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Transvenous embolization of a carotid-cavernous fistula via the inferior ophthalmic vein: illustrative case

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BACKGROUND A 49-year-old woman with a history of hypertension presented to the emergency department with right eye redness, proptosis, orbital fullness, and blurry vision. She had initially been diagnosed with an orbital pseudotumor, and the symptoms worsened over a course of steroids. Computed tomography angiography raised concern for a carotid-cavernous fistula (CCF), which was subsequently confirmed by digital subtraction angiography.

OBSERVATIONS She underwent fistula coil embolization via the internal maxillary artery and inferior ophthalmic vein (IOV). At the 2-month follow-up, she reported complete resolution of diplopia, orbital fullness, and proptosis. An ophthalmology examination revealed normal visual fields bilaterally.

LESSONS CCF embolization is rarely performed through the IOV, with only 5 reported cases in the literature. This case demonstrates that the procedure can be easily performed if the anatomy is favorable over the superior ophthalmic vein, with the illustration of good cosmetic outcomes.

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KEYWORDS indirect carotid-cavernous fistula; transvenous embolization; carotid-cavernous fistula; direct inferior ophthalmic vein catheterization

Carotid-cavernous fistulas (CCFs) are vascular malformations characterized by abnormal connections between the cavernous sinus (CS) and the carotid artery. CCFs can be classified into direct or indirect based on the nature of the connection. Presenting symptoms include pulsatile exophthalmos, orbital bruit, and cranial nerve palsies.[1] Management aims to restore normal hemodynamic flow and alleviate associated symptoms. It could be conservative management, endovascular embolization, surgical repair, or a combined approach.[2] Transvenous embolization (TVE) of CCFs via the inferior ophthalmic vein (IOV) is a minimally invasive treatment for these vascular malformations.

Illustrative Case

A 49-year-old woman with a history of hypertension presented to the emergency department with right-eye redness, proptosis, orbital fullness, and blurry vision. These symptoms had started about 5 months earlier after a fractured and infected tooth extraction and had significantly worsened for 1 day. Previous evaluation showed an intraocular pressure of 15 mm Hg and 46 mm Hg in the left and right eyes, respectively. Brain magnetic resonance imaging was concerning for idiopathic orbital inflammatory syndrome, which was treated with prednisone with limited effect. On physical examination, right-eye proptosis and chemosis were noted (Fig. 1A), as well as an orbital bruit on auscultation. Neurological examination findings were significant for right lateral gaze palsy and decreased sensation in the V1 and V2 distributions. Computed tomography angiography (CTA) revealed a proptotic right globe, enlarged extraocular muscles, dilation of the right ophthalmic veins, and tenting of the right optic nerve (Fig. 2A and B). A concern for a CCF was raised, and the patient underwent diagnostic angiography, which confirmed the diagnosis of a Barrow type D CCF (Fig. 3A). Subsequent fistula embolization via the right internal maxillary artery (IMAX) and IOV was performed. The patient's symptoms completely resolved at the 2-month postoperative clinic follow-up. CTA at the 6-month follow-up showed complete resolution of the right proptosis (Fig. 2C and D). Unfortunately, at the 18-month mark, the patient had developed the new onset of mild right ptosis, myosis, and double vision with right lateral gaze concerning for Horner's syndrome and a partial cranial

ABBREVIATIONS CCF = carotid-cavernous fistula; CS = cavernous sinus; CTA = computed tomography angiography; DSA = digital subtraction angiogram; ECA = external carotid artery; ICA = internal carotid artery; IMAX = internal maxillary artery; IOV = inferior ophthalmic vein; SOV = superior ophthalmic vein; TVE = transvenous embolization.

