

Orbital apex osteodural fistula - An unusual surgical access

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Dural arteriovenous fistulas (DAVFs) are fistulas connecting the branches of dural arteries to dural veins or a venous sinus. Osteodural fistulas are a rare subset of this group of diseases. We wish to report a rare case of an osteodural arteriovenous fistula at the foot of the superior ophthalmic vein (SOV), treatment of which required an unusual surgical approach via the orbit and SOV. Though access for endovascular treatment via the SOV for treatment of caroticocavernous fistulas is reported, the external approach is relatively infrequently performed, outside Europe and the Americas, with this being the first reported procedure from the Indian subcontinent. We wish to explain the steps of this unusual surgical access and highlight the salient precautions and pitfalls in the technique.

Key words: Coil embolization, dural arteriovenous fistula, superior ophthalmic vein

We report the first case of an orbital apex osteodural arteriovenous (AV) fistula in Indian literature, in which after an unsuccessful attempt at endovascular embolization via the transvenous-transfemoral route, the superior ophthalmic vein (SOV) was accessed surgically, and a retrograde venous embolization was performed as an emergency intervention.

Case Report

A 34-year-old lady, 3 months postpartum, presented with 2 months of protrusion, pain, redness of the left eye. Her best-corrected visual acuity was 20/20, N6 and 20/200, N12 in the right and left eyes, respectively. The right eye was normal. The left eye was proptosed 2 mm compared to the right with prominent corkscrew episcleral vessels [Fig. 1]. Pupillary reactions, ocular motility, color vision, and gonioscopic examination were normal. Intraocular pressure was 12 mmHg OD and 24 mmHg OS with prominent pulsations of applanation mires. Fundus and automated perimetry were

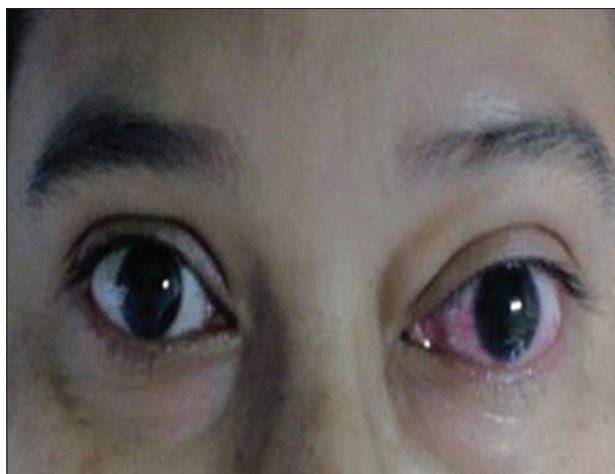


Figure 1: Prominent corkscrew vessels with proptosis

normal. No history of trauma. Thyroid profile was normal. A low flow Caroticocavernous fistula left orbit was diagnosed.

Magnetic resonance imaging showed prominent extra-ocular muscles, dilated SOV in the left orbit [Fig. 2]. Magnetic resonance angiography (MRA) revealed the presence of an arteriovenous (AV) fistula at the orbital apex connecting SOV and internal carotid artery (ICA) [Fig. 3]. Digital subtraction angiography (DSA) showed a high flow fistula at orbital apex, supplied by dural branches from cavernous segment of ICA, accessory meningeal, middle meningeal, and the third part of internal maxillary arteries [Fig. 4]. The fistula drained into SOV, angular, superficial temporal, and facial veins. There was no reflux into ipsilateral or contralateral cavernous sinuses or cerebral veins. The inferior petrosal sinuses were not visualized.

Under general anesthesia, endovascular embolization was attempted. A transfemoral venous access using the facial vein to reach the orbital venous system was taken. However, loopiness and tortuosity of the orbital venous system tributaries made attempts of a good distal microcatheter placement at the foot of SOV unsuccessful. Hence, SOV was accessed by an alternative approach.

