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ORIGINAL ARTICLE

PTA Stent of Dural Sinuses in Brain DAVF

A Report of 4 Cases

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Abstract

Background and Purpose Type I and IIa dural arteriovenous fistulas (DAVFs) have a low hemorrhagic risk, but are often the cause of debilitating tinnitus that requires treatment. While Onyx® and PHIL™ (Precipitating hydrophobic injectable liquid) transarterial embolization represent the first endovascular option, there are occasional cases where performing angioplasty and stenting of the affected sinus may lead to satisfactory results.

Material and Methods We retrospectively analyzed four consecutive cases of patients with DAVF-induced pulsatile tinnitus secondary to type I and II DAVFs who were treated with angioplasty and stenting of the sinus only. All the patients had clinical and radiological long-term follow-up.

Results We noticed a significant radiological and clinical improvement in all the cases. Of the patients two were completely cured at follow-up with eradication of the neurological symptoms as well as the fistula, one was retreated with Onyx® for a very small residual shunt despite having no more tinnitus, and one showed improvement in venous drainage (from type IIa+b to type I fistula) without resolution of the fistula.

Conclusion In cases of type I and II DAVFs associated with sinus stenosis, angioplasty and stenting alone seem to be safe and effective. This treatment probably compresses the venules within the sinus walls, promoting thrombosis of the shunts thus solving the underlying cause of the fistula.

Keywords Treatment of DAVF · Sinus stenting · PTA stent · Type I fistula

Introduction

The natural history of dural arteriovenous fistulas (DAVFs) is variable and principally dependent on their pattern of venous drainage. The Cognard classification system takes this pattern of venous drainage and outcome into account. Cognard type I and IIa lesions have a very low risk of hemorrhage because the venous drainage is anterograde and flows into a dural venous sinus [1–3]. Despite their benign nature, they often need to be treated because of the symptoms they cause and for the unlikely chance they can develop into malignant venous drainage. The aim of endovascular treatment is to occlude the shunt and maintain sinus patency, without compromising the venous drainage of the brain [9] which is mandatory if the sinus is functional. This can be achieved with Onyx® (Medtronic Inc. Miami, FL, USA) or PHIL™ (Precipitating Hydrophobic Injectable Liquid) (MicroVention Inc. Tustin, CA, USA) embolization with the help of temporary sinus balloon occlusion if needed; however, avoiding Onyx® leakage into the sinus and thus worsening the stenosis or occluding it completely might be challenging.

In this study, we propose an alternative approach to treatment of certain Cognard type I and IIa lesions with associated venous stenosis causing pulsatile tinnitus, venous angioplasty and percutaneous transluminal angioplasty stenting (PTA stent). Based on the pathophysiology of such le-

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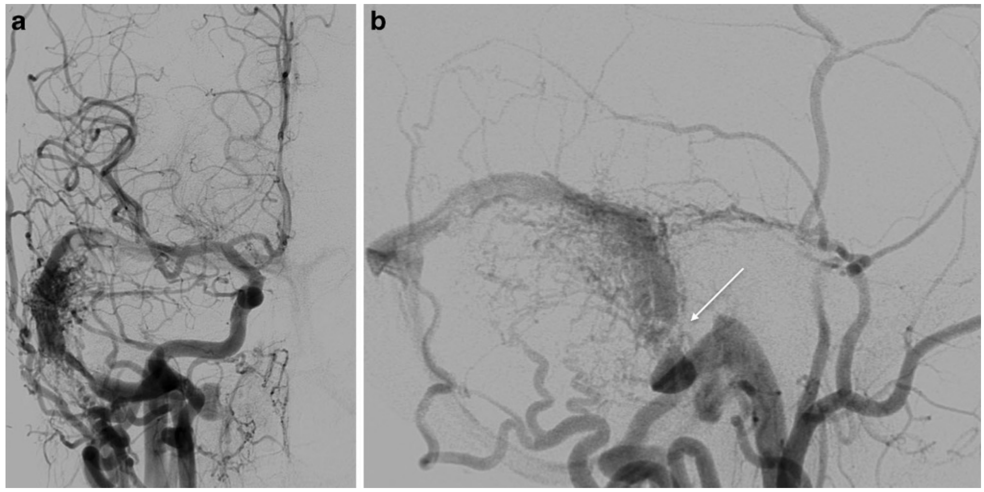

Fig. 1 DAVF of the right transverse sigmoid sinus; pretreatment anteroposterior (AP) (**a**) and lateral (**b**) projections during right common carotid injection. The *arrow* in (**b**) highlights the stenosis of the sinus