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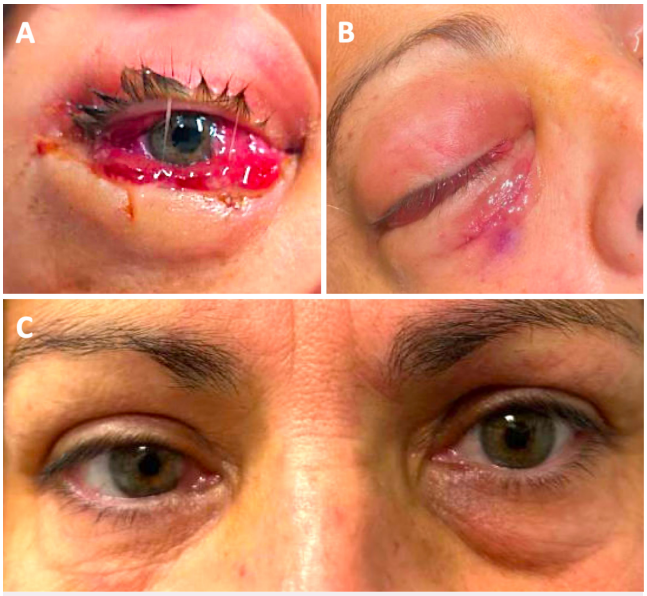

FIG. 1. A: Significant right-eye proptosis and chemosis on initial evaluation. **B:** Immediate postoperative image of the surgical access site. **C:** Photograph at the 18-month follow-up with an excellent cosmetic outcome.

nerve 6 palsy. No evidence of recurrence was found on imaging. The patient was started on a steroid taper with a moderate improvement of symptoms.

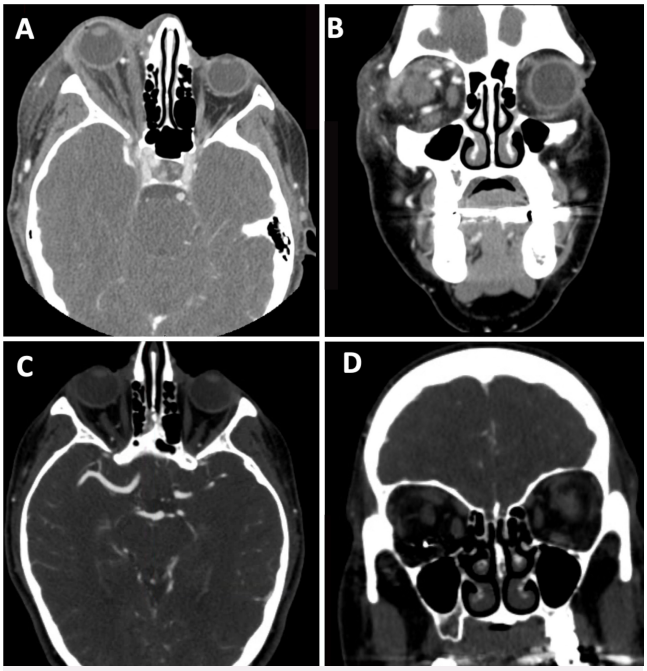

FIG. 2. Axial (**A**) and coronal (**B**) preoperative CT angiograms demonstrating significant proptosis as well as a high level of vascularity within the right globe. Axial (**C**) and coronal (**D**) postoperative CT angiograms demonstrating resolution of proptosis, resolution of vascularity seen previously, and evidence of embolic material within the IOV.

