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Case Report

A Case of Intraosseous Petrous Bone Arteriovenous Fistula Complicated by Transient Worsening of Ipsilateral Hearing Following Transvenous Coil Embolization

Daisuke Satoh,[1] Tetsuo Sasaki,[2] Takehiro Yako,[1] Kazuo Kitazawa,[1] and Shigeaki Kobayashi[1]

Objective: We report a rare case of intraosseous arteriovenous fistula (AVF) in the petrous bone occluded by transvenous coil embolization, complicated by transient hearing loss postoperatively.

Case Presentation: A 55-year-old female patient underwent medical examination for vertigo and headache. CT showed an osteolytic lesion in the right petrous bone. CTA and DSA revealed an AVF that had caused bone erosion. We performed transvenous coil embolization to obtain complete occlusion of the fistula. Vertigo disappeared soon after the procedure, but hearing loss in the right side worsened to near deafness by that night. We started steroid pulse therapy and heparinization. The hearing gradually recovered to the preoperative level in 10 days.

Conclusion: It is important to pay attention to possible hearing loss in cases of transvenous coil embolization for intraosseous AVF in the petrous bone.

Keywords ▶ petrous, intraosseous, arteriovenous fistula

Introduction

The petrous bone forms a part of the skull base between the sphenoid and occipital bones, and the internal auditory canal is located near the center of the posterior surface of the petrous bone. While intracranial dural arteriovenous fistulas (AVFs) commonly occur in the cranial venous sinuses, intraosseous occurrence is rare, especially in the petrous bone. We encountered a case of intraosseous petrous bone AVF, which was successfully obliterated by transvenous coil embolization, with transient worsening of the ipsilateral hearing postoperatively.

Case Presentation

A 55-year-old female patient underwent a medical examination for vertigo and headache in our hospital. She had a history of bilateral otitis media with cholesteatoma that was operated on 40 years previously. Hearing loss on both sides progressed after the operation with total hearing loss in the left ear and significant loss in the right ear. CT showed osteolysis of the right petrous bone, involving the right internal auditory canal and extending to the adjacent right jugular foramen; the lesion was not contiguous with the previously operated site for the cholesteatoma (**Fig. 1**). MRI revealed an AVF around the petrous bone, with no visible signs of a neoplastic process. CTA and DSA revealed that the AVF was supplied by the bilateral ascending pharyngeal arteries, branches of the right middle meningeal artery, recurrent arteries of the right maxillary artery, right anterior tympanic artery, right stylomastoid artery, mastoid branch of the right occipital artery, meningohypophyseal trunk arising from the right internal carotid artery and branches of the right vertebral artery, and so on,

[1]*Department of Neurosurgery, Aizawa Hospital, Matsumoto, Nagano, Japan*
[2]*Department of Neurosurgery, Ina Central Hospital, Ina. Nagano, Japan*

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Corresponding author: Daisuke Satoh. Department of Neurosurgery, Aizawa Hospital, 2-5-1, Honjou, Matsumoto, Nagano 390-8510, Japan
Email: ds726@m7.dion.ne.jp

