

## Case Report

# Total temporary occlusion of blood flow for several hours to treat a giant deep arteriovenous malformation: A series of multiple operations to save a young life

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## Abstract

**Background:** The treatment of giant deep arteriovenous malformations (AVMs) remains challenging.**Case Description:** We report a case of giant deep AVM diagnosed in a 9-year-old girl, for whom the AVM rupture occurred 9 years later. At the age of 9, the girl developed mild left hemiparesis. Magnetic resonance imaging revealed a giant deep AVM. The patient underwent one course of stereotactic radiotherapy followed by serial imaging. At the age of 18, we admitted her to our department with left hemiparesis and a loss of consciousness. Computed tomography showed intracerebral hemorrhage related to AVM. The treatment process proved challenging, with recurrent intracerebral hemorrhages. During the second operation, we used total temporary occlusion for almost 4 hours. Eventually, after 4 rounds of embolizations, 4 microsurgical operations, and a month-and-a-half after admission, AVM was successfully occluded. Five years after this treatment, the patient regained the ability to walk without assistance, although a moderate disability with visual changes remained (Modified Rankin Scale score 3).**Conclusion:** This case illustrates that the cumulative risk of rupture of a high-grade AVM in young patients is evident, while treatment may prove successful with satisfactory results.**Key Words:** Arteriovenous malformation, deep AVM, intracerebral hemorrhage, microneurosurgery

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## BACKGROUND

Intracranial arteriovenous malformation (AVM) is a tangle of abnormal blood vessels with no intermediary capillary system. A vast majority of all lesions are thought to have a congenital nature, however, at present, some authors report noncongenital cases.<sup>[1,7,10]</sup> 45–72% of the cases manifested with hemorrhage, and go seizures with 18–35% of the cases. Less common symptoms are chronic headaches and focal neurological deficits with 6–14 and 3–10% of the cases, respectively.<sup>[9]</sup>

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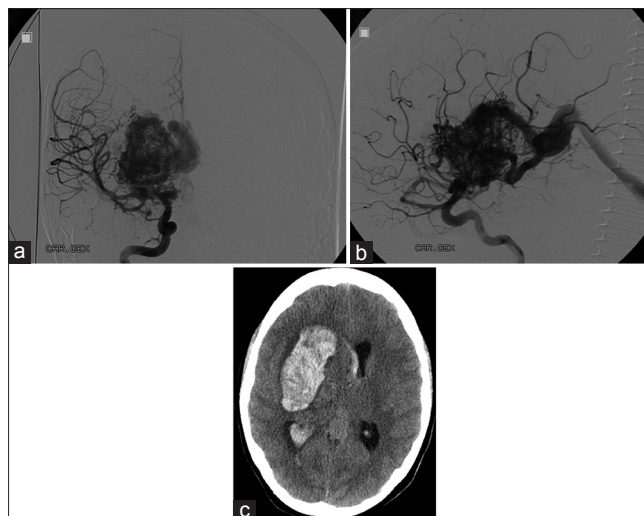
AVM is considered to be giant when nidus is 6 cm or more in maximum diameter.<sup>[2]</sup> Giant AVMs constitute a small percentage of all AVMs. At the same time, they are complex due to a higher risk of morbidity and mortality compared with the smaller ones. Giant AVMs located in the deep brain area (thalamus and basal ganglia) are still difficult to treat. They typically have high-flow, deep venous drainage, and extend into eloquent areas. These features associated with higher rates of postoperative complications even in young, otherwise healthy patients.<sup>[11,14]</sup> Spetzler–Martin grades IV–V AVMs are the most difficult for microsurgical removal. For such AVMs, personalized treatment is necessary and life expectancy should be compared with a cumulative risk of rupture. Combined therapies provide an effective way to obliterate the AVM completely.<sup>[2]</sup> The use of preoperative Onyx embolization might decrease surgical complications.

