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Case Report

A Case of Cavernous Sinus Dural Arteriovenous Fistula Draining Solely to the Superior Ophthalmic Vein with Normal Cerebral Venous Flow from the Superficial Middle Cerebral Vein to the Inferior Petrosal Sinus due to a Septum in the Cavernous Sinus

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Objective: We report here an atypical case of cavernous sinus dural arteriovenous fistula (CSDAVF) with a septation that separates the cavernous sinus (CS) into two components, namely, normal cerebral venous drainage and shunted blood drainage into the superior ophthalmic vein (SOV) alone. The CSDAVF was successfully treated by selective transvenous embolization (TVE) through the septum with the trans-inferior petrosal sinus (IPS) approach.

Case Presentation: A 74-year-old woman presented with right exophthalmos and tinnitus on the right side. Neuroradiological examination showed CSDAVF mainly supplied by multiple feeders from the bilateral ascending pharyngeal artery and meningohypophyseal trunk with a shunted pouch located medial-dorsally to the right CS. Blood from the CSDAVF drained via the anterior component of the CS to the right SOV only. Normal cerebral venous blood from the right superficial middle cerebral vein drained through the dorsolateral component of the right CS into the right IPS. These findings suggest that a septal barrier exists between the outflow tract of the dural arteriovenous fistula and the normal cerebral venous outflow tract within the CS. The CSDAVF was successfully treated by selective TVE through the septum with the trans-IPS approach after detailed evaluation of 3D rotational angiography (3DRA) and MRA/MR venography (MRV) cross-sectional images. The patient's symptoms improved, and she was discharged uneventfully.

Conclusion: Septation within the CS can completely separate the drainage route of the CSDAVF from the normal cerebral drainage route. Successful catheterization to the shunted pouch through the septum with the IPS approach and selective embolization were possible with detailed evaluation of anatomy on MRA/MRV cross-sectional images and 3DRA images.

Keywords ▸ CSDAVF, septum in the cavernous sinus, normal cerebral venous flow, TVE, SOV

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Introduction

Blood from the cavernous sinus (CS) normally drains posteroinferiorly via the inferior petrosal sinus (IPS) and laterally via the emissary vein of the foramen ovale and the pterygoid plexus into the internal jugular vein (IJV) and/or the external jugular vein. In cases of cavernous sinus dural arteriovenous fistulas (CSDAVF), as the disease progresses, obstructive changes occur in the IPS and other antegrade drainages, resulting in reflux into the superior ophthalmic vein (SOV) and cerebral veins.[1] On the other hand, it is well known that the CS develops from the primitive

