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Case Report

Spontaneous cerebrospinal fluid otorrhea and pneumocephalus on the contralateral side of the previous cranial surgery

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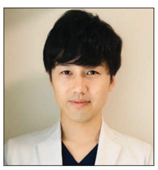

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ABSTRACT

Background: Cerebrospinal fluid (CSF) leaks and pneumocephalus commonly occur due to head trauma or surgical procedures. Spontaneous CSF (sCSF) leaks, however, occur without any clear etiology and are relatively uncommon.

Case Description: An 84-year-old woman presented with the right-sided otorrhea. The patient had a history of a ventriculoperitoneal shunt placement following a subarachnoid hemorrhage treated by clip ligation of a left-sided ruptured cerebral aneurysm 7 years before presentation, with shunt catheter ligation after evidence of intraventricular pneumocephalus 6 years before presentation. At admission, computed tomography (CT) imaging of the head showed enlargement of the lateral ventricles, a right mastoid fluid collection, and a defect of the superior wall of the right petrous bone. We performed a right temporal craniotomy for the repair of the CSF leak. Intraoperatively, it was noted that temporal lobe parenchyma herniated into the mastoid air cells through lacerated dura and a partially defective tegmen mastoideum. The leak point was successfully obliterated with a pericranial graft and reinforced by a collagen sheet and fibrin glue. There was no recurrence of otorrhea postoperatively.

Conclusion: This report presents a very unique case of a patient with a CSF leak and pneumocephalus occurring on the contralateral side of a previous cranial surgery. We accurately identified the defect site with CT imaging and repaired the CSF leak by temporal craniotomy. Awareness of the mechanisms by which sCSF leaks can be caused by aberrant arachnoid granulations is imperative for neurosurgeons.

Keywords: Aberrant arachnoid granulation, Cerebrospinal fluid otorrhea, Pneumocephalus, Spontaneous cerebrospinal fluid leak

INTRODUCTION

Cerebrospinal fluid (CSF) leaks and pneumocephalus occur due to abnormal communication between the subarachnoid space and the adjacent paranasal sinuses.[1] If left untreated, these conditions can lead to life-threatening meningitis, brain infections, or seizures. Although most cases result from a surgical procedure or skull fracture, certain cases have no obvious causes.[7] Here, we describe a unique case of a patient with delayed right-sided spontaneous CSF (sCSF) otorrhea and pneumocephalus occurring after ventriculoperitoneal (VP) shunt placement following clip ligation for a left-sided aneurysmal subarachnoid hemorrhage (SAH).

CASE PRESENTATION

An 84-year-old woman presented to our hospital complaining of the right-sided clear otorrhea. The patient had a history of a VP shunt placement for hydrocephalus after a clip ligation for a left internal carotid-posterior communicating artery aneurysmal SAH 7 years before presentation. At 6 years before presentation, she had undergone ligation of the shunt catheter after complaining of the right-sided tinnitus with evidence of intraventricular pneumocephalus [Figure 1]. Since then, she had been asymptomatic and had no known history of head trauma. Computed tomography (CT) imaging of the head at admission showed enlargement of the lateral ventricles, a right mastoid fluid collection, and a defect of the superior wall of the right petrous bone [Figures 2-5].

Figure 1: Computed tomography imaging of the head 6 years before presentation showing intraventricular pneumocephalus and the VP shunt catheter (white arrow).

Figure 2: Computed tomography imaging of the head showing enlargement of the inferior horn of the lateral ventricles (black dotted circles).

Otoscopy revealed bilateral partially perforated tympanic membranes.

