Posterior Inferior Cerebellar Artery Aneurysm Associated With Persistent Primitive Hypoglossal Artery

—Case Report—

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Abstract

A 49-year-old woman presented with subarachnoid hemorrhage (SAH) from an aneurysm associated with a persistent primitive hypoglossal artery (PPHA) manifesting as sudden onset of headache, but without neurological deficits. Conventional computed tomography (CT) of her head showed no abnormality but lumbar tap indicated SAH. Three-dimensional (3D)-CT angiography showed a PPHA originating from the internal carotid artery and an aneurysm of the posterior inferior cerebellar artery at the junction with the remnant hypoplastic vertebral artery. 3D-CT angiography was essentially useful for presurgical planning to determine the extent of craniotomy and the space for possible temporary clipping, and confirmed the diagnosis of aneurysmal SAH. The aneurysm was clipped and she returned to her job 4 weeks later. Cerebral angiography is the golden standard technique to diagnose PPHA, but 3D-CT angiography can be recommended for presurgical evaluation, especially in patients with complex and anomalous anatomical structures.

Key words: persistent primitive hypoglossal artery, three-dimensional computed tomography angiography, subarachnoid hemorrhage

Introduction

Persistent primitive hypoglossal artery (PPHA) is a rare anomalous remnant with an incidence of 0.027% to 0.26%.

PPHA is the second most frequent type of persistent embryonic carotid-basilar anastomosis after persistent primitive trigeminal artery.1-4,6,7 PPHA may be associated with vascular anomalies such as arteriovenous malformation or cerebral aneurysm, with the risk of intracranial hemorrhage. The ipsilateral vertebral artery (VA) is often found to be hypoplastic.1,4 PPHA is an anomalous structure, so the anatomical relationship with the skull base and surrounding vascular structures will be atypical and complex, and thus such structural information is essential for the planning of neurosurgical procedures.

We present a case of subarachnoid hemorrhage (SAH) originating from an aneurysm associated with PPHA, which demonstrated the importance of three-dimensional (3D)-computed tomography (CT) angiography for evaluating anomalous anatomical structures.

Case Report

A 49-year-old woman suffered sudden onset of headache, and was referred to our hospital. She was completely conscious with no neurological deficits on admission. Head CT detected no hemorrhage but lumbar puncture revealed SAH. Aortography showed a PPHA originating from the right internal carotid artery at the C2 level and a saccular aneurysm (Fig. 1). The bilateral VAs were hypoplastic and a catheter could not be inserted into these vessels. Selective angiography of the right PPHA
Fig. 1 Aortogram revealing a persistent primitive hypoglossal artery (arrows) and an aneurysm (arrowheads), as well as the hypoplastic bilateral vertebral arteries.

Fig. 2 Selective angiogram of the right persistent primitive hypoglossal artery showing the aneurysm arising from the right posterior inferior cerebellar artery at the junction with the remnant vertebral artery (arrowheads).

Fig. 3 Postero-superior view of three-dimensional computed tomography angiography before surgery revealing the persistent primitive hypoglossal artery, aneurysm, posterior inferior cerebellar artery, and a space adequate for temporary clipping (arrow).

Fig. 4 Operative photograph demonstrating that partial drilling of the condylar fossa provided enough space for temporary clipping of the parent posterior inferior cerebellar artery (PICA) and clipping of the aneurysm (An), which arose at the junction with the remnant hypoplastic vertebral artery (arrowheads). The distal vertebral artery was not identified during the surgery. PPHA: persistent primitive hypoglossal artery.

showed that the aneurysm arose from the right posterior inferior cerebellar artery (PICA) at the junction with the remnant hypoplastic VA (Fig. 2). Left internal carotid angiography proved to be unremarkable.

3D-CT angiography clearly delineated the aneurysm and the PPHA, which penetrated the right hypoglossal canal and joined the lower portion of the basilar artery. The PPHA arose from the internal carotid artery at the C2 level and ran parallel to the cervical spine to enter the posterior cranial fossa through the hypoglossal canal (Fig. 3). The aneurysm was localized laterally to medulla oblongata, and the parent PICA projected from the PPHA at the orifice of the hypoglossal canal, indicating a potential location for temporary clipping during the surgery. Presurgical planning based on the 3D-CT angiography findings established the necessary extent of craniotomy and partial resection of the occipital bone, and the space for temporary clipping.

Surgical intervention was performed 14 days after the onset of SAH. After unilateral suboccipital craniotomy, the occipital bone was drilled to allow the exploration of the aneurysm and its parent artery (Fig. 4). Partial drilling of the condylar fossa provid-
ed enough space for temporary clipping of the parent PICA and clipping of the aneurysm. Complete neck clipping was successfully performed (Fig. 5). The postoperative course was uneventful. She recovered rapidly and was discharged without neurological deficits. She returned to her job as a high school teacher 4 weeks after the surgery.

Discussion

Cerebral angiography is the golden standard technique to diagnose cerebral aneurysms, including PPHA.\(^7\) 3D-CT angiography is a minimally invasive method to detect intracranial aneurysms, but may not detect small multiple unruptured aneurysms,\(^2\) multiple clips,\(^6\) or some aneurysms such as posterior communicating artery aneurysms,\(^5\) so conventional angiography may still be necessary. After the introduction of helical scanning CT and various other improvements, 3D-CT angiography has become a reliable method for the diagnosis of most cerebral aneurysms.\(^6\) In our case, only 3D-CT angiography would have been adequate for the diagnosis and presurgical evaluation, rather than also performing cerebral angiography. 3D-CT angiography successfully delineated the aneurysm, the parent arteries, and surrounding bony structures, and was helpful to confirm the diagnosis of aneurysmal SAH that was not ascertained with conventional CT.

Cerebral angiography may be less helpful than 3D-CT angiography to evaluate the anatomical relationship between the skull base bony structures and blood vessels, which may be anomalous but essential for preoperative evaluation. 3D-CT angiography can be recommended for presurgical evaluation, especially in patients with complex or anomalous anatomical structures.

References


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