Online April 29, 2021
Journal of Neuroendovascular Therapy 2021; 15: 811–817
DOI: 10.5797/jnet.cr.2020-0099

Case Report

A Case of the So-Called Posterior Condylar Canal Dural Arteriovenous Fistula with an Osseous Shunt

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Objective: The authors describe a case of the so-called dural arteriovenous fistula (DAVF) around the posterior condylar canal (PCC).

Case Presentation: A 71-year-old woman presented with pulse-synchronous bruit on the left side. Conventional DSA demonstrated the following: There were many feeders, including the ascending pharyngeal artery (APA), the occipital artery (OA), and the vertebral artery (VA), to the DAVF around the PCC. Shunt flow from the posterior condylar vein (PCV) drained the suboccipital cavernous sinus (SCS) and sigmoid sinus (SS), and there was venous reflux into the inferior petrosal sinus (IPS). The patient was diagnosed with PCC DAVF and underwent transvenous embolization (TVE) with coils. Intraoperative 3D-rotational angiography (RA) and axially reconstructed images revealed an osseous shunt within the occipital bone adjacent to the PCC. The arteriovenous (AV) shunt and other symptoms disappeared after occluding the drainage route from the osseous shunt to the PCV.

Conclusion: There are only three previous reports of PCC DAVF, being rare. However, no report clearly described the shunt point of PCC DAVF. 3D-RA and axially reconstructed images were useful to find and treat the shunt point.

Keywords▶ dural arteriovenous fistula, posterior condylar canal, osseous shunt

Introduction

Recently, peri-anterior condylar confluence (ACC) dural arteriovenous fistulae (DAVFe), including ACC DAVFe, have been investigated. In this study, we report a patient in whom transvenous embolization (TVE) of a DAVF, characterized by osseous shunt formation adjacent to the posterior condylar canal (PCC) and outflow to the posterior condylar vein (PCV), was performed, and review the few published reports of similar cases.

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Received: June 13, 2020; Accepted: February 15, 2021
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Case Presentation

Patient: A 71-year-old woman.
Medical history: Hypertension, hyperlipidemia, and thyroid tumor.
Family history: Not contributory.
Present illness: Left tinnitus developed 1 month prior and she consulted a local otorhinolaryngology clinic. Brain MRI revealed aggregation of abnormal blood vessels, suggesting a DAVF. She was referred to our hospital.
Physical examination: There was no abnormality other than pulsatile left tinnitus.
Imaging findings: Conventional angiography demonstrated that feeders, including the bilateral ascending pharyngeal arteries (APAs), left occipital artery (OA), bilateral anterior meningeal arteries (AMAs), left deep cervical artery, bilateral dorsal meningeal arteries (DMAs), and muscular branch of the left vertebral artery (VA) aggregated at the fistula pouch (shunt point) and flowed into the left PCV.

