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Case Report

Subarachnoid Hemorrhage Related to a Ruptured Anterior Spinal Artery Aneurysm Associated with Bilateral Vertebral Artery Occlusion

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Objective: We report a rare case of a ruptured anterior spinal artery (ASA) aneurysm caused by bilateral vertebral artery (VA) occlusion.

Case Presentations: A 78-year-old man suddenly developed severe headache and slight hemiparesis, and was admitted to our hospital. Computed tomography (CT) revealed subarachnoid hemorrhage, mainly in the posterior fossa. On emergency angiography, the right VA terminated at the origin of the posterior inferior cerebellar artery (PICA), and anastomoses between the PICA and the anterior inferior cerebellar artery (AICA) were observed, in addition to a saccular 3-mm aneurysm with bleb originating from the PICA-AICA anastomosis. Left vertebral arteriography demonstrated that the left VA was occluded segmentally at the V4 level and revealed a tortuous arterial network filling the distal VA. Based on the location of the bleeding, the right VA aneurysm was considered to have ruptured. After balloon test occlusion of the right VA, parent artery occlusion was performed without complications. The patient had no neurological changes immediately after surgery, but several hours later, he stopped breathing. Retrospective analysis revealed an ASA aneurysm, which was determined to be the bleeding source. Although conservative treatment was performed, he died the fourth day after onset without neurological improvement.

Conclusion: In cases of subarachnoid hemorrhage associated with bilateral VA occlusion, an aneurysm formed by hemodynamic stress may be the source of hemorrhage. It is important to suspect aneurysms in the extracranial collaterals, such as the ASA, and intracranial collaterals such as the PICA-AICA anastomosis.

Keywords ▶ anterior spinal artery aneurysm, subarachnoid hemorrhage, PICA-AICA anastomotic artery, VA rete mirabile

Introduction

Cases in which the source of subarachnoid hemorrhage is a spinal artery aneurysm are rare.[1] Anterior spinal artery (ASA) aneurysms are complicated by vascular anomalies,[1] such as spinal arteriovenous anomalies and coarctation of the aorta, autoimmune disease,[2] angitis,[3] such as connective tissue disease or infection[4] in many cases, but only a few case reports on the rupture of an ASA aneurysm related to bilateral vertebral artery (VA) occlusion have been published.[5–8]

In this study, we report a patient with subarachnoid hemorrhage in whom occlusion of the bilateral VA trunks and a right posterior inferior cerebellar artery (PICA)–anterior inferior cerebellar artery (AICA) anastomotic artery aneurysm were detected, and coil embolization was performed considering it to be the source of hemorrhage, but postoperative detailed examination suggested an ASA aneurysm as the source of hemorrhage.

Case Presentation

A 78-year-old man. Headache, consciousness disorder, and weakness of the right half of the body suddenly developed, and he was brought to our hospital by ambulance. He had a history of myocardial infarction, diabetes mellitus, and

