

Case Report

# Otogenic Pneumocephalus Associated with a Ventriculoperitoneal Shunt

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Otogenic pneumocephalus is a condition of intracranial air originating from the middle ear or mastoid air cells. This communication between the intracranial cavity and the pneumatic cavities is usually associated with trauma after cranial fractures or iatrogenic trauma. We present a rare case of otogenic pneumocephalus arising in the left posterior fossa from well-pneumatized mastoid air cells. The patient complained of roaring tinnitus that developed 29 months after ventriculoperitoneal shunt insertion due to brain tumor surgery. High resolution computed tomography scan of the temporal bones revealed a large pneumocephalus below the left tentorium, and a bony dehiscence route was clearly identified in a sagittal view. A left mastoidectomy with preservation of the posterior wall of the external auditory canal was performed, and the expected bony dehiscence site was identified in the posterior fossa dura plate, just posterior to the posterior semicircular canal, below the Donaldson's line. This communication was sealed with a temporalis muscle plug from the deep temporalis muscle fascia and bone dust. Pneumocephalus may be caused by negative intracranial pressure in a patient with very well-pneumatized mastoid bone, and it can be a possible cause of 'wind-like' sound in the ear.

**Key Words.** *Pneumocephalus, Ventriculoperitoneal shunt*

## INTRODUCTION

Pneumocephalus is defined as the presence of air or gas in the intracranial cavity. In the absence of intracranial tumors or infection, it is usually caused by head and facial trauma or surgical interventions, and more rarely, infection of pneumatized cavities, paranasal tumors, or radionecrosis (1-4). In other cases with no pathologic antecedent events, pneumocephalus may be exceptionally formed by anatomic malformations located in the pneumatic cavities, or spontaneously (1, 3, 4). Pneumocephalus following ventriculoperitoneal (VP) shunt is a rare but well-described complication. The first case of air entry via a skull base defect,

not via direct distal shunt catheter, was reported by Pitts et al. in 1975 (5).

Herein, we report a rare case of delayed pneumocephalus presenting 30 months after insertion of the VP shunt secondary to brain tumor surgery and related to the bony dehiscence in well-pneumatized mastoid cavity. The initial manifestation was a roaring, 'wind-like' sound in the left ear, and the case was managed successfully by surgical repair.

## CASE REPORT

A 33-yr-old man was referred for the management of pneumocephalus in the left posterior fossa, with the symptom of intermittent, roaring, 'wind-like' noise in his left ear that started 29 months after VP shunt surgery, persisting for 8 months in total. He underwent right parietotemporal craniotomy, removal of a choroid plexus papilloma in the right ventricle, and VP shunt insertion in the left side due to hydrocephalus at the age of 30.

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After tumor removal and craniotomy, he had intermittent seizures but the neurological examinations performed by a neurologist were normal. Anticonvulsant was prescribed and brain computed tomography (CT) showed no evidence of pneumocephalus. Thirty months after shunt insertion, a large pneumocephalus was noted in the left posterior fossa, and consultation was done to our department. On clinical examination, there were no abnormal findings in his neurologic status, otologic examination, or in shunt function. He had no history of ear infection, otologic surgery, or barotrauma. His hearing was normal in pure tone audiometry. A high resolution computed tomography (HRCT) scan and contrast-enhanced magnetic resonance image (MRI) of temporal areas revealed about a 4×3 cm pneumocephalus adjacent to an apparent bony defect in the posterior fossa of the left mastoid bone (Fig. 1). There was no radiologic evidence of mastoiditis, otitis media, trauma, or neoplasm.

During the left mastoidectomy with preservation of the posterior wall of the external auditory canal (EAC), mastoid air cells were well-pneumatized and large, without any evidence of infl-

ammation, trauma, or neoplasm. Some mucosa along the mid-portion of the posterior fossa was swollen and conglomerated, so it was scraped off with cotton ball. After that, a definite 4×4 mm bony defect was noted along the posterior fossa bony plate, anterior to the sigmoid sinus, just posterior to the posterior semicircular canal, below the Donaldson's line (Fig. 2). The dura was identified through this opening, but there was no cerebrospinal fluid (CSF) leakage when the dura was pressed with a micro-elevator. Some temporalis muscle and deep temporalis muscle fascia were inserted to seal the defect site and a mixture of bone dust and fibrin glue was applied.

After surgery, the symptoms gradually improved and hearing was preserved. He was discharged 8 days after the operation in a satisfactory condition. Follow-up HRCT scan of temporal bone was taken one month later, and there was no remnant air in the intracranial cavity (Fig. 3A). Brain CT taken by the Neurosurgery Department 12 months postoperatively confirmed complete resolution of the pneumocephalus (Fig. 3B). Tinnitus decreased and was not troublesome any more.