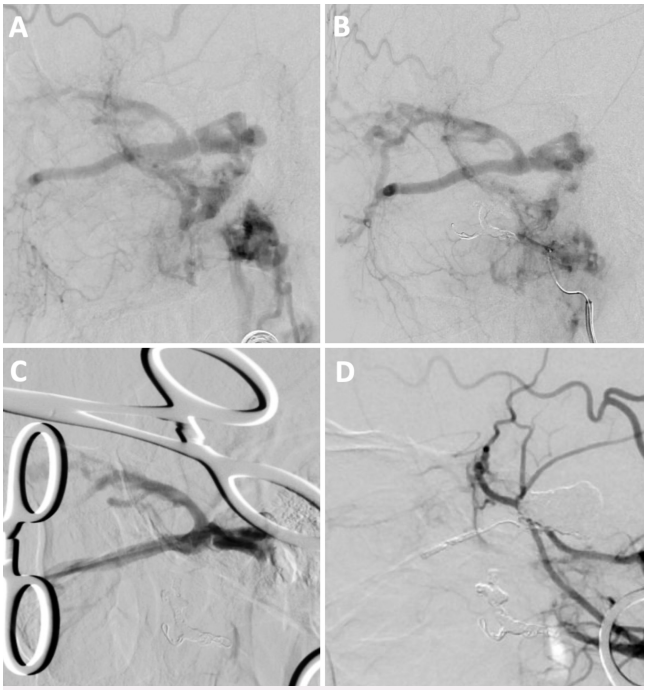

FIG. 3. Intraprocedural DSAs of the right CCF. **A:** Lateral projection of the mid–arterial phase of the CCF prior to transarterial embolization. **B:** Lateral projection of the mid–arterial phase of the CCF after transarterial embolization showing a decrease of flow through the fistula, although significant drainage through the SOV and IOV remains. **C:** Lateral projection of the catheterization of the right CS via direct catheterization of the IOV. **D:** Lateral projection of the CCF after TVE, with complete resolution of drainage through the SOV and the IOV.

The patient underwent diagnostic cerebral angiography, which confirmed a Barrow type D right CCF (Fig. 3A), with both the superior ophthalmic vein (SOV) and IOV as the main draining veins. During the same procedure, embolization of the main external carotid artery (ECA) feeder right IMAX using coils was also performed, which yielded decreased flow to the fistula (Fig. 3B). The patient was brought back 2 days later for a second-stage embolization via the transvenous route. Transarterial access was obtained through the right femoral artery. The right ECA was selected, and a digital subtraction angiogram (DSA) was obtained. Because of the tortuous nature of the SOV and the relatively straight course of the IOV, the IOV was chosen for direct catheterization (Fig. 4). Under the guidance of the roadmap, the IOV was identified and marked on the skin of the lower eyelid. A blade was used to make a transcutaneous incision on the right medial to the central lower eyelid. The orbicularis was incised, and the orbital septum was incised with the Bovie and Westcott scissors. Curved iris scissors and cotton swabs were then used to bluntly dissect past the postseptal plane into the orbital fat. Small tortuous vessels were noted and cauterized. Bipolar cautery was used to achieve hemostasis; 4-0 silk sutures were used as traction sutures. Blunt dissection was then performed until the dilated tributary was located and skeletonized. The arterialized IOV was catheterized with a pediatric micropuncture needle. A 0.018-inch nitinol wire was then advanced without difficulty for approximately 6 cm. The micropuncture needle was then exchanged for a stiff 4-Fr dilator that was then secured

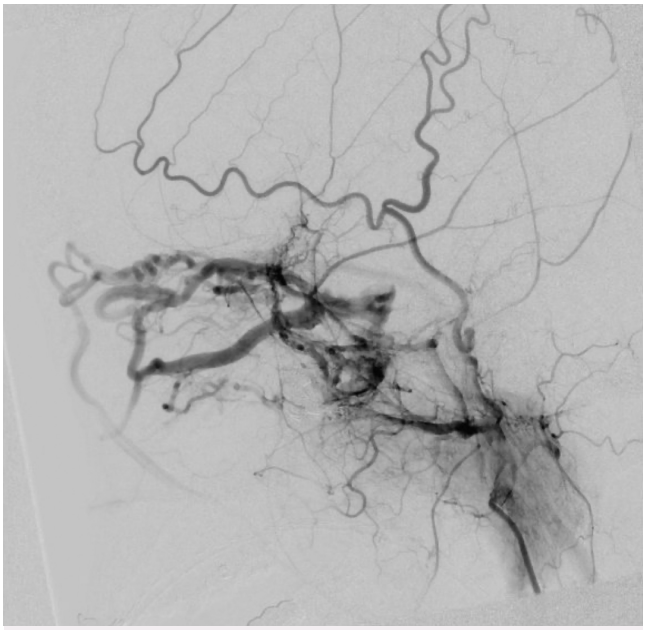

FIG. 4. Lateral projection DSA of the right SOV and IOV prior to transvenous catheterization and TVE. Note the significant tortuosity of the distal SOV and the relatively straight course of the IOV.

