# Cerebral arteriovenous malformation recurrence

- Delayed but Dangerous: Chronic Encapsulated Expanding Hematoma as a Reversible Cause of Steroid-Resistant Extensive Edema Following Stereotactic Radiosurgery for Cerebral Arteriovenous Malformation
- Case Report: Multiple cerebral infarctions in a patient with hereditary hemorrhagic telangiectasia following a fall
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Stereotactic radiosurgery (SRS) has been recently described but mainly limited to AVMs deemed too risky to approach in an open fashion and limited to 2.5cm-3cm in size. The delayed protection from hemorrhage (approximately 2-3 years) and high marginal failure/recurrence rate are the greatest concerns <sup>1)</sup>.

Follow-up imaging has an integral role after cerebral arteriovenous malformation resection and is sometimes not performed for a sufficient period, leading to delayed detection of recurrence and an increased likelihood of a recurrent ruptured cerebral arteriovenous malformation <sup>2</sup>.

While there is general consensus on the importance of follow-up after surgical removal of an AVM, there is a lack of consistency in the duration of that follow-up  $^{3)}$ .

An inherent preference exists for MR angiography (MRA) surveillance rather than arteriography. The validity of this strategy is uncertain.

The true postoperative incidence of arteriovenous malformation (AVM) recurrence in the pediatric population remains largely unreported. Some literature suggests that delayed imaging studies should be obtained at 6 months to 1 year after negative findings on a postoperative angiogram.

## Systemic review

The Department of Neurosurgery, University of Miami Miller School of Medicine, Miami, Florida, performed a systematic review using articles obtained through a search of the literature contained in the MeSH database, according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.

Search results revealed 1052 articles, 13 of which described 31 cases of AVM recurrence meeting the

criteria for inclusion in this study. Detection of AVM occurred significantly earlier (mean  $\pm$  SD, 3.56  $\pm$  3.67 years) in patients with follow-up imaging than in those without (mean 8.86  $\pm$  5.61 years; p = 0.0169). While 13.34% of patients who underwent follow-up imaging presented with rupture of a recurrent AVM, 57.14% of those without follow-up imaging presented with a ruptured recurrence (p = 0.0377).

Follow-up imaging has an integral role after AVM resection and is sometimes not performed for a sufficient period, leading to delayed detection of recurrence and an increased likelihood of a ruptured recurrent AVM <sup>4)</sup>.

### **Case series**

Järvelin et al. studied the rate of recurrence of surgically treated bAVMs at Kuopio University Hospital (KUH) in 1981-2021. The study population was collected retrospectively from KUH databases and presented a cohort of 135 surgically treated bAVMs with complete occlusion of the lesion. We also performed a systematic literature review on this topic. In our series, 6 out of 135 (4.4%) patients with angiographically confirmed removal of the lesion later developed a recurrent bAVM with a median time to diagnosis of recurrence of 7.46 years. In pediatric patients, the rate was 5 out of 17 (29.4%). bAVM recurrence was associated with age (p = 0.001) and initial hemorrhagic presentation (p = 0.039). Median age of the study population was 37 years (min 0, max 70), and 51/135 (37.8%) of the patients were female. Seventeen (12.6%) of the 135 bAVM patients were considered pediatric (18 years old or younger) at the time of the operation. In the literature review, 79 of 1739 (4.5%) of surgically treated patients later developed a recurrence with a mean delay of 3.1 years until diagnosis of recurrence. Young surgically treated bAVM patients with a hemorrhagic presentation at initial diagnosis are at a relatively high risk of bAVM recurrence. Follow-up imaging should be arranged for these patients in order to prevent rupture from a recurrent bAVM and subsequent morbidity <sup>5)</sup>.

#### 2016

Two hundred three patients underwent AVM resection between 1995 and 2012. Seventy-two patients met eligibility criteria. There were 3 recurrences (4%). Deep venous drainage and diffuse type of AVM nidus were significantly associated with recurrence. Although preoperative embolization did not reach statistical significance as an independent risk factor, radiographic data supported its role in every case, with the site of recurrence correlating with deep regions of nidus previously obliterated by embolization.

AVM recurrences in the adult population may have a multifactorial origin. Although deep venous drainage and diffuse nidus are clearly risk factors, preoperative embolization may also be a contributing factor with the potential for recurrence of unresected but embolized portions of the AVM. Follow-up angiography at 1 to 3 years appears to be warrant <sup>6</sup>.

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#### 2015

A retrospective chart review was performed on pediatric patients treated for cerebral AVMs at a single institution from 1998 to 2012. Patients with complete obliteration of the AVM nidus after treatment and more than 12 months of follow-up were included in the analysis. Data collection focused on recurrence rates, associated risk factors, and surveillance methods.

