Acta Neurochir (2010) 152:1745–1753
DOI 10.1007/s00701-010-0722-6

CASE REPORT

Tentorial dural fistula with giant venous ampulae treated with embolisation and surgery. A case report

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Received: 28 February 2010 / Accepted: 14 June 2010 / Published online: 16 July 2010

Abstract Tentorial dural arteriovenous fistulas are rare and complex lesions in deep locations with unusual vascular anatomy and critical surrounding neuroanatomy. A rare case presenting a complex fistula with a giant venous draining ampulae, causing headaches and visual troubles is presented. We describe the case of a 52-year-old woman admitted in our department for headaches and visual troubles. Magnetic resonance imaging and cerebral angiography showed a tentorial dural arteriovenous fistula draining in a giant tentorial venous ampulae and leptomeningeal veins. The patient was embolised via an arterial route with a good clinical and radiological result. However, 4 days later she presented a sudden change of her clinical status with coma, left hemiparesis and a right midriasis. The *cerebral computed tomography* scan showed a huge occipital haemorrhagic mass and a severe cerebral oedema. An emergent surgical procedure was decided realising evacuation of the occipital haematoma and a complete resection of the giant venous ampoule. The neck of the ampulae was sutured and clipped at its dural entrance. Postoperatively a new embolisation was realised because of persistent of a small dural fistulae *with occipital leptomeningeal drainage*. The patient recovered rapidly with only a residual hemianopsy. Treatment of dural AV malformation represent a serious challenge. Our report describes an unusual case of a tentorial dural complex fistula treated by an endovascular procedure with secondary clinical aggravation that needed emergent surgical therapy. Even in a case for good immediate radiological result after endovascular procedure, dural arteriovenous fistulas with giant venous ampulae and important venous engorgement, need closed follow-up, because of the possibility of aggravation secondary to venous thrombosis and haemorrhage. Treatment and patophysiology of the aggravation mechanism are discussed.

Keywords Tentorial dural fistula · Dural arteriovenous malformation · Transarterial embolisation · Giant venous ampulae

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Introduction

Intracranial dural arteriovenous fistulas (DAVF) account for 10% to 15% of all brain arteriovenous malformations (AVMs) and consist of single or multiple fistulas whose niduses lie entirely within the dura [1, 58].

Tentorial dural arteriovenous fistulas (TDAVF) are a subgroup of dural fistulas, compromising about 4–8% of all dural arteriovenous fistulae [1, 5, 13, 38, 58]. They represent rare and complex lesions in deep locations with unusual vascular anatomy and critical surrounding neuroanatomy [35].

Tentorial DAVFs are located in the tentorial dura mater and are fed primarily from branches of the meningohypophyseal trunk, middle meningeal artery and occipital artery.

Venous drainage varies greatly and depends on the tentorial location of the fistula. They typically drain into deep cortical and leptomeningeal veins [5]. Fistulas with leptomeningeal venous drainage, variceal or aneurysmal venous dilations, and galenic often present an aggressive behaviour with haemorrhage or progressive neurological deficit [1].

Case report

A 52-year-old right handed woman was admitted to our department for progressive diffuse headaches, visual troubles (left homonym hemianopsy) and small difficulties using the left arm, lasting from 1 month. The patient complained also for a left eye pain. Neurological examination revealed a left homonym hemianopsy and mild left brachial monoparesis. She had no past medical history.

The cerebral computed tomography (CT) showed an important hyperdense occipital lesion. The lesion was round and homogenous, 6 cm diameter (Fig. 1). Magnetic resonance imaging (MRI) revealed an important vascular occipital mass (Fig. 2) with flow void on fluid-attenuated inversion recovery sequence (Fig. 3) localised in the right occipital lobe, on the tentorium.

Cerebral angiography showed a complex tentorial arteriovenous fistula with a giant venous varix (type IIb in Borden classification, type IV according Cognard's classification). There were feeding arteries coming from both occipital arteries, the right and left middle meningeal arteries, the right tentorial branch of the meningohypophyseal trunk and the meningeal branch of the right ophthalmic artery. Injection of the vertebral arteries showed also a vascularisation of the fistulae via the posterior meningeal branch of the right vertebral artery and a small contribution from meningeal branch of the left vertebral artery. The venous drainage was thrown a right giant tentorial venous ampulae which drained in the right lateral sinus via a stenosed tentorial sinus, and also via occipital leptomeningeal veins which drained in the superior sagittal sinus (Fig. 4).

