

Posterior Inferior Cerebellar Artery Thrombosed Aneurysm Associated with Persistent Primitive Hypoglossal Artery Successfully Treated with Condylar Fossa Approach

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A 68-year-old woman presented with generalized seizure due to the left internal carotid artery (ICA) aneurysmal compression of the ipsilateral medial temporal lobe. Computed tomography angiography (CTA) revealed multiple aneurysms of the right persistent primitive hypoglossal artery (PPHA), the right ICA, and the right anterior cerebral artery (ACA). The right PPHA originated from the ICA at the level of the C1 and C2 vertebral bodies and passed through the hypoglossal canal (HC). The PPHA aneurysm was large and thrombosed, which was located at the bifurcation of the right PPHA and the right posterior inferior cerebellar artery (PICA), projecting medially to compress the medulla oblongata. Since this patient had no neurological deficits, sequential imaging studies were performed to follow this lesion, which showed gradual growth of the PPHA aneurysm with further compression of the brain stem. Although the patient remained neurologically intact, considering the growing tendency clipping of the aneurysm was performed. Drilling of the condylar fossa was necessary to expose the proximal portion of the PPHA inside the HC. The key of this surgery was the preoperative imaging studies to fully understand the anatomical structures. The PPHA was fully exposed from the dura to the corner its turning inferiorly without damaging the occipital condylar facet. Utilizing this technique, the neck ligation of the aneurysm was safely achieved without any surgical complications.

Keywords: persistent primitive hypoglossal artery, cerebral aneurysm, thrombosed aneurysm, condylar fossa approach

Introduction

Persistent primitive hypoglossal arteries (PPHAs) are the second most common persistent embryonic carotid-basilar anastomoses after trigeminal arteries.^{1–3} They are occasionally associated with cerebrovascular disorders such as arterial aneurysms, arteriovenous malformations and ischemic diseases.^{4–14} The prevalence of PPHAs has been estimated

0.025–0.26%, and the rate of aneurysms on them has been reported as frequent as 22–25%.^{15–17} When treating the vascular lesions related to the PPHA, special consideration is required, because the PPHA commonly conveys the only blood supply to the posterior circulating system, which usually lacks the posterior communicating arteries and the contralateral vertebral artery. In addition, since it traverses the hypoglossal canal (HC),³ when the intracranial proximal portion of the PPHA can be seen sufficiently, identifying this parental artery at the extracranial portion utilizing skull base technique is necessary.

We experienced a rare case of the PPHA-posterior inferior cerebellar artery (PICA) aneurysm successfully treated with clipping using condylar fossa approach.

Case Report

A 68-year-old woman presented with generalized seizure due to the left internal carotid artery (ICA) aneurysmal compression of the ipsilateral medial temporal lobe. Computed tomography angiography (CTA) revealed multiple aneurysms of the right PPHA, the right ICA, and the right anterior cerebral artery (ACA). This right PPHA originated from the ICA at the level of the C1 and C2 vertebral bodies and passed through the HC (Fig. 1). The PPHA aneurysm was large and thrombosed, which was located at the bifurcation of the right PPHA and the right posterior inferior cerebellar artery (PICA), projecting medially to compress the medulla oblongata. Since this patient had no neurological deficits, sequential imaging studies were performed to follow this lesion, which showed gradual growth of the PPHA aneurysm with further compression of the brain stem in 11 months (Fig. 2). Although the patient remained neurologically intact, considering the growing tendency clipping of the aneurysm was performed. The patient was placed on the operative table in the park bench position with the head positioned lateral in order to make it easier to secure the proximal ICA at the cervical level if needed. Intraoperative neurophysiological monitoring was performed using facial motor evoked potentials (MEPs) and brain stem auditory evoked potentials. Facial MEPs were recorded from the orbicular oris and orbicularis oris muscle through needle electrodes. To avoid lower cranial nerve injury neural integrity monitor (NIM) 3.0 with automatic periodic stimulation (Medtronic Inc., USA)

