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Transarterial embolization of dural arteriovenous fistula of the inferior petrosal sinus: A report of two cases

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ABSTRACT

Dural arteriovenous fistula (DAVF) of the inferior petrosal sinus (IPS) is very unusual. Endovascular embolization is a good option for the treatment of DAVF. Until now, DAVFs of the IPS have only been reported sporadically. We reported two such cases. Case 1 was a 48-year-old man with headache and diplopia. Angiography confirmed a DAVF of the distal IPS, mainly supplied by the occipital artery (OA), and the IPS was occluded, which retrogradely drained into the cavernous sinus (CS) and then into the cortical vein. The DAVF in case 1 was embolized completely via the OA to cast Onyx-18. Case 2 was a 69-year-old female who had red and swollen eyes. Angiography confirmed a DAVF of the proximal IPS, mainly supplied by the accessory meningeal artery (AMA), which drained into the CS and then into the ophthalmic vein, and the IPS was occluded. The DAVF in case 2 was embolized completely via the AMA to cast Onyx-18. After treatment, these two patients had uneventful recoveries. In our report, these two cases indicated that the DAVFs of the proximal and distal IPS shared different origins of feeding arteries. When the IPS is occluded, the transarterial approach via the main feeder, such as OA and AMA, can be feasible to cure the DAVF of the IPS.

1. Introduction

Dural arteriovenous fistulas (DAVFs) are abnormal connections between the dural arteries and dural venous sinuses or leptomeningeal veins [1]. DAVFs often involve the regions of the transverse, sigmoid and cavernous sinuses [2]. Rarely, DAVFs can be located at the inferior petrosal sinus (IPS) [3]. The IPS extends from the posterior part of the cavernous sinus (CS) to the anterior superior aspect of the jugular bulb of the internal jugular vein (IJV) [4]. The IPS is a complex anatomical structure with variations [5].

DAVF of the IPS is complex [6]. DAVF of the IPS may be associated with a high Cognard grade, and treatment is often necessary [6, 7]. Due to the deep location and complex anatomy of the IPS, surgical treatment is challenging. Similar to other intracranial DAVFs, endovascular embolization is also a good option for DAVFs of the IPS. Especially since the introduction of Onyx (Medtronic, Irvine, CA, USA), it has been considered the preferred embolic agent for DAVF due to its better control [8]. Here, we report two patients who had rare DAVFs of the IPS treated with endovascular embolization with Onyx-18.

2. Case presentation

Case 1: A 48-year-old man of Han nationality with an unremarkable medical history presented with headache and diplopia for 2