Because of the complex anatomy of the fistula, an endovascular procedure via a transarterial route was decided initially. Firstly, it was realised as a selective catheterization of the superior branch of the right middle meningeal artery. During the embolisation of these branches, a hyperdrainage in the lateral sinus with migration of the Onyx (Micro Therapeutics Inc., Irvine,

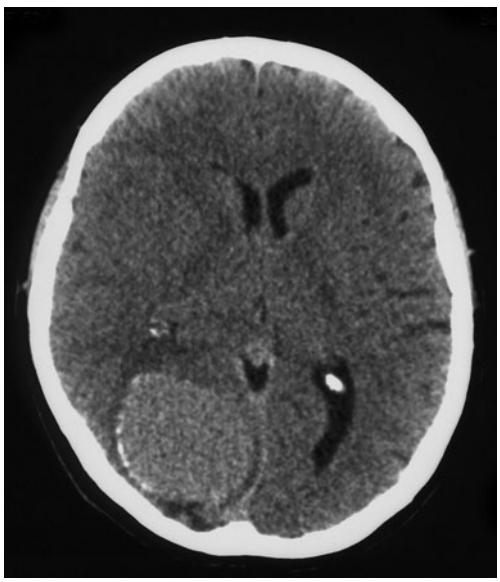

Fig. 1 Initial computed tomography showing an important round hyperdense occipital mass, 6 cm diameter

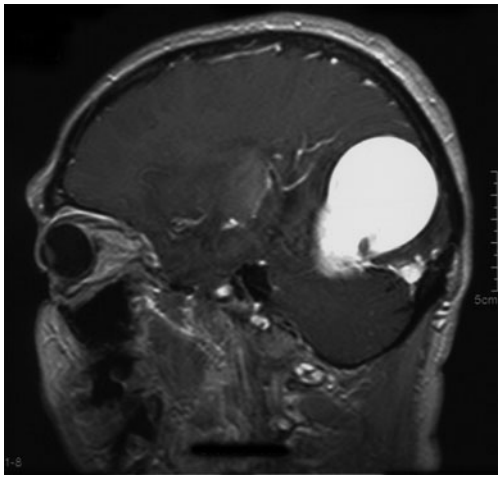

Fig. 2 T1 Gadolinium ponderate sagital magnetic resonance imaging sequence on admission showing an important occipital vascular mass

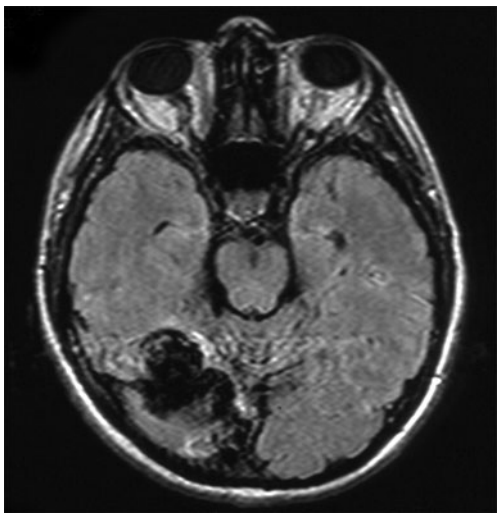

Fig. 3 Axial brain magnetic resonance imaging fluid-attenuated inversion recovery sequence showing signs of flow void in right occipital area