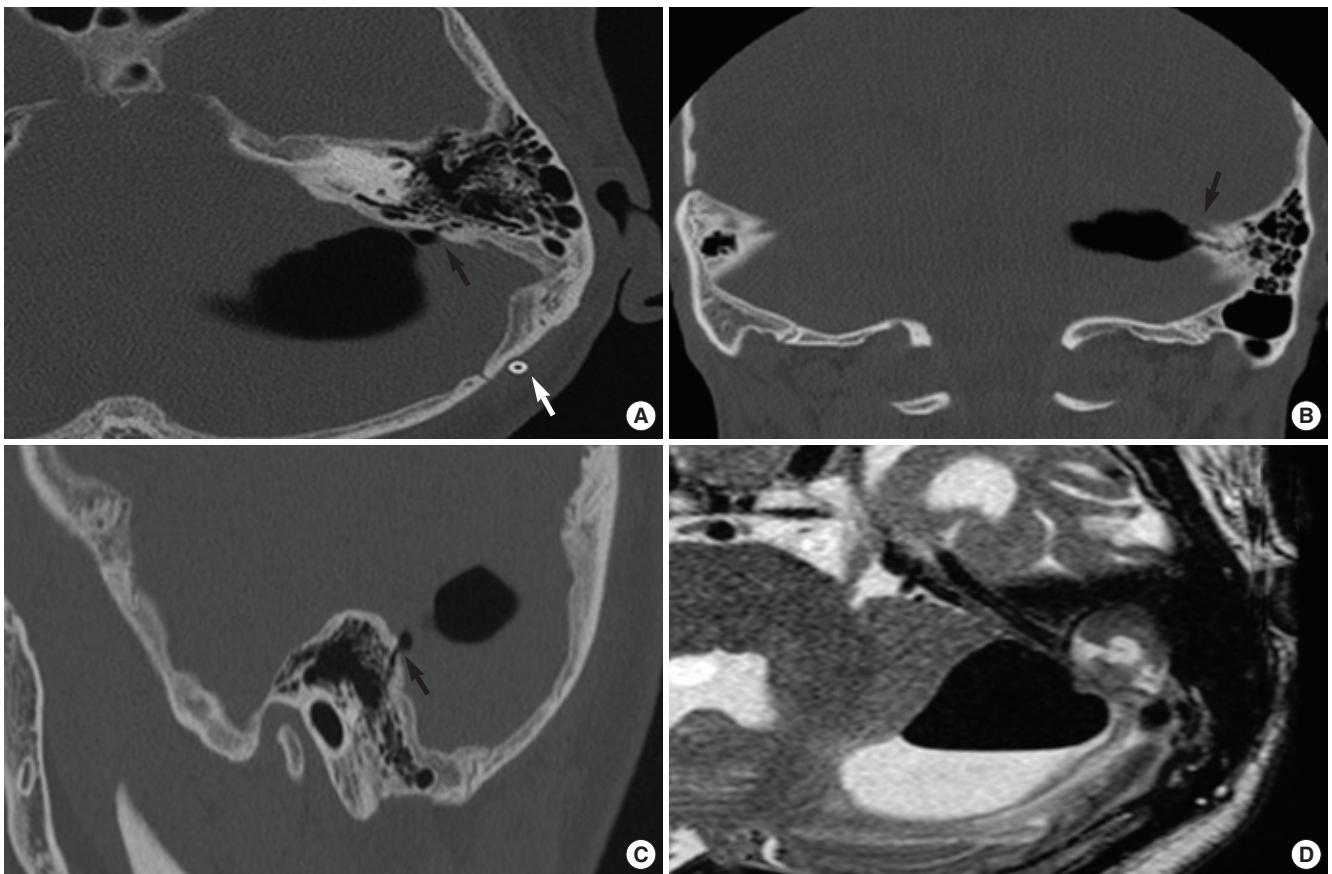


Fig. 1. High resolution CT scan of the temporal bones. (A) Axial finding shows a large pneumocephalus adjacent to left posterior fossa (black arrow) and the well-pneumatized temporal bone. Previously inserted ventriculoperitoneal shunt is noted (white arrow). (B) Coronal finding suggests the possible communication (arrow) between the pneumocephalus and the air cells of the mastoid. (C) Sagittal finding shows the definite bony defect (arrow) in the left temporal bone. (D) Axial T2-weighted temporal bone MRI demonstrates a 3.7×3.9×2 cm large cystic lesion with air fluid level inferior to left tentorium.