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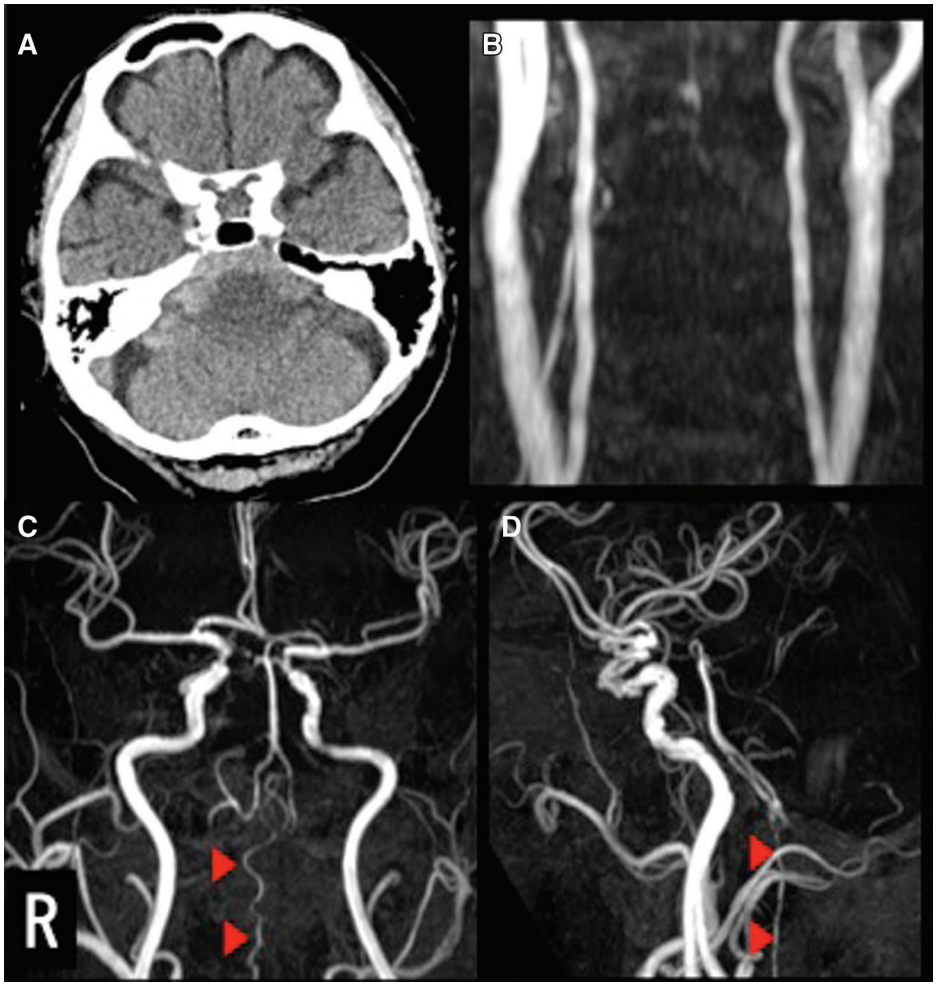

Fig. 1 (**A**) CT revealed subarachnoid hemorrhage, mainly in the posterior fossa. (**B, C, D**) Cervical MRA (**B**) and brain MRA (**C**, anterior view; **D**, lateral view) showed occlusion of the bilateral VA and collateral flow signal via the ASA (arrowhead). ASA: anterior spinal artery; CT: computed tomography; MRA: magnetic resonance imaging; VA: vertebral artery

cerebellar infarction. He previously took biaspirin. On arrival, his Japan Coma Scale (JCS) score was 3 and manual muscle testing (MMT) demonstrated 4/5, suggesting right hemiplegia. Computed tomography (CT) revealed right posterior fossa-dominant subarachnoid hemorrhage (**Fig. 1**). Mild right hemiplegia was noted on arrival and cephalic magnetic resonance imaging (MRI) was performed. However, there was no acute-phase finding as an etiological factor for paralysis other than subarachnoid hemorrhage. On magnetic resonance angiography (MRA), the bilateral cervical VAs were clearly visualized, but there was no continuity into the cranium (**Fig. 1**).

On the same day, emergency digital subtraction angiography (DSA) was performed. Right vertebral arteriography demonstrated occlusion of the V4 VA trunk. An artery reaching the AICA from the right PICA bifurcation adjacent to the site of occlusion and an aneurysm measuring 3 mm at the origin of this artery were observed (**Fig. 2**). On left vertebral arteriography, the distal VA was visualized via a large number of net-like vessels branching from the V4 segment (**Fig. 3**). The aneurysmal site was consistent with the localization of subarachnoid hemorrhage detected on CT and MRI, suggesting aneurysmal rupture-related subarachnoid hemorrhage. Endovascular treatment was performed.

