A Case of Vertebral Artery Dissecting Aneurysm with Double Origin of the Posterior Inferior Cerebellar Artery Causing Subarachnoid Hemorrhage

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Double origin of the posterior inferior cerebellar artery (DOPICA) is rare but is associated with intracranial aneurysm formation. A 46-year-old man was brought to our hospital with severe subarachnoid hemorrhage (SAH). Digital subtraction angiography revealed right vertebral artery dissecting aneurysm (VADA) and DOPICA. The aneurysm involved the distal component of the posterior inferior cerebellar artery. The proximal component covered the original flow angiographically. Endovascular internal trapping and parent artery occlusion were performed for the aneurysm and the right vertebral artery (VA). Flow of the posterior inferior cerebellar artery was preserved by the proximal component of the posterior inferior cerebellar artery after trapping. The patient unfortunately died and autopsy revealed ischemic change in the right medulla oblongata. The ischemic change was considered to occur at the treatment according to the pathological findings. Sacrifice of one component of DOPICA to treat VADA with the expectation of preserved flow via the other component should be considered cautiously in terms of the neurovascular anatomy.

Keywords: vertebral artery dissecting aneurysm, double origin of posterior inferior cerebellar artery, subarachnoid hemorrhage, internal trapping

Introduction

Double origin of the posterior inferior cerebellar artery (DOPICA) is a rare developmental anomaly with an incidence of 1.45–6%.1–4 However, DOPICA has a high rate of association with intracranial aneurysms including vertebral artery dissecting aneurysm (VADA) and saccular aneurysm of the vertebral artery (VA) and PICA.2,3,5,6 Only five cases of VADA with DOPICA causing subarachnoid hemorrhage (SAH) have been reported. Internal trapping was performed in all cases, but the strategy and outcome are unclear. We report another case of ruptured VADA with DOPICA and discuss the strategy for this disease in terms of the neurovascular system.

Case Report

A 46-year-old man was referred to our hospital after onset of severe headache followed by unconsciousness. His pupils were miosis and the light reflexes were absent. Computed tomography (CT) revealed massive SAH (Hunt and Kosnik grade V) mainly in the posterior fossa with acute hydrocephalus (Figs. 1A and 1B). After external ventricular drainage, digital subtraction angiography showed the right VADA and two components of the PICA arising from the same VA. The origin of the distal component of the PICA (DCPICA) was involved in the dissecting lesion. The origin of the proximal component of the PICA (PCPICA) was the extradural segment of the VA (V3 segment). The two components of the PICA converged at the tonsillomedullary segment (Figs. 2A and 2B). The projection of the left VA revealed no aneurysm in the left VA and the retrograde flow to the right VADA.

We planned endovascular internal trapping and parent artery occlusion from the aneurysm including the origin of the DCPICA to the right VA just a little distal to the origin of the PCPICA because flow in the PICA would be ensured via the opening PCPICA. The procedure was performed under general anesthesia. A 6-French guiding catheter was placed in the proximal right VA. Two microcatheters were passed into the dissecting lesion proximally and distally. After caging from the distal microcatheter, detachable coils were deployed from the proximal microcatheter to the dissecting aneurysm, partial coil anchoring into the origin of the DCPICA, and back into the proximal parent artery (Fig. 2C).

Postoperative angiography showed that anterograde flow to the cerebellum through the PCPICA was preserved but the DCPICA’s cortical branch could not be observed, whereas the retrograde flow to the DCPICA could reach only up to the distal portion of the lateral medullary segment (Fig. 2D). Postoperative CT scan did not show apparent ischemia due to pulmonary edema gradually progressed so that postoperative MRI could not be taken for his severe status, and he eventually died on the day 11 after onset. Autopsy revealed thick hematoma mainly around the right VA and in the ventricle, and the VADA was filled with coils. There was a tear in the
course of the autopsy because the adhesion between the VA and surrounding tissues due to massive SAH, and the detailed distribution of the perforating branches is unknown, but a few perforating branches were confirmed near the lesion (Fig. 3A). Microscopic examination revealed medullary ischemia in the right side. The lateral area in the right
side of the medulla oblongata had more hyperplastic reactive astrocytes and macrophages compared to the anterolateral area (Figs. 3B and 3C). The right cerebellum perfused by the PICA was not infarction.

**Discussion**

DOPICA consists of two components arising from the same VA proximally and distally to each other and converging at the distal PICA. The PCPICA is a remnant of embryological anastomoses between the lateral spinal artery (LSA) and the PICA. The LSA originates from either the PICA or the intracranial VA and passes lateral to the medulla oblongata. The DCPICA is the original embryological PICA, which derives from a hypertrophied radiculopial artery.