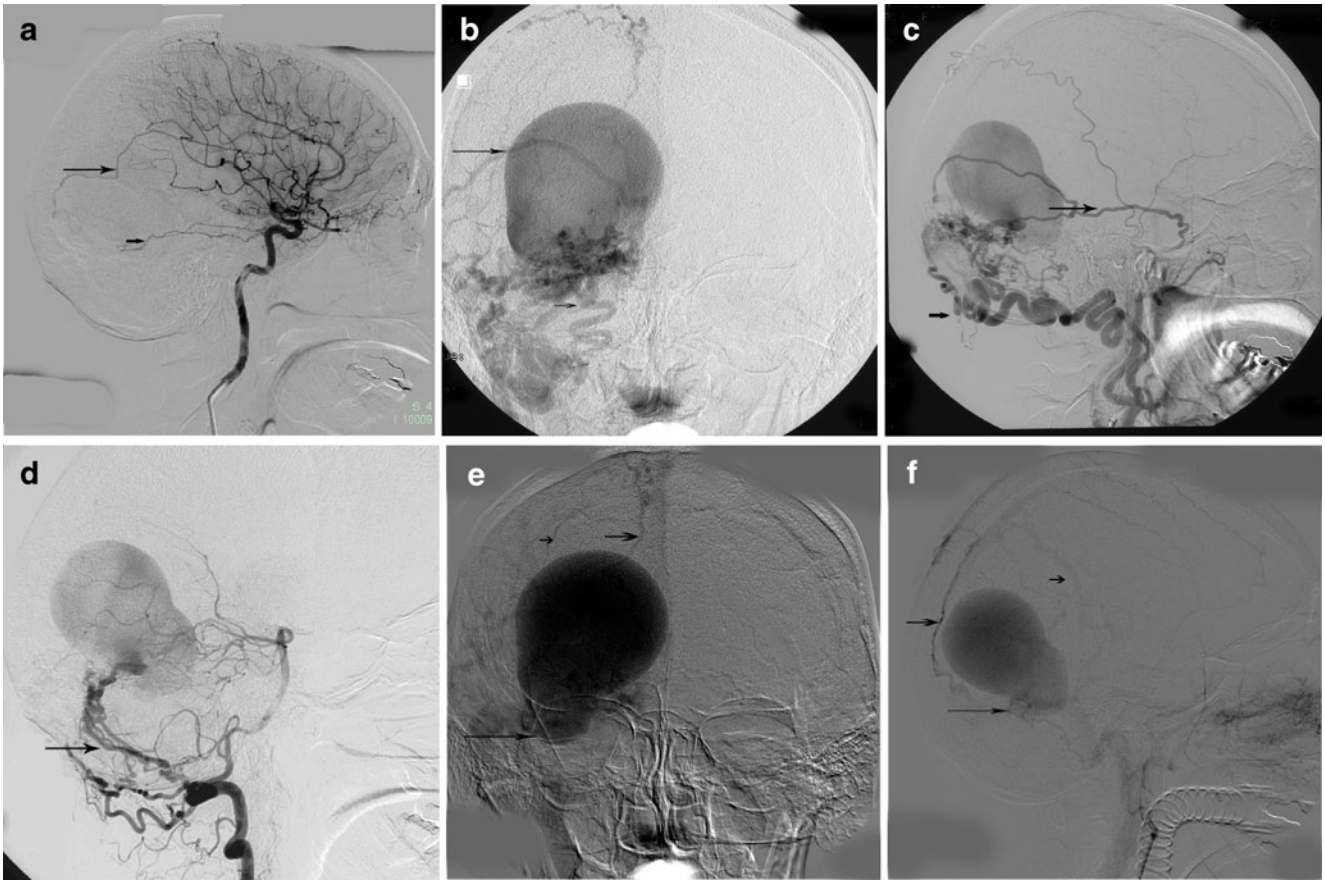

Fig. 4 Initial right internal cerebral artery angiograms: lateral (**a**) view demonstrating very small arterial supply to the fistula from the tentorial artery (*short arrow*) and meningeal branch of the ophthalmic artery (*long arrow*). Right external cerebral angiograms anteroposterior (**b**), lateral (**c**) showing an important tentorial dural arterio venous fistula fed by the middle meningeal artery (*long arrow*) and right occipital artery (*short arrow*), and draining in a huge tentorial venous ampulae, which drains in the right lateral sinus (via a stenosed tentorial sinus) and also in occipital leptomeningeal veins. Lateral (**d**) angiogram view of the right vertebral artery showing the fistula fed by the meningeal branch of the vertebral artery (*arrow*). Anterioposterior (**e**) and lateral (**f**) angiography (venous phase) showing a giant venous ampulae draining in the right transverse sinus, via a stenosed tentorial sinus (*long arrow*) and leptomeningeal veins draining in the superior sagittal sinus (*small and medium arrow*)

CA) was found. Then it was decided to embolise the patient's inferior branch of the right middle meningeal artery using Onyx. The meningeal branch of the right vertebral artery was embolised with Glubran glue realising a radiological disappearance of the fistula.

