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## Case Report

# Transorbital hybrid approach for endovascular occlusion of indirect carotid-cavernous fistulas–Case report and systematic literature review<sup>☆</sup>

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## ABSTRACT

Carotid-cavernous fistulas (CCF) are vascular malformations characterized by an aberrant shunt between one or more sources of arterial inflow and the cavernous sinus (CS). They are subdivided into direct and indirect fistulas. This last one, called dural CCF involve dural fistulous connections between branches of the internal carotid artery or the external carotid artery. When conventional routes are not eligible, surgical exposure of the vein is the only access to the fistula. We present the case of a patient successfully treated for right sided dural CCF, by a hybrid approach. Furthermore, through a literature review, we analyze the possible risks and benefits associated with this approach.

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## Introduction

A carotid-cavernous fistula (CCF) is an abnormal arteriovenous connection between the vein drained to the cavernous sinus (CS) and the cavernous segment of the internal or ex-

ternal carotid artery, accounting for 10%–15% of all intracranial vascular malformation (ICA) [1,2]. Fistulas could be classified according to the intracranial localization or to the venous drainage type [3]. Patients with dCCF are often diagnosed with a significant time delay, for nonspecific and mild clinical symptoms. The goal of treatment of high flow CCF is to

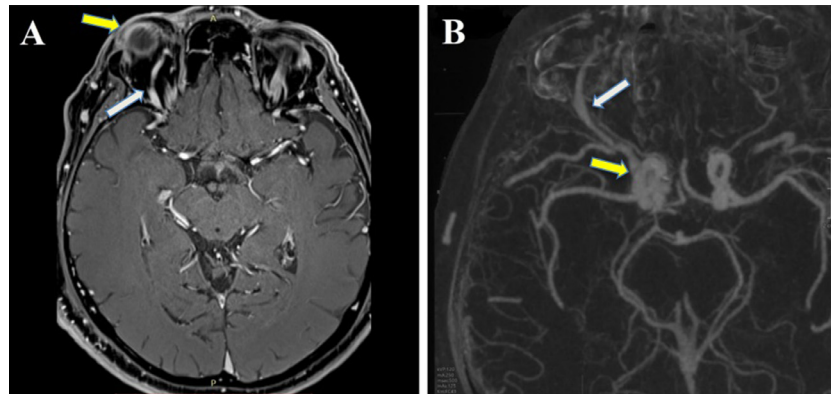
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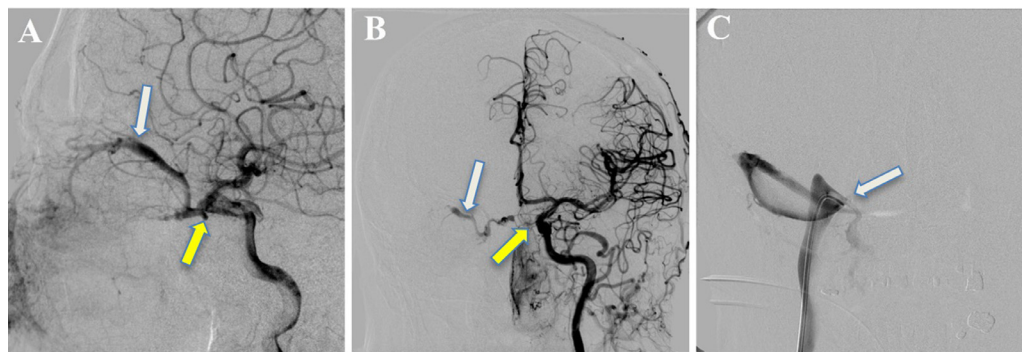
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**Fig. 1 – (A) Contrast-enhanced magnetic resonance brain imaging reveals a proptosis (yellow arrow) and an enhancement of the right SOV (white arrow) (B) CT angiography revealed early enhancement of the right CS (yellow arrow) with dilated right SOV (white arrow).**



**Fig. 2 – (A) DSA of the right internal carotid artery (ICA) in lateral projection showing dural arteries of the carotid siphon feeding the right cavernous sinus shunts (yellow arrow) and deeply located dilated superior ophthalmic vein (SOV) (white arrow) (B) DSA of the left ICA in anteroposterior projection showing irrigation via small dural branches of the left siphon draining (yellow arrow) in the right CS and the right SOV (white arrow) (C) The lack of access to the inferior petrosal sinus from the catheter in the right jugular bulb (white arrow).**