A 5Fr Destination (Terumo Corporation, Tokyo, Japan) was inserted into the right VA and a 4Fr diagnostic catheter was inserted into the left VA. On contralateral angiography occluded at a site proximal to the cerebral aneurysm (V4 right VA) using a Scepter XC (MicroVention, Aliso Viejo, CA, USA), the area distal to the site of right VA occlusion was visualized via the union. We considered embolization of the parent artery (right VA) involving the aneurysm possible. It was embolized until antegrade flow from the aneurysm toward the proximal side of the VA disappeared (Target

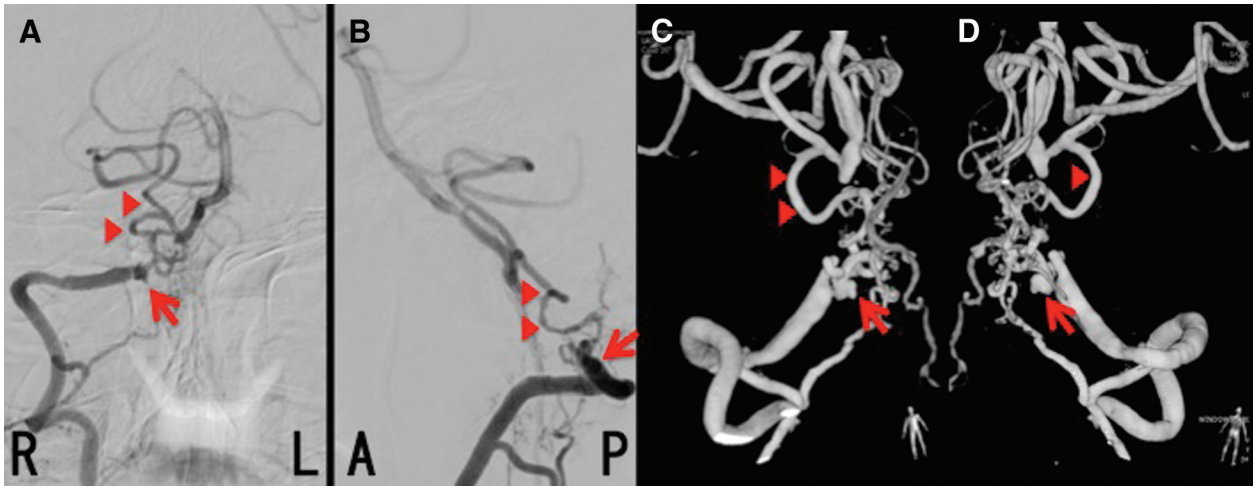

Fig. 2 Rt. vertebral angiography (**A**, anterior view; **B**, lateral view; **C** and **D**, 3D-DSA) showed that the rt. VA terminated at the origin of the PICA and anastomoses of the PICA-AICA were present (arrowhead), in addition to a saccular 3-mm aneurysm with bleb originating from the PICA-AICA anastomosis (arrow). 3D-DSA: three-dimensional digital subtraction angiography; AICA: anterior inferior cerebellar artery; PICA: posterior inferior cerebellar artery

