A Shunt of the Diploic Vein of the Orbital Roof Accompanying a Cavernous Sinus Dural Arteriovenous Fistula: A Case Report

Rie Aoki, Kittipong Srivatanakul, Takahiro Osada, Takatoshi Sorimachi, and Mitsunori Matsumae

Objective: In cavernous sinus dural arteriovenous fistula (CSdAVF), multiple shunt points are often demonstrated on the angiogram. In some cases, shunt points may be located not only at the CS itself but at the part surrounding the CS. We report a rare case of CSdAVF with a shunt at the diploic vein of the orbital roof (DVOR).

Case Presentation: A 64-year-old man presented with exophthalmos, conjunctival injection, and abducens nerve palsy. The shunt points were suspected to be located at the anterior lateral part and posterior medial part of the left CS on the angiogram and the drainage route was the left superior ophthalmic vein (SOV) and the veins of the posterior fossa via the left petrosal vein. We embolized the suspected shunt points in the CS transvenously. Another shunt point was identified anterior to the CS by superselective angiography. By meticulous review of the angioarchitecture, using 3D rotational angiography and multi-planar reconstruction (MPR) images, another shunt point was found at the DVOR fed by branches of the ophthalmic artery and sphenopalatine artery branches.

Conclusion: We report a case of CSdAVF with multiple shunt points including a shunt located at the DVOR. To the best of our knowledge, this is the first case report with CSdAVF with a shunt point at the DVOR. In cases of CSdAVFs, careful analysis of the angioarchitecture using 3D rotational angiography, MPR images, and superselective angiography is necessary to elucidate the presence of shunts in rare locations.

Keywords: diploic vein of the orbital roof, cavernous sinus dural arteriovenous fistula, endovascular treatment

Introduction

Cavernous sinus dural arteriovenous fistula (CSdAVF) is the most common type of dAVF in Japan.1) Embolization of the whole sinus has been recommended.2) However, a large amount of coils and longer procedure time are required to obtain complete obliteration of the shunt with transvenous sinus packing. Recently, superselective embolization of the shunted parts of the CS has been introduced.3) The shunt points in CSdAVF are often multiple.4)

In rare cases, the shunt is not located on CS itself but outside of the CS.5,6) We report a rare case of CSdAVF with one of the shunts localized at the diploic vein of the orbital roof (DVOR).

Case Presentation

A 64-year-old man presented with left exophthalmos, conjunctival injection, and abducens nerve palsy. Time of flight MRI demonstrated a high-intensity lesion around the left CS and dilation of the left superior ophthalmic vein (SOV). Angiography showed two shunt points, at the anterolateral and at the posteromedial wall of the left CS. The shunt drained through the left SOV and the veins of the posterior fossa via a bridging vein (Fig. 1).

A 4 Fr intermediate catheter was positioned into the inferior petrosal sinus through a 6 Fr Roadmaster guiding catheter (Goodman Co., LTD., Aichi, Japan). An Excelsior SL-10 (Boston Scientific, Natick, MA, USA) microwire catheter, through the 4 Fr catheter, was introduced into the anterior shunt point which was causing the reflux to the SOV.
We embolized this part of the CS with coils. After embolization of the anterior part, another shunt point anterior to that became apparent by a superselective angiography of the left sphenopalatine artery. This newly discovered shunt point was later on identified to be at the DVOR. Although we tried to embolize the DVOR, it was not reachable transvenously due to the previously placed coil mass. We alternatively embolized the distal branches of the left sphenopalatine artery with coils and injected pieces of Gelfoam (Upjohn, Kalamazoo, MI, USA) at the third portion of the internal maxillary artery to obliterate the feeding arteries supplying the shunt. Subsequently, additional coil embolization to the posterior part of the CS including the posterior shunt point was done. However, a faint early venous filling of the SOV remained on the control left internal carotid artery angiogram done immediately after the embolization, which, after being retrospectively analyzed, is suggestive of a small residual shunt at the DVOR (Fig. 2). The symptoms improved completely 3 days after the procedure. A cerebral angiography performed 3 months after the procedure demonstrated complete disappearance of the shunt. Meticulous re-evaluation of 3D rotational angiogram and multi-planar reconstruction (MPR) images led to the discovery of the shunt point at the DVOR (Fig. 3). MPR revealed transosseous feeders from the left external carotid artery converging to the shunt point at the DVOR (Fig. 4). 3D rotational angiogram demonstrated that the DVOR had a connection to the sinus of the lesser sphenoid wing and the anterolateral part of the CS (Fig. 3). The DVOR had no direct communication to the SOV. The SOV joined the CS at the anteroinferior aspect. In other words, the CS is the only connection between the DVOR and the SOV. The persistent reflux to the SOV at the end of the procedure was assumed to be caused by not having enough coil density in the part of the CS between the DVOR and the SOV. However, as there was no other drainage route, the coils promoted further thrombosis and led to the complete disappearance of the shunt.

