Regression of a Flow-Related Ophthalmic Artery Aneurysm After Treatment of a Frontal DAVS

A Case Report

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Summary

We report a case of a frontal dural arteriovenous shunt or fistula (DAVS) adjacent to the left side of the cribriform plate, with bilateral supply from multiple arteries, the most prominent being the dural branches originating from the anterior ethmoidal artery coming from the left ophthalmic artery. Before treatment there was an eight mm flow-related arterial aneurysm proximally on the left ophthalmic artery. After transarterial embolization of the DAVS with N-butyl cyanoacrylate and polyvinyl alcohol, minimal shunting still remained. At follow-up angiography six months after the treatment, the shunt was obliterated and the ophthalmic artery aneurysm had regressed completely. Our case illustrates that complete obliteration of a DAVS may be achieved even though arteriovenous shunting remains at the end of the procedure. Furthermore, a flow-related arterial aneurysm, may not warrant any specific treatment. Elimination of the high flow situation can lead to complete regression of these aneurysms.

Introduction

Dural arteriovenous fistulae or shunts (DAVS) in the adult population can cause neurologic symptoms and intracranial haemorrhage^{1,2}.

Generally believed to be acquired lesions, a DAVS may be related to traumatic brain injury or cranial surgery³⁻⁶ but in most cases the pathogenesis is unknown. Several hypotheses have been proposed as to the development and natural history of non-traumatic DAVS, e.g. dural sinus thrombosis and venous hypertension^{7,8}.

There are two major classifications of DAVS^{1,9} but the principal divider is whether cortical venous reflux, i.e. retrograde flow of arterial blood into the pial venous system, is present or not. If present, the annual risk of intracranial haemorrhage has been reported to be as high as 8.1%¹⁰ and subsequently there is an absolute indication for urgent treatment.

Flow-related aneurysms, i.e. aneurysms proximally located on a main feeding artery, belong to the complex changes in the wall of a vessel exposed to a pathologic high flow situation, such as in an AVM or DAVS¹¹. Though previously debated, it has been shown that unruptured flow-related aneurysms do not usually warrant any specific treatment as the majority of them disappear spontaneously after treatment of the distal arteriovenous shunt^{12,13}. We here present a case with complete regression of a flow-related ophthalmic artery aneurysm six months after endovascular treatment of a left frontal DAVS.

Case Report

The patient was a 77-year-old woman with a medical history of well controlled hypothyroidism, who presented with a one-year history of progressive memory loss. In addition, she experienced sporadic episodes of strange smelling sensations and disturbed spatial recognition that was interpreted as possibly being seizure-related.

The neurologic examination confirmed memory problems but was otherwise normal. A contrast enhanced CT-scan was indicative of a left-sided ophthalmic artery aneurysm and also revealed a left frontal edema with tortuous vessels interpreted as dilated veins. On MRI with gadolinium, the aneurysm was verified measuring approximately 8 mm in diameter and with a narrow neck. There was still a left frontal lobe edema as shown by high signal intensity on T2 weighted images as well as tortuous and ectatic dural and cortical veins with a large venous pouch. The findings were consistent with a DAVS with probable cortical venous reflux. The aneurysm was believed to be flow-related rather than a separate entity. The patient was referred to our institution for angiographic evaluation and endovascular treatment, primarily of the DAVS.

A cerebral angiography followed by embolization was then performed with the patient under general anaesthesia. The frontal DAVS was confirmed, supplied by bilateral dural branches from the anterior ethmoidal arteries coming from the ophthalmic arteries, particularly on the left side. Additionally, there was supply from branches of the internal maxillary artery bilaterally, from the left middle meningeal artery and bilaterally from the sphenopalatine arteries. The frontal branch of the right middle meningeal artery supplied the anterior falcine artery, which provided blood to the fistula in a retrograde fashion. The feeders converged to a single fistulous point draining into an enlarged and ectatic cortical vein with a large venous pouch and onwards into the superior sagittal sinus. In addition, there was an arterial aneurysm on the left ophthalmic artery, located 3-4 mm from the origin at the internal carotid, measuring approximately 8 mm in diameter (figure 1A). This aneurysm was believed to be flow-related.

