Resolution of an Anterior-Inferior Cerebellar Artery Feeding Aneurysm with the Treatment of a Transverse-Sigmoid Dural Arteriovenous Fistula

Case Report

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Abstract

The authors describe a 27-year old man who developed an unruptured anterior-inferior cerebellar artery feeding aneurysm from a transverse-sigmoid dAVF and its subsequent resolution with the treatment of the dAVF. The patient, with a known history of left transverse and sigmoid sinus thrombosis, presented with pulse-synchronous tinnitus. Angiography revealed an extensive dAVF, with feeders from both the extracranial and intracranial circulations, involving the right transverse sinus, the torcula, and the left transverse/sigmoid sinuses. Multimodal endovascular and open surgical therapy was employed. Prior to a planned second-stage treatment for the left sigmoid sinus component, the dAVF improved significantly, but a small flow-related aneurysm developed on the left AICA feeding the petrous dural region in the interval. Resection of the involved sigmoid sinus resulted in resolution of the aneurysm. This is the first reported case of an unruptured feeding-artery aneurysm in an intracranial dAVF that resolved spontaneously with the treatment of the dAVF. Until more is known about its natural history, the decision on when and whether to treat an unruptured dAVF feeding-artery aneurysm must be made on an individual basis.

Running Head: Dural Arteriovenous Fistula with a Feeding-Artery Aneurysm

Key Words: Aneurysms; Dural Arteriovenous Fistulas, Endovascular therapy, Surgery
Introduction

Dural arteriovenous fistulas (dAVFs) are vascular abnormalities of the dura mater that typically involve the dural sinuses, most commonly the transverse and sigmoid sinuses.\(^1\) dAVFs are acquired lesions, often developing after dural sinus thrombosis (DST), and have a variable presentation ranging from a pulse-synchronous tinnitus to intracerebral hemorrhage.

Unlike with arteriovenous malformations (AVMs), concurrent arterial aneurysms and intracranial dAVFs are extremely rare.\(^2-4\) The two other patients detailed in the literature both presented with subarachnoid hemorrhage (SAH). We present a unique case of a patient who developed an unruptured anterior-inferior cerebellar artery (AICA) feeding-artery aneurysm from a transverse-sigmoid dAVF that resolved spontaneously with the treatment of the dAVF. The unique issues surrounding the management of feeding-artery aneurysms associated with an intracranial dAVF are discussed.

History and Examination

This 27-year-old man originally presented with an acute headache, vomiting, and meningismus in August 2003. On the following day, he developed horizontal diplopia and was found to have bilateral 6th nerve palsies and papilledema. A diagnosis of intracranial hypertension was made, and he was started on Diamox. The origin of the intracranial hypertension was initially thought to be doxycycline-related.\(^5\)

Further evaluation with a magnetic resonance venogram (MRV) revealed thrombosis of the left transverse and sigmoid sinus. The patient was started on Coumadin. His hypercoagulable work-up was remarkable for hyperhomocysteinemia. His diplopia
and papilledema resolved with acetazolamide therapy, but several months later, despite maximal medical therapy, he had recurrence of his headaches and diplopia, and he developed visual field deficits associated with severe papilledema. As a result, he underwent a right optic nerve sheath fenestration. Although the papilledema improved, the remainder of the patient’s signs and symptoms of elevated pressure persisted.

Ten months after his initial presentation, the patient developed a palpable and prominent left occipital artery with an audible bruit. Cerebral angiogram revealed an extensive dAVF involving the proximal right transverse sinus, the torcular, the left transverse sinus, and the left sigmoid sinus. Supply to the arteriovenous fistula was from the occipital arteries, the middle meningeal arteries, the left posterior meningeal artery, and a muscular branch off the left vertebral artery. Supply was also present from the tentorial branches and a prominent left anterior-inferior cerebellar artery (Fig. 1). No cortical reflux was seen.

Because maximal medical therapy and optic nerve sheath fenestration had not relieved the situation and there was the potential for deterioration of the left optic nerve function, multimodal and staged therapy was planned. The patient underwent staged transarterial embolizations, followed by a bilateral occipital craniotomy for skeletonization and placement of interpositional dura xenograft (bovine pericardium) at the involved transverse dural sinuses and the left sigmoid sinus. The goal of the operation was to disconnect all of the involved sinuses from aberrant arterial feeders and to prevent future recurrences with the use of interpositional dura xenograft. Postoperative angiography showed marked improvement of the dAVF with small residual feeders to the transverse and sigmoid sinus component on the left. The patient’s follow-up angiogram 4
months later, however, revealed slightly more prominent feeders and an interval development of a 3-mm left AICA feeding-artery aneurysm beyond the floccular segment (Fig. 2). He subsequently underwent a planned second-stage transtemporal (presigmoid, retrolabyrinthine) approach for resection of the involved sigmoid sinus on the left. The sinus was removed, from the entrance of the vein of Labbe to the jugular bulb. The left transverse sinus was then coil-embolized through a transvenous approach. A 1-month follow-up angiogram revealed complete resolution of the feeding-artery AICA aneurysm (Fig. 3). The patient’s symptoms, papilledema, sixth nerve palsies, and bruit also resolved completely.

