Re-Growth of a Posterior Inferior Cerebellar Artery Aneurysm after Resection of the Associated Posterior Fossa Arteriovenous Malformation

HOSAM AL-JEHANI^{1,2}, DONATELLA TAMPIERI¹, MARIA CORTES¹, DENIS MELANÇON¹

¹ Department of Neuroradiology, Montreal Neurological Institute and Hospital, McGill University; Montreal, Quebec, Canada

² Department of Neurosurgery, King Fahad University Hospital, Dammam University; Al-Khobar, Saudi Arabia

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Summary

Arteriovenous malformation (AVM)-related aneurysms have been described in the literature. Their behavior varies based on their location in relation to and the activity of the shunting through the index AVM. The intuitive expectation supported by numerous reports is that these aneurysms should regress if the AVM is excluded from the circulation.

We describe a case of 46-year-old man who presented with a posterior fossa AVM with an aneurysm on the posterior inferior cerebellar artery feeding the AVM. The nidus of the AVM was successfully excluded by glue embolization, with initial regression of the PICA aneurysm on serial imaging. Five years after the endovascular treatment, the aneurysm showed significant re-growth necessitating endovascular coiling. This case presents the re-growth of an AVM-related aneurysm and emphasizes the importance of long-term follow-up of such aneurysms even if the AVM is completely excluded.

Introduction

The association of aneurysms and arteriovenous malformations (AVMs) has been reported in 2.3-16.7% of patients harboring an AVM^{1,2}.

Posterior inferior cerebellar artery (PICA) aneurysms associated with AVMs are uncommon lesions. Only 29 cases have been reported

in the literature since the first case was described in 1956³⁻²². The hemodynamic stress due to the increased blood flow caused by the presence of the AVM seems to play a major role in the development of these aneurysms²³. If this hemodynamic stress were reduced after resection of the AVM then one would suspect that the flow-related artery aneurysms should resolve spontaneously²⁴. This would have important implications with respect to treatment options.

We report a case in which a flow-related aneurysm located at the PICA origin feeding an AVM initially regressed in size after complete embolization followed by surgical resection of the AVM, prompting an observatory strategy. Five years later, follow-up vascular imaging revealed re-growth of the aneurysm to the same size as previously seen without any recurrence of the original AVM. We review the literature on this phenomenon and summarize treatment recommendations.

Case Report

This 46-year-old left-handed man presented to his family physician with a six-month history of non-specific sensory changes affecting the left hemibody, leg more than arm, and to a lesser degree the left side of the face. The patient experienced no motor weakness in any muscle group and the remainder of the neurological examination was unremarkable. His medical

Figure 1 Initial cerebral angiography. This oblique left vertebral artery injection shows the vermian AVM with the large proximal PICA aneurysm.

Figure 2 Three-year follow-up angiography. This lateral left vertebral artery injection shows a significant reduction of the size of the proximal PICA aneurysm with no residual or recurrent AVM.

Figure 3 Five-year follow-up CT angiography. This CT angiography demonstrates a significant increase in the size of the proximal PICA aneurysm with no evidence of recurrent AVM.

Figure 4 Post-coiling cerebral angiography. This oblique left vertebral angiography shows the coiling of the PICA aneurysm with preservation of the parent vessel.

history was significant for diabetes on dietary control and no positive family history was elicited of any vascular malformation or strokes. Plain CT and CT Angiogram (CTA) revealed an inferior vermian arteriovenous malformation with a flow-related aneurysm at the left

PICA origin. Conventional DSA angiography confirmed the presence of the aneurysm, which measured 11 mm in diameter, and demonstrated the ectasia of the left PICA as a unique feeder to the AVM.

