

## Multimodal treatment of cerebral AVMs: Case report

Salma Mrichi \*, Mohammed Yassaad Oudghiri, ElMehdi Hakkou, Mahjouba Boutarbouch, Adyl Melhaoui, Yasser Arkha and Abdessamad Elouahabi

*Specialty Hospital, Department of Neurosurgery, Mohamed V University, Rabat, Morocco.*

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### Abstract

Cerebral arteriovenous malformations located within eloquent areas are a permanent cause for concern. One that has forced all health providers to step up their game in order to improve clinical outcomes and prevent serious complications among patients. Awake surgery for cerebral mapping has changed dramatically the course of AVM history. Our case report discusses this very same technique.

**Keywords:** Arteriovenous Malformation; Awake Surgery; Radiosurgery; Brain Mapping

### 1. Introduction

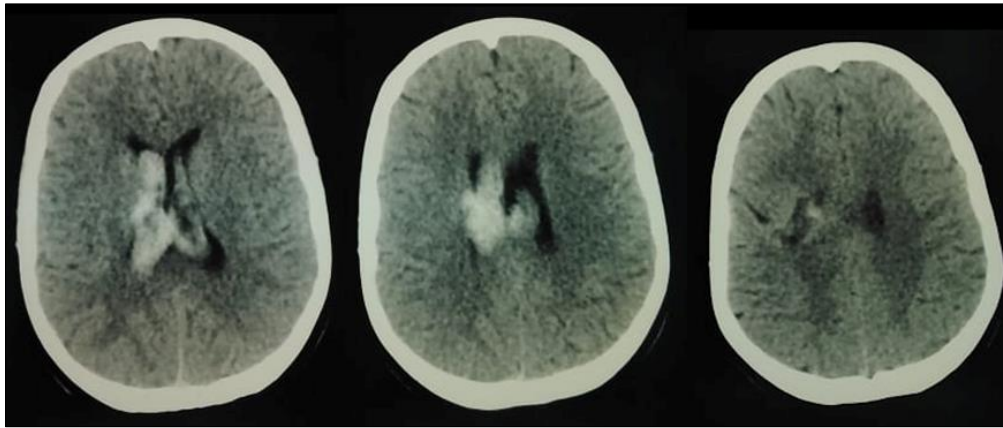
Arteriovenous malformation is a vascular abnormality characterized by a complex angioarchitecture in which there is hardly any interposition of the capillary network between cerebral arteries and veins. It was described by Harvey Cushing and Dandy Walter in the early 20th century. Not until 1986 that a classification of these cerebral AVMs was proposed by Spetzler and Martin [1]. This classification is used to predict the relative risk of morbidity and mortality among patients with cerebral AVMs.

As we know, these vascular malformations are classified according to 3 characteristics based on size, venous drainage and location within the eloquent or non-eloquent brain areas. Consequently, AVMs can be classified into 5 grades for which several therapeutic modalities can be proposed such as microsurgery, radiosurgery, embolization and finally multimodal treatment combining at least two of these techniques. Surgical treatment has long been at the heart of this disorder's management. It is considered as the gold standard for the treatment of AVMs. Whenever necessary, it can be associated with preoperative embolization or complementary radiosurgery, especially for low-grade AVMs.

