Spontaneous resolution of a flow-related ophthalmic-segment aneurysm after treatment of anterior cranial fossa dural arteriovenous fistula

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Abstract

Background: The natural history of proximal, feeding-artery aneurysms after successful obliteration of high-grade, anterior cranial fossa dural arteriovenous fistulas (dAVFs) has not been well documented.

Case Description: A 52-year-old Caucasian male presented with an unruptured anterior cranial fossa (dAVF) and an associated aneurysm. Cerebral angiography revealed a large, contralateral, carotid-ophthalmic segment aneurysm, enlarged feeding ophthalmic arteries, as well as cortical venous drainage. Successful surgical obliteration of the dAVF was undertaken to eliminate the risk of hemorrhage.

Conclusion: The carotid-ophthalmic aneurysm regressed significantly after surgical obliteration of the dAVF and a follow-up, planned coiling procedure to address the carotid-ophthalmic aneurysm was abandoned. This represents the first reported case of a near complete, spontaneous resolution of an unruptured carotid-ophthalmic aneurysm associated with a high-grade anterior cranial fossa dAVF.

Key Words: Anterior cranial fossa, cortical venous drainage, dural arteriovenous fistula, flow-related aneurysms

INTRODUCTION

Dural arteriovenous fistulas (dAVFs) comprise 10-15% of all arteriovenous malformations (AVMs)¹⁴ and are abnormal arteriovenous (AV) shunts most frequently located in the walls of the cavernous, sigmoid, and transverse sinuses that are characteristically fed by meningeal arteries.³ Surgery, trauma, infection, and sinus thrombosis have all been proposed as possible etiologies for dAVFs.¹¹ In 1963, the French neurosurgeon, Lepoire first described anterior cranial fossa dAVFs.¹⁷ Today, anterior cranial fossa dAVFs are recognized as a unique subgroup of vascular malformations due to their high rate of hemorrhage.⁹,¹² Eight percent of all dAVFs appear in the anterior cranial fossa, typically, in the fifth to sixth decades of life with strong male predilection.¹⁰ The majority of dAVFs result in hemorrhage and nearly half of these hemorrhages are attributable to feeding artery aneurysms.⁸ Given the variable complexity of dAVFs with concomitant aneurysms and their high propensity to hemorrhage, expeditious, multi-disciplinary management of these unique vascular lesions is advisable to prevent significant
neurologic morbidity and mortality. No consensus guidelines for treatment of dAVFs and concomitant aneurysms exist, but management options include endovascular treatment and stereotactic radiosurgery as well as microsurgical obliteration, which offers a high cure rate. Treatment of the dAVF and all associated aneurysms may not be necessary in one setting as microsurgical obliteration of the dAVF may result in complete resolution of the feeding artery aneurysms, thus obviating the need for longer and more complex surgical exposures or secondary endovascular treatments. While complete regression of a posterior fossa, flow-related aneurysm has been described, we believe this to be the first reported case of near-complete, spontaneous resolution of a large flow-related ophthalmic aneurysm following treatment of a high-grade, unruptured, anterior cranial fossa dAVF.

**CASE REPORT**

A 52-year-old, right-handed, Caucasian male was referred to our outpatient neurosurgery clinic for evaluation of progressively worsening headaches over a 2-week period. On examination, the patient did not have exophthalmoses, chemosis, or bruit, and was otherwise neurologically intact. There was, however, a prominent right superficial temporal artery that engorged with valsalva-type maneuvers.

Cerebral angiography revealed a dAVF in the anterior cranial fossa. The fistula was fed by bilateral ophthalmic arteries via the anterior ethmoidal branches [Figure 1a and b], as well as a frontal branch of the right superficial temporal artery [Figure 2]. The fistula drained to the superior sagittal sinus via cortical veins with associated venous varix dilatation (Cognard type IV lesion). Injection of the left ICA also revealed a large, wide-necked, 10-mm carotid-ophthalmic aneurysm [Figure 3] incorporating the parent artery [Figure 4a and b]. Given the angioarchitecture of the carotid-ophthalmic aneurysm, microsurgical obliteration of the fistula and the aneurysm was undertaken instead of any endovascular treatment to minimize the risk of ophthalmic artery occlusion and blindness.

The patient’s dAVF was successfully obliterated using standard microsurgical techniques through a right pterional craniotomy. Clipping of the contralateral carotid-ophthalmic aneurysm was attempted but since it required significant brain retraction, an intraoperative decision was made to abandon aneurysm clipping with the understanding that the patient could undergo coiling of the aneurysm shortly after surgery. Obliteration of the AV fistula was confirmed intraoperatively by fluorescein angiography. The patient was discharged home after an uneventful hospital course and a short-term, follow-up visit revealed resolution of the patient’s headaches.