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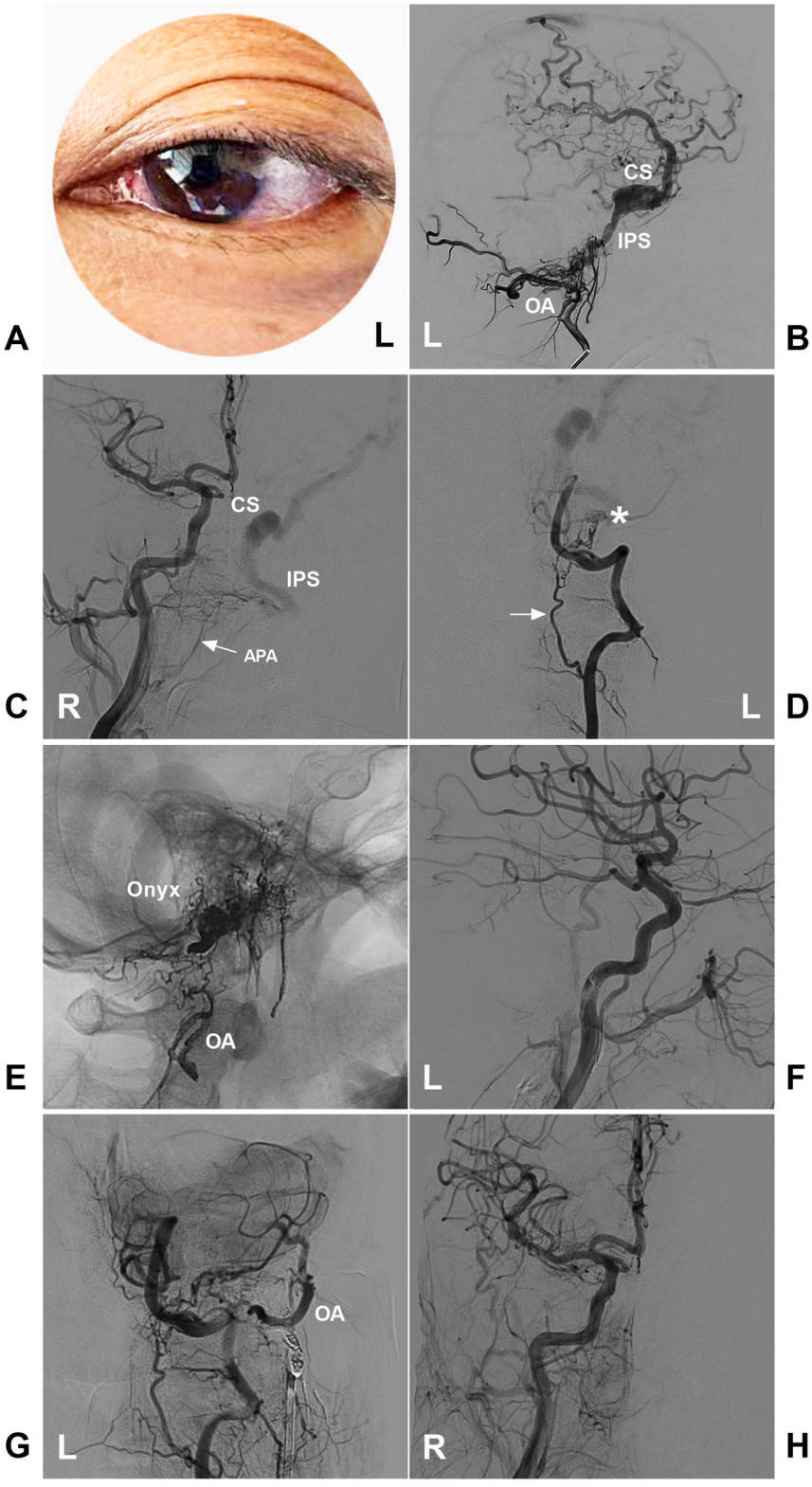

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Fig. 1. Images of case 1. A: Photo showing mild conjunctival congestion of the left eye. B: DSA of the left OA showing an IPS DAVF. The DAVF was supplied by multiple OA meningeal branches. The IPS termination was occluded and did not drain into the internal jugular sinus but retrogradely drained into the CS and then went into the cortical veins. C: DSA of the right carotid artery showing the neuromeningeal branch of the right APA (arrow) supplying the DAVF crossing the middle line. D: DSA showing that the left VA gives rise to the meningeal branch (arrow) to supply the DAVF (asterisk). E: X-ray film showing casting Onyx via OA meningeal branches after three procedures. F–H: Postoperative DSA showing that the DAVF was completely obliterated. F: DSA of the left carotid artery. G: DSA of the left VA. H: DSA of the right carotid artery. In the G image, anastomosis between the VA muscle branch and OA can be seen. **Abbreviations:** APA: ascending pharyngeal artery, CS: cavernous sinus, DAVF: dural arteriovenous fistula, DSA: digital subtraction angiography, IPS: inferior petrous sinus, L: left, OA: occipital artery, R: right, VA: vertebral artery.

months. He was healthy and denied having a history of chronic diseases. He had no history of head injury, drug abuse or surgical treatment of craniocerebral disease. On physical examination, the left eye was mildly proptotic with conjunctival congestion (Fig. 1A), and the left eyeball had limited abduction. No bruit could be auscultated over the orbit and the temporal bone. In addition, no other positive signs can be found. Head computed tomography (CT) was normal.

Digital subtraction angiography (DSA) confirmed a left DAVF, and the primary feeding artery was the meningeal branch of the left occipital artery (OA) (Fig. 1B). Other feeding arteries included the neuromeningeal branch of the right ascending pharyngeal artery (APA) (Fig. 1C) and the meningeal branch of the vertebral artery (Fig. 1D). The DAVF drained into the cavernous sinus (CS) and the cortical veins (Fig. 1B). The DAVF was Cognard Type IIa + b. Onyx-18 (Medtronic, Irvine, CA, USA) embolization via the OA was planned.