### 2. Patient and observation

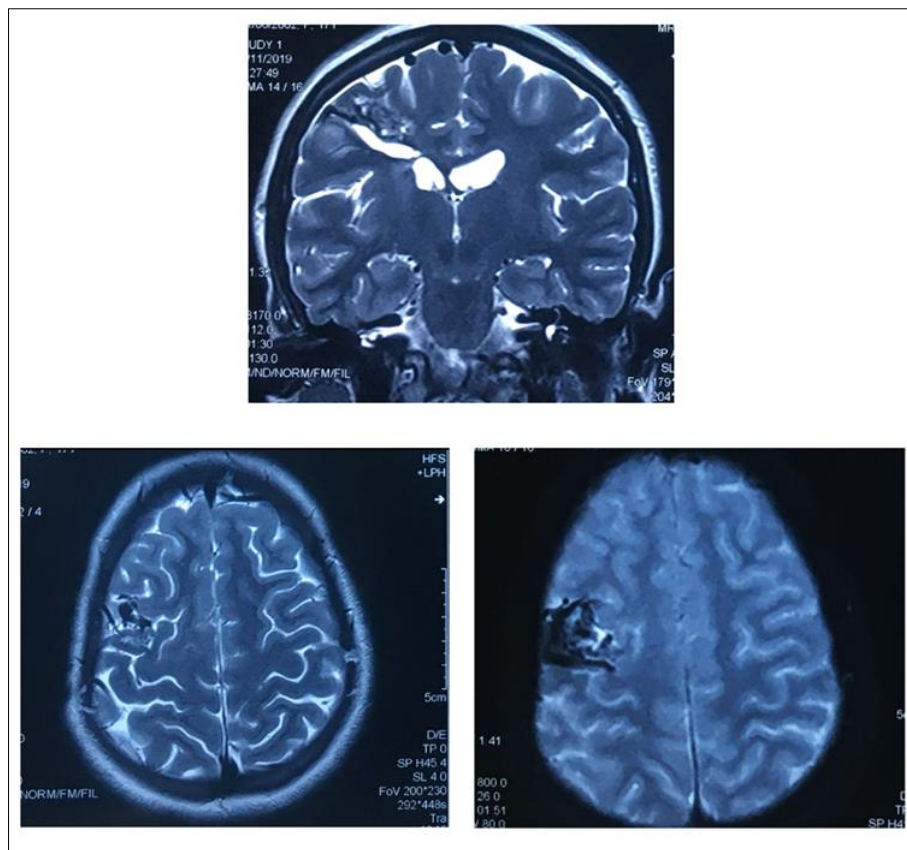
This is a case of an 18-year-old female, with a history of hemorrhagic stroke at the age of 12 years, expressed clinically by a left hemiparesis impeding walking ability with partial recovery. At the age of 16, she had a second hemorrhagic stroke with a left brachial monoparesis following the onset. She had her first consultation in emergency room after this event. Two years after this episode, she experienced Bravais-Jacksonian seizures, controlled with antiepileptic drugs. She underwent an imaging workup which included a cerebral CT scan (Figure 1). There was evidence of an intracerebral hematoma in the right perolondic cortex breaking into the right lateral ventricle.

\* Corresponding author: Salma Mrichi  
Specialty Hospital, Department of Neurosurgery, Mohamed V University, Rabat, Morocco.



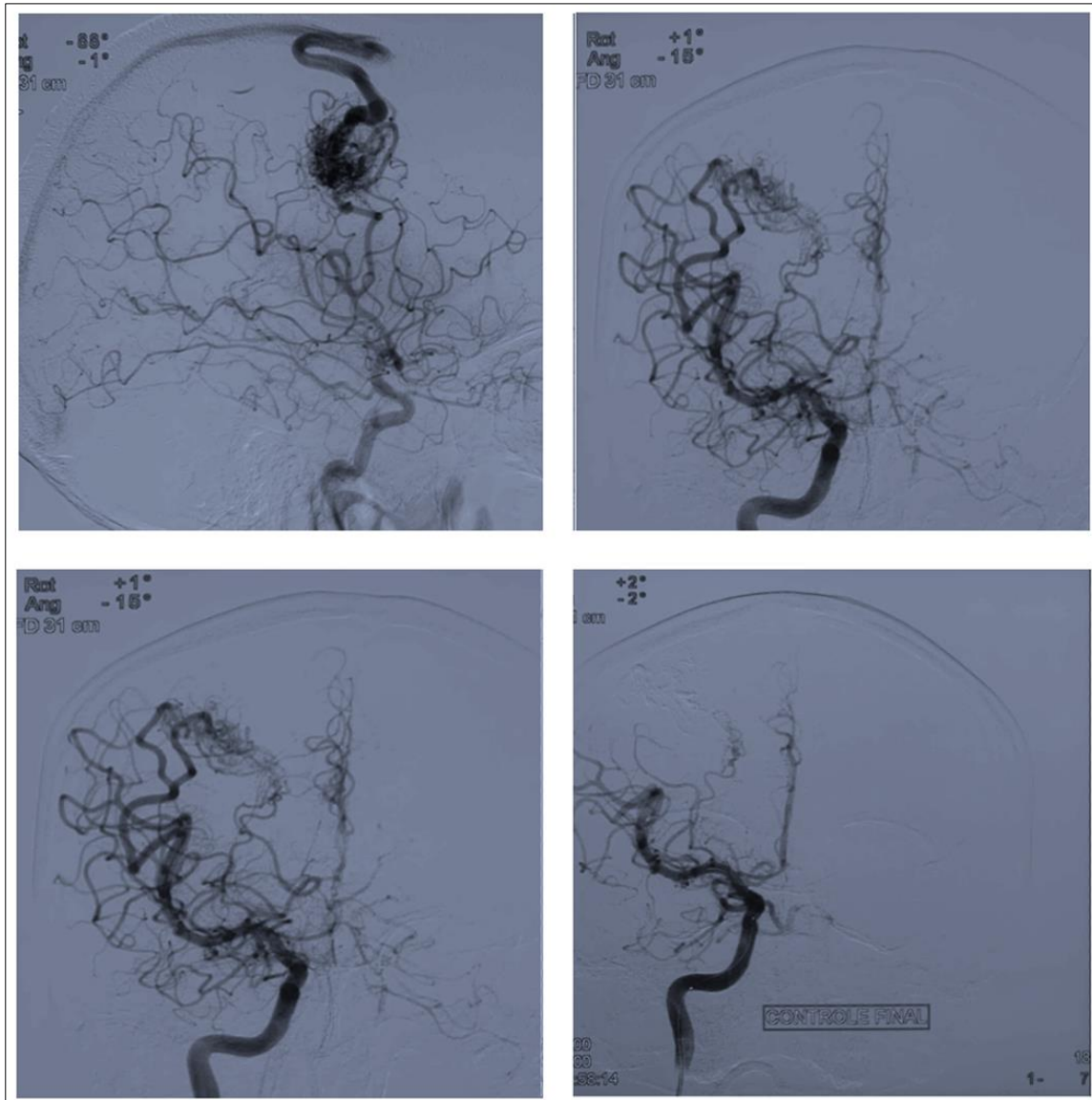
**Figure 1** Brain CT scan showing right frontal hematoma rupturing into the right ventricle