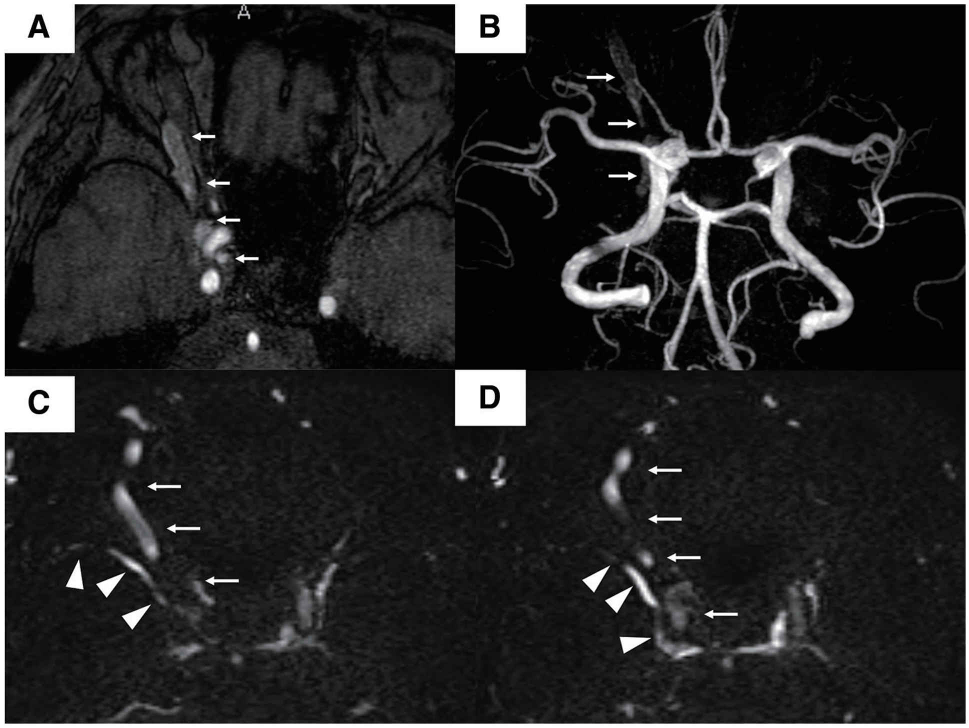

Fig. 1 3D TOF MRA axial source image (**A**) and MIP image (**B**) show a high signal in the right CS and dilated right SOV (white arrows). The source image shows that the high signal leading to the SOV was stronger in the medial part of the right CS. This suggests a shunted pouch located at the medial portion of the right CS. MRV oblique–axial thin MIP images (**C** and **D**) show that the SMCV joins the lateral part of the CS (white arrowheads) and the medial component of the CS continues to the SOV (white arrows). Both components of the CS are separated (**C** and **D**). CS: cavernous sinus; MIP: maximum intensity projection; MRV: MR venography; SMCV: superior middle cerebral vein; SOV: superior ophthalmic vein; TOF: time of flight

venous plexus, and therefore, the CS often contains trabeculations that may almost completely divide the cavernous.[2,3) A septation can separate the drainage route of CSDAVF from normal cerebral drainage of the CS to the IPS. We report here a relatively rare case of CSDAVF draining retrogradely into the SOV, which was separated by a septum from the CS of normal cerebral venous drainage to the IPS without obstructive changes.

Case Presentation

A 74-year-old woman presented with right exophthalmos and tinnitus on the right side for a few months. Although the tinnitus spontaneously disappeared within a month, MRI suggested right CSDAVF. She was referred to our department for the treatment of the CSDAVF. On admission, the patient was clear and had right exophthalmos, chemosis of the right eye, decreased vision, and omnidirectional diplopia. Maximum intensity projection and axial source images of 3D time of flight MRA showed high signal structures in the medial part of the right CS contiguous with the dilated right SOV (**Fig. 1A** and **1B**). Axial images of MR venography (MRV) showed that the superficial middle cerebral vein (SMCV) connects to the lateral part of the CS, which was separated from the drainage routes of the CSDAVF to the SOV (**Fig. 1C** and **1D**). Cerebral angiography showed the CSDAVF mainly supplied by multiple feeders from the right ascending pharyngeal artery (APA) and the meningohypophyseal trunk, which form a shunted pouch in the posteromedial portion to the right CS (**Fig. 2A**–**2D**). Shunted blood from the CSDAVF drained anteriorly and inferiorly along the medial surface of the cavernous portion of the internal carotid artery (ICA), and then anterolaterally to connect to the right SOV alone (**Fig. 2A**–**2F**). No other drainage routes of the CSDAVF were observed on angiography. The venous phase of right common carotid artery angiography showed normal cerebral venous drainage from the right SMCV through the right CS and the right IPS to the IJV, suggesting a septum existed between the drainage route of the CSDAVF and the normal cerebral venous drainage route within the CS (**Fig. 2E**–**2H**). Multiplanar reconstruction images of 3D rotational angiography (3DRA) of the right common carotid artery depicted the detailed anatomical relationships of the shunted pouch with surrounding