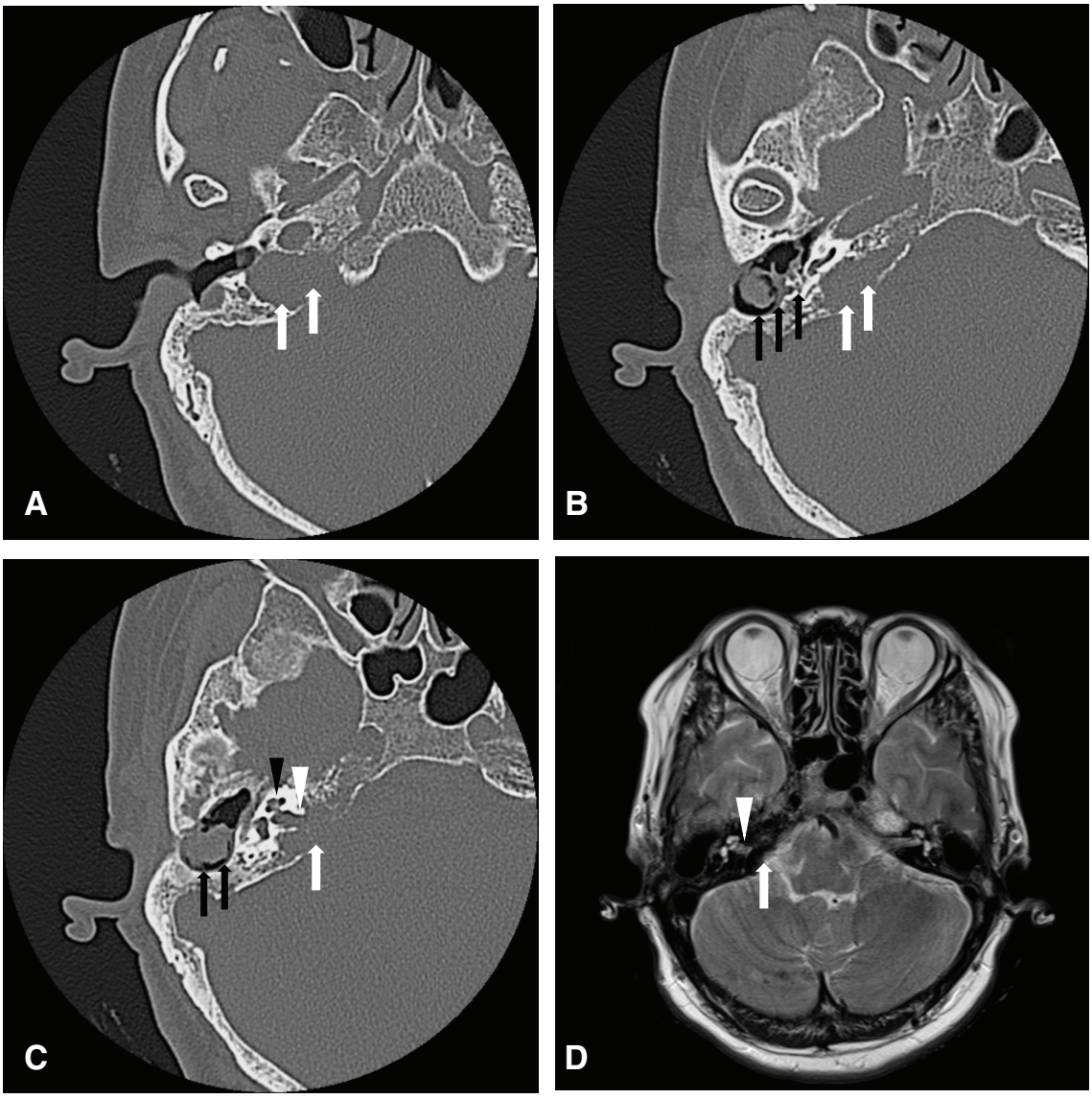

Fig. 1 (**A**–**C**) CT shows the partial osteolysis of the right petrous bone (white arrows) involving the internal auditory meatus (white arrowhead) and the previous operative site for cholesteatoma (black arrows). The black arrowhead indicates the cochlea. (**D**) MRI-T2WI shows disruption of the internal auditory meatus (arrowhead) by osteolysis (arrow). T2WI: T2 weighted image

draining into the right jugular vein; no reflux of the shunt flow to cortical veins was evident (**Figs. 2**–**4**). The eroded bone became clear with a venous pouch where most feeders converged (**Figs. 2** and **4**). Although she noticed pulsatile tinnitus in the right ear, she did not report it to be noisy because of the hearing disorder. Because the bone erosion was progressive, we performed transvenous coil embolization with the patient's consent.

Under local anesthesia, a 6-Fr 80-cm Flexor Shuttle Guiding Sheath (COOK MEDICAL, Bloomington, IN, USA) was inserted into the right femoral vein and 4-Fr introducer sheathes were inserted into the right femoral artery and the right radial artery. Following venous and arterial access, 3000 U of heparin was injected. The 6-Fr Flexor Shuttle Guiding Sheath was guided to the right jugular vein. A 4-Fr modified Simmons-type catheter (Carry; UTM, Aichi, Japan) was guided to the right external carotid artery from the right radial area and a 4-Fr JB2 type catheter (Nishiya; Medikit, Tokyo, Japan) was guided to the left ascending pharyngeal artery from the right femoral route for angiography.

