenic ventriculitis

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Introduction

Otitis media, including cholesteatoma and mastoiditis, can lead to intracranial complications such as meningitis, brain abscesses, epi- and subdural empyema, and sigmoid sinus thrombophlebitis (Kuo et al. 2015; Koshihara et al. 2022). Pyogenic ventriculitis (PV) is an inflammation of the ependymal lining of the cerebral ventricles (Hall and Munakomi 2024). It is usually secondary to meningitis caused by bacteria and viruses, cerebral abscess with intraventricular rupture, complications of neurosurgery, etc. (Tunkel et al. 2017). This disease presents several symptoms, such as fever, headache, nuchal rigidity, or decreased mental status, and could lead to poor neurology, hydrocephalus, and death if not treated (Shang et al. 2018). A few case reports of PV also mention otitis media or mastoiditis (Hatakeyama et al. 2014; Tokimura et al. 2019; Kiamos et al. 2022). There has been only one reported case of cholesteatoma causing PV in the literature (Feinberg et al. 2022). Additionally, previous case report mainly describes neurological findings and do not provide details regarding the otological and otosurgical findings.

We encountered an extremely rare case of middle ear cholesteatoma leading to PV, who was treated with a com-

ination of surgery and antimicrobials.

Case Presentation

A 52-year-old male presented to department of general medicine in our hospital with posterior cervical pain, fever, and anorexia. His medical history included craniotomy due to intracranial hemorrhage and cerebral contusion, and right temporal bone fracture, resulting from a traffic accident at the age of 18, leading to subsequent complications such as right hearing loss, left hemiparesis, right blepharoptosis, and midline fixation of the right eyeball. Additionally, he has a history of symptomatic epilepsy and glaucoma. He received supplemental fluid due to suspected crowned dens syndrome or meningitis. Computed tomography (CT) imaging of the head and neck was inconclusive. However, due to persistent fever, he was transferred to the Department of Neurology on the 9th day of hospitalization. Meningeal irritation signs were noted, and cerebrospinal fluid (CSF) examination revealed elevated mononuclear cell count and mild decrease in glucose levels. Blood and CSF cultures yielded negative results. Intravenous amphotericin B and acyclovir were initiated. However, on the 15th day of hospitalization, he developed with vomiting, worsening fever, and deteriorating level of consciousness (Glasgow Coma

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Scale of 12; eye-opening: 3, best verbal response: 4, and best motor response: 5). Brain magnetic resonance imaging (MRI) fluid attenuated inversion recovery (FLAIR) image showed linear high-signal areas on the brain surface from bilateral temporal lobes to the right parietal and occipital lobes, that suggested meningitis. CSF analysis showed an increase in polymorphonuclear cells with decreased glucose levels, prompting treatment adjustment to meropenem, vancomycin, and dexamethasone for bacterial meningitis. Brain MRI diffusion weighted image (DWI) on the 19th day revealed new high-signal lesions in the bilateral subventricular horns of the lateral ventricles (Fig. 1b).

On the 23rd hospital day, the patient was referred to the otolaryngology department due to the suspicion of an abscess or mastoiditis. Otoloscopic examination revealed swollen right external auditory canal (EAC), obscured tympanic membrane, and debris-filled EAC with otorrhea. Biopsy of the EAC tissue confirmed cholesteatoma. The otorrhea culture showed *Staphylococcus aureus*. CT of the right temporal bone showed bone erosion of EAC, defect of the tympanic tegmen, and soft tissue shadows from EAC to

the mastoid cavity (Fig. 2a, b). We did not perform a pre-operative hearing test because the patient's level of consciousness deteriorated. He was diagnosed with middle ear cholesteatoma, and mastoidectomy with canal wall down approach was performed on the 27th day. Intraoperatively, cholesteatoma and granulation were found in the mastoid cavity, epitympanum, and mesotympanum (Fig. 3a). Despite dural exposure in the epitympanum, there was no evidence of cerebrospinal fluid leakage or defect. Bone-wax-like tissue was observed on the tegmen side of the mastoid cavity (Fig. 3b), suspected to be residue from prior surgery. Facial canal defect was noted, but the nerve integrity monitor (NIM) responded well. Ossicles were largely absent, with remnants resembling a stapes footplate (Fig. 3c). All the tympanic membrane was missing except for the lower annulus tympanicus. The ossicles were not manipulated, and the meatoplasty was performed by applying the temporal fascia with fibrin glue. Postoperatively, ear lavage and an ofloxacin drop were alongside antibiotics. Consciousness improved (Glasgow Coma Scale of 15) on the 32nd day, with resolution of MRI lesions on the 33rd