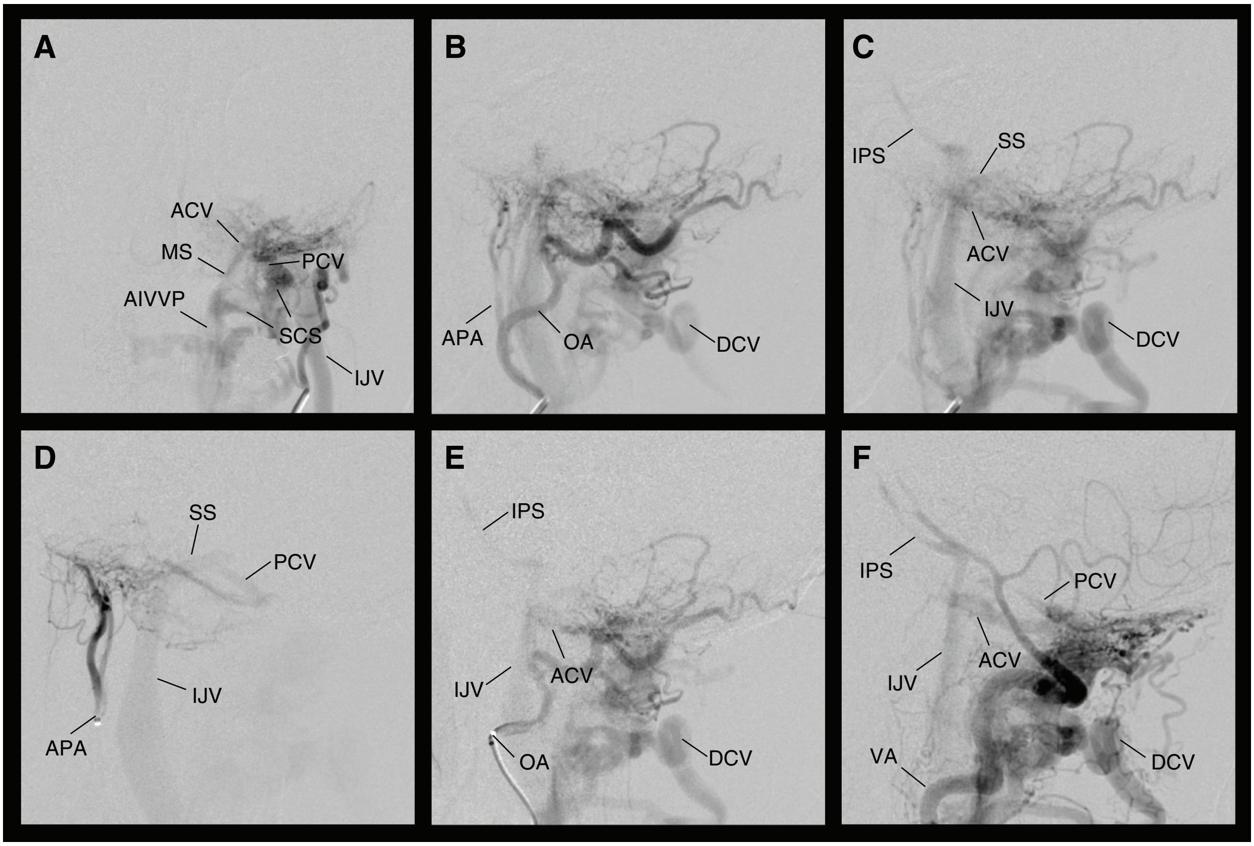

Fig. 1 (**A**) Anteroposterior view on angiography of the left common trunk of the APA and the OA. (**B**, **C**) Lateral view on angiography of the left common trunk of the APA and the OA. Many feeders from the APA and OA aggregated around the PCV. The fistulous flow drained through the PCV in two directions, the SS and the SCS. The fistulous flow reached the SCS, draining into both the DCV and the IJV via the MS and the ACV. Reflux into the IPS was confirmed. (**D**) Lateral view on angiography of the left APA. The fistula flow from the APA drained into the SS and the IJV. (**E**) Lateral view on angiography of the left OA. The fistula flow mainly reached the SCS from the OA draining into the IJV via the MS and ACV. Reflux into the IPS was confirmed. (**F**) Lateral view on angiography of the left VA. Many feeders from the VA aggregated around the PCV. The fistula flow drained through the PCV in two directions, the SS and the SCS. The amount of drainage to the SCS was greater than that of the SS. The fistulous flow from the SCS drained into both the DCV and the IJV via the MS and ACV. Reflux into the IPS was confirmed. ACV: anterior condylar vein; AIVVP: anterior internal vertebral venous plexus; APA: ascending pharyngeal artery; DCV: deep cervical vein; IJV: internal jugular vein; IPS: inferior petrosal sinus; MS: marginal sinus; OA: occipital artery; PCV: posterior condylar vein; SCS: suboccipital cavernous sinus; SS: sigmoid sinus; VA: vertebral artery