Next, two microcatheters (Excelsior 1018; Stryker, Kalamazoo, MI, USA) were navigated to the venous pouch where most of the feeders converged and were located adjacent to the upper medial part of the right jugular bulb over a 0.014-inch micro guidewire (Transend EX Floppy; Stryker) from the 6-Fr Flexor Shuttle Guiding Sheath. In

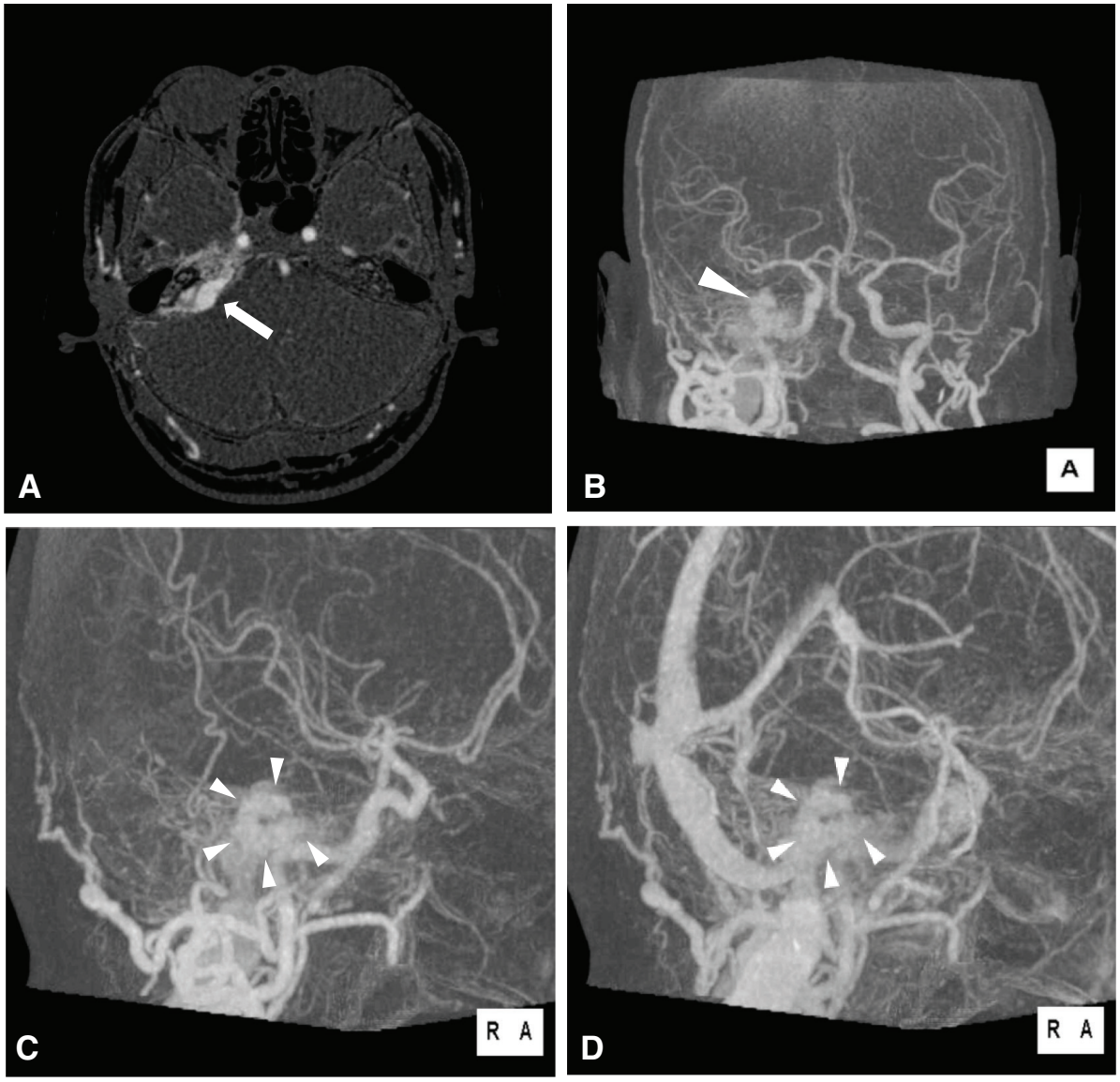

Fig. 2 (**A**, **B**) CTA revealing an AVF around the petrous bone. The bone erosion part (arrow) becomes clear with a venous pouch (arrowhead) where most of the feeders