After embolisation, the patient still presented headaches with no neurological change. The postembolisation angiography showed a complete obliteration of the fistula (Fig. 5).

Four days later, she presented a sudden change of her clinical status with coma, a right midriasis and left hemiparesis. The emergent computed tomography showed an important haemorrhagic occipital mass, important occipital oedema and subfalcorial herniation (Fig. 6).

A surgical option was decided in the emergency room. The patient was operated in prone position using a right occipital supratentorial approach. An occipital craniotomy was realised. After opening the dura, a huge cerebral

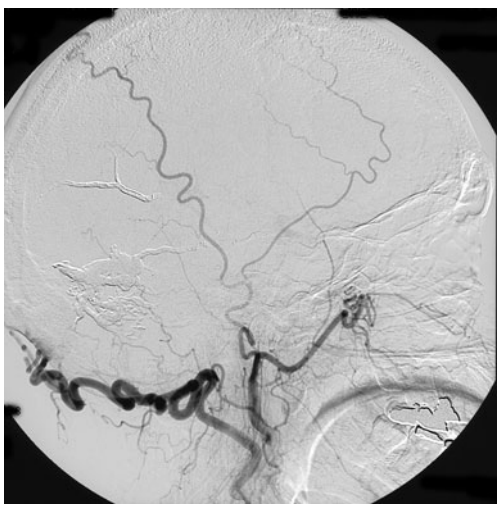

Fig. 5 Postembolisation right external carotid artery angiogram lateral view, showing complete interruption of the fistula

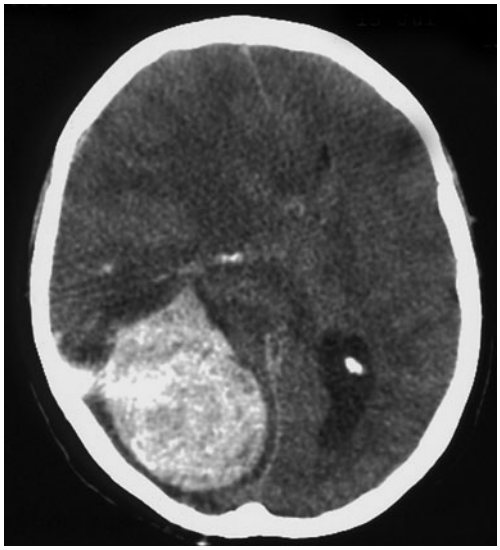

Fig. 6 Computed tomography realised in emergency 4 days post-embolisation, showing an important occipital haemorrhagic mass, occipital oedema and also midline shift with subfalcorial herniation

oedema and intracererbral occipital haemorrhage were noticed. The haematoma was evacuated under operative microscope. There was an important occipital clot in the venous ampulae and around it. The venous ampulae continued with a sinus in the tentorium, which drained in the right transverse sinus. Delicate aspiration of haemorrhage was realised and also the opening of the venous ampoulae. A constant haemorrhage persisted during surgery. Initially, the clot was evacuated from the ampoule and then the wall was completely resected not far from the base where it entered the dura. A small haemorrhage persisted at the neck of the ampulae and also on the tentorium. Electro-coagulation of small vessels on the tentorium was realised. Because of the persistent venous haemorrhage at the neck of the varix, it was decided to suture the neck under microscope. A microvascular continuous suture was carefully realised, then a straight clip was also applied to reinforce the neck suture and the haemorrhage completely stopped. The brain presented a normal pulsatility without oedema.

Control computed tomography showed disappearance of the cerebral oedema and of the occipital mass (Fig. 7).

Postoperatively, the patient recovered progressively. A postoperative angiography was realised and showed a small residual fistulae which drained in a cortical paramedian leptomeningeal vein (Fig. 8a). This drainage vein was observed also on the diagnostic angiography (Fig. 4f, medium arrow).

