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Short clinical case

Management of two cavernous sinus dural arteriovenous fistulae by direct microsurgical approach and catheterization of the superior ophthalmic vein

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ABSTRACT

Background. – In case of cavernous sinus dural arteriovenous fistula, transvenous embolization of the cavernous sinus via the inferior petrosal sinus is generally sufficient. However, when inferior petrosal sinus access is challenging, various alternative approaches have been reported, with corresponding difficulties and risks.
Case reports. – We report the management of two cases of life-threatening cavernous sinus dural arteriovenous fistula revealed by a typical cavernous sinus syndrome. Conventional approaches were unsuccessful, and a direct microsurgical approach was performed, with catheterization of the superior ophthalmic vein. This combined approach safely accessed the cavernous sinus, and obtained complete occlusion of the fistulae by Onyx® embolization.
Conclusions. – This procedure could be an interesting alternative option in the treatment of cavernous sinus dural arteriovenous fistula when conventional approaches are not possible.

1. Introduction

Cavernous sinus dural arteriovenous fistula (CS-DAVF) raised various treatment challenges for many years [1]. Currently, treatment is dominated by endovascular procedures, of which transvenous embolization (TVE) is the technique of choice, usually conducted via the inferior petrosal sinus (IPS). Other transvenous approaches could be used when the IPS is occluded: superior petrous sinus (SPS) or facial vein (FV) [2–9]. Trans-arterial embolization with coils or liquid agents can be used when the fistula is in one of the branches of the external carotid artery (ECA); when the fistula derives from the branches of the internal carotid artery (ICA), the transvenous approach is often preferred because of the risk of ICA reflux and stroke [10]. Surgery is generally reserved for cases in which endovascular procedures are not possible. Radiosurgery is indicated when the emergency situation may be delayed due to an action interval of months to years, and is thus reserved to low-flow fistula. Whatever the treatment, CS-DAVF can be closed completely in most cases, restoring normal orbital status and resolving visual deficits [11]. In the present cases, because conventional endovascular approaches were impossible and because the degree of urgency was high, a direct microsurgical approach was used with catheterization of the superior ophthalmic vein (SOV) to fill the CS with Onyx® embolization liquid and completely occlude the fistulae, following Jiang et al. [12].

2. Short case reports

First report: a 75-year-old post-menopausal right-handed woman presented with a painful red left eye with loss of visual acuity. She was under treatment for hypertension and followed for hypothyroidism; she had never smoked, was not known or treated for glaucoma, and had no history of ocular surgery. She had had left eye pain, with headache and photophobia for two weeks, initially treated by eye drops and local corticosteroids but without efficacy. Exophthalmia associated with chemosis were noted initially. She also presented cavernous sinus syndrome (CSS), characterized by pulsatile exophthalmia, ophthalmoplegia with involvement of the left VI (horizontal binocular diplopia) and the left III cranial nerves (mydriasis and proptosis). Ophthalmic examination revealed 360° "jellyfish head" venous dilation, clear cornea, papillary drusen by

