Spontaneous Closure of a Type 2a Dural Arteriovenous Fistula Following Late Recanalization of the Occluded Sinus

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Summary

We describe the rare spontaneous resolution of a type 2a dural AVF that coincided with recanalization of the previously thrombosed sigmoid sinus after ten years of conservative management. The factors potentially responsible for spontaneous fistula obliteration are discussed and the therapeutic implication of this observation is considered.

Introduction

Spontaneous closure of dural arteriovenous fistulas is relatively rare, but has been documented in an increasing number of case reports and small series¹⁻⁹. Most cases have occurred with Cognard¹⁰ type 1 AVFs and precipitating factors include venous outflow thrombosis, haemorrhage, cerebral angiography and trauma^{6-8,11}. We describe the rare spontaneous closure of a type 2a fistula associated with recanalisation of the occluded dural sinus and discuss the implication of this observation.

Case Report

A 51-year-old woman presented in 1998 with a six-week history of bilateral pulsatile tinnitus. Eight weeks prior to the onset of tinnitus she described two weeks of left sided headache. A bruit was audible over her left mastoid bone. T2-weighted brain MRI demonstrated increased signal within the distal left transverse

sinus, sigmoid sinus and jugular bulb. Enlarged flow voids were observed within the wall of the sinus on coronal T2 imaging. The sinus was iso-intense with muscle on T1-weighted imaging; the jugular bulb was distended and enhanced with intravenous contrast. These radiological features were interpreted as recent venous sinus thrombosis and a dural AVF within the wall of the left lateral sinus.

A catheter angiogram performed the same day confirmed complete occlusion of the sigmoid sinus and a type 2a dural AVF within the distal left transverse sinus (Figure 1). Arterial pedicles to the fistula were derived from the occipital artery, posterior division of the left middle meningeal artery, artery to the free edge of the tentorium and both posterior meningeal arteries. Venous drainage was entirely retrograde into the contralateral transverse sinus and there was no reflux into the cortical veins.

Endovascular coiling of the transverse sinus was considered in view of the retrograde filling of the contralateral sinus, but the procedure was not undertaken because of the risk of compromising the large vein of Labbé that drained into the sinus. The patient's symptoms were not severe, and because of the unfavourable anatomy of the sinus, a conservative approach was adopted.

The patient represented in 2008 with increasingly severe occipital headaches. After nine days the headaches abated spontaneously and the tinnitus improved. A catheter angiogram performed at this time demonstrated complete resolution of the dural AVF. The left sigmoid sinus had largely recanalized with the result

that antegrade venous flow was established once again in the posterior fossa. There was no evidence of persistent arteriovenous shunting (Figure 2).

Discussion

Previously regarded as congenital vascular malformations, dural AVFs are now widely accepted as acquired lesions that result from an insult to the dural sinuses and / or cortical veins^{11,12}. Clinical reports indicate that venous thrombosis, infection, trauma and surgery predispose to fistula formation, but the aetiology of many dural AVFs remains unexplained¹⁰, despite the observation that certain low-flow dural AVFs, such as those located at the cavernous sinus, can improve or resolve with simple measures such as carotid compression¹, there are few reports of fistulas at other sites resolving spontaneously. The cases of spontaneous fistula closure that are described have mostly been observed in type 1 fistulas and resolution has usually occurred approximately 18 months from diagnosis⁶.

The mechanism by which dural AVFs spontaneously resolve is poorly understood. Factors that have been implicated in previous reports include (i) venous outflow thrombosis¹¹ (ii) haemorrhage⁸, (iii) cerebral angiography⁷ and (iv) trauma⁶. Whilst it seems intuitive that simple, low-flow fistulas with a single venous outlet are more likely to resolve than larger, more complex fistulas, such as those located on the larger dural sinuses, the evidence for this is lacking and it is usually assumed, rather than proven, that this occurs as a result of venous outflow thrombosis.

The majority of the reported dural AVFs that have resolved have been type 1 lesions and have obliterated in the absence of intracranial haemorrhage. The arterial pedicles to the fistulas are extradural and not subject to potential vasoactive agents within the cerebrospinal fluid and, whilst in theory an intracerebral clot may compress an intradural vein associated with a non-type 1 fistula, in clinical practice it does not appear that either direct mass effect, or inflammation / vasospasm within the draining vein is a common explanation for fistula closure. In three previously reported type 3 dural AVFs, occlusion occurred in all cases as a result of 'thrombosis', not haemorrhage, one in the context of cerebral angiography^{5,7,11}.

It has also been postulated that post-traumatic lesions are more likely to resolve than more complex spontaneous fistulas^{6,13}. Traumatic lesions may represent a more favourable subgroup, in part due to the more simple anatomy that is sometimes observed when there is a single venous outlet into the relatively smaller skull base veins. Acute inflammation and subsequent scar may predispose to spontaneous resolution and it is possible that the incidence of these fistulas is underestimated as many lesions may be sub-clinical^{6,13}.

Complete resolution of certain dural AVFs can occur without occlusion of the draining sinus, possibly due to compression of the arteriovenous connections within the diseased sinus wall as it undergoes 'structural or conformational changes'^{1,6,8}. Satomi et al. demonstrated the natural history of benign dural AV fistulas (i.e. those without cortical venous drainage) with over 98% tolerable and self-limiting disease¹⁴. They concluded that such benign lesions could be monitored with close clinical follow-up and further radiological assessment in the setting of symptom deterioration¹⁴. Kim et al. demonstrated a 4% conversion rate of a benign dural AVF into an aggressive lesion⁹.

We are aware of only very limited reports of spontaneous fistula resolution associated with spontaneous sinus recanalization^{2,4} and suggest this is a further, albeit extremely rare, method by which dural AVFs may spontaneously resolve. Mori et al. concluded that the sinus occlusion was caused by the high arterial flow to the fistula and its disappearance made recanalization of the sinus possible². We suspect that the reverse is true, and that recanalization can promote dural AV resolution. In the same way that venous hypertension can initiate fistula formation¹⁵ (even in the absence of thrombosis) we postulate that persisting hypertension within the sinus is required to maintain the patency of certain dural AVFs. Kultluk et al. described two recanalizing sigmoid dural AVFs one of which was associated with disappearance of the fistula⁴; the second fistula persisted following recanalization demonstrating that sinus re-opening is not dependent upon closure of the fistula but this also demonstrates that immediate closure of the fistula is not inevitable⁴.

The observation that sinus recanalization can lead to fistula closure has important therapeutic consequences. Although there is a move away from sinus sacrifice as a treatment for

Figure 1 Lateral (A) and AP (B) projections of a catheter cerebral angiogram with selective left external carotid artery injection. The dural AVF can be clearly identified with predominant arterial supply from the occipital artery. The left sigmoid sinus is occluded and there is retrograde drainage via the right transverse sinus.

Figure 2 Follow-up catheter cerebral angiogram following spontaneous relief of tinnitus associated with a severe occipital headache. Lateral view external carotid artery (A) and AP left internal carotid artery (B) injection, venous phase, demonstrate complete resolution of the dural AVF and re-canalisation of the previously occluded sinus.

symptomatic or aggressive lesions, in favour of liquid embolization of arterial pedicles, this case implies that occlusion of certain dural AVFs could be achieved by sinus recanalization if this can be achieved by therapeutic means such as balloon angioplasty and stenting

¹⁶. It may be the case that such therapeutic measures may not only restore antegrade sinus flow and prevent cortical venous reflux, but may also initiate a process by which the fistula spontaneously resolves without the need to always tackle the arterial pedicles.

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