Ruptured AVM associated with a large intraparenchymal hematoma needs urgent surgical intervention. In such a case, surgery is a life-saving procedure, and the main aim is removal of hematoma. Resection of AVM depends upon patient's condition, the complexity of the AVM, and surgeon's and department's experience. We present one of the cases with unique staged multidisciplinary treatment. Outcomes were assessed with the Modified Rankin Scale (mRS) score after the operation and at late follow-up.

## CASE DESCRIPTION

The 9-year-old otherwise healthy girl presented with minimal physical clumsiness and progressive mild left hemiparesis. Magnetic resonance imaging (MRI) revealed a giant deep AVM. This lesion was considered a grade-V AVM according to Spetzler–Martin scale. Surgical treatment of the AVM was declined as it was too risky. Radiosurgical treatment was instituted due to the size and location of the AVM. She received one course of radiotherapy (20 Gr) resulting in a slight reduction in the size of the AVM. Last follow-up digital subtraction angiography (DSA) at the age of 12 showed no change in AVM [Figure 1a and b]. At the age of 18, she admitted to our hospital with loss of consciousness and left hemiparesis (mRS score 5). A computed tomography (CT) scan revealed large intracerebral hemorrhage [Figure 1c] due to ruptured arteriovenous malformation.

The decision on the surgical treatment of the AVM was made with a help of endovascular embolization. Onyx embolization was admitted with minor success. Treatment continued with microsurgery. The patient was taken to the operative theater for the removal of intracerebral hematoma and resection of the AVM. During the evacuation of hematoma major intraoperative bleeding (1 L) occurred from Onyx embolized intranidal aneurysm. Bleeding was effectively held by emergency



**Figure 1: Angiographic images: digital subtraction angiography (DSA) (posterioranterior view) (a) and DSA (lateral view) (b) 6 years before the AVM rupture; preoperative axial computed tomography (c) large basal ganglia-putamenal intracerebral hemorrhage**

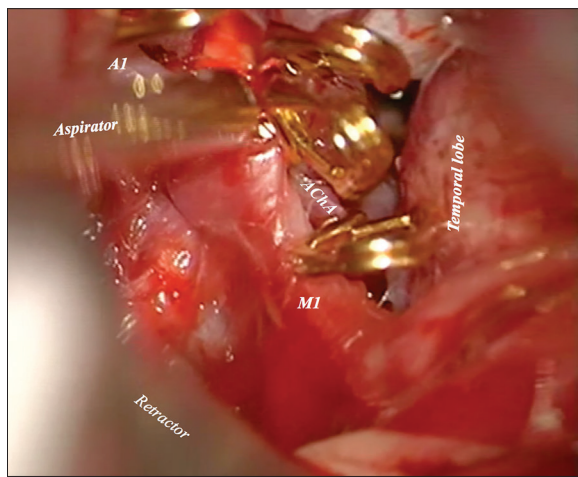
clipping of some feeders originating from the middle cerebral artery (MCA). Resection of the AVM could not be performed because of the unstable medical conditions of the patient.

Postoperatively, the patient clinical condition was the same neurologically as during admission. However, 3 days after the first operation, the patient's condition deteriorated suggesting intracranial hypertension. The patient underwent a placement of external drainage catheter with the intracranial pressure monitor. Intracranial hypertension was eliminated. Further treatment continued with a new round of endovascular modality. A new attempt of Onyx embolization through the posterior cerebral artery (PCA) was made. On the same day, the patient underwent a second microsurgical operation using the same craniotomy. We used total temporary clipping under mild hypothermia (33°C) for almost 4 hours; P2 segment of PCA, A1 segment of anterior cerebral artery, anterior choroidal artery (AChA), and the supraclinoid segment of internal carotid artery (ICA) [Figure 2]. Most of the lateral part of AVM underwent removal [Figure 3a]. A postoperative angiogram demonstrated remaining medial and some of the lateral parts of AVM [Figure 3b]. MRI with diffusion-weighted imaging (DWI) performed next day showed a lack of global ischemia [Figure 3c]. The next day the patient underwent the third operation using the same craniotomy. Remaining part of the medial AVM was completely removed, and only a small lateral part was left.