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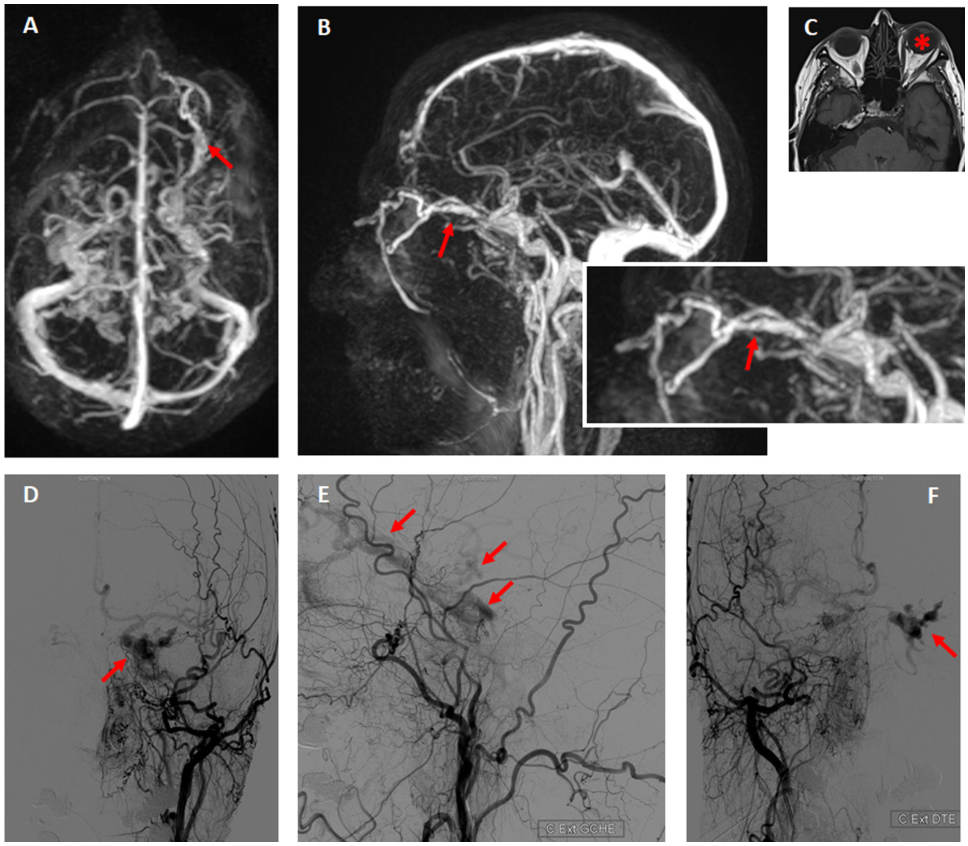

Fig. 1. First report: characterization of the CF-DAVF. MR angiography showing left CS-DAVF on axial (A) and sagittal (B) views. Varicosous dilation of the left SOV (magnification of B). There is extensive early enhancement of the left CS and an ectasic aspect of the ophthalmic vein which also shows early enhancement with gadolinium. (C) Low-grade left exophthalmia (red asterisk, MRI, T1-sequence). Arteriography of the left ECA confirming the dural fistula on frontal (D) and sagittal (E) views. The dural fistula is also enhanced by catheterization of the right ECA (F). Red arrows = fistula.

autofluorescence, impaired left eye visual acuity at 4/10 and lower left homonymous quadranopsia, with the bottom of the eye normal. Left CS dural fistula was suspected. Low-grade left exophthalmia was observed on MRI (Fig. 1C). MR angiography showed left ventral CS-DAVF (Fig. 1A and B) with retrograde reflux in the SOV (magnification of Fig. 1B). Dynamic slices showed extensive early enhancement of the left CS and SOV ectasia which also showed early enhancement with gadolinium. Left ECA arteriography confirmed the fistula (Fig. 1D), also enhanced by catheterization of the right ECA (Fig. 1F). Symptoms were not alleviated by acetazolamide. The first attempt consisted in retrograde embolization via the left IPS (red arrow) then the right, but anterior compartmentalization by thrombus made catheterization impossible (Fig. 2A). Direct ultrasound-guided puncture of the left FV failed due to insufficient FV dilation despite an a priori favorable radiological aspect (Fig. 2B). Another attempt at retrograde venous catheterization of the left and right FVs by femoral puncture also failed, as the veins were too small (Fig. 2C and D). Given the visual prognosis, it was decided to perform a microsurgical approach to the left SOV to reach and occlude the CS with Onyx® embolization liquid (Medtronic, Minneapolis, Minnesota, USA) (Fig. 2E, and zoom on Fig. 1B). The procedure was performed in a neurosurgical operating room under microscopy with 3D fluoroscopy monitoring. After orbital removal (Fig. 2F), the SOV was exposed by the assistant using ophthalmological retractors, and punctured by the neurosurgeon with a pink Cathlon® catheter (BD Insyte™ Autoguard™ 20 G, Becton Dickinson, Franklin Lake, New Jersey, USA). It was easy to locate the arterialized SOV under the microscope, because it was ectasic due to the venous congestion. The catheter was then held in place manually throughout the procedure; it would have been difficult to secure the catheter otherwise, as there was no structure to which it could be sutured. The neuroradiologist the directly injected the Onyx® into the SOV until the CS was completely filled (Fig. 2G). Progression into the CS was monitored by intermittent digital subtraction angiography (DSA) via the carotid artery access. Postoperative arteriography showed complete exclusion of the fistula on sagittal view after injection of contrast medium in the left ICA (Fig. 2H), within the right ICA on frontal view (Fig. 2I), and within the left ECA on frontal (Fig. 2J) and sagittal (Fig. 2K) views. The catheter was removed at the end of the procedure, without bleeding. The orbital bone was then repositioned, followed by classic closure. Immediately after procedure, the patient developed major palpebral congestion that rapidly resolved. Pain and exophthalmia resolved completely a few hours post-procedure. Oculomotor palsy and proptosis resolved within 2 months. Slight sensory disorder of the left supraorbital region persisted. There was no esthetic blemish due to the orbital approach. At 1 year, visual acuity was stable and normal for age, and the dural fistula was still excluded on control MRI and arteriography.