converge. (**C**, **D**) CTA RAO 45° revealing the venous pouch presenting as a C-shaped space structure (arrowheads). (**C**) Arterial phase and (**D**) venous phase. A: anterior; AVF: arteriovenous fistula; R: right; RAO: right anterior oblique

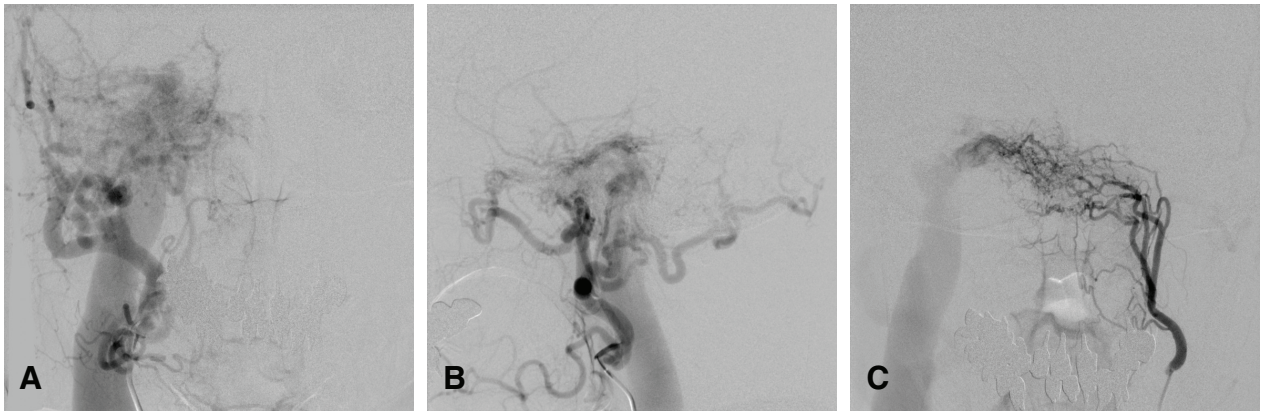

Fig. 3 (**A**, **B**) Right external carotid artery angiogram showing an intense AVF. (**A**) Anterior-posterior view and (**B**) lateral view. (**C**) Selective angiogram of the left ascending pharyngeal artery showing an AVF on the contralateral side. AVF: arteriovenous fistula