Three days after admission, we performed a right temporal craniotomy for a CSF leak repair. A lumbar drain was placed preoperatively. When the right temporal lobe was retracted, we identified the site of the leak, and it was noted that temporal lobe parenchyma herniated into the mastoid air cells through lacerated dura and a partially defective tegmen mastoideum [Figures 6 and 7]. We interrupted the herniated parenchyma using bipolar forceps and tacked the pericranial graft to the dura with 7-0 PRONOVA® Poly

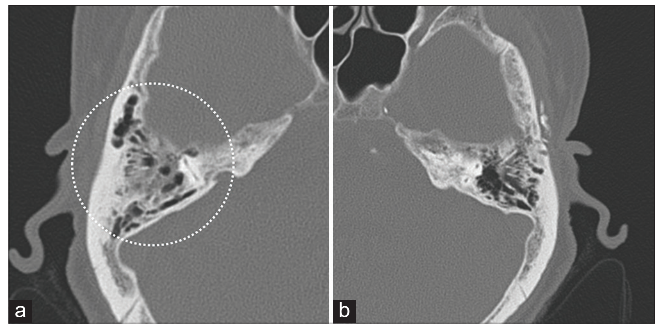

Figure 3: Axial temporal bone computed tomography imaging showing a mastoid fluid collection (white dotted circle). (a), right side and (b), left side.

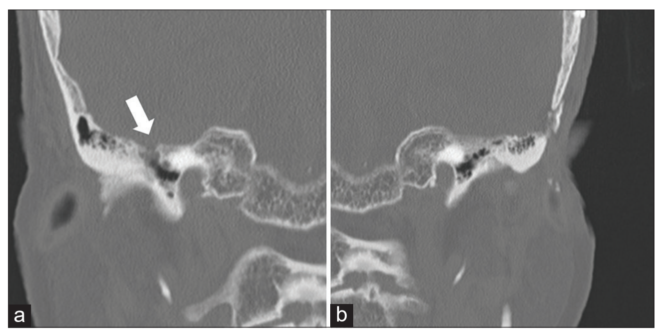

Figure 4: Coronal temporal bone computed tomography imaging showing a mastoid fluid collection and a defect of the petrous bone (white arrow). (a), right side and (b), left side.

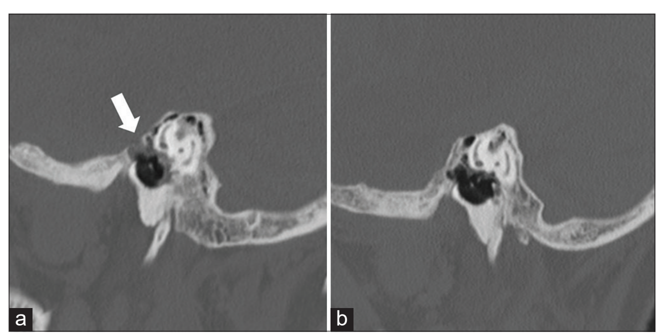

Figure 5: Sagittal temporal bone computed tomography imaging showing a mastoid fluid collection and a defect of the petrous bone (white arrow). (a), right side and (b), left side.

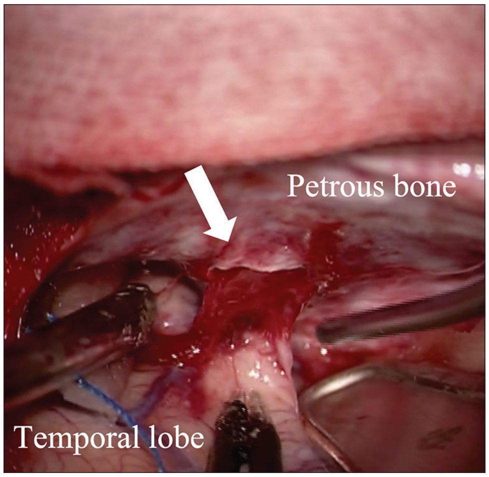

Figure 6: Intraoperative microscopic image showing a temporal lobe parenchymal herniation into the mastoid air cells through lacerated dura and a defect site of petrous bone (white arrow).

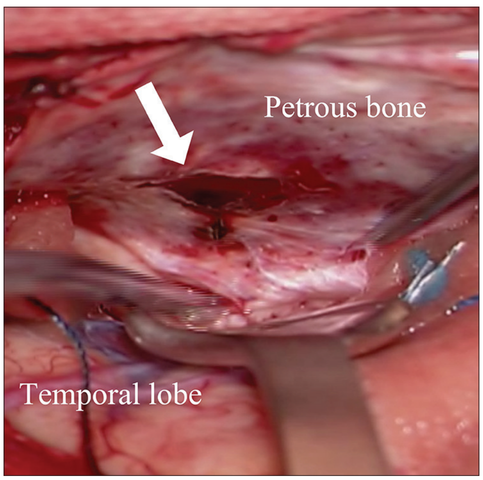

Figure 7: Intraoperative microscopic image showing lacerated dura after herniated temporal lobe was interrupted (white arrow).