Second report: like the first case, this was a 75-year-old postmenopausal right-handed woman who presented with a painful red right eye with chemosis, exophthalmia (Fig. 3A) and ocular hypertonia, without diplopia or loss of visual acuity. Right ECA and ICA arteriography showed enhancement of a right ventral CS-DAVF with retrograde reflux in the SOV and inferior ophthalmic veins (Fig. 3B–E). Transient improvement in symptom occurred under curative-dose anticoagulant and acetazolamide. Embolization failed after attempting to catheterize i) the right IPS, ii) the right FV, iii) and the right ECA. Again, a combined approach to expose and catheterize the right SOV with Onyx® was performed,

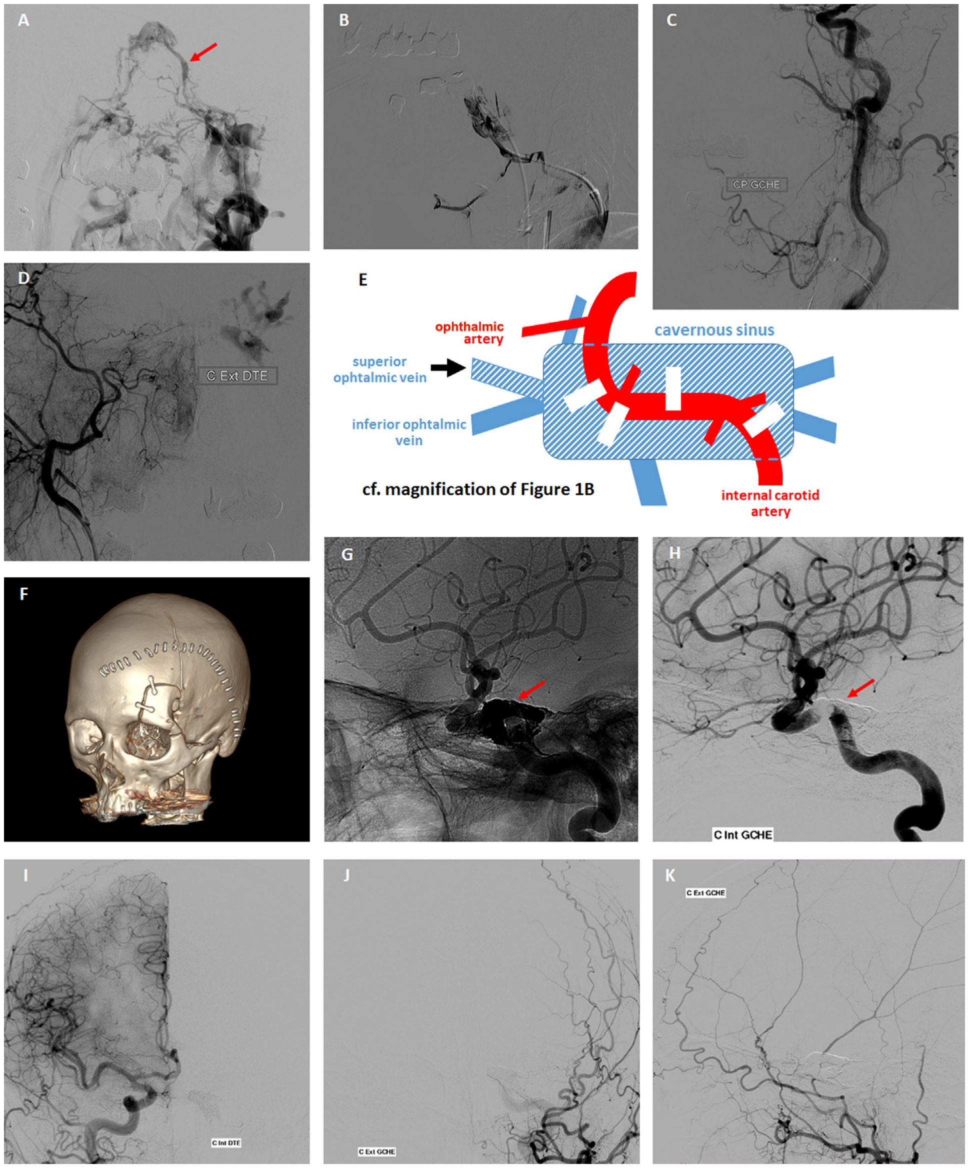

Fig. 2. First report: treatments. A. First attempt at retrograde embolization via the left IPS (red arrow) then the right, but anterior compartmentalization by thrombus made catheterization impossible (lateral view). B. Attempt at direct ultrasound-guided puncture of the left FV (sagittal view). C and D. Attempt at retrograde venous catheterization of the left and right FV by femoral puncture, but veins were too small (frontal views). E. Microsurgical approach to the left SOV followed by retrograde venous catheterization of the CS (hatched, sagittal view) by micro-catheter and injection of Onyx® (black arrow, and cf. magnification of Fig. 1B). F. 3D bone reconstruction showing left orbital removal. G. Complete filling of the CS. Arteriography showing complete exclusion of the CS-DAVF after injection of contrast medium within the left ICA on sagittal view (H), within the right ICA on frontal view (I), and within the left ECA on frontal (J) and sagittal (K) views.