A transverse, 2 cm superomedial lid crease incision was made [Fig. 5]. Incision was deepened [Fig. 6]; the orbicularis oculi muscle was dissected and split [Fig. 7]. Orbital septum was identified, incised [Figs. 8 and 9], and orbital fat was bluntly separated with care not to damage vascular structures. A small arterialized venous branch was discovered and dissected off investing fat and connective tissue, to localize the main trunk of SOV. An arterialized giant SOV was exposed [Fig. 10]. 3-0 Silk loops were passed to straighten and control the vein [Fig. 11].

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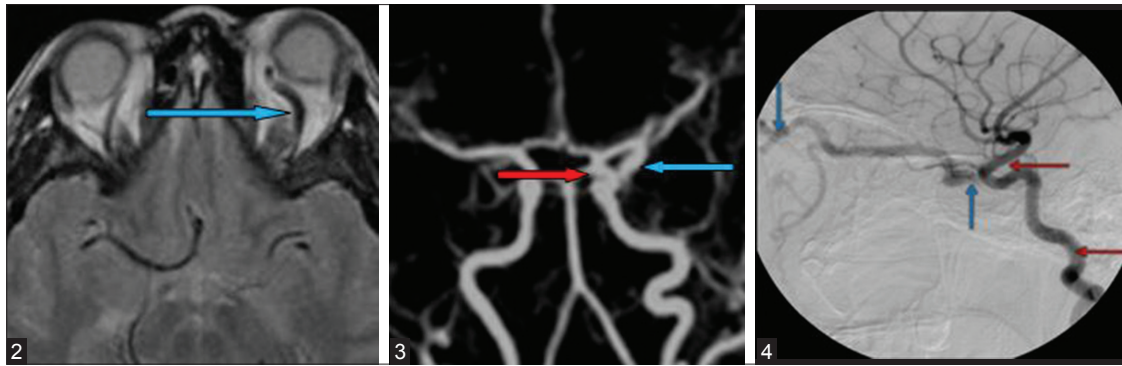


Figure 2-4: (2) Magnetic resonance imaging–dilated left superior ophthalmic vein; (3) Magnetic resonance angiography–arteriovenous fistula; (4) Digital subtraction angiography–left osteodural A-V fistula

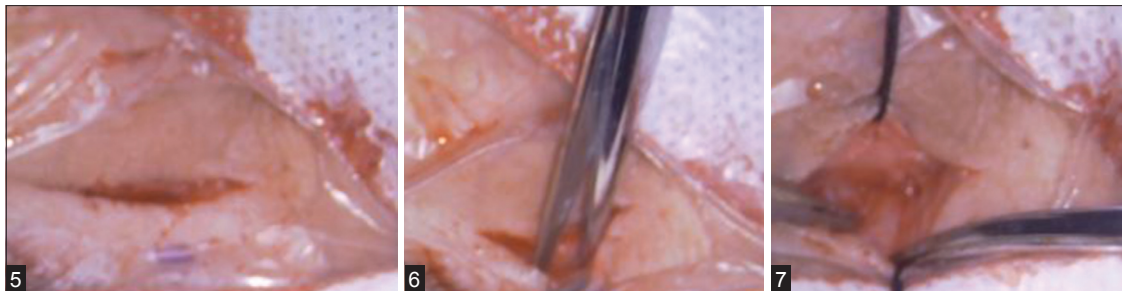


Figure 5-7: (5) Superior lid crease incision; (6) Blunt dissection; (7) Orbicularis oculi identified