Subsequently, the arterial blood either drained from the suboccipital cavernous sinus (SCS) and sigmoid sinus (SS) into the internal jugular vein (IJV) on the cardiac side, or regurgitated into the inferior petrosal sinus (IPS) (**Fig. 1**). Based on these results, a diagnosis of PCC DAVF was made. The left APA formed a common trunk with the OA and branched. On selective angiography of respective blood vessels, a feeder from the left APA flowed into the PCV, draining to the IJV via the SS. A feeder from the left OA flowed into the PCV, draining to the cardiac side through the extracranial drainage route or draining to the IJV/regurgitating to the IPS via the SCS, marginal sinus (MS), and anterior condylar vein (ACV). A feeder from the left VA flowed into the PCV, draining in two directions: the SS and extracranial PCV sides. However, the greater portion of shunt flow went to the extracranial PCV side, draining to the IJV/regurgitating to the IPS via the SCS, MS, and ACV, as described for shunt flow from the OA.

Treatment: Under local anesthesia, TVE was performed. A 4-Fr long sheath was inserted into the right femoral artery and a 5-Fr Fubuki guiding sheath 80 cm (Asahi Intecc, Aichi, Japan) was inserted into the left femoral artery. In addition, a 5-Fr Fubuki guiding sheath 90 cm was inserted into the right femoral vein. While maintaining the activated coagulation time (ACT) at 2.0–2.5 (control value) through systemic heparinization, the following procedure was conducted: The 5-Fr Fubuki guiding sheath was guided into the left common carotid artery (CCA) through the left femoral artery. To this guiding sheath, an XT-27 (Stryker, Kalamazoo, MI, USA) was guided and the left APA was selected using a CHIKAI 0.014 (Asahi Intecc). In addition, a 4-Fr JB-1 catheter (MEDIKIT, Tokyo, Japan) was inserted into

