CASE REPORT

Ethmoidal dural arteriovenous fistula with unusual drainage route treated by transarterial embolization

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Accepted 10 March 2014

SUMMARY

Ethmoidal dural arteriovenous fistulas (AVFs) are rare intracranial lesions associated with a high risk of intracranial hemorrhage. In particular, this entity with reflux drainage directly into the ophthalmic vein is extremely rare. We report a case of ethmoidal dural AVF with direct drainage of the superior ophthalmic vein (SOV) and inferior ophthalmic vein (IOV), successfully treated by endovascular surgery. A 58-year-old man presented with progressive diplopia. Angiography and contrast-enhanced CT showed an ethmoidal dural AVF supplied via the bilateral anterior ethmoidal arteries and venous drainage through the left SOV and IOV. A transarterial approach through the bilateral anterior ethmoidal arteries was used to place the microcatheter close to the fistula site. After intra-arterial embolization with 20% N-butyl cyanoacrylate, the dural AVF was completely occluded. In patients with good vascular access, endovascular transarterial embolization may be an effective and less invasive treatment strategy for ethmoidal dural AVF.

BACKGROUND

Dural arteriovenous fistula (AVF) in the anterior cranial fossa is a rare entity,^{1,2} and frequently receives arterial blood supply from ethmoidal branches of the ophthalmic artery. The anterior

ethmoidal artery usually passes through the orbitocranial canal and flows into the dura mater of the anterior cranial fossa. This canal also contains some veins connecting to the dural venous plexus and ophthalmic veins.³

As there is no dural sinus in the anterior cranial fossa, these types of fistulas often drain directly into the frontal cortical veins. However, on extremely rare occasions, venous drainage is directly into the ophthalmic vein. We present the first description of a patient with ethmoidal dural AVF with an unusual drainage route involving the superior ophthalmic vein (SOV) and inferior ophthalmic vein (IOV), in whom successful treatment was achieved by endovascular treatment using a transarterial approach.

CASE PRESENTATION

A 58-year-old man presented with an 8-month history of progressive diplopia. He was found to have profound left-sided exophthalmos and chemosis, as well as all-round partial rectus palsy (figure 1A). Internal carotid artery (ICA) angiography (figure 1B,C) and superselective bilateral ophthalmic artery angiography (figure 2A–D) showed the presence of an AVF supplied by the bilateral anterior ethmoidal branches of each ophthalmic artery. These ethmoidal arteries were divided beyond the origin of the central retinal artery.

Figure 1 External appearance of the patient's eyes and angiographic and CT findings. (A) Severe chemosis of the left eye at the time of initial presentation. (B, C) Internal carotid artery angiography (ICAG) shows arteriovenous fistula (AVF) (bold arrow): (B) right ICAG, (C) left ICAG. (D) CT angiography with three-dimensional reconstruction shows dural AVF identified in the median ethmoid bone around the anterior cranial fossa (bold arrow). Red blood vessels indicate arteries and blue indicate draining veins.

To cite: Inoue A, Tagawa M, Kumon Y, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-011098

Figure 2 Supraselective angiographic findings. (A) Right anterior oblique view and (B) lateral projection of the right ophthalmic artery (white dashed arrow) injection shows the anterior ethmoidal fistula (bold arrow) with feeding artery of the right anterior ethmoidal artery (white arrowhead). (C, D) Left anterior oblique view (C-1: early phase; C-2: late phase) and lateral projection (D-1: early phase; D-2: late phase) of the left ophthalmic artery (black dashed arrow) injection demonstrating the same ethmoidal dural arteriovenous fistula supplied by the left anterior ethmoidal artery (black arrowhead) and venous drainage through the left superior ophthalmic vein (white arrow) and inferior ophthalmic vein (black arrow). Both anterior ethmoidal arteries were divided beyond the central retinal arteries (right: white open arrow, left: black open arrow).

Figure 3 Intraoperative supraselective angiographic findings. Lateral projection of the ophthalmic artery (white dashed arrow) injection (A: right; B: left) shows the tip of the microcatheter (black arrow) and the cast of n-butyl cyanoacrylate (black arrowhead).

Figure 4 Angiographic findings and external appearance of the patient's left eye. (A, B) Internal carotid artery angiography (ICAG) after transarterial embolization using n-butyl cyanoacrylate demonstrating occlusion of the fistula without residual arteriovenous shunting: (A) right ICAG, (B) left ICAG. (C) One month after endovascular treatment, chemosis of the left eye shows marked improvement.

The venous drainage suggested the left SOV and IOV. No ophthalmic veins drained into the cavernous sinus; instead, drainage was into the facial vein. In addition, there was no obvious cortical draining vein. CT angiography with three-dimensional (3D) reconstruction indicated that the fistula was located in the median ethmoid bone around the anterior cranial fossa (figure 1D). We therefore diagnosed ethmoidal dural AVF.