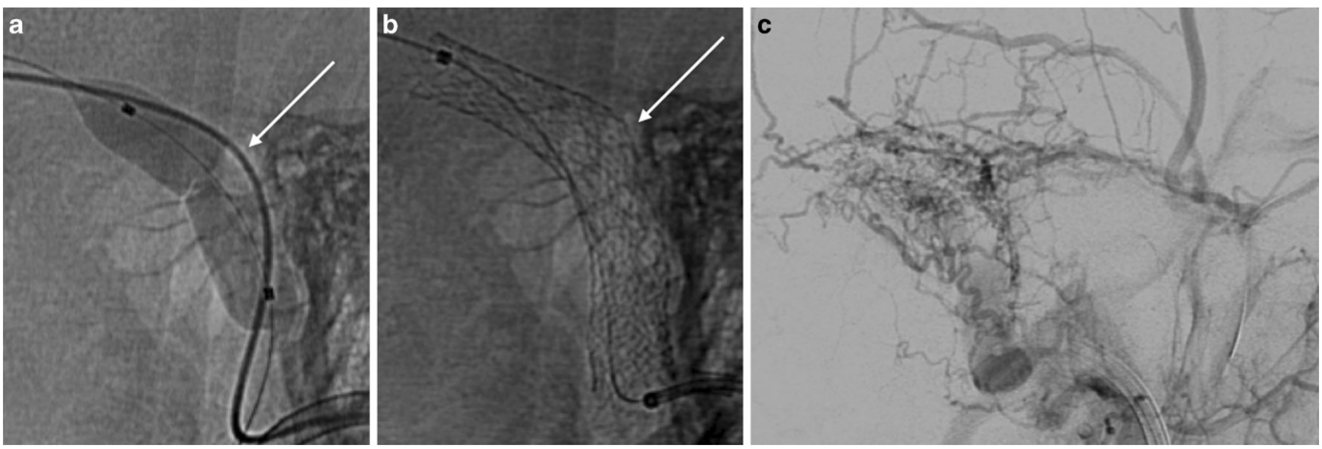

Fig. 2 **a** PTA of the stenosis: the *arrow* demonstrates the stenosis. **b** Deployment of the stent, the *arrow* points to where the stenosis was before being treated. **c** Contrast stasis on the final angiogram, sign of effective compression of the venules by the stent

sions, it is conceivable that such treatment could close the fistula by compressing the venules in the sinus wall which are the nidus of the fistula as well as relieve the venous stenosis which could be contributing to the pulsatile tinnitus.

Materials and Methods

This study was approved by our institutional review board (IRB). A total of four patients (3 male, 1 female; mean age 58 years) with type I and II DAVFs underwent endovascular treatment with angioplasty and stenting of the involved sinuses as sole treatment. All of them were admitted or referred to our neurovascular unit after computed tomography (CT) or magnetic resonance imaging (MRI) had shown the presence of the fistula. After a digital subtraction angiography (DSA) examination was carried out, our interventional neuroradiology, neurosurgical and neurological teams discussed the indications and the therapeutic strategies for each case. All the endovascular procedures were performed with the patient under general anesthesia and with systemic heparinization. With the arterial transfemoral approach, a 5F diagnostic catheter was inserted into the common carotid artery of the affected side for controls, while the whole procedure was carried out via an 8F femoral vein approach. An 8F guiding catheter was placed in the jugular bulb and a 7 × 40 mm or 8 × 40mm Precise Stent (Cordis Corporation, Miami, FL, USA) was tracked over a 14 Traxcess® (MicroVention Inc. Tustin, CA, USA) extra-support guidewire. A balloon angioplasty was performed after stent placement in all the cases.

All the patients were woken up in the angio-suite after undergoing a flat-panel CT and then hospitalized in the intensive care unit (ICU) for at least one night before being transferred to the neurosurgical ward. Systemic heparinization at anticoagulant doses was administrated in all of the patients for 7 days in addition to aspirin 160 mg for 3 months. One patient received oral anticoagulant therapy for 3 months and no aspirin.

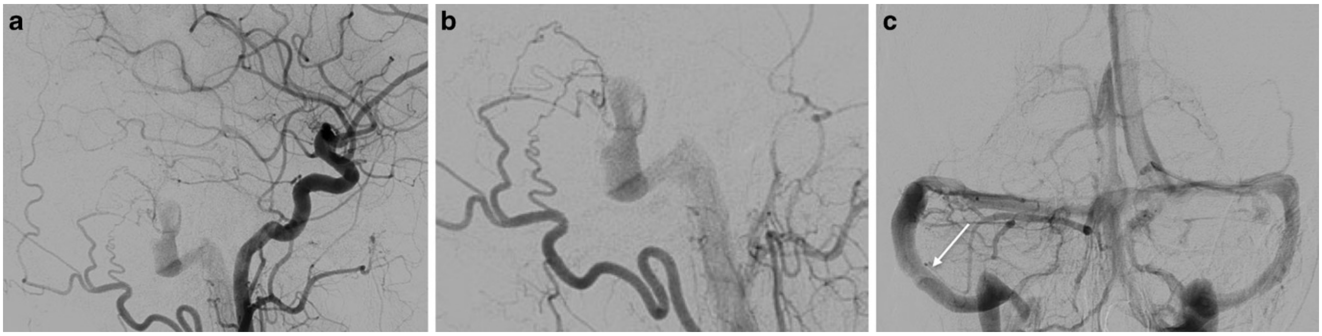

Fig. 3 At 3-month follow-up. Lateral (**a**) and lateral magnified (**b**) view showing a small DAVF remnant with faint opacification of the sinus on right common carotid injections. **c** Townes projection during right vertebral artery injection, the *arrow* shows that the stenosis is no longer present