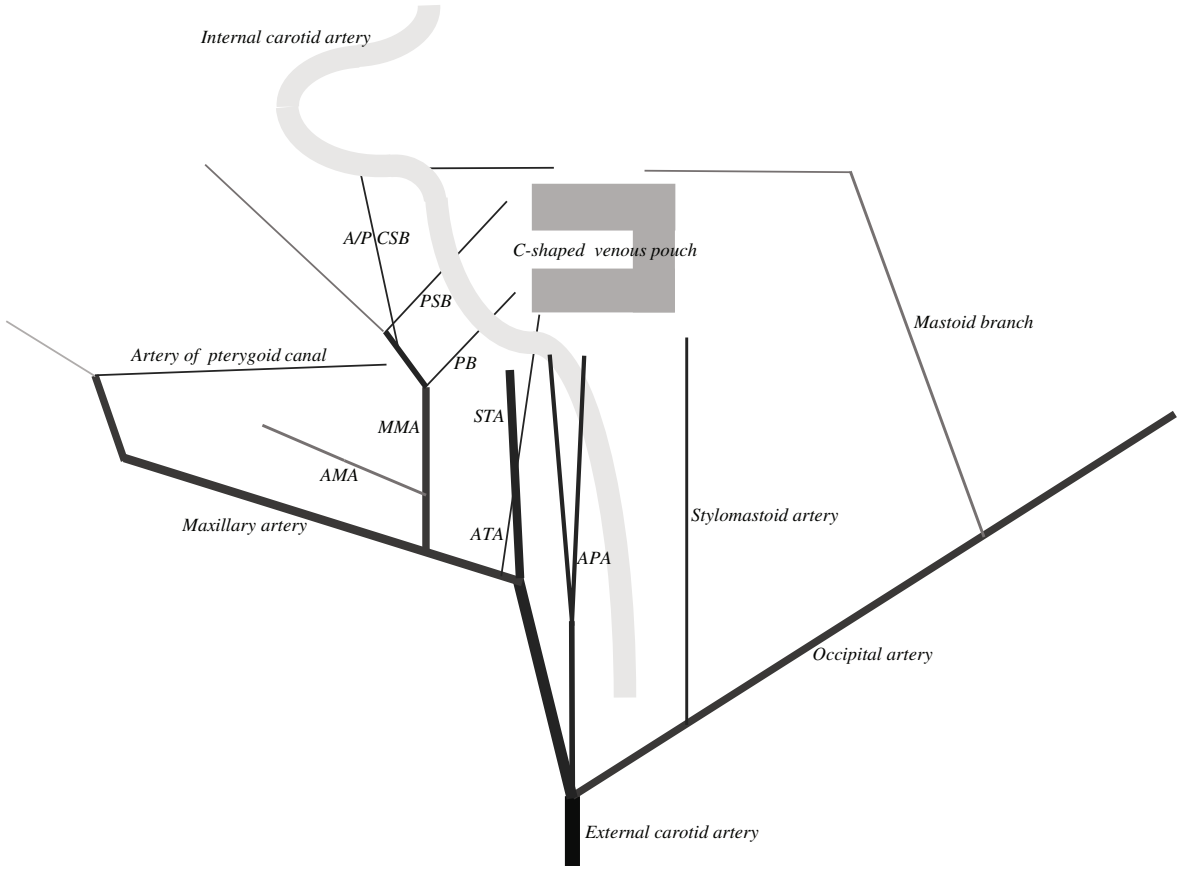

Fig. 4 Schematic representation of the right external carotid artery angiogram of this case. Detailed analysis of the original DSA and 3D-CTA images indicates that the main feeders are the ascending pharyngeal artery, branches of the middle meningeal artery, recurrent arteries of the maxillary artery, anterior tympanic artery, stylomastoid artery, and mastoid branch of the occipital artery. A/P CSB: anterior/posterior cavernous sinus branch; AMA: accessory meningeal artery; APA: ascending pharyngeal artery; ATA: anterior tympanic artery; MMA: middle meningeal artery; PB: petrosal branch; PSB: petrosquamous branch; STA: superficial temporal artery

addition, through the 6-Fr Flexor Shuttle Guiding Sheath, we placed a balloon catheter (Hyperform 7 mm × 7 mm; Medtronic, Minneapolis, MN, USA) from the right sigmoid sinus to the right jugular bulb to prevent migration of the embolized coils from the venous pouch to the right jugular bulb.

The venous pouch presented a C-shaped space structure (**Fig. 2**). In our analysis, the feeders seemed to converge mainly in both ends and upper lateral part of the C-shaped venous pouch; hence, we guided each microcatheter to both ends of the C-shaped venous pouch and performed coil embolization with the balloon to assist in the patency of the venous sinus. We paid special attention to hearing, vertigo, and facial palsy during coil embolization. Finally, the AVF was completely occluded with 26 coils without obstruction of the venous sinus (**Fig. 5**).

Vertigo disappeared soon after the procedure while the right hearing disorder worsened that night to near deafness. The right pure tone average was 71.8 dB preoperatively, and 98.8 dB on postoperative day 1. We consulted otolaryngologists and started steroid pulse therapy and continuous intravenous heparin infusion (20000 U/day) to lessen the possible mechanical compression of the embolized coils on the auditory nerve as well as to improve the disturbed circulation around the cochlea following coil embolization. Right-sided hearing began to improve around postoperative day 5 and almost recovered to the preoperative level (73.8 dB) on day 10.

Follow-up MRI 1 year after embolization showed no recurrence of the AVF and the right internal auditory meatus and auditory nerve passing through the central cavity of the C-shaped embolized coil mass (**Fig. 5**).

