

## Case report

## Bilateral ethmoidal dural arteriovenous fistula: Unexpected surgical diagnosis

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## ABSTRACT

Dural arteriovenous fistulae (DAVFs) are infrequent lesions, the most common locations of which are the cavernous, sigmoid and transverse sinuses. The cribriform plate is one of the less frequent sites for DAVFs, where they entail a high hemorrhage risk. Feeding arteries for ethmoidal DAVFs can be uni- or bilateral. However, the draining fistulous system has classically been described as unilateral. The authors report the second case in literature of bilateral ethmoidal DAVF, which is defined as that with bilateral draining veins. The present case was diagnosed only after surgical exploration of both cribriform plates. No preoperative radiological test could detect the presence of a bilateral venous draining system from the ethmoidal DAVF. Possible reasons for that lack of presurgical diagnosis are discussed. Bilateral surgical exploration of the anterior cranial fossa is recommended when dealing with ethmoidal DAVFs, even when they seem to be unilateral on preoperative studies.

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## 1. Introduction

Dural fistulae (DAVFs) are defined as abnormal arteriovenous connections confined to a certain region of pachymeninges [4,6]. They comprise 10–15% of all intracranial vascular malformations [15]. Cavernous, sigmoid, and transverse sinuses are their most frequent locations [2,4,11]. DAVFs can be clinically grouped into benign (asymptomatic, bruit, headache, tinnitus or ocular symptoms) and aggressive (hemorrhage or dementia) [2,4–7,12,13,19]. The venous drainage pattern has been reported as the main feature determining the DAVFs clinical presentation [2–5,7,9,12,17–20].

Anterior cranial fossa DAVFs have been scarcely reported [11,13,14]. They have a higher risk of hemorrhage than DAVFs in other locations [1,8,11,13,14,16]. Their arterial supply usually comes from the ethmoidal or the ophthalmic arteries, and it is often bilateral [1,11,13]. On the other hand, their venous drainage is far more commonly unilateral [10,11,13], being accomplished through cortical veins of the frontal lobes into the superior sagittal, inferior sagittal or cavernous sinuses [8,11,13].

We report the second case of bilateral ethmoidal DAVF and review the literature on this regard.

## 2. Case report

## 2.1. Clinical onset

A 41-year-old man was assisted at our department after a generalized tonic-clonic seizure. The patient was fully conscious when explored. No focal deficit was observed on thorough neurological examination. He only complained about mild bifrontal headache and episodic amnesia. The patient's previous clinical history was unremarkable, except for a moderate frontal head trauma 21 years before.

## 2.2. Radiological study

Initial CT scan displayed a left polar frontal hemorrhage, associated to an acute laminar subdural hematoma in the anterior interhemispheric fissure and the left frontal convexity.

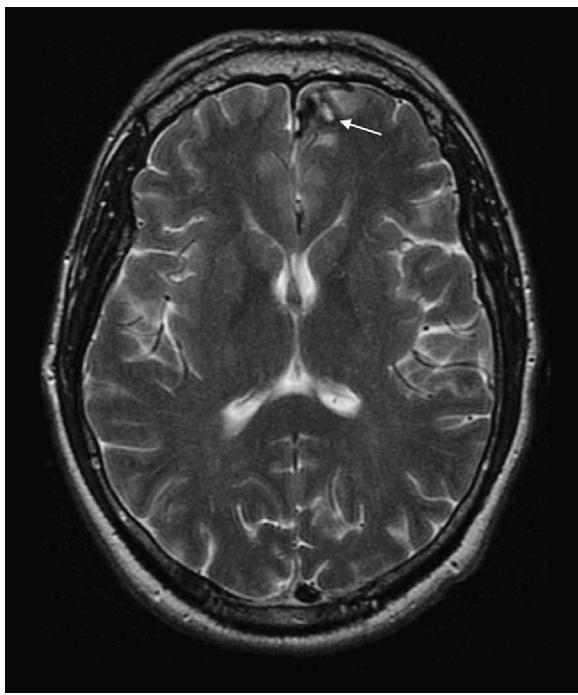
MRI only showed a round parenchymal lesion in the left frontal lobe (Fig. 1), while MRA disclosed the whole anatomy of an ethmoidal DAVF (Fig. 2). The round lesion seen on MRI turned out to be a venous aneurysm of the DAVF (Fig. 2 right).