Discussion

In CSdAVF, shunt points located outside the CS are rare. Watanabe et al.7) reported a case of CSdAVF with multiple shunts involving the CS and the sphenoparietal sinus treated in separate sessions. In a case series of CSdAVF reported by Satow et al.,3) 7 of 14 shunt points were mentioned to be located outside the CS, at the paracavernous sinus but these shunts can be considered to be adjacent to the CS wall. Remote shunts mimicking CSdAVF have been reported. Hwang et al.6) reported two cases of a shunt located at the ophthalmic sheath. Kobkitsuksakul et al.5) reported 10 cases of extra-CS shunts in 350 patients initially diagnosed as CSdAVF, two of these extra-CS shunts were located close to the CS, one intraorbital and another at the lesser sphenoid wing. In patients with CSdAVF, multiple shunt points are frequently observed.4)

In planning the embolization for patients with dAVF, in principle, we analyze the angioarchitecture of dAVF meticulously using rotational angiography to clarify the exact shunt points. Based on the preprocedural analysis of the present patient, we expected the shunt points to be at the anterior and posterior part of the CS. Immediately after
Fig. 2  (A) A left external carotid angiogram performed after the embolization of the shunt point located at the anterior part of the CS. The angiogram shows a separate shunt point located at the DVOR (arrow) (lateral view). (B) Superselective angiogram of the left sphenopalatine artery (left oblique view). The tip of the microcatheter (arrow) was introduced into the left sphenopalatine artery. This angiogram shows a shunt point (arrowhead) at the DVOR. (C) After transarterial embolization of the sphenopalatine artery branches, a left external carotid angiogram shows the disappearance of the blood supply from the external carotid artery (lateral view). (D) A left internal carotid angiogram shows that the reflux to the SOV (arrow) still remains in the late arterial phase (lateral view). DVOR: diploic vein of the orbital roof; CS: cavernous sinus; SOV: superior ophthalmic vein.

Fig. 3  (A) Left superior oblique view of the fusion of the ECA and ICA 3D rotational angiogram showing the shunt point (arrowhead) located at the anterior part of the DVOR (asterisk) and the reflux to the SOV (double arrowheads). The shunt at the DVOR drains through a venous connection at the tip of the anterior clinoid process (dotted line) directly into the lateral part of the CS (arrow) dorsal to the entrance of the SOV (double arrow). (B) Left superior oblique view of the fusion of the ECA, ICA, and coils (light blue) 3D rotational angiogram showing the location of the coils after embolization. Coils were placed into the entrance of the SOV, the lower part, the medial part and the posterolateral part of the CS. No coils were placed into the DVOR nor into the connection of the DVOR to the CS. (C) MPR (2.3-mm thick) oblique image shows transosseous feeders (arrow) converging to the shunt point (arrowhead) located at the DVOR (asterisk). This image clearly demonstrates that this vein is completely surrounded by the bone. (D) MPR (6-mm thick) axial image demonstrates the feeders (arrow), the shunt point (arrowhead), the DVOR (asterisk), and the sinus of the lesser sphenoid wing (double arrowheads). The outlet to the CS (not seen here) through the tip of the anterior clinoid process is at the point of the curved arrow. (E) The left posterior oblique view of the fusion of the ECA with the bone 3D rotational angiogram shows that the DVOR (asterisk) has a connection to the sinus of the lesser sphenoid wing (arrow). ECA: external carotid artery; ICA: internal carotid artery; DVOR: diploic vein of the orbital roof; CS: cavernous sinus; MPR: multi-planar reconstruction; SOV: superior ophthalmic vein.
A Shunt of the Diploic Vein of the Orbital Roof

A Shunt of the Diploic Vein of the Orbital Roof

embolization of the anterior part of the CS including the shunt already identified, another shunt, which was presumably located more anterior to the known embolized shunt, became apparent. The exact position of this shunt point could not be confirmed during the procedure. Meticulous retrospective review of the 3D rotational angiogram and MPR images revealed another additional shunt point at a diploic vein running inside the bone of the orbital roof (Fig. 3). This vein fits the description of the DVOR reported by Ruiz et al. He demonstrated that this vein was one of the tributaries of the sinus of the lesser sphenoid wing. The DVOR can connect with frontal diploic veins and supraorbital veins (Fig. 4).2) Feeders supplying the shunt originated from the ipsilateral ophthalmic artery and bilateral sphenopalatine artery branches. The shunt at the DVOR drains through a venous connection at the tip of the anterior clinoid process directly into the lateral part of the cavernous sinus dorsal to the entrance of the SOV. In this case, the DVOR also has a connection to the sinus of the lesser sphenoid wing (Figs. 3 and 4).

In some cases of CSDAVF, the shunt points are not always located in the CS itself. In this case, the shunt point was located at the DVOR. We were not able to locate the exact shunt point because we were not aware of the possibility of shunts in this area. Inappropriate embolization of CSDAVF could obstruct venous outflows, which may increase cortical venous reflux leading to the risk of cerebral hemorrhage. Furthermore, the deployed coil mass could block the catheterization to the residual shunt point. Therefore, appropriate embolization strategy should be developed prior to the embolization procedure by preprocedural meticulous evaluation of dAVF angioarchitecture. In our experience, careful analysis of the 3D rotational angiogram and its MPR images is useful in identifying the precise location of the shunt points in dAVFs. In this specific case, fusion of bilateral external carotid arteries and the left internal carotid artery 3D rotational angiograms demonstrated precise angioarchitecture of the CS and the DVOR. The DVOR was not recognized until we made fusion MPR images of the shunt with the bone. Adding the spatial information of the shunt related to the bone led to the final diagnosis of the shunt located inside the orbital roof, in other words, the DVOR. Superselective angiography of the feeders is also helpful in obtaining precise information on the angioarchitecture of CSDAVF during the procedure (Fig. 2). In addition to meticulous analysis of the 3D images and intraprocedural superselective angiography, one must keep in mind the existence of unexpected shunt points outside the CS.

Conclusion

We report a case of CSDAVF with an additional shunt located at the DVOR. To the best of our knowledge, this is the first case report on a CSDAVF associated with an extra-CS shunt at the DVOR. In treating the patients with CSDAVF, careful analysis of 3D rotational angiography and MPR images prior to the procedure is essential to elucidate the precise angioarchitecture and one must be aware of unusual additional shunt points outside the CS.

Disclosure Statement

The authors declare no conflict of interest.
References