A new complement of embolisation of the right occipital artery was realised using Glubran glue, giving a complete eradication of the fistula. The angiographic control 1 day and 3 months later showed a complete eradication of the fistulae (Fig. 9).

The patient presented a good recovery and was discharged to a rehabilitation centre with small visual troubles and a slight gait disability. One year after the treatment, she returned to a normal life with only a residual left hemianopsy. The control angiography (3 months after surgery) showed no residual fistulae.

Discussion

Intracranial dural arteriovenous fistulas (DAVF) account for 10 to 15% of all brain AVM and consist of single or multiple fistulas whose niduses lie entirely within the dura [1, 58]. Tentorial dural arteriovenous malformation represent a subgroup of dural fistulas compromising about 4–8% of all dural arteriovenous fistulae [1, 5, 13, 38, 58]. They represent rare and complex lesions in deep locations, with unusual vascular anatomy and critical surrounding neuroanatomy [35]. Tentorial DAVF are located in the tentorial dura mater and are fed primarily from branches of the meningohypophyseal trunk, middle meningeal artery and occipital artery. Venous drainage varies greatly and depends on the tentorial location of the fistula. However, they typically drain into cortical and leptomeningeal veins [5, 36, 45, 55]. Tentorial dural arteriovenous fistulas draining into the spinal venous system are rare lesions [8].

Tentorial DAVF usually have aggressive neurological behaviour with haemorrhage or progressive neurological deficit. Because their complex anatomy, their management represents a challenge and controversial exists regarding the optimal treatment strategy [58].

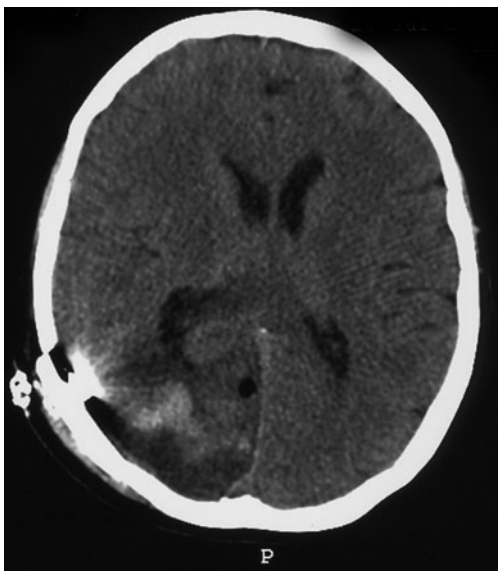

Fig. 7 Postoperative CT cerebral scan showing resection of the occipital mass

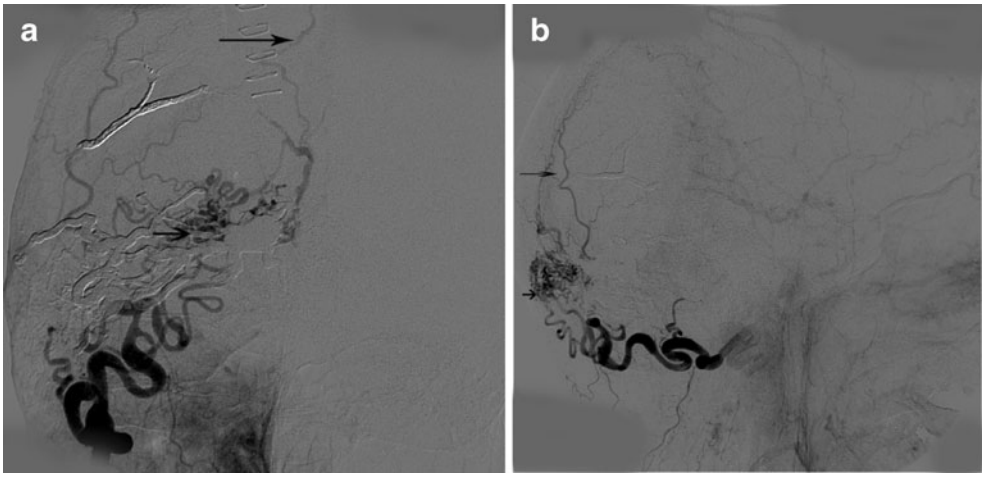

Fig. 8 Postoperative angiograms: right external carotid artery anteroposterior (**a**) and lateral (**b**) views, shows a small residual fistula (*short arrow*) fed by the right occipital artery with cortical paramedian venous drainage (*long arrow*)

Classification systems

It exists multiple classification of dural fistula trying to predict which patients are most likely to develop progressive neurological symptoms and/or haemorrhages [7, 13, 35, 48]. There are two widely used angiographic classification systems for DAVF: the Borden–Shucart and the Cognard classifications [7, 13]. Both the Borden–Shucart and Cognard systems highlight the importance of cortical venous drainage [7, 13, 17].