The AVM was surgically excised via a sub-

Table 1 Classification systems for AVM-related aneurysms

Arteriovenous malformation-associated aneurysm grouping of Hayashi et al²⁶.	
Group A	Aneurysms located on a major feeding artery itself
Group B	Aneurysms located on a proximal portion of the feeding system
Group C	Aneurysms located on arteries hemodynamically unrelated to the AVM
Cunha et al. categories of aneurysms associated with arteriovenous malformations²⁷	
Type I	Proximal on ipsilateral major artery feeding the AVM
Type IA	Proximal on major artery related but contralateral to the AVM
Type II	Distal on superficial artery feeding the AVM
Type III	Proximal or distal on deep artery feeding the AVM ("bizarre")
Type IV	On artery unrelated to the AVM

occipital craniotomy 24 hours after glue embolization of the AVM. It was difficult to clip the aneurysm primarily as the neck anatomy with the exiting PICA posed considerable risk of parent vessel occlusion. Treatment options were discussed with the patient and because the aneurysm was felt to be flow-related it was decided to monitor the size of the aneurysm with vascular imaging (conventional DSA and CTA). An angiogram performed three months postoperatively showed no residual AVM and a slight reduction in aneurysm size, and angiography three years later showed a 75% reduction in the size of the aneurysm. (Figure 2).

During the years the patient was in good and stable neurological condition. Follow-up CTA at five years after resection of the AVM interestingly revealed a re-growth of the aneurysm to 10.2 mm, similar to the size the lesion had when the AVM was present (Figure 3). In consideration of this unusual evolution the treating interdisciplinary vascular team recommended coiling of the aneurysm. The patient was submitted to an uneventful endovascular coil embolization of the aneurysm with preservation of the PICA (Figure 4)

Discussion

The association between intracranial aneurysms and AVMs has been reported by a number of authors. Aneurysms are associated with AVMs in 2.3-16.7% of cases. These aneurysms were classified into type I which are dysplastic or remote, unrelated to the inflow vessel (24%), type II which are proximal, arising at the circle of Willis at the origin of a vessel sup-

plying the AVM, type III which are pedicular, arising from the distal portion of the feeding pedicle (57%), and type IV which are intranidal, within the AVM nidus itself (19%)²⁵.

Other classifications proposed earlier failed to include the intranidal type therefore they were considered incomplete, particularly for the evaluation of the morbidity of the lesion (Table 1)^{26,27}.

Thompson et al. reported an annual risk of bleeding of 7% when an association of an aneurysm and a brain AVM is documented²⁸. No relation was found to the size of the aneurysm or the feeding vessel harboring the aneurysm. The primary event was an intracranial bleed, which was found in 51% of the patient population studied. Of these intracranial bleeds, the AVM was the culprit of the hemorrhage in 68% of the cases while in 22% of cases the aneurysm was the source of the bleed²⁸. With respect to PICA aneurysms associated with AVMs, Kaptain et al. found that 81% of this subset of aneurysms are located distally along the course of the PICA and 84% of these patients presented with subarachnoid hemorrhage²⁹.

Our patient presented with an interesting evolution of a proximal PICA aneurysm related to an AVM. The initial regression of this proximal aneurysm following the excision of the AVM is unusual. In the literature proximal aneurysms rarely regress completely (4%), and the size decreased in only 17% of cases. Distal flow-related aneurysms regress in 80% of cases with complete AVM obliteration³⁰. In the past many authors had recommended the need to treat the aneurysms first because of the notion that they could play a major role in bleeding and the fear of increased risk of bleeding from the aneurysm

following obliteration of the AVM due to a sudden increase in vascular resistance¹. This initial regression of the PICA aneurysm prompted us to recommend a conservative strategy with regular vascular imaging follow-up. Despite its initial regression in size, the PICA aneurysm showed re-growth to a size similar to that seen when the AVM was present, raising several hemodynamic possibilities.

Three theories have been suggested to explain the pathogenesis of the association between aneurysms and AVM³¹. The first, by Boyd-Wilson in 1959, suggested that AVM-associated aneurysms were a coincidental occurrence³². This theory has largely been disregarded because the incidence of aneurysms in patients with AVMs is significantly greater than in the general population³³. Anderson and Blackwood proposed a second theory which suggested that a congenital defect is responsible for the development of the AVM and the aneurysm³⁴. This form of "progressive multiform angiogenesis" is theoretically possible, but to date there has been no direct evidence for a specific developmental defect or responsible gene to explain the association between AVMs and aneurysms³⁵. The hyperdynamic theory proposed by McKissock and Paterson in 1956 suggests that aneurysm formation is due to the increased hemodynamic stresses placed on the walls of major AVM feeding vessels¹⁴.

