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Case Report

A Case of an Intraosseous Arteriovenous Fistula at the Squamous Part of the Occipital Bone with Spontaneous Occlusion of Diploic Venous Drainage

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Objective: An intraosseous arteriovenous fistula (AVF) is a rare fistula with an intracranial shunted pouch. A case of an intraosseous AVF at the squamous part of the occipital bone with spontaneous occlusion of diploic venous drainage is described.

Case Presentation: The patient, a Japanese woman in her 80s, presented with headaches at the back of the head and a history of multiple unruptured cerebral aneurysms but no recent head trauma. MRA showed abnormal signals in the occipital diploic region, and DSA showed an intraosseous AVF with a shunted pouch in the squamous part of the occipital bone near the inion. This was not seen on MRA 6 months earlier. One month later, follow-up examinations showed spontaneous occlusion of the diploic venous drainage, leading to a change in retrograde drainage into the superior sagittal sinus. Transvenous coil embolization was performed, and the shunted pouch was completely occluded. Postoperatively, the patient's symptoms resolved, and subsequent follow-ups showed no recurrence of the AVF.

Conclusion: This case suggested that the vascular architecture of intraosseous AVFs might change over a short period. Transvenous embolization was effective in obliterating the intraosseous shunted pouch.

Keywords ▶ intraosseous arteriovenous fistula, diploic arteriovenous fistula, transvenous embolism

Introduction

Intraosseous arteriovenous fistulas (AVFs) are an uncommon variant of arteriovenous shunts, characterized by an intraosseous shunted pouch,[1)] and they have attracted increasing attention in contemporary medical literature. However, the natural course of intraosseous AVFs is unclear.[2,3)] A case of an intraosseous AVF at the squamous part of the occipital bone with spontaneous occlusion of diploic venous drainage is reported.

Case Presentation

A Japanese woman in her 80s visited our hospital with a complaint of headaches at the back of the head. She had a medical history of multiple unruptured cerebral aneurysms and no history of head trauma for several years. At the most recent MRA follow-up 6 months earlier, no abnormalities were noted (**Fig. 1A**), but the head MRA on admission showed abnormal signals at the occipital diploic region (**Fig. 1B** and **1C**). DSA showed an arteriovenous shunt at the occipital diploic region fed by feeders from bilateral occipital arteries (**Fig. 2A** and **2B**). The fistula drained into the left transverse sinus via the confluence of sinuses and the occipital diploic vein. No venous congestion was observed (**Fig. 2C**). Three-dimensional rotational

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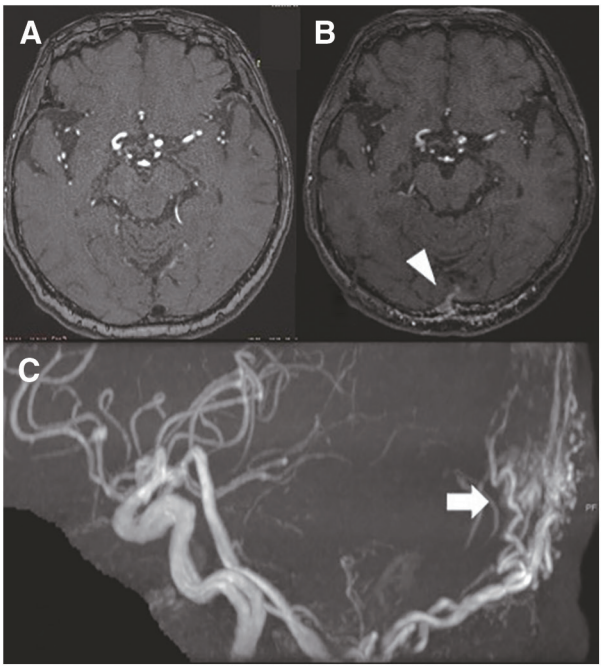

Fig. 1 MRA 6 months before admission (**A**) shows no abnormal high-signal intensity areas. MRA on admission (**B**: axial image, **C**: maximum intensity projection image) shows abnormal signals in the occipital region (arrow and arrowhead).