Angiography showed the DAVF anatomy in detail. Multiple feeding arteries came bilaterally from the ethmoidal arteries. These feeding arteries pierced the cribriform plate and the orbital roofs to

**Abbreviations:** AVM, arteriovenous malformation; CT, computed tomography; CTA, computed tomography angiography; DAVF, dural arteriovenous fistula; ECA, external carotid artery; ICA, internal carotid artery; MRA, magnetic resonance angiography; MRI, magnetic resonance image.

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**Fig. 1.** Axial T2-weighted MRI shows a round lesion in the left frontal pole lobe (white arrow), which corresponds to the parenchymal hematoma in evolution. The fistulous vein is seen as a hypointense area medially to the hematoma.

enter the skull. The DAVF nidus was detected as a mesh of engorged vessels in the dura mater covering the cribriform plate and its neighborhood (Fig. 3A–C). Venous drainage was made through a cortical frontal vein on the left side. This vein had a huge aneurysmatic dilation, which was considered the bleeding source for its close relation to the hematoma (Fig. 3A–C). Some other small veins were detected at the midline coming up from the nidus, though it was not possible to ascertain their side with regard to the falk. According to these angiographic features the DAVF was classified as Borden Grade IIIb and Cognard Grade IV [3,5].

CTA with bone window and coronal reconstruction of the anterior cranial fossa displayed the arteries entrance into the skull through several orifices at the left cribriform plate. The most conspicuous fistulous point, from where the main vein originated, was

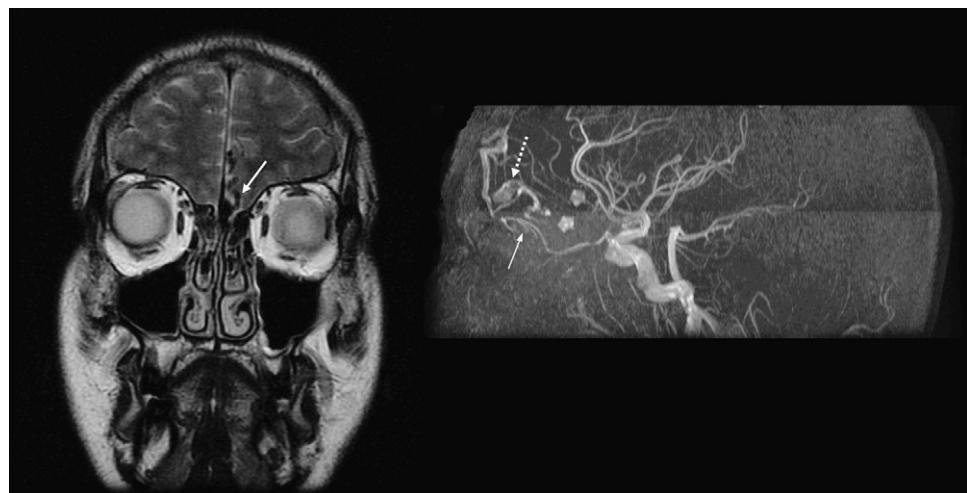
visible at 1.2 mm lateral to the crista galli process on the left side. Venous drainage was only detected unilaterally on the left side (Fig. 4).