Three Onyx castings via meningeal branches of the OA were performed to obliterate the DAVF, including two procedures via an Echelon-10 microcatheter (Medtronic, Irvine, CA, USA) and one procedure via a Marathon microcatheter (Medtronic, Irvine, CA, USA) (Fig. 1E). The Onyx casting penetrated the fistula (Fig. 1F–H). After embolization, Xper-CT was performed to show the DAVF at IPS termination (Fig. 2). Postoperatively, the patient recovered gradually. One month later, he was normal.

Case 2: A 69-year-old female of Han nationality with a history of hypertension presented with redness and swelling of the right eye for 3 months. She had no history of head injury or surgical treatment of craniocerebral disease. On physical examination, her right eye was mildly proptotic with conjunctival congestion (Fig. 3A), and her eye movement was normal. No bruit could be auscultated over the orbit and the temporal bone. In addition, no other positive signs were found. Head CT was normal.

DSA of the right carotid artery confirmed a DAVF, and the draining veins included the ophthalmic vein and the cortical vein (Fig. 3B). The right accessory meningeal artery (AMA) was the primary feeding artery, and the APA was also the feeding artery (Fig. 3C). The DAVF was Cognard Type IIa + b. Onyx-18 embolization via AMA was planned. Onyx castings were performed to obliterate the DAVF, including only one procedure via a Marathon microcatheter (Fig. 3D and E). The Onyx casting penetrated the fistula (Fig. 3F). Postoperative DSA confirmed that the fistula was completely obliterated (Fig. 3G and H). After embolization, Xper-CT was performed to show the DAVF at the beginning of the IPS (Fig. 4). Postoperatively, she recovered gradually. Two months later, she was normal.

3. Discussion

Embryologically, the IPS develops as a new structure that connects the anterior head vein, precursor of the CS, and jugular bulb

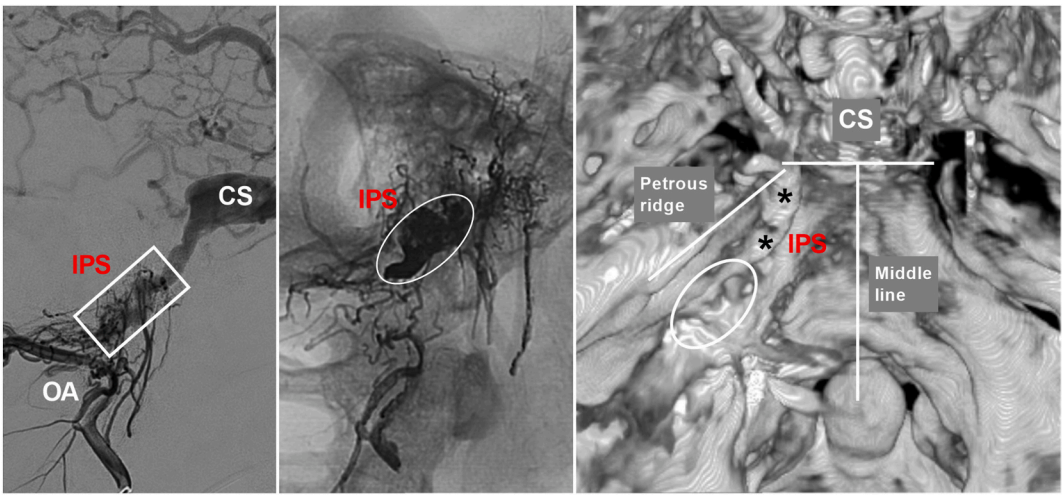

Fig. 2. Determination of DAVF location in case 1. Left panel: DSA showing multiple slim OA meningeal branches around the IPS termination (frame) and retrograde drainage into the CS. Middle panel: X-ray film showing that the IPS termination was occluded by Onyx (ellipse). Right panel: Xper-CT showing DAVF location at left IPS termination (ellipse); the asterisks indicate the Onyx in the IPS. **Abbreviations:** CS: cavernous sinus, CT: computed tomography, DAVF: dural arteriovenous fistula, DSA: digital subtraction angiography, IPS: inferior petrous sinus, OA: occipital artery.

