

Case Report

Medial lenticulostriate artery aneurysm presenting with isolated intraventricular hemorrhage

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Abstract

Background: Isolated intraventricular hemorrhage (IVH) secondary to lenticulostriate artery aneurysm rupture is extremely rare. Thus, the diagnostic imaging modalities and therapeutic interventions utilized in the management of such cases are not clearly defined.

Case Description: Here we describe a case of isolated or primary IVH (PIVH) in a 71-year-old woman presenting with severe headache. Emergent catheter cerebral angiography, performed after nondiagnostic computed tomography angiography (CTA), revealed the bleeding source to be a 4 × 2.6 mm distal medial lenticulostriate artery aneurysm that ruptured directly into the lateral ventricle. The poorly accessible location of the aneurysm for both endovascular and direct surgical treatment argued for conservative management. A good clinical outcome was obtained with rapid angiographic resolution of the ruptured aneurysm.

Conclusion: Thus, lenticulostriate artery aneurysm rupture must be given diagnostic consideration in cases of isolated IVH. Emergent catheter cerebral angiography should be performed in cases such as this when noninvasive imaging is unrevealing. Conservative management may be a reasonable therapeutic option in patients with this kind of aneurysm, and spontaneous resolution can be observed.

Key Words: Angiography, cerebral aneurysm, fibromuscular dysplasia, intraventricular hemorrhage, lenticulostriate artery, pseudoaneurysm

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INTRODUCTION

Isolated or primary intraventricular hemorrhage (PIVH) is defined as bleeding within the ventricles of the brain without associated parenchymal or subarachnoid hemorrhage demonstrated on noncontrast head computed tomography (CT).^[2,3,7,8] PIVH is uncommon

and reported to account for only 2–4% of all intracranial hemorrhages.^[3,5,8] Aneurysms are second only to arteriovenous malformations (AVM) as the bleeding source in cases of PIVH.^[5,37] Other etiologies of PIVH include intraventricular tumor, venous thrombosis, coagulopathy, moyamoya disease, hypertension, or trauma.^[3,7,8,23,30,34] Here we present a rare case of PIVH

resulting from rupture of a medial lenticulostriate cerebral aneurysm. Also, the utility of emergent catheter cerebral angiography for diagnosis and guiding the management of this case is demonstrated.

CASE REPORT

History and examination

The patient was a 71-year-old woman with a history of hypertension, hyperlipidemia, congestive heart failure, and atrial fibrillation who presented to the Emergency Department (ED) after 1 day of severe headache. The headache was acute in onset and associated with one episode of vomiting. No history of trauma, loss of consciousness, visual changes, seizure activity, language deficit, or motor weakness was elicited. The patient had a blood pressure of 116/59 and the neurological examination was nonfocal. Initial laboratory studies showed an INR of 3.0 consistent with her history of daily warfarin therapy for atrial fibrillation. Noncontrast head computed tomography (CT) revealed symmetric IVH with moderate blood in the lateral, third, and fourth ventricles [Figure 1a and b]. No parenchymal or subarachnoid blood was present. Computed tomographic angiography (CTA) revealed a 4-mm right-sided middle cerebral artery (MCA) M2 segment aneurysm which was thought to be an unlikely source of the hemorrhage given the absence of adjacent parenchymal or subarachnoid blood.

Management

Warfarin was withheld and initial pharmacological therapy included intravenous administration of 10 mg vitamin K and 4 U fresh frozen plasma (FFP) to reduce the INR below 1.5. Because the patient remained neurologically intact and had no evidence of hydrocephalus, no extraventricular drain was required. Catheter cerebral angiography was performed revealing the hemorrhage source to be a 4 × 2.6 mm aneurysm of a distal medial lenticulostriate artery arising from the proximal A1 segment of the right anterior cerebral artery (ACA) [Figure 1c and d]. Diagnostic considerations included pseudoaneurysm, mycotic aneurysm, or degenerative lesions such as a Charcot–Bouchard aneurysm. Additional angiographic findings included two saccular right-sided MCA aneurysms (4 mm and 2 mm), one fusiform aneurysm of the right-sided MCA posterior temporal branch (1–1.5 mm), and fibromuscular dysplasia (FMD) of the extracranial carotid and vertebral arteries [Figure 2]. Further clinical evaluation including echocardiography and blood cultures to rule out septic embolism secondary to endocarditis was unrevealing.