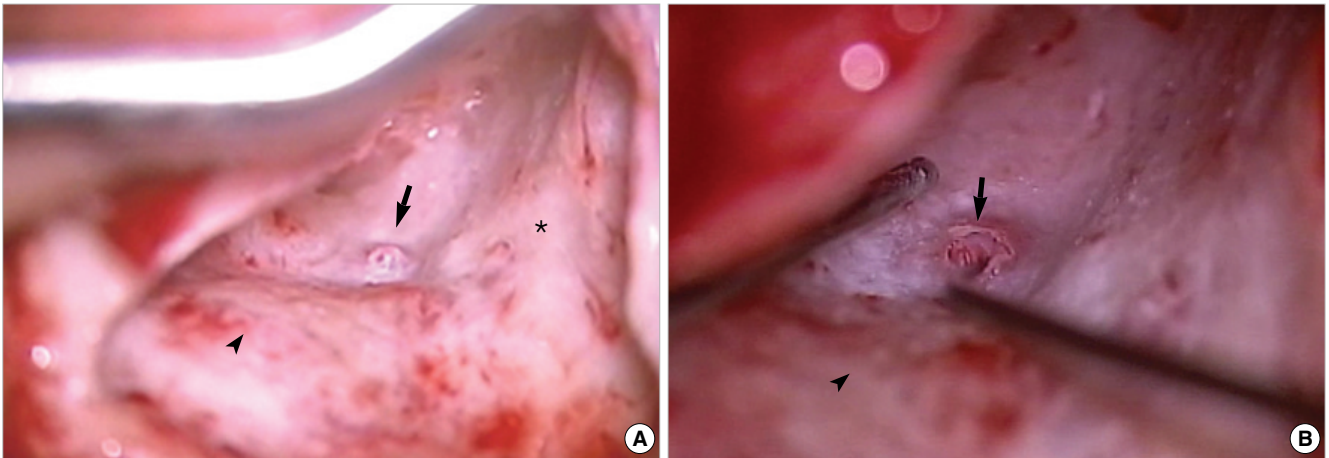


Fig. 2. Intraoperative findings. (A) There was swollen, conglomerated mucosa (arrow) was noted in the posterior fossa bony plate, anterior to the sigmoid sinus (arrowhead), inferior to the middle fossa dural plate (asterisk). (B) The defect (arrow) was about 4 × 4 mm size and there was no evidence of cerebrospinal fluid leak. Sigmoid sinus (arrowhead).

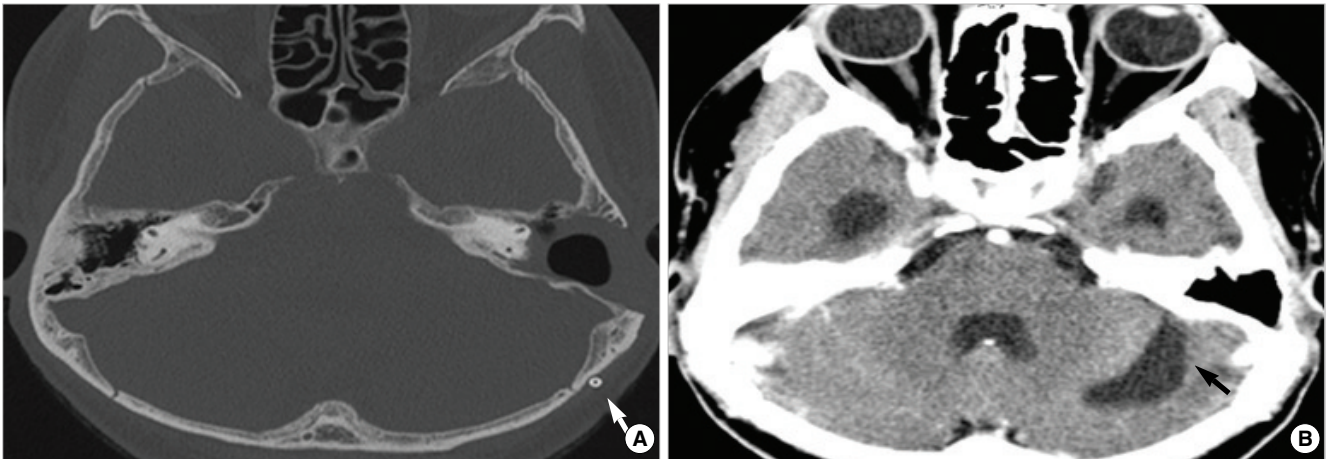


Fig. 3. (A) Follow-up Temporal bone CT scan 1 month after the surgery. There is no evidence of recurrent or remnant pneumocephalus. This shows left canal-up mastoidectomy state and ventriculoperitoneal shunt placement (arrow). (B) Follow-up brain CT scan 12 months after the surgery. There is no evidence of pneumocephalus, and the space is replaced with cerebrospinal fluid (arrow).