under the same conditions, without complications. There was complete immediate postoperative remission of pain (Fig. 3F and G) and complete resolution of ophthalmological symptomatology with a conserved visual acuity in the following months. Angiograms again showed complete stable exclusion of the CS-DAVF at 1 year.

3. Discussion

The traditional treatment for CS-DAVF is endovascular occlusion [1]. The most commonly used venous route is the IPS. Other routes such as the SPS or FV can be used in second line [2]. In some patients, however, venous routes are not feasible because of venous sinus thrombosis, tortuosity, stenosis or previous embolization. Direct transorbital embolization has also been proposed, but may induce eye and cranial nerve injury, causing blindness or even life-threatening ICA injury [3–9,11]. If all these techniques fail, the last option consists in a surgically approach to the orbit, exposing the SOV for direct puncture [12]. We successfully performed such surgery without cranial nerve injury in these two similar cases. However, the surgery was made easier by the fact the SOV was dilated, making it easily identifiable and accessible under an operating microscope; the risk of SOV rupture was increased but, in view of the visual prognosis and the fact that there was no other back-up, the risk/benefit ratio was in favor of the procedure. To help achieve accurate puncture pathway and depth, Dashti et al. accessed the SOV via direct percutaneous transorbital puncture [13]. Liu et al. went further, reporting a safer and more precise direct transorbital approach under fluoroscopy superimposed on the 3D-reconstructed skull image combined with DSA [14]. Neuronavigation systems could provide an interesting alternative in cases where the SOV is atretic or inaccessible to direct surgical procedures. The progression of Onyx® into the CS must be monitored by intermittent carotid artery angiography. In case of retrograde reflux during proximal injection into the SOV, the procedure must