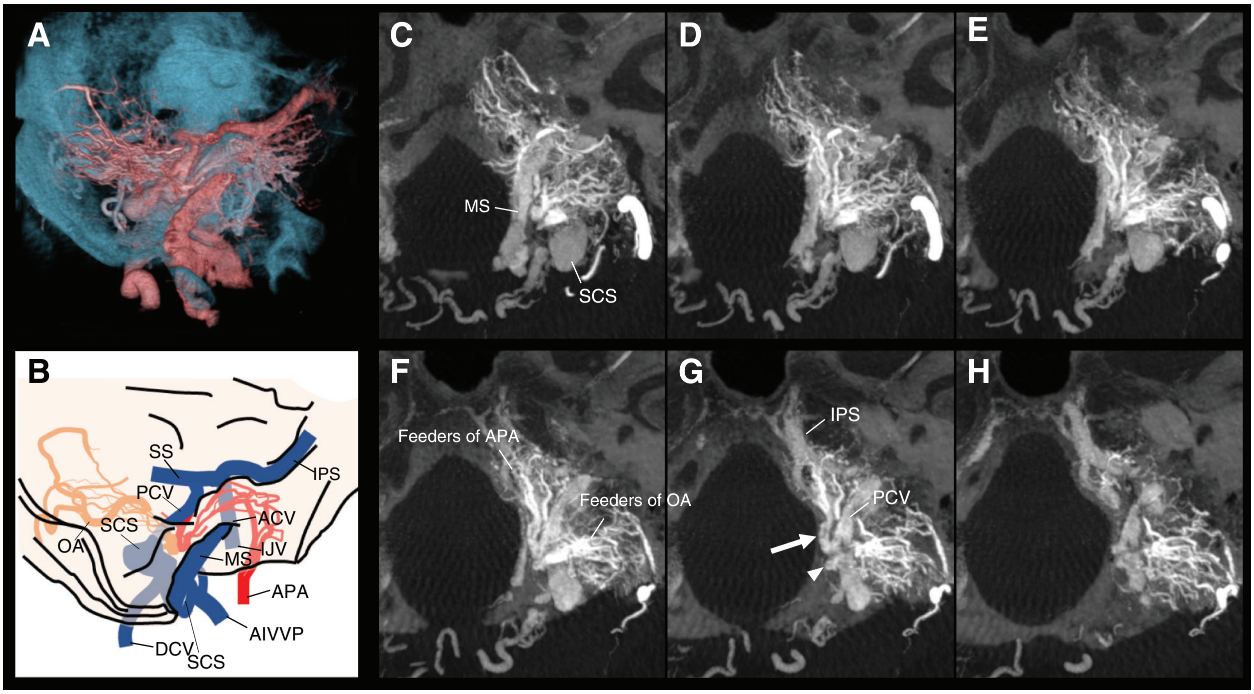

Fig. 2 (**A**) 3D-RA via the left common trunk of the APA and the OA. The figure shows the rear upper view from the medial side. (**B**) Illustration of 3D-RA via the left common trunk of the APA and the OA. (**C–H**) Axially reconstructed image of 3D-RA via the left common trunk of the APA and the OA. Two fistula pouches (white arrow and white arrowhead) were located in the occipital bone medial to the PCC. One fistula pouch (white arrow) mainly received flow from the AV shunt from the medial feeders, whereas the other (white arrowhead) mainly received flow from the lateral feeders. ACV: anterior condylar vein; AIVVP: anterior internal vertebral venous plexus; APA: ascending pharyngeal artery; AV: arteriovenous; DCV: deep cervical vein; IJV: internal jugular vein; IPS: inferior petrosal sinus; MS: marginal sinus; OA: occipital artery; PCC: posterior condylar canal; PCV: posterior condylar vein; RA: rotational angiography; SCS: suboccipital cavernous sinus; SS: sigmoid sinus

the right CCA for angiography. Under a roadmap, the 4-Fr JB-1 catheter + 5-Fr Fubuki coaxial system was guided into the left IJV using a Silverway (Asahi Intecc). Initially, an Echelon 14 pre-shaped 45 (Medtronic, Minneapolis, MN, USA) was placed in the left IJV and guided through the SS into the PCV using a Synchro[2] (Stryker, Kalamazoo, MI, USA), by selective angiography through the bilateral APAs as appropriate. The shunt point was embolized with coils based on the information obtained on 3D rotational angiography (3D-RA) and axially reconstructed images (**Fig. 2**).

As coils, we used a Target 360 NANO (Stryker) and SMART COIL WAVE Extra Soft (Penumbra, Alameda, CA, USA) measuring 1 mm in diameter. Angiography confirmed a decrease in the shunt volume from the bilateral APAs. Although PCV obliteration was considered as an option, we were unable to evaluate the shunt point at this point. Therefore, the 4-Fr JB-1 catheter was guided to the origin of the left VA, and left vertebral angiography (VAG) and 3D-RA were performed (**Fig. 3**). When conducting 3D-RA, the Echelon 14 pre-shaped 45 was placed in the extracranial PCV beyond the intra-PCC PCV through the IJV to clarify the shunt point/venous structures. The tip of the other catheter was guided to the MS via the ACV through the IJV. Beyond this site, resistance was noted and only a microguidewire was guided into the extracranial PCV. Based on the findings on 3D-RA of the left OA (common trunk with the APA) and axially reconstructed images (**Fig. 2**), the shunt distribution primarily consisted of two affluents: aggregation of feeders medial to the PCV and that of feeders lateral to the PCV. In each affluent, osseous shunt formation was observed in an area medial to the PCV and one affluent was adjacent to the other in the anteroposterior direction. The two affluents drained from different points to the PCV (**Figs. 2** and **3**). As PCV obliteration was suggested to lead to disappearance of the shunt, management of the PCV was prioritized. The Echelon 14 pre-shaped 45 was inserted into the extracranial PCV beyond the intra-PCC PCV in preparation for the existence of the extracranial shunt, and coiling of the PCV was performed from the extracranial orifice toward the SS side via the another microcatheter (**Fig. 4**). As coils, we used a Target XL 360 Soft (Stryker) measuring 3 mm in diameter, SMART COIL Extra Soft (Penumbra) measuring 2 mm in diameter, and Barricade COMPLEX FINISHING COIL