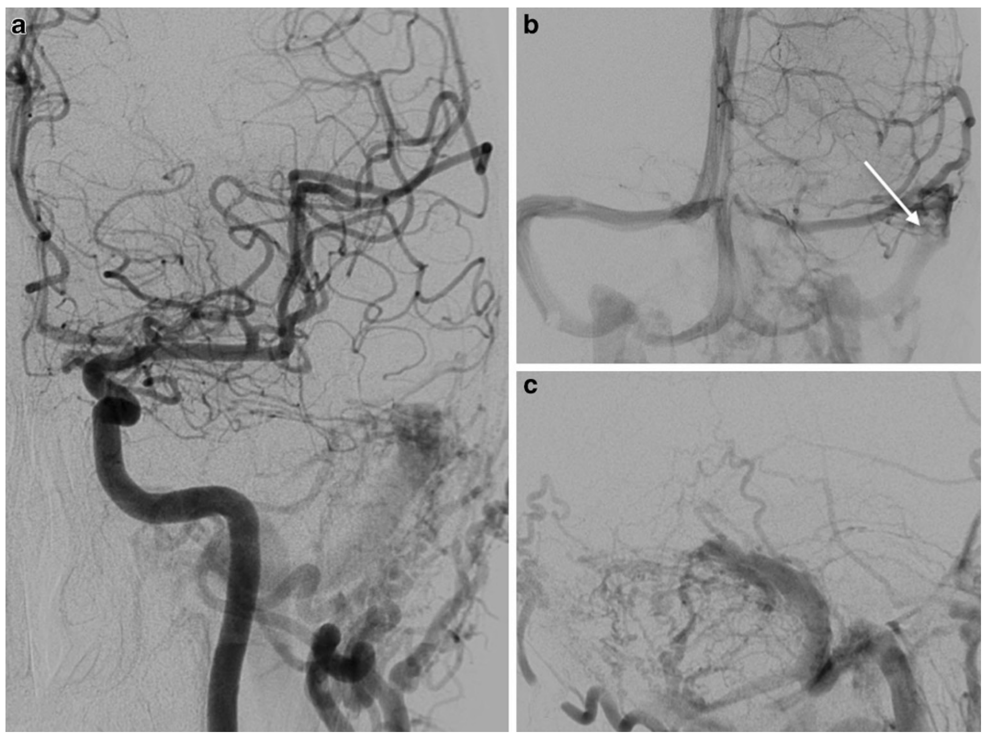

Fig. 4 DAVF of the left transverse sigmoid sinus; pretreatment anteroposterior (AP) (**a**), AP magnified (**b**) and lateral magnified (**c**) projections obtained during left common carotid artery injections. The *arrow* in **b** highlights the stenosis of the sinus

Results

In all the cases we noticed a significant improvement of the venous drainage with a reduction of the pathological shunts due to disappearance of many arterial feeders.

Case 1

A 45-year-old woman was referred to us due to right-sided pulsatile bruit. The MRA and DSA examinations revealed the presence of a type I DAVF involving the lateral and sigmoid sinuses with evidence of a significant stenosis (>70%) associated with compartmentalization of the right lateral sinus. After the PTA and stenting had resolved the stenosis, we noticed an improvement of venous drainage. At the end of the procedure, a reduction of the arterial feeders was clearly evident and the patient reported complete resolution of the bruit at discharge. The 3-month DSA control showed a dramatic reduction of the arteriovenous shunt. On the long-term MRA follow-up (5 years), no arteriovenous shunts were visible and the patient was angiographically cured (Fig. 1, 2, 3).

Case 2

A 66-year-old man was referred to us due to sudden and transitory onset of left temporoparietal headache associated with a pulsatile ipsilateral bruit. The patient underwent a brain MRI and a cerebral DSA that showed the presence of a type I fistula of the left lateral and sigmoidal sinuses