Clinical course

As the distal location of the ruptured aneurysm within the ventricular wall made it a poor candidate for surgical or endovascular treatment without arterial sacrifice,

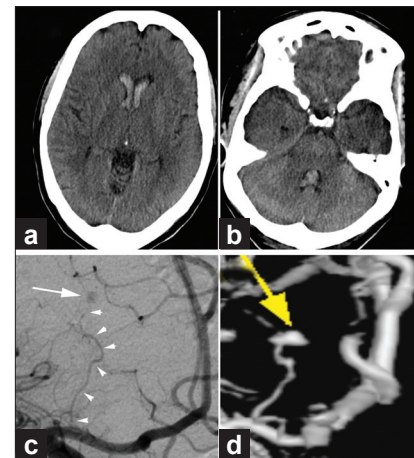


Figure 1: CT and angiographic evaluation. Nonenhanced head CT demonstrates acute hemorrhage within the frontal horns of the lateral ventricles (a) and in the fourth ventricle (b). Catheter cerebral angiography with a right internal carotid injection in the oblique view shows aneurysmal dilatation along the distal aspect (arrowheads) of a lenticulostriate artery arising from the A1 segment of the right anterior cerebral artery (ACA). Three-dimensional vessel reconstruction (d) clearly demonstrates the origin of the parent medial lenticulostriate vessel from the horizontal segment of the ACA with aneurysmal dilatation distally (arrow)

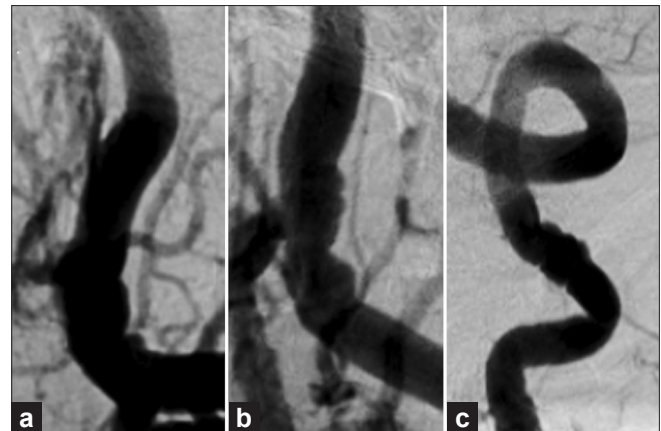


Figure 2: Angiographic fibromuscular dysplasia. A characteristic “string of beads” appearance is seen in the right (a) and left (b) cervical internal carotid arteries in the lateral view after an internal carotid artery contrast injection. Similarly, vessel contour irregularity is also demonstrated in the cervical vertebral artery in AP view after a left vertebral contrast injection

conservative management was thought to be the most appropriate. The patient remained neurologically stable in the Neurological Intensive Care Unit (NICU) and did not develop hydrocephalus. Repeat catheter angiography 5 days after initial presentation showed no evidence of the previously noted distal medial lenticulostriate aneurysm [Figure 3a]. The patient was discharged 8 days after initial presentation with an intact neurological exam. The medicine consult service recommended treatment of her atrial fibrillation with aspirin 325