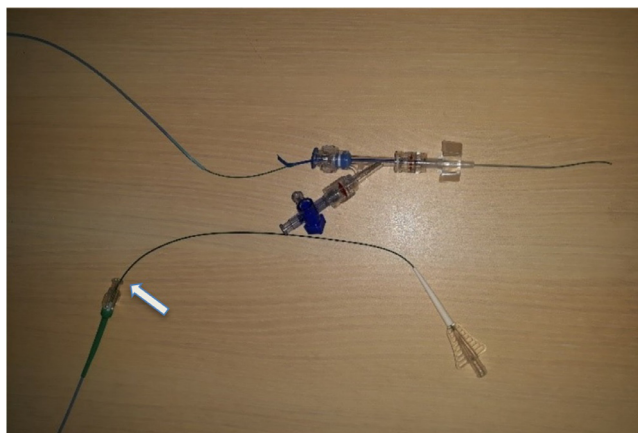
obliterate the arteriovenous shunt at the site of the fistula, preserving the patency of the ICA as much as possible. Endovascular embolization has become the standard treatment of dural carotid cavernous fistula (DCCF). Numerous endovascular routes are able to obtain the cavernous sinus access, including the inferior petrosal sinus, superior petrosal sinus, facial and ophthalmic veins, pterygoid plexus, and the vein of Labbe. Despite the emergence of the aforementioned endovascular procedures, in some cases the complex anatomy can preclude conventional transarterial or transvenous approaches. In these cases, the recent advances in transorbital techniques become a treatment option for the management of CC.

We present our experience of transorbital CCF embolization and demonstrate that this approach is a viable alternative to conventional methods for treatment of selected CCF cases. We supplement our case with a literature review.

### Case report

A 62-year-old man with a posterior fossa cavernoma and type I Arnold-Chiari malformation presented a progressive visual

loss and right monocular pain. On clinical examination, his visual acuity was 0.7 in RE; whit diplopia, chemosis and exophthalmos. A fundoscopy exam did not showed signs of disc edema, optic atrophy, or abnormalities in arterial and venous pulsation. Standard blood test was unremarkable except for moderate leukopenia ( $3.76 \times 10^9/L$ ) and thrombocytopenia ( $138 \times 10^9/L$ ). The magnetic resonance imaging (MRI) demonstrated the posterior fossa cavernoma with no signs of bleeding, as well as herniated cerebellar tonsils in the spinal canal to the level of the lower edge of the posterior arch of the atlas, without syringomyelia. The angiography (MRA) showed exophthalmos with an ectasia of the right SOV (Fig. 1), without signs of orbital soft tissue swelling. He was, initially, treated with 3% solution of boric acid, neomycin and dexamethasone eye drops 4 times a day. Due to further worsening of visual acuity on the right eye, additional tests were performed. The patient subsequently underwent a diagnostic cerebral angiography, which showed engorgement of the right ophthalmic vein and proptosis. So, the diagnosis of a dCCF of the right CS was suspected, and the patient was medicated with acetylsalicylic acid (100 mg) and clopidogrel (75 mg), 5 days before the embolization.



**Fig. 3 – Prior to the procedure, it was verified that the microcatheter Headway DUO is able to pass through the 17G (white) cannula. Headway DUO was previously coaxially inserted into 5-Fr diagnostic vertebral catheter for better support (white arrow).**

Digital subtraction angiography (DSA) demonstrate the existence of an dural CCF of the right CS or Barrow type B according to Barrow classification (Fig. 2). The decision was made to embolize the right sided CCF.

The diagnostic catheter was advanced through the external jugular vein into the right facial vein over a guidewire. Venous outflow anatomy of the CCF and anatomy of the periorbital cortical veins were assessed via contrast administration through the right ICA.

An initial attempt was made to catheterize or reopen inferior or superior petrosal sinus on both sides failed (Fig. 2). Both angular veins were inaccessible due to small caliber and tortuosity. In the absence of the venous access to the right CS, further intervention was discontinued. After discharge, conservative treatment with intermittent manual self-compression of the carotid arteries was recommended to the patient.

As no signs of improvement were reported. Therefore, for the lack of transvenous route into the right CCF, the decision was made to directly access the CS through a surgical and endovascular method combined "transorbital hybrid approach."

Prior to the procedure, it was verified that the microcatheter Headway DUO 0.0165" (MicroVention/Terumo, Tustin, California) is able to pass through the 17G (white) cannula. Also, the system of telescopic catheters was tested before the procedure (Fig. 3).

Surgical exposure of the SOV was performed under general anesthesia. A small curvilinear incision was performed over the shorn eyebrow for aesthetic reasons, and subcutaneous tissues were carefully dissected to expose the vein.

