

## Fatal rupture of a brain arteriovenous malformation flow-related aneurysm during microcatheter removal: a rare complication

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Intracranial aneurysms are relatively frequently encountered in patients with brain arteriovenous malformations (BAVMs). They may be located on the circle of Willis, on arterial feeders, or even inside the nidus. Because BAVM-associated aneurysms represent a risk factor of bleeding, the question of the timing and modality of their management remains a matter of debate in unruptured BAVMs. The authors present a case of fatal periprocedural rupture of a flow-related aneurysm (FRA) during the removal of the microcatheter after injection of a liquid embolic agent. A 40-year-old man was treated at the authors' institution for the management of a Spetzler-Martin Grade III left unruptured frontal BAVM, revealed by seizures and a focal neurological deficit attributed to flow steal phenomenon. After a multidisciplinary meeting, endovascular treatment was considered to reduce the flow of the BAVM. A proximal FRA located on the feeding internal carotid artery (ICA) was purposely left untreated because it did not meet the criteria of the authors' institution for preventative treatment (i.e., small size [2.5 mm]). During embolization, at the time of microcatheter retrieval, and after glue injection, the aneurysm unexpectedly ruptured. The aneurysm's rupture was attributed to the stress (torsion/flexion) on the ICA caused by the microcatheter removal. Despite the attempts to manage the bleeding, the patient eventually died of the acute increase of intracranial pressure related to the massive subarachnoid hemorrhage. This case highlights a previously unreported mechanism of FRA rupture during BAVM embolization: the stress transmitted to the parent artery during the removal of the microcatheter.

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**KEY WORDS** brain arteriovenous malformation; flow-related aneurysm; fatal; complication; microcatheter removal; hemorrhage; vascular disorders

**I**NTRACRANIAL aneurysms may be observed concomitantly with brain arteriovenous malformations (BAVMs).<sup>1</sup> In some cases, the association of both lesions is fortuitous; in other cases, these aneurysms are either inside the BAVM or related to the increased flow on arterial feeders: the so-called “flow-related” aneurysms (FRAs). Flow-related aneurysms constitute a particular subtype of BAVM-associated aneurysms and they should be considered separately from intranidal and unrelated aneurysms. When unruptured, the management of these aneurysms and the timing for their treatment is controversial and remains a matter of debate.

Indeed, some authors<sup>2</sup> have recommended treating

FRAs before the BAVM because they may rupture after the closure of arteriovenous shunts due to increasing pressure in the parent vessel. Other authors<sup>6</sup> have argued that after BAVM embolization, these aneurysms may spontaneously regress due to hemodynamic changes in the feeding arteries. In this paper we report an unusual cause of rupture of a proximal FRA during microcatheter removal, after glue embolization, leading to a fatal subarachnoid hemorrhage.

### Case Report

#### History and Examination

A 40-year-old man was treated at Pitié-Salpêtrière Hos-

**ABBREVIATIONS** ACoA = anterior communicating artery; BAVM = brain arteriovenous malformation; DSA = digital subtraction angiography; FRA = flow-related aneurysm; ICA = internal carotid artery.