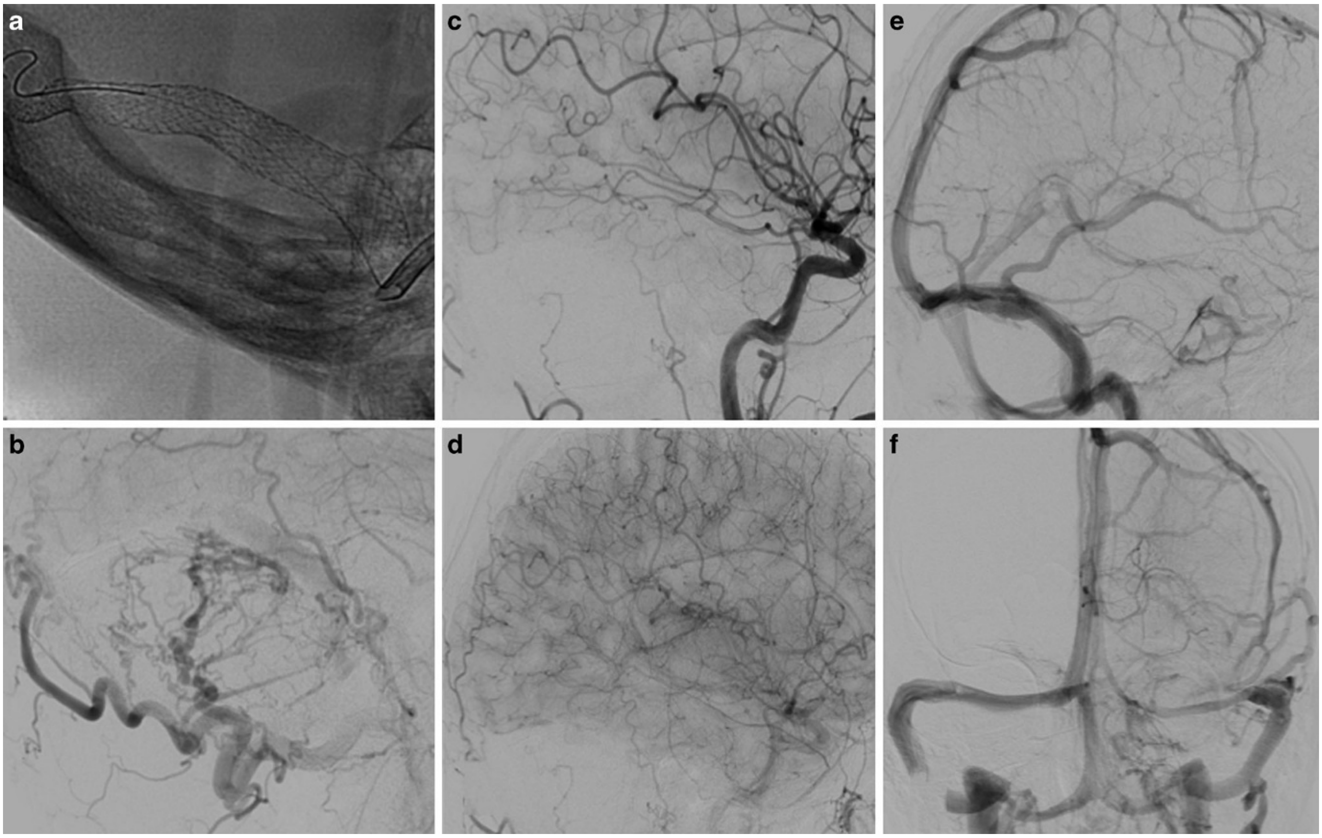

Fig. 5 Deployment of the stent (**a**) and immediately postprocedural angiogram showing arterial stasis on the left common carotid injection (**d**). The 3-month DSA, lateral (**b**, **c**, **e**) and Towne projections (**f**) during left common carotid injection showing disappearance of the DAVF

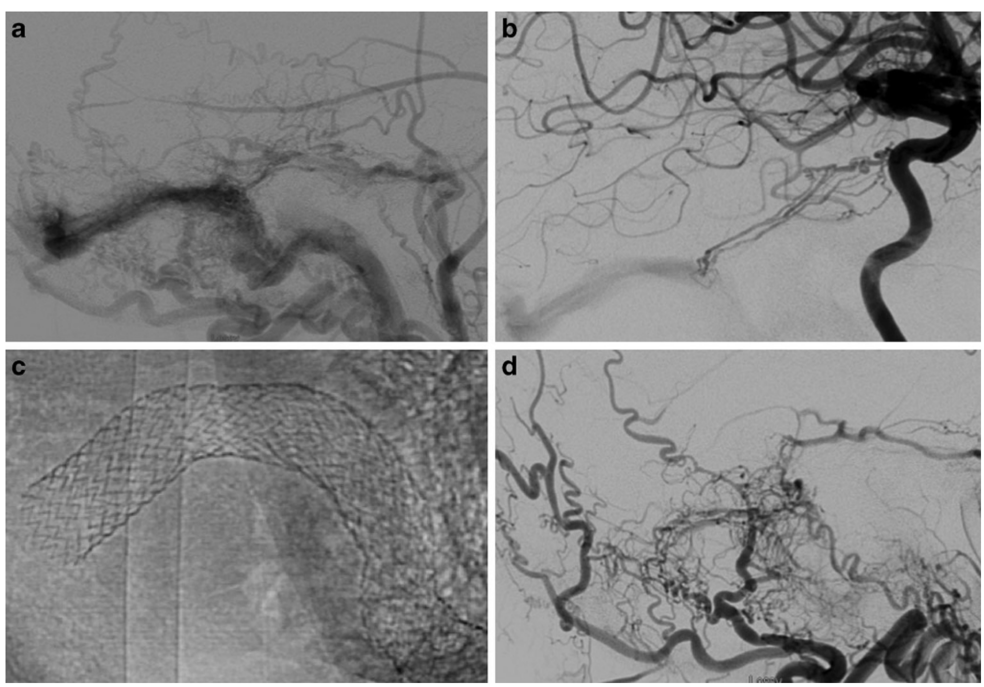

Fig. 6 Cognard IIa left lateral-sigmoid DAVF; pretreatment lateral angiogram of the left common carotid (**a**) and selective injection of the ipsilateral internal carotid (**b**) demonstrating tentorial supply to the fistula. **c** Stent deployment. **d** Post-procedural left external carotid injection demonstrating arterial stasis

