CASE REPORT

Unexpected early radiographic findings associated with a ruptured blister-like carotid wall aneurysm

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SUMMARY

A 33-year-old man presented with aneurysmal subarachnoid hemorrhage from a ruptured, blister-type sidewall internal carotid artery (ICA) aneurysm. Balloon-assisted coiling was performed with residual neck. He subsequently developed severe vasospasm requiring intra-arterial therapies on multiple occasions, during which it was noted that despite widespread vasospasm, a focal segment of the ICA at the site of the aneurysm showed no significant spasm, suggesting underlying vessel abnormality. He was discharged without deficit and scheduled for flow diversion given concern over this potentially pathologic segment of vessel. At time of scheduled flow diversion 6 weeks later, a de novo unstable-appearing 6 mm stalk-like pseudoaneurysm was identified in this segment. Both aneurysms and the diseased vessel were successfully treated with Pipeline stenting, with excellent clinical and angiographic result. This case highlights the need for close angiographic follow-up when there is a heterogeneous vasospastic response in arterial segments adjacent to a ruptured aneurysm.

BACKGROUND

Blister-type cerebral aneurysms pose significant challenges and are notoriously difficult to treat. We present the case of a ruptured blister-type aneurysm treated with balloon-assisted coiling with subsequent development of severe vasospasm in which there was disproportionate dilatation adjacent to the aneurysm when compared with the rest of major cerebral vessels. Recognition of this finding prompted a plan for early vascular remodelling of this segment with flow diversion. Such action led to the early identification and treatment of a rapidly growing pseudoaneurysm at this location. Had this feature not been recognized and appropriately triaged to short interval follow-up, planned routine follow-up at 3 or 6 months may have resulted in catastrophic rupture of the rapidly enlarging pseudoaneurysm.

Case presentation

A 33-year-old man presented as a Hunt and Hess grade 4, Fisher grade 3 subarachnoid haemorrhage with associated hydrocephalus (figure 1A). He had a history of cocaine use. On arrival, he was intubated and localizing bilaterally. An external ventricular drain was placed emergently. He was taken for diagnostic angiogram after initial CT angiography revealed an aneurysmal dilation in the region of the

left internal carotid artery (ICA) communicating segment.

INVESTIGATIONS

Catheter angiography demonstrated a 3×3 mm blister-type sidewall aneurysm arising from the proximal left intradural ICA (figure 1B,C). Balloon-assisted coiling was performed using a compliant 4 mm balloon. During placement of the first coil loops, intraprocedural rupture occurred with coil extrusion into the subarachnoid space. The aneurysm dome was successfully coiled with final angiography demonstrating a residual neck (Raymond grade II; figure 1D). The patient began following commands and was successfully extubated. On postrupture day 8, the patient developed symptomatic vasospasm with aphasia resistant to hypertension therapy that necessitated emergent vasospasm treatment with intra-arterial spasmolytic administration and balloon angioplasty. This procedure was repeated four times during the ensuing 11 days due to severe, recurrent, symptomatic spasm. At each treatment, an isolated segment of the left ICA at the site of the aneurysm showed relative focal dilatation when compared with the immediate proximal and distal segments of the carotid artery (figure 2A). This observation suggested an underlying vessel wall abnormality at this site. Balloon angioplasty was therefore not performed at or near this segment due to concern for potential vessel rupture. The patient responded favorably with each endovascular treatment, returning to a normal neurological examination.

The external ventricular drain was weaned successfully and he was discharged home on postrupture day 20 neurologically normal. Prior to discharge, the patient was informed that an additional flow diversion procedure was necessary in the near future in order to definitively treat the aneurysm and to remodel the diseased ICA segment. He was consented and scheduled for Pipeline flow diversion of the diseased ICA segment at 4 weeks and was loaded on dual antiplatelet agents 1 week prior to his procedure.

At the time of the subsequent procedure, 6 weeks after discharge from the hospital, initial angiography demonstrated an irregular de novo 6 mm × 3 mm stalk-like pseudoaneurysm arising from the previously identified segment of abnormal ICA near the previously coiled aneurysm neck (figure 2B,C).

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Figure 1 (A) Presenting CT demonstrating diffuse subarachnoid hemorrhage and hydrocephalus. (B) Angiogram working view showing laterally directed blister-like aneurysm. (C) Angiogram lateral view showing relationship of aneurysm (double-density; arrow) to posterior communicating artery and bulging of the internal carotid artery in the aneurysmal segment (dotted arrow). (D) Postcoiling angiogram demonstrating residual neck with coils in the dome and subarachnoid space (arrow).

TREATMENT

The patient underwent uncomplicated vascular remodelling and aneurysm embolization using a single Pipeline Flex flow diverting stent (Medtronic; Irvine, California, USA).

