CASE REPORT

Subarachnoid hemorrhage then thrombosis of posterior inferior cerebellar artery dissection: is early surgical exploration warranted?

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SUMMARY

The natural history of spontaneous cerebral artery dissection and thrombosis remains uncertain. Concurrent subarachnoid hemorrhage further complicates the therapeutic approach. Thus the best strategy for managing patients with acute vessel thrombosis in the setting of subarachnoid hemorrhage is unclear. Here we present a case of spontaneous posterior inferior cerebellar artery dissection presenting with subarachnoid hemorrhage and acute thrombosis. Although the patient was initially managed conservatively, angiographic follow-up demonstrated recanalization of the diseased vessel, necessitating definitive treatment. Thus we propose that angiographic follow-up is necessary in the management of patients with subarachnoid hemorrhage in association with apparent vessel thrombosis.

BACKGROUND

Dissecting aneurysms of intracranial arteries are rare but may result in significant morbidity and mortality. These lesions may present with ischemia, subarachnoid hemorrhage, or, infrequently, a combination of the two. Dissecting aneurysms appear most commonly in the vertebrobasilar circulation with a documented incidence of 3.2–4.5% among patients presenting with subarachnoid hemorrhage, the majority of which develop on the vertebral or basilar arteries.^{1–2} Posterior inferior cerebellar artery (PICA) dissection represents a small subset of these entities with an estimated incidence of 0.5% of subarachnoid hemorrhage patients.³ Although PICA dissecting aneurysms have been previously reported, they rarely present with vessel occlusion.⁴ To our knowledge, this is the first documented case of a dissecting PICA aneurysm causing subarachnoid hemorrhage that recanalized after presenting as an acute arterial occlusion.

CASE PRESENTATION

The patient is a middle aged individual (age/gender anonymized) with no significant past medical history who presented with severe headache. On examination, the patient was lethargic with a left abducens nerve palsy and increased tone in the lower extremities. Non-contrast head CT showed thick subarachnoid blood, primarily in the right prepontine and premedullary cisterns, along with intraventricular hemorrhage and hydrocephalus (figure 1A). A CT angiogram (not shown) was also performed, revealing absent filling in the right PICA but no definite hemorrhage source. With

external ventricular drainage, the patient's mental status improved. Digital subtraction angiography (DSA) confirmed occlusion beyond the origin of the right PICA (figure 1B), suggesting dissection and thrombosis. Although the very proximal portion of the right PICA initially demonstrated minimal contrast filling, repeat DSA performed 1 week later showed no antegrade flow and persistent thrombosis. MRI and MR angiography were subsequently performed, further supporting the diagnosis of PICA thrombosis (figure 1C). A decision was made to manage the patient conservatively as the diseased vessel demonstrated no significant flow, presumably minimizing the potential for repeat subarachnoid hemorrhage. The patient

Figure 1 Subarachnoid hemorrhage, posterior inferior cerebellar artery (PICA) dissection, and thrombosis. (A) Non-contrast head CT demonstrates subarachnoid hemorrhage in the right prepontine cistern along with intraventricular hemorrhage in the fourth ventricle. Mild hydrocephalus and third/lateral intraventricular hemorrhage was also present (not shown). (B) Right vertebral arteriogram in the lateral projection of the initial digital subtraction angiogram reveals absent antegrade flow in the right PICA (arrow). (C) Axial fluid attenuated inversion recovery MRI through the origin of the right PICA demonstrates a lobular hypointensity (arrow) consistent with a thrombosed dissected vessel. Absent flow was seen through this segment on MR angiography (not shown). (D) Right vertebral arteriogram in an oblique projection 6 months later shows that the right PICA (arrow) has recanalized and that the proximal portion has residual irregularity, consistent with a partially healed dissection.

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Figure 2 Postoperative angiography. (A) Skull radiograph shows the location of the two aneurysm clips (arrows) used to trap the diseased posterior inferior cerebellar artery (PICA) segment. (B) Right vertebral arteriogram demonstrates opacification at the origin (arrow) of the right PICA and no flow within the trapped segment. (C) Distal reconstitution (arrow) of the right PICA is seen to occur in a slightly delayed fashion via collaterals.

ultimately required placement of a ventriculoperitoneal shunt and was discharged home neurologically intact.