### 2.3. Evolution

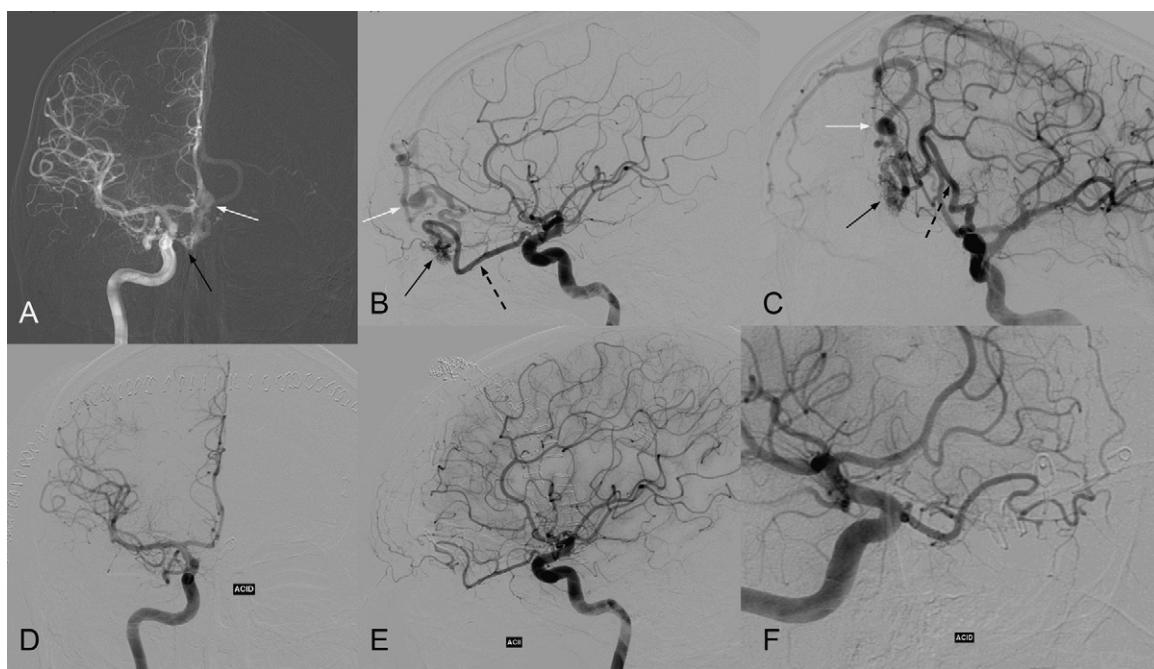
The patient kept asymptomatic thereafter. Surgical management was decided after analysis of all clinical and radiological data.

### 2.4. Surgical procedure

The patient was positioned supine. The head was fixed on a three-pin head holder with pronounced extension but no rotation. A bicoronal skin incision with a myofascial flap was performed. Then, a low bifrontal craniotomy was done. The inner table at the orbital ridge was dissected extradurally and later drilled away in order to get a better access to the anterior cranial fossa. The dura mater was opened bilaterally in a linear fashion parallel to the orbital ridge. The sagittal sinus and the falk were not divided. Once intradurally, thorough exploration of both anterior cranial fossae was accomplished. For that, each frontal lobe was gently retracted upward for better exposure. The DAVF nidus was encountered bilaterally, but with left predominance. An “arterialized” vein (reddish and beating) was easily found on the left side and followed from the nidus to its entrance into a frontobasal sulcus (Fig. 5A). After dissection of this sulcus the huge venous aneurysm became exposed. The vein was clipped just after its origin at the dura mater (Fig. 5C). Afterwards, the nidus was completely coagulated with bipolar forceps. All these maneuvers made the vein progressively turn bluish and “beatless” while the aneurysm shrank. The right anterior cranial fossa was explored latter. Then, another arterialized vein going up from the cribriform plate was encountered (Fig. 5B). This vessel was mirror-like positioned compared to its left counterpart, though the right vessel was smaller. The right vein was also clipped after its origin at the dura mater (Fig. 5D). All engorged dural vessels at the falk and the right anterior cranial fossa suggestive of DAVF nidus were also coagulated with bipolar forceps. Intraoperative cerebral angiography showed complete exclusion of the DAVF, with distal preservation of the ophthalmic arteries (Fig. 3D–F).



