

## Clinical Article

# Brain stem venous congestion due to dural arteriovenous fistulas of the cavernous sinus

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## Summary

**Background.** Venous congestion of the brain stem due to dural arteriovenous fistulas (DAVFs) in the cavernous sinus is rare and presents therapeutic challenges. To assess the prognosis of patients with symptomatic DAVFs and brain stem dysfunction, we evaluated the degree of venous ischemia by examining pre- and post-treatment magnetic resonance images (MRI) in 2 patients presenting with venous congestion of the brain stem.

**Methods.** A 56-year-old woman with left hemiparesis and a 70-year-old woman with gait disturbance attributable to right cavernous sinus DAVFs were referred to our hospital. In both cases, T2-weighted magnetic resonance imaging (MRI) disclosed a hyperintensity lesion in the brainstem due to venous congestion.

**Findings.** Both patients underwent open surgery for direct embolization of the cavernous sinus because there were no approach routes for transvenous embolization. The patient whose pretreatment MRI demonstrated Gd enhancement continued to manifest neurological deficits and persistence of the abnormal hyperintensity on post-treatment T2-weighted MRI. In the other patient whose pretreatment MRI showed no Gd enhancement, treatment produced a complete response of her neurological deficit and disappearance of the abnormal hyperintensity area.

**Conclusions.** We tentatively conclude that lesions corresponding to hyperintensity areas on non-Gd-enhanced, T2-weighted MRI may reflect a reversible condition whereas lesions identified as hyperintense areas on GD-enhanced T2-weighted MRI may be indicative of irreversibility.

**Keywords:** Dural arteriovenous fistulas; MRI; retrograde leptomeningeal venous drainage; venous congestion.

## Introduction

Dural arteriovenous fistulas (DAVFs) of the cavernous sinus are abnormal communications between the dural branches of the internal or external carotid arteries. They can lead to chemosis, proptosis, and bruit and, rarely,

to infarction or haemorrhage [1, 3, 6, 9]. Intracranial DAVFs with cortical or deep venous drainage can produce haemorrhages, seizures, and neurological deficit [7]. Their pathophysiological manifestations are attributable to venous congestion which in turn may be exacerbated by veno-occlusive disease. We report 2 patients with DAVFs who manifested rapidly progressing symptoms related to brainstem dysfunction such as dysarthria and ataxic gait. Both underwent open surgery for direct embolization of the cavernous sinus because there



Fig. 1. Case 1: Right internal carotid artery angiogram (lateral view) demonstrating that the carotid-cavernous fistula drains only into the petrosal vein (arrow) and empties retrogradely into the cerebellar vein (arrow head)

were no approach routes for transvenous embolization. To determine the prognosis of patients with symptomatic DAVFs we evaluated the degree of venous ischemia on pre- and post-treatment magnetic resonance images (MRI).

## Case reports

### Case 1

This 56-year-old woman initially experienced redness in her right eye, proptosis, double vision, and a progressive decrease in visual acuity.

Two weeks before admission she noted mild left-sided hemiparesis. Cerebral angiography revealed a right cavernous sinus DAVFs fed by branches of the intercavernous and external carotid arteries. It drained only through the cerebellar veins, including a lateral mesencephalic vein connected to the basal vein via the right petrosal vein (Fig. 1). Axial T2-weighted MRI showed a hyperintense brain stem lesion (Fig. 2a). On contrast-enhanced T1-weighted MRI there was moderate enhancement in the brain stem (Fig. 2b). A percutaneous transvenous approach was attempted, however, the catheter failed to reach the cavernous sinus. Therefore we performed a suboccipital craniectomy and introduced a microcatheter directly into the hemispheric branch of the right petrosal vein. The cavernous sinus was packed with Guglielmi detachable coils.

