

Spontaneous Thrombosis of a Residual Arteriovenous Malformation in Eloquent Cortex after Surgery: Case Report

Bernard R. Bendok, M.D., Christopher C. Getch, M.D.,
Mir Jafer Ali, B.A., Todd Parish, Ph.D., H. Hunt Batjer, M.D.

Departments of Neurological Surgery (BRB, CCG, MJA, HHB) and Radiology (TP),
Northwestern University Medical School, Chicago, Illinois

OBJECTIVE AND IMPORTANCE: The presence of a residual arteriovenous malformation (AVM) on postoperative angiograms is typically an indication for prompt return to the operating room to complete resection, because of the risk of early hemorrhage. This approach, however, may involve risks of neurological deficits when the residual AVM is in eloquent cortex. We present a case of complete thrombosis of a residual AVM after surgery. This residual AVM tissue was located in eloquent cortex. Complete spontaneous thrombosis of residual AVMs after surgery has only rarely been reported. This phenomenon raises questions regarding the most appropriate management for residual AVMs in eloquent cortex.

CLINICAL PRESENTATION: The patient was a 43-year-old, right-handed, male patient with an AVM centered in the left precentral gyrus. The patient presented with medically intractable seizures and progressive right hemiparesis. After AVM resection, angiography revealed a residual AVM with early venous drainage. Angiography performed 1 week later demonstrated a persisting AVM nidus without early venous drainage. Angiography performed 3 months later demonstrated complete thrombosis of the residual AVM.

INTERVENTION: The patient has been monitored for more than 1 year, without additional symptoms or therapy.

CONCLUSION: We continue to advocate prompt return to the operating room when postoperative angiography reveals a residual AVM with persistent shunting. When the residual AVM is in eloquent cortex and is small, with a single draining vein, however, observation of the patient (with strict blood pressure control) and repeat angiography after 1 week represent an alternative strategy that is supported by this case report. As this case demonstrates, it is possible for small residual AVMs to thrombose. This may avert the need for reoperation for residual AVMs in eloquent cortex, with the potential for neurological deficits.

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Key words: Eloquent cortex, Residual arteriovenous malformation, Thrombosis

The exact natural history of residual arteriovenous malformations (AVMs) after partial resection is not clearly known. Anecdotal evidence suggests that

early postoperative hemorrhage is associated with residual AVMs (2, 4). This early risk may be attributable to the presence of persistent high flow through the nidus in

the setting of surgically induced venous outflow impairment. Most neurosurgeons, therefore, advocate immediate return to the operating room for resection of any residual AVMs observed on postoperative angiograms. This has been our approach, in fact. If the residual AVM observed on postoperative angiograms is in confirmed eloquent cortex, however, a dilemma arises, i.e., resecting the residual AVM increases the likelihood of deficits, whereas not treating the residual AVM may result in hemorrhage. A conservative approach may entail repeating angiographic evaluations within several days, to determine whether thrombosis has occurred. The frequency with which thrombosis of AVMs occurs after partial resection is not known. There are three reports in the literature of spontaneous thrombosis of residual AVMs after partial resection (1, 2). To this limited documented experience with this phenomenon we add another case. This case raises questions regarding the most appropriate way to manage residual AVMs in eloquent cortex after AVM resection.

CASE REPORT

The patient was a 43-year-old, right-handed, male patient with a 5-cm-diameter AVM centered in the left precentral gyrus. The AVM was initially diagnosed at another institution in the early 1980s, during a seizure evaluation. The AVM was partially embolized, with open and endovascular techniques, at that time. The patient was left with dense paresis of his right arm, which partially improved with time. The patient was left with a known residual AVM. The patient was referred to our center approximately two decades after his initial treatment, because of his refractory seizure disorder (with several episodes each day of partial seizures on the right side) and progressive right hemiparesis. The seizures occurred daily despite the use of four antiepileptic medications. During our evaluation, the patient was noted to have significant right hemiparesis, which was greater in his arm than in his leg. The patient also exhibited mild right facial weakness and