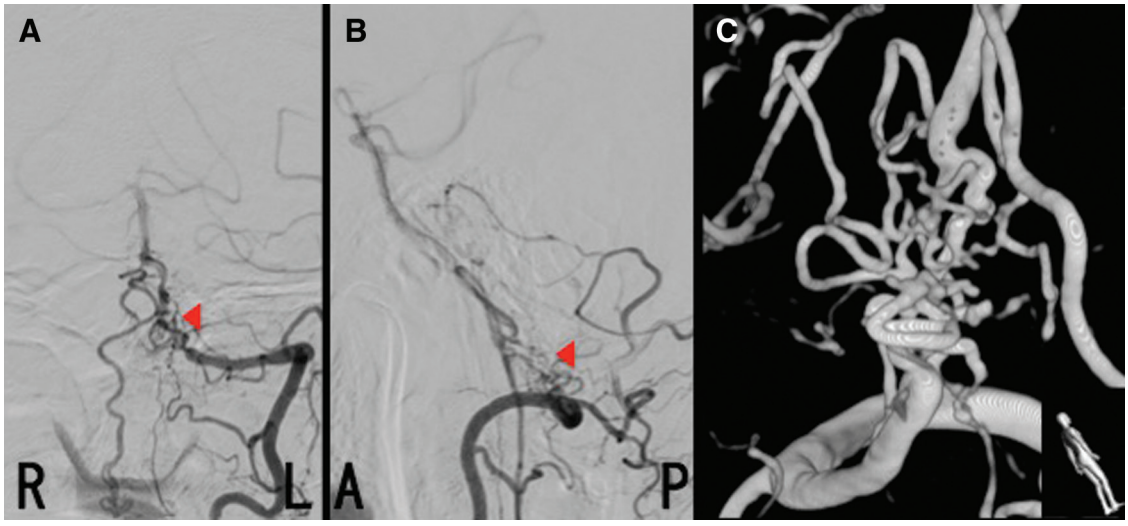

Fig. 3 Lt. vertebral angiography (**A**, anterior view; **B**, lateral view; **C**, 3D-DSA) showed that the lt. VA was occluded at the V4 segment and there was a tortuous arterial network filling into the distal VA (arrowhead). 3D-DSA: three-dimensional digital subtraction angiography; VA: vertebral artery

NANO 2.0 mm × 3 cm: 4, 3.0 mm × 6 cm, 2.0 mm × 4 cm: 2, Stryker, Kalamazoo, MI, USA). On final right vertebral arteriography, the aneurysm was not visualized and the artery reaching the AICA from the PICA at the periphery of the aneurysm was visualized through a collateral pathway mediated by the right C2 radicular artery (**Fig. 4**).

He was stable until 4 hours after embolization when respiratory arrest suddenly developed. No changes in intracranial subarachnoid hemorrhage were noted on emergency CT or MRI. However, extensive hematoma involving the cervical cord and intramedullary to subarachnoid regions was detected (**Fig. 5**). DSA images were reconfirmed. Right vertebral arteriography on parent-catheter insertion and left vertebral arteriography, which was conducted by occluding the right VA with a balloon, revealed the ASA flowing into the cranium via the segmental artery. An ASA aneurysm measuring 8 mm and protruding to the right posterior area at the C3 cervical cord level was visualized (**Fig. 6**). In addition, we reconfirmed emergency cervical MRA findings on arrival. At the same site, an aneurysm was visualized (**Fig. 1**). Thus, the rupture of the ASA aneurysm may have caused subarachnoid hemorrhage, and rerupture of this aneurysm after embolization of the unruptured VA aneurysm may have led to respiratory arrest. The

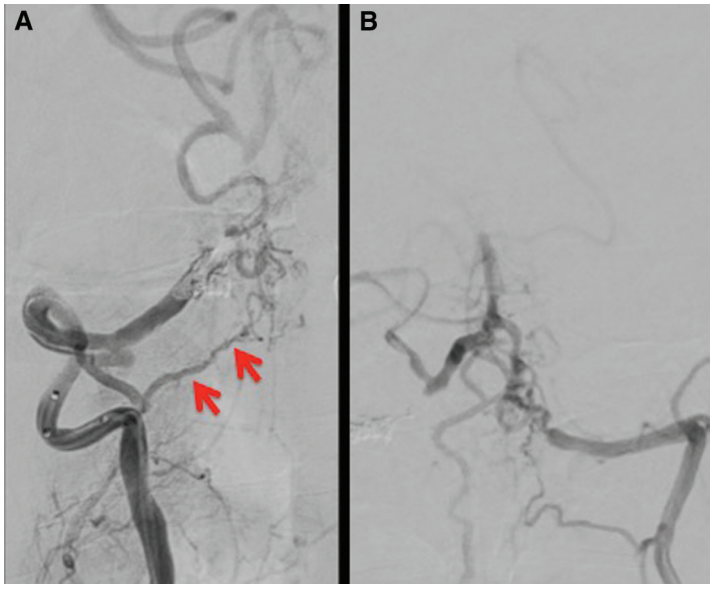

Fig. 4 Rt. VA parent artery occlusion including the aneurysm was performed (**A**, working angle). Antegrade collateral circulation via the radicular artery (arrow) and retrograde collateral circulation from the lt. VA (**B**, anterior view) were observed. VA: vertebral artery