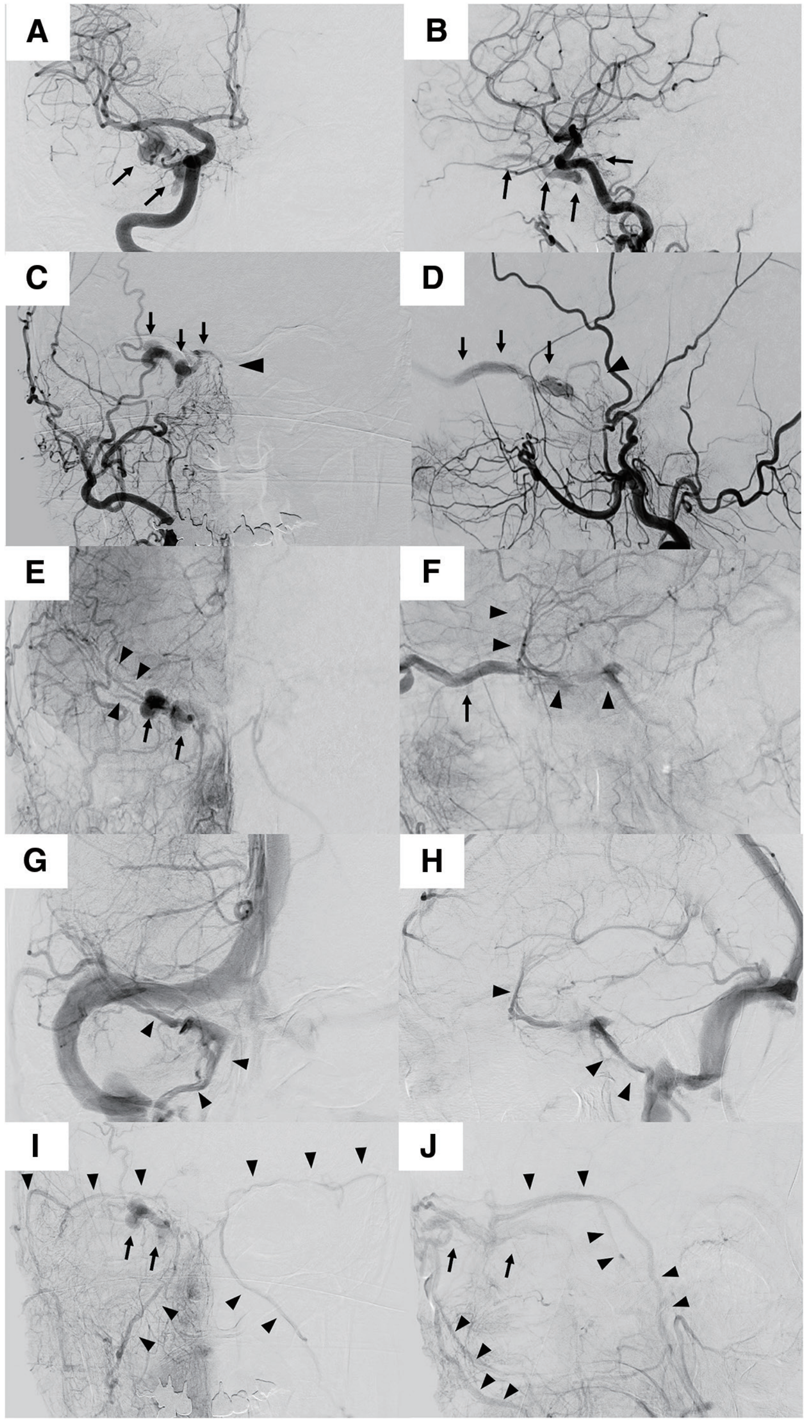

Fig. 2 Right internal carotid angiography (**A** and **B**) and right external carotid angiography (**C** and **D**) at the arterial phase showed a CSDAVF supplied by the right MHT (**A** and **B**), APA, MMA, artery of foramen rotundum, and artery of pterygoid canal (**C** and **D**). The shunt points were located in the right dorsal side of the CS and medial to the right ICA (black arrowheads in **C** and **D**). The CSDAVF drains to the right SOV alone (black arrows in **A**–**D**). The right common carotid angiography at the venous phase shows normal cerebral venous flow from the right SMCV through the right CS to the right IPS (black arrowheads in **E**–**H**). Drainage routes from the CSDAVF via the SOV (black arrows in **E** and **F**) and the small facial vein and superficial temporal vein are also noted. The right external carotid angiography at the venous phase shows reflux cerebral venous flow to the SOV (black arrows in **I** and **J**) and drains into small bilateral relatively small facial veins and superficial temporal veins (black arrowheads in **I** and **J**). APA: ascending pharyngeal artery; CSDAVF: cavernous sinus dural arteriovenous fistula; ICA: internal carotid artery; IPS: inferior petrosal sinus; MHT: meningohypophyseal trunk; MMA: middle meningeal artery; SMCV: superior middle cerebral vein; SOV: superior ophthalmic vein