and connected to a continuous heparinized drip. A super-selective DSA showed excellent positioning of the sheath in the junction of the SOV and IOV (Fig. 3C). The CS was then catheterized using a Prowler 14 microcatheter (Cerenovus) and a Synchro-2 microwire (Stryker) under the guidance of the roadmap obtained via the injection through the Envoy MPD catheter (Cerenovus) in the right ECA. The microcatheter was advanced as distally as possible, and the CS was densely coiled. A postembolization DSA of the right ECA and internal carotid artery (ICA) was obtained, which showed complete obliteration of the fistula without early opacification of venous drainage (Fig. 3D). The dilator was removed, and the IOV was ligated with silk ties. The wound was washed out, and the incision site was closed using a 7-0 running Monocryl suture subcutaneously, followed by the application of medical-grade skin glue (Fig. 1B).

Patient Informed Consent

The necessary patient informed consent was obtained in this study.

Discussion

The etiology of CCFs can vary and commonly includes trauma, idiopathic connective tissue disorders such as Ehlers-Danlos syndrome or fibromuscular dysplasia, ruptured aneurysms, inflammatory conditions such as vasculitis or infection, congenital factors, or iatrogenic injury, etc.[1] The clinical presentations for anterior-draining fistulas tend to involve ophthalmic symptoms such as proptosis, blurry vision, ocular bruit, strabismus, chemosis, diplopia, as well as headaches.[1–3]

Visual loss and intracranial hemorrhage secondary to venous hypertension remain the most dreadful outcomes. The diagnosis of CCF typically involves a combination of clinical evaluation, imaging studies, and specialized tests. The ophthalmology service is often the first to be consulted for eye symptoms, and intraocular pressure measurement often yields an elevated result. Magnetic resonance angiography or CTA are noninvasive techniques that typically would reveal dilation of the feeding arteries, engorgement of the draining veins, and proptosis of the eyeball.[4] Due to its spatial and temporal resolution, digital subtraction angiography remains the gold-standard method in the diagnosis of CCF. Conservative management along with carotid compression therapy can be applied in low-flow, low-risk patients for a chance of spontaneous closure.[3,5,6]

Endovascular treatment of the CCF is the mainstay and can be achieved via an arterial, venous, or combined approach. Coils, liquid embolic agents, detachable balloons, and flow-diverter stents can all be used in various combinations to achieve the goal of occluding the fistula while maintaining the patency of the ICA.[1,7] In rare cases, however, with an intact circle of Willis and robust collateral flows, the ICA of interest could potentially be sacrificed without significant complications.[8]

Observations

This presented case was managed in a staged fashion via both arterial and venous approaches. Due to the indirect nature of the fistula, the CS could not be embolized via the arterial approach, so coil embolization was performed in the first stage to decrease the flow from the IMAX. In the second stage of TVE, the anatomy was also unfavorable for a transfemoral venous approach due to small petrosal sinuses observed on the first DSA. Attempting for an extended time to catheterize these sinuses could lead to unwanted radiation exposure and injury. In general, the SOV is larger than the IOV and is more commonly used for direct puncture for embolization of the CCF. However, in this particular case, a direct transorbital approach through the right IOV was selected due to its larger size and straighter course to the CS than the SOV. Anatomically, the IOV ultimately terminates into the pterygoid plexus via the inferior orbital fissure and the CS via the superior orbital fissure.[9] A roadmap obtained after establishing access using the 4-Fr dilator provided guidance to the CS.

Lessons

TVE of a CCF through the IOV is a technique rarely reported in the literature. We found a total of 5 cases that described direct puncture of the IOV for the treatment of indirect CCFs[10–14] (Table 1). In all cases, the fistulas were successfully embolized with favorable clinical outcomes and no reported complications. In 2 cases described by Oono et al.[11] and Dashti et al.,[13] the IOV was accessed as part of retreatment after the failure of initial embolization and as an initial treatment in 3 other cases described by Cecchini et al.,[10] Schmidbauer et al.,[14] and Michels et al.[12] Catheterization through the SOV was considered in all cases, but the vein was found to be too tortuous and partially thrombosed or stenotic in 4 cases and too deep within the orbital apex in the case described by Dashti et al.[13] While direct catheterization has been shown to be relatively safe and effective due to a low number of reported cases and availability of other endovascular options, it remains a viable backup solution in select cases.