**Fig. 2.** (Left) coronal T2-weighted MRI shows the primary fistulous draining vein (white arrow), which is exclusively visible on the left side of the falk. (Right) lateral view of magnetic resonance angiography (MRA) displays the gross anatomy of the DAVF. The nidus is detected at the cribriform plate (white straight arrow). A venous aneurysm is present at the primary fistulous vein (white dashed arrow).



**Fig. 3.** Cerebral angiography. (A) Antero-posterior view of right internal carotid artery (ICA) angiography shows the right feedings arteries (black arrow) to the DAVF and the venous aneurysm (white arrow). (B) Lateral view of left ICA angiography displays the fistulous nodule (black arrow), fed by the left ophthalmic artery (black dashed arrow), and the venous aneurysm (white arrow). (C) Left oblique antero-inferior projection of the left ICA angiography demonstrates the DAVF nodule (black arrow), the feeding left ophthalmic artery (black dashed arrow) and the venous aneurysm (white arrow), with no visible right venous drainage. Post-operative angiographies (D–F) confirm complete exclusion of the DAVF; (D) right ICA antero-posterior view; (E) left ICA lateral view; (F) right ICA lateral expanded view.

### 2.5. Post-operative course

The patient recovered with no problems. Complete cerebral angiography, including both external carotid arteries, was performed six days after surgery. Complete exclusion of the DAVF was confirmed. The patient was discharged home on the seventh day after surgery.

### 2.6. Follow-up

Periodical clinical follow-up has proved the patient to be asymptomatic after 16 months. A control angiography, performed 14 months after surgery, demonstrated complete DAVF resolution (Fig. 6).

## 3. Discussion

Ethmoidal DAVFs comprise 5% of all cranial DAVFs [8,11,13,14]. Despite their rarity they present the highest hemorrhage risk among DAVFs (79%) because of a high frequency of leptomeningeal venous drainage [1,8,11,13,14,16]. Their fistulous connection is located at the cribriform plate [10,11,13]. This anatomical region is not close to any venous sinus. Therefore, venous drainage is more easily performed through cortical veins and eventually to the sagittal or cavernous sinuses [10,11,13]. Cortical venous involvement is associated with venous hypertension and venous aneurysms development, which portend the aforementioned high hemorrhagic risk [11]. This fact warrants ethmoidal DAVF treatment.

