

SURGICAL NEUROLOGY INTERNATIONAL

SNI: Neurovascular

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# Case Report

# Dural arteriovenous fistula in the superior orbital fissure: A case report

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Received: 06 February 18 Accepted: 12 April 18 Published: 07 May 18

### Abstract

**Background:** Dural arteriovenous fistulas (dAVFs) are extremely rare in the superior orbital fissure, and they exhibit ocular symptoms similar to the dAVF in the cavernous sinus because of the intraorbital venous congestion. Hence, the distinction of these conditions is imperative because of some inherent differences in endovascular treatment techniques.

**Case Description:** A 58-year-old woman presented with a gradually worsening left eyeball protrusion and conjunctival congestion. The digital subtraction angiography revealed a dAVF with a shunting point in the left superior orbital fissure. Moreover, the inferolateral trunk of the left internal carotid artery and the left middle meningeal artery were involved as feeding arteries. Shunting blood flow drained into the facial vein through the superior ophthalmic vein (SOV) but not into the cavernous sinus, which was located just posterior to the superior orbital fissure. We performed transvenous coil embolization in the SOV through the facial vein, and the symptoms disappeared completely.

**Conclusion:** We experienced a case of a dAVF in the superior orbital fissure. This case presented a possibility of the presence of one subtype of the dAVF in the part of the cavernous sinus separated at the superior orbital fissure in front. Transvenous coil embolization in the SOV through the facial vein efficiently occluded the fistula.

**Key Words:** Dural arteriovenous fistula, superior orbital fissure, superior ophthalmic vein, transvenous coil embolization



### **INTRODUCTION**

Dural arteriovenous fistulas (dAVFs) are abnormal direct connections between arteries and veins in the dura mater,<sup>[11]</sup> with the cavernous sinus as one of their most common location. dAVFs of the cavernous sinus (CSdAVFs) are often characterized by symptoms such as chemosis, diplopia, and proptosis.<sup>[11]</sup> Although dAVFs with a shunting point in the superior orbital fissure are sporadic, these might mimic symptoms of CSdAVFs by the intraorbital venous congestion.<sup>[8]</sup> While

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How to cite this article: Yamamoto Y,Yamamoto N, Satomi J,Yamaguchi I, Korai M, Kanematsu Y, et al. Dural arteriovenous fistula in the superior orbital fissure: A case report. Surg Neurol Int 2018;9:95.

http://surgicalneurology int.com/Dural-arteriovenous-fistula-in-the-superior-orbital-fissure:-A-case-report/

#### Surgical Neurology International 2018, 9:95

endovascular therapy effectively treats dAVFs around the intraorbital space, its pathological shunting point and the approach route during the treatment differ from CSdAVFs. Here, we report the case of a patient with a dAVF in the left superior orbital fissure who underwent transvenous coil embolization in the superior ophthalmic vein (SOV) through the facial vein approach.

## **CASE REPORT**

A 58-year-old woman without a previous medical and traumatic history presented with gradually worsening pulsatile proptosis and chemosis in her left eye. The initial examination revealed eye movement disturbance (abduction and elevation) and diplopia. Although ophthalmologic examination showed no visual acuity reduction or defect of visual field, it showed dilation of the retinal vein. The three-dimensional time-of-flight magnetic resonance (MR) angiography identified a high-intensity signal in the left SOV and the inferior ophthalmic vein (IOV) [Figure 1]. The digital subtraction angiography (DSA) after 1 week revealed a dAVF with a shunting point in the left orbital fissure; the inferolateral trunk (ILT) of the left internal carotid artery and the left middle cerebral artery (MMA) were involved as feeding arteries and shunted blood flow drained into the left angular vein and facial vein through the SOV. The ophthalmic artery was not involved in the shunt. We determined no drainage into the left cavernous sinus and no cortical venous reflux and deep venous reflux, and grade IIa of the Cognard classification. The left cavernous sinus was used as the antegrade intracranial venous return pathway in the venous phase of the DSA [Figure 2].

We planned transvenous coil embolization in the SOV at the left superior orbital fissure. Accordingly, we placed a 6-Fr guiding catheter (ENVOY, Codman Neurovascular) in the left internal jugular vein under local anesthesia and by the right femoral vein approach. We tried to precede a microcatheter using an approach route from the inferior petrosal sinus and the cavernous sinus to the SOV.