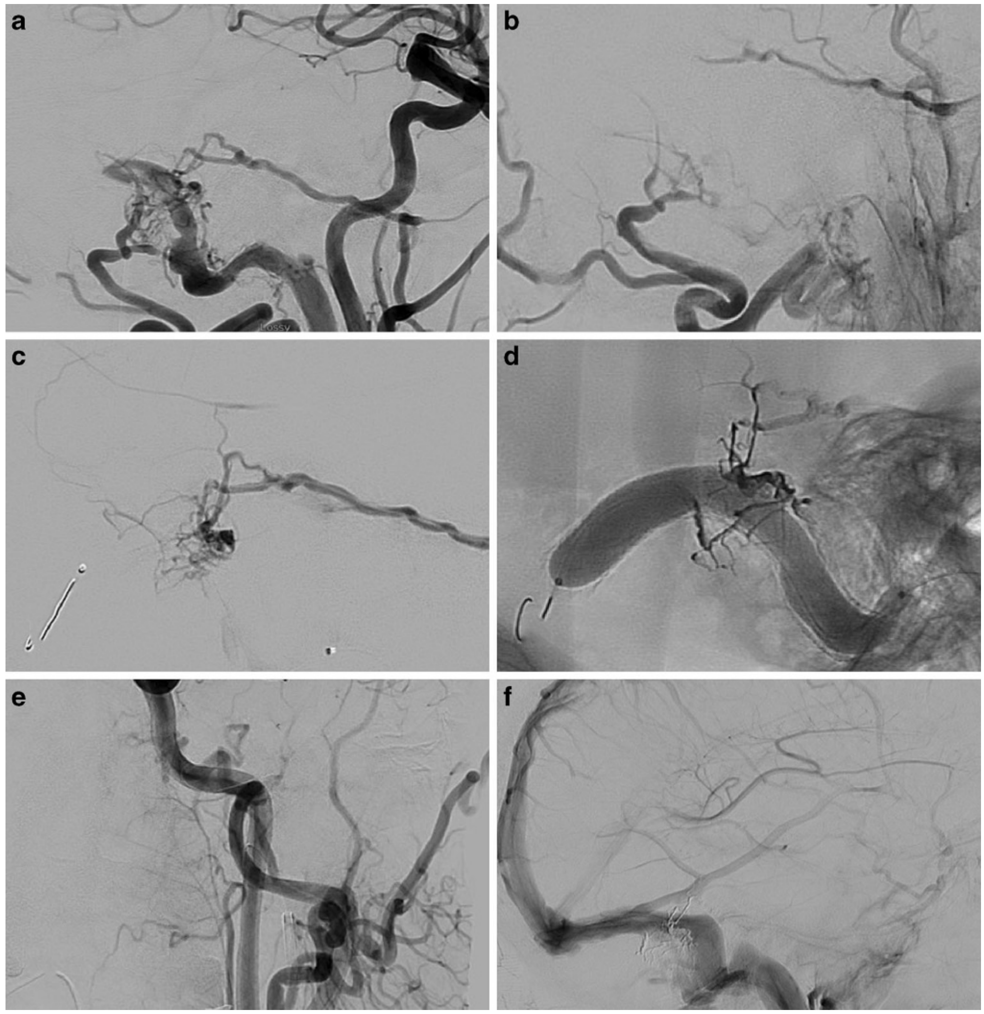

Fig. 7 **a**,**b** A 3-month DSA demonstrating significant reduction in number of the feeders. Selective microcatheterization of the middle meningeal artery (**c**) and injection of Onyx® while the sinus is temporarily occluded with a Copernic balloon (Balt Extrusion, Montmorency, France) (**d**). Note the small amount of Onyx® needed to occlude the fistula in (**d,e,f**) long-term follow-up, demonstrating the complete and stable occlusion of the fistula

and a high-grade stenosis (>70%) caused by a large arachnoid granulation within the transverse sinus, at the origin of the vein of Labbé. The sinus stenosis was treated by angioplasty and placement of two stents. After deploying two stents and performing angioplasty of the stenosis, we noted near complete occlusion of the shunts and sensible stagnation of contrast within the arteriolar network around the sinus. The patient had no more neurological symptoms at discharge. At the 3-month DSA, no shunts were visible. At the 2-year follow-up, no clinical symptoms or signs of recurrence on the MRA were present (Figs. 4 and 5).