Follow-up DSA 6 months after discharge was notable for right PICA recanalization with the proximal 8 mm of the vessel demonstrating areas of focal outpouching, stenosis, and curvilinear filling defects, consistent with dissection flaps (figure 1D). Return of blood flow within this persistently dissected PICA raised concern for recurrent hemorrhage, and therefore operative intervention was undertaken. Using a far lateral suboccipital craniectomy, the aneurysmal portion of the right PICA was identified and trapped without incident. Care was taken to preserve the most proximal PICA segment from which small medullary perforators arose. Postoperative angiography confirmed successful trapping of the dissected PICA segment and distal reconstitution of PICA territory blood flow from collaterals. The proximal PICA demonstrated contrast filling after vertebral artery injection, suggesting that there was adequate flow to brainstem perforators arising from this segment (figure 2). The patient tolerated the procedure well and was discharged home in excellent condition on the third postoperative day.

DISCUSSION

The natural history of thrombosed intracranial vessels secondary to dissection may follow three distinct pathways: (1) the vessel may remain stably thrombosed, (2) it may recanalize with intimal healing,⁵ or (3) it may recanalize without intimal healing.⁶⁻⁷ While dissecting intracranial aneurysms without parent vessel thrombosis are typically surgically occluded acutely, the appropriate management of spontaneously thrombosed dissecting aneurysms is unclear. Dissecting aneurysms are rare entities that predominantly occur in the vertebrobasilar circulation, with isolated PICA dissecting aneurysms comprising an even smaller subset of such lesions.³⁻⁵ As such, the natural history and optimal management strategy for these lesions remains controversial. Two main subtypes of intracranial arterial dissection have been described. Type 1 dissections typically present with vessel occlusion and ischemia, and occur when vessels dissect between the internal elastic lamina and media, causing stenosis of the true arterial lumen. Type 2 dissections often present with subarachnoid hemorrhage and occur within the media or adventitia of the vessel.⁸⁻⁹ Type 2 dissections occur most commonly in the posterior circulation, due to the relatively thin media and adventitia in these vessels.¹⁰

The case presented highlights the management dilemma encountered in treating patients with subarachnoid hemorrhage and acute thrombosis of the dissected vessel. On the one hand,

the thrombosed vessel has effectively occluded the bleeding source, thereby mitigating the need for immediate operative intervention. On the other hand, the stability of the thrombus is unclear, necessitating close angiographic follow-up if conservative management is elected. Indeed, while a significant risk of rebleeding for vertebral artery dissecting aneurysms (from 24% to 65% in some series) has been reported, the low incidence of dissecting PICA aneurysms precludes robust analysis.^{2-6,11-13} A retrospective series by Ramgren *et al* including both vertebral artery and PICA dissecting aneurysms documented early rebleeding (within 12 days) in 31% of patients, but found no outcome differences between patients treated conservatively compared with those managed with endovascular therapy.¹⁴ Another series by Takagi *et al* examining the rebleeding risk due to dissecting vertebral artery aneurysms identified a 35% rebleeding risk, with 6% of rebleeds occurring at times greater than 1 week from initial hemorrhage.¹⁵ These results were echoed in a study by Mizutani *et al* who noted a rebleeding risk of 65%, 70% of which occurred within 24 h of initial rupture. In this study, only 10% of hemorrhages occurred at time points greater than 1 week after the initial rupture.²

The optimal approach to the management of dissecting PICA aneurysms must take into account several factors, including the patient's neurological status at presentation, the angiographic characteristics of the lesion, and the anatomic location of the dissection. In cases of subarachnoid hemorrhage without parent

Learning points

- ▶ Dissecting aneurysms of the posterior inferior cerebellar artery that present with subarachnoid hemorrhage and acute vessel thrombosis are rare entities, with the potential for significant associated morbidity and mortality.
- ▶ The natural history of such lesions is unknown, however, resulting in unclear therapeutic pathways.
- ▶ An initial conservative approach with serial angiographic follow-up is a reasonable and safe strategy for managing this pathology.
- ▶ Definitive vessel occlusion should be performed at the first sign of recanalization if vessel healing has not occurred.
- ▶ Although early operative intervention to permanently obliterate the thrombosed vessel is an alternative strategy, additional studies are needed to support such an approach.

vessel thrombosis, the high risk of rebleeding makes early intervention with either endovascular or open surgical techniques prudent.^{2 11 14–18} Conversely, in cases presenting with ischemia due to thrombosis, conservative measures with selective use of antiplatelet therapy is reasonable.^{3 7 19} In unique cases such as this, where the presentation is a mix of vessel thrombosis and hemorrhage, a nuanced approach must be used. In cases such as this, delayed surgical intervention is a reasonable approach as it may facilitate detailed angiographic characterization of the recanalized vessel, allowing for preservation of critical brainstem perforators.

Contributors JAE is responsible for the conception and design of this manuscript, and is the designated guarantor. NGA and JAE acquired the case data and drafted the original manuscript. PMM and ESC critically revised this article and were responsible for the clinical management of the case presented.

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