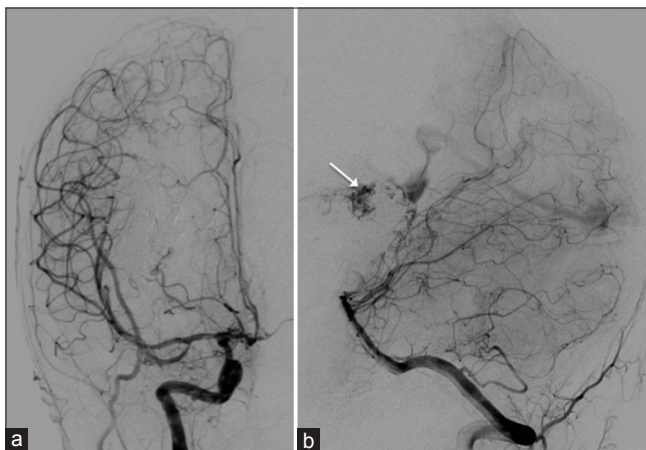
Three weeks after the third operation, shunt for cerebrospinal fluid diversion was inserted, and the patient remained conscious with left hemiparesis. The patient was transported to the department of neurology

for rehabilitation. However, few days later, new minor bleeding occurred, adjacent to the residual part of the AVM. Two embolizations were performed through MCA and PCA. Residual part filling via MCA was completely occluded, however, small residual filling via PCA remained [Figure 4a and b].

Shunt for cerebrospinal fluid diversion was removed. Ten days later, the patient underwent the last microsurgical operation. Eventually, AVM was completely occluded [Figure 5a-c]. The patient was discharged with strong left hemiparesis, visual changes, memory difficulties, and aphasia (mRS score 4). These symptoms gradually improved, and 5 years after her initial surgery, she could walk without assistance, however, visual changes and memory difficulties remained (mRS score 3) [Figure 6].



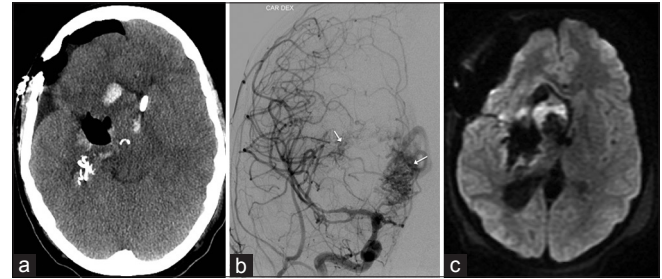
**Figure 2:** Intraoperative photography during second operation: Total temporary clipping – AI, supraclinoid segment of internal carotid artery and anterior choroidal artery (P2 segment not visualized)



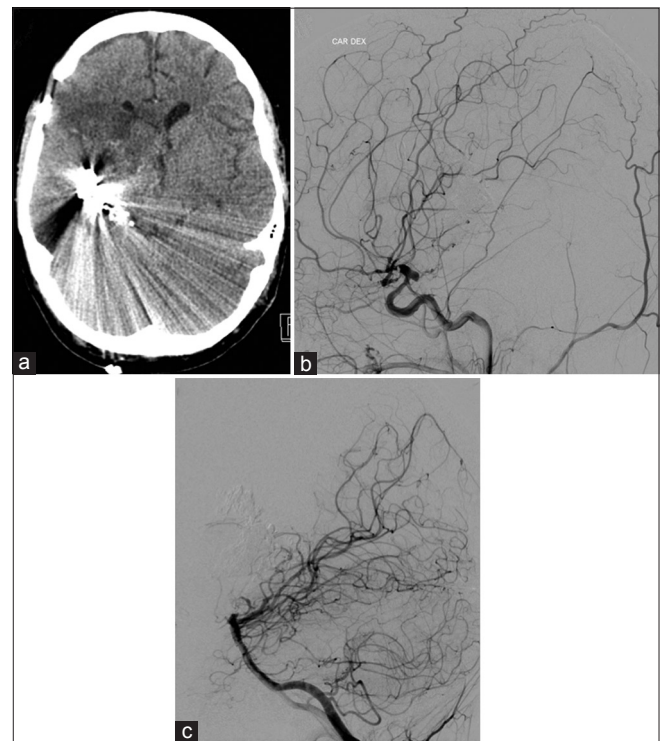
**Figure 4:** Postoperative angiographic images after the 4<sup>th</sup> embolization: Digital subtraction angiography (DSA) (posterioranterior view) (a) no filling of arteriovenous malformation through middle cerebral artery; DSA (lateral view) (b) small residual part filling through branch of posterior cerebral artery (arrow)