Brain MRI identified an AVM in the right precentral gyrus (Figure 2). Additional functional MRI showed an activation of the sensory-motor cortex of the hand and foot located immediately behind the AVM.



**Figure 2** T2 sagittal and axial and T2\* imaging demonstrates numerous serpiginous flow-voids consistent with a right-sided arteriovenous malformation with a small saccular apparent aneurysm

In order to study the angioarchitecture of the AVM, a cerebral angiography was performed. It showed a right frontal AVM supplied by a frontal branch of the middle cerebral artery and two lenticulostriate arteries (Figure 3). The AVM had a small compact nidus measuring less than 3 cm. It is drained by a dilated cortical vein draining into the superior sagittal sinus. The AVM is therefore classified as Spetzler II

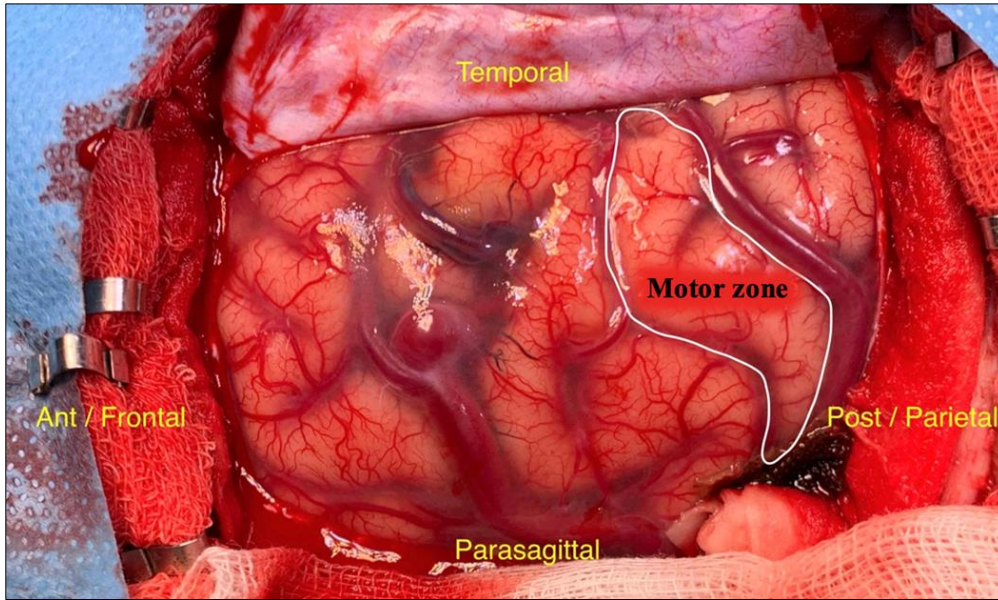


**Figure 3** *Upper images:* Cerebral angiography showing arterial supply from frontal branch of the middle cerebral artery and two lenticulostriate arteries. The AVM has superficial drainage via markedly enlarged cortical vein, which drain in the superior sagittal sinus. *Lower images:* Cerebral arteriography demonstrating supra selective embolization of AVM