TREATMENT

As the lesion was symptomatic, endovascular surgery was planned in our hospital. A 4 Fr Cerulean G40 catheter (Medikit, Tokyo, Japan) was inserted through a 6 Fr Launcher guiding catheter (Medtronic Vascular, Danvers, Massachusetts, USA) into the femoral artery and advanced until the tip reached the ICA. This procedure was performed bilaterally. An Excelsior SL-10 Microcatheter (Stryker Neurovascular, Fremont, California, USA) was used to selectively catheterize the left ethmoidal artery and a Marathon Flow Directed Micro Catheter (Covidien, Irvine, California, USA) was also introduced into the right ethmoidal artery in a similar procedure via each ophthalmic artery. Next, 0.5 mL of n-butyl cyanoacrylate (NBCA) (Histoacryl Tissue Adhesive; Aesculap AG, Tuttlingen, Germany) mixed 1 : 4 with iodized oil (Lipiodol Ultra-Fluide; Guerbet, Aulnay-Sous-Bios, France) to allow fluoroscopic visualization and to prolong NBCA polymerization was injected from the bilateral ethmoidal arteries (figure 3A,B). After embolization, cerebral angiography showed no arteriovenous shunt, compatible with radiographic cure of the fistula (figure 4A,B). By the next morning, diplopia, chemosis and proptosis had begun to improve. The patient was discharged 1 week after undergoing this procedure. At clinical follow-up 4 weeks later, the blurred vision and ocular findings had returned almost to normal (figure 4C).

DISCUSSION

Dural AVFs occur in the anterior ethmoidal region with an incidence of about 5%.^{1 4-6} In recent years the site of this fistula has been able to be accurately determined by contrast-enhanced CT with 3D construction. This modality is very useful for diagnosing ethmoidal dural AVF.⁷ Angiography of the ethmoidal dural AVF typically shows that the fistula is supplied by the anterior ethmoidal arteries or falcine branches of the ophthalmic artery. As there is no dural sinus around the anterior cranial fossa, venous drainage generally flows directly through the intracranial cortical veins, which ultimately drains into the superior sagittal sinus, inferior sagittal sinus or cavernous sinus. In the present case, the AVF was clearly recognized in the anterior cranial fossa on contrast-enhanced CT. Furthermore, the feeding arteries of this AVF were the bilateral anterior ethmoidal

branches of the ophthalmic artery, and the draining veins were only the left SOV and IOV, representing an extremely rare situation. No previous reports appear to have been described of an ethmoidal dural AVF with such an exceedingly interesting drainage route.

In previous reports, ethmoidal dural AVF has been a particularly difficult lesion to cure with neuroendovascular techniques. For several reasons, endovascular surgery of this fistula has not been widely performed.^{4-6 8 9} First, and most importantly, several authors have warned of the risk of embolization of the central retinal artery, a distal branch of the ophthalmic artery. Second, the small caliber and tortuous course of the ophthalmic artery, ethmoidal artery and various venous drainage routes can make selective catheterization very difficult. Third, unlike embolization of other dural AVFs in which the blood supply originates from the external carotid artery, embolization of ethmoidal dural AVFs through an ICA branch carries the risk of embolic agents refluxing into the central circulation.

However, Ronit *et al* reported that disconnection of these dural AVFs using transarterial catheterization through the ophthalmic artery and subsequent injection of NBCA was possible with a reasonable success rate and a low risk of complications.⁵ Although a transvenous approach has often been performed by other groups,¹⁰ they described that the transarterial approach route is usually shorter with easier and safer access via the ethmoidal artery.⁵ In addition, among a number of embolization materials they favored the use of glue, particularly NBCA, rather than particles. Particle embolization is associated with a risk of unintended occlusion of the central retinal artery and ICA, resulting in visual loss and a number of severe complications.⁶ Mixtures of NBCA, lipiodol and glacial acetic acid have a prolonged polymerization time and enhanced viscosity, which improves the accuracy and safety of embolization.⁵ However, the transarterial approach is not without risk, so interventionalists should be familiar with the anatomy of the central retinal artery origin, be able to choose an appropriate concentration of NBCA mixture and be able to maneuver the catheter into a proper position with respect to the fistula point. In our case the blood supply was from both sides but involved only one of the anterior ethmoidal arteries, and these arteries were divided a long way beyond the origin of the central retinal artery. Furthermore, the venous drainage route was very complex, but the feeding arteries were not tortuous and we were able to reach close to the fistula point relatively easily. We therefore performed endovascular surgery by transarterial embolization using NBCA and the postoperative course was favorable.

We have described an extremely rare case of ethmoidal dural AVF with an unusual draining route involving the SOV and IOV. In patients with good vascular access enabling insertion of the

microcatheter close to the fistula point a long way beyond the central retinal artery, endovascular surgery comprising transarterial embolization using NBCA may be an effective and less invasive treatment strategy for ethmoidal dural AVF.

Learning points

- ▶ Ethmoidal dural arteriovenous fistulas (AVFs) with reflux drainage directly into the ophthalmic vein is an extremely rare entity and no previous reports appear to have been described.
- ▶ Contrast-enhanced CT with three-dimensional reconstruction allows for accurate determination of the site of the ethmoidal AVF, and this modality has been a very useful tool for diagnosing this entity in recent years.
- ▶ In patients with good vascular access enabling insertion of the microcatheter close to the fistula point a long way beyond the central retinal artery, endovascular surgery with transarterial embolization using n-butyl cyanoacrylate may be an effective and less invasive treatment strategy for ethmoidal dural AVFs.

Contributors MT and YK made the diagnosis. MT, AI (main author) and KS performed the operation.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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