Secondary Spontaneous Thrombosis of a Giant Aneurysm Located Distally on a Feeding Artery after Embolization of an Associated Arteriovenous Malformation

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Summary

Giant aneurysm located in the distal part of the feeding artery associated with a cerebral arteriovenous malformation is rare and the treatment is clinically challenging.

We report the spontaneous and complete thrombosis of a flow-related giant aneurysm immediate up-stream to a cerebral arteriovenous malformation by embolization of that malformation alone in a patient presenting with complex partial seizure and no history of intracranial haemorrhage. We obviated the need to directly intervene on the giant aneurysm, thus reducing unnecessary procedure related risks to the patient. Follow up one year later confirms the thrombosis and show shrinkage of the mass. The patient is asymptomatic.

Introduction

The association between cerebral arteriovenous malformations (AVM) and intracranial aneurysms has been well documented in the literatures¹⁻¹¹. Among them is a subgroup of giant aneurysms associated with cerebral AVM, which is very rare⁴⁻⁷.

The treatment strategy for patients with both cerebral AVM and aneurysm is still controversial, especially in those with no history of

aneurysmal rupture^{8,10}. We keep on applying the targeted embolization that we developed 20 years ago and maintain particularly in cerebral AVM and AA¹⁵. If the associated aneurysm is giant in nature, it could impose additional challenge to the clinical management, be it surgical, endovascular, or combined approach. We report the endovascular strategy in a case of cerebral AVM associated with a feeding artery giant aneurysm, in which embolization was targeted towards the AVM only.

Case Report

A 27-year-old lady was referred for further management of her cerebral AVM, presented with complex partial seizure that was poorly controlled by anticonvulsant medications. There was no history of subarachnoid or intracerebral haemorrhage. Pre-embolization diagnostic cerebral angiogram demonstrated a large cerebral AVM over the right Sylvian fissure, supplied by branches of the right middle cerebral artery. In addition, there was a bilobed giant aneurysm measuring 3 cm x 2.5 cm x 2.5 cm in dimensions in a feeding artery, locating just up-stream to the nidus of the AVM (figure 1). Large draining veins were seen leading from the AVM to the superior sagittal sinus.

Figure 1 A) Frontal and B) lateral views of digital subtraction cerebral angiograms demonstrating the architecture of the cerebral AVM and the associated up-stream giant aneurysm. C) Medial and D) lateral 3 dimensional reconstruction images of the cerebral AVM and the associated giant aneurysm.

We decided to perform embolization of the AVM first, in view of an absence of history of haemorrhagic complication and a possibility of subsequent spontaneous thrombosis of the associated up-stream giant aneurysm. Superselective catheterization of the right middle cerebral artery was performed with microcatheters (Baltacci 1.8), which were then navigated through the giant aneurysm and reached the sites of entry of the feeding arteries into the AVM. A total of 1.4 ml of Histoacryl mixed

with 50% of Lipiodol was injected into the AVM, with good penetration of the nidus. Successful embolization of 80% of the AVM was achieved. Cerebral angiogram performed immediately after embolization already showed the expected stasis of blood-flow within the giant aneurysm, with characteristic blood-contrast level identified (figure 2).

A CT scan of the head 48 hours after the embolization confirmed complete thrombosis of the giant aneurysm, with no increase in mass

effect observed (figure 3). A follow-up cerebral angiogram 2 weeks post-embolization showed persistent thrombosis of the giant aneurysm, as well as a small residual AVM (figure 4). The patient was neurologically intact on discharge and further embolization was planned for the residual AVM. On follow-up 1 year later (figure 5) the thrombosis and shrinkage of the giant aneurysm is complete, the AVM residual although small has been further embolized. The venous shunt is dramatically decreased. The patient is asymptomatic.

Discussion

The aneurysms associated with AVM can be classified by their location with respect to the nidus.

The emerging uniform classification system divided such aneurysms into four groups^{9,12}: dysplastic or remote, which is unrelated to the inflow vessels; flow-related proximal, arising at the circle of Willis origin of an artery supplying the AVM; flow-related pedicular, arising from the midcourse or distal branch of a feeding artery; intranidal, within the AVM nidus. Those arising on the arterial feeders were considered to be flow-related and their development were likely related to haemodynamic stress. This classification system has an advantage of avoiding mixing up of pedicular and intranidal aneurysms, which had very different properties but had been summarized together by some authors².