### 3.1. Ethmoidal DAVFs pathophysiology

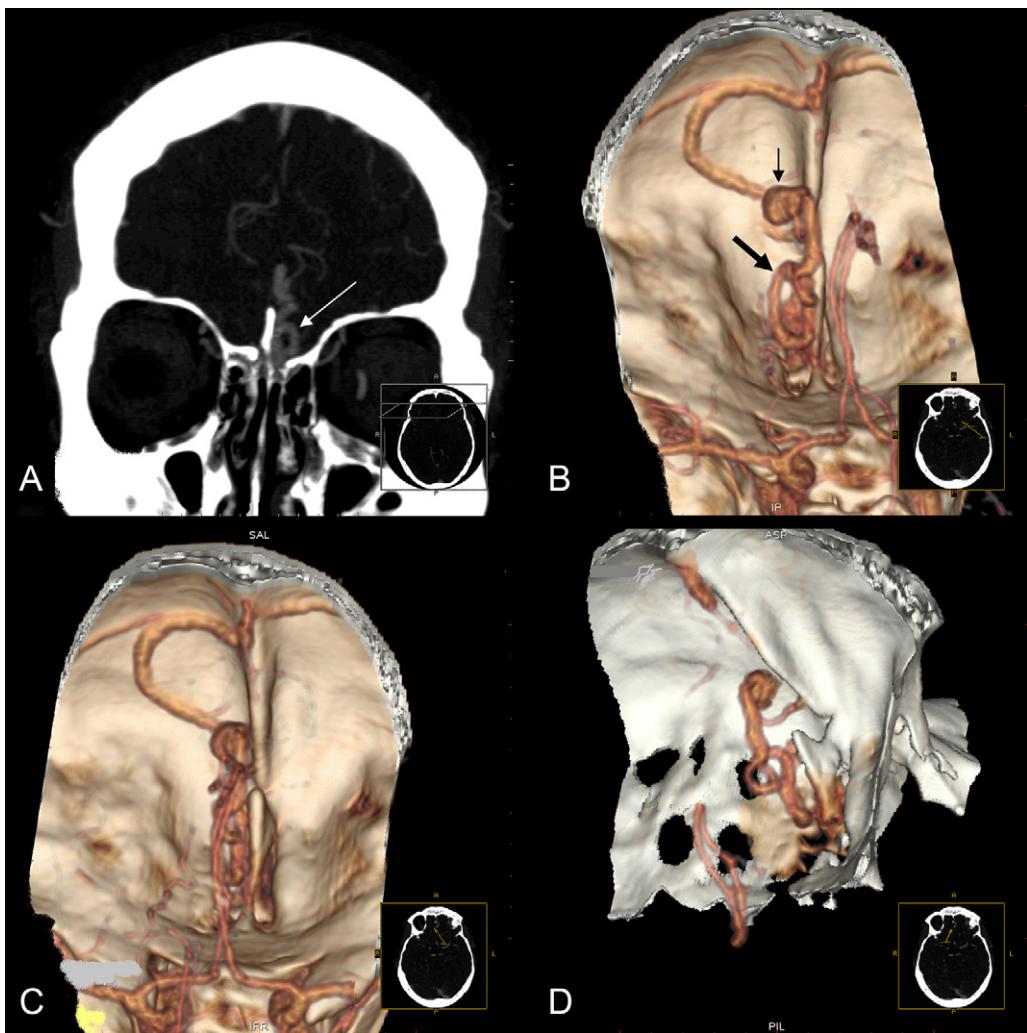
Ethmoidal DAVFs may be fed by both ICA and ECA branches [1,8,10–13]. ICA branches are the most frequent suppliers, especially the anterior ethmoidal artery [1,8,11,12]. However, feeders can also be recruited from the internal maxillary, middle meningeal or superficial temporal arteries [1,8,10–12]. This arterial supply is

commonly bilateral, but with one predominant side [1,11,12]. Thus, the prevailing side labels the ethmoidal DAVFs as right or left.

Ethmoidal DAVFs venous drainage is commonly performed through frontal cortical veins [1,8,10–12]. The venous system complexity may be quite variable, but theoretically it is always unilateral [1,8,11]. That would mean that the whole venous system is situated on one side of the falx. The first case of “bilateral ethmoidal DAVF” challenged this concept [8]. In that case, the DAVF persistence after disconnection of its right vein permitted the detection by intraoperative angiography of a previously unnoticed vein on the left cribriform plate [8]. Our case is another example of bilateral venous system in DAVFs.

### 3.2. Radiological diagnosis

Angiography is the gold-standard for DAVFs diagnosis, showing its vascular anatomy. However, angiography did not detect the contralateral vein in our case, which can be explained by three main reasons. First, angiography is limited to differentiate between sides in complex medial lesions. Second, hyperselective catheterization of ethmoidal DAVFs is rather difficult, which hinders from obtaining detailed images of isolated veins. Third, complex DAVFs may have two types of venous drainage. *Primary drainage* would be performed through the main vein displayed on angiography. *Secondary drainage* would consist of smaller veins that can be hardly detected on non-selective angiographies. The primary drainage is more visible on angiography for both its bigger size and greater flow, which may act as a “venous stealing phenomenon” that keeps blood from draining through secondary veins. Thus, secondary drainage patency could be disclosed after primary drainage occlusion [8]. Once the primary vein is occluded secondary veins become the only draining pathway, with an increasing flow that eventually becomes visible on angiography. This could explain why the “secondary” right vein in our case could not be detected before the “primary” left vein had been occluded.



