

Spontaneous Cystic Arteriovenous Malformation—A Novel Treatment Alternative

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Background: Brain arteriovenous malformation (BAVM) is a life-threatening vascular congenital malformation due to the risk of intracerebral hemorrhage. The formation of a spontaneous cyst within a BAVM, without history of hemorrhage or gamma knife surgery, is rare and has an unknown pathophysiology. We suggest a novel theory of spontaneous cystic BAVM formation, subsequently treated with endovascular embolization followed by surgery with a favorable long-term outcome. *Methods:* Review of the literature using PUBMED database and comparison between clinical presentation, diagnostic imaging studies, and treatment options. *Results:* The high flow causing a shear stress over the drainage vein may be responsible for venous ectasia and thus incrementing hydrostatic intranidal pressure causing a serous effusion that develops into a pseudocyst. We hypothesize that hemodynamic factors may be responsible for both origin and growth of such cystic lesions. *Conclusions:* Endovascular treatment allowed us to achieve cyst volume reduction by managing its unique flow characteristics. To our knowledge, this is the first case report of combined treatment with endovascular and surgical approach of a cystic BAVM. **Key Words:** Intracranial arteriovenous malformations—angiography—digital subtraction—brain cyst—embolization—therapeutic.

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Introduction

Brain arteriovenous malformation (BAVM) is a threatening vascular anomaly with a 3% annual risk of intracerebral hemorrhage.^{1,2} BAVM can present with epilepsy, chronic headache, and progressive neurological focal

deficits as well.³ The estimated prevalence is .1%-4% in the general population. About 90% are localized in supratentorial structures as a single mass-occupying lesion.^{4,5} BAVMs are also responsible for 1%-2% of strokes and 9% of all subarachnoid hemorrhage.⁶

The angioarchitecture shows single or multiple direct connections between arteries and veins without an interposed capillary network, usually associated with gliosis and calcification of the surrounding brain tissue.⁶ A variety of flow-related phenomena such as the presence of nidus or venous aneurysms, as well as the amount or localization of vein drainage, increase the bleeding risk up to 25%.^{7,8}

Cyst formation is a well-known complication following BAVM's gamma knife surgery (GKS) with an incidence of 1.6%.⁹ Spontaneous cyst formation, without previous history of hemorrhage or GKS, is a rare presentation with an unknown pathophysiology. The lack of an epithelial wall defines these lesions as a pseudocyst, containing a serous or serohemetic fluid with fibrosis, capillaries, and hemosiderin deposits over the cyst's border zone.

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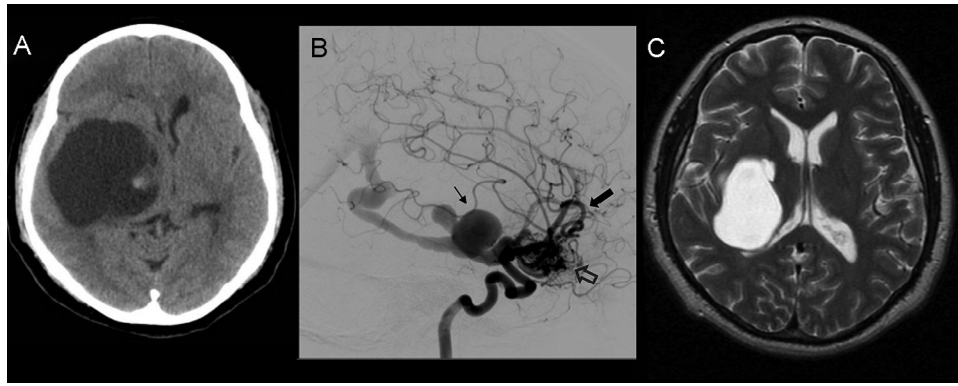


Figure 1. (A) Noncontrast CT: large hypodense space occupying lesion with CSF-like content. 13-mm midline shift. (B) DSA. Lateral view. Large plexiform AVM with main afferent from anterior cerebral artery (black arrow) and secondary afferent from fronto-basal branches on the MCA. Nidus volume smaller than 6 cm (empty arrow). Voluminous venous varices (small arrow) and drainage into a dilated vein of Galen. (C) MRI T2-WI. Follow-up before surgery. Cyst volume reduction and resolution of the midline shift after the endovascular embolization. Abbreviations: AVM, arteriovenous malformation; CSF, cerebrospinal fluid; CT, computed tomography; DSA, digital subtraction angiography; MCA, middle cerebral artery; MRI, magnetic resonance imaging.