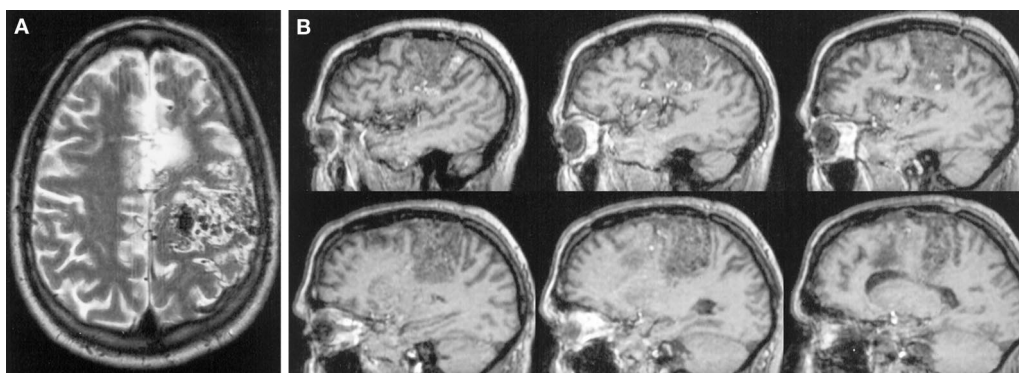


FIGURE 1. Preoperative T2-weighted axial (A) and T1-weighted sagittal (B) magnetic resonance imaging scans.

mild expressive aphasia. Magnetic resonance imaging (Fig. 1) and angiography (Fig. 2) revealed a 5- × 4- × 4-cm AVM centered in the left precentral gyrus. The AVM was fed by multiple branches of the middle cerebral artery and the anterior cerebral artery, with some supply from lenticulostriate branches. The AVM drained to the superior sagittal sinus via two cortical veins. Before surgery, the patient underwent three embolization sessions with *n*-butylcyanoacrylate. During the first embolization (14 d before surgery), a middle cerebral artery branch was embolized with coils and glue. During the second embolization (12 d before surgery), embolization of more middle cerebral artery branches was achieved with glue. During the third embolization (8 d before surgery), an anterior cerebral artery branch was embolized with glue. Preoperative functional magnetic resonance imaging findings suggested that the motor cortex was the gyrus immediately posterior to the posterior margin of the AVM (Fig. 3). During surgery, leptomeningeal collateral vessels were noted in

the motor cortex, but the AVM nidus itself was clearly demarcated from the precentral gyrus. The posterior dissection plane was pristine, and the senior author had no suspicion that a residual AVM could be located posteriorly. No unusual bleeding was observed in this area. The patient awakened very well, with minimal enhancement of his baseline neurological deficit. During surgery, what appeared to be a leptomeningeal collateral vessel along the motor cortex was left in place. On postoperative Day 1, angiography revealed a residual AVM nidus at the posterior margin of the resection cavity, with drainage into the vein of Trolard (Fig. 4). At that time, several options were contemplated, as follows: 1) a return to the operating room for AVM resection, 2) radiosurgical treatment of the residual AVM, 3) embolization of the residual AVM, or 4) close observation in the intensive care unit with blood pressure control, with plans to repeat angiographic assessments in 1 week.

Resection of the AVM was considered by the team to involve a high risk of

neurological deficits. There were several reasons why we suspected that the posterior aspect of the AVM was intimately associated with the motor cortex. First, the patient had presented with progressive right hemiparesis. Second, the equivalent area in the contralateral hemisphere was activated with left body movement (Fig. 3). The fact that there was no activation near the AVM with right hand motion could have been a function of the flow dynamics in the AVM.

Embolization was considered by our interventional specialist not to be an option. Because the residual nidus was thought to be in the motor cortex, we decided to monitor the patient, with strict blood pressure control (100/60 to 120/80 mm Hg). Because the patient had already been extubated and was faring well neurologically before an-

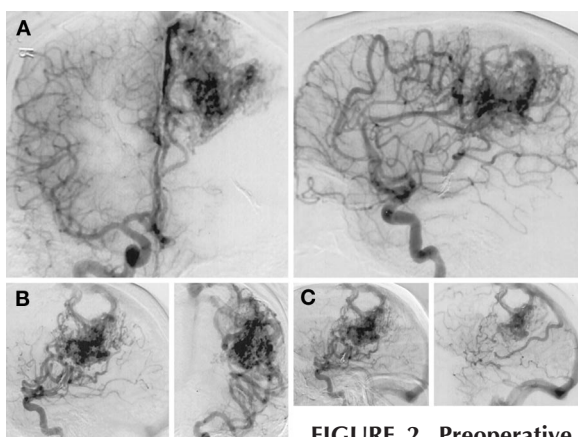


FIGURE 2. Preoperative early arterial-phase (A), late arterial-phase (B), and venous-phase (C) angiograms.

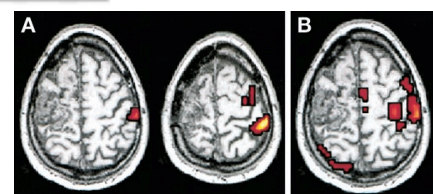


FIGURE 3. Functional magnetic resonance imaging scans. A, scans demonstrating displacement of right hand function to the right hemisphere. B, testing of left hand function, demonstrating the motor cortex in the right hemisphere. In a comparison of the sides, it was suggested that the posterior AVM most likely involved the gyrus just anterior to the motor cortex. No left hemisphere activation was achieved with right body function, which might have been attributable to the high level of blood flow through the AVM.