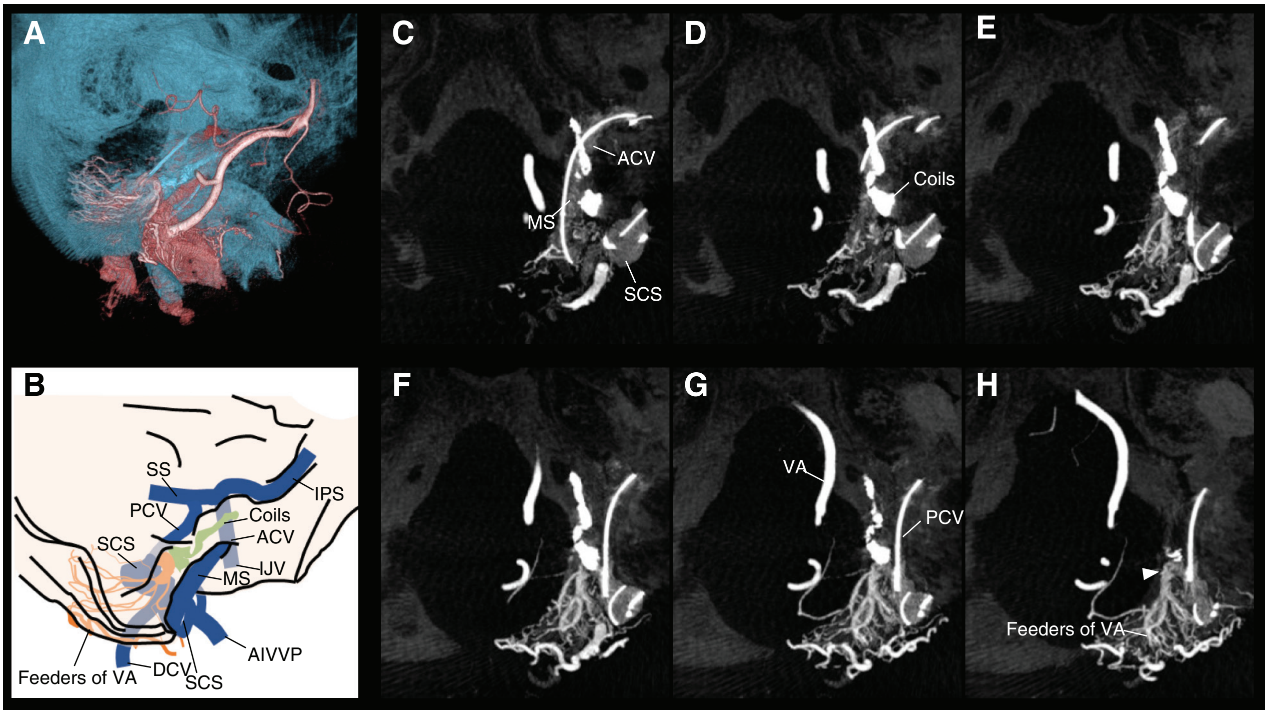

Fig. 3 (**A**) 3D-RA via the left VA. The figure shows the rear upper view from the medial side. (**B**) Illustration of 3D-RA via the left VA. (**C–H**) Axially reconstructed image of 3D-RA via the left VA. The fistula pouch (white arrowhead) was located in the occipital bone medial to the PCC and shared a common trunk with the feeders from the OA. One microsystem (microcatheter and microguidewire) was located in the SCS via the PCV. Another was located in the same area via the ACV and MS. ACV: anterior condylar vein; AIVVP: anterior internal vertebral venous plexus; DCV: deep cervical vein; IJV: internal jugular vein; IPS: inferior petrosal sinus; MS: marginal sinus; OA: occipital artery; PCC: posterior condylar canal; PCV: posterior condylar vein; RA: rotational angiography; SS: sigmoid sinus; SCS: suboccipital cavernous sinus; VA: vertebral artery