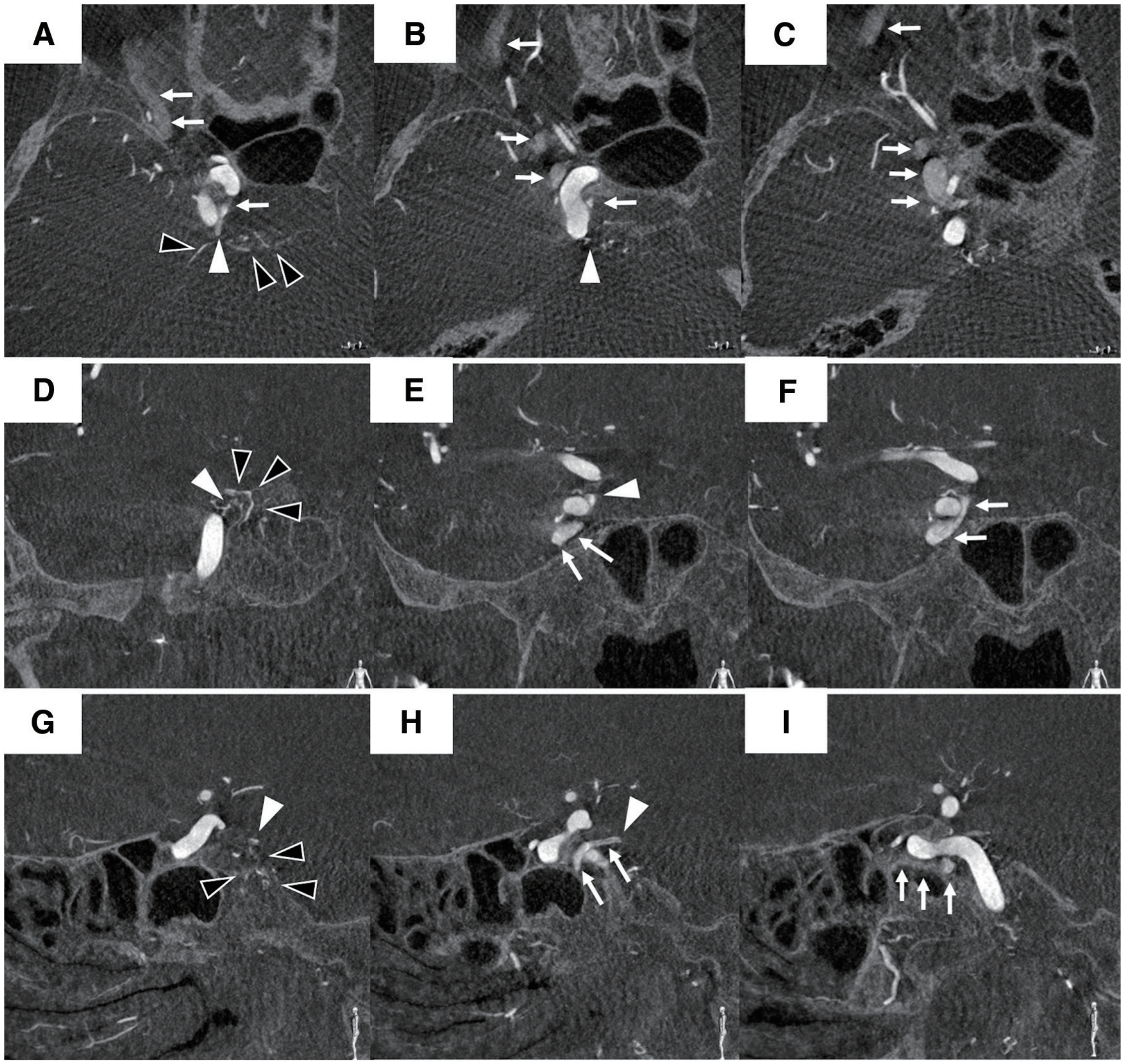

Fig. 3 Axial (**A**–**C**), coronal (**D**–**F**), and sagittal (**G**–**I**) MPR images of 3DRA show the detailed anatomy of the feeders (black arrowheads surrounded by white), shunted pouch (white arrowheads), drainage vein (white arrows), and surrounding structures. 3DRA: 3D rotational angiography; MPR: multiplanar reconstruction