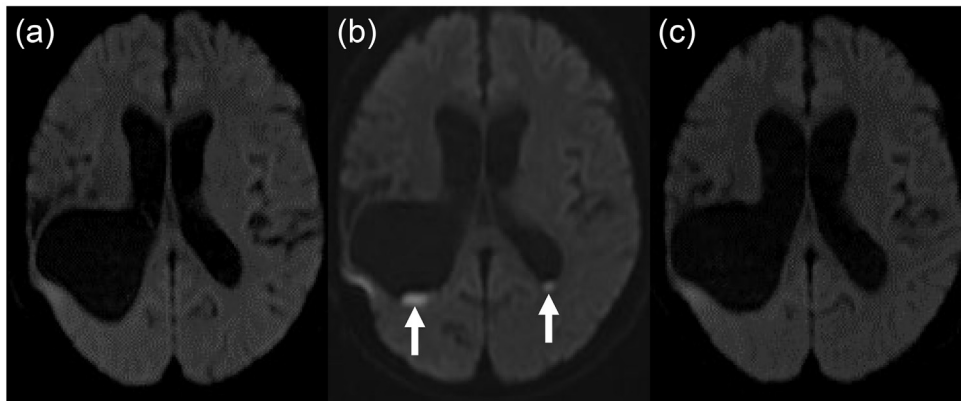


Fig. 1. Axial view of magnetic resonance imaging (diffusion-weighted image) of brain. (a) On the 15th day. (b) On the 19th day, white arrows show high-signal new debris like lesions in the bilateral subventricular horns of the lateral ventricles. (c) On the 33rd day, the high-signal lesions resolved.

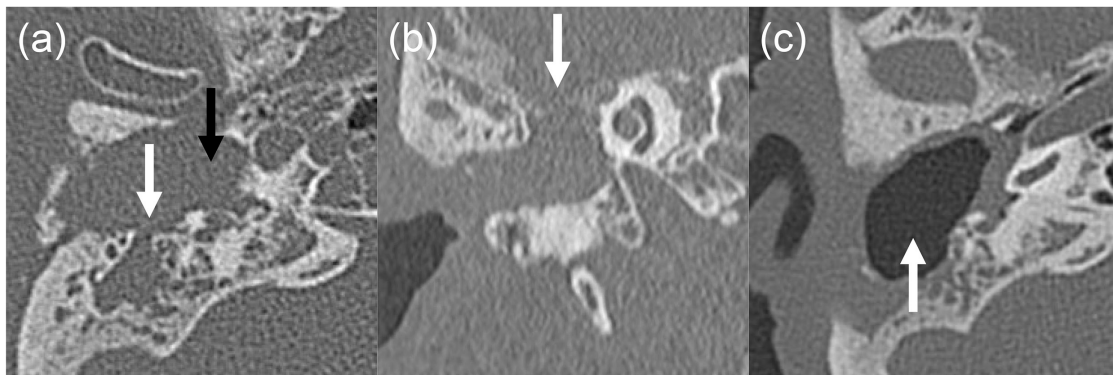


Fig. 2. Computed tomography imaging of right temporal bone. (a) Axial view on the 23rd day: Bone destruction of external auditory canal (EAC) (white arrow), and soft tissue shadows from EAC to the mesotympanium (black arrow) and the mastoid cavity were observed. (b) Coronal view on the 23rd day: Defect of tympanic tegmen (white arrow) was observed. (c) Axial view on the 68th day: The external and middle ear cavity were aerated (white arrow) after surgery.