The reported success rate for closing direct fistulas is 67%–99% and that for indirect fistulas is 50%–100%.[2,8] For patients with failed or unavailable endovascular treatment, surgical vessel ligation versus trapping could be considered as an alternative.

TABLE 1. Systematic review of indirect CCFs treated through direct catheterization of the IOV

Authors & Year	Age (yrs)/Sex	Presenting Symptoms	FND	CCF Arterial Supply	CCF Venous Drainage	Reason for Direct IOV Access	Prior Treatment	Access	Embolic Material	Postop Course	FU Time
Present case	49/F	Rt conjunctival injection, proptosis, blurry vision, decreased sensation	CN V1–2 & CN VI palsy	Rt IMAX	Rt IOV & SOV	Small bilat IPSs, small & tortuous SOV	Yes, TAE performed as a staged procedure to decrease flow	Incision along medial to central lower eyelid; the orbicularis followed by orbital septum incised	Platinum coils	Initial resolution of symptoms w/ delayed CN VI palsy & Horner's syndrome	1.5 yrs
Cecchini et al., 2012[10]	61/M	Rt chemosis, proptosis, & ocular pain	None	Rt IMAX & MMA, dural branches from bilat ICAs	Rt IOV	IPS w/ no communication w/ CCF; SOV partially thrombosed	No	Incision underneath eyelid, just lat to midorbit	GDCs	Resolution of all symptoms	6 wks
Dashti et al., 2011[13]	39/M	Chemosis, proptosis, double vision	Mild vision loss in rt eye	Meningeal branches of lt cavernous ICA	Contralat SOV	SOV deep w/in orbital apex; unable to traverse occluded lt IPS	Yes, TAE w/ short-term recanalization (next day)	Entry point at lat third of lower eyelid	Onyx	Vision improved, chemosis & diplopia resolved	2 wks
Michels et al., 2007[12]	76/F	Periocular pain, diplopia, ocular injection, proptosis, swelling	CN VI palsy, mild vision loss in rt eye	Rt MMA	Primarily rt IOV, minimal SOV outflow	SOV small, tortuous & partially thrombosed; IPS occluded, facial & angular veins tortuous	No	Transconjunctival swinging eyelid approach to access inferonasal orbital space	Platinum coils	Improvement in CN VI palsy, resolution of ocular swelling & injection	2 mos
Schmidbauer et al., 2000[14]	69/F	Rt proptosis & conjunctival injection	Mild vision loss in rt eye	Bilat ICAs, lt ECA	Rt IOV	Small caliber of arterial inflow, no access through transvenous route, SOV of smaller caliber	No	An infraciliary skin incision in middle of lower eyelid & incision of orbital septum	Platinum coils	Resolution of symptoms	2 wks
Oono et al., 1998[11]	52/M	Diplopia, rt periocular pain, conjunctival injection	Partial CN VI palsy	Dural branches of ICA & branches of IMAX	Bilat IPS & internal jugular veins initially, SOV & IOV at recurrence	Occlusion of IPS & ECA routes due to prior embolization; SOV tortuous & partly stenotic	Yes, TAE w/ short-term recanalization (2 mos)	Horizontal skin incision of lower lid & incision of orbital septum	Platinum coils	Resolution of CN VI palsy & conjunctival congestion	3 yrs

CN = cranial nerve; FND = focal neurological deficit; FU = follow-up; GDC = Gugliemi detachable coil; IPS = inferior petrosal sinus; MMA = middle meningeal artery; TAE = transarterial embolization.

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Disclosures

Dr. Waldau reported personal fees from Stryker and Siemens outside the submitted work.

Author Contributions

Conception and design: Waldau. Acquisition of data: all authors. Analysis and interpretation of data: all authors. Drafting the article: Liang, Moskalik, Taylor. Critically revising the article: Waldau, Liang, Moskalik. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Waldau. Statistical analysis: Moskalik. Administrative/technical/material support: Waldau, Moskalik. Study supervision: Waldau.

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