neurological severity was marked and conservative treatment, such as respiratory control, was conducted, but there was no improvement. The patient died 4 days after onset.

Discussion

Ruptured ASA aneurysms are rare. Approximately 50% of spinal artery aneurysms are complicated by spinal arteriovenous anomalies or vascular lesions such as coarctation of the aorta.[1] In the case of vascular-lesion-related development, hemodynamic stress on the ASA functioning as an artery flowing in a congenital vascular anomaly or a collateral pathway related to a vascular anomaly may lead to aneurysm formation.[9] In the absence of vascular lesions, aneurysms complicated by autoimmune disease,[2] angitis,[3] or infection[4] have been reported in addition to idiopathic (solitary) aneurysms.[4] In many ASA aneurysms, hemodynamic stress is involved in the pathogenesis in comparison with cerebral aneurysms.[9]

ASA aneurysms related to congenital anomalies of the VA are markedly rare. Only one case report on an ASA aneurysm related to vertebral rete mirabile was published by Nagahata et al.[5] They reported the hemodynamic development/rupture of aneurysms of the anterior spinal arteries functioning as collateral pathways for bilateral vertebral retes , and emphasized that ASA aneurysms should be suspected when VA abnormalities are observed in the presence of non-typical subarachnoid hemorrhage.

In the present case, a diagnosis of ruptured ASA aneurysm complicated by bilateral VA occlusion, right PICA-AICA anastomotic artery, and left vertebral rete mirabile was made based on cerebral angiography. The PICA-AICA anastomotic artery may be the remnant of PICA-AICA anastomosis as a cerebral arterial anastomosis in the embryonic phase.[10] This anastomosis disappears in weeks 6–8 of gestation corresponding to the 4th phase of Streeter's cerebrovascular genesis. PICA-AICA anastomotic artery is rare, although the incidence is unclear.[10] On the other hand, vertebral rete mirabile refers to rete of the VA, but it remains to be clearly defined unlike rete mirabile of the internal carotid artery. To date, only 12 or 13 patients have been reported. In them, a portion of the VA was visualized like retes.[5,11,12] The pathogenesis may be similar to that of rete mirabile of the internal carotid artery; the VA once formed, may regress in the embryonic or perinatal phases after regression of a primitive vessel, and to compensate for this, vertebral rete mirabile may develop.[13] To our knowledge, vertebral rete mirabile was complicated by carotid rete mirabile in all patients; no study has reported such abnormality of the VA alone. In the present case, the left VA was morphologically connected to the periphery via a large number of net-like vessels, suggesting rete mirabile. On the other hand, several studies suggested that arteriosclerosis-related bilateral VA occlusion induced an ASA aneurysm; three patients have been reported.[6–8] Concerning the pathogenesis, the involvement of a hemodynamic mechanism in aneurysms of the ASA as a collateral pathway was proposed. These studies also emphasized that ASA aneurysms in addition to VA dissection should be suspected when subarachnoid hemorrhage related to bilateral VA occlusion is observed. In the present case, sufficient detailed examination was unable to be performed, and neither the mechanism nor timing of VA occlusion was able to be clarified. However,