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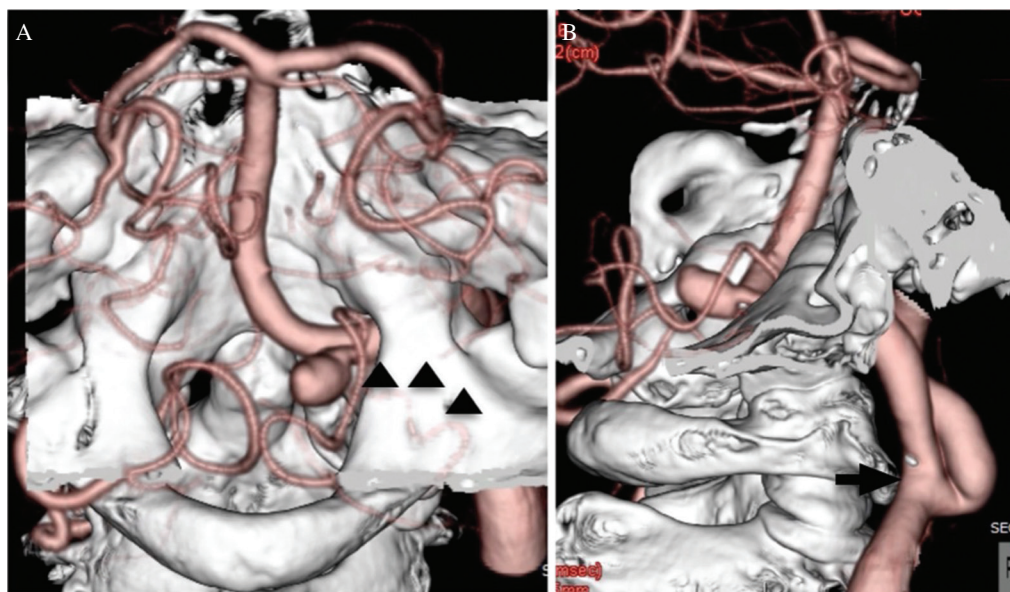


Fig. 1 Three-dimensional computed tomography angiography (3D-CTA) showed the persistent primitive hypoglossal artery originating from the right internal carotid artery between the C1 and C2 vertebra level (arrow), which traverse the hypoglossal canal (arrow heads).

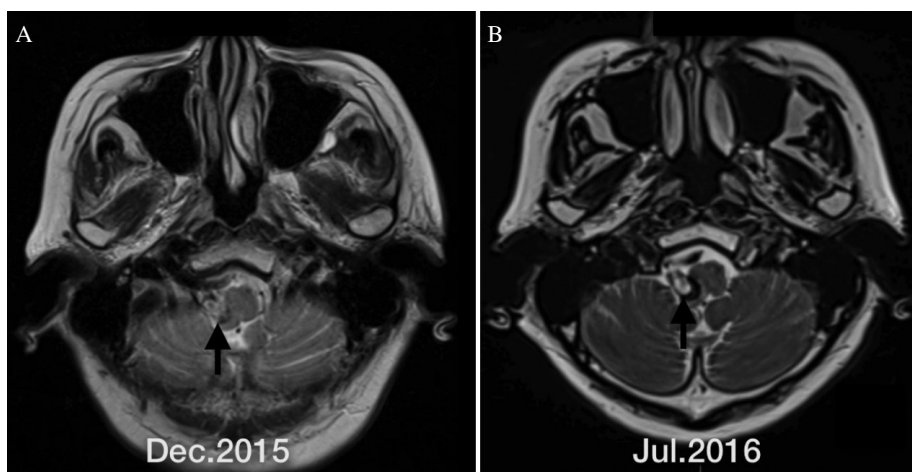


Fig. 2 MRI T2WI revealed the persistent primitive hypoglossal artery - the posterior inferior cerebellar artery aneurysm (arrow) had grown and compressed the brain stem for the 11 months.

was employed. The patient is intubated with NIM contact-reinforced electromyogram (EMG) endotracheal tube (Medtronic Inc., USA). After unilateral suboccipital craniotomy, drilling of the condylar fossa was performed to expose the proximal portion of the PPHA inside the HC. With identifying the hypoglossal nerve by electrical stimulation, the PPHA was exposed from the entry to the dura mater to the corner its turning inferiorly without damaging the occipital condylar facet (Fig. 3). Bleeding from the venous plexus covering the PPHA inside the HC was controlled utilizing bipolar electrocautery. The aneurysm was identified after opening the dura mater. Fortunately the intracranial portion of the PPHA could be secured for proximal control. Neck clipping of the aneurysm was safely performed (Fig. 4). Intraoperative indocyanine green (ICG) video angiography was performed, which showed the appropriate patency of the PPHA and the PICA, along with the complete collapse of the aneurysm. CTA was postoperatively performed to reconfirm these findings (Fig. 5). Her postoperative course was uneventful. She was discharged in a month with no neurological deficits.