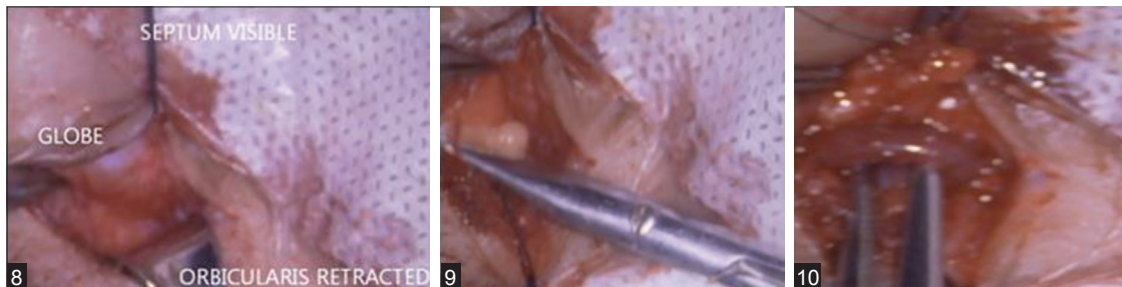


Figure 8-10: (8) Orbital septum exposed; (9) Orbital septum incised; (10) Exposed superior ophthalmic vein

After this, an intact 10 mm segment, between two silk loops, was handed over to the interventional radiologist. The SOV was punctured under naked eye, using a micropuncture access set, and a 5 F vascular sheath introduced into SOV [Fig. 12]. Thereafter, a dual tip microcatheter (Echelon-10, EV3) was introduced to the foot of SOV, and multiple detachable platinum coils were used to achieve embolization and complete exclusion of the fistula [Fig. 13]. The microcatheter was withdrawn, and SOV was ligated with 6/0 vicryl suture [Fig 14]. Hemostasis was achieved with bipolar cautery and ligation of larger tributaries. Single layer closure of skin was performed with vertical mattress sutures with 6/0 vicryl [Fig. 15].

On postoperative day 1 [Fig. 16], extraocular movements were full. Pupillary reactions were normal. Periorbital dependent edema, ecchymosis, and conjunctival chemosis was present with a healthy, supple wound. Edema settled over 7 days, with healing of the wound and good cosmetic outcome [Fig. 17].

One month later, she developed proptosis and

redness [Fig. 18]. MRA and DSA revealed recurrence. Endovascular embolization was performed using the accessory meningeal artery route, 1 cc of liquid embolic agent onyx (Ethylene-vinylalcohol copolymer) (Onyx, Micro Therapeutics, Irvine, California), injected with complete exclusion. Good postoperative recovery with regression of proptosis and disappearance of prominent episcleral congestion noted, 1 month postprocedure. She is on regular follow-up since last 6 months and is asymptomatic [Fig. 19].

Discussion

Dural arteriovenous fistulas are rare (10–15% of intracranial AV fistulas).^[1] Osteodural, (cartilaginous) epidural A-V fistulas belong to the ventral epidural group.^[2] These are lesions wherein intracranial arteriovenous shunts fed by dural arteries (that also supply the bone) are located within or near bony structures along the convexity or at the skull base. The orbital apex is a rare location.^[3,4]

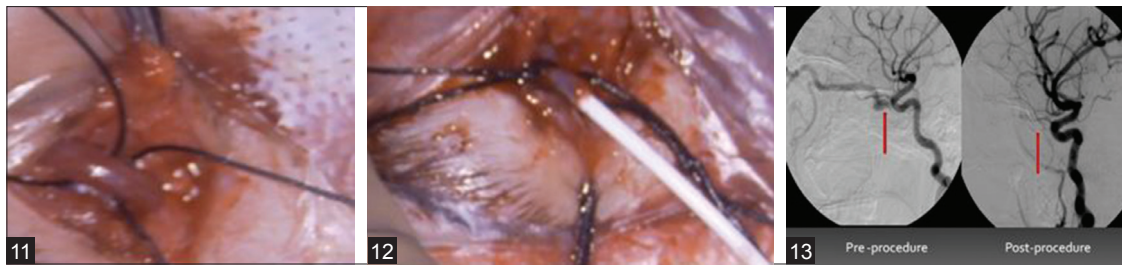


Figure 11-13: (11) Silk ties around the superior ophthalmic vein; (12) Introduction of microcatheter; (13) Coil embolization

