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Case Report

Transvenous embolization of a direct carotid-cavernous fistula through the pterygoid plexus approach ^{☆,☆☆}

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ABSTRACT

We present a transvenous embolization technique for a direct carotid-cavernous fistula through the pterygoid plexus to the cavernous sinus which only 2 cases have been previously reported in the English literature. This method is appropriate when transarterial techniques or other attempts at transvenous access have failed due to vessel tortuosity, hypoplasia, stenosis, or occlusion.

A middle-aged female patient presented with progressive left exophthalmos with conjunctiva chemosis and bruit after sustaining a falling injury. Digital subtraction angiography revealed Barrow type A carotid-cavernous fistula. The drainage route passed through a distal thrombosed superior ophthalmic vein that ended deep in the orbit. No other patent venous sinuses connected to the cavernous sinus, except for a small tract of pterygoid plexus. After failure of transarterial approach and other methods of transvenous access, we attempted to superselectively access to the cavernous sinus by applying transpterygoid technique with embolization using detachable coils. The transpterygoid venous approach to accessing the cavernous sinus represents an alternative approach when other techniques fail.

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Background

An arteriovenous fistula of cavernous sinus is comprised of an abnormal connection between internal carotid artery (ICA)

or external carotid artery (ECA) and cavernous sinus (CS). Carotid-cavernous fistulas (CCFs) can be classified as direct (feeders from the main trunk of ICA) or indirect (feeders from the dural branches of ICA, ECA, or both) and are usually divided into 4 subtypes according to the Barrow classification [1]. Type A represents direct connection between the ICA and CS, whereas types B, C, and D are indirect fistulas and considered cavernous sinus dural arteriovenous fistulas (CSDAVF). Type B fistulas are located between meningeal branches of the ICA and CS. Type C fistulas are located

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between meningeal branches of the ECA and CS. Type D fistulas involve abnormal communication between the CS and meningeal branches of both ICA and ECA. These abnormal connections to the CS cause venous hypertension, which may result in venous congestive symptoms, cranial nerve compromise, perfusion reduction, and haemorrhage [2]. Endovascular embolization through arterial or venous approach is considered first-line treatment for CCFs. Transvenous embolization is usually performed through the inferior petrosal sinus (IPS) or superior ophthalmic vein (SOV). If transvenous access to the CS is not possible, approaches from other CS connections may be used. Transvenous embolization through the pterygoid plexus has been previously described twice: in one case, a CSDAVF was embolized through contralateral pterygoid plexus [3]; in the other, a direct CCF was treated through ipsilateral pterygoid plexus [4]. Here, we describe the successful ipsilateral transpterygoid plexus embolization of direct CCF after transarterial catheterization failed and no opacification of the IPS and SOV was present.

Case presentation

A middle-aged female presented with 2-month history of progressive left exophthalmos with chemosis after a fall. An orbital bruit was auscultated. Neurological examination indicated normal consciousness without cranial nerve deficits. Computed tomography angiography and magnetic resonance imaging showed left middle cranial fossa subdural hematoma and a prominent left CS with left SOV engorgement. Digital subtraction angiography showed direct left CCF originating from the inferior aspect of posterior horizontal C4 segment in the cavernous ICA (Fig. 1A) with venous drainage from the CS into SOV and pterygoid plexus. The IPS did not opacify and thrombosis of the left distal SOV without connection to the facial vein was observed (Fig. 1B).

Both femoral artery and vein punctures were performed. An ENVOY 6-Fr guiding catheter (Codman Neuro, Raynham, MA) was positioned in the left cervical ICA and a coaxial Excelsior microcatheter (Stryker Neurovascular, Fremont, CA) passed into the posterior genu of the ICA (C4 segment). Another 5-Fr guiding catheter was placed in the left internal jugular vein. Navigation of microcatheter into the fistula failed because of an acutely angulated posterior genu and small fistula ostium. Scepter balloon (Microvention, Tustin, CA) assisted microcatheter navigation was then attempted. The tip of microcatheter was positioned at the ostium but could not enter CS (Fig. 2A). We then attempted to pass the venous guiding catheter into the nonvisible IPS coaxially with a 0.035-inch guidewire, which also failed. Careful evaluation of multiphases of the angiogram revealed a transforaminal channel passing from the CS to the pterygoid plexus. An Excelsior microcatheter was manipulated over a 0.014 guidewire through the guiding catheter located in the external jugular vein and advanced through the left retromandibular vein, maxillary vein, and pterygoid plexus into the CS (Fig. 2B). A total of 6 Target Detach-

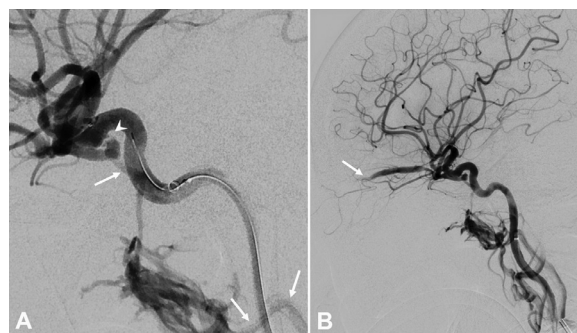


Fig. 1 – Pre-embolization digital subtraction angiography with close-up view. (A) Lateral projections reveal a single channel connecting the CS and pterygoid plexus and drainage into the maxillary vein, retromandibular vein, and external jugular vein (arrow). A small fistulous ostium originates from the inferior aspect of posterior horizontal C4 segment of the cavernous ICA (arrowhead). (B) No opacification of the IPS was present, and the SOV ends behind the eyeball with distal thrombosis and no connection to the facial vein (arrow).