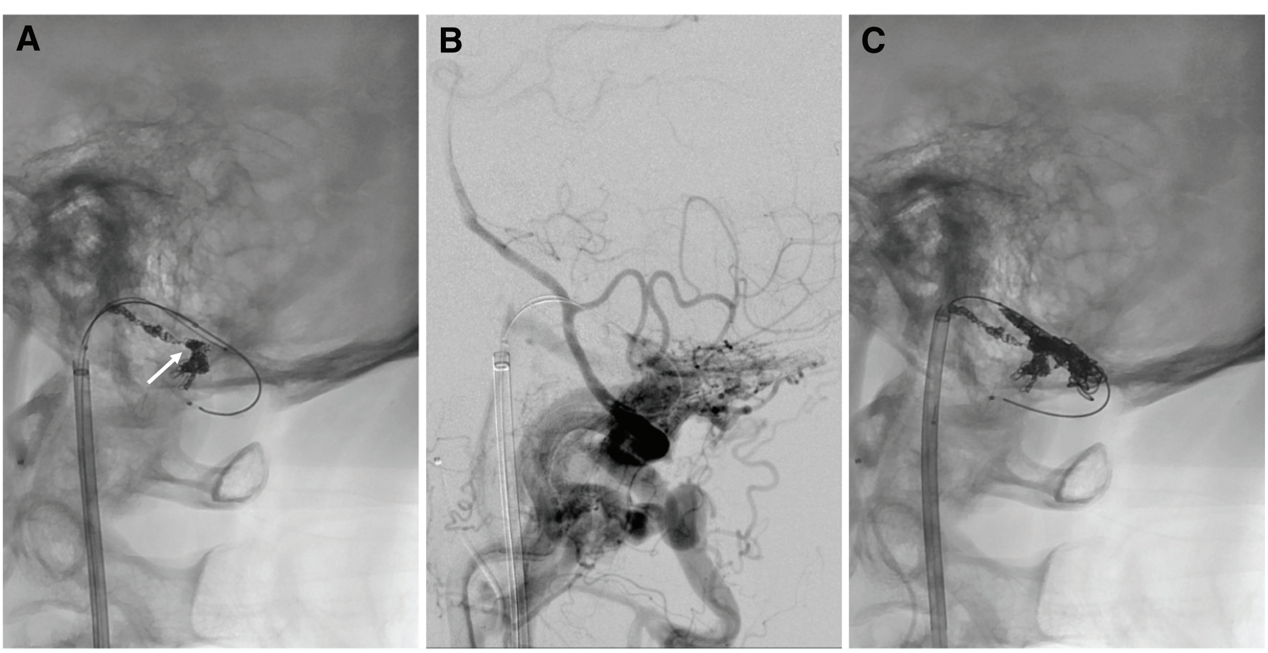

Fig. 4 Lateral view. (**A**) The microcatheter was located in the PCV. Another was located in the SCS via the PCV. The coil mass represents the fistula pouch that received flow from the AV shunt from the medial feeders (white arrow). (**B**) Lateral view on angiography of the VA. (**C**) The PCV was occluded with coils. AV: arteriovenous; PCV: posterior condylar vein; SCS: suboccipital cavernous sinus; VA: vertebral artery