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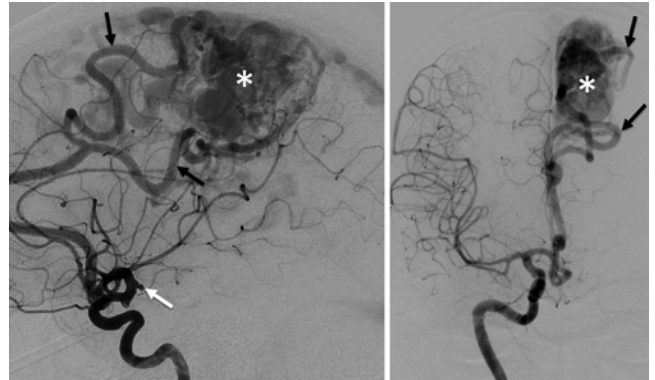
pital for the management of a Spetzler-Martin Grade III<sup>7</sup> unruptured frontal BAVM revealed by seizure and resistance to antiepileptic drugs, later associated with a right lower-limb persistent hyposthenia, possibly due to flow steal phenomenon. No previous bleeding was depicted on pretreatment MRI. The medical chart was discussed in a multidisciplinary meeting. Because the patient's symptomatology was partly attributed to steal phenomenon, embolization was considered to reduce the flow of this BAVM. Digital subtraction angiography (DSA) was performed, which confirmed the presence of the frontal BAVM fed by dilated branches from the left pericallosal artery, and by frontal branches of the left middle cerebral artery. It is noteworthy that the right internal carotid artery (ICA) also supplied the BAVM via a dilated right A<sub>1</sub> segment and a patent anterior communicating artery (ACoA). The nidus measured 35 mm and presented multiple direct intranidal arteriovenous shunts (Fig. 1). In addition, the angiography showed a proximal 2.5-mm FRA with a 2-mm neck located on the posterior aspect of the terminal segment of the right ICA. A huge kinking of the left cervical ICA was also observed (not shown) on diagnostic DSA.

Functional 3-T MRI was performed, which showed the close relationship of the nidus with the primary motor cortex of the right lower limb (not shown). The case was then discussed in a multidisciplinary meeting involving neurosurgeons, interventional neuroradiologists, radiosurgeons, and neuroanesthesiologists; endovascular embolization was considered as a suitable therapeutic option. The strategy proposed was to treat only the intranidal high-flow arteriovenous shunts to reduce the steal phenomenon. Because the aneurysm was small, conservative management was chosen.

#### Operation and Postoperative Course

Preoperative DSA showed morphological stability of the aneurysm and, as planned, treatment was directed toward the BAVM. General anesthesia was induced and full heparinization started, i.e., activated clotting time between 2- and 3-fold of the baseline. A 6-Fr Envoy guiding catheter was positioned in the proximal petrous segment of the right ICA. Then, a flow-dependent microcatheter (1.2F Magic, Balt Extrusion) was navigated distally through the right A<sub>1</sub> segment and the ACoA toward a branch of the left pericallosal artery feeding a high-flow direct arteriovenous shunt. The navigation process was smooth and performed by an experienced operator with more than 20 years of experience in the treatment of AVMs; far above the circle of Willis, a Mirage microguidewire (ev3/Covidien) was added to the system to progress further by giving some additional proximal support.

Once the microcatheter reached a juxta-nidal position, an injection of 0.5 ml of Glubran 2 (GEM) at 30% concentration, diluted with lipiodol, was performed under a blank roadmap. Such a concentration was chosen to limit venous migration of the embolic agent because of the high-flow characteristic of the arteriovenous shunt (Fig. 2 left). The microcatheter was rapidly retrieved under fluoroscopic control. No microcatheter sticking or breakage occurred, but a minor movement of the vessels was observed along the path of the microcatheter. Subsequent angiographic



**FIG. 1.** Digital subtraction angiography lateral (left) and anteroposterior (right) projections of the right ICA before endovascular treatment. The BAVM (asterisk) is mainly fed by branches from the left pericallosal artery (black arrows) and presents a mixed cortical and deep (toward the vein of Galen) venous drainage. The left anterior cerebral artery and the BAVM are opacified by injection in the right ICA via a patent ACoA. The right ICA also presents a 2.5-mm aneurysm (white arrow).