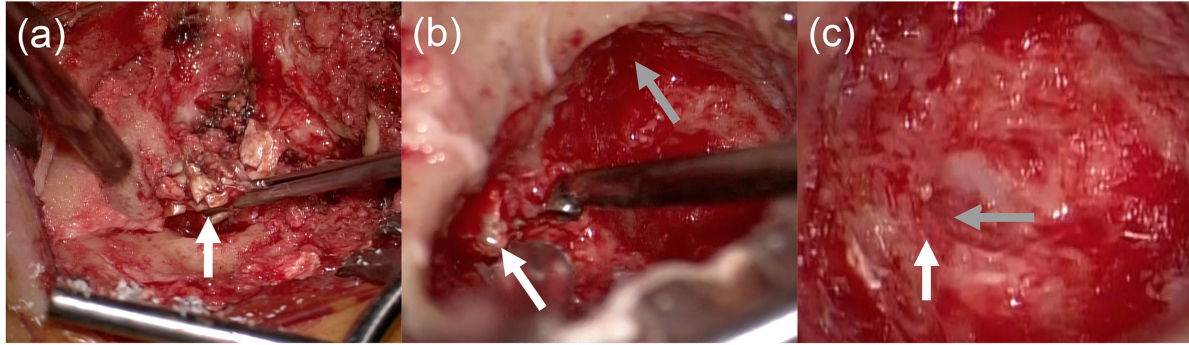


Fig. 3. Surgical findings.

(a) Mastoid cavity was fulfilled with cholesteatoma and granulation (white allow). (b) A bone wax-like tissue on the tegmen side of the mastoid cavity (white allow) and a dural exposure in the epitympanum (gray allow) were observed. (c) After removing cholesteatoma and granulation, facial nerve (white allow) and the foot plate of stapes (gray allow) were observed.

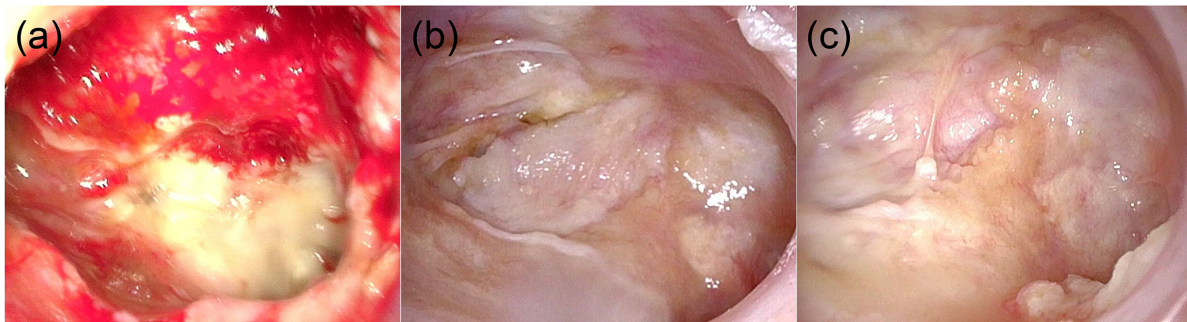


Fig. 4. Endoscopic findings of right ear after surgery.

(a) On the 40th day (b) On the 68th day (c) On the 117th day.

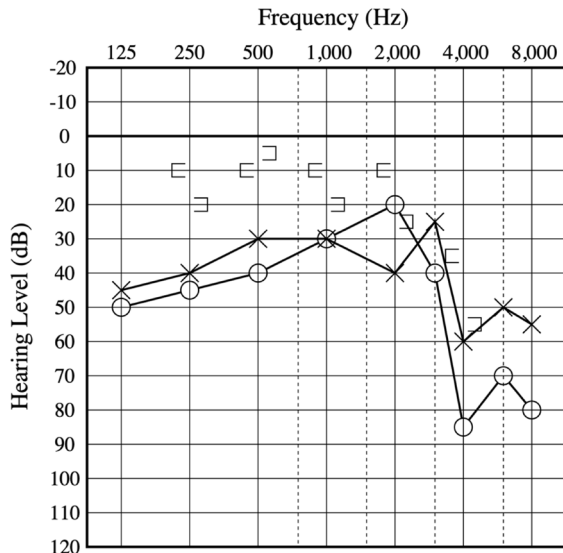


Fig. 5. The result of hearing test on the 61st day.