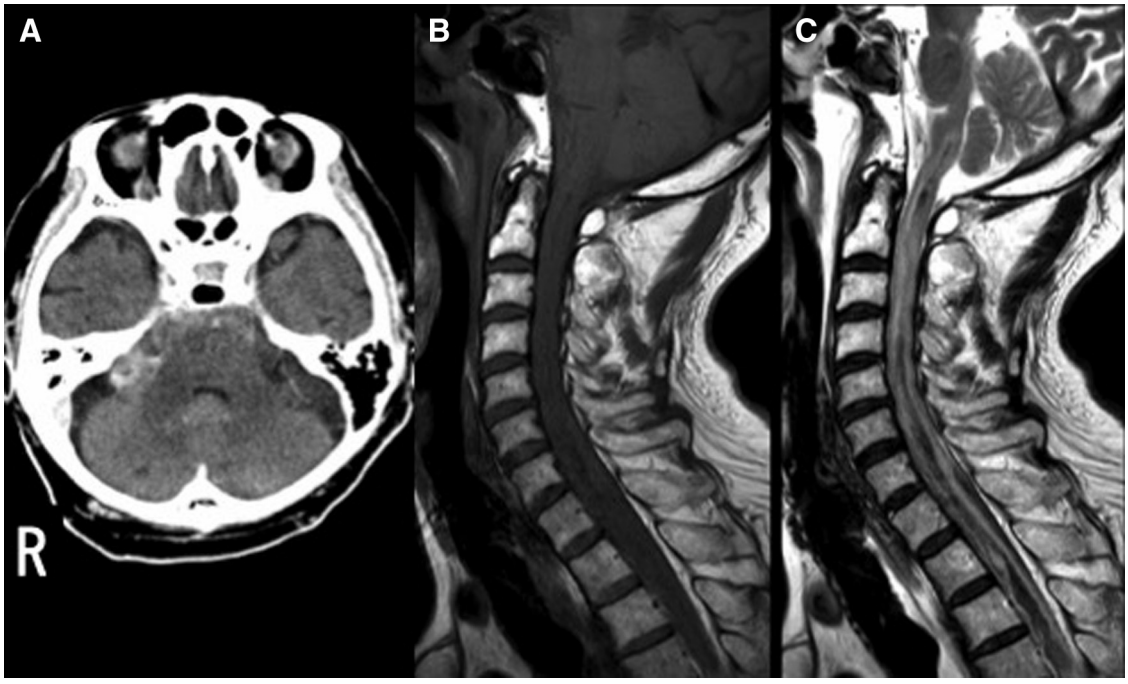

Fig. 5 Head CT (**A**) showed no change in subarachnoid hemorrhage, but cervical MRI (**B**, T1-weighted image; **C**, T2-weighted image) demonstrated bleeding from the spinal cord into the subarachnoid space. CT: computed tomography; MRI: magnetic resonance imaging