Discussion

Malik et al. first described two cases of intraosseous AVFs that were treated by surgical excision in 1994. The intraosseous AVFs were completely removed by resection of the pathologic bony portion.[1] Jung et al. reported six cases of intraosseous AVFs successfully treated with transvenous embolization and

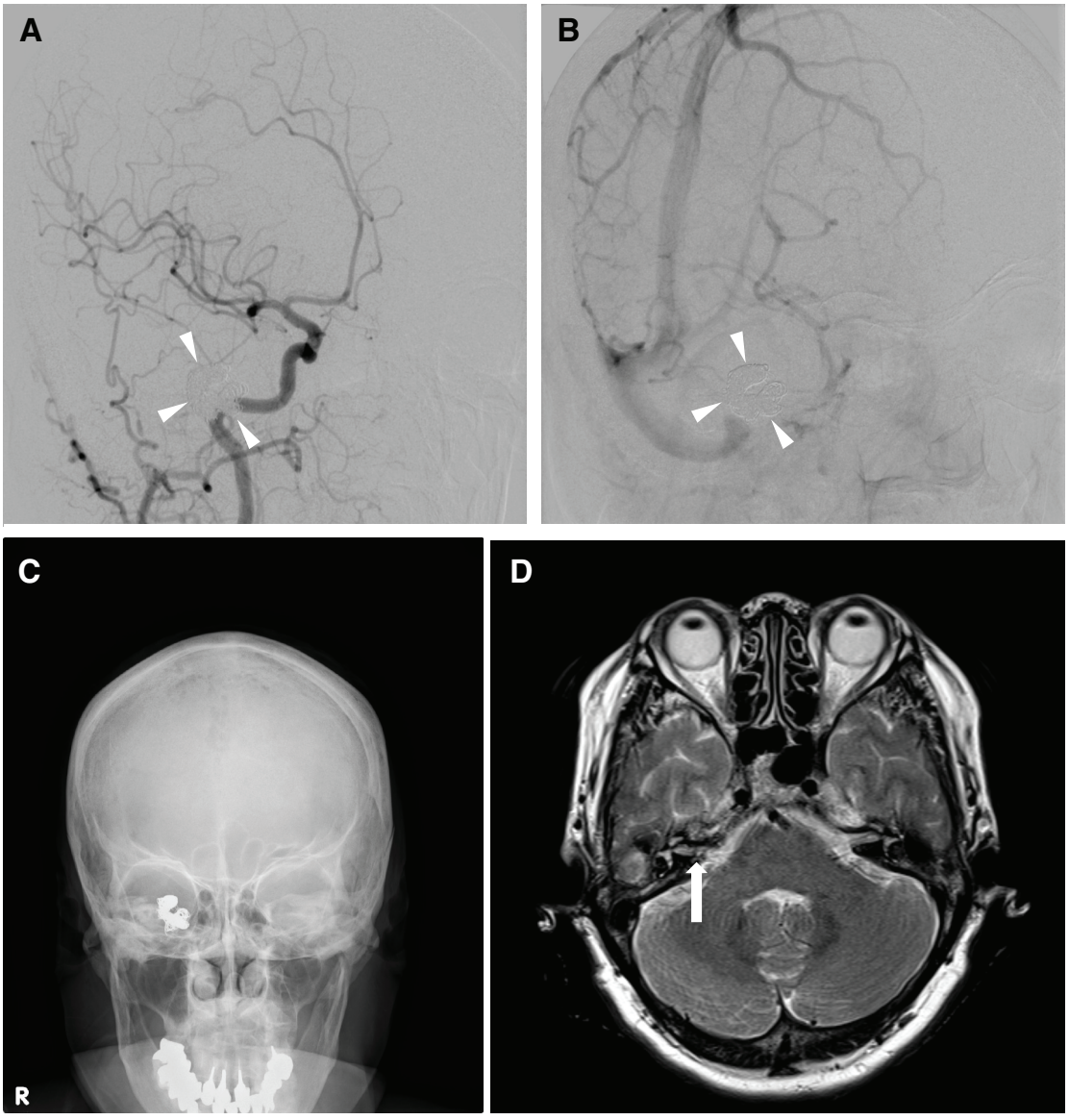

Fig. 5 (**A**, **B**) Postoperative right common carotid artery angiogram revealing a complete obliteration of the AVF (arrowheads indicate the C-shaped coil mass). (**A**) RAO 45° arterial phase and (**B**) RAO 45° venous phase. (**C**) Postoperative X-p. (**D**) Follow-up MRI 1 year after embolization shows the right internal auditory meatus (arrow) preserved with the auditory nerve passing through the central cavity of the C-shaped coil mass. AVF: arteriovenous fistula; R: right; RAO: right anterior oblique

concluded that the intraosseous dilated venous pouch could be the target lesion for endovascular treatment.[2)]

Although anterior condylar confluence (ACC) dural AVFs sometimes have an intraosseous venous pouch where the feeders converge around the hypoglossal canal,[3)] intraosseous AVF located in the petrous bone is rare; only one case has been reported to the best of our knowledge.[2,4)] Although the DSA findings in this case somewhat resembled those of ACC dural AVFs, bone erosion was located mainly in the petrous bone, and neither the bone erosion nor AVF was confirmed around the ACC and the hypoglossal canal on the contrast-enhanced CT. In contrast, several cases of arteriovenous malformation in the internal auditory meatus with dilatation of the internal auditory canal have been reported.[5–8)]

This patient had a history of bilateral otitis media with cholesteatoma and underwent surgery for the cholesteatoma. In this case, the osteolytic portion did not extend to the previously operated site for the cholesteatoma on preoperative CT examination. An association between otitis media with cholesteatoma that was operated on and intraosseous AVF is not definite in this case, but as Mironov reported chronic otitis media as a predisposing history for focal phlebothrombosis, it might have some relationship with the formation of dural AVFs.[9)]

