Angiographically occult, progressively expanding, giant vertebral artery aneurysm

Case report

RICARDO J. KOMOTAR, M.D., J MOCCO, M.D., SEAN D. LAVINE, M.D., AND ROBERT A. SOLOMON, M.D.

Department of Neurological Surgery, Neurological Institute of New York, Columbia University College of Physicians and Surgeons, New York, New York

Hunterian ligation is a well-known treatment for complex aneurysms not amenable to direct microsurgical clip application. After proximal parent vessel occlusion, cerebral angiography is typically used to confirm aneurysm thrombosis. The authors report on a vertebral artery (VA) aneurysm that had progressively expanded and caused brainstem compression after hunterian ligation, despite nondiagnostic findings on both conventional and computed tomography (CT) angiography at multiple time points.

This 64-year-old woman underwent hunterian ligation of a 1.8-cm VA aneurysm at the origin of the right posterior inferior cerebellar artery. An immediately postoperative conventional angiogram and follow-up CT angiograms obtained 5 and 6 years postligation confirmed complete obliteration of the lesion. Nine years after the initial surgery, however, the patient experienced neurological deterioration. Although CTs showed substantial aneurysm enlargement together with pontine compression, angiograms once again demonstrated complete right VA occlusion with no retrograde filling of the aneurysm. On reexploration, the aneurysm was effectively debulked, clipped, and obliterated. Arterial bleeding was found in the lesion neck, as was evidence of microrecanalization.

Hunterian ligation for complex aneurysms carries the risk of microrecanalization and lesion expansion despite nondiagnostic angiography. Although this ligation procedure remains a viable treatment option in carefully selected patients, an extended follow-up evaluation period may be required even when imaging suggests aneurysm obliteration.

KEY WORDS • expanding giant aneurysm • vertebral artery • angiography

MICROSURGICAL clip application is the optimal method for definitive aneurysm treatment. Clip-assisted reconstruction, however, may not be technically feasible because of aneurysm size, location of critical branch vessels, and presence of thrombus or calcified atheroma in the lesion neck. In these complex cases, hunterian ligation is a well-known alternative treatment. With this approach, parent vessel occlusion proximal to the lesion promotes aneurysm thrombosis and relies on retrograde flow to maintain adequate perfusion. To confirm lesion obliteration or detect residual aneurysm, cerebral angiography is performed postoperatively; complete thrombosis represents the definitive cure. Here, we present the case of a VA aneurysm that progressively expanded and caused brainstem compression after hunterian ligation, despite nondiagnostic findings on both conventional and CT angiography at multiple time points.

Abbreviations used in this paper: CT = computed tomography; MR = magnetic resonance; PICA = posterior inferior cerebellar artery; VA = vertebral artery.
She remained asymptomatic for 5 years, but then began to experience progressive gait instability. Head CTs obtained at this time demonstrated a 2-cm calcified lesion impinging on the right side of the brainstem, just distal to the aneurysm clip. Additional CT angiograms—obtained at this time, and another 1 year later—revealed complete lesion thrombosis with only a small lumen of retrograde flow supplying the PICA. Without a clear indication for surgery, the patient was conservatively treated and closely monitored.

Three years later, however, she experienced progressive neurological deterioration, including gait instability, right facial nerve palsy, and difficulty swallowing. Head CTs obtained at another hospital showed massive aneurysm enlargement, with a 3.2-cm calcified mass compressing the midpons. The patient was transferred to Columbia University Medical Center, where MR imaging and cerebral angiography results once again demonstrated complete occlusion of the right VA with no retrograde filling of the aneurysm (Fig. 2). At this point, surgical decompression of the lesion appeared to be the most appropriate therapeutic option.

**Operation.** The patient was taken to the operating room for exploration and aneurysm clip application. Once exposed, the lower cranial nerves were seen to be draped around the large mass, with cystic changes in the right facial nerve and scarring of the ninth, 10th, and 11th cranial nerve complex to the dome of the aneurysm, which was deeply imbedded in the pons. The aneurysm was effectively debulked, leaving only a tiny rim of tissue attached to the brainstem. Notably, arterial bleeding was found in the lesion together with evidence of microrecanalization. The aneurysm was then successfully clipped and obliterated, leaving the PICA patent.

**Postoperative Course.** After surgery, the patient experienced some minor worsening of her neurological status, with increased facial weakness and difficulty swallowing. However, she was uneventfully extubated on postoperative Day 2, and her neurological condition rapidly improved thereafter. At the time of transfer to a rehabilitation program, she was able to handle a liquid diet and ambulate with minimal assistance.