structures (**Fig. 3**). After evaluation of those images, we performed selective transvenous embolization (TVE) of the shunted pouch for the treatment of the CSDAVF (**Fig. 4**). Under general anesthesia, a 4F guiding sheath (FUBUKI; Asahi Intecc, Aichi, Japan) was placed into the right external carotid artery, and a 6F guiding sheath (FUBUKI) was guided into the right IJV using a right femoral approach. The embolization procedure underwent systemic heparinization with activated clotting time monitoring. A microcatheter (Eschelon-10; Medtronic, Minneapolis, MN, USA) was introduced through the 6F guiding sheath into the right CS via the right IPS using a 0.014 in. microguidewire (CHIKAI black; Asahi Intecc), and then it was introduced through the presumed septum into the shunted pouch deeply close to the shunt points (**Fig. 5A** and **5B**). Subsequently, selective TVE of the shunted pouch with microcoils was performed. Immediately after placement of 10 microcoils, the CSDAVF disappeared (**Figs. 4C** and **5C**–**5F**). Normal venous return from the right SMCV via the right CS was preserved (**Fig. 5G** and **5H**). Postoperatively, the patient's symptoms improved, and she was discharged uneventfully (to home). Three months after embolization, her symptoms completely disappeared and MRA showed no recurrent CSDAVF.

Discussion

The CS is a dural sinus surrounded by the periosteum and dura mater, which extends from the superior orbital fissure to the pyramidal part of the temporal bone on the dorsal