## DISCUSSION

Otogenic pneumocephalus associated with VP shunt is a rare event. Ruge et al. (6) summarized a few cases of pneumocephalus associated with VP shunt, but these included cases who had been treated for trauma and who had directly retrograde airflow in shunts that had perforated the colon or bronchus postoperatively. Pneumocephalus related to shunt surgery can develop secondary to a combination of the siphon effect of shunting and a skull base defect (5). Excessively negative intracranial pressure by the siphon effects of shunts, in certain situations, allows air ingress to fill the vacuum by the skull base defect. The bone defect may be congenital but more often occurs as a result of bony erosion by long-standing raised intracranial pressure by hydrocephalus (5, 6). The common sites of congenital skull base defects are anterior fossa, followed by the middle fossa (5) and tegmen

tympani in temporal bone (7). The bone defect more often occurs in hyper-pneumatized mastoid cavity with thin intercellular septa, thus facilitating its rupture with increased endotympanic pressure (1, 3, 5).

In this case, the patient had removal of a choroid plexus tumor in the ventricle via a craniotomy and VP shunting for hydrocephalus. His neurosurgical history could indicate neoplastic fistula, but the primary site of the tumor was far from the identified pneumocephalus, and no evidence of tumor was found adjacent to the bone defect site. Moreover, posterior fossa is a rare site of pneumocephalus related to VP shunt, with the first case reported by Kanner et al. (8) in 2000. This pneumocephalus developed two years later after shunt insertion, so the chronic negative intracranial pressure by shunt may cause the air leak through the bony dehiscence site in the posterior fossa. The mucosa around the fistula site was slightly swollen and conglomerated due to

long-standing irritation, so it was easy to find the defect site. This bone defect could have been congenital and/or the result of previous hydrocephalus. During mastoidectomy, the absence of CSF leakage in the mastoid cavity may have been due to the presence of a ball-valve mechanism over the fistula by brain tissue or a dura leaflet or due to the persistent pressure gradient caused by the shunt.

Clinical presentations vary depending on the location and mass effect of the pneumocephalus (3, 4). Jooma and Grant described the splashing sound inside the head, 'bruit hydroaérique', as a pathognomic symptom of pneumocephalus (5), and acoustic phenomena like this case have been reported (4). In the posterior fossa pneumocephalus, if intracranial pressure increases by pneumocephalus and tension pneumocephalus develop, headache is the most common symptom (1), and neurologic symptoms such as vomiting, blurring vision, dizziness, syncope, aphasia, hemiplegia, ataxia can also occur (4, 5, 9). In this case, a non-specific wind-like, roaring sound of the left ear was the only symptom of pneumocephalus. His intermittent seizure and dizziness existed prior to shunt insertion, and it occurred intermittently after sealing of the bone defect.

Pneumocephalus can be treated by surgical repair to identify the communication and seal it (1, 4). Otogenic pneumocephalus can be managed more successfully by surgical repair without craniotomy or duroplasty (5). Autologous materials such as cartilage, free fascia, temporal muscle (1) and bone dust, fibrin glue, and/or bone wax can be used for sealing. But if there are difficulties in surgical approach or no definite fistula site is identified, puncture of intracranial air and closure of mastoid air cells or closure of the Eustachian tube may be alternative options. In these situations, conservative management relieving intracranial pressure and preventing the brain from infection (2, 4) are important. For VP shunts, temporary shunt externalization or ligation with an anti-siphon device are sometimes needed, and insertion of a high pressure valve or anti-siphon device is considered a prima-

ry measure (3).

In conclusion, although extremely rare, an otogenic posterior fossa pneumocephalus associated with VP shunt may be a possible diagnosis in patients with acoustic phenomena and other non-specific neurological symptoms. Fine-cut reformatted CT scan is a useful diagnostic aid, but a precise localization of the fistula may be difficult. In the present case, we were able to localize the site of the fistula by HRCT and satisfactorily managed this patient by sealing the defect easily through a transmastoid extracranial approach. The development of spontaneous or secondary pneumocephalus must be considered and monitored in neurosurgical patients with shunt insertion.

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