**Discussion**

Optimal treatment of cerebral aneurysms requires their exclusion from the intracranial circulation. However, certain aneurysms cannot be successfully clipped or embolized because of atherosclerosis or calcification within the lesion neck, inaneurysm thrombosis, or anatomical factors such as a wide aneurysm neck. 

In these complex cases, proximal arterial (hunterian) ligation is an acceptable alternative. This technique, which involves occlusion of the parent vessel proximal to the lesion, promotes aneurysm thrombosis and relies on retrograde flow to maintain adequate perfusion. Subsequent lesion obliteration is confirmed on follow-up cerebral angiography, with absent aneurysm filling considered to represent a definitive cure.

We present the unique case of a VA aneurysm that progressively expanded after hunterian ligation despite persistent negative angiography findings at multiple time points. In the patient in the present report, complete vessel occlusion without retrograde aneurysm filling was demonstrated twice on conventional angiography and twice on CT angiography during a 9-year period. Nonetheless, the aneurysm continued to expand, leading to brainstem compression and eventual neurological deterioration. After surgical reexploration, lesion debulking, and direct microsurgical clip application, the patient recovered. It is important to note that brisk arterial bleeding was found at the neck of the aneurysm.

Hunterian ligation, if performed correctly, can be expected to lead to high rates of aneurysm thrombosis in a relatively short period of time. In one of the largest series to date, Steinberg and colleagues reported an 87% complete and a 13% incomplete rate of giant VA aneurysm thrombosis after ipsilateral parent vessel occlusion, usually within a few months. 

Incomplete thrombosis was found to have minimal therapeutic benefit, however: 67% of patients with incomplete thrombosis suffered neurological complications, 86% of which were fatal. Thus, anything short of complete aneurysm thrombosis after hunterian ligation may be indicative of treatment failure and warrants further evaluation.

Although complete aneurysm thrombosis after hunterian ligation has traditionally been considered the definitive cure, it is important to note that authors of several studies have documented recanalization with or without subsequent lesion rupture or enlargement, even after complete thrombosis of giant aneurysms. Recanalization eventually became evident on cerebral angiography in each of these cases—an outcome substantially different from that in the...
the precise mechanism responsible for the growth. Review pathological specimens of thrombosed aneurysms, hypothesized that not the possible role of the vasa vasorum in such cases, because these methods completely exclude lesions from the intracranial circulation. Unfortunately, this option was not applicable in the present case, given that the origin of the PICA at the base of the aneurysm prevented such an approach.

Although intracranial aneurysm enlargement is commonly attributed to repeated hemodynamic stress on the vessel wall, the precise mechanism responsible for the growth of thrombosed aneurysms with mass effect has yet to be clearly described. On reviewing the CT and MR imaging characteristics of such lesions, Schubiger and colleagues postulated that highly vascularized walls of giant intracranial aneurysms are analogous to chronic subdural hematoma membranes in that they undergo recurrent hemorrhages that eventually cause aneurysm growth. Nagahiro and colleagues reviewed pathological specimens of thrombosed giant VA aneurysms and suggested that the development of intrathrombotic capillary channels can lead to lesion expansion. Recently, Iihara and associates noted the possible role of the vasa vaso-ram in such cases, because the results of histological examination after definitive resection of a partially thrombosed aneurysm revealed inflammatory cells and neovascularization with proliferation of the vasa vaso-ram. Additionally, Kwan and colleagues hypothesized that either subtotal aneurysm thrombosis or recanalization may allow arterial pulsations to be continuously transmitted within the lumen, thereby gradually weakening the walls of the aneurysm through this so-called water-hammer effect. Although each of these factors may have been present to some degree in the currently featured case, which differs from those previously reported, this latter mechanism is, in retrospect, the most favored; that is, microrecanalization with brisk arterial blood flow was the most likely cause for aneurysm growth.

Conclusions

In summary, data in the present case illustrate a potential pitfall of hunterian ligation for complex aneurysms: lesion expansion may occur despite nondiagnostic findings on both conventional and CT angiography. Furthermore, our intraoperative findings support microrecanalization as a key factor in the enlargement of angiographically occult aneurysms. Although hunterian ligation remains a viable treatment option in carefully selected patients, extended follow-up evaluation may be required even when imaging results suggest complete aneurysm obliteration.

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Address reprint requests to: Robert A. Solomon, M.D., The Neurological Institute, Department of Neurosurgery, Columbia University Medical Center, 710 West 168th Street, Room 439, New York, New York 10032. email: ras5@columbia.edu.