Metallic hemostat was placed and DSA was repeated through the 5Fr diagnostic catheter from the right ICA (Fig. 4). Following surgical exposure, SOV was cannulated with an 17G cannula. In the first attempt, insertion of the microwire ASAHI CHIKAI 0.010" (Asahi-Intecc, Aichi, Japan) was unsuccessful due to lack of its stiffness. In the second attempt, microwire ASAHI CHIKAI 0.014" (Asahi-Intecc, Aichi, Japan) successfully passed through cannula into SOV and right CS (Fig. 4). Hemo-

static valve was attached on the cannula, and microcatheter Headway DUO advanced over the microwire into the right CS. Vertebral 5F catheter previously telescopically positioned over Headway DUO, was inserted into the hemostatic valve, as a support to the microcatheter. CS was then obliterated with the coils, and then D.M.S.O. with E.V.O.H. copolymer (Squid 12 Balt, Montmorency, France; Fig. 4). These coils were submerged in thrombin prior to deployment to promote thrombosis. Postembolization angiography demonstrated no significant residual shunting from the right ICA through the CCF (Fig. 4). The venous catheter and sheath were removed, the vein was ligated, and the eyebrow incision was closed.

The patient's visual acuity improved immediately, and intraocular pressure was normalized. One year after the intervention, brain MRI with angiography confirmed stable obliteration of the arteriovenous shunt. Follow-up at three weeks demonstrated complete resolution of diplopia (Fig. 5).

At the latest follow-up at 24 months, the patient presented no recurrences or new symptoms

## Discussion

Carotid cavernous sinus fistula (CCF) represents abnormal connections between the external and/or internal CA and the cavernous sinus [1]. CCFs have been classified according to the hemodynamic properties, anatomy of the fistula and venous pattern. Hemodynamic classification separates CCFs into high-flow and dural fistulas. According to the angiographic findings, Barrow et al. [2], provided a detailed anatomical classification. In 1995, Cognard et al. [3], developed a different classification system based on venous flow pattern in association with a prognostic judgment. Borden's classification [4] suggests that the most important predictive factor of aggressive clinical behavior is the presence of leptomeningeal venous drainage.

However, these classifications lacked predictive factors for the clinical presentation, natural history, and hemorrhagic risk. Therefore, Leone et al. [5], validating the Thomas classification, provide useful informations regarding the anatomy of the fistula and the venous hemodynamics, and its relationship with the clinical presentations.

Symptoms depend on rate of flow, location of venous drainage of CCF, inflammation, and pressure within the venous system. Anterior drainage into the superior ophthalmic vein is associated with ocular and neuro-ophthalmological symptoms while retrograde cortical venous flow presents with neurological symptoms because of venous congestion or infarction within the supra- or infra-tentorial compartments.

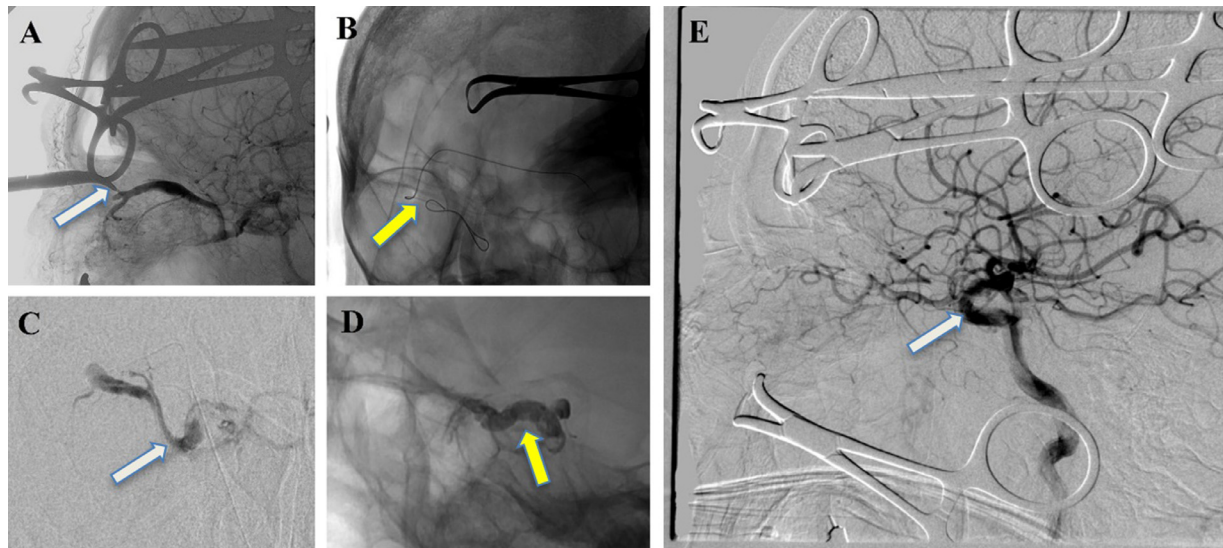
The goal of the treatment is the complete occlusion of the fistula with preservation of normal flow of blood in ICA.