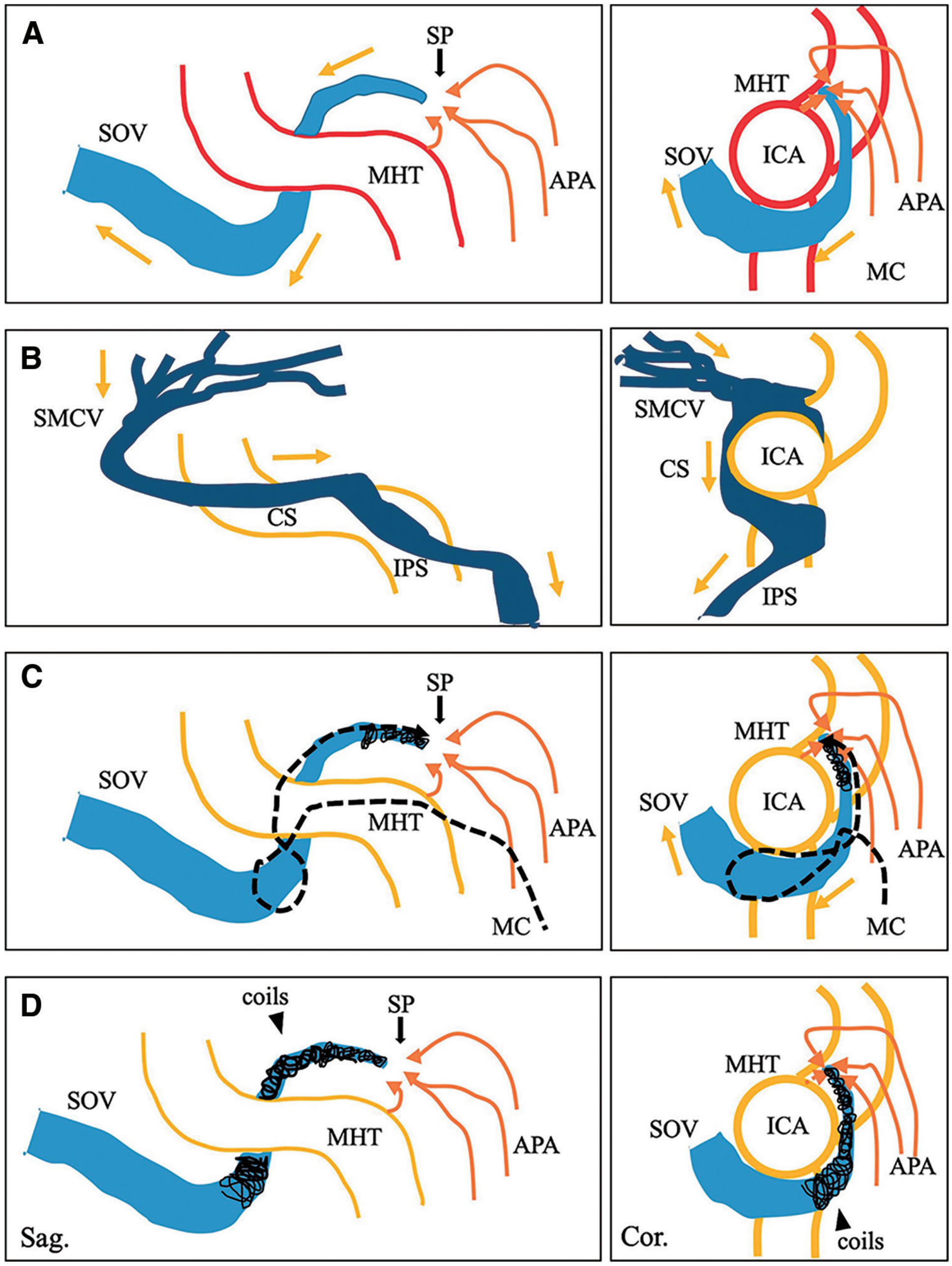

Fig. 4 Schematic drawings of the anatomical and hemodynamic features of the presented case of CSDAVF and treatment strategy. A shunted pouch receiving blood from multiple feeders on the dorsal and medial sides of the CS, which runs anterior–inferiorly along the medial surface of the right ICA and turns anterolaterally at the inferior aspect of the ICA to join the SOV (**A**). The normal venous drainage from the SMCV runs posteriorly in the lateral part of the CS to join the IPS (**B**). Microcatheter entry path (black dotted arrow) and starting site of coil embolization (**C**). The area embolized with microcoils is from the shunted pouch and partially from the medial component of the CS. This eliminated the CSDAVF (**D**). APA: ascending pharyngeal artery; Cor.: coronal image; CS: cavernous sinus; CSDAVF: cavernous sinus dural arteriovenous fistula; ICA: internal carotid artery; IJV: internal jugular vein; IPS: inferior petrosal sinus; MC: microcatheter; MHT: meningohypophyseal trunk; Sag.: sagittal image; SMCV: superior middle cerebral vein; SOV: superior ophthalmic vein; SP: shunt point

side.[4] The CS often has many trabeculations, as a remnant of its primary feature of the venous plexus. It is well known that the CS is formed by fusion of three different venous systems during its development process. First, the prootic sinus, a partial remnant of the primary head sinus, receives the SOV. Second, osseous veins from the medial part of the sphenoid bone join the prootic sinus to form the primary CS. Third, the primitive tentorial sinus receiving the superficial and deep telencephalic veins fuses to the lateral part of the primary CS and forms the adult configuration of