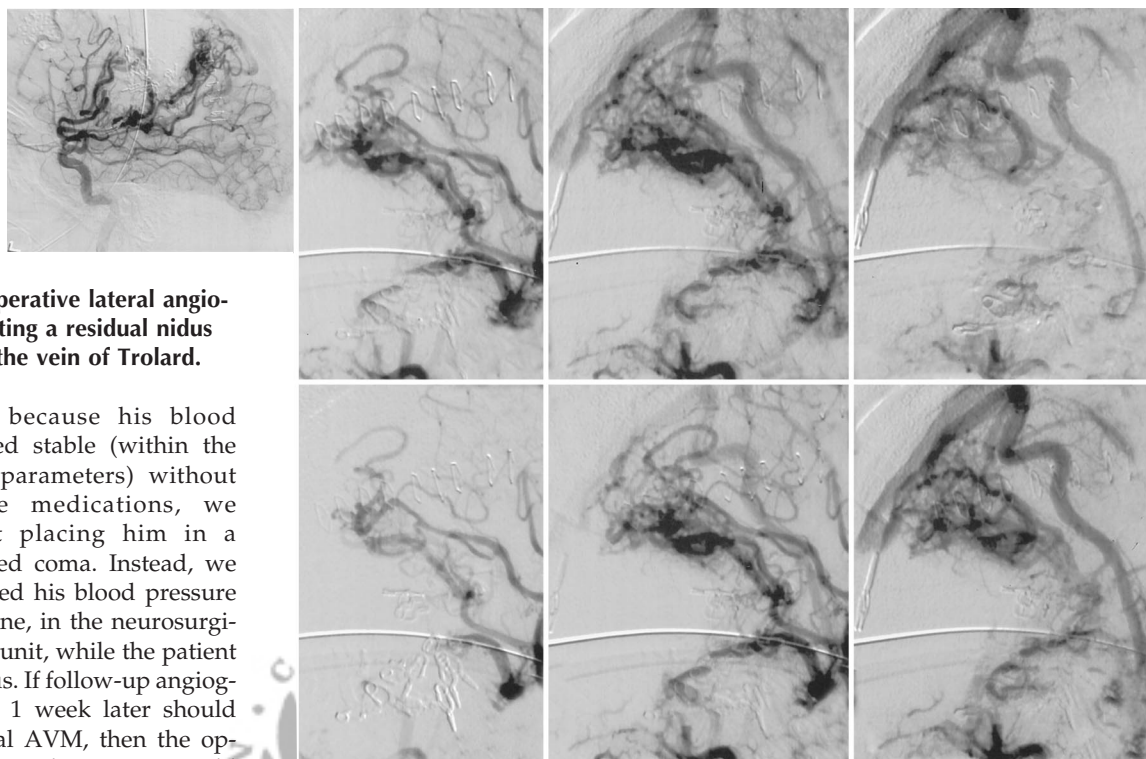


FIGURE 4. Postoperative lateral angiograms, demonstrating a residual nidus draining through the vein of Trolard.

giography and because his blood pressure remained stable (within the aforementioned parameters) without antihypertensive medications, we decided against placing him in a barbiturate-induced coma. Instead, we carefully monitored his blood pressure with an arterial line, in the neurosurgical intensive care unit, while the patient was kept conscious. If follow-up angiography performed 1 week later should indicate a residual AVM, then the options of surgery or radiosurgery would be considered.

Eight days after surgery, residual abnormal vessels were still observed but the early venous drainage was absent (Fig. 5). The vein of Trolard filled during the venous phase. The patient underwent follow-up angiography 3 months after surgery (Fig. 6), which demonstrated resolution of the abnormal vessels and the absence of early venous drainage. All angiography was performed by an interventional neuroradiologist with significant experience in the angiographic diagnosis and endovascular treatment of AVMs.