Figure 1: (a and b) Three-dimensional time-of-flight MR angiography (a, axial view and b, sagittal view) reveals high signal in the superior orbital fissure (*arrow*) to the SOV and the IOV. The high signal is continuous with the cavernous sinus (*double arrows*) posterior to the superior orbital fissure

However, as the connection between the cavernous sinus and the SOV could not be opened, any guidewire could not pass through it. Hence, we shifted to the anterior approach, placed a 4-Fr intermediate catheter (Cerulean G; Medikit) in the left facial vein, and preceded a microcatheter (Renegade; Stryker) to the shunting point in the SOV. We embolized the SOV using nine detachable coils (Orbit Galaxy 3-8 mm, Codman; Target nano 2–3 mm, Stryker) [Figure 3]. After the embolization, the fistula from the ILT and the MMA was completely occluded. In the venous phase of the angiography, the left cavernous sinus was maintained as the antegrade pathway of the cerebral venous return [Figure 4]. After embolization, ocular symptoms, such as chemosis and proptosis, promptly improved in few days, and the patient did not report any recurrence until 1 year of follow-up.

#### **DISCUSSION**

This dAVF case was characterized by the presence of a shunting point in the superior orbital fissure and separated from the cavernous sinus. dAVFs with a shunting point around the orbit are sporadic. Intraorbital AVF is only one out of 350 patients who received DSA suspected of cavernous sinus dAVF<sup>[8]</sup> Intraorbital AVF has a direct connection between the SOV/IOV and intraorbital arteries such as the ophthalmic artery<sup>[2-4,6]</sup> and its branches, e.g. the central retinal artery.<sup>[9,10]</sup> In this case, there was no involvement of the ophthalmic artery, and feeding arteries were the ILT and the MMA, which are otherwise commonly involved in CSdAVFs.<sup>[11]</sup> Although the first MR angiography indicated a connection between the SOV and the cavernous sinus, its connection disappeared in the DSA after 1 week. Based on these findings, we hypothesize



Figure 2: (a) The left internal carotid artery angiography (ICAG) reveals an arteriovenous fistula between the ILT of the internal carotid artery and the SOV. (b) Shunting blood flow drains from the SOV to the facial vein through the angular vein and not into the cavernous sinus. (c) In the venous phase of the ICAG, the cavernous sinus is revealed as normal intracranial venous return pathway (lateral view)

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Figure 3:A 4-Fr intermediate catheter (Cerulean; *arrow*) is placed in the left facial vein.A microcatheter (Renegade; the first marker: *arrowhead*; the second marker: *double arrows*) is placed near the shunting point in the superior orbital fissure (lateral view)

that this case was originally a dAVF with a shunting point in the superior orbital fissure located at the frontal tip of the cavernous sinus. The SOV was separated from the cavernous sinus by thrombosis and blood flow changes in the superior orbital fissure. In the antegrade drainage subtypes of the CSdAVFs, reduction of visual acuity may occur due to elevation of ocular pressure secondary to orbital venous congestion, venous stasis retinopathy, and retinal hemorrhage.<sup>[11]</sup> Even in the intraorbital AVF, it can cause severe visual acuity reduction<sup>[12]</sup> and visual field disturbance.<sup>[3]</sup> In our case, there was no obvious decrease in visual acuity, but considering its antegrade venous outflow, there was a possibility that visual symptoms have appeared over the long term.

Endovascular therapy for dAVFs is broadly categorized into transarterial and transvenous embolization. In this case, the ILT of the carotid artery and the MMA were involved as feeding arteries. However, transarterial embolization, especially using liquid embolizing materials, would be dangerous in this condition because the ILT branch (the anteromedial branch that runs through the superior orbital fissure) has a potential anastomosis with the ophthalmic artery.<sup>[7]</sup> Moreover, transarterial embolization from the MMA poses a high risk of ophthalmic ischemia because of the anastomosis between the MMA and the recurrent meningeal artery, meningolacrimal artery, and lacrimal artery.<sup>[5]</sup> In this case, the separated cavernous sinus was used as the antegrade cerebral venous return pathway; therefore, it was considered desirable to preserve as much as possible. We chose selective coil embolization in the SOV by the transvenous approach through the facial vein. In transvenous coil embolization of CSdAVFs, this anterior approach is sometimes used as an alternative route when the inferior petrosal sinus is occluded.<sup>[1]</sup> One of the disadvantages of the anterior approach is its specific anatomic configurations: long distance from the



Figure 4: (a) The common carotid artery angiography (CCAG) after coil embolization exhibits complete obliteration of the arteriovenous fistula. (b) The venous phase of the CCAG demonstrates the antegrade venous flow in the left cavernous sinus (lateral view)

femoral puncture site and a substantial corner angle in the angular vein. In this case, we managed to place a microcatheter in the SOV by refining supportability using a 4-Fr intermediate catheter in a 6-Fr guiding catheter.

#### CONCLUSION

We experienced a rare dAVF case with a shunting point in the superior orbital fissure. The report suggests that detailed evaluations of the DSA and other neuroradiological imaging are imperative to distinguish dAVFs from CSdAVFs. From the perspective of the location of the shunting point and the type of feeding arteries, this case was considered a rare subtype of CSdAVF, and transvenous coil embolization by the facial vein approach an effective treatment of this condition.

#### **Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

#### **Financial support and sponsorship** Nil.

#### **Conflicts of interest**

There are no conflicts of interest.

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