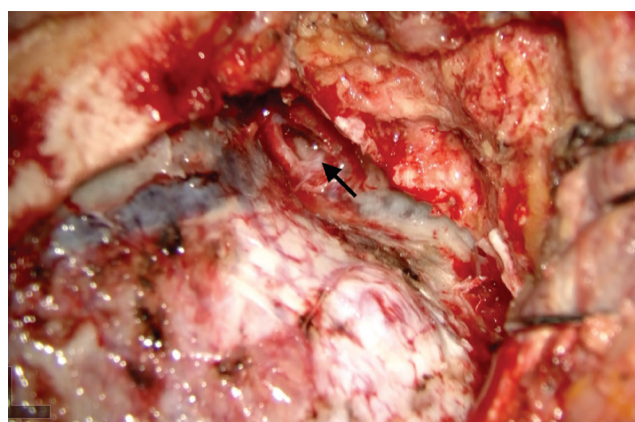


Fig. 3 The persistent primitive hypoglossal artery (arrow) was exposed laterally from the dura to the corner its turning inferiorly without damaging the occipital condylar facet.

Discussion

The persistent primitive hypoglossal artery is one of rare vascular anomalies. The incidence has been reported as

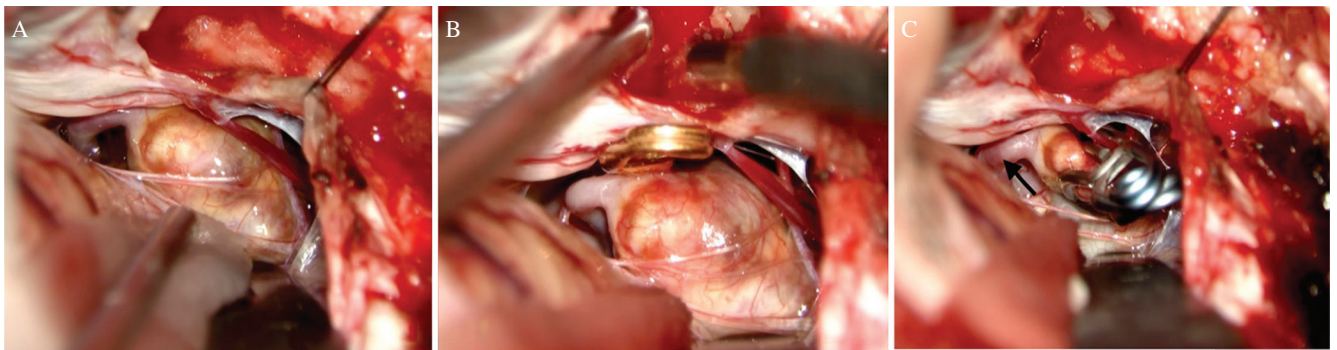


Fig. 4 By securing proximal portion of the persistent primitive hypoglossal artery, neck clipping of the aneurysm was safely performed. (A) PICA-PPHA aneurysm existed close to the hypoglossal canal. (B) Temporary clip could be placed on the intracranial portion of the PPHA. C: Neck clipping was performed while preserving PICA (arrow) and PPHA flow.

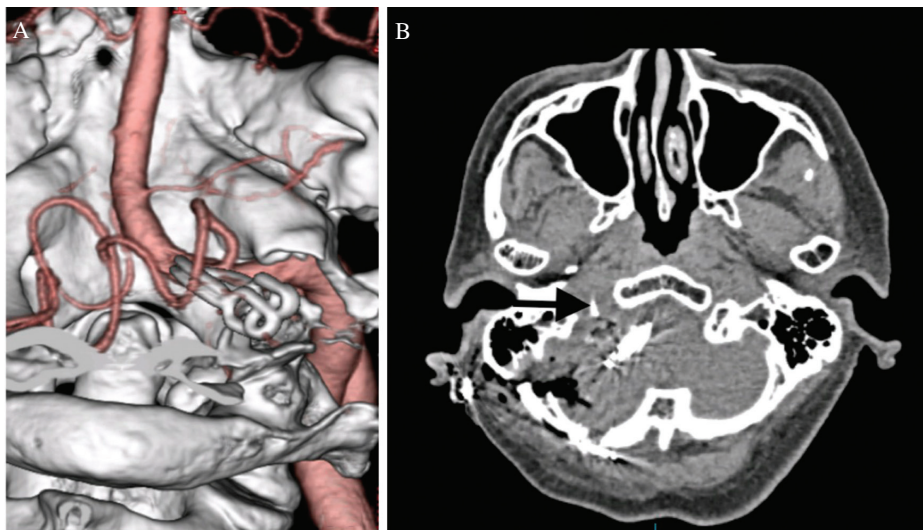


Fig. 5 Post-operative 3D-CTA showed the range of skull base drilling and appropriate clipping of the PICA-PPHA aneurysm. (A) PICA-PPHA aneurysm was obliterated with appropriate clipping. (B) CT bone imaging showing the range of skull base drilling.