The scheduled postoperative cerebral angiography with intent to coil the carotid-ophthalmic aneurysm a mere 28 days following surgery revealed no fistulous connection, return of bilateral ophthalmic arteries to normal, and disappearance of the prominent right superficial temporal artery.
their normal caliber, and an 85% reduction in size of the aneurysm. The small, residual aneurysm appeared round with a smooth, regular outline [Figure 5]. Because of this dramatic reduction in size of the aneurysm and its benign angiographic morphology, we elected not to coil the aneurysm and instead opted for conservative management with serial imaging and aggressive follow-up.

DISCUSSION

Recent reports have demonstrated that 13-21% of dAVFs harbor associated aneurysms[23] and that a third of these aneurysms appear on the feeding arteries and the remaining two-thirds on remote arteries.[8] It is also estimated that dAVFs with concomitant aneurysms have a hemorrhage rate as high as 58%; half of these hemorrhages have been attributed to a proximal feeding-artery aneurysm while the other half to the dAVF itself.[8] Different mechanisms have been proposed to explain the relationship of the AV fistulas and aneurysms. Hemodynamic stress, genetic or developmental defects, and pure coincidental occurrences are some of the theories put forward. However, no theory can satisfactorily explain the development of both feeding artery and remote aneurysms and this phenomenon deserves further investigation.

Cortical venous hypertension and resultant intracranial hemorrhage is the major cause of morbidity and mortality associated with most dAVFs.[18] Due to the limited available data on dAVFs associated with flow-related aneurysms, no consensus guidelines exist for the treatment of such dAVFs.[7] As a result, data from reports on pial AVMs and associated aneurysms have been extrapolated to help direct the management of complex dAVFs with related aneurysms.[13]

Patients with concurrent pial AVMs and flow-related aneurysms have an annual hemorrhage risk of approximately 7-20%.[15,24] To further complicate the management of dAVFs with associated aneurysms, treatment of pial AVMs associated with aneurysms has been controversial. Thompson et al. have advocated treating the aneurysm first[24] while others have suggested that obliteration of the AVM nidus could result in resolution of flow-related aneurysms.[15,22] Meisel et al. have further demonstrated that complete obliteration of a pial AVM nidus results in 50% reduction in the size of prenidal aneurysms in about 3.5 years.[20] However, no study to date has demonstrated the effects of dAVF obliteration on a flow-related aneurysm and no report is available on how treatment of a large, feeding-artery aneurysm affects the natural history of a distal dAVF.

Only one report exists in the literature describing an anterior cranial fossa dAVF associated with a ruptured ophthalmic artery aneurysm. The authors treated the aneurysm by endovascular means followed by gamma knife radiosurgery of the AV fistula.[1] Only 13 cases have been reported in the literature describing an anterior cranial fossa dAVF associated with an aneurysm and different treatment modalities have been employed for the AV fistula and the associated aneurysms, underscoring the lack of guidelines for treating these unique vascular pathologies.

We report the case of a 52-year-old Caucasian male with headaches and prominent scalp vasculature who was found to have a large, unruptured left carotid-ophthalmic aneurysm in addition to a high-grade anterior cranial fossa dAVF. Although studies have demonstrated that safe embolization of vascular lesions supplied by ophthalmic arteries can be achieved,[14,16] our patient chose to undergo surgery given the senior author’s (GM) success with microsurgical obliteration of dAVFs as well as the potential risk of blindness inherent in endovascular treatment of any aneurysm that incorporates the ophthalmic artery. We initially intended to address both vascular lesions in one setting through a right pterional craniotomy but abandoned the
ophthalmic artery aneurysm clipping because it became apparent intraoperatively that successful clipping would require extensive brain retraction that could potentially increase surgical morbidity. To address the untreated ophthalmic artery aneurysm, we scheduled the patient for a short-term cerebral angiography with intent to treat the aneurysm. Much to our surprise, the postoperative angiogram revealed near-complete resolution of the ophthalmic aneurysm in an unexpectedly short time forcing us to abort any coiling procedure. Aneurysm remodeling or partial thrombosis of the aneurysm dome could be possible explanations for the rapid reduction in the size of the ophthalmic aneurysm, however, we do not have a short-term, follow-up computed tomography (CT) or magnetic resonance imaging (MRI) study after the postoperative angiogram to confirm this. We have elected to follow the patient’s small, residual aneurysm because it remains asymptomatic.

**CONCLUSION**

Anterior cranial base dAVFs associated with flow-related aneurysms carry a high morbidity and mortality risk due to their high propensity to hemorrhage and, as such, require aggressive, multidisciplinary treatment. While no consensus guidelines exist on the management of high-grade dAVFs associated with feeding artery aneurysms, successful microsurgical obliteration of anterior cranial fossa dAVFs may result in near complete resolution of parent artery aneurysms.

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