There is a wide range of reported incidence of aneurysms associated with cerebral AVM, from 3% to as high as 58%¹³⁻¹⁴. The cause of such a large discrepancy could be related to low interobserver agreement, inconsistent definitions for the concurrent aneurysms in different studies and also probably referral bias to the tertiary treatment centers¹¹. When narrowing down to the subgroup of giant aneurysms associated with cerebral AVM, the incidence became very small. In one series, Meisel found that 4% of the proximal aneurysms were larger than 10 mm, whereas 12% of the intranidal ones were larger than 10 mm¹⁵.

Boyd-Wilson in 1959³ first described a patient harboring an aneurysm, which was 1.8 cm in diameter and was associated with a cerebral AVM. Although by definition the aneurysm was not strictly a giant aneurysm. Subsequently there were a few more case reports published,

Figure 2 Lateral view of the venous phase of the cerebral angiogram after embolization of the cerebral AVM, showing blood-contrast level within the giant aneurysm, indicating immediate cessation of blood flow within the aneurysmal lumen.

Figure 3 CT image of the brain obtained 48 hours post-embolization of the cerebral AVM, indicating complete thrombosis of the giant aneurysm. Note the hyperdense glue-lipidol mixture within the AVM nidus.

Figure 4 A) Frontal and B) lateral views, obtained two weeks post-embolization of the cerebral AVM. The giant aneurysm was no longer opacified with contrast. A small residual AVM was still present.

including the one by Cunha E Sa et Al. in 1992⁶, in which he described three cases of giant aneurysms associated with cerebral AVM in their series of 39 patients. Two of the giant aneurysms were of proximal type and one belonged to the dysplastic type.

More recently, Bapuraj reported a case of spontaneous thrombosis of a giant aneurysm associated with AVM, and he classified the lesion as intranidal type⁷.

The treatment strategy for patients who have both intracranial aneurysms and AVM, especially for those who have unruptured aneurysms, is still controversial. For patients who presented with haemorrhage, most will agree on the strategy of treating the lesion that was responsible for the haemorrhage¹⁰. If bleeding is suspected to be due to rupture of the concurrent aneurysm, the aneurysm is treated first. Conversely, if the AVM is the suspected culprit, the treatment is aimed primarily at the AVM. However, if the source of bleeding cannot be determined because of the close proximity between the AVM and the aneurysm, then treatment strategy is controversial^{10,15}, yet we always treated the AVM first (targeted embolization) and never observed secondary aneurysm rupture.

In patients with unruptured aneurysms like the one in our case, the therapeutic decision is also debatable. In a large series involving 149 proximal aneurysms associated with cerebral AVM, with embolization targeted towards the AVM only, there was no rupture of any untreated proximal aneurysms after partial embolization of the AVM¹⁵. Based on these facts and long lasting experience, we embolized the cerebral AVM first, leaving the proximal giant aneurysm untouched. Another consideration was the relatively lack of significant mass effect imposed by the giant aneurysms, implying it was not a quickly enlarging lesion and was not in impending rupture state.

The immediate occurrence of stasis of blood flow within the giant aneurysm after successful embolization of the AVM in our patient clearly demonstrated the fact that we could eliminate the shear stresses related to the blood passing into the "flow-related" aneurysm once the AVM was treated. After removal of the abnormal haemodynamic stress that we believed to be a major etiologic factor in the development of this aneurysm, spontaneous thrombosis and subsequent regression was expected to follow. There were several studies demonstrated similar aneurysmal regression after surgical resection¹⁶⁻¹⁷ or embolization⁸⁻¹⁵ of a distal AVM.

The percentage of spontaneous thrombosis in giant aneurysms increased with size and varied between 52.4% and 83%¹⁸, which is much higher than that of the smaller aneurysms. Therefore, the fact that in our case the concurrent aneurysm was giant in nature can be an-

Figure 5 A) CT control 1 year later. B-D) lateral views. The giant aneurysm is no longer opacified and has completely shrunk. A small residual AVM is still present.

other favorable factor for the occurrence of spontaneous thrombosis after treatment of the distal AVM.

The importance of follow-up CT scan and cerebral angiogram to demonstrate complete thrombosis of the giant aneurysm needs to be stressed, as partial thrombosis of the aneurysm or recanalization of the thrombosed lumen does not protect against subarachnoid haemorrhage¹⁷. On follow-up the thrombosis and shrinkage of the giant aneurysm was complete, then further treatment to prevent aneurysmal rupture is not warranted.

Conclusions

The immediate stasis of blood flow and subsequent complete thrombosis of the unruptured flow-rate giant aneurysm after embolization targeted at the associated cerebral AVM eloquently demonstrated the fact that the abnormal haemodynamic stress inducing these aneurysms could be corrected with primary AVM treatment.

This avoided the need to intervene on the giant aneurysm either surgically or endovascularly and thus reduced the unnecessary associated operative risk to the patient.

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