A total of 45 patients with a mean age of 11.7 years (range 0.5-18 years) were treated for AVMs via surgical, endovascular, radiosurgical, or combined approaches. Total AVM obliteration on posttreatment digital subtraction angiography (DSA) was confirmed in 27 patients, of whom the 20 with more than 12 months of follow-up were included in subsequent analysis. The mean follow-up duration in this cohort was 5.75 years (median 5.53 years, range 1.11-10.64 years). Recurrence occurred in 3 of 20 patients (15%). Two recurrences were detected by surveillance DSA and 1 at the time of rehemorrhage. No recurrences were detected by MRA. Median time to recurrence was 33.6 months (range 19-71 months). Two patients (10%) underwent follow-up DSA, 5 (25%) had DSA and MRI/MRA, 9 (45%) had MRI/MRA only, 1 (5%) had CT angiography only, and 3 (15%) had no imaging within the first 3 years of follow-up. After 5 years posttreatment, 2 patients (10%) were followed with MRI/MRA only, 2 (10%) with DSA only, and 10 (50%) with continued DSA and MRI/MRA.

AVM recurrence in children occurred at a median of 33.6 months, when MRA was more commonly used for surveillance, but failed to detect any recurrences. A recurrence rate of 15% may be an underestimate given the reliance on surveillance MRA over angiography. A new surveillance strategy is proposed, taking into account exposure to diagnostic radiation and the potential for catastrophic rehemorrhage<sup>8)</sup>.

#### 2012

A total of 28 patients (13 female, 15 male) underwent an AVM resection. In 18 patients (64.3%) an intraoperative angiogram was obtained. In 4 cases the intraoperative angiogram revealed residual AVM, and repeat resections were performed immediately. Recurrent AVMs were found in 4 children (14.3%) at 50, 51, 56, and 60 weeks after the initial resection. Recurrence risk was 0.08 per personyear. No patient with normal results on an angiogram obtained at 1 year developed a recurrence on either a 5-year angiogram or one obtained at 18 years of age. All patients with recurrence had a compactness score of 1 (diffuse AVM); a lower compactness score was associated with recurrence (p = 0.0003).

All recurrences in this cohort occurred less than 15 months from the initial resection. The authors recommend intraoperative angiography to help ensure complete resection at the time of the surgery. Follow-up vascular imaging is crucial for detecting recurrent AVMs, and conventional angiography is preferred because MRA can miss smaller AVMs. One-year follow-up imaging detected these recurrences, and no one who had negative results on an angiogram obtained at 1 year had a late recurrence. However, not all of the patients have been followed for 5 years or until 18 years of age, so longer follow-up is required for these patients. A lower compactness score predicted recurrent AVM in this cohort <sup>9</sup>.

#### Case reports

A 24-year-old woman was diagnosed with a cerebral arteriovenous malformation(AVM)in the right

parietal lobe(Spetzler-Martin grade I). The AVM was treated with stereotactic radiosurgery and was observed to have completely disappeared 3 years after radiosurgery. At the age of 35 years, the patient complained of a headache, and was referred to our hospital. A plain CT scan demonstrated a large cyst with niveau formation in the right parietal lobe. Cerebral angiography identified no recurrence of AVM. However, contrast MRI revealed an enhanced lesion on the surface of the cyst. The patient underwent cyst fenestration and total removal of the obliterated nidus through a right parietal craniotomy. Residual blood flow was confirmed in the obliterated nidus during surgery. The postoperative course was uneventful, and the headache was completely resolved. The patient was discharged without any neurological deficits. On pathological examination, a large number of small vessels were observed within the obliterated nidus. Immunohistochemistry demonstrated that these vessels were positive for CD31, CD34, and VEGFR-2, suggesting that endothelial progenitor cells may be involved in occult recurrence, cyst formation, and late bleeding after stereotactic radiosurgery targeting cerebral AVMs<sup>10</sup>.

A 25-year-old man with cerebellar hemorrhage secondary to a recurrent AVM. His current admission with bleeding from a ruptured cerebellar AVM followed a previous presentation 15 years earlier with the similar clinical picture of AVM rupture within the same vascular territory. At that time, he was managed conservatively with follow-up digital subtraction angiography (DSA) 2 years later, confirming no residuum of the AVM. At the current presentation, he had DSA confirming AVM recurrence. He was managed by complete excision of the AVM via a suboccipital craniotomy.

This case illustrates the need for long-term imaging follow-up to exclude recanalization even many years after AVM obliteration  $^{11}$ .

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