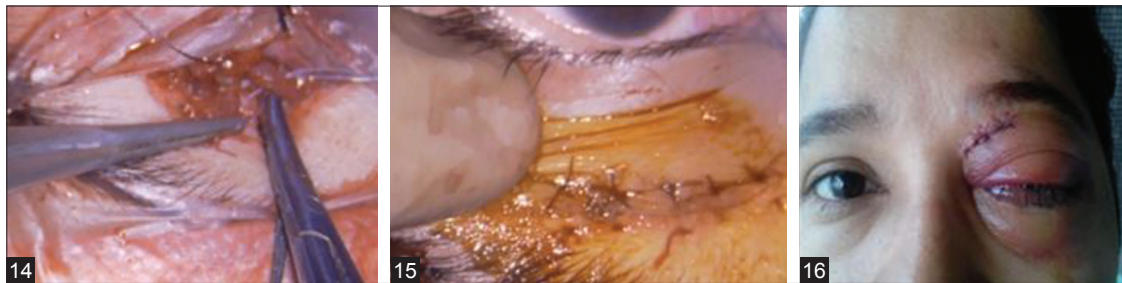


Figure 14-16: (14) Superior ophthalmic vein ligation; (15) Closure; (16) Postoperative day 1

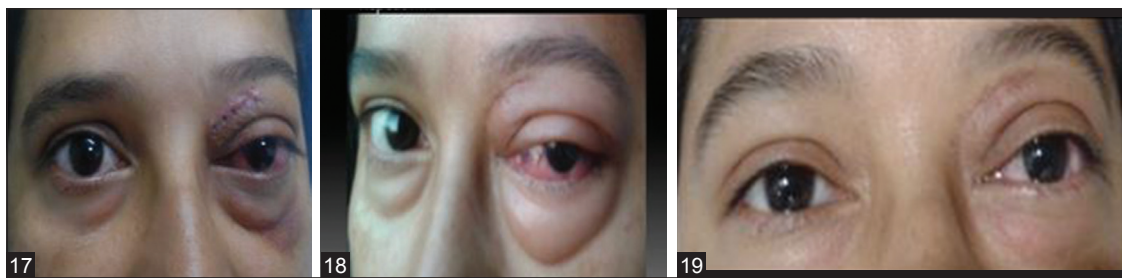


Figure 17-19: (17) Postoperative day 7; (18) Recurrence; (19) Resolution

Management by endovascular route is routine. This case was noteworthy in that we resorted to open orbital surgical cannulation of SOV to gain endovascular access. The location of SOV in the superomedial orbit necessitates an anterior orbitotomy and localizing the incision with the supratrochlear notch as the center of incision is important. Exposure and incision of the orbital septum are standard, but location of SOV in the orbital fat with variations of structure can complicate access. It requires fairly long and meticulous dissection to obtain the first glimpse. Once a large tributary is seen, it is easy to dissect and expose this surprisingly large vessel. Patients with thin, thrombosed veins are more difficult to expose and require lateral orbitotomy or a failure to cannulate.^[5]

Surgical ties and proper isolation are important to help straighten the segment, control flow and obtain hemostasis. The cannulation of the vein was done by the naked eye; the vein sustained a double puncture resulting in bleeding after withdrawing the cannula. Hence, venous cannulation should be done under the microscope or a loupe. Further, since the tortuosity of SOV can distort anatomy making it impossible to discern the correct direction of flow by gross anatomy, assessing the direction by studying the angiograms carefully is of paramount importance.

Postoperative swelling and proptosis resolve slowly compared to resolution in purely endovascular treatment. Localized recurrence due to loose fit of the coil in the deep part of the fistula was noted and was tackled by endovascular transarterial access and occlusion by onyx.^[6]

The lack of online surgical guidelines or videos makes this procedure difficult for nonorbital surgeons. Hence, the surgical details and availability of online videos will help surgeons when such situations arise.

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.

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Conflicts of interest

There are no conflicts of interest.

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