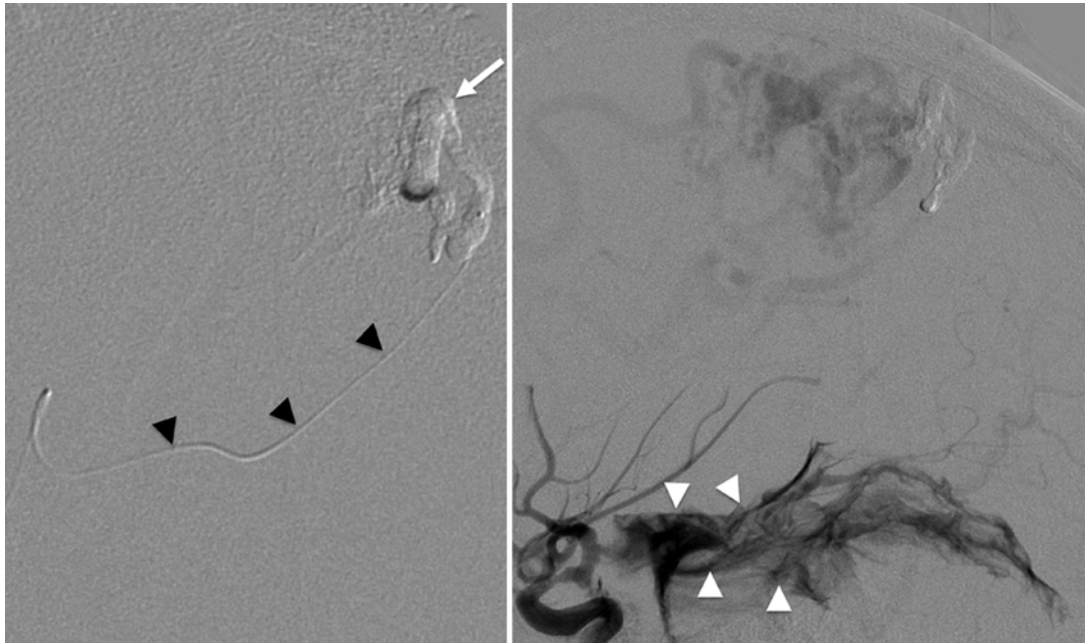
control showed a massive extravasation of contrast media, freely flowing from the dome of the now ruptured ICA aneurysm (Fig. 2 right).

Right cervical ICA DSA performed just after rupture demonstrated a twisting of the intrapetrous segment of the ICA (Fig. 3). After attempts to stop the acute bleeding by interventional means (i.e., navigation of a nondetachable balloon), it spontaneously stopped within 5 minutes. While hypertensive peaks were not registered before rupture, shortly after an arterial pressure increase and associated bradycardia were evident and were attributed to the response to increased intracranial pressure (Cushing's reflex). The patient was kept in a barbiturate-induced coma, but due to the poor prognosis suggested by the postoperative brain CT scan (Fig. 4 left), by the MRI performed at Day 5 (Fig. 4 right), and by the absence of awakening after stopping sedation, we chose to limit aggressive intensive care treatment and the patient eventually died.

## Discussion

The prevalence of BAVM-associated aneurysms has typically been reported in approximately 15%<sup>5,6</sup> of patients with BAVMs (range 2.7%<sup>4</sup> to 58%<sup>8</sup>). According to some authors, BAVM-associated aneurysms should be divided into unrelated aneurysms, FRAs (located on the arterial feeders), and intranidal aneurysms.<sup>5,6</sup>

The management of an FRA is a dilemma, balanced between a uselessly aggressive treatment of an aneurysm that may spontaneously regress, versus cautious observation of an aneurysm that may acutely bleed. Flow-related aneurysms have been subclassified by Redekop et al.<sup>6</sup> into proximal (aneurysms of intracranial ICAs and the vertebrobasilar system up to and including all the circle of Willis and the M<sub>1</sub> segment) or distal (when above these locations). According to their case series, after BAVM obliteration, or nidus reduction greater than 50%, distal aneurysms are likely to regress whereas proximal aneurysms are likely to remain unchanged. On the other hand,



**FIG. 2.** Endovascular treatment of the BAVM. **Left:** Snapshot of the *N*-butyl cyanoacrylate (NBCA) injection; blank roadmap in lateral projection showing the course of the microcatheter (*arrowheads*) and the cast of glue injected (*arrow*). **Right:** Control digital subtraction angiogram in lateral projection after microcatheter retrieval. Acute subarachnoid hemorrhage from the tip of the aneurysm's sac is noted (*arrowheads*).