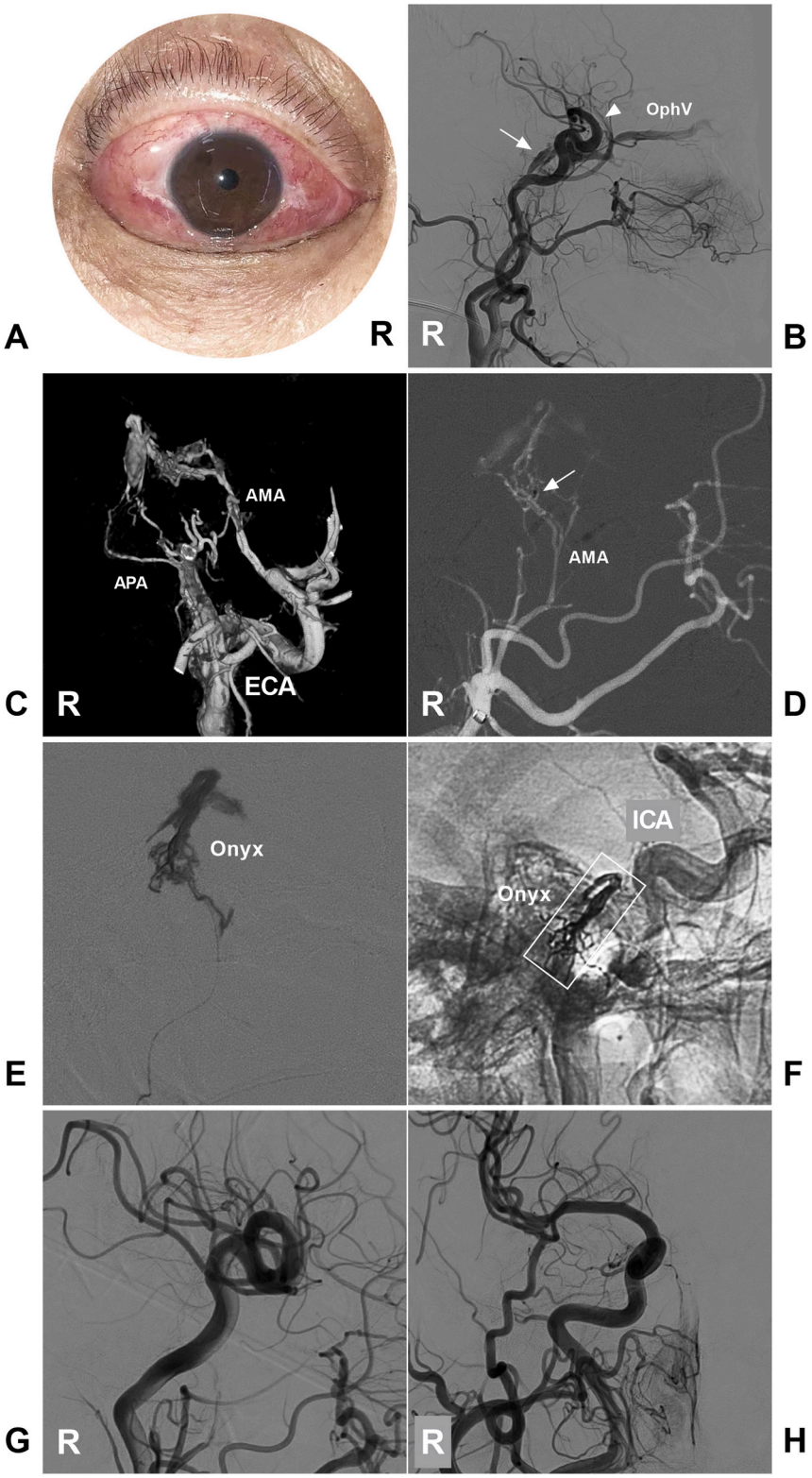

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Fig. 3. Images of case 2. A: Photo showing mild conjunctival congestion of the right eye. B: DSA of the right carotid artery showing a DAVF (arrow) draining to the OphV and cortical vein (arrowhead). C: Three-dimensional reconstructive DSA of the right ECA showing that the AMA was the main feeding artery as the DAVF, and the APA was also involved in the feeding artery. D: Roadmap navigation film of the right ECA showing that the Marathon microcatheter tip (arrow) accessed the DAVF via the AMA. E: Empty roadmap film showing Onyx casting. F: Postoperative unsubtracted DSA of the right carotid artery showing Onyx casting in the DAVF (frame). G–H: Postoperative DSA of the right carotid artery of oblique view (G) and anterior posterior view (H) showing that the DAVF was completely obliterated. **Abbreviations:** AMA: accessory meningeal artery, APA: ascending pharyngeal artery, DAVF: dural arteriovenous fistula, DSA: digital subtraction angiography, ECA: external carotid artery, ICA: internal carotid artery, OphV: ophthalmic vein, R: right.

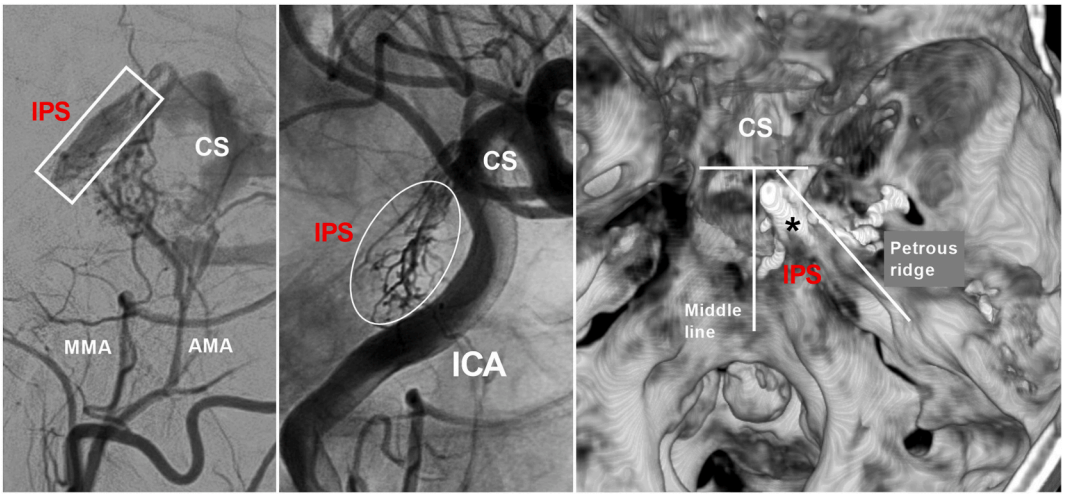

Fig. 4. Determination of DAVF location in case 2. Left panel: DSA showing multiple slim branches of AMA around the beginning of the IPS (frame) and retrograde drainage into the CS. Middle panel: unsubtracted DSA of the ICA showing the onyx casting beside the IPS beginning (ellipse). Right panel: Xper-CT showing DAVF location at the beginning of the right IPS (asterisk). **Abbreviations:** AMA: accessory meningeal artery, CS: cavernous sinus, CT: computed tomography, DAVF: dural arteriovenous fistula, DSA: digital subtraction angiography, ICA: internal carotid artery, IPS: inferior petrous sinus, MMA: middle meningeal artery.

vesicle vein [9]. Normally, the IPS drains into the CS and receives inflow from auditory structures, the brainstem, and the inferior cerebellar surface along its course prior to emptying into the IJV [4,5].

The anatomy of the IPS is complex. In Gebarski et al., which was an angiographic study of the IPS, 90% of IPSs were well formed, and size asymmetry was present in 39% of IPSs. In asymmetrical IPSs, the right IPS is larger than the left IPS in 75% of IPSs; the width and depth of the superior portion of the IPS are 6–16 mm and 2–9 mm, respectively, and the diameter of the inferior portion is 2–7 mm [10]. Miller et al. divided IPSs into four types: Type I, the IPS had anastomosis with the anterior condylar vein and then drained into the vertebral venous plexus, but the anastomosis was small; Type II, the anastomosis was large; Type III, the IPS was a network or group of small vessels, and the size of anastomosis was not important; and Type IV, there was no connection between the IPS and the IJV [11]. Type I-IV anatomy was observed in 45%, 24%, 24% and 7% of patients, respectively [9].