Her postoperative course was uneventful, however, follow-up revealed only partial resolution of her symptoms and signs consisting of slight

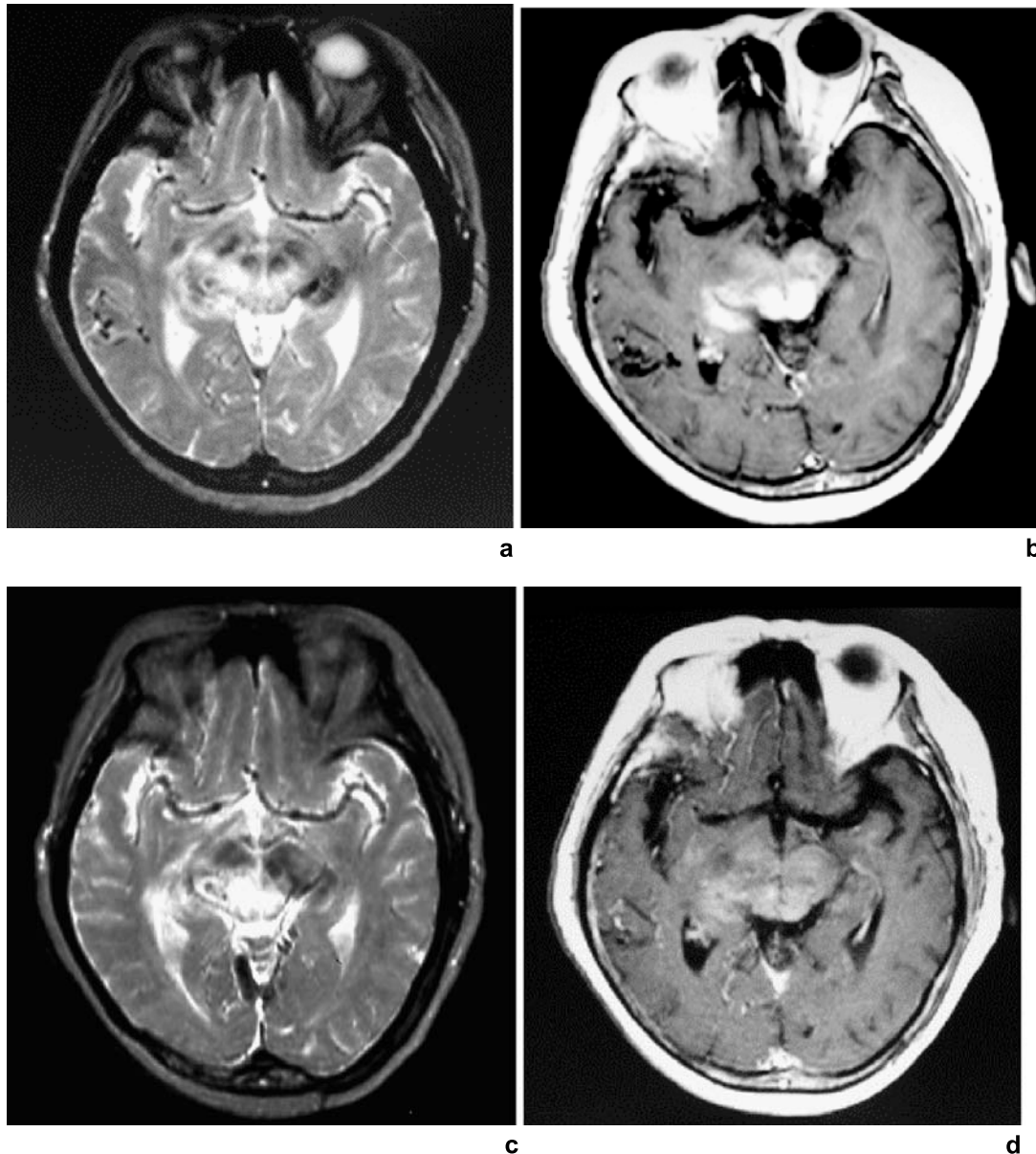


Fig. 2. Case 1: (a) Axial T2-weighted MRI showing diffuse hyperintensity of the brain stem. (b) Contrast-enhanced axial T1-weighted MRI showing enhancement in the brain stem. (c) Axial T2-weighted image obtained approximately 1 month after surgery shows residual hyperintensity in the brain stem. (d) Contrast-enhanced axial T1-weighted MRI obtained approximately 1 month after surgery shows residual enhancement in the brain stem

improvement of her left-sided hemiparesis. Although on cerebral angiograms obtained immediately after surgery the DAVFs had disappeared, T2-weighted MRI performed 1 month after surgery showed a residual hyperintensity lesion in the brain stem (Fig. 2c). On T1-weighted MRI there was persistent, albeit slightly decreased enhancement in the brain stem (Fig. 2d).