DOPICA is a risk factor for pathogenesis of VA/PICA dissecting aneurysm or saccular aneurysm in the posterior circulation. DOPICA was present in 4.1% of patients with intracranial aneurysms. When a long proximal component of the PICA arises from the extradural VA, VA or DCPICA is easy to stretch or compress over the dura mater and brainstem, and will eventually dissect. Treatment for conventional VADA without DOPICA includes various strategies such as proximal ligation, endovascular internal trapping, trapping combined with revascularization (occipital artery-PICA bypass), or stent-assisted coil embolization. However, all five reported cases of VADA with DOPICA causing SAH were treated by endovascular internal trapping. Treatment was successful in four cases and no death occurred, with good postoperative courses and neurological freedom from events (Table 1).

Unfortunately, our patient died and autopsy revealed ischemic change in the right side of the medulla oblongata. Moreover, two areas in the right side of the medulla oblongata (anterolateral area and lateral area) may have suffered ischemia at different times because the lateral area contained more hyperplastic reactive astrocytes and macrophages than the anterolateral area. Ischemia on the lateral area might have been caused by vasospasm or cerebral herniation after SAH, before obstruction of the perforators of the VA or the proximal segment of the involved DCPICA after coiling resulted in medullary ischemia. The parent artery occlusion in this case was applied just distal to the origin of the PCPICA. Although this procedure might be a risk of ischemia due to the perforators’ obstruction, it was difficult to identify the origin of the false lumen of the VADA.
The perforators to the medulla oblongata originate from the VA, the bulbar branch, or from the PICA, especially the anterior and lateral medullary segments. The middle group of bulbar branches mainly distributes to the anterolateral area of the medulla oblongata. On the other hand, the perforators arising from the lateral medullary segment of the PICA distribute to the lateral area of the medulla oblongata. Nevertheless, the formation of the perforators in DOPICA is still unclear. The DCPICA is embryologically a true PICA so that the DCPICA is likely to be the main supplier of perforators to the medulla oblongata. Therefore, we can expect retrograde flow from the opening component of the PICA to the occluded component after endovascular internal trapping in patients with DOPICA.

We performed treatment with a similar procedure to previous cases, but medullary ischemia occurred. Needless to say, non-involved type VADA as in other cases carry little risk of medullary ischemia because the origins of the perforators are unrelated to endovascular internal trapping and remain patent. In the type involving DCPICA, the two components of the DOPICA converge at the lateral medullary segment as in two cases, and no neurological deficit happened after treatment. However, in a previous and the present cases, the two components of the DOPICA converged at the tonsillomedullary segment, so only the DCPICA passed to the convergence point and the perforators arising from the DCPICA distributed to the medulla oblongata over a relatively wide range. Therefore, preservation of the retrograde flow to the DCPICA and the anterograde flow to the converging PICA from the PCPICA is crucial if endovascular internal trapping is attempted in the case of VADA involving the DCPICA. Preservation of both directions of flow is likely to depend on the convergence point of the DOPICA and the dominance of the components of the DOPICA. Even if the PCPICA seems to preserve the perfusion of the PICA angiographically, sacrifice of the DCPICA and dependence of the PCPICA alone should be carefully considered including the anatomical features.

Conflicts of Interest Disclosure

The authors declare that they have no financial or other conflicts of interest in relation to this research and its publication. All authors who are members of The Japan Neurosurgical Society (JNS) have registered online Self-reported COI Disclosure Statement Forms through the website for JNS members.

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Table 1  Reported cases of VADA with DOPICA

| Author | Age/sex | HH grade | Location of aneurysm | Convergence | Postoperative complication |
|--------|---------|----------|----------------------|-------------|---------------------------|
| Kwon et al. (2007) | 50/Male | III | DCPICA involved | LMS | No neurological deficit |
| Fan et al. (2011) | 58/Male | – | Pre-DCPICA | AMS | No neurological deficit |
| Koh et al. (2012) | 42/Male | – | DCPICA involved | TTS | No neurological deficit |
| Kawabata et al. (2016) | 61/Female | V | DCPICA involved | TMS | Small infarction of the PICA territory |
| Present case | 48/Male | V | DCPICA involved | TMS | Medullary infarction |

AMS: anterior medullary segment, DCPICA: distal component of posterior inferior cerebellar artery, HH: Hunt and Hess; LMS: lateral medullary segment, TMS: tonsillomedullary segment, TTS: telovelotonsillar segment, VADA: vertebral artery dissecting aneurysm.

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