Disappearance of a Ruptured Distal Flow–Related Aneurysm after Arteriovenous Malformation Nidal Embolization

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Key words
- Arteriovenous malformation
- Endovascular treatment
- Flow-related aneurysm
- Intraventricular hemorrhage

Abbreviations and Acronyms
ACA: Anterior cerebral artery
AVM: Arteriovenous malformation
DSA: Digital subtraction angiography
EDAS: Encephaloduroarteriosynangiosis
IVH: Intraventricular hemorrhage
MCA: Middle cerebral artery
STA: Superficial temporal artery

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Citation: World Neurosurg. (2015) 84, 5:1496.e1-1496.e6. http://dx.doi.org/10.1016/j.wneu.2015.05.065

Journal homepage: www.WORLDNEUROSURGERY.org

Available online: www.sciencedirect.com

Aneurysms associated with arteriovenous malformations (AVMs) are well represented in the literature. Their exact etiology is poorly understood, but likely global hemodynamic changes coupled with vascular wall pathology play into their formation. Flow-related and intranidal aneurysms, in particular, appear to have an increased risk for hemorrhagic presentation. Treatment strategies for these aneurysms are particularly challenging. We report the case of an AVM-associated aneurysm causing intraventricular hemorrhage that disappeared after embolization of unrelated, distal feeding pedicles to the nidus, at a site distant from the aneurysm. We also review the literature with regards to these so-called "disappearing aneurysms" in the context of AVMs and other vascular pathologies.

Intraventricular hemorrhage associated with arteriovenous malformations (AVMs) has been reported previously in the literature and can be a presenting symptom secondary to direct AVM hemorrhage into the ventricle or secondary to ruptures of aneurysms associated with AVMs (17, 22). Aneurysms associated with AVMs are classified depending on their relationship to the AVM nidus and/or feeder vessels (10, 11, 14). They are considered weak points within the vessel wall that increase the risk of intracranial hemorrhage. Hemodynamic changes, such as increased blood flow or changes in intravascular pressure, are associated with rupture of these AVM-associated aneurysms. Treatment strategies for these aneurysms are particularly challenging (25). Here, we report the case of an AVM-associated aneurysm causing intraventricular hemorrhage that disappeared after subsequent embolization of the nidus from pedicles not directly feeding the aneurysm. We also review the literature with regards to these so-called "disappearing aneurysms" in the context of AVMs and other vascular pathologies.

CASE REPORT

A 57-year-old female with a known diagnosis of a large left parietal AVM presented with acute onset of headache, nausea, and photophobia. Noncontrasted computed tomography head imaging demonstrated intraventricular hemorrhage (IVH), left greater than right, without evidence of subarachnoid hemorrhage (Figure 1A). The patient's initial diagnosis occurred more than 30 years prior, a time when there were no endovascular treatment options. Open treatment options were thought too morbid given the size of the lesion and its proximity to the post–central gyrus and visual cortex. The patient was last seen by the senior author 10 years before her current hospital presentation and had been routinely followed by her outpatient neurologist. The patient also had a history of seizure disorder well controlled with a single agent. She had no known history of hemorrhage from this AVM.

The day after admission, the patient underwent digital subtraction angiography (DSA), which demonstrated a 5.4 cm × 5.2 cm × 5.8 cm AVM within the left posterior parietal lobe with predominant feeders from the left anterior cerebral artery (ACA), middle cerebral artery (MCA), and posterior cerebral artery (PCA) with marked dilation of the ACA and MCA feeders in particular (Figure 2). There was predominant drainage via the superior sagittal sinus and Labeé vein with a smaller portion of venous outflow from the deep venous system. Functional magnetic resonance imaging demonstrated that motor and speech areas were anterior to the nidus, and there was no involvement of the occipital lobe. Given these findings, the patient was deemed to have a Spetzler-Martin grade III AVM (21).

The patient was monitored for development of hydrocephalus and ultimately discharged to inpatient rehabilitation 10 days after presentation. Discussion in our institution's neurovascular conference recommended staged embolization treatment followed by surgical resection.

Two weeks after discharge, while awaiting her first staged embolization, the patient re-presented with acute-onset severe headache. Repeat computed tomography imaging again demonstrated acute IVH, this time confined to the left ventricle (Figure 1B). DSA demonstrated similar