This could be achieved by glue, liquid emboli, coil, or mesh.

Historically, CCA ligation was the choice for the treatment of patients with CCF. In 1933, Hamby and Gardner described the first successful intracranial ligation of the carotid artery for the treatment of this disease [6].

To date, endovascular techniques are the mainstay approach for CFFs, by occluding the arterial branches supplying the fistula (transarterial embolization) or, more commonly,





**Fig. 4 – (A) DSA identification of the superior ophthalmic vein (SOV) at the tip of the hemostat plier (white arrow) (B) Transvenous microwire looping into the cavernous sinus (CS) through SOV (yellow arrow) (C) Microcatheter injection with opacification of CS and SOV, tip of the microcatheter positioned at the posterior part of CS (white arrow) (D) CS filled with coils and liquid embolic agent E.V.O.H. (Squid 12), resulting in complete obliteration of the fistula (yellow arrow) (E) Visualization of the right internal carotid artery (white arrow) with complete closure of the fistula.**



**Fig. 5 – Procedure outcomes and recovery (A) Chemosis, proptosis and paresis of the right CN IV before embolization (white arrow) (B) Resolution of chemosis and proptosis 1 month after embolization (yellow arrow).**

the cavernous sinus that harbors the fistulous communications (transvenous embolization). Transarterial access is, often, used when the CCF originates from branches of the ECA, as well as in select cases of direct fistulas.

When the CCF originates from branches of the ICA, the transarterial embolization increase the risk of cranial nerve palsy and stroke, for the obliteration of meningeal and pial vessels. In this case, and also when ECA embolization is ineffective, is necessary the venous route.

It has become the treatment of choice for symptomatic CCFs due to its better results compared with other therapeutic modalities.

Transvenous access can be established through the femoral vein, internal jugular vein, or direct puncture to affected sinus by burr hole, craniotomy, or ultrasound-guided puncture of a pericranial venous pouch. In the majority of cases, venous access to the cavernous sinus may be gained via the ipsilateral inferior petrosal sinus with a success rate between 60% and 76% [7,8].

The surgical intervention is adopted when the endovascular treatment is impossible.

In these cases, it is necessary a direct cut down on the superior ophthalmic vein (SOV). That is the side of retrograde cannulation for the cavernous sinus dural fistulas. Hanneken et al. in 1989, proposed the superior ophthalmic vein approach how direct access to cavernous sinus [1,9,10]. This procedure cannot be performed if the SOV is thrombosed, and there are a number of potential complications, which include difficulty in determining the direction of the flow, orbital hemorrhage, orbital infection, possible injury to trochlea and loss of vision associated with acute orbital congestion secondary to thrombosis of the ophthalmic veins [11]. According to different authors, an orbital approach for transvenous embolization has been demonstrated to be a safe procedure that improves pre-operative ocular morbidities in the majority of patients.

Direct hybrid approaches, associated endovascular technique with the cannulation of the superior ophthalmic vein (SOV) by direct percutaneous puncture to reduce the net distance and catheter manipulations to reach the cavernous sinus [12–17].

This approach is useful to achieve complete obliteration of a fistula owing to inadequate initial embolization, recurrence in the setting of access occlusion from previous embolization, or complex fistula pouches requiring access to multiple CS compartments.

This approach may include risk of damage to the trochlea or other orbital structures, resulting in retroocular hemorrhage and/or infection; lesions of the supraorbital nerve or elevator upper eyelid muscle [18]. Furthermore it could be complicated by intraorbital hemorrhage [17], vision loss and secondary neurovascular glaucoma [10], acute exophthalmos [19], cavernous sinus occlusion [20], late occlusion of the central retinal vein and tromboflebitis [21].

In conclusion, our case shows feasibility and safety of the hybrid transorbital approach for dural CCF not successful and properly treated by conventional endovascular access to the cavernous sinus.

## Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper

## Author contributions

Acquisition of data: Vukasinovic I, Bascarevic V, Nedeljkovic Z, Nedeljkovic A, Grujicic D. Analysis and interpretation of data: all authors. Drafting the article: all authors. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Study supervision: Grujicic D.

## Patient consent

Written informed consent to publish this case and use anonymized radiologic material was obtained from the patient.

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