#### Case 2

This 70-year-old woman initially experienced double vision, right-sided chemosis, and exophthalmos. She reported to our hospital with a 2-month history of gait disturbance and mild truncal ataxia. Cerebral angiography showed DAVFs of the right cavernous sinus supplied by the dural branches of the internal and external carotid arteries bilaterally. In the venous phase the petrosal sinuses were not visualized. The fistula drained primarily into the deep Sylvian vein via the right sphenoparietal sinus. The draining route continued to the pontomesencephalic veins (Fig. 3). T2-weighted MRI disclosed a hyperintensity area in the midbrain (Fig. 4a). On contrast-enhanced T1-weighted MRI no enhancement was recognized (Fig. 4b). As the percutaneous transvenous approach failed because the catheter could not be inserted into the cavernous sinus, she underwent direct surgery. The right cavernous sinus was packed with sponges via the orbit-zygomatic approach.

Her postoperative course was uneventful and her symptoms disappeared completely. Postoperative angiograms revealed no evidence of fistula recurrence. T2-weighted MRI performed approximately 1 month post-treatment, revealed no abnormal hyperintensity area (Fig. 4c). On T1-weighted MRI there was no persistent enhancement in the brain stem (Fig. 4d).

#### Discussion

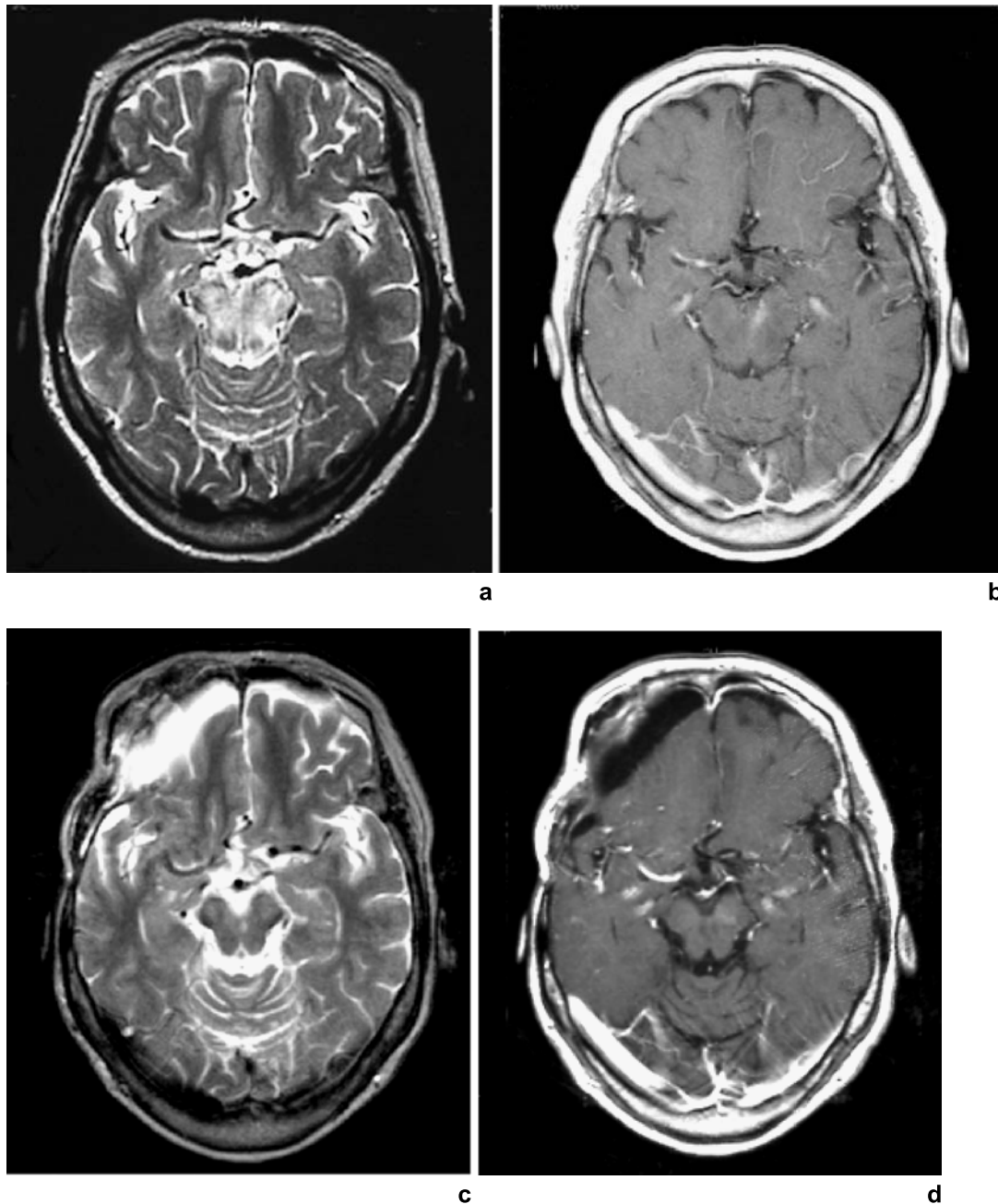
Patients with DAVFs and cerebral ischemia and/or cerebral haemorrhage reportedly manifested regional cerebral blood flow (rCBF) abnormalities due to venous drainage disturbance [1, 5, 10, 14]. The angiographic characteristics of DAVFs with disturbed rCBF include retrograde leptomeningeal venous drainage (RLVD)



