Spontaneous regression of brain arteriovenous malformations

A clinical study and a systematic review of the literature

Introduction

The prevalence of brain arteriovenous malformations (AVM) is 0.02% [4], with an incidence of between 1.1 and 1.3 per 100,000 persons-years [5, 60]. An intracerebral hemorrhage is the main mode of presentation [1, 20, 26, 28, 51] with a prevalence of 0.68 per 100,000 and an incidence of 0.51 per 100,000 person-years [5, 60]. The median age of presentation is 28 and 38 years for a hemorrhagic and a non-hemorrhagic presentation, respectively [60]. Spontaneous and complete regression of an AVM is a well recognized, but rare phenomenon, with an estimated prevalence of 0.8 to 1.3% [2, 50]. Several causes of spontaneous regression have been postulated, such as premature atherosclerosis [8], embolus [30], turbulence in feeding vessels [15], elevated estrogens [57], hemodynamic changes associated with surgery [38] and mass effect due to haemorrhage from the AVM [43].

We describe the case of an adult patient, referred to our clinic for treatment of his brain AVM, by whom com-
plete regression had occurred, and present a systematic review of the literature.

**Patients and methods**

A computerized search of both the National Library of Medicine database of literature and Embase was performed. The search was limited to human studies. We used the medical subject heading 'intracranial arteriovenous malformations' combined with the terms 'regression' and 'obliteration'. Regression was defined as the disappearance of an AVM without prior intervention or therapy. Obliteration was defined as the disappearance of an AVM after prior therapy. Thrombosis of an AVM was only used when this was pathologically demonstrated. Brain AVMs were defined as parenchymal or pial AVMs. Excluded were articles that dealt with cases other than brain AVMs, cases of partial regression and all cases in which patients had undergone treatment. Treatment was defined as surgical, endovascular or radiation therapy to the AVM and included removal of an AVM related hematoma and clipping of aneurysms proximal to the AVM. VP-Shunt placement or exploratory surgery in which the AVM was not manipulated was not regarded as treatment. Duplicate references, as well as redundant publications were discarded. The abstracts were reviewed, and articles unrelated to the specific topic were excluded. To identify additional eligible studies, the reference lists were screened for journal articles.

An analysis of clinical, radiological, and pathological features of all cases was performed. Details included were: patient age at diagnosis of regression, gender, clinical presentation which led to the diagnosis of the AVM, localization and lateralization of the AVM, previous treatment, nidus size, Spetzler-Martin classification, number of arterial feeders, pattern of venous drainage, and number of draining veins (single or multiple). Events possibly leading to regression and timespan between diagnosis of the AVM and diagnosis of regression were noted. In our database of over 300 patients with a brain AVM, referred for stereotactic radiosurgery, we identified one patient in whom complete spontaneous regression had occurred.

**Case report**

A 51-year old male (patient 1, Table 1) presented with seizures, attributed to a left-temporal AVM, Spetzler-Martin grade II (Figs. 1A and B), with drainage in the direction of the transverse sinus (Fig. 1C). Two months later the patient had an acute moment of complete disorientation, with hemianopia and dysphasia, without headaches, nausea, vomiting, or loss of consciousness. Four months after this episode, he was referred to our institute for radiosurgical treatment. The preplanning-MR-angiography showed remains of an old hematoma, surrounded by gliosis, but no aberrant vessels. The planning DSA confirmed these findings (Figs. 1D and E).

![Fig. 1](image)

A small AVM is visible in the left temporal lobe. The AVM has a maximal diameter of approximately 1 cm (A & B). Drainage occurs through a cerebellar vein in the direction of the transverse sinus (C). Approximately two months after this angiogram the AVM ruptured, resulting in mild clinical symptoms. On the planning DSA performed 4 months later, the AVM is no longer demonstrable (D & E).