suture (Ethicon, Inc., Somerville, NJ) to cover the defect site [Figure 8]. A collagen sheet was placed over the pericranial graft, and fibrin glue was injected to reinforce the closure. An abdominal fat graft was harvested to obliterate the defect site extradurally [Figure 9]. Furthermore, a temporalis muscle flap was sutured to the dura to close the extradural space [Figure 10]. The lumbar drain was removed on postoperative day 5. The postoperative course was uneventful, and there was no recurrence of CSF otorrhea. CT imaging of the head performed at 2 months after the operation revealed resolution of the mastoid fluid collection and disappearance of the defect site of the petrous bone [Figure 11].

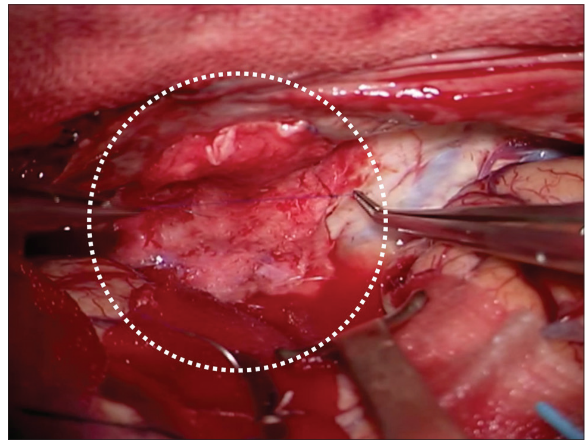

Figure 8: Intraoperative microscopic image showing the defect site covered by the pericranial graft (white dotted circle).

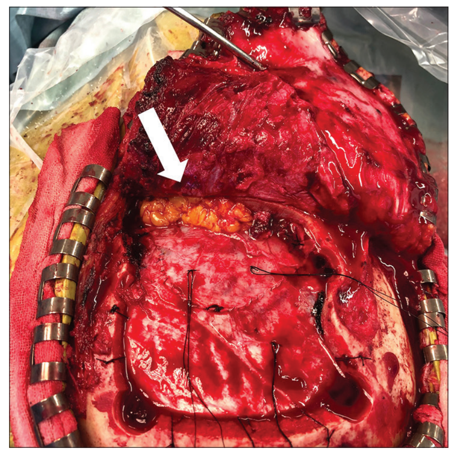

Figure 9: Intraoperative image showing the extradural space closure using an abdominal fat graft (white arrow).

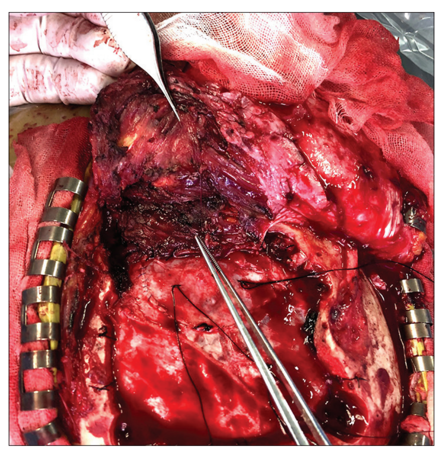

Figure 10: Intraoperative image showing a temporalis muscle flap sutured to the middle cranial fossa dura.

DISCUSSION

CSF leaks result from abnormal communication between the subarachnoid space and the adjacent paranasal sinuses through osteodural defects. The etiologies of CSF leaks are commonly classified as either congenital or acquired.