Fig. 3. Case 2: Right external carotid arteriogram (lateral projection) showing DAVFs of the right cavernous sinus with venous drainage into the right deep Sylvian vein (arrow). The draining veins continue to the pontomesencephalic veins (arrow head)

[1, 10]. DAVFs with RLVD tended to be associated with variceal or aneurysmal venous dilatation [2, 4] and some patients developed venous hypertension resulting in neurological deficits due to haemorrhage, brain edema, and/or ischemia [2, 18]. Willinsky *et al.* [20, 21] suggested that in patients with DAVFs, the presence of a pseudophlebitic pattern (PPP) was indicative of venous congestion due to tortuous, engorged pial veins and that this condition may produce ischemia in the venous circulation of the brain. They offered this phenomenon, rather than leptomeningeal venous drainage, as a better prognostic indicator in patients with intracranial DAVFs.

While signal abnormalities on MRI are useful for the identification of DAVFs [13, 21], the degree of rCBF disturbance in the presence of DAVFs with RLVD remains to be elucidated. Willinsky *et al.* [21] reported that in DAVFs with cortical venous drainage, T2-weighted MRI identified areas of hyperintensity in the venous territory; they proposed this as direct evidence for venous hypertension and congestive encephalopathy. We already reported that there is a correlation between the pattern of venous drainage and MRI findings in DAVFs with RLVD [11, 12]. Patients with an accessory route to another sinus developed haemorrhagic episodes while those without manifest venous congestion, reflected by a tendency to exhibit hyperintense areas on T2-weighted MRI. According to Kawaguchi *et al.* [13], MRI findings are useful for determining the degree of venous ischemia and abnormal hyperintensity on T2-weighted MRI and Gd-DTPA contrast enhancement are suggestive of venous ischemia. In the course of observation, their patients with Gd-DTPA enhancement on T2-weighted MRI tended to develop haemorrhage in the enhanced areas. Similar findings were reported in patients with spinal DAVFs [8, 15, 16]. The diffuse enhancement of the cord is thought to be attributable to venous engorgement and secondary ischemia since infarcted regions in the spinal cord are known to contrast-enhance. Spinal cord enhancement is thought to reflect ischemic changes. Venous engorgement is usually longstanding and the healing process seems to be slow. Contrast enhancement of the cord may gradually decrease if venous engorgement is relieved by embolization. This suggests the presence of irreversible ischemic changes in the spinal cord. Others [17, 19] reported pontine venous congestion in 4 patients with DAVFs with convergent venous drainage into the basal vein of Rosenthal or the cortical veins of the posterior fossa. All of these patients manifested hyperintensity areas in their pontine lesions on T2-weighted MRI and Gd



**Fig. 4.** Case 2: (a) Axial T2-weighted MRI showing diffuse hyperintensity of the midbrain. (b) Contrast-enhanced axial T1-weighted MRI showing no enhancement of the midbrain. (c) Axial T2-weighted image obtained approximately 1 month after surgery shows disappearance of the midbrain lesion. (d) Contrast-enhanced axial T1-weighted MRI obtained approximately 1 month after surgery showing no enhancement in the brain stem

enhancement. In 2 of the 4 patients the hyperintense area on T2-weighted MRI and the neurological symptoms persisted post-treatment.