Similar to the hypothesis of DAVFs, due to primary IPS thrombosis, after partial recanalization, DAVFs of the IPS may develop [3,6,12,13]. In our report, the IPSs in patients 1 and 2 were occluded, which supports the evidence of DAVF formation from thrombosis and recanalization of the dural sinus.

DAVF of the IPS is very unusual. In 1990, Barnwell et al. first reported them as a separate entity [6]. Until now, no exact frequency of the DAVF of the IPS was known; they are reported sporadically, only including the reports by Barnwell et al., in 1990 [6], Yamada et al., in 1994 [3], Mironov et al., in 1994 [12], Kato et al., in 2002 [13], and Gentric et al., in 2013 [14]. Therefore, our report is important to increase the understanding of this rare entity.

DAVFs can be located at the beginning or termination of the IPS, which results in different origins of the feeding artery or the involvement of the draining vein [3,6,12,13]. Under normal circumstances, the main dural arteries of the petroclival region are from the OA, middle meningeal artery (MMA)/AMA, APA, and meningeal branches of the internal carotid artery (ICA) [15,16]. Therefore, in DAVFs of the IPS, these dural arteries can be involved. When the DAVF was close to the IPS termination, the meningeal branch of the OA and the neuromeningeal branch of the APA were often involved (case 1); when the DAVF was close to the CS at the beginning of the IPS, the petrous and posterior branches of the MMA/AMA were often involved (case 2). In case 1, the neuromeningeal branch of the contralateral APA and meningeal branch of the ipsilateral vertebral vein (VA) were involved, which was uncommon.

The venous drainage of the DAVFs of the IPS was determined by IPS occlusion. If the distal IPS was occluded, the venous drainage was superiorly retrograde through the IPS into the CS; if the proximal IPS was occluded, the venous drainage may be an inferior antegrade flow to the sigmoid sinus and IJV. If the IPS is patent, venous drainage may be bilateral. In Barnwell et al., who reported six DAVFs of the IPS, three had bidirectional venous drainages, two drained inferiorly into the jugular bulb, and one drained superiorly

into the CS and superior ophthalmic vein (OphV) [6]. In our two cases, the draining veins were all toward the CS.

DAVFs of the IPS termination need differential diagnosis with 4 types of DAVFs in the lateral foramen magnum region, including anterior condylar confluence (ACC) and anterior condylar vein (ACV) DAVFs, posterior condylar canal (PCC) DAVFs, marginal sinus DAVFs, and jugular foramen DAVFs [17]. These DAVFs in the lateral foramen magnum region commonly had lower locations than the DAVF of the IPS termination, and the angioarchitectures were different. Moreover, their angioarchitectures were more complex than the DAVF of the IPS termination.

ACC and ACV DAVFs share a similar angioarchitecture; the APA is often the main feeding artery, and the IJV and/or vertebral venous plexus often act as the main draining veins. In PCC DAVFs, the APA is often the main feeding artery, and the posterior condylar vein often acts as the main draining vein into the IJV or suboccipital cavernous sinus. In marginal sinus DAVFs, the APA and OA are often the most common feeders, and venous drainage mainly was into the jugular bulb. In jugular foramen DAVFs, the APA is the most common feeder, and the draining vein is mainly into the IJV [17]. These DAVFs in the lateral foramen magnum region can drain into the venous systems of the brainstem and vertical cord, resulting in neurological deficits of the brainstem and vertical cord, which is different from the DAVFs of the IPS termination [18].

The signs and symptoms of a DAVF of the IPS were determined by the venous drainage pattern. When a DAVF drains into the CS, the clinical presentations are similar to those of a DAVF involving the CS and include ipsilateral exophthalmos, bruit, decreasing vision and abducens nerve paresis, which present with false-localizing ocular symptoms [3,13]. Even rarely, due to patent intercavernous channels, the symptoms can be bilateral or contralateral [12].

The symptoms of patient 2 were similar to those of CS DAVF and cortical vein reflux. The symptoms of patient 1 were unique; after a high-flow arteriovenous shunt into the CS, the blood flow was directly diverted into the superficial cortical vein bed of the cerebral hemisphere, resulting in extensive venous congestion [19].