Our fistula drained essentially in a giant venous ampulae, then threw a small stenosed tentorial sinus in the transverse sinus, but also in occipital leptomeningeal veins. The ampulae increased progressively and started to make a mass effect on the occipital lobe explaining the symtomathology of the patient. We have classified it as Borden IIb, Cognard IV fistula.

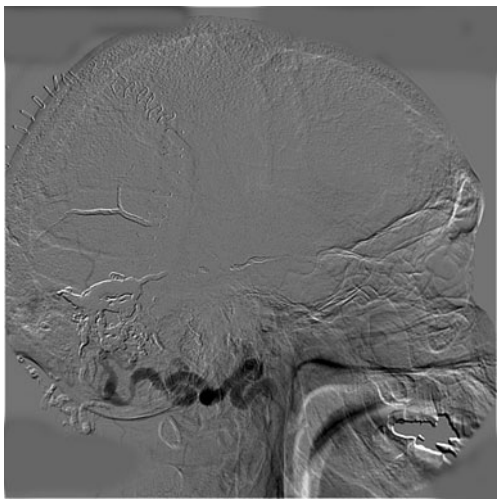

Fig. 9 Right external cerebral angiography (*lateral view*), realised after a complementary embolisation: showing complete disappearance of the tentorial fistula

Pathogenesis, history

Although the pathogenesis of dural AVMs remains controversial, most authors now think that they are acquired; resulting from spontaneous or traumatic sinus thrombosis that revascularizes and in the process develops a dural blood supply with microshunts between the dural layers [11, 36, 44, 46].

Posttraumatic dural arteriovenous fistula of the tentorium and straight sinus are rarely described in the literature [59].

In our case, there was a stenosis of a tentorial sinus which drained in the right transverse sinus, but no major sinus thrombosis. The junction between this tentorial sinus and the right transverse sinus was stenosed explaining the size of the ampulae and the fact the blood remains within before draining in the transverse sinus (Fig. 4c).

A small increase in the degree of stenosis within a draining sinus may lead to a significant rise in the pressure proximal to the restriction, resulting in the development and recruitment of retrograde draining veins and the formation of venous varices or aneurysms, which have high rates of haemorrhage [53].

Lewis reported that 31 (57.4%) of 54 tentorial DAVFs were associated with a venous pouch and 40 (74%) of them presented with intracranial haemorrhage [36].

The natural history and clinical manifestations of DAVMs are largely dictated by the venous drainage of each lesion. A progressive focal neurological deficit or haemorrhage is much more frequently observed in lesions of the petrosal or straight sinus, in fistulae with leptomeningeal venous drainage, with variceal or aneurysmal venous dilations, or in galenic drainage [6, 9, 16, 21, 26, 31, 51, 52].

In one of the largest series, the risk of bleeding is evaluated at 1.8% per year [9]. The source of bleeding is not the fistula but rather the distended leptomeningeal venous varices [38].

Duffau showed that AV *fistulas* with retrograde cortical venous drainage present also a high risk of early rebleeding (35% within 2 weeks after the first haemorrhage), with graver consequences than the first haemorrhage. They therefore advocate complete and early treatment in all cases of AVF with cortical venous drainage revealed by an ICH [21].

Clinics

TDAVFare lesions which can have aggressive neurological behaviour, causing haemorrhage or progressive focal neurological deficits (visual troubles (including bilateral proptosis, bilateral episcleral and retinal venous congestion, conjunctival injection optic disc palor, quadrantanopia and concentric narrowing of the visual field), hemisensory disturbance, hemifacial spasm, atypical trigeminal neuralgia [5, 19, 29, 30, 43, 45, 46, 50]. Rare cases with spinal cord myélopathiy [8, 47], pachymeningitis [25] or even dementia [42] are described in the literature.