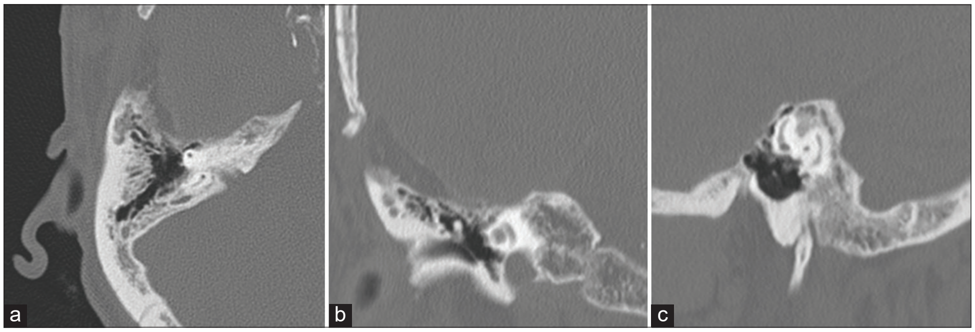

Figure 11: Postoperative temporal bone CT imaging showing resolution of the mastoid fluid collection and disappearance of the defect site of the petrous bone. (a), axial, (b), coronal and (c), sagittal.

Acquired CSF leaks can be further subdivided into traumatic, nontraumatic, and spontaneous subtypes.[1] sCSF leaks occur without any clear causes, including trauma, surgery, or other inciting events.[7] sCSF leaks were previously presumed to be rare; however, recent research has reported a relatively higher incidence, reaching approximately 20% of all CSF leaks.[2] This patient's CSF leak occurred on the contralateral side of the previous clip ligation surgery; therefore, we determined that the prior surgical procedure was not related to the etiology of this CSF leak and that this patient had experienced a sCSF leak.

Although increased CSF leaks are thought to occur spontaneously, certain factors may predispose patients to this type of leak. It has been demonstrated that aberrant arachnoid granulations (AGs) that are surrounded by thin bony surfaces, including the cribriform plate, tegmen mastoideum, or sella turnica, can penetrate the adjacent bone to form fistulas.[3] AGs are normally projections of the arachnoid membrane into the dural venous sinuses; however, AGs are not associated with venous structures in the middle cranial fossa (MCF) and can, therefore, enlarge by eroding surrounding bony structures.[5] One study reported that the size of AGs is related to the extent of bone erosion, with patients who have AGs larger than 3 mm^3 found to have bone erosion.[4] The resultant bony erosion can lead to communication of the CSF space with the adjacent paranasal sinuses.[4,5]

In this case, we consider that subclinical osteodural defect has been existed in the MCF, and there was an aberrant AG, which presented as a parenchymal herniation adjacent to the defect [Figure 6]. Then, the VP shunt generated a negative pressure, which may have enabled air entry from the mastoid air cells into the lateral ventricles, resulting in intraventricular pneumocephalus. After the shunt catheter ligation, the gradually increasing intracranial pressure and the constant pulsation of the CSF may have eventually resulted in the erosion of the adjacent petrous bone.[6] In addition, a certain degree of localized subclinical meningitis may have led to cerebral tissue deterioration and adhesion to the adjacent bone, resulting in fistula formation between the mastoid air cells and the inferior horn of the lateral ventricles.

During repairs of temporal CSF leaks, either the MCF approach or the transmastoid approach are typically employed. The MCF approach, which facilitates extensive exposure of the MCF, a multilayer repair, and preservation of hearing, is preferred by many neurosurgeons and was adopted in this case. The transmastoid approach, which is an otological procedure, is less invasive and easier to be performed; however, the entire floor of the MCF can be difficult to visualize.[6] Perez *et al*. reported an 86% success rate for transmastoid repairs with mastoid air cell obliteration using an abdominal fat graft in uncomplicated sCSF leak cases.[8] As there is no clear consensus regarding the surgical approach in these cases, it depends on the surgeon's preference. Although neurosurgeons generally prefer the MCF approach to repair the temporal bone CSF leaks, a transmastoid approach should also be considered as an alternative option. In fact, it might be reasonable to adopt the transmastoid approach in cases of a single defect and to reserve the MCF approach for recurrent cases or for cases of multiple large defects.

CONCLUSION

In this patient, we successfully repaired a temporal bone CSF leak due to an aberrant AG by performing a temporal craniotomy. It is imperative for neurosurgeons to be aware of the mechanism by which sCSF leaks can be caused by aberrant AGs, and the surgical options for repairs of temporal bone CSF leaks.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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