High Cognard grade DAVFs with cortical venous drainage, such as in cases 1 and 2, are associated with bleeding risk, especially for patient 1, who had extensive venous congestive encephalopathy, and treatment is necessary and needs to be prompt to reduce the bleeding risk and cognitive function impairment [19]. For the treatment of DAVFs, endovascular embolization is a good choice, and the approaches include either transarterial, transvenous, or both combined [3,13].

For DAVFs of the IPS, the choice of endovascular approach was determined by the draining vein pattern. If the IPS was patent, transvenous coiling of the IPS with/without transarterial embolization was a good choice. If the IPS is occluded, the transarterial approach may be the only choice. In Barnwell et al., six DAVFs of the IPS were reported. Depending on whether the IPS was patent, five patients accepted endovascular embolization, and of them, one accepted transarterial embolization, two accepted transvenous embolization, and two accepted combined transarterial/transvenous embolization [6]. In DAVFs with IPS occlusion, if the feeding arteries are too slim to perform transarterial embolization, the transorbital approach can be chosen. For instance, in Gentric et al.'s report of DAVFs of the IPS that had IPS occlusion, direct puncture of the OphV accessed the IPS past the CS, and after coiling the IPS, the DAVF was cured [14].

Our two patients had to undergo a transarterial approach due to IPS occlusion. Many dural feeding arteries can be used to perform embolization for a DAVF of the IPS, depending on the hyperplastic status of the main feeding artery. In patient 2, the AMA was hyperplastic and acted as the main feeding artery; it served as the ideal transarterial approach. The AMA may arise from the MMA or the maxillary artery, and it traverses the foramen ovale to supply the semilunar ganglion and adjacent dura as an important addition to the MMA [20,21]. For DAVFs with the MMA/AMA as a supplier, the MMA/AMA are often thick and straight and can be used as the gold standard to cast Onyx [22]. In our previous report, in all of the DAVFs with MMA/AMA as the feeder to cast Onyx, complete embolization was achieved in the first procedure in 64.4% of the patients, and success was eventually achieved after the first attempt or a subsequent attempt in 74.1% of the patients [2].

For patient 1, the OA was chosen as the transarterial approach because the MMA/AMA were not the supplying vessels, but complete embolization was still achieved. The OA was not a poor artery for the embolization of a DAVF [23]. In particular, a very high complete occlusion rate of the DAVF was obtained when using Scepter dual-lumen balloon catheter (MicroVention, Aliso Viejo, CA, USA)-assisted embolization via the OA [20]. However, caution should be taken when using the Scepter balloon catheter to perform embolization via the OA because the Scepter balloon catheter is stiff, which reduces its trackability in a tortuous OA. Using transosseous Onyx penetration with a Scepter catheter in the proximal OA can be dangerous due to the potential presence of either seen or unseen occipito-vertebral anastomoses [21]. For instance, in case 1, the VA had dangerous anastomosis with the OA (Fig. 1G). Because the Marathon microcatheter could access the fistula, we did not choose the Scepter balloon catheter.

Generally, endovascular embolization via transarterial or transvenous approaches was effective, including in the two patients in our report [6,12–14]. However, not all endovascular embolization can be successful; thus, radiotherapy can be a candidate therapy in those instances. For instance, Yamada et al. reported that embolization via the OA and APA was performed to obliterate a DAVF of the IPS, but the embolization was incomplete, and later radiotherapy cured the DAVF [3]. In addition, during transarterial embolization for DAVF of the IPS with liquid embolic material, the potential risk of cranial nerve palsy due to the migration of liquid embolic material to the vasa nervorum had to be considered [24].

4. Conclusion

In this report, these two cases indicated that the DAVFs of the proximal and distal IPS shared different origins of feeding arteries. When the IPS is occluded, the transarterial approach via the main feeder, such as the OA and AMA, can be feasible to cure DAVFs of the IPS. However, because this is a report, the conclusion should be cautiously interpreted. In addition, no cases of surgical treatment were compared with cases of endovascular embolization, which was also a limitation of our study.

Ethics approval and consent to participate

Ethics approval was not necessary in the authors' institution, as the present study is a case report. All methods were performed in accordance with the relevant guidelines and regulations. Written informed consent was obtained from the participants.

Consent for publication

Written informed consent for publication was obtained from all the participants (or their first-degree relatives).

Author contribution statement

All authors listed have significantly contributed to the investigation, development and writing of this article.

Data availability statement

Data will be made available on request.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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