The literature cited aggressive neurologic behaviour rates in 55–97% of cases and haemorrhage rates in 38% to 79% of cases [31, 35, 36, 46, 52].

There is a male preponderance in dural fistulas with aggressive behaviour (55–85%) [14, 15, 40].

In this case, there were progressive neurological symptoms due to the mass effect of the varix with headaches, left hemianopsia and ocular pains. Due to the leptomeningeal drainage and a giant venous ampulae, a haemorrhagic risk existed, so a rapid treatment after the diagnosis was decided.

Imaging

Six-vessel catheter angiography remains the most accurate method for the detection and classification of a DAVF.

Recent advances in CT angiography and MR angiography have allowed improved lesion detection with these modalities. These tools, particularly CT angiography, may aid in surgical planning by locating draining veins relative to brain structures. MRI and MR angiography can show dilated and tortuous flow void [27, 30, 44, 46, 58] and can be used also for the follow-up. Complementary informations are also given by SPECT which can show venous congestion [30]

In our case, MRI showed a huge vascular mass. Investigations were completed with six vessels angiography.

Therapy

Because of their complex anatomy, optimal management of this fistulas is a serious challenge.

Due to their aggressive behaviour, tentorial DAVF are treated aggressively when diagnosed, even in the absence of haemorrhage. The goal of treatment is permanent and complete obliteration of flow through the arteriovenous shunt [16, 26, 41, 52, 58].

Optimal management of this fistulas rest also controversial due to their complex anatomy [14, 24, 26].

Therapeutical options of TDAVF include endovascular therapy [6, 18, 31, 41, 55], surgery [24, 35], radiosurgery [10, 43, 49] or combination of them [3, 23, 30, 38, 48, 52, 58, 59].

Endovascular therapy

Endovascular therapy has become the predominant therapy for intracranial DAVF because their arterial supply from the external carotid artery (ECA) can be embolised safely, and their location on dural venous sinuses facilitates access and occlusion through that sinus.

The transarterial [28, 29, 31, 34, 45, 46, 48, 55], transvenous embolisation [12, 32, 37, 56] or the combination of them [3, 31, 33, 36, 38, 53, 54] result in high obliteration rates for most DAVF, but tentorial DAVF are an exception. Endovascular procedure can use coils, glue, particules; *n*-butyl cyanoacrylate or Onyx [18, 28, 29, 58]. It is possible to use more than one session for the therapy of the fistula [6, 29].

Even if embolisation is incomplete, it minimises the bleeding risk for subsequent surgical therapy. Transarterial embolisation also aided in simplifying the angioarchitecture of more complex lesions [38].

The arterial supply of DAVF can be extensive, involving meningeal arteries from the internal carotid artery (ICA) and vertebral artery that are difficult to cannulate and riskier to embolise than ECA feeders. In these cases transvenous navigation was chosen and reported in literature. The transvenous approach was preferred when the venous anatomic features were not too tortuous to allow catheterization of the draining vein at the point of the fistula [37].

The overall morbidity and mortality rate of the endovascular treatment is between 9–10.5% reported in the literature [31, 52] and complication can occur in 22.5% of cases [41, 57]. Residual small fistula can be treated by radiosurgery or surgery [31].

Postembolisation complications are described in the literature as: cerebral infarction, trigeminocardiac reflex or venous rupture with death. Venous rupture can appear if the fistula is subtotally obliterated, because of Onyx migration which can occlude the distal draining vein and may lead to the venous varix rupture [31].

Surgery

The management of tentorial DAVF may require often microsurgical interruption, unlike most other DAVF

because of the anatomy of venous drainage in the subarachnoidian veins rather than venous sinuses [30, 38, 47].

Surgery is usually used complementary to embolisation. It is generally recognised that arterial embolisation alone allows complete and permanent obliteration of the fistula in less of 50% of cases [14, 15]. In fact, failure of distal superselective catheterization is not uncommon, and complete occlusion of the fistula can be very difficult. If the occlusion remains partial, this invariably results in the recruitment of other feeding vessels, often from the ICA and the VB complex, thus making the angioarchitecture of the dural AVF more complex. Furthermore, recanalisation of embolised vessels is possible.