The average hearing level of the right ear was 30.0 dB (averages of 500, 1,000, and 2,000 Hz).

day (Fig. 1c). Postoperative ear findings (Fig. 4) and CSF examination gradually improved. Audiogram on the 61st day showed 30.0 dB (averages of 500, 1,000, and 2,000 Hz) in the right ear, comparable to the left ear (Fig. 5). Antibiotics were discontinued on the 65th day, and the

patient was transferred to a convalescent facility on the 73rd day. This study was approved by the Iwate Medical University Hospital Institutional Review Board.

Discussion

We encountered PV as an intracranial complication of cholesteatoma. Cholesteatoma may progress silently until it reaches a critical size, posing a risk to intratemporal structures. This can result in various intra- and extracranial complications, including meningitis, brain abscesses, epidural abscesses, septic cavernous sinus thrombosis, and acute mastoiditis with a subperiosteal abscess (Prasad et al. 2013; Kuo et al. 2015). While headache, vomiting, and fever are atypical presentations of cholesteatoma, their occurrence suggests potential intratemporal or intracranial complications (Kuo et al. 2015). PV involves inflammation of the ependymal lining of the cerebral ventricles and can be secondary to various factors such as meningitis, cerebral abscess with intraventricular rupture, trauma, catheter-related issues, CSF leaks, or complications of neurosurgery (Tunkel et al. 2004; Hall and Munakomi 2024). When meningitis does not respond to antibiotics, PV should be considered (Hall and Munakomi 2024). Risk factors include compromised immunity (cancer, HIV, diabetes, alcoholism, etc.) and virulence of the causative organism (Hall and Munakomi 2024). *Streptococcus pneumoniae*

and gram-negative rods are commonly implicated in PV following head trauma, while oral flora bacteria (*Streptococcus pneumoniae*, *Haemophilus influenzae*, and *Streptococcus pyogenes*) are associated with infections in patients with skull base fractures and persistent CSF leaks (Martin et al. 2018). *Staphylococcus aureus*, which typically lives on the skin's surface, was detected in our case. Watanabe et al. (2004) previously described two cases of middle ear cholesteatoma-related brain abscesses, in which *Staphylococcus aureus* was identified through bacterial culture. PV lacks a standardized diagnostic criteria, and patients typically present with fever and meningism symptoms such as nuchal rigidity, headache, photophobia, decreased mental status, seizures, or moribund (Hall and Munakomi 2024). Early diagnosis is crucial for appropriate management, often aided by imaging and CSF examination (Fukui et al. 2001). The presence of ventricular debris in diffusion-weighted MRI is a common imaging finding in PV (Fukui et al. 2001). Antimicrobial therapy, typically comprising vancomycin and an anti-pseudomonal beta-lactam, is essential (Tunkel et al. 2004, 2017). Treatment duration may vary based on clinical response.

Several reports describe meningitis or brain abscesses as intracranial complications of otitis media or mastoiditis, with or without cholesteatoma (Penido Nde et al. 2005, Sun and Sun 2014). Feinberg et al. (2022) reported a case of a 57-year-old male with brain abscess and PV, where mastoidectomy was performed to address cholesteatoma. However, details regarding cholesteatoma progression or associated complications were not provided. The lack of a widely accepted diagnostic criteria for ventriculitis may contribute to the scarcity of research on its association with cholesteatoma. In our case, PV was not initially detected, and the patient did not report any aural symptoms. Given his history of hearing loss following head trauma, it is possible that subjective symptoms were not prominent until the condition worsened considerably. Early identification and elimination of the infection source are critical.

Conclusion

In conclusion, we report a case of meningitis and subsequent PV secondary to cholesteatoma. Despite delays in identifying the infection source due to the patient's history of head trauma, prompt antibiotic therapy and surgical intervention led to successful treatment. Although PV as a consequence of cholesteatoma is rare, it underscores the importance of considering ear disease as a potential source of infection for timely diagnosis and management of intracranial infections.

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Conflict of Interest

The authors declare no conflict of interest

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