Case 3

This 60-year-old man presented with left-sided pulsatile bruit. After an MRI was performed, the patient underwent a cerebral DSA that showed a type IIa DAVF extending to the left lateral and sigmoid sinuses fed by internal, external and posterior meningeal branches. The drainage was both anterograde towards the jugular vein and retrograde towards the contralateral lateral sinus. No reflux into the superior sagittal sinus or into the deep system or the cortical veins was present. After performing a PTA and deploying a stent at the level of the stenosis in one of the two channels of the transverse sinus, we noted significant stasis at the level of the arteriolar network. At discharge the patient reported complete disappearance of the bruit. At the 3-month MRA, there was still evidence of a fistula and for this reason a second treatment was scheduled. The fistula appeared to be significantly reduced in size, as there was no supply from the posterior circulation and a dramatic reduction of the feeders from the external and internal carotid arteries were demonstrated. The shunt was fed by a small branch from the middle meningeal artery. We injected 1 ml of Onyx® with temporary in-stent balloon occlusion of the sinus. On follow-up (6 months), the fistula was completely cured and the patient continued to be free of pulsatile tinnitus (Figs. 6 and 7).

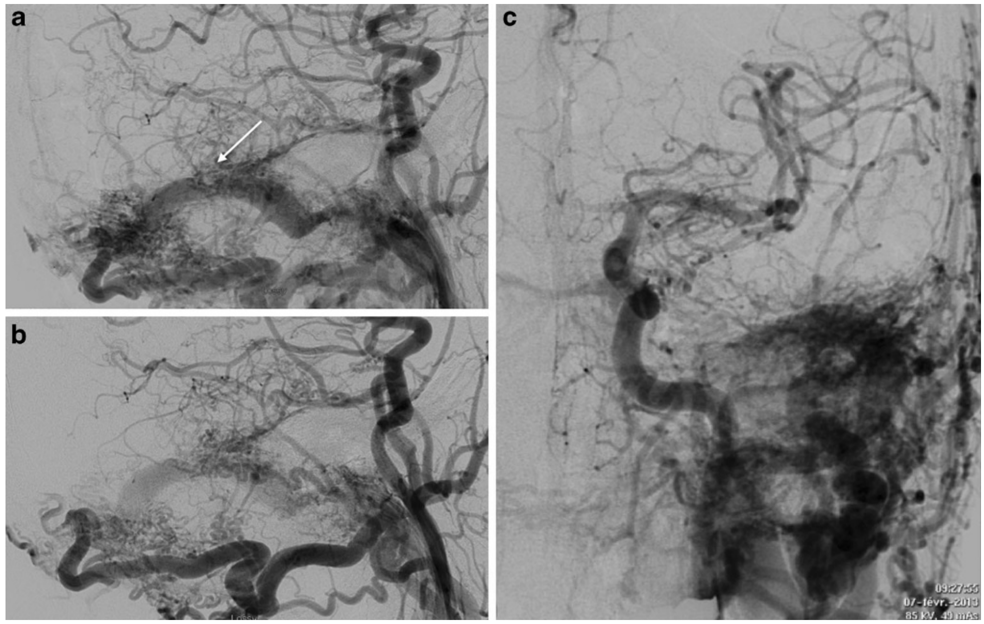

Fig. 8 Extensive DAVF involving the left transverse and sigmoid sinus in addition to the jugular bulb. **a**,**b** Lateral projections taken during left common carotid injections. The *arrow* in (**a**) highlights the presence of the stenosis. **c** Towne projection

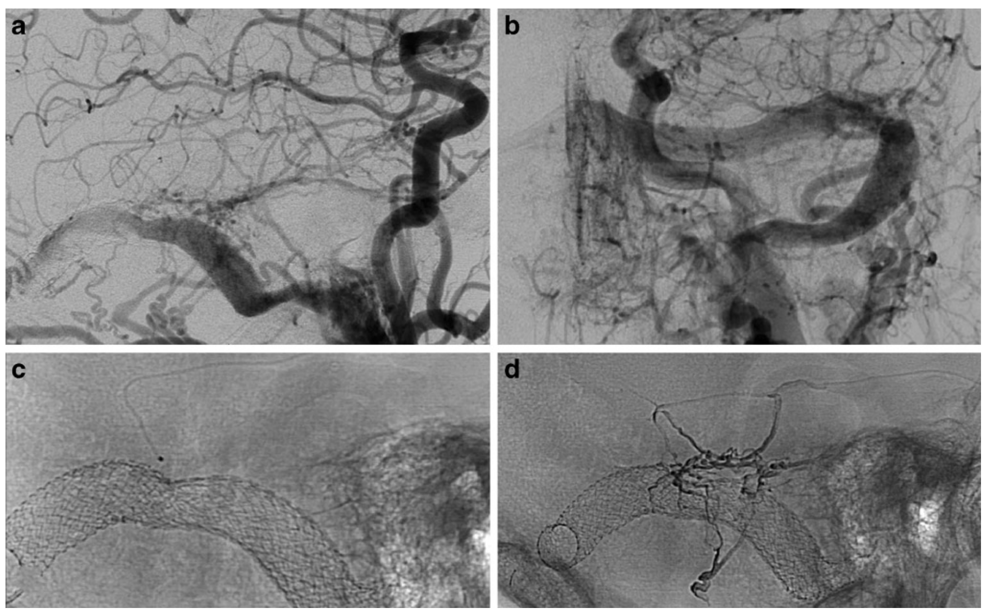

Fig. 9 Massive reduction of flow in the DAVF at the 3-month follow-up (**a**,**b**). Selective catheterization of the feeder from the middle meningeal artery (**c**) and final cast of Onyx® (**d**). Note the small quantity of Onyx® needed to occlude this compartment