The surgical approach in case of giant dural fistulae rest challenging. Some authors used vascular doppler, intraoperative digital angiography or endoscopy in surgery [2, 24, 30]. Low-flow deep hypothermic cardiopulmonary bypass can be used to control intraoperative bleeding for surgical excision of a giant intracererbral dural arteriovenous fistula [4, 22].

Surgical options described in the literature are: excision of the fistula, surgical skeletonization of the dural sinus, or surgical interruption of leptomeningeal veins [14–16, 35, 38–40, 52]. These options are realised using different approaches: supracerebellar-infratentorial, torcular, supratentorial-infraoccipital, extended retrosigmoid approach, pterional or subtemporal, transpetrous or cranio-orbito zygomatic approach [19, 20, 26, 33, 35, 47, 58].

We operated our patient in emergency in a prone position and we have a good access on the falcotentorial junction.

Collice et al. recommend excision of the sinus in fistulas that drain into a large dural sinus with retrograde filling of leptomeningeal veins; and the simple surgical interruption of the draining veins as they exit the dural wall in fistulas with pure leptomeningeal drainage [14, 15]. By occluding the venous drainage the shunts are obliterated, with subsequent regression of the feeding arteries [32, 51].

Surgical goal of obliterating of the draining vein as its exits the dura is the strategy adopted by many authors [14–16, 35, 38, 52]. In the series of Lawton, fistulas were treated microsurgically by simple interruption of the draining vein, 94% being completely obliterated [35].

Because of a complex anatomy of the fistulae in our case, an endovascular treatment was decided initially and was realised with a good radiological result.

The aggravation appeared in a delayed fashion, 4 days after the embolisation. It shows that patients with important venous engorgement and venous ampulae with mass effect need close follow-up even if they do neurologically well. A fast discharge for this kind of patients is not recommended.

This aggravation was the consequence of the venous ampulae thrombosis with an important cerebral oedema and bleeding.

Post treatment thrombosis with venous engorgement and bleeding was described in the literature and can be a potential cause of rapid clinical aggravation. This condition was found in cases with important preoperative venous engorgement [14, 15].

During surgery haemorrhage was also found around this giant ampulae, as a result of diapedesis and venous rupture following the thrombosis. After haemorrhage aspiration and resection of the venous ampulae, the intracranial hypertension was released but persisted the ampulae's neck situated on the medial upper surface of the tentorium, with a continous small haemorrhage. The neck of the ampulae was sutured under microscope with a 4.0 thread and a great aneurysmal clip was simply adjusted to reinforce the suture, at the place where the venous ampulae entered the dura.

The angiographical postoperative control showed a small residual fistula alimented by the occipital artery and with an occipital leptomeningeal drainage (Fig. 8a). This residual fistula was not treated with a complementary embolisation, and it shows that we must not trust entirely on an early angiographical control.

In our case, the advantage of preoperative embolisation was important in reducing considerably the debit of the fistula. However, due to the importance of the venous varix and its mass effect, an immediate surgical approach of the fistulae after or before the embolisation could be discussed as another management option. This therapeutical option was initially not chosen because of the good neurological status of the patient, high operative haemorrhagic risk and complexity of this fistula.

Transarterial embolisation and even surgery can leave small residual arterio venous shunts because of the complexity of arterial anatomy. For this reason, a close fellow-up in the immediate posttreatment period and long-term angiographical controls is recommended.

This illustrative case shows that on the management of this dural AV fistula rest an important challenge. However, cooperation between neurosurgeon, neuroradiologist and anesthesist is needed. It is recommended that these patients be treated in dedicated neurovascular centres. Regular angiographic controls after 3 months and 1 year are also recommended to eliminate residual fistulas.

Conclusion

Tentorial dural arterio venous fistulas with leptomeningeal drainage and venous ampulae are rare lesions with progressive and aggressive neurological evolution. Treatment is challenging and still often needs a combination of

endovascular and surgical approach depending on the anatomy of the fistula. This article shows that even in case of a good immediate radiological result after endovascular procedure, giant arterio venous fistulas need closed follow-up in our experience because of possible aggravation risk due to venous thrombosis with haemorrhage.

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