The main contributor to the DAVF, from the left ophthalmic artery, was embolized with a

50/50 mixture of N-butyl cyanoacrylate (NBCA, Braun, Aesculap, Tuttlingen, Germany) and Lipiodol ultra-fluide (Laboratoire Guerbet, Aulnay-Sous-Bois, France). It was not possible to achieve complete flow-arrest conditions with a wedged catheter position^{14,15} during the embolization. Even so, the glue reached the vein and the venous pouch and a subsequent angiogram revealed significant flow reduction. There was, however, some residual shunting, mainly from the external carotid artery feeders bilaterally, and those were therefore embolized with polyvinyl alcohol, sized 150-250 microns (PVA, "Contour", Target Therapeutics, Fremont, CA, USA). At this point, even though there was still some residual shunting from small branches originating from the right ethmoidal artery and the left sphenopalatine artery, we decided that further embolization was unnecessary. There was no attempt to treat the ophthalmic artery aneurysm. A CT-scan six days after the embolization showed embolic material in the draining veins but no contrast enhancement, i.e. in concordance with further thrombosis of the DAVS. The ophthalmic artery aneurysm was unchanged.

Follow-up cerebral angiography was performed slightly less than six months after the treatment (figure 1B) and showed occlusion of the DAVS. Furthermore, the left ophthalmic artery aneurysm had regressed completely and the left ophthalmic artery was almost normalized in size.

Discussion

DAVS with cortical venous involvement, either directly or via dural sinus reflux, carry a high risk for intracranial haemorrhage¹⁰. In addition, symptoms of the disease, e.g. neurologic deficits and seizures due to venous congestion and hypertension, can be devastating for the patient¹.

Curative treatment is consequently warranted and embolization, transarterial or transvenous, has become the primary treatment modality in many centers. Transarterial embolization with a liquid agent requires penetration of the glue to the proximal aspect of the draining vein, thereby eliminating the pathologic arteriovenous connection¹⁵. Even under such circumstances, it might, however, be beneficiary to embolize collateral contributors with PVA before occluding the pathologic shunt with NBCA

Figure 1 Lateral view of pre-embolization left ICA angiogram (A) shows the DAVS fed by dural branches from the anterior ethmoidal artery. Note the flow-related aneurysm located proximally on the ophthalmic artery. At follow-up cerebral angiography six months after the treatment (B), the shunt was totally occluded and the ophthalmic artery aneurysm had regressed completely.

through the main feeder. This is especially important in high-flow DAVS to better control the glue injection and avoid fragmentation of the glue column. In our patient, the flow was relatively slow and no preparative PVA embolization was performed.

The NBCA transversed well into the vein as a single column but there was still some residual shunting through multiple tiny dural feeders, probably since there was more than one fistulous connection. After treatment of these collateral contributors with PVA, minimal shunting remained but the flow was markedly reduced. Under the assumption that progressive thrombosis would lead to obliteration of the shunt, no further embolization was performed. Indirect signs of thrombosis and no contrast medium in the former draining veins were evident on the CT-scan six days after the treatment.

Flow-related arterial aneurysms have been well described and evaluated, especially in connection with brain AVMs, and are generally assumed to carry a low risk of rupture^{13,16}. They are usually located on main feeders of an intracranial arteriovenous shunt and are expres-

sions of a vascular remodelling process triggered by the increased shear stress in the vessel. Such progressive vessel changes secondary to flow have been demonstrated in experimental arteriovenous shunts¹¹. Tears in the internal elastic lamina of the afferent artery appear after two to five days with high flow^{17,18}. Further changes involve all layers of the vessel and may progress to a point where the layers are destroyed and the vessel is actually unidentifiable as being an artery or a vein¹⁹. Localised fibrointimal thickening as well as uneven stretching and distension create wall thinning and aneurysms²⁰. These histopathological changes in an AVM nidus and its afferent and efferent vessels have been proposed to be secondary to the high flow^{11,19,20} and increased shear stress. However, the vessels adapt to the increased flow by widening, and at rest shear stress in these vessels is not significantly higher than in arteries supplying normal brain tissue²¹. Thus, additional factors such as lack of autoregulation with augmented flow changes during normal blood pressure variations or disturbed laminar flow due to tortuous vessels may interact with the endothelium and promote vascular remodel-

ling. Consequently, if the triggering factor, i.e. the increased shear stress, is removed, the vessel wall will again adapt and remodel whereby the flow-related aneurysms in the majority of cases will disappear.

Our case had a presumed flow-related aneurysm located on the ophthalmic artery, that had disappeared six months after the flow in the artery had been normalized. Thus, flow-related aneurysms, being normal vessel-wall adaptations to increased shear stress, may be treated by removal of this pathological trigger, i.e. treatment of the AV-shunt.

Conclusions

Transarterial embolization is an effective treatment for DAVS and progressive thrombosis may lead to obliteration even if minimal shunting remains at the end of the treatment procedure. Flow-related arterial aneurysms in relation to DAVS or brain-AVMs result from a vascular remodelling process in the normal vessel wall probably triggered by increased shear stress. Elimination of the shunt is the primary treatment, since, as in our case, it often leads to disappearance of the aneurysms.

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