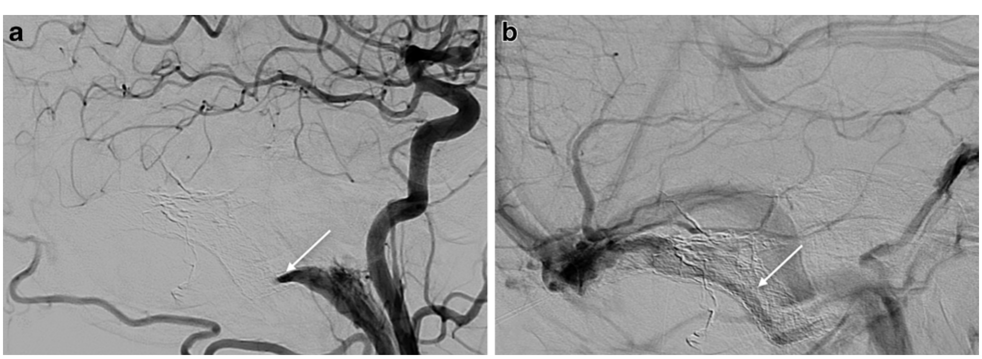

Fig. 10 **a** Stable occlusion of the previous compartment treated with PTA stenting and Onyx®, DAVF remnant at the jugular bulb where the sinus has not been covered. **b** Venous phase which demonstrates the patency of the sinus that now drains the parenchyma. The *arrows* in (**a**) highlight that the fistula switched into a type I and in (**b**) that the distal sinus is patent

Table 1 Summary of the series published in the literature [6–8]

Author	Number of cases	Cognard class		Angiographic result		Clinical result	
Levrier et al. [8]	10	I	2	Cured	4	No symptoms	7
		IIa	5				
		IIb	1	Shunt reduction	6	Symptoms improved	2
		IV	2				
						Unchanged	1
Takada et al. [7]	1	IIa+b	1	Cured	1	No symptoms	1
Liebig et al. [6]	4	Not specified	4	Cured	3	Not specified	4
				Shunt reduction	1		
Our series	4	I	2	Cured	2	No symptoms	3
		IIa	1	Shunt reduction	2	Symptoms improved	1
		IIa+b	1				

Case 4

This 61-year-old man was referred to our institution for left retroauricular pain associated with pulsatile bruit. A brain MRI and a DSA revealed the presence of a type IIa+b fistula that involved the posterior aspect of the jugular bulb and the whole left lateral and sigmoidal sinuses with significant impairment of left hemispheric drainage. The left transverse sinus was characterized by the presence of a significant stenosis (>70%) and was functionally excluded. We performed a PTA of the stenosis and then we implanted two stents that could cover all the affected sinuses with the exception of the jugular bulb where the shunts remained untreated. We noticed stagnation of contrast in the arterial feeders and disappearance of the cortical reflux. At the 3-month DSA, the portion of the fistula covered with stents appeared to be significantly changed because there was no more involvement of the posterior circulation feeders and the ones from the anterior circulation were definitely decreased in number; moreover, there was a reduction of venous congestion and the left transverse sinus, which could now partially drain the ipsilateral hemisphere; however, there was still residual fistula at the jugular bulb where the sinus was not covered by a stent. Despite transformation of the fistula into a type I with a very low hemorrhagic risk, the patient continued suffering from bruit and asked for a second treatment. We proceeded with Onyx® embolization via the middle meningeal artery and coiling of the sinus at the jugular bulb (Fig. 8, 9, 10).

Discussion

Our small series of four patients with extensive DAVFs and sinus stenosis treated with primary PTA and stenting of the involved sinuses demonstrated good results of this technique, which has already been reported in the literature ([6–8]; Table 1). Patients number 1 and 2 had resolution of clinical symptoms at discharge and were completely cured at the long-term follow-up (2 and 5 years). In the third case, there was progressive reduction of the shunts with almost complete occlusion on the 6-month DSA. Adjunctive Onyx® embolization resulted in a complete cure of the fistula. Finally, the last patient's fistula switched from type IIa+b to type I, meaning that there was no more cortical venous reflux and the potential hemorrhagic risk was improved. Unfortunately, the stents did not cover the entirety of the involved sinus and this was the reason for the incomplete resolution of the problem.