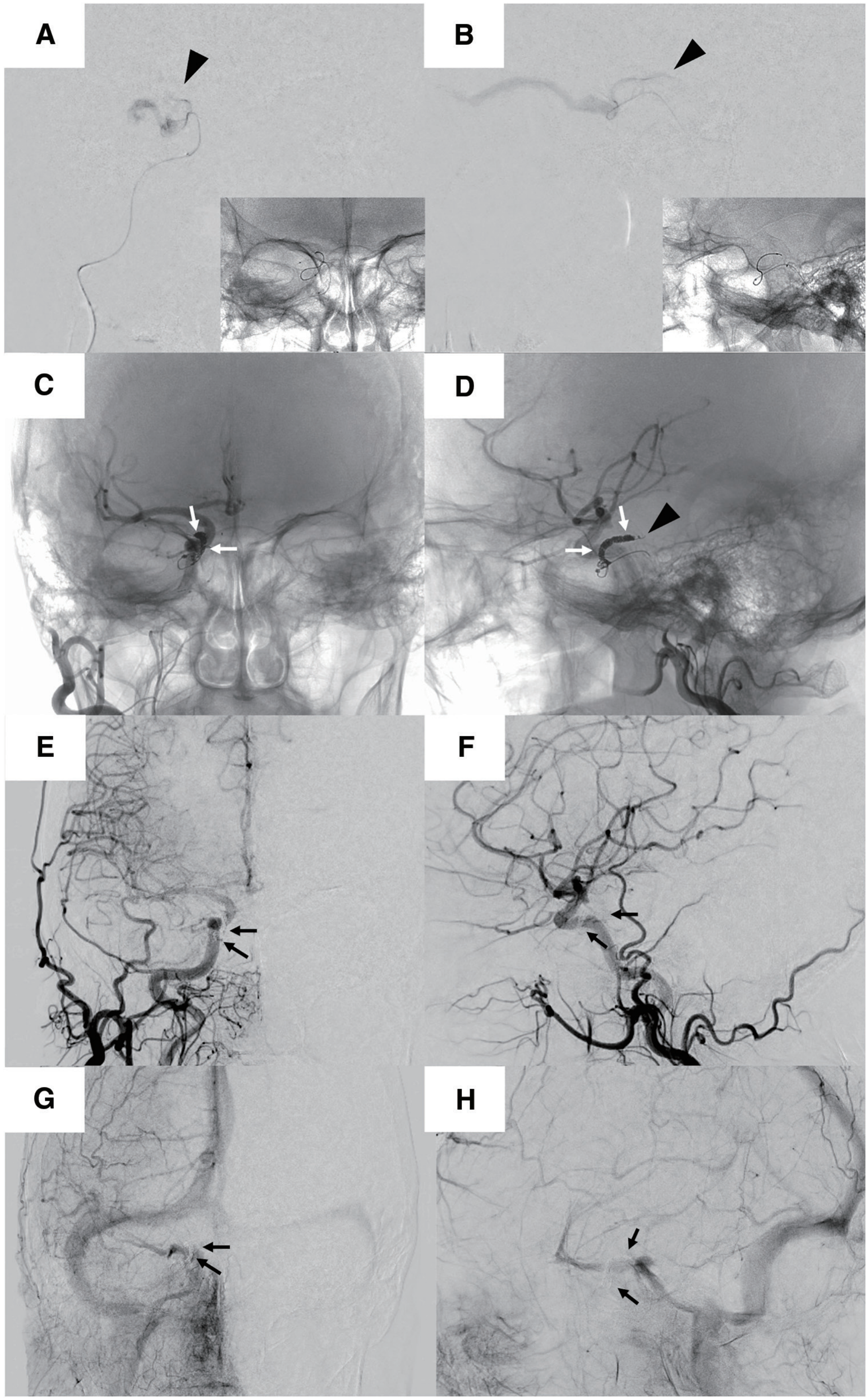

Fig. 5 A microcatheter was guided and placed via the ipsilateral IPS through the septum in the CS to the distal portion of the shunt pouch (black arrowheads) (**A** and **B**). Microcoils were placed in the shunted pouch and partially in the medial component of the CS (white arrows) from the shunt point (black arrowhead) (**C** and **D**). CCAG immediately after selective TVE shows complete occlusion of the CSDAVF (**E** and **F**). CCAG at the venous phase shows that normal venous drainage through the right CS tract is preserved (**G** and **H**). Black arrows in **E**–**H** indicate microcoils. CCAG: common carotid artery angiography; CS: cavernous sinus; CSDAVF: cavernous sinus dural arteriovenous fistula; IPS: inferior petrosal sinus; TVE: transvenous embolization

CS.[5)] Among them, the telencephalic veins become SMCV, and the uncal vein, which is responsible for the normal cerebral venous drainage to the CS, is particularly important, and various anatomical variations have been reported by anatomical and diagnostic imaging techniques.[2,3,6–13)]