0.025–0.26%, the rate of accompanying cerebral aneurysm including at another arteries has been considered to be 26.9–53%.^{3,15–17} There have been only a few reports of successful treatment for PICA-PPHA bifurcation aneurysms. The criteria for identification of PPHAs are 1. It is arising from the cervical part of the internal carotid artery at C1–C2 vertebra level. 2. It enters the posterior cranial fossa through the hypoglossal canal along with the accessory nerve. 3. The basilar trunk appears filled only beyond its anastomosis with PPHA. Since the vertebral arteries and the posterior communicating arteries may be hypoplastic or absent,³ the treatment for these rare aneurysms is challenging considering their complex anatomical structure and the possible insufficiency of the posterior circulation. The aneurysm of our case was thrombosed in addition to that, but we concluded that open surgery was still feasible. The key to the successful treatment of this case was the meticulous preoperative planning based on CTA and computed tomography venography (CTV) and the correct understanding of the anatomical structure.^{5,18,19} The 3D-CTA showed that this aneurysm originated from the PPHA-PICA bifurcation just distal of the entry of the HC. As the length of the proximal portion of the intracranial PPHA looked too short to grasp easily, we adopted condylar

fossa approach to identify the PPHA inside the HC.²⁰ Since difficulty was predicted grasping the proximal portion of the PPHA inside the HC, the patient was positioned with the head rotated lateral, so that the PPHA was grasped more proximally using anterolateral approach. The HC was expanded by the PPHA and it was easier to drill away the condylar fossa to grasp the PPHA inside the HC. While drilling the condylar fossa electrical stimulation of XII was useful to detect the depth of the HC. Wide drilling of the HC required extra attention not to cause instability of the atlanto-occipital joint. The lateral rim of the foramen magnum or the posterior rim of the jugular foramen should remain not drilled.²¹

CTV was useful to understand the venous system around the condylar fossa: i.e. the posterior condylar emissary marginal sinus, the occipital sinus, the vertebral venous plexus and the jugular bulb etc.^{3,19} The PPHA without the ordinal venous plexus surrounding the V3 portion of the vertebral artery is known to have unusual venous system.¹⁹ In our case, the preoperative CTV showed a developed posterior condylar vein, but no venous structure inside the bone around the condylar fossa (Fig. 6) However, we encountered bleeding from the developed diploic vein while drilling away the condylar fossa, which was controlled with bone wax.

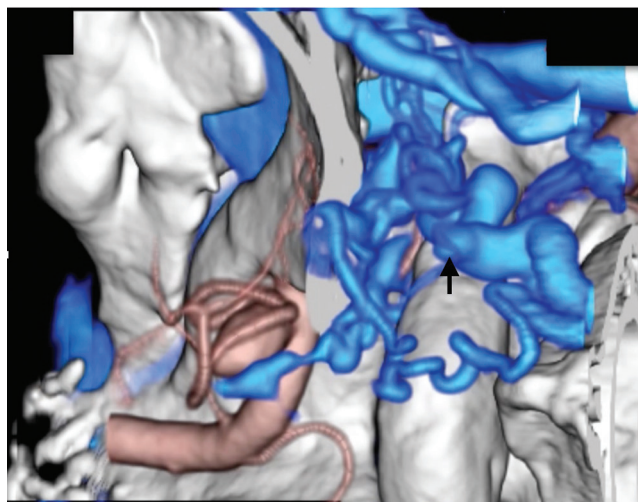


Fig. 6 The preoperative computed tomography venography showed developed posterior condylar vein (arrow), there was no venous structure inside the bone at condylar fossa.

Developed venous plexuses surrounding the PPHA such as vertebral venous plexus were also identified. To minimize the venous injury, these plexuses were partially coagulated and dissected open to expose the PPHA. After securing the proximal portion of the PPHA this way, neck clipping of the PICA-PPHA aneurysm was safely performed (Fig. 5)

It is full understanding of the unusual anatomical structures based on the preoperative imaging studies along with the appropriate choice of the surgical strategies including skull base techniques that are essential to safe open surgery.

Conflicts of Interest Disclosure

The authors declare that they have no conflicts of interest. 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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