## DISCUSSION

Cerebral giant deep arteriovenous malformations are complex lesions with highly challenging treatments. In addition, giant AVMs and deep-seated AVMs have consistent risk factors for future bleeding.<sup>[6]</sup> According to the current data, the rate of rupture of AVMs is 2.4–4%,<sup>[6,12]</sup> for AVM Spetzler–Martin Grade IV; for grade V risk is higher at 3.3%.<sup>[8]</sup> Fleetwood *et al.* studied 96 patients with basal ganglia and thalamus AVMs. They revealed that annual hemorrhage rate after detection



**Figure 3:** Postoperative images after the 2<sup>nd</sup> operation: axial CT scan (a) air instead of main part of arteriovenous malformation (AVM); digital subtraction angiography (posterioranterior view) (b) medial and lateral residual parts of AVM (arrows); axial diffusion weighted imaging scan (c); only focal ischemia adjacent to remnant part in basal ganglia



**Figure 5:** Postoperative images after the 4<sup>th</sup> operation: Axial CT scan (a) occluded part of the arteriovenous malformation (AVM); digital subtraction angiography (lateral view) through the middle cerebral artery (b) and through posterior cerebral artery (c) – AVM not filling





**Figure 6:** The patient – five years after initial surgery

of the lesion was 9.8% per patient per year, and among those who suffered hemorrhage, 85.5% were hemiparetic or hemiplegic.<sup>[5]</sup>

Although it is probable that deep AVMs carry a higher hemorrhage risk, it is clear that these AVMs also carry a higher surgical treatment risk.<sup>[15]</sup> Careful preoperative analysis the risks and benefits of all available treatment options should be made before surgery.<sup>[4]</sup> Hemorrhage may encourage surgical intervention, it can also create extra-anatomic surgical route to the lesion.<sup>[3]</sup>

In general, for deep AVMs without safe surgical access, initial treatment of choice should be radiosurgery and/or endovascular intervention. Nevertheless, radiosurgery carries a relatively lower long-term obliteration rate and has a high risk of radiation-related complications compared with AVMs in other locations. Potts *et al.* found that 18% of patients initially treated only with radiosurgery of deep AVMs have post-treatment hemorrhage and the annual rate is 3.9%.<sup>[13]</sup> Thus, in each particular case, it is necessary to weigh the risks of different options of treatment and natural history of these malformations.

As illustrated in this report, specific clinical situations may compel surgical intervention,<sup>[4]</sup> with surgery becoming a life-saving procedure. Usually using preoperative endovascular embolization reduces blood flow to an AVM, thereby decreasing intraoperative blood loss. Sometimes, similar to our case, a vascular structure such as an intranidal aneurysm filled with an endovascular agent leads to a major bleeding. This bleeding is very difficult to handle because bipolar coagulation fails to compress such structure. The solution of choice is an application of permanent vascular clips.

## CONCLUSION

This case demonstrates a complex problem that encountered during treatment of giant deep AVM. Such kind of AVMs should have staged multimodality treatment and final aim is complete occlusion. This eliminates the risk of future hemorrhages that is crucial for young individuals with a long life expectancy. Even a small remnant part of an AVM can cause hemorrhage.

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## Conflicts of interest

There are no conflicts of interest.

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