Most of the symptoms are due to the progressive growth and mass effect over encephalic structures, with seizures and focal progressive neurological deficits as the main presentation. Cystic BAVMs have been reported as an infrequent finding that requires surgical resolution, although there is no evidence of the natural history, bleeding risk, long-term outcomes, or alternative treatments.¹⁰⁻¹⁴

The most common options to achieve BAVM occlusion include conventional surgery, endovascular embolization, and GKS as well as a combination between those techniques. Endovascular embolization is a proper option for deep, small BAVMs with few feeders. The objective is to reduce the arterial inflow to decrease the capillary pressure that lowers the bleeding risk. BAVMs with a Spetzler & Martin (S&M) score ≥ 3 require prior embolization to reduce the bleeding risk during the surgical procedure.¹⁵

Case Description

A 40-year-old man with history of arterial hypertension and no family history of cardiovascular or cerebrovascular diseases was admitted presenting 2 months of mild headaches, progressive left-sided weakness, and left homonymous hemianopia. A noncontrast computed tomography of head demonstrated a large hypodense, cystic-like mass-occupying lesion with an approximate volume of 147 mL with a nodular hyperdense lesion inside (Fig 1, A). The digital subtraction angiography showed a deep, plexiform, right frontotemporal BAVM with middle cerebral artery feeders, and multiple intranidal aneurysms with Galen vein drainage, S&M grade 3 because of the medium size between 3 and 6 cm, the location in a noneloquent area, and the deep venous drainage (Fig 1, B). A partial endovascular embolization using ONYX (eV3 Endovascular, Plymouth, MN) was performed, with immediate inflow reduction. There was worsening of symptoms in the first 3 days after the procedure followed by improvement in

the left-sided weakness in the next few days. A month later, a second embolization was performed, achieving a near complete obliteration of the feeders with minimal arteriovenous malformation (AVM) inflow. The patient had a good clinical and functional recovery with persistent left homonymous hemianopia. A brain magnetic resonance imaging 7 months after the second endovascular embolization demonstrated a significant reduction in the cyst size and a complete resolution of the midline shift (Fig 1, C). Conventional open neurosurgery was performed with the excision of a deep AVM surrounded by a cerebrospinal fluid-filled cavity. The biopsy study reported multiple sized vascular-like structures filled with black synthetic material (ONYX) and multinucleated giant cells (Fig 2). Five years later, the patient remained with incomplete left homonymous hemianopia without recurrence of neurological symptoms.

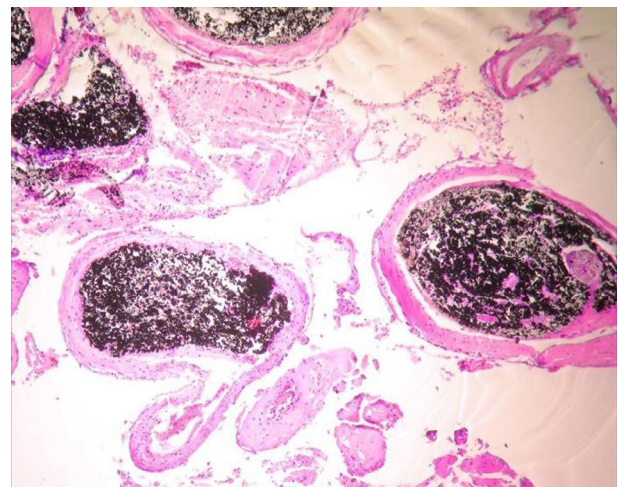


Figure 2. BAVM histopathology (hematoxylin and eosin): Multiple disorganized vascular structures containing Etinil-Vinil-Alcohol (ONYX) surrounded by fibrous tissue. Abbreviation: BAVM, brain arteriovenous malformation.

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