A careful analysis of the case was carried out by a multidisciplinary team of neurosurgeons and neuroradiologists. The therapeutic decision was based on the following factors: the fact that there had been two episodes of bleeding, the patient's age, the AVM was located in an eloquent area, the AVM was grade II according to Spetzler, and the risk of rebleeding. We therefore proposed to our patient an awake surgery with preoperative embolization by supra-selective catheterization and injection of Onyx 18 particles. A 60% exclusion of the AVM was achieved through endovascular embolization, 48 hours before surgery (Figure 3).

Intraoperatively, after opening the dura mater, we observed a large arterialized vein running along the cerebral cortex, representing the draining vein of the AVM to the superior sagittal sinus. In addition, we could observe cortical vessels embolized by onyx particles (Figure 5). We performed a cortical stimulation to locate the frontal motor zone. We witnessed several Jacksonian seizures during stimulation that were stopped by flushing the surface of the brain with ice-cold saline. We access the AVM using a transsulcal approach following the draining vein. During AVM removal, there was minimal bleeding due to preoperative embolization. Total removal of the AVM was achieved.

There was no abnormal neurological event in the postoperative period. Anticonvulsant treatment was reinforced. At discharge, the patient had a grade 1 mRS. She is scheduled for a follow-up cerebral arteriography in one month.



**Figure 4** Intraoperative view showing motor zone located by cortical brain stimulation. *Ant = anterior; Post = posterior*

### 3. Discussion

In the literature, several papers have compared and contrasted surgical treatment versus embolization alone or stereotactic radiosurgery. One can mention of course the famous ARUBA trial, criticized for quite some time [2],[3]. This trial is specific to unruptured AVMs, unlike our present case. According to this study, medical treatment would be the most advisable option for AVMs given that the mortality and stroke rates were 3 times higher among patients who underwent surgery or embolization. However, when we take a closer look at the patient selection process, we realize that this study highlights the results of an inappropriate therapeutic choice. The outcomes are not conclusive and do not reflect the real performance of microsurgical treatment. The authors reported that 68% of the patients in the interventional arm were Spetzler class A, and only 4% received surgical treatment.

Later on, it became clear that surgical treatment is recommended for class A AVMs. Indeed, the Spetzler classification published in 2011 came to a similar conclusion.

In order to have clear guidelines for the management of cerebral AVMs, the same team from the Barrow Neurological Institute proposed a second classification to categorize the Spetzler and Martin grades into three classes [4] as follows : Class A AVMs, which include Spetzler I and II, should be treated surgically. Class B AVMs or Spetzler III may benefit from multimodal treatment, such as surgery and embolization. To evaluate and understand the current trends in the management of cerebral AVMs, a paper published by the UCLA team gathered all the series of low-grade cerebral AVMs treated either by microsurgery, embolization using onyx and/or radiosurgery [5]. Table 1 summarizes the results of the three therapeutic modalities.

**Table 1** Outcomes according to the Potts et al. [5]

Treatment strategy	No. of patients	Morbidity (%)	Mortality (%)	Cure rate (%)	Hemorrhage rate (%)
Microsurgery	1235	2.2	0.3	98.5	0.3
Endovascular	1297	6.2	1.6	29.5	8.0
Radiosurgery	1057	6.5	1.2	75.2	7.2

For low grade AVMs, it appears that endovascular treatment and radiosurgery carried a rate of morbidity three times higher than microsurgery, and a rate of mortality 4 to 5 times greater. A comprehensive review of the literature was published on pubmed and EMBASE covering studies conducted between January 2000 and March 2011 [6]. This review

includes 137 studies including 142 cohorts, 13,698 patients, 46,314 patient per years of follow up. The authors confirm the above-mentioned results. Highly satisfactory occlusion rates were reported in the surgically treated groups with levels of 96% following microsurgery, 38% following stereotactic radiosurgery and 13% following embolization.