**Fig. 4.** (A) A Computed Tomography–Angiography (CTA) coronal slice shows the fistulous draining vein (white arrow) located on the left cribiform fossa. (B) A postero-lateral left view of a three-dimensional (3D) CTA reconstruction displays the draining fistulous system, with a primary vein (thick black arrow) and the venous aneurysm (small black arrow). (C) A posterior projection of the 3-D CTA shows no venous fistulous drainage at the right cribiform plate. (D) A right postero-lateral view of the 3-D CTA confirms no fistulous vein emerging at the right side of the crista galli process.

MRI could theoretically set the exact location of both primary and secondary veins. However, neither MRI nor MRA could clearly detect secondary veins in our case, probably due to their small size and slow flow.

### 3.3. Ethmoidal DAVFs treatment

Three options are currently available for ethmoidal DAVFs management. Surgery has proved the highest healing rate [1,8,11–14,16,20]. Surgical DAVF disconnection can be achieved by venous disconnection [6,7,19] and nidus obliteration [6,7,11,13,14,16,19]. The first maneuver is mandatory and should be done just at the fistula site, both by clipping [6,8] or division of the vein [6,11,13,14,16]. The latter is optional and could be obtained after dura mater coagulation [6,7,11,13,14,16,19]. We consider that dural coagulation secures DAVF exclusion in cases with multiple feeding arteries and a broad dural settlement (Borden subtype b), and when venous occlusion cannot be accomplished at the fistula site.

Embolization rarely achieves complete occlusion of ethmoidal DAVFs [1,4,8,12,13,16,20] due to a difficult endovascular access [1,4,8,12–14]. Moreover, there is a high risk of visual impairment for retinal arteries occlusion [1,4,8,11–14], and parenchymal injury

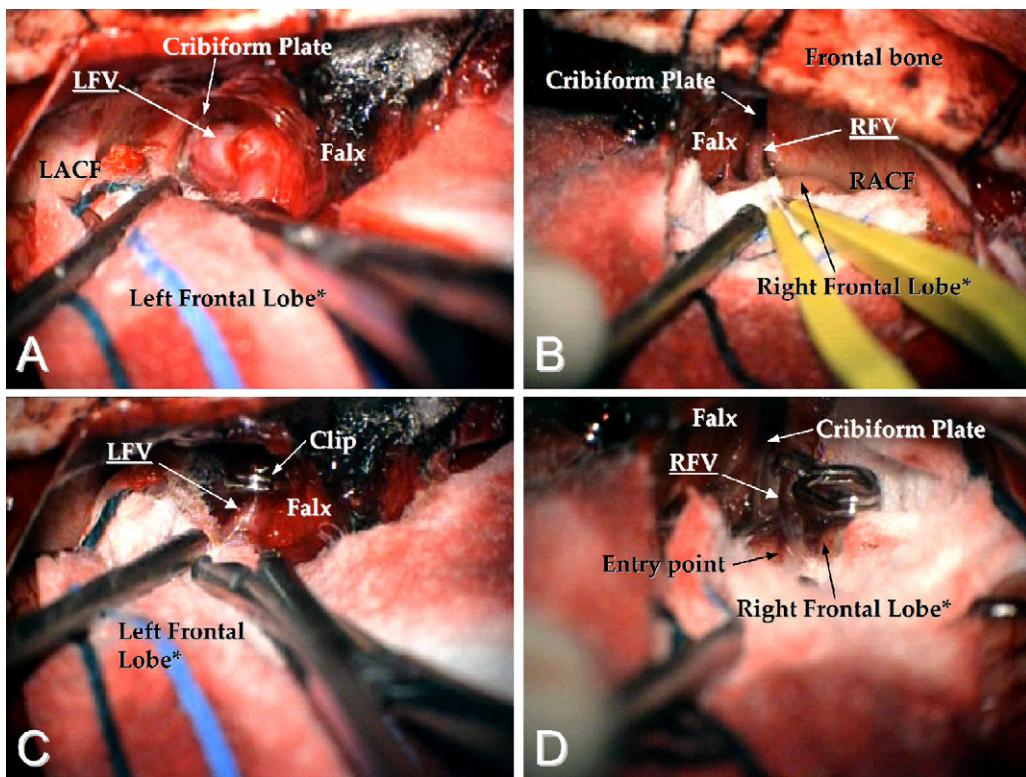
after ICA damage or embolic material reflux [1,4,8]. Thus, the role of endovascular approaches in ethmoidal DAVFs is reducing the arterial supply, defining the vascular anatomy [8] or being an alternative when surgery is not feasible.