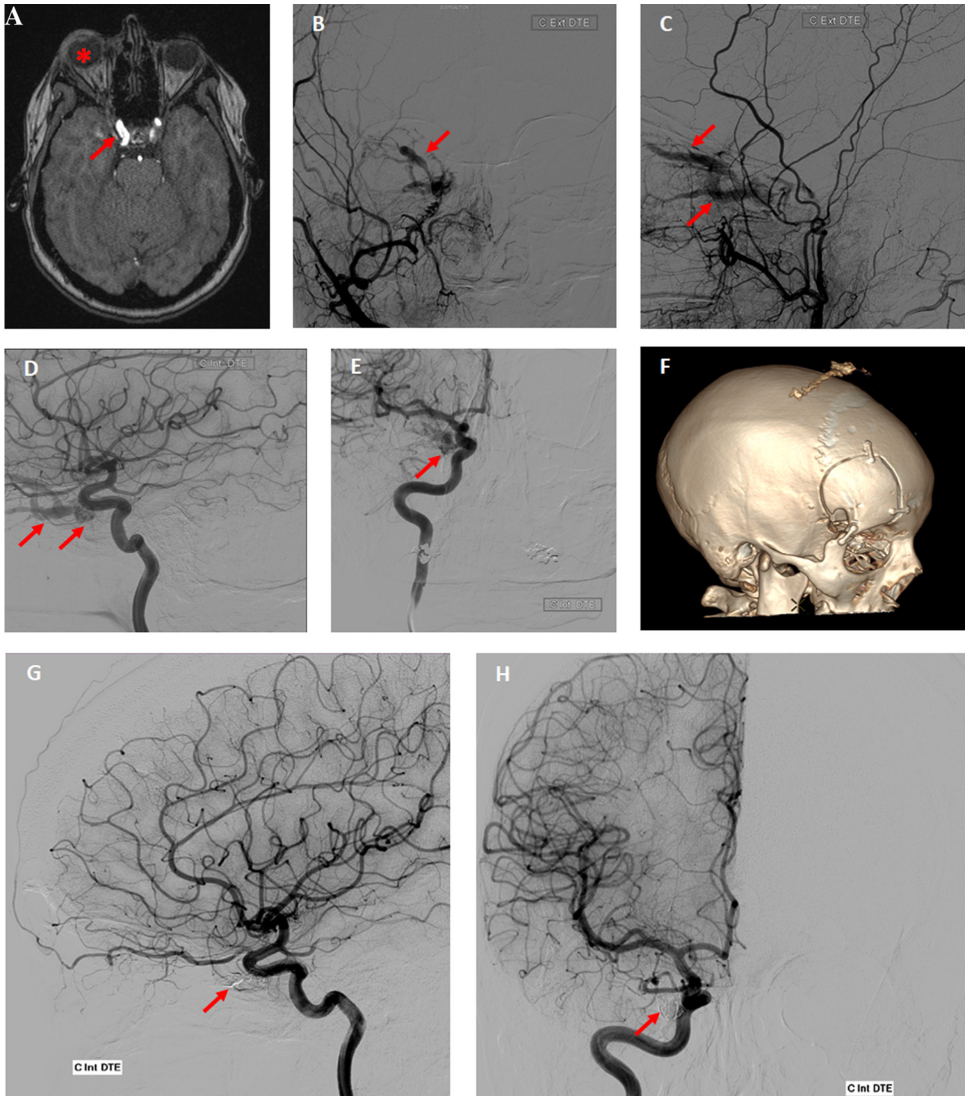

Fig. 3. Second report. MR angiography showing right CS-DAVF (red arrow) on axial view A. Note exophthalmia (red asterisk, MRI, T1-sequence with gadolinium). Arteriography of the right ECA showing enhancement of a fistula (red arrows) on frontal views at various acquisition times (B and C). The dural fistula (red arrows) is also enhanced by catheterization of the right ICA (D and E). F. Right orbital removal giving access to the right SOV. (G and H) Complete exclusion of the CS-DAVF with Onyx® (red arrows).

be stopped and the catheter be replaced. In addition, Onyx® can continually block the sinus during needle withdrawal, significantly reducing the severity of retrobulbar hematoma compared with conventional transorbital coil embolization [14]. Toxicity has been reported with Onyx® in the treatment of CCF, but it is not possible to say whether the toxicity was due to the product itself or to filling the CS [15]. After 1 year's follow-up, no residual shunt was observed on angiograms. Imaging follow-up comprised one MRI and one angiography, postoperatively to check complete exclusion of the fistula and at 1 year. Subsequent follow-up will be mainly based on clinical examination and annual MRI; no arteriography required. We consider that no CS enhancement after gadolinium injection is sufficient for MRI follow-up, as described in the literature [16,17]. Follow-up is imperative. In case of recurrence of symptoms or CS gadolinium enhancement on MRI, repeat arteriography is urgently required to update the situation. Finally, there was no esthetic blemish following orbital bone removal; if at any time blemish occurs, it is possible to fill the defects with fat or substitute.

4. Conclusion

The traditional treatment for CS-DAVF is endovascular occlusion. The goal is to interrupt the fistulous communications and decrease pressure in the CS. The most commonly used venous route is the IPS. Other routes such as the SPS or FV can be used in second line. If all these techniques fail, the SOV provides an alternative approach, but requires surgical exposure. Here, we demonstrated that this combined procedure is safe and completely excludes CS-DAVF.

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Disclosure of interest

The authors declare that they have no competing interest.

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