Discussion

Dural arteriovenous fistulas represent 10% to 15% of all intracranial vascular malformations. They consist of pathologic vascular channels within the dura mater and usually involve the walls of dural sinuses. Patients typically present between 40 and 60 years of age at various clinical stages, ranging from a pulse-synchronous bruit to a neurologic deficit to intracerebral hemorrhage.¹

All dAVFs are acquired, and many are preceded by DST.⁶⁻⁸ It is believed that venous hypertension from DST enlarges the normally present microscopic arteriovenous shunts in the walls of the sinus, leading to the formation of dAVFs. In addition, venous hypertension induces ischemia, which may also play an important role in the pathogenesis of dAVFs by stimulating angiogenic growth factors and angiogenesis.⁹
Several grading schemes\textsuperscript{10-12} have been proposed to predict the clinical course of dAVFs. They all suggest that cortical venous drainage, a high-grade feature, is associated with a significant risk of intracerebral hemorrhage (33\% to 42\%).\textsuperscript{1,13}

Dural arteriovenous fistulas involving the transverse and sigmoid sinuses are the most common, representing up to 50\% of cases. The epicenter is usually at the junction of the transverse and sigmoid sinus. Only 10\% to 15\% of these lesions exhibit aggressive behavior.\textsuperscript{1} Their arterial supply is typically from both the extracranial (branches of the occipital and middle meningeal artery, posterior auricular artery, and ascending pharyngeal artery) and the intracranial system (posterior meningeal branches of the vertebral artery and marginal tentorial branches of the meningohypophyseal trunk).

Although there are reports of spinal dAVF with associated feeding artery aneurysms,\textsuperscript{14,15} there are only a few reported cases of feeding-artery aneurysm associated with intracranial dAVFs.\textsuperscript{2-4} The two cases detailed in the literature involved an AICA and a posterior-inferior cerebellar artery (PICA) feeding-artery aneurysm. Both aneurysms presented with SAH and were treated surgically with good outcomes.\textsuperscript{2,3} In contrast, our patient developed an asymptomatic AICA feeding-artery aneurysm from a transverse-sigmoid dAVF that resolved spontaneously with the treatment of the dAVF.

The etiology of feeding-artery aneurysms is controversial, but the main theory focuses on flow-related stress on the arterial feeders.\textsuperscript{16,17} Based on the morphology of the feeding vessel, we believe that the feeding-artery aneurysm in our case is also flow-related and is a result of the hemodynamic stress on the anterior inferior cerebellar artery caused by the high-flow dAVF.
Intracranial hemorrhage resulting from venous hypertension is a main cause of morbidity and mortality related to dAVF. Cortical venous drainage, as mentioned, is the major risk factor. At present, the additional effect of concurrent arterial aneurysms on the hemorrhagic risk of dAVFs is unknown, although these feeding-aneurysms can clearly hemorrhage.\textsuperscript{2,3} The literature on AVMs and concurrent aneurysms may shed some light on the issue. Aneurysms associated with AVMs have been reported in 5\% to 7\% of cases.\textsuperscript{18,19} and several retrospective and prospective studies suggested a higher risk of hemorrhage in patients with AVM and concurrent feeding-artery and intranidal aneurysms.\textsuperscript{18-21} Brown et al.\textsuperscript{22} showed an increased annual risk of 7\% for intracranial hemorrhage in the setting of an unruptured AVM with a concurrent intranidal or feeding-artery aneurysm, giving an annual hemorrhage rate of 10\%.

The treatment of feeding-artery aneurysms with AVMs is controversial. Some recommend a conservative approach as they believe that these are flow-related lesions that may regress or remain stable once the AVM is removed.\textsuperscript{11} Others propose that they should be treated immediately because of their risk of hemorrhage.\textsuperscript{18,23} At present, there is no treatment guideline regarding concurrent arterial aneurysm and dAVF. In our case, we elected to follow the aneurysm, because of its small size and its lack of symptoms, and because we believed it was a flow-related lesion that would regress once the dAVF was obliterated. In support of our theory, the aneurysm regressed spontaneously after the staged, multimodal treatment of the dAVF. Until more data are available regarding concurrent aneurysms and dAVFs, however, the decision of when and whether to treat an unruptured dAVF feeding-artery aneurysm must be made on an individual basis.
Acknowledgments

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References


Figure Legends

**Figure 1.** Cerebral angiogram left maxillary (A) and vertebral (B) injections revealed an extensive dAVF involving the torcula and the left transverse and sigmoid sinus with feeders from both extracranial and intracranial circulations.

**Figure 2.** Follow-up angiogram, left vertebral injection, after the first-staged operation revealed an interval development of a 3-mm left AICA feeding-artery aneurysm beyond the floccular segment (arrows). (A) Anteroposterior view; (B) lateral view.

**Figure 3.** Follow-up angiogram, left vertebral injection, after the second-staged procedure demonstrated complete spontaneous resolution of the AICA feeding-artery aneurysm (arrows).