CASE REPORT

Combined surgical and endovascular approach to treat a carotid cavernous fistula with associated brainstem venous congestion

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ABSTRACT

Endovascular embolization is the standard approach for management of carotid cavernous fistulas (CCFs) due to the ease of access and reduced level of risk associated with the procedure compared with open surgery. We present here a case of a CCF that eventually led to the development of brainstem venous congestion from perimedullary venous drainage. This fistula was not amenable to endovascular embolization due to lack of either ophthalmic vein or petrosal sinus drainage. Therefore, a craniotomy with direct puncture of the cavernous sinus was performed, followed by coil embolization to completely treat this fistula. This case demonstrates an uncommon progression of venous drainage to Cognard grade V, rare development of symptomatic brainstem venous congestion and a unique method to combine an open surgical approach with endovascular embolization to treat CCFs.

BACKGROUND

This case is important for several reasons. First, it describes a unique clinical presentation of a carotid cavernous fistula as the patient presented with orthostatic hypotension, which to the best of our knowledge has not been described in the literature before. Second, it also demonstrates an innovative treatment strategy of carotid cavernous fistulas (CCF) when conventional options are not possible. There have been many cases that describe a transvenous embolization approach through an ophthalmic or facial vein. There have also been cases that describe a surgical cut down of an ophthalmic vein or cortical vein to access the cavernous sinus. The case presented here is unique as the above options were not possible so a craniotomy with direct puncture of the cavernous sinus had to be performed to access it.

CASE PRESENTATION

A 70-year-old female presented with a 6-month history of diplopia from a mild right sixth nerve palsy. CT angiogram (CTA) and MRI suggested the possibility of a right CCF, which was confirmed by digital subtraction angiography (DSA) (figure 1A, B). The fistula was fed by multiple meningeal branches from both internal carotid arteries (ICAs) and the right external carotid artery (ECA), making it a Barrow type D. Venous drainage was consistent with a Cognard grade III. Of note, no ophthalmic veins

or petrosal sinuses were present, and retrograde leptomeningeal flow was seen in the veins of the cerebellar hemisphere. Two attempts at transfemoral endovascular embolization were made through the right and left inferior petrosal sinuses (IPS) using a combination of microcatheter-microwire systems. These included an Excelsior SL-10 microcatheter, a Tracker-17 microcatheter, an Excelsior-1018 microcatheter and a Prowler 10 microcatheter. Multiple wires, including a Transcend platinum wire, a Headliner 0.016 double angle wire and a Headliner 0.012 90° wire were used. Exchange length wires were also used in order to maintain access while switching out for the various catheters

Figure 1 (A) Axial CT angiogram revealing asymmetric filling of the right cavernous sinus compared with the left in keeping with a right-sided carotid cavernous fistulas (CCF). (B) Right internal carotid digital subtraction angiography (DSA) in lateral projection at the time of the patient's initial presentation showing CCF with drainage into cerebellar veins within posterior fossa (arrow head). (C) Lateral radiograph taken at the time of attempted transfemoral embolization of the fistula demonstrating the microwire (arrow head) inside the cavernous sinus, but failure to advance the microcatheter tip (arrow) into the same venous pouch. (D) T2-weighted sagittal MRI of cervical spine displaying hyperintensity and extensive signal abnormality within the medulla and cervical cord. (E) Preoperative right internal carotid DSA in lateral projection at the time of the patient's re-presentation demonstrating CCF with draining perimedullary (arrow) and cerebellar (arrow head) veins. (F) Preoperative right external carotid DSA in lateral projection at the time of the patient's re-presentation demonstrating draining perimedullary vein (filled arrow) and reflux in vein of Galen (arrow head) and straight sinus (unfilled arrow).

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described above. These included Xcelerator 10, Transcend floppy and V-18 Control wires. During these attempts, the microwire was navigated into the cavernous sinus, but we were unable to advance any microcatheter beyond the IPS (figure 1C). A more direct transcranial access was discussed, but further attempts were withheld at the patient's discretion, due to paucity of symptoms at that time.

One year later, the patient developed postural syncope and intractable nausea. This progressed to the point that the patient was unable to maintain an erect position due to severe orthostatic hypotension and was essentially bedridden. Repeat imaging revealed extensive edema involving the medulla and upper cervical spinal cord with pronounced spinal medullary venous drainage of the known fistula, which was confirmed by DSA (figure 1D–F). This pattern of venous drainage was now consistent with a Cognard grade V. Due to the progression of her symptoms, the decision was made to obtain direct cavernous sinus access through a craniotomy followed by endovascular coiling of the fistula.

INVESTIGATIONS

CT angiogram.

MRI.

Digital subtraction angiography.