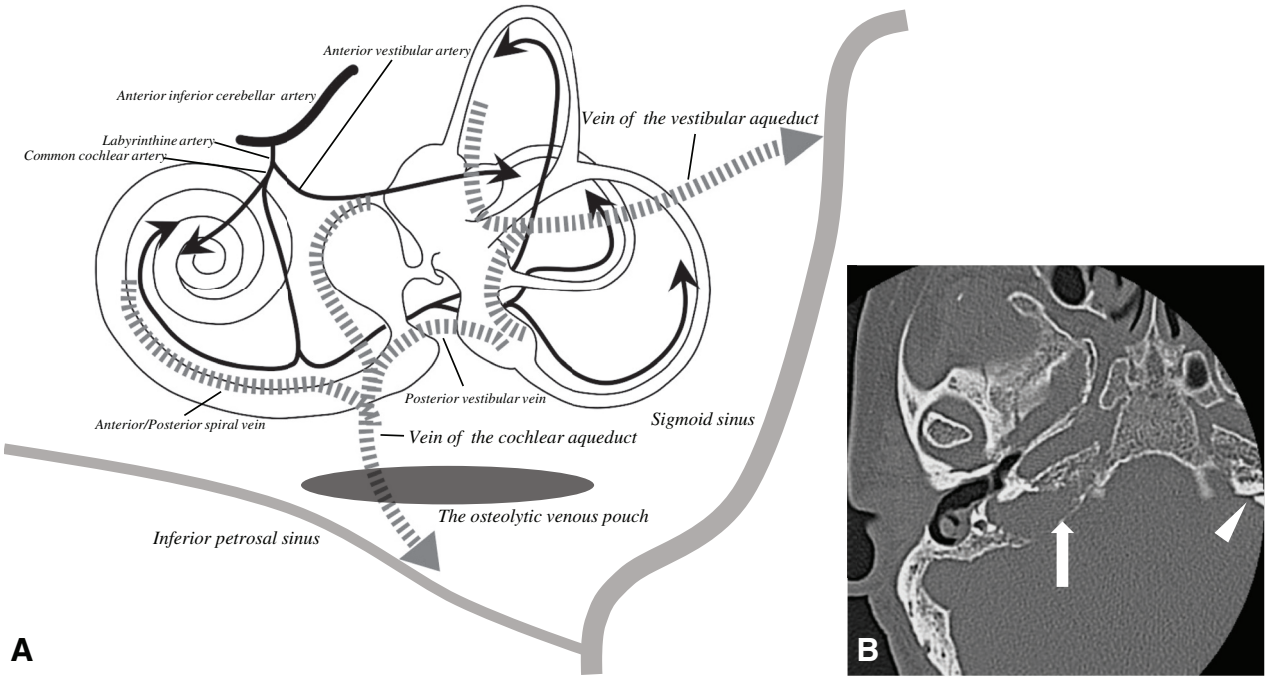

Fig. 6 (**A**) Schematic representation of the vascular anatomy of the inner ear (Modified from Hiramatsu H et al.[10]). The osteolytic venous pouch involves the vein of the cochlear aqueduct. (**B**) Preoperative CT shows the osteolytic portion (arrow) involving the right cochlear aqueduct (arrowhead indicates the left cochlear aqueduct).

This intraosseous AVF was completely occluded with transvenous coil embolization into the C-shaped venous pouch where the feeders converged; however, her right hearing disorder worsened several hours after the procedure. Kim et al. reported an intraosseous AVF located at the petrous apex, describing that the patient had noticed hearing loss 4 months before surgery, and the loss persisted after the successful obliteration of the AVF by transvenous coil embolization.[4] They speculated that the mechanism of hearing loss involved either direct compression of the auditory nerve or interruption of the arterial blood supply to the auditory nerve or cochlea by the dilated draining vein or venous pouch. In our case, the embolized part of the eroded bone was around the right internal auditory meatus in which the auditory nerve should have originally run; therefore, we initially considered the possibility of mechanical compression on the coils on the nerve, because of which we used steroid pulse therapy. However, the follow-up MRI 1 year after embolization showed that the right internal auditory meatus preserved with the auditory nerve passing through the central cavity of the C-shaped embolized coil mass. In addition, the right-sided hearing almost recovered to the preoperative level in a relatively brief period of 10 days, so we conclude that mechanical compression was not the reason for her transient hearing disorder.

The other possible cause for the transient hearing loss was an interruption of the arterial blood supply to the cochlea and/or the influence of venous congestion; therefore, we started continuous intravenous heparin infusion at that time. However, we confirmed the preservation of the internal auditory meatus on the postoperative follow-up MRI 1 year later, which suggests that the arterial blood supply to the cochlea was most likely preserved and sufficient. In contrast, there are two other case reports about hearing disorder after transvenous coil embolization for dural AVFs.