The rationale of deploying a stent across the stenosis is both to compress the sinus wall and to resolve the venous hypertension from the venous stenosis. It has already been reported that stenting a sinus leads to compression of the venules (30 μm) within the sinus wall, thus lowering the blood flow within the shunts resulting in subsequent shunt thrombosis[10, 11]. As demonstrated in a previous series and confirmed by our data, this is a slow process and it can takes months, but it can be effective[6–8]. The importance of this mechanism in curing the lesion is well highlighted by case number 4 where the sinus covered by the stent (which created a constant radial force against the walls over time) was cured and the portion of the sinus where angioplasty was only carried out at the jugular bulb remained unchanged. Moreover, when a stent is deployed across a venous stenosis, the venous hypertension is reduced or resolved. This is important because it is thought that venous hypertension plays an important role in DAVF development [12, 13]. Histopathological examinations have found an association of DAVFs with thickening venous walls, which are signs of pre-existing venous hypertension. It was postulated that sinus hypertension might force open pathological connections between arteries and veins within the dura, that lead to dilatation of the venules and DAVF occurrence. In addition, increased intraluminal pressure in the vessels stimulates angiogenesis [14] and the new vessels formed around the sinus can connect in an abnormal manner. Another factor which can influence DAVF formation is that the increase in venous pressure can reduce the perfusion pressure, resulting in hypoxia which stimulates angiogenesis [15] through angiogenic factors. New abnormal vessels can form aberrant connections leading to AVF formation [13–16]. A vicious cycle can then be created by the recruitment of arterial blood from dural branches caused by thickening and steno-occlusive disease of the sinus itself [10]. This theory does not reject the importance of venous thrombosis in the pathogenesis of DAVFs [8, 17–19], but in some cases sinus thrombosis can occur during the development of DAVFs and not be the trigger [10, 11].

The rationale and the results published in previous small series and confirmed by our results, do not justify extensive use of this technique for all kind of fistulas. In fact, as previously stated, PTA stenting needs time to be effective and any kind of aggressive fistula with significant hemorrhagic risk represents a contraindication because an immediate cure has to be achieved in these cases. On the contrary, for Cognard type I and IIa lesions, the goal of the treatment is only to alleviate the symptoms (tinnitus 61%, orbital symptoms 35%, headache 32%) [1], which are often intolerable for the patient. A critical point in treating these kinds of fistula is preserving the normal sinus anatomy: this is mandatory in cases of a functional sinus, but it is recommended in any case in order to preserve cortical vein origins.

In our opinion Onyx® embolization with sinus protection with a dedicated balloon (Copernic) generally remains the first option for treating these lesions [4, 5, 9], but obtaining satisfactory penetration of embolic material into the shunts while preserving the patency of the sinus can be challenging in cases of extensive involvement of the sinus. In these peculiar cases, PTA stenting (when a stenosis is visible) can be a valid alternative, considering also that DAVFs with multiple feeders extending over the whole sinus need several vials of Onyx® to be treated and sometimes long, expensive procedures are required. The advantages of PTA stenting include cost and time saving while at the same time not precluding a second treatment with transarterial Onyx® injection in case of residual feeders at long-term follow-up (as demonstrated in cases 3 and 4). This procedure is also relatively safe as demonstrated by the many series recently published on idiopathic intracranial hypertension (IIH) where excellent results have been reported [20, 21]. The IIH is a different disease, but the underlying cause seems to be similar to DAVF, i.e. venous hypertension. Restenosis rates for dural venous sinus stenting are very low. In one large series, Ahmed et al. [20] reported a very low in-stent stenosis rate of showing that the radial force of the stents currently on the market is enough to guarantee a long-lasting patency of the sinus.

Regarding antiplatelet and anticoagulation strategies, all our patients received full heparinization for 7 days. In three cases they were given aspirin 160 mg for 3 months and in one case oral anticoagulant for 3 months but no aspirin. There are no trials concerning the best medical therapy for patients with venous stenting, but the major series in the literature suggest dual antiplatelet therapy [20, 21]. In our series, after full anticoagulation for 1 week, monoantiplatelet therapy seemed to be effective to guarantee stent patency. Also, the patient who received oral anticoagulation did not experience stent occlusion.

Our study has several limitations; it is a small series of patients and is retrospective. Follow-up is limited. In addition, there was not a standardized imaging and clinical follow-up protocol. Further studies are needed to confirm our results.

Conclusion

In conclusion, PTA and stenting of the sinuses in the case of type I and II dural arteriovenous fistulas associated with a significant stenosis seem to be a safe and effective alternative to transarterial Onyx® embolization. Considering that this approach does not lead to the immediate cure of the patient, more aggressive types of fistula (type > IIa) are probably not amenable to this treatment strategy. Further studies are needed to confirm our results.

Conflict of Interests L. Renieri, C. Michelozzi, W. Brinjikji, J. Darcout, A. Guenego, I. Vukasinovic, P. Tall, F. Bonneville, A-C. Januel, C. Cognard declare they have no competing interests.

Ethical standards All procedures described in this article were carried out in accordance with national law and the Helsinki Declaration of 1964 (in its current revised form). Informed consent was obtained from all patients included in the study.

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