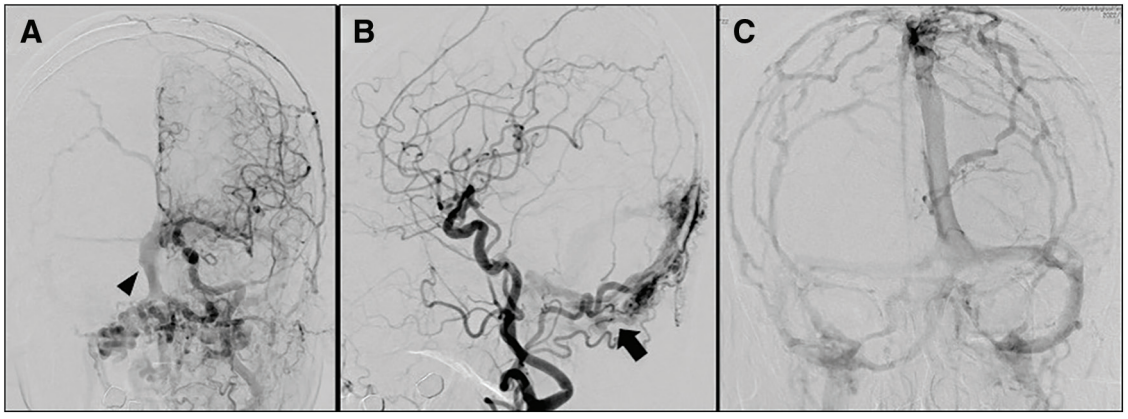

Fig. 2 Left common carotid arteriography (**A**: anterior-posterior view, **B**: lateral view). An arteriovenous shunt at the occipital region is fed by feeders from left occipital arteries (arrow). The fistula drains into the left transverse sinus via the confluence of sinuses and the occipital diploic vein (arrowhead). No venous congestion is observed (**C**).

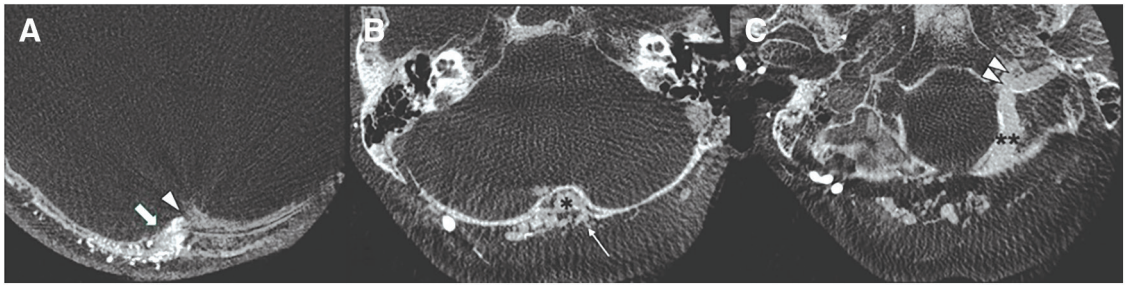

Fig. 3 Axial images of 3D rotational angiography. (**A**) The shunted pouch is in the squamous part of the occipital bone (white arrow). The arteriovenous shunt drains into the confluence of sinuses (white arrowhead) and the occipital diploic vein. (**B**) Diploic drainage descends in the median occipital bone (asterisk), connecting to the dilated external occipital vein (thin arrow) through the occipital foramina. (**C**) On the caudal side, the diploic drainage travels the left edge of the foramen magnum (double asterisk) and drains into the anterior condylar vein (double arrowhead).

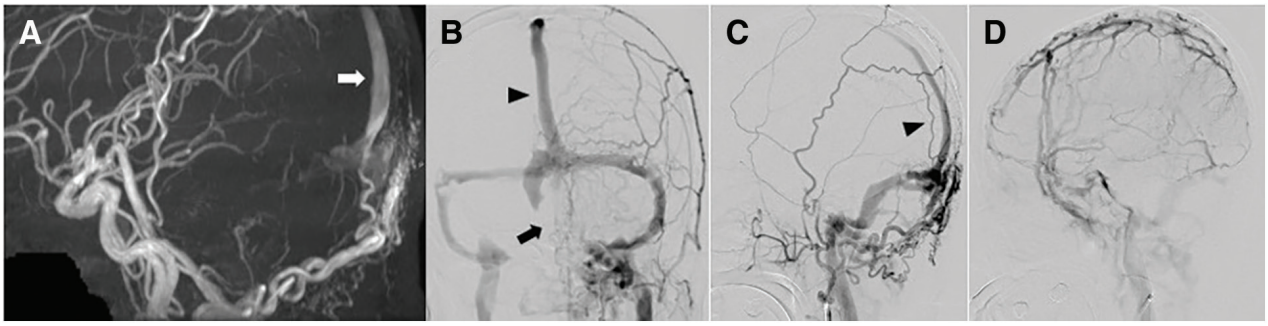

Fig. 4 Follow-up MRA after 1 month shows a hyperintense signal in the SSS (white arrow) that was not observed in the previous MRA (**A**). Left external carotid arteriography (**B**: anterior-posterior view, **C**: lateral view) shows that the diploic venous drainage is occluded (**B**, arrow), accompanied by retrograde drainage into the SSS (**B**, arrowhead and **C**, arrowhead). Left internal carotid angiography shows that the anterior part of the SSS drains into the anterior temporal diploic vein and emissary veins without venous congestion (**D**). SSS, superior sagittal sinus