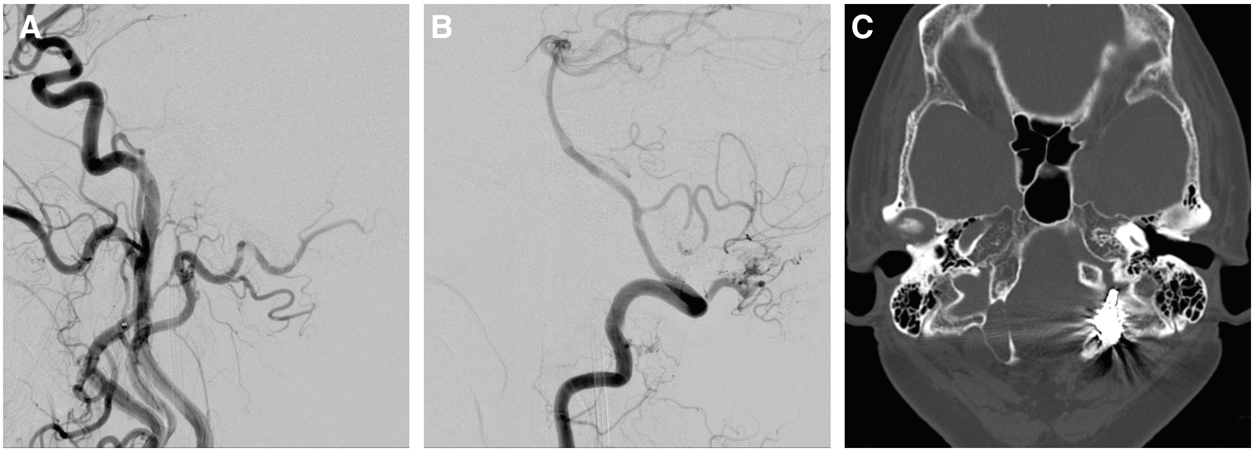

Fig. 5 (**A**, **B**) Lateral view on angiography of the left CCA and VA. The AV shunt and patient's symptom disappeared after occluding the PCV from the osseous shunt. (**C**) Postoperative CT showed the coil mass located in the PCC. AV: arteriovenous; CCA: common carotid artery; PCC: posterior condylar canal; PCV: posterior condylar vein; VA: vertebral artery

(Balt USA, Irvine, CA, USA) measuring 2 mm in diameter. After confirming shunt occlusion using angiography, the procedure was completed (**Fig. 5**).

Course: After treatment, pulsatile left tinnitus, as a complaint, promptly disappeared. The course was favorable and the patient was discharged 7 days after surgery.

Discussion

The PCC is a great intracranial emissary tract. According to a previous review using CT, its structure can be confirmed on the unilateral side in at least 81% of patients (bilateral: 31%, unilateral: 50%).[1] This tract involves the course of the PCV. The PCV originates from the most medial area of the jugular bulb (JB) or SS, draining to the SCS or paravertebral vein through the PCC.[2,3]

Recently, peri-ACC DAVFe, including ACC DAVFe, have been investigated. To our knowledge, only three patients with DAVFe characterized by shunt formation in the PCC, as demonstrated in the present case, have been reported, including the first patient presented by Kiyosue et al.[4] in 2007; this disease is rare (**Table 1**).[4–6] As its characteristics, pulsatile tinnitus initially develops in many patients,[4–6] and feeders include the APA, OA, AMA, posterior meningeal artery (PMA), DMA, and VA. They aggregate at the shunt point, draining from the PCV to the SS or SCS. Mondel et al.[7] and Shambanduram et al.[8] reported patients with PCC DAVFe. In these patients, subarachnoid hemorrhage was initially noted, but this was related to draining into the intracranial cavity via a bridging vein, differing from the previously reported PCC DAVF patients. Feeders did not flow into the PCV, but seemed to form the shunt at the intracranial bridging vein via the PCC. So, their cases were not DAVFe that formed an osseous shunt and flowed into the PCV as in the past three cases and our case, suggesting a different entity.

Of the previously reported patients with PCC DAVFe, the shunt point was not clearly described. Sasaki et al.[5] reviewed this issue, suggesting that both the dura mater and periosteum are involved in the pathogenesis of ACC-adjacent DAVFe involving the PCC based on the following three points: (1) the jugular foramen (JF) and the JB to PCC area are covered with the dura mater; (2) on the inner surface of the JF, the periosteum and dense perineural connective tissue are present, and the inner surface of the PCC is also covered with the periosteum; and (3) the dura mater anatomically resembles the periosteum. However, no study has accurately reported the degree to which the inner surface of the PCC is covered with the periosteum. Furthermore, the above hypothesis does not explain PCC-adjacent bone shunt formation, as demonstrated in the present case.