OUTCOME AND FOLLOW-UP

The patient underwent diagnostic angiography 4 weeks from flow diversion demonstrating complete occlusion of both aneurysms with successful vascular remodelling of the ICA (Raymond grade I; [figure 2D](#)). He remains neurologically intact.

DISCUSSION

Cerebral aneurysms are known to have a 10%–15% risk of delayed recurrence following coil embolization.¹ Although the exact pathophysiologic mechanism accountable for aneurysm recanalization following coiling is not completely understood, coil compaction and/or further aneurysm growth at the neck are largely thought to be responsible. This process usually occurs slowly over a period of months, with 12.7% of completely coiled aneurysms showing recanalization at 6 months.² Recanalization of a coiled aneurysm in the first 8 weeks is rare. Further, rapid de novo aneurysm formation at the site of a ruptured aneurysm immediately following treatment is exceedingly rare.

Histopathologically, blister-like aneurysms consist of a thin adventitia that covers a focal intima and media defect.³ In this case, the patient developed severe vasospasm following

Figure 2 (A) Oblique angiogram during vasospasm treatment showing severe vasospasm of internal carotid artery (ICA) segments proximal and distal to the aneurysm (arrows) and relative lack of spasm at the site of the aneurysm (dotted arrow). (B,C) Oblique view and three-dimensional angiogram at time of elective flow diversion demonstrating a new pseudoaneurysm arising from superior surface of ICA. (D) Repeat oblique angiogram 4 weeks after Pipeline demonstrating complete occlusion of both aneurysms with successful vessel remodelling.

aneurysm rupture, requiring multiple endovascular balloon angioplasty and spasmolytic infusions. During these treatments, it was noticed that the ICA segment at the site of the blister aneurysm remained persistently dilated, unlike the ICA segments both proximal and distal to the aneurysm which had narrowed and undergone spasm, suggesting an underlying vessel wall abnormality at this site. This is further evidenced by the presence of the ruptured blister aneurysm, which are associated with defects in the intima and media.³

The lack of arterial vasospasm occurring at this site can be hypothesized to be due to multiple potential etiologies. One explanation is the diseased ICA segment harbored underlying histopathologic abnormalities such as the absence of media and elastic lamina that allowed for initial blister aneurysm formation. This resulted in absence of spasm secondary to a limited muscular layer, and then allowed for rapid de novo pseudoaneurysm formation from the involved vessel segment. The convex bulging of the ICA adjacent to the aneurysm prior to intervention was also another factor which could suggest underlying vessel disease. Another possible etiology could be secondary to injury to the vessel/dissection via balloon angioplasty during coiling. This may have had a lasting, prolonged effect on muscularis function followed by rapid de novo pseudoaneurysm formation afterwards. This explanation is considered less likely given the widespread and routine use of balloon-assisted coiling for ruptured aneurysms, wherein this finding is exceedingly rare; and a compliant 4 mm balloon was used in a vessel of normal diameter (4.1 mm) at the time of coiling without angiographic evidence of wall change post-coiling. As it was thought that the lack of spasm at this segment was due to underlying vessel wall dysplasia, the patient was

consented for early flow diversion embolization to remodel the diseased vessel wall and definitively treat the aneurysm neck remnant just 6 weeks after discharge. This strategy allowed for diagnosis and treatment of the large, de novo stalk-like pseudoaneurysm that formed adjacent to the blister-like aneurysm. Had this patient undergone standard follow-up imaging schedule (3 to 6 months), it is possible that this patient would

have sustained recurrent subarachnoid hemorrhage from pseudoaneurysm rupture.

This case is novel and interesting for several reasons. First, the rapid formation of a de novo, stalk-like 6 mm pseudoaneurysm immediately adjacent to a blister aneurysm over only a 6-week time period is exceedingly rare. Second, the focal absence of vasospasm at the arterial segment was recognized to represent underlying histopathological vessel wall abnormality and early retreatment with flow diversion was recommended, averting potential catastrophic rupture of the de novo pseudoaneurysm.

Learning points

- ▶ Blister-like aneurysms consist of a thin adventitia that covers a focal intima and media defect.
- ▶ Recognition of focal absence of vasospasm at the segment of vessel containing a blister aneurysm may suggest a wider segment of diseased vessel at this location with predisposition to pseudoaneurysm formation.
- ▶ Early angiographic follow-up and/or vascular remodelling with flow diversion is important when such underlying vessel disease is suspected.
- ▶ In this case, identification of focal, inappropriate spasm reaction allowed for early identification and treatment of a de novo pseudoaneurysm adjacent to the ruptured aneurysm with excellent clinical result.

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