angiography (3D-RA) demonstrated that the shunted pouch was formed within the squamous part of the occipital bone near the inion; thus, the fistula was diagnosed as an intraosseous AVF (**Fig. 3A**). Diploic drainage descended in the median occipital bone, connecting to the dilated external occipital vein through the occipital foramina (**Fig. 3B**). On the caudal side, the diploic drainage traveled the left edge of the foramen magnum and drained into the anterior condylar vein (**Fig. 3C**). There was no evidence of cerebral venous thrombosis and sinus stenosis.

One month later, MRA showed a hyperintense signal in the superior sagittal sinus (SSS) that was not observed on the previous MRA (**Fig. 4A**). DSA showed that diploic venous drainage was occluded, accompanied by retrograde drainage into the SSS (**Fig. 4B** and **4C**). The anterior part of the SSS drained into the anterior temporal diploic vein and emissary veins without venous congestion (**Fig. 4D**).

Endovascular procedure

Under general anesthesia, transvenous embolization (TVE) was performed with the left femoral approach. A 6Fr guiding sheath (ASAHI FUBUKI Dilator Kit; ASAHI INTECC, Aichi, Japan) was placed in the left internal jugular vein, and a 3.2Fr distal access catheter (TACTICS; Technocrat Corporation, Aichi, Japan) was placed in the left transverse sinus. A microcatheter (Excelsior SL-10; Stryker, Kalamazoo, MI, USA) with a 0.014-inch micro guidewire (ASAHI CHIKAI; ASAHI INTECC) was advanced into the shunted pouch via the confluence of sinuses, and then the shunted pouch was occluded using coils (**Fig. 5A**–**5C**). Intraprocedural cone-beam CT showed that the coils did not protrude beyond the bony margin into the confluence of sinuses (**Fig. 5D**). The final angiogram showed complete occlusion of the intraosseous AVF, and the confluence of sinuses was preserved (**Fig. 5E** and **5F**). Postoperatively, the patient's headache resolved, and there were no complications. One-year follow-up showed no recurrence.

Discussion

An intraosseous AVF is a rare fistula with an intracranial shunted pouch, and several case reports have been published since 1994 when Malik et al. first described it.[1] It is known that the hypoglossal canal is a common site for intraosseous AVFs,[4,5] where there is a particularly highly developed network of veins within the bone.[6] Hiramatsu et al. found that most AVFs that occur not only around the condyle but also on the clivus and the dorsum sellae have a shunt in the osseous part.[7] Other AVFs within the pyramidal and sphenoid bones have also been reported.[4,8] Thus, recently, there have been an increasing number of reports of AVFs occurring in osseous parts. Many cases of intraosseous AVFs occur at the base of the skull, and some consider that the base of the skull is a common site for intraosseous AVFs due to the many emissary veins.[4]

Intraosseous AVFs that involve diploic veins, as in the present case, have been reported as "diploic" AVFs.[2,3,9–12] Tokuyama et al. summarized 20 cases of diploic AVFs; the cause of onset was unknown in most cases, although some were due to trauma or pregnancy, and 11 of the 12 cases involved the frontal or parietal regions.[2] Involvement of the occipital diploic vein, as in the present case, is very rare.[13]

The occipital diploic vein courses in the superoinferior direction in the median occipital bone. A previous MRI study demonstrated that 82% of occipital diploic veins had a channel with the confluence of the sinuses, as in the present case.[14] Occipital diploic veins drain into extracranial