On the other hand, Mizutani et al.[9] recently investigated the intraosseous venous structure in the jugular tubercle (JT). Using CT-digital subtraction venography, they confirmed an intraosseous vein in the JT superior/medial to the hypoglossal canal in 46% of normal controls. This structure was connected with the ACV and other adjacent venous structures. They termed this structure the jugular tubercle venous complex (JTVC). It was connected with the PCV in 39% of normal controls. Furthermore, they reported that, of their series, nine fistulous pouches were confirmed in seven patients with ACC

Table 1 Summary of PCC DAVF

Case	Symptom	Feeding artery	Draining vein	Treatment/result
54 years, M Kiyosue et al. 2007[4]	Tinnitus	APA	SS	TVE/cured
		AMA	Paravertebral veins	
		OA	OS	
		DMA		
26 Years, M Sasaki et al. 2008[5]	Tinnitus	APA	SS	TVE/cured
		OA	SS and IJV	
Age unknown, F Maus et al. 2016[6]	Tinnitus	APA	SCS	TVE/cured
		VA	SS and IJV	
		ECA (other branches)		
71 Years, F Present case	Tinnitus	APA	SS and IJV	TVE/cured
		AMA	SCS	
		Muscular branch of VA	(IPS→CS)	
		OA		
		DMA		

AMA: anterior meningeal artery; APA: ascending pharyngeal artery; CS: cavernous sinus; DAVF: dural arteriovenous fistula; DMA: dorsal meningeal artery; ECA: external carotid artery; IJV: internal jugular vein; IPS: inferior petrosal sinus; OA: occipital artery; OS: occipital sinus; PCC: posterior condylar canal; SCS: suboccipital cavernous sinus; SS: sigmoid sinus; TVE: transvenous embolization; VA: vertebral artery

DAVFe, and that their sites consisted of the JTVC, ACV, and other bones, accounting for 33.3% each. A venous channel in the osseous layer is embryologically similar to that in the dural layer, being based on the meninx primitiva. The meninx primitiva later differentiates into layers, followed by bone development in the outermost dense skeletogenous layer and dura-mater formation in the inner layer.[10–12] Based on this, Mizutani et al.[13] hypothesized that a physiological arteriovenous (AV) shunt of an osseous vein is embryologically present, leading to AVF development mediated by a pathogenesis resembling that of intracranial DAVFe.

In the present case, intraoperative 3D-RA and axially reconstructed images confirmed two osseous shunts in the bone adjacent to the medial side of the PCV, with outflow to the PCV; we considered shunt points to be present in the bone adjacent to the PCC, differing from standard DAVFe. The osseous shunts were observed in the bone dorsal to the JT and medial to the PCC, but the embryological presence of a physiological AV shunt of an osseous vein may have played a role in AVF development via a pathogenesis resembling that of intracranial DAVFe, as proposed by Mizutani et al.[13]

For treatment, three methods, endovascular surgery, microsurgery, and radiosurgery, are considered. Microsurgery is anatomically difficult and may induce hemorrhage or inferior cranial nerve injury. Radiosurgery requires a specific time until the effects are observed and may induce inferior cranial nerve injury. Therefore, endovascular surgery may be the first option. However, TVE may be safer if the fistula site can be reached. In previous reports, there were two patients in whom transarterial embolization (TAE) with N-butyl-2 cyanoacrylate (NBCA) was performed in an area adjacent to this site,[7,8] but this was because transvenous approaching was difficult in both due to a bridging vein being a drainer.

TAE using liquid agents, such as NBCA and ONYX, may lead to migration to areas other than the target shunt or neuropathy related to occlusion of a nerve-nourishing branch. This must always be considered.

Conclusion

We performed TVE of a PCC DAVF characterized by osseous shunt formation in the bone adjacent to the PCC, with outflow to the PCV. It was difficult to evaluate the state of AVF using conventional cerebral angiography. Intraoperative 3D-RA and axially reconstructed images facilitated accurate evaluation.

Disclosure Statement

The authors declare no conflict of interest.

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