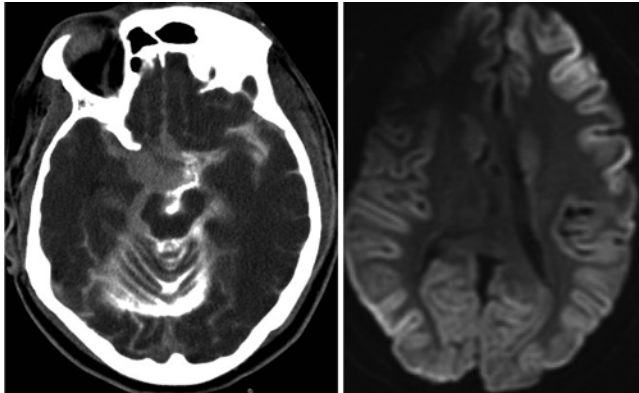
experimental observation<sup>3</sup> shows that during BAVM obliteration distal feeders are subjected to profound pressure elevation, while less significant alterations act in proximity and below the circle of Willis.

In our institution, the strategy concerning unruptured FRAs is to treat distal aneurysms at the same time as BAVM treatment, usually by glue injection, and to treat proximally located aneurysms before BAVM only when they present with a diameter greater than 5 mm and/or irregular shape. Conversely, in patients who present with hemorrhage, all aneurysms that are believed to be the source of bleeding are treated first.

In the current case, the proximal FRA of the ICA presented no criteria for selective treatment before nidus embolization. Retrospectively, it would have been safer to embolize the BAVM from the left ICA. However, the huge kinking of the cervical ICA would have been difficult or even impossible to cross, leading to a much more challenging catheterization of the BAVM's feeders. Indeed, supple guiding catheters were not available when we treated the patient and a regular guiding catheter would not have been able to be navigated distally to the kinking. Finally, we may have not used full heparinization during the procedure. Because the blood flow in the parent artery was so high, a thromboembolic complication would have been unlikely. On the contrary, heparin may have promoted or at least increased the bleeding from the aneurysm. The instantaneous bleeding after the microcatheter removal, the associated mechanical stretching observed along the parent artery, and the proximity of the aneurysm to the dural ring (1.5 cm) that could act as a point of fixation, suggest a cause-effect relationship between the catheter retrieval and the aneurysm rupture. Although it is impossible to as-



**FIG. 3.** Right ICA digital subtraction angiogram in lateral projection performed just after the FRA rupture showing a twisting of the cervical ICA (*arrow*).



**FIG. 4.** **Left:** Axial brain CT scan obtained after the procedure showing a diffuse subarachnoid hemorrhage and contrast media extravasation in the subarachnoid space. **Right:** Axial brain diffusion-weighted 3-T MR image obtained on Day 5 after the endovascular treatment showing a hyperintense signal involving the entire brain cortex, suggestive of irreversible brain anoxia.

certain whether all or just some of these factors coincided to determine the rupture, it appears very probable that the rupture was linked to the retrieval of the microcatheter.

To our knowledge this mechanism of BAVM-associated aneurysm rupture has not been previously reported. This fatal complication should prompt interventional neuroradiologists to carefully consider the presence of proximal FRAs during treatment planning. Indeed, even if hemodynamic alterations induced by AVM obliteration are unlikely to be significant at a proximal level, other mechanisms may induce rupture of a proximal FRA. Thus, if an alternative approach is feasible—for example, from the contralateral side—this should be taken into account and if the route ultimately chosen harbors FRAs, then special care during retrieval of the microcatheter is advisable, or maybe the use of a distal access catheter could be considered.

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## Author Contributions

Conception and design: Clarençon, Gabrieli. Acquisition of data: Sourour. Drafting the article: Clarençon, Gabrieli. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Clarençon.

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