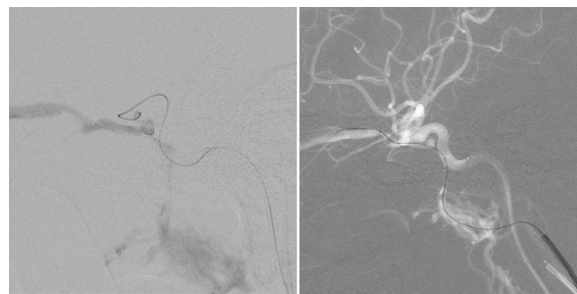


Fig. 2 – Attempted transarterial and transvenous catheterization. (A) Navigation of the microcatheter was possible only at the small fistulous ostium because of the acutely angulated posterior genu location. (B) The microcatheter passed into the CS through the external jugular vein, retromandibular vein, maxillary vein, and pterygoid plexus.

able Coils (Stryker Neurovascular) were used to perform coil embolization.

Postembolization angiography showed complete CCF occlusion (Fig. 3). No procedure-related complications occurred. The patient's chemosis and exophthalmos resolved completely 3 days after embolization and she remained neurologically intact.

Discussion

Endovascular embolization is the treatment of choice for CCF. Approaches include transarterial catheterization of feeders or fistula ostium [5] and transvenous access, which is most often achieved through IPS or SOV [6]. Transarterial embolization

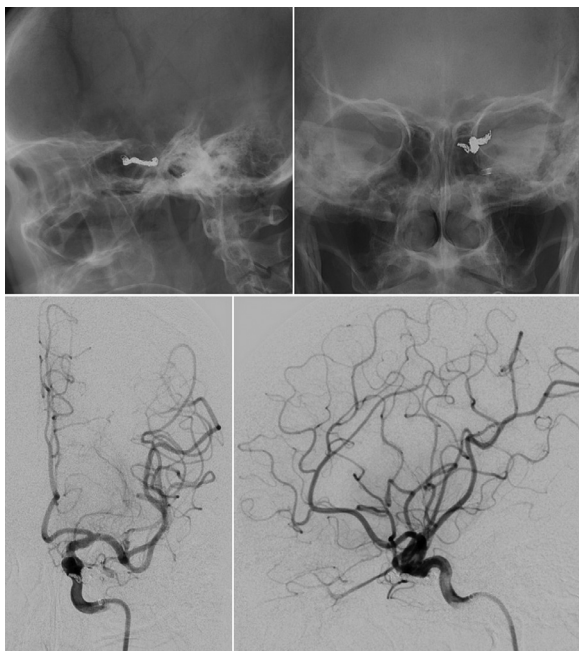


Fig. 3 – Postembolization angiography. Follow-up left ICA angiography after Guglielmi detachable coil embolization revealed complete occlusion of the carotid-cavernous fistulous flow.

with detachable balloons or coils is safe and effective for high-flow direct CCFs and achieves sustained effects [7]. However, this approach may be unsuitable in patients with complex vascular or fistula anatomy, such as tortuous, atherosclerotic ICA, or unfavorable positioning of fistula ostium. Transvenous embolization has become the primary therapeutic option because of its high success rate and safety [8]. Thomas et al. [2] proposed a CCF classification based on venous drainage and noted anterior (superior and inferior ophthalmic veins), posterior (inferior and superior petrosal sinuses), and inferior (pterygoid and parapharyngeal plexuses and cortical veins) drainage routes. All are potential transvenous routes for embolization. The pterygoid plexus communicates with the CS through emissary veins which pass through foramen of Vesalius/Lacerum and drains posteriorly into maxillary vein, retro-mandibular vein and external or internal jugular vein. In our patient, the transarterial approach failed due to unfavorable positioning and small fistula between ICA and CS. Although the IPS and SOV are preferable for transvenous access, in our patient, the IPS and superior petrosal sinus were not patent and catheterization failed. Besides, the SOV ended behind the eye and nonopacification of facial or angular vein, showing this was a suboptimal route for transvenous catheterization or surgical cutdown procedure [9]. Tang et al. [10] reported an indirect CCF treated using endoscope-assisted transsphenoidal puncture of CS for embolization. However, because of the invasiveness and the need for a hybrid operating room, we elected to navigate microcatheter through the retromandibular vein, maxillary vein, pterygoid plexus, and emissary vein into the CS for embolization.

Conclusion

The transpterygoid plexus approach can provide alternative access to the CS for embolization if transarterial catheterization is not feasible or transvenous approach through the IPS and SOV fails.

Declarations

Ethics approval and consent to participate

According hospital's ethical policy for case reports, this study had received the patient consent form agreement.

Consent for publication

This study had received the publication consent form agreement.

Availability of data and material

All relevant data is provided in the manuscript. However, additional supplementary data can be provided from the corresponding author upon request.

Author's contribution

Conception and design: CHO, WCL. Acquisition of data: CLL, PLL, CHO. Analysis and interpretation of data: CHO, WCL, TYC. Drafting the article: WCL, CHO. Critically revising the article: CHO, TYC. Technical/device/material support: PLL, CLL. Study supervision: CHO. All authors read and approved the final manuscript.

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