Extracranial Vertebral Artery Aneurysm Causing Spinal Subarachnoid Hemorrhage
—Case Report—

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Abstract
A 48-year-old female presented with spinal subarachnoid hemorrhage (SAH) due to the rupture of an extracranial vertebral artery (VA) aneurysm. The aneurysm arose from the junction of the third and the fourth segments of the left VA just inside the dura mater, and was partially coated with Biobond. This is only the second such case reported. We recommend complete four-vessel angiography in evaluating patients with SAH, with special attention given to the extracranial VA if spinal SAH is suspected.

Key words: extracranial aneurysm, vertebral artery, subarachnoid hemorrhage

Introduction
Spontaneous subarachnoid hemorrhage (SAH) originates from an underlying intracranial aneurysm in approximately 80% of patients. Approximately 10% of all intracranial aneurysms occur in the posterior circulation and can be divided into three groups based on surgical approach: 1) upper basilar artery aneurysms, 2) mid-basilar trunk aneurysms, and 3) lower basilar artery and vertebral artery (VA) aneurysms. These aneurysms are all located in the intracranial subarachnoid space and rupture inevitably results in SAH. Extracranial VA aneurysm is an extremely rare cause of spinal SAH. We report a patient with an extracranial VA aneurysm which ruptured into the spinal subarachnoid space.

Case Report
A 48-year-old hypertensive female developed a severe occipital headache during an argument with her colleagues. She was transferred to our hospital because of persistent headache 5 days after the onset. Neurological examination on admission revealed an alert woman with questionable neck stiffness.

Computed tomography showed no abnormalities. Lumbar puncture demonstrated an opening pressure of 220 mmH2O and bloody cerebrospinal fluid. Emergency transfemoral cerebral angiography including the bilateral carotid arteries and left VA showed no intracranial aneurysms or vascular malformations. Left vertebral angiography, however, revealed a small funnel-shaped aneurysm at the junction of the third and the fourth segments of the VA (Fig. 1). On the following day, right vertebral angiography by retrograde brachial artery injection showed no vascular lesion.

She was taken to the operating room immediately after the second angiography. With her in the prone position, a midline suboccipital craniectomy and partial removal of the posterior arch of C1 were performed. The cerebrospinal fluid was mildly bloody. The origin of the posterior inferior cerebellar artery was identified and no aneurysm was found. Careful inspection revealed a VA aneurysm where the left VA pierced the dura to enter the spinal canal. The aneurysm was partially buried in the dural tissue but was mainly located intradurally. The bleb was covered with a small clot. A small artery was found adjacent to the aneurysm, but there was no obvious relationship between this artery and the aneurysmal neck (Fig. 2). The intradural portion of the
bleb and adjacent dura were coated with Biobond® (Yoshitomi Pharmaceutical Industries Ltd., Osaka).

Her headache subsided over a week, and her recovery was uneventful. Postoperative angiography including the bilateral carotid arteries and left VA 19 days after surgery showed no change in the size or shape of the coated aneurysm. No pathological finding was observed other than this aneurysm.

**Discussion**

Only two extracranial non-traumatic VA aneurysms resulting in intraspinal SAH have been reported, that of Barnett and the present patient. Jewel described a patient with a ruptured extradural VA aneurysm in a similar location to ours, but presenting with brainstem dysfunction due to arterial spasm secondary to spinal epidural hemorrhage.

The majority of extracranial VA aneurysms are traumatic pseudoaneurysms, usually developing after penetrating wound caused by gunshot, stabbing, or laceration. Non-penetrating injuries such as chiropractic manipulation, cervical vertebral fractures, and dislocation or implantation of radioactive seeds may cause similar aneurysms. Non-traumatic aneurysms are less frequent, and may result from inflammatory lesions including osteomyelitis, endarteritis, and necrotizing arteritis, while congenital aneurysms are associated with inherited disorders, such as the Ehlers-Danlos syndrome, neurofibromatosis, and fibromuscular dysplasia. Our patient had no history of craniocervical trauma, inflammatory diseases, or hereditary disorders.

The pathogenesis of this aneurysm is unclear. Although cerebral aneurysms occur almost exclusively at the bifurcations of arteries, vertebral angiography showed no arterial branch arising around the aneurysmal neck in this patient. The VA passes a funnel-shaped dural foramen to enter the spinal canal. The posterior spinal artery arises from the VA just outside the dura mater and enters the spinal canal through this foramen. Another extradural branch of VA in this region is the posterior meningeal artery. The small artery found close to the aneurysm might be one of these branches, and therefore responsible for the aneurysm formation. Possibly vertebral angiography failed to opacify the arterial branch due to vasospasm. Constriction of the parent artery by surrounding dural tissue and subsequent hemodynamic stress to the arterial wall are alternative explanations for the genesis of the aneurysm.

The incidence of SAH of unknown etiology has decreased due to the improvement of diagnostic techniques. The importance of complete four-vessel study and repeat angiography has been emphasized. SAH may originate from spinal cord lesions, such as arteriovenous malformations and neoplasms, so the
spinal canal should be investigated especially when the history or physical examination indicate a spinal SAH. Our case shows that an extracranial VA aneurysm can cause SAH. Careful angiographic inspection of extracranial VA is mandatory when evaluating such patients.

References

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