In the present case, the shunted pouch located posteromedially to the CS receives blood from CSDAVF via duro-osseous veins, which run anteriorly along the medial surface of the ICA to join the SOV. This drainage venous route may represent the primary CS. The SMCV flows into the lateral part of the CS and forms a normal cerebral drainage route through the IPS, indicating that it is an embryologically new part of the CS. Both compartments may be separated, but incompletely at the initial stage of the development of CSDAVF because the patient initially had tinnitus, which disappeared later without occlusion of the IPS. The ocular symptoms worsened simultaneously. This suggests that compartmentalization was completed when the tinnitus disappeared. The postulated pathophysiology of the compartmentalization is as follows. First, inflammation and/or thrombus obstruct local venous perfusion in the CS (probably in some duro-osseous venous channels), resulting in increased venous pressure and angiogenic factors, and an arteriovenous shunt was formed in the bone, periosteum and/or dura mater, including walls of CS. At this time, the CSDAVF drained through the IPS to the IJV, and the tinnitus was present. Next, progression of DAVF led to further elevation of venous pressure and angiogenesis, resulting in obstruction of the CS by thrombus and proliferation of fibrous tissue due to high-flow vasculopathy, which caused complete septation in the CS. At that time, the tinnitus disappeared and there was no more outflow of DAVF to the IPS; it was solely to the SOV.

The latero-cavernous spinus (LCS) is a variant drainage route of the SMCV (and often the uncal vein), which is located laterally to the CS and drains into either the superior petrosal sinus, pterygoid plexus, or the posterior part of the CS. It was a type of remnant of the primitive tentorial sinus, and a dural layer separates the LCS from the CS. For this case, it was obviously demonstrated on frontal view of the right internal carotid angiography that the SMCV drains directly into the lateral portion of the CS without dural septation. This finding was unlikely to the LCS.[12,14)]

Regarding the TVE technique for this type of CSDAVF with septation, it requires successful navigation of a microguidewire and a microcatheter through the septum into the shunted component in the CS. Because the access route from the IPS to the shunted component through the septum cannot be demonstrated on angiography, it should be assumed based upon the anatomical relationship between the shunted component and the component of normal cerebral drainage depicted on pretherapeutic images, including cross-sectional images from 3DRA and MRA/MRV. For this case, the drainage route from the SMCV joins the lateral portion of the CS, and the CSDAVF drainage runs anteroinferiorly and then anterolaterally to join the SOV; therefore, we sought a "hole" that once existed in the septum by directing a microguidewire posteromedially from the lateral part of the CS toward the shunted component. It may be useful to use a high-torque transmissive and shapable microguidewire for this purpose. After successful navigation of the microguidewire, the subsequent procedure is similar to the usual selective TVE technique, advancing a microcatheter as distally close to the shunt points as possible, packing the shunted pouch with small and soft coils. If the CSDAVF remained after TVE via this transseptal approach, the DAVF may be drained into the normal CS via the penetrated septum. This potentially causes cortical venous reflux when the steno-occlusive change further occurs in the antegrade drainage from the CS via the IPS.

Trans-SOV approaches from the facial or superficial temporal vein or by direct puncture are alternative options. Both the facial vein and the superficial temporal vein were too small and tortuous for the transvenous approach in the present case; therefore, a direct trans-SOV approach is the alternative approach route when the trans-IPS approach fails or is difficult. Trans-SOV has the advantage of allowing for a microcatheter to easily be navigated to the shunted pouch, which is usually located in the posterior portion of the CS. However, it has the potential risk of orbital and/or palpebral hemorrhage during and after the procedure, and of worsening of visual acuity due to increasing central retinal venous pressure with blocking of the SOV drainage by catheterization.

Conclusion

We demonstrated a relatively rare case of CSDAVF in which normal cerebral venous flow was maintained by the septal wall within the CS, but reflux into the SOV resulted in ocular symptoms. Preoperative imaging evaluation of cross-sectional 3DRA and MRA/MRV images is useful for guiding the microcatheter to the shunted pouch via the septal wall that exists within the CS.

Disclosure Statement

The authors declare that they have no conflicts of interest.

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