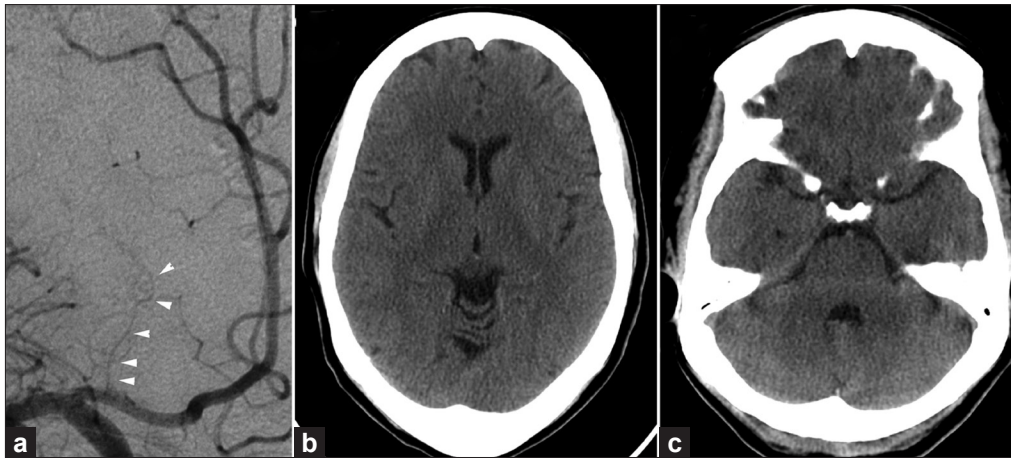


Figure 3: Follow-up imaging. Catheter angiography performed 5 days after initial presentation demonstrates the interval resolution of the aneurysmal dilatation of the distal tip of the right medial lenticulostriate artery (arrowheads; a). Repeat noncontrast head CT performed 2.5 months after discharge shows the resolution of the prior intraventricular hemorrhage (b and c)

mg daily and discontinuation of anticoagulation with warfarin. A follow-up noncontrast head CT performed 2.5 months after discharge showed no stigmata of the IVH [Figure 3b and c].

DISCUSSION

Isolated IVH is rare and only one previous report in a patient with Moyamoya disease has been ascribed to distal lenticulostriate artery (LSA) aneurysm rupture.^[16] Parenchymal and subarachnoid hemorrhages are more commonly associated with LSA aneurysm rupture.^[1,4,6,9,11-17,19,20,22,24,25,27-29,31,33,35,36] Similar to other types of intracranial hemorrhages, PIVH presents with acute onset of severe headache, altered sensorium, vomiting, vertigo, focal neurologic deficits, coma, or sudden death.^[2,3,7,8,10,21,30] Mortality rates vary by etiology and may be as high as 40%.^[2,5] Obstructive or communicating hydrocephalus occurs in 62% of cases and is an indicator of poor prognosis.^[2,5,8,30] In their systematic review of the literature, Flint *et al.*^[5] found that up to 34% of PIVH cases develop hydrocephalus requiring temporary external ventricular drainage although a permanent ventriculoperitoneal shunt was required less often.^[10]

As demonstrated in this case, catheter cerebral angiography was essential to determine the underlying lesion resulting in PIVH. Flint *et al.*^[5] found that catheter angiography was utilized in about 52% of PIVH cases with the identification of a bleeding source in 56%. Similarly, catheter cerebral angiography had a yield of 65% in a prospective series of PIVH patients by Zhu *et al.*^[37] Based on these studies it was suggested that routine catheter angiography may be warranted in all cases of PIVH. Indeed the inconclusive CTA findings in our case provided additional justification for use of catheter cerebral angiography. Furthermore, catheter angiography

enabled diagnosis of concurrent fibromuscular dysplasia (FMD) and multiple unruptured cerebral aneurysms in the patient we have reported.

The identification of FMD in this patient raised the question of whether pseudoaneurysm rupture was responsible for the unusual hemorrhage in this case. Pseudoaneurysms of the cerebral vasculature, which are defined by the absence of all three vessel wall layers (intima, media, and adventitia), most commonly result from trauma but have been reported to occur spontaneously in the setting of FMD.^[18,26,32] The angiographic appearance of cerebral pseudoaneurysms is typically that of a poorly defined globular sac with delayed contrast filling and emptying. The definitive determination of whether our case represents a true or pseudoaneurysm rupture requires pathological analysis of the diseased vessel, which was not available in this case. Nonetheless, the extremely delicate and friable nature of pseudoaneurysms in this location provided additional support for a conservative management strategy.

CONCLUSION

Distal lenticulostriate aneurysm rupture may result in isolated IVH. Catheter cerebral angiography should be performed in cases such as ours when noninvasive imaging does not reveal a source for hemorrhage. Given the relatively inaccessible location of these lesions for direct surgical or endovascular occlusion, conservative management may represent a reasonable therapeutic strategy.

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