Various treatment strategies have been proposed to treat this uncommon association. We think that if the aneurysm is likely not the source of bleeding based on the blood distribution on CT, the treatment plan should be directed towards treatment of the AVM, unless the aneurysm is accessible via same approach, whether endovascularly or through an open microsurgical technique²⁶. This was reported to have been done safely with observation of regression of the aneurysm as early as ten days post-treatment of the AVM in both the anterior and posterior circulation^{23,29,36}. On the other hand, and in fear of progression and rupture, at least in the short-term, Lasjaunias et al. proposed the utilization of a nitroglycerin drip post obliteration of the AVM to avoid the stump effect (transient hyperpulsation) by maintaining the blood pressure between 90-110 mmHg for the initial 48 hours after treatment of the AVM². This is supported by the recent work of Gao et al. showing that the predicted pressure changes after the obliteration of the AVM are relatively minor near the circle of

Willis are but are more profound along the feeding vessels approaching the nidus level³⁷.

The significant regression noted at the three-year follow-up conventional DSA and later re-growth at the nine-year control of this proximal flow-related PICA aneurysm raises several possibilities and interesting theories. The initial regression is a true phenomenon based on the serial gold standard imaging using digital subtraction angiography. To attribute the lack of visualization as the result of partial aneurysm thrombosis is a hypothesis that should be considered. However, we do not believe that any thrombosis occurred since the patient remained clinically stable throughout the years and none of the vascular imaging controls (CTA or conventional DSA) were triggered by a change in the clinical condition. However such "ghost aneurysms" have been reported in the literature and represent a diagnostic and therapeutic dilemma³⁸. We attribute the progressive decrease in the size of the aneurysm to the regression of the blood flow through the PICA since the excision of the AVM reduced blood flow and led to a reduction of the wall shear pressure on the PICA and on the aneurysm. This mechanism is supported by the progressive reduction in size of the PICA concomitant with the reduction in the size of the aneurysm. The explanation of the delayed re-growth or re-expansion of the aneurysm could be hypothesized as the result of the long-term expansion of the aneurysm with a weak wall submitted to the wall shear stress imposed by the upstream blood flow in the parent vessel³⁹. We postulate that the aneurysm re-expanded in spite of the reduction of the blood flow and shear stresses at the PICA origin because its wall is weak and unable to contract due to the loss of physiological reactivity. In this particular case, in which the initial driving force for maintenance of the aneurysm, namely the AVM, was excised, we raise the possibility that chronic submission of the aneurysm to such a wall stress resulted in a long term failure in the integrity of the blood vessel wall element leading to the re-expansion observed⁴⁰.

This report is the first to illustrate the unusual evolution of a flow-related proximal PICA aneurysm, which re-expanded following initial regression after resection of the associated AVM. We analyze the possible mechanisms responsible for this behavior, certainly the weakened wall of the aneurysm, which cannot tolerate normal in flow pressure in the long-term after initial transitory improvement seems to

be the most plausible theory. We emphasize the significant implication of this phenomenon and the need for continuous follow-up of this type of patient with residual or untreated vascular pathology regardless of their initial behavior. We recommend regular follow-up, well beyond five years to ensure adequate monitoring of the aneurysm and appropriate treatment if lack of regression or expansion of these lesions is noted. Conventional DSA is a valuable investigation, however since these patients must undergo several long-term follow-up examinations, CTA or magnetic resonance angiography (MRA) can be the non-invasive modality to monitor the aneurysm in the long-term, reserving conventional DSA for cases of suspicion or re-intervention.

Conclusion

The association between PICA aneurysms and AVMs of the posterior fossa is uncommon. The re-growth of this type of flow-related aneurysm is reported and the possible mechanisms responsible for it are evaluated. We recommend regular long-term follow-up vascular imaging in these cases to assess the evolution of the lesion and plan for treatment if re-expansion is noted.

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Donatella Tampieri, MD, FRCPC
 Department of Diagnostic
 and Interventional Neuroradiology
 Montreal Neurological Institute/Hospital
 3801 University St.
 Montreal, QC, Canada
 Tel: 1-514-398-1908
 Fax: 1-514-398-7213
 E-mail: donatella.tampieri@mcgill.ca