TREATMENT

The procedure was performed in our hybrid operating room, fully equipped to function as an endovascular suite with a single plane angiography unit. After induction, the right groin was prepared in a sterile manner to obtain arterial access through a 5 French femoral sheath. A modified right orbitozygomatic craniotomy was then performed, followed by extradural removal of the anterior clinoid process, which exposed the lateral aspect of the cavernous sinus. Neuronavigation and micro-Doppler were then used to assess the location of the ICA. Under fluoroscopic guidance, the anterolateral cavernous sinus was punctured with a 16-gauge intravenous catheter (Smiths Medical ASD, Southington, Connecticut, USA) and connected to a pressurized heparin flush through a rotating hemostatic valve (MBA Hemostasis valve, Merit Medical Systems South Jordan, Utah, USA). The angi catheter was held in place by a titanium mesh that was screwed to the orbital rim. Through this setup, an Excelsior SL-10 microcatheter (Stryker Neurovascular, Fremont, California, USA), over Synchro 2 soft 0.014 microwire (Stryker Neurovascular), was then introduced into the posterior venous pouch and the entire CCF was coiled to obliteration (figure 2A–C). Final cure was confirmed with angiographic runs from bilateral ECAs and ICAs (figure 2D, E). Fortunately, there was no significant bleeding when the angi catheter was removed from the venous pouch, but Gelfoam and fibrin glue was used to cover the puncture site.

OUTCOME AND FOLLOW-UP

The patient's diplopia and orthostatic hypotension resolved. Repeat MRI done 14 days postoperatively showed resolution of fluid-attenuated inversion recovery abnormality in the medulla and the upper cervical spinal cord (figure 2F).

DISCUSSION

CCFs are uncommon lesions, comprising up to 10%–16% of cerebral arteriovenous malformations.¹ When initially diagnosed, our patient had a Barrow type D fistula, with venous drainage consistent with a Cognard grade III, which eventually progressed to a Cognard Grade V. CCFs of higher grades

Figure 2 (A) Intraoperative angiography showing direct puncture of the cavernous sinus with an angi catheter. (B) Intraoperative angiography demonstrating embolization of the CCF with coils. (C) Intraoperative image showing orbitozygomatic approach with angi catheter inserted into the cavernous sinus. Titanium mesh fixed to the skull was used to stabilize the angi catheter in position during embolization. (D) Postprocedural angiography in anterior-posterior projection demonstrates successful embolization of the fistula. (E) Postprocedural angiography in lateral projection demonstrates successful embolization of the fistula. (F) Two-week postoperative T2-weighted sagittal MRI of cervical spine showing significant reduction in signal change within the medulla and cervical cord.

(Cognard grades IIb to V) have cortical venous reflux, which can be associated with hemorrhagic complications and congestive venous hypertension.^{1 2} Our case is unusual in that the venopathy resulted in edema of the medulla and upper cervical cord, probably due to drainage through spinal perimedullary veins. The development of brainstem venous congestion from CCFs is a rare finding and to the best of our knowledge a CCF causing orthostatic hypotension has not been described before.³

There have been several different approaches to the cavernous sinus described in the literature when conventional options are not possible. Transvenous embolization through a direct puncture of the ophthalmic veins, facial vein or cavernous sinus has been well reported and found to be successful.^{4–6} Another approach to the cavernous sinus that has been well documented is a surgical cut down of the superior ophthalmic or cortical sylvian veins.^{1 7 8} Although less common, a direct transforamen ovale puncture of the cavernous sinus or an endoscope-assisted transphenoidal puncture of the cavernous sinus have also been reported in the literature, with successful outcomes.^{9 10} Our case reveals a unique situation where conventional endovascular embolization through a transfemoral approach was not successful due to the inability to visualize patent petrosal sinuses and the inability to guide the microcatheter into the fistula. Accessing the ophthalmic veins or a cortical sylvian vein through a percutaneous transorbital approach, or via a surgical cut-down, was not possible as these veins were not filling on angiography. Therefore, the only remaining option was to gain direct surgical access to the cavernous sinus. To the best of our knowledge, this open surgical approach with direct puncture of the cavernous sinus and subsequent endovascular embolization of a CCF has not been described before. This case demonstrates both a unique clinical presentation of a CCF and a novel way to combine a direct surgical approach with endovascular embolization to treat CCFs when conventional treatment options are not possible.

Learning points

- ▶ Brainstem venous congestion is rare in the setting of carotid cavernous fistulas, but when it occurs it should be identified as it can be quite symptomatic
- ▶ Carotid cavernous fistulas should be monitored for both clinical and radiological progression
- ▶ This case shows a novel approach to treat carotid cavernous fistulas when conventional options are not possible

Competing interests None declared.

Patient consent Obtained.

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