DISCUSSION

A recent review of the world literature by Abdulrauf et al. (1) revealed

only 24 cases of spontaneous angiographic obliteration of AVMs. Of these cases, only three involved patients who had undergone partial resection of their AVMs. The characteristics of those three patients, as well as ours, are presented in Table 1. The time to known angiographic obliteration for these four patients ranged from 3 months to 8 years. We demonstrated the absence of early venous drainage 8 days after surgery and the complete resolution of abnormal vessels 3 months after surgery. All four residual AVMs were classified as small by the treating surgeons. The AVMs were all associated with one draining vein, and three of the four appeared to have had limited arterial supply. The natural history of residual

AVMs after surgery has been reported to be associated with subsequent hemorrhage (2). For our patient, a residual early draining vein associated with the persisting nidus, which was observed on postoperative Day 1, was noted to fill only during the venous angiographic phase 8 days later. In the angiographic evaluation performed 3 months later, the abnormal vasculature was observed to be spontaneously thrombosed. A significant amount of angiographic experience is needed to skillfully perform angiography and interpret the results after AVM resection. Abnormal vasculature surrounding AVMs can be confused with residual AVMs, and prolonged injection times can give the false appearance of an early draining vein. Our patient's angiograms were obtained and interpreted by interventional neuroradiologists who are involved in diagnosing and treating more than 30 AVMs each year.

Thrombosis of the residual AVM for our patient raises questions regarding the management of residual AVMs in eloquent cortex immediately after surgery and the natural history of spontaneously thrombosed residual AVMs.

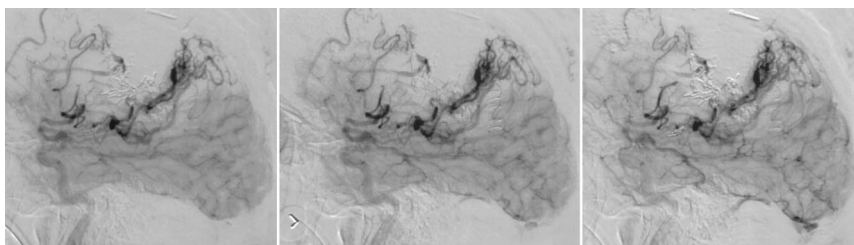


FIGURE 5. Angiograms obtained 8 days after surgery, showing the absence of early venous drainage but the persistence of abnormal vessels.

TABLE 1. Features of Arteriovenous Malformations Documented to Have Thrombosed after Partial Resection^a

Series (Ref. No.)	Age (yr)/ Sex	Presentation	Location	No. of Feeding Arteries	No. of Draining Veins	Interval to Known Thrombosis
Abdulrauf et al., 1999 (1)	58/F	SAH, IVH	Cerebellar	2	1 (transverse sinus)	5 mo
Abdulrauf et al., 1999 (1)	25/M	ICH	Cerebellar	Not noted	1	5 yr
Lakke, 1970 (3)	33/F	ICH	Frontal	1	1	8 yr
Bendok et al., 2002 (present report)	43/M	Seizures	Parietal	Small abnormal vessels	1 (vein of Trolard)	3 mo

^a SAH, subarachnoid hemorrhage; IVH, intraventricular hemorrhage; ICH, intracerebral hemorrhage.

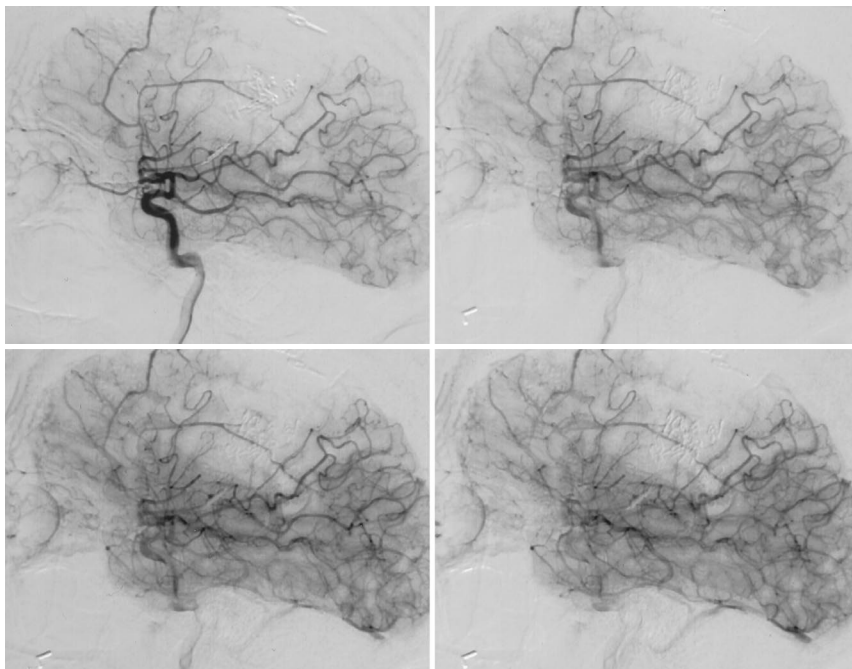


FIGURE 6. Angiograms obtained 3 months after surgery, demonstrating the absence of both abnormal vessels and early venous drainage.