[10,11] Hiramatsu et al.[10] reported a case of hearing disorder after transvenous coil embolization of the inferior petrosal sinus (IPS) for the ACC dural AVF. They speculated that the hearing disturbance was due to venous circulatory failure of the inner ear caused by occlusion of the IPS because venous drainage from the cochlea enters the IPS and/or jugular bulb passing through the vein of the cochlear aqueduct (inferior cochlear vein). In our case, the embolized C-shaped venous pouch was located adjacent to the upper medial part of the right jugular bulb, and preoperative CT showed an osteolytic portion involving the right cochlear aqueduct. Therefore, we considered that embolization of the C-shaped venous pouch resulted in occlusion of the right vein of the cochlear aqueduct, causing venous congestion of the cochlea (**Fig. 6**).

Although the right-sided hearing fortunately almost recovered to the preoperative level in this case, we should not neglect the possibility of hearing loss in cases of transvenous coil embolization for intraosseous AVF in the petrous bone.

In our case, we speculate that the C-shaped venous pouch was formed by the intraosseous AVF, whereby the internal auditory meatus, a hollow tunnel, did not involve the AVF. Therefore, our case is different from cases of arteriovenous malformation in the internal auditory meatus with dilatation of the internal auditory canal.[5–8)]

Conclusion

We encountered a rare case of intraosseous petrous bone AVF that caused transient hearing loss after transvenous coil embolization. We speculated that the transient hearing disorder was caused by venous congestion of the cochlea following transvenous coil embolization. Physicians should pay attention to possible hearing loss when performing transvenous coil embolization for intraosseous AVF in the petrous bone.

Disclosure Statement

The authors declare no conflict of interest.

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