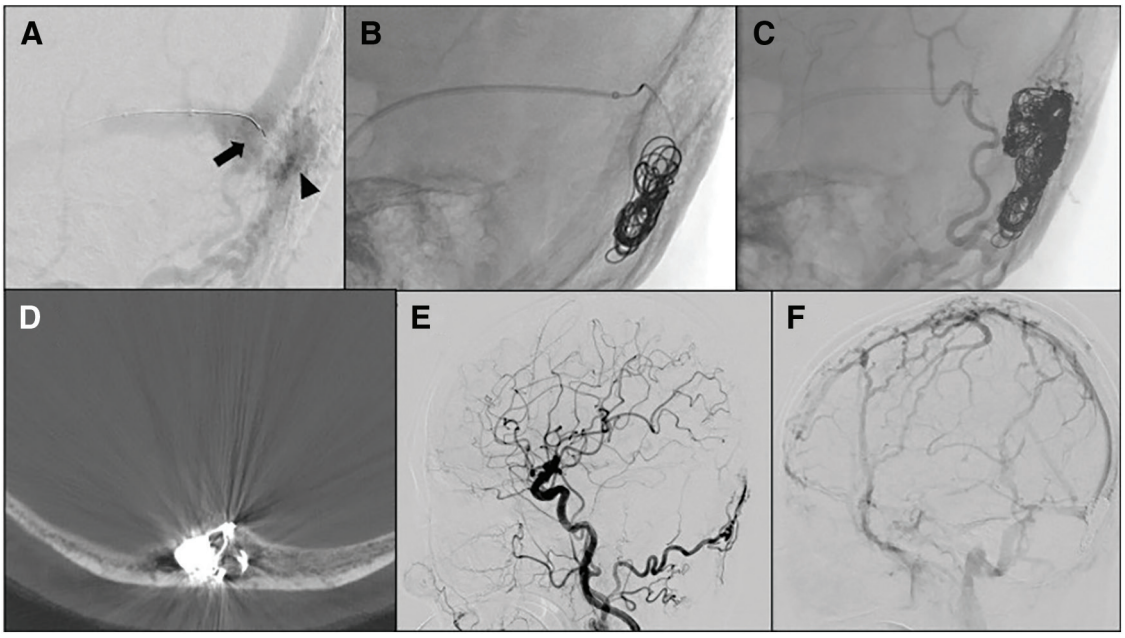

Fig. 5 Intraprocedural images. (**A**) A microcatheter is advanced into the shunted pouch (arrowhead) via the confluence of sinuses (arrow). (**B** and **C**) The shunted pouch is occluded using coils. (**D**) Intraprocedural cone-beam CT shows that the coils do not protrude beyond the bony margin into the confluence of sinuses. (**E** and **F**) The final angiogram shows complete occlusion of the intraosseous arteriovenous fistula, with the confluence of sinuses preserved.

occipital veins via occipital emissary veins running through the occipital foramina located in the squamous part of the occipital bone.[14,15]

The natural course of intraosseous AVFs is unclear. In the present case, the AVF occurred spontaneously within 6 months, and the diploic venous drainage was occluded spontaneously the next month. Rivera-Lara et al. proposed three potential venous outflow patterns: (1) dural sinus drainage only, (2) extracranial drainage only, and (3) dural sinus and extracranial drainage.[3] After spontaneous occlusion of diploic venous drainage, the venous outflow pattern changed from type (3) to type (1) in the present case. In some dural AVFs, the drainage route is known to change due to thrombosis, and the present case suggested that the vascular architecture of diploic AVFs might also change within a short period through a similar mechanism.[16]

It was challenging to identify the location of the intraosseous shunted pouch on conventional angiography, but 3D-RA clearly showed not only the vascular architecture but also the positional relationship between the skull and the shunted pouch and the site of outflow into the venous sinus.

Endovascular treatment for the intraosseous AVF, including TVE and transarterial embolization (TAE), has been reported.[2,3,5,7,8,10–13] Patients with intraosseous AVFs at the skull base mainly received TVE.[5,7,8] On the other hand, it was often challenging to approach intraosseous AVFs in the frontal or parietal bones transvenously. Such cases received TAE using N-butyl-2-cyanoacrylate or Onyx without recurrence and complications.[2,3,10,11] In the present case, since the channel between the confluence of the sinuses and the occipital diploic vein was wide enough to introduce a microcatheter into the shunted pouch, TVE was used. Moreover, TAE with liquid embolic materials has a risk of migration of embolic materials into the confluence of the sinuses. Cone-beam CT during the procedure was useful to confirm that the coils did not protrude beyond the bony margin into the venous sinus.

Conclusion

A case of an intraosseous AVF with a shunt pouch in the squamous part of the occipital bone was described. This case occurred within 6 months, and the diploic venous drainage was occluded spontaneously the following month. TVE was effective in obliterating the intraosseous shunted pouch.

Disclosure Statement

The authors declare that they have no conflicts of interest.

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