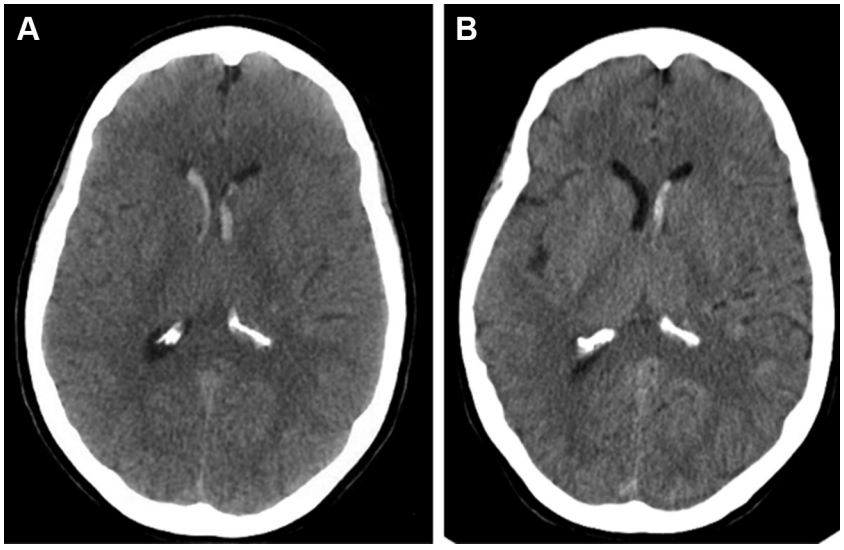

Figure 1. Axial-view, noncontrasted computed tomography (CT) head images from the patient. (**A**) Initial presentation with bilateral intraventricular hemorrhage (IVH), more in the left than right ventricle. The patient was medically managed and observed for signs of hydrocephalus. Her last CT before discharge showed resolution of the IVH. (**B**) Two weeks after discharge, the patient re-presented with acute-onset headache and hadleft-only acute IVH.

arterial flow patterns when compared with prior angiography. On 3D-DSA, a 5-mm anterior choroidal artery aneurysm, anatomically near the atria of the left ventricle, was found. Careful review of this second DSA showed dysplastic features of the left anterior choroidal artery that provided small pedicle feeders to the AVM. The aneurysm was located distally on the left anterior choroidal artery, some distance from the vessel origin off the MCA. On review of the patient's first DSA, this aneurysm was previously present but smaller and partially obscured, likely secondary to thrombosis (**Figure 2**). The periventricular location of this aneurysm was likely the source of the patient's recurrent IVH. Despite providing feeders to the AVM nidus, this left anterior choroidal artery was diminutive in caliber with a tortuous course. There was concern that any attempts to access the vessel and potentially treat this distal aneurysm would lead to vessel dissection or perforation and devastating associated stroke. Thus the decision was made to target the AVM nidus instead. The patient subsequently underwent AVM embolization of a distal M3 branch supplying the pre-Rolandic region with Onyx-18 (ev3, Plymouth, Minnesota, USA) resulting in approximately 40% reduction in nidus size.

Three days later, a follow-up DSA was performed to further delineate the distal left anterior choroidal aneurysm with possible treatment. Multiple magnified views of the area of interest failed to demonstrate continued presence of this aneurysm (**Figure 3**). The patient was ultimately discharged to inpatient rehabilitation 2 weeks after her second IVH.

Two months later, second-stage Onyx embolization was undertaken via left MCA posterior branch feeders, resulting in another 10%–20% reduction in nidus size. Multiple views of the distal left anterior choroidal artery again did not demonstrate presence of the previously seen aneurysm. A third-stage Onyx embolization was undertaken almost 6 months after the initial hemorrhage and again did not demonstrate evidence of this aneurysm. The aneurysm has remained resolved without direct treatment of the aneurysm. The patient is currently anticipated to undergo craniotomy for AVM resection.

DISCUSSION

The incidence of AVM-associated aneurysms has been quoted in the literature as ranging from 2.7%–51.5% (10, 11, 15, 20, 22). Our internal review over the past 18 months for AVMs undergoing DSA indicates an associated aneurysm rate of 13%; 71% of these aneurysms were type II Perata et al. (14) and type IIa Redekop et al (19), and the remainder were type III Perata et al. (14) and type IIb Redekop et al (19). None were like our currently described aneurysm. None of our aneurysms regressed with direct AVM nidal treatment as the primary treatment (internal unpublished data). It is hypothesized that hemodynamic stress secondary to the high-flow nature of AVMs likely causes continual damage to the endothelia of blood vessels, leading to aneurysm formation, and over time these weakened blood vessels have a propensity to rupture (1, 7, 10, 16, 25). To our knowledge, this is the first reported case of resolution of ruptured AVM-associated aneurysm after treatment of nidus from unrelated, distant arterial feeders.

Classification Systems

Various classification systems exist to characterize AVM-associated aneurysms, though a definitive system is still lacking given the uncertainty regarding the actual etiology of these aneurysms (10, 11, 14, 19). The first was proposed by Lasjaunias et al., who described 3 types of AVM-associated aneurysms: 1) distal or intralesional, 2) proximal aneurysms directly on vessels supplying the AVM, and 3) remote or dysplastic aneurysms unrelated to inflow vessels. Further refinement of this classification separates whether "flow related" aneurysms originated from vessels off the circle of Willis that directly supplied the AVM or originated more distally in the feeding pedicle (**Table 1**) (14, 19). Our patient's aneurysm is difficult to classify on the basis of the existing systems as the anterior choroidal in our patient arose off the ICA/MCA junction, the MCA was a feeder vessel to the AVM, but the anterior choroidal was not a feeder to the AVM. It is not entirely "unrelated" to the AVM according to the classification systems; however, it is not truly a "distal pedicle" aneurysm, either. It shares features