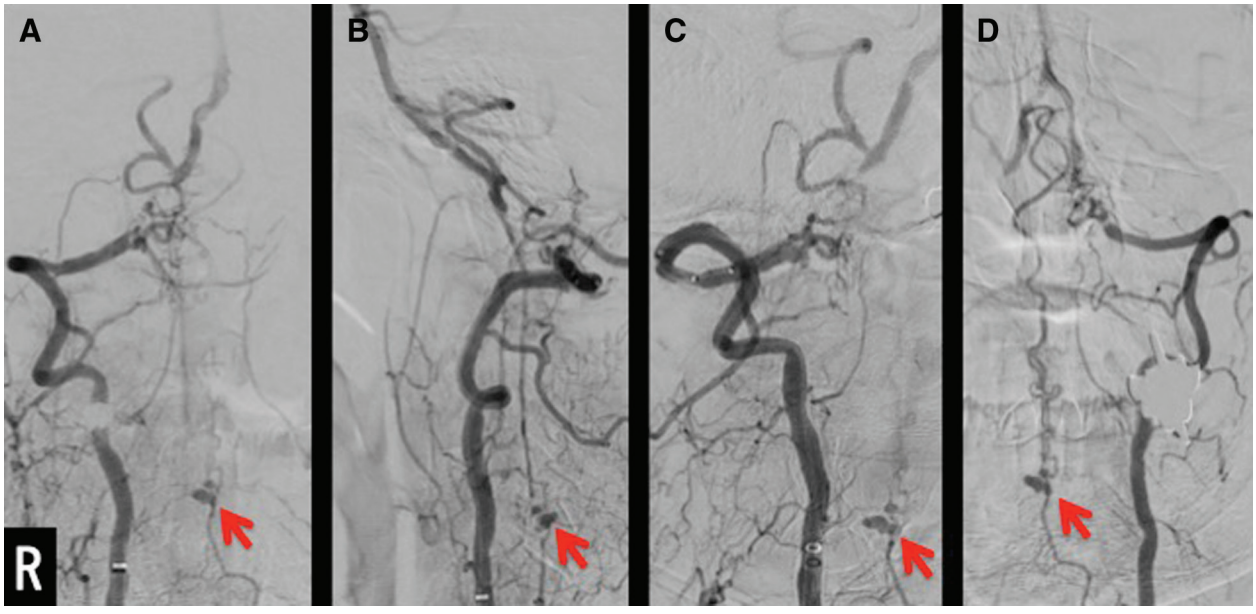

Fig. 6 Retrospective analysis of rt. (**A**, anterior view; **B**, lateral view; **C**, rt. front oblique) and lt. vertebral angiography (**D**, anterior view) revealed an ASA aneurysm at the level of C3 (arrow). ASA: anterior spinal artery

occlusion of the bilateral VA trunks may have led to the formation of the right PICA-AICA anastomotic artery and left vertebral rete mirabile as collateral pathways, whereas a collateral pathway mediated by the ASA may have developed due to insufficient intracranial blood flow, resulting in aneurysm formation.

Treatments for ASA aneurysms include direct surgery, endovascular treatment, and conservative treatment.[4,6,14,15]. Ashour et al.[6] reported a patient with a ruptured ASA aneurysm complicated by bilateral VA occlusion, and recommended that direct surgery be performed rather than endovascular treatment from the viewpoint of parent artery

flow preservation if an aneurysm in a collateral pathway is hemodynamically formed. Furthermore, another study found that relatively small, neck-free, fusiform aneurysms were frequent.[4] Direct surgery is appropriate in many patients, but most ASA aneurysms protrude from the median to anterior lateral area, as demonstrated in the present case. An approach from the posterior area sometimes makes it difficult to visualize the aneurysm. If a parent artery can be preserved, endovascular treatment may be considered as a treatment option.[14,15]

In the present case, a cerebral aneurysm was complicated by a right PICA-AICA anastomotic artery. Aneurysms at this site are rare, and to our knowledge, only three patients have been reported.[10,16,17] None of the patients had ASA aneurysms. A PICA-AICA anastomotic artery aneurysm ruptured only in one patient; Fujimura et al.[17] hypothesized that a hemodynamic mechanism was involved in the formation/rupture of an aneurysm in the PICA-AICA anastomotic artery as a collateral pathway. In the present case, mild right hemiplegia was noted at the time of onset, and MRI revealed no causative focus in the cranium. In addition, abnormalities in the courses of the bilateral VAs were observed, we should have suspected an aneurysm in the spinal artery, leading to detailed examination. If further detailed examination of the spinal cord had been conducted, the aneurysm in the right PICA-AICA anastomotic artery may have been evaluated as unruptured.

Conclusion

In patients with subarachnoid hemorrhage related to bilateral VA occlusion, an aneurysm formed in the presence of hemodynamic stress may be the source of hemorrhage. It is important to search for the source of hemorrhage, considering aneurysms of an extracranial collateral pathway, such as the ASA, in addition to those of an intracranial collateral pathway such as PICA-AICA anastomosis.

Disclosure Statement

The authors declare no conflict of interest.

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