Radiosurgery shall not be considered as an actual treatment option for DAVFs [1]. Only few cases with complete occlusion have been reported [1,14]. Furthermore, a latency period until healing is not acceptable for lesions that have already bled or have a high bleeding risk [1].

### 3.4. Surgical approaches for ethmoidal DAVFs

Three surgical approaches are used for ethmoidal DAVFs. With the *orbitozygomatic approach* less brain retraction is achieved by means of greater bone removal [8]. However, the orbit content may be injured and the falx must be incised to explore the contralateral fossa [8], which implies an added technical difficulty. Moreover, it is not a familiar approach for most neurosurgeons.

The *anterior interhemispheric approach* spares the frontal sinus with less risk of CSF rhinorrhea, intradural infection and mucocele development [14]. However, it has a high risk of sagittal sinus and bridging veins injury. Surgery is performed through a long corridor, between frontal lobe and falx, increasing the risk of brain



**Fig. 5.** Surgical high-magnification photographs. (A) Left anterior subfrontal view under operating microscope of the left fistulous vein (LFV). The left frontal lobe, which is covered by cottonoids (\*), has been retracted in order to show the left anterior cranial fossa (LACF) and the cribiform plate, from where the LFV comes up into the brain. (B) Right anterior subfrontal view, under operating microscope, of a previously undetected right fistulous vein (RFV). The right frontal lobe is gently retracted with help of cottonoids (\*). Thus, the right anterior cranial fossa (RACF) and the right cribiform plate are exposed. This maneuver shows the RFV trajectory from the cribiform plate to the right frontal lobe. (C) Same view as picture A, but under higher magnification. The LFV has shrunk and turned bluish immediately after clipping at the fistula site. (D) Same view as picture B, but under higher magnification. The RFV has diminished and turned bluish just after clipping. The vein entry point into the frontal lobe is clearly shown.



**Fig. 6.** Control angiography, 14 months after surgery, still shows complete DAVF resolution. (Left) lateral projection of the right internal carotid artery. (Right) antero-posterior view of the left internal carotid artery.

damage. Bilateral exploration is laborious since bilateral C-shaped dura mater openings are necessary.

The *bilateral subfrontal approach* is the most common approach for ethmoidal DAVFs [13]. It exposes both anterior fossae equally, falx division is not always necessary, there is little risk of sagittal sinus injury, orbit content is spared, and it is more familiar for most neurosurgeons. However, frontal sinus might be opened and brain retraction should be greater to expose the anterior cranial fossa. This latter obstacle is solved drilling the inner table of the

frontal skull base, which permits a better access with less brain manipulation.

### 3.5. Summary

Prevalence of bilateral ethmoidal DAVFs may be underestimated. They should be suspected when complex venous drainage is observed in preoperative images. Intraoperative angiography could be a great help to detect these DAVFs. Bilateral exploration of the

anterior cranial fossa should be mandatory in those cases with high index of suspicion and whenever intraoperative angiography is not available, even if there is no suspicion of bilateral drainage on pre-operative MRA, CTA or angiography. Final treatment consists in occlusion of any abnormal draining vein coming from the cribriform plate into the brain. Dura mater coagulation at the fistula site is optional. Surgical management would be preferred over endovascular or radiosurgical approaches because of its higher curative rate with a lower rate of complications.

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