Although in general we still advocate prompt return to the operating room for resection of residual AVMs, we question this policy for small residual AVMs in eloquent cortex that are associated with a single draining vein. If thrombosis is demonstrated on subsequent angiograms for such patients, they must be monitored for several years, to confirm the absence of further natural history risk.

ACKNOWLEDGMENTS

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COMMENTS

This case demonstrating spontaneous thrombosis of a residual arteriovenous malformation (AVM) after subtotal surgical resection raises an important question: what should be done with a residual AVM that lies in eloquent cortex? A residual AVM poses a significant danger of rehemorrhage, particularly when draining veins have been occluded and venous outflow has been compromised. Therefore, prompt return of the patient to the operating room for the resection of a residual AVM is advocated. This case, although interesting and unusual, should not, in my opinion, alter the practice of aggressively treating patients with residual AVMs. Neither small nidus size nor a single draining vein justifies a conservative approach, because there are data associating these factors with increased hemorrhage risk. Certain eloquence of adjacent brain should give the surgeon pause, but in these cases I would still be inclined to operate or to treat the residual AVM with radiosurgery.

Michael T. Lawton

San Francisco, California

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Reprint requests: H. Hunt Batjer, M.D., Department of Neurological Surgery, Northwestern University Medical School, 233 East Erie Street, Suite 614, Chicago, IL 60611. Email: hbatjer@nmff.org

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Bendok et al. report a case of what they interpret as a residual AVM that spontaneously thrombosed with conservative management. I completely agree with the way that this case was handled. I think that this case may well fit into the case series that my colleagues and I recently described (1). It seems in *Figure 4B* that there was a prolonged arterial

injection, because there is blood in the feeding arteries leading to the malformation and well out into the venous phase of the angiogram. This prolonged injection may falsely lead to the appearance of an early draining vein. In performing immediate postoperative angiography, it is important that contrast be administered as a short bolus and that the dye's transit through the circulation be monitored carefully to see whether there is an early draining vein. Hyperemia after AVM resection sometimes reduces the transit time. If the injection technique is not perfect, then the angiographic results may occasionally be difficult to interpret properly.

An angiogram obtained 1 week after surgery clearly shows resolution of the residual abnormal vessels. This finding is in accord with my own experience. I agree with the authors' assumption that the resection of all abnormal vessels, especially if injury to critical functional cortex is required, may not be in the patient's best interest. It is evident that, in many cases, residual abnormal vessels left behind after AVM resection spontaneously regress and disappear. Questions remain unanswered, however. For example, what are the criteria that need to be applied to direct further resection of abnormal vessels seen on

intraoperative angiograms or on an immediate postoperative angiogram? Further thoughtful analysis of clinical experiences such as the one presented in this report will help to answer such questions.

Robert A. Solomon
New York, New York

1. Solomon RA, Connolly ES Jr, Prestigiacomo CJ, Khandji AG, Pile-Spellman J: Management of residual dysplastic vessels after cerebral arteriovenous malformation resection: Implications for postoperative angiography. *Neurosurgery* 46:1052-1062, 2000.

The authors report an interesting patient with a formidable parietal AVM that was thought to have been resected completely. The routine postoperative angiogram, however, demonstrated persistent filling and shunting of the motor lesion. The authors elected to continue observation of the patient in the intensive care unit without returning to the operating room, for fear of producing a neurological deficit. Follow-up angiography performed 1 week after surgery demonstrated shunting without early venous drainage. Continued observation revealed complete thrombosis of the remaining segment of the AVM.

The finding in this case highlights an interesting feature of AVM compartmentalization. Although the authors "got away" with this strategy, it is not recommended as a general strategy for managing a residual lesion after AVM resection, as the authors themselves point out. When one studies immediate postoperative angiograms, a number of unusual features can be identified that later resolve. When large AVMs with collateral supply from cortical feeding vessels are resected, the small feeding vessels can produce a blush around the area of AVM resection. In patients in whom there is no early venous drainage, my colleagues and I have observed a number of these abnormalities and then restudied them approximately 1 week after surgery, only to find that the blush has resolved. The fear in these situations is that a small segment of the AVM has been disconnected from the venous drainage and therefore remains at high risk for hemorrhage. If this is the case, the authors' strategy of maintaining relative hypotension is to be followed. The authors have demonstrated excellent management of this very difficult situation.

Christopher S. Ogilvy
Boston, Massachusetts



Ebola virus.