As to the use of embolization in the management of cerebral AVMs, it would be indicated as a preoperative treatment prior to microsurgery as well as radiosurgery, as palliative treatment for symptomatic lesions unsuitable for surgery, and as a first-line treatment for small, deep, and difficult-to-access AVMs. History narrates the first cases of endovascular treatment in the holistic and multimodal management of cerebral AVMs. To date, it is the only FDA approved indication for Onyx use in the United States. There is a wide variety of embolic agents used worldwide. Nowadays, n-butyl cyanoacrylate (NBCA), ethylene vinyl alcohol copolymer, platinum coils, and polyvinyl alcohol particles are agents commonly used to embolize AVMs. Onyx is one of the newer liquid embolic agents. It has the advantage of being non-adhesive. Unlike NBCA, which hardens almost immediately, Onyx has a very slow solidification rate. The slow solidification rate and the lack of adherence allow for more extended and repeated injections from the same point, which ultimately results in deeper penetration of this agent, reaching a larger portion of the nidus. However, one must keep in mind that these are theoretical advantages, as there is currently no published evidence to support such claims. The overall strategy for preoperative embolization includes occlusion of deep arterial branches not encountered until the very final stages of surgical resection. In addition, it aims to secure AVM-associated aneurysms, especially if they are remotely located away from the AVM. In contrast to curative embolization, in which penetration and obliteration of the nidus are required, successful preoperative embolization can be achieved by decreasing the arterial supply to the AVM by occluding its feeding pedicles. The latter will allow limited embolization of the feeding arteries, known as supra-selective embolization. When practiced, it is critical to ensure that cumulative risks associated with endovascular treatment and surgery do not exceed the risk of treating the very same AVM without embolization.

Microsurgical resection of AVMs in eloquent areas can be difficult. In the early 80s, an idea was gradually gaining ground among neurosurgeons interested in brain AVMs. According to them, there is a modification of the classical brain architectural features seen in patients with AVMs. The advent of functional MRI in the late 90s confirmed these suspicions, proving that translocation of the language area in cerebral AVMs is quite common. Moreover, the availability of this new tool led us to rethink the known anatomical patterns regarding the location of eloquent areas and to consider more extensively awake surgery. We would like to emphasize that awake surgery in the management of AVMs is not common practice. This procedure requires a team of experts including anesthesiologists and dedicated neurosurgeons. Several centers, have attempted similar procedure. The Gamble team published in 2015 a series of 42 cases of AVMs [7]. The focus of this study was to investigate the value of Language mapping, both cortical and subcortical during awake surgery. It involved Spetzler Grade II and III AVMs. This technique allowed a safe surgical excision with well-defined margins thus reducing the postoperative morbidity rate. Cortical and subcortical stimulation were considered to significantly affect the patients' outcome in 67% of cases.

A retrospective study covering 13 years extending from 2003 to 2016 at the Prince of Wales Hospital in Hong Kong [8], reported Spetzler II and III grade AVMs management options. The objective of this study was to qualitatively distinguish between brain mapping via awake surgery and radiosurgery treatment. These two options seem to be the most comparable regarding targeted treatment. Comparative analysis revealed a better outcome of awake surgical treatment compared to radiosurgery with a 100% obliteration rate versus 96% for radiosurgery.

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#### 4. Conclusion

State-of-the-art treatment of cerebral AVMs is a multimodal business, one which requires a multidisciplinary team dedicated to cerebrovascular neurosurgery, endovascular intervention, and radiation therapy to deliver a full spectrum of therapeutic options and to determine the most appropriate treatment regimen based on the patient's characteristics and AVM morphology. As for preoperative embolization, its effectiveness has been demonstrated, but one must be mindful of the cumulative risk of surgery combined with embolization versus surgery alone.

Awake surgery in selected cases of AVMs within eloquent areas may improve patients' outcomes by preserving speech and motor functions during resection.

## Compliance with ethical standards

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Salma Mrichi: Conceptualization, data collection, paper preparation, writing; Mohammed Yassaad Oudrhiri: Conceptualization, ElMehdi Hakkou: Conceptualization, Mahjouba Boutarbouch: study design; Adyl Melhaoui: study design; Yasser Arkha: Validation. Abdessamad El Ouahabi: Validation, Writing - review & editing.

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The authors did not receive any funding for the preparation of this case report.

### *Statement of informed consent*

Informed consent and verbal permission were obtained from the patient and her family before the submission of this article. In addition, this article follows both the Consensus-based Clinical Case Reporting Guideline and the Recommendations for the Conducting, Reporting, Editing, and Publication of Scholarly Work in Medical Journals.

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