Our patient with preoperative Gd enhancement on MRI experienced only partial resolution of her neurological deficits; the abnormal hyperintensity continued to be present on postoperative T2-weighted MRI. Her symptoms worsened over time and MRI study was preceded

by a 2-month history of gait disturbance and mild truncal ataxia. On the other hand, in the patient without preoperative Gd enhancement, treatment produced a complete response and post-operative MRI disclosed no hyperintensity. MRI study was performed 2 weeks after the onset of mild left-sided hemiparesis.

Based on these and findings reported by others, we posit that among lesions that correspond to hyperintense

areas on pre-treatment T2-weighted MRI, those without Gd enhancement are reversible, while those with enhancement are not. We propose that the presence or absence of Gd enhancement may be useful for assessing venous ischemia, time-course changes, the degree of emergency and urgency of treatment, and the effectiveness of it. We suggest that Gd enhancement on pre-treatment T2-weighted MRI and the duration of symptom exacerbation are important prognostic indicators in patients with DAVFs.

Although further investigations are necessary for a better understanding of venous ischemia in patients with DAVFs, those whose DAVFs with hyperintensity areas on MRI are treated before the manifestation of Gd enhancement may achieve a good outcome.

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## Comments

The authors report on 2 patients with venous congestion and abnormal signals on MR of brain stem/midbrain due to dural arteriovenous fistulae. The key message of this case-report is the possible prognostic value of enhancement on MR indicative of reversibility of the lesion and/or clinical symptoms and signs. Although described earlier, this could be of clinical interest (even though there are only 2 cases).

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The cases however are well analysed as far as MRI abnormal signals and post operative changes are concerned. The associated thrombosis of all the cavernous sinus outlets is remarkable. Analysis of the drainage of the hemispheric structure on the same side would probably be very interesting to look at. Most of the time such lesions with complex thrombosis of multiple outlets do not produce the hyperemia and ischemia at the level of the brain stem but rather to the spinal cord. Several reports have shown intra cranial dural shunts of the cavernous sinus and adjacent tent to produce cervical myelopathy by congestive reflux into the petrous vein and the posterior fossa superficial venous network down to the spinal cord venous plexus.

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In the current issue of *Acta Neurochirurgica*, Dr Kai and al report 2 cases of brain stem congestion due to dural arteriovenous fistulas of the cavernous sinus (CCF). These 2 well documented cases clearly demonstrate a retrograde filling of subarachnoid veins responsible for venous

congestion of the brain stem. A large cohort of publications have pointed-out the close relationships between the pattern of subarachnoid venous drainage of a dural fistula and the incidence of haemorrhage and venous congestion. Nevertheless, descriptions of brain stem congestions remained few, contrasting with the relatively high frequency of dural carotid cavernous fistulas (12% approximately among all intracranial dural AVF). In a personal work to be published in the next issues of the Journal of Neuroradiology, we describe a similar case of a venous congestion of the brain stem and of a hemispheric territory related to the basal vein. In this paper we review 7 additional cases [1] including 2 direct CCF [2] and 5 dural CCF [3–5]. The current article and the review of previously published cases suggest two points for us:

The first one involves the thrombotic complications of this disease. In cases of brain stem oedema associated with CCF, a retrograde filling of subarachnoid veins may be visible or not. In our review, only 4 cases harboured continuity between the cavernous sinus and the subarachnoid veins (through the superior petrosal sinus and the petrosal vein). The remaining 4 patients lacked this angiographic pattern. In these cases, suggesting occurrence of a thrombosis, the brain stem lesions were supposed to pursue their own evolution, unrelated to the CCF itself and its proper treatment. In the case reported by Gregoire, clinical worsening and brain stem MRI signal abnormalities followed embolization of the fistula [5]. In our opinion, as far as the subarachnoid drainage of the fistula is not visible on the angiogram and when ophthalmological complications are not feared, one can reasonably question an invasive treatment aiming to occlude the fistula itself. Conversely anticoagulation may then be discussed, since, in the absence of haemorrhage, the benefit of this treatment is usually accepted in intracranial venous thrombosis. The thrombosis of arterialized veins may be one of the major complications and may explain sequelae despite a spontaneous or therapeutic cure of the fistula.

The authors have closely linked together the contrast enhancement of the brain stem with a worse prognosis. In our review of the literature, 3 cases demonstrating MR contrast enhancement were available for clinical follow-up data. Two patients improved at 4 and 10 months respec-

tively, contrasting with our case were a mild dystonia remained 4 years later. We can only suggest that one month follow-up is a very short time for assessing such a prognostic implication owing to the small number of cases.

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