Persistent Primitive Hypoglossal Artery Aneurysms
—Report of Two Cases—

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Abstract
The authors present two patients with subarachnoid hemorrhage caused by ruptured intracranial saccular aneurysms of the persistent primitive hypoglossal artery. A standard unilateral suboccipital approach in one patient resulted in incomplete neck clipping since the operative field was restricted by a protruding jugular tubercle. Successful aneurysmal neck clipping was achieved in the second patient via a unilateral-transcondylar-suboccipital approach with resection of the jugular tubercle and rim of the foramen magnum.

Key words: intracranial aneurysm, primitive hypoglossal artery, subarachnoid hemorrhage, transcondylar approach, neck clipping

Introduction
The persistent primitive hypoglossal artery (PHA) is a remnant of the embryonal carotid-basilar anastomosis. It is usually a rare incidental finding during cerebral angiography. The incidence of intracranial aneurysms associated with persistent PHA is high.

Here we report two cases of this rare anomalous artery associated with ruptured aneurysms and subarachnoid hemorrhage (SAH).

Case Reports
Case 1: A 33-year-old male suffered a sudden onset of severe headache and nausea. On admission the same day, he was alert but his consciousness level deteriorated quickly presumably due to aneurysmal rebleeding. Computed tomographic (CT) scans demonstrated high-density areas in the basal cisterns indicating SAH. Right carotid angiography demonstrated a persistent PHA (Fig. 1 left). This artery originated from the internal carotid artery (ICA) at the C2 vertebral level and supplied the basilar artery (BA) and all its branches. A saccular aneurysm was observed at the junction of the PHA

Fig. 1 Case 1. left: Right carotid angiogram, lateral view, showing the persistent primitive hypoglossal artery (PHA), originating from the internal carotid artery at the C2 vertebral level. right: Selective angiogram of the PHA, anteroposterior view, demonstrating a saccular aneurysm (arrow) at its junction with the posterior inferior cerebellar artery (PICA). BA: basilar artery.
and the posterior inferior cerebellar artery (PICA) (Fig. 1 right). The aneurysm was 8 × 6 mm and the fundus projected medially. The aneurysmal neck was 9 mm from the midline and 10 mm above the foramen magnum. Catheterization of the left vertebral artery (VA) was impossible. Preoperative condition was Hunt and Kosnik grade III.

A right suboccipital craniectomy was performed on the day of admission. The lateral cerebellomedullary cistern was exposed with gentle cerebellar retraction. The operative field was very restricted since the jugular tubercle, which formed the roof of the enlarged hypoglossal canal, was well developed and protruded. Therefore, both the proximal portion of the intracranial PHA and the aneurysm could not be visualized adequately. The jugular tubercle was removed using a high-speed air drill, but the aneurysm could not be seen well from this angle without further brainstem retraction. Aneurysmal neck clipping was accomplished with great difficulty because of the narrow operating field.

On the 30th postoperative day he suddenly became comatose. CT scans showed considerable SAH. Angiography revealed the incomplete aneurysmal neck clipping. His neurological condition deteriorated greatly and no further operative treatment was performed. He died 3 years later.

**Case 2:** A 39-year-old female was hospitalized for sudden onset of severe headache and vomiting on the same day. On admission, she was alert and well oriented, with no neurological deficit. No nuchal rigidity was found. CT scans revealed high-density areas in the basal cisterns. Right carotid angiography demonstrated a persistent PHA originating from the ICA at the C2 vertebral level and finally joining the BA (Fig. 2 left). It supplied most of the posterior circulation. The left VA was impossible to catheterize. A magnified straight anteroposterior view of the selective angiogram obtained by PHA catheterization revealed a small aneurysm located at the junction with the BA (Fig. 2 right). The aneurysm was 2 × 3 mm with the dome projecting ventrally. The aneurysmal neck was 5 mm from the midline and 15 mm above the foramen magnum. Retrograde filling through the PHA and BA junction also demonstrated the left hypoplastic VA. Preoperative condition was Hunt and Kosnik grade II.

On the day of admission, a right suboccipital craniectomy was performed with a paramedian skin incision in the semiprone park bench position. The ipsilateral posterior rim of the foramen magnum was removed and a hemilaminectomy of the atlas performed. The opening of the foramen magnum was extended anterolaterally toward the jugular foramen just behind the occipital condyle using a high-speed air drill. The inner one-third of the occipital condyle and the jugular tubercle were carefully removed (Fig. 3 upper). After the dural incision, the lateral cerebellomedullary cistern was opened. The extensive bone removal provided a wide exposure around the brainstem front. The aneurysm could be recognized easily (Fig. 3 lower). Premature rupture occurred during neck dissection, but was easily controlled with temporary clipping of the PHA proximal to the aneurysm. A Yasargil clip was successfully applied to obliterate the aneurysmal neck.

Postoperative angiography demonstrated complete neck clipping. She was discharged without neurological deficit.

**Discussion**

Forty (26%) of 155 previously reported cases of this anomalous artery were associated with intracranial saccular aneurysms. Nine cases of persistent PHA aneurysms are summarized with the present two cases in Table 1.1-3,6,7,10,13-15) The incidence of intracranial aneurysm associated with persistent PHA is relatively high. Their mean age was 43 years, somewhat younger than for aneurysms at other locations. All 11 patients presented with SAH, caused by ruptured PHA aneurysms in nine cases and ruptured aneurysms at other locations in two cases. Nine

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aneurysms were at the PHA-BA junction and two were at the PHA-PICA junction. The ventral surface of the medulla oblongata, including the vertebrobasilar junction where PHA aneurysms often develop, cannot be approached easily by a standard suboccipital craniectomy because the jugular tubercle or occipital condyle obstruct the operating field. Therefore, extensive bone removal is necessary to avoid excess retraction which may injure the brainstem.\textsuperscript{5,8,11,12} There are two key points in this approach: firstly, removal of the rim of the foramen magnum laterally to the jugular foramen, and secondly, removal of the inner third of the occipital condyle and the jugular tubercle. This procedure allows easy access to the ventral brainstem more inferolaterally without brainstem retraction. Case 2 demonstrated the usefulness of this procedure.

The transoral transclival approach has also been used for aneurysms located in the midline at the level of the lower third of the clivus. However, there is a high risk of meningitis associated with postoperative cerebrospinal fluid leakage because wound closure is fairly difficult.\textsuperscript{3} The limited surgical field is also a problem as the maximum bone removal is only 15 mm. This reduces intraoperative safety if a premature aneurysmal rupture occurs during operation.

An advantage of the transcondylar approach is the securing of the parent vertebral artery proximal to the aneurysm at an early stage. Also, midline vertebral or vertebrobasilar junction aneurysms can safely be approached with a wider exposure of the bilateral vertebral and basilar arteries through this transcondylar approach.

**Table 1** Persistent primitive hypoglossal artery aneurysms

<table>
<thead>
<tr>
<th>Author (Year)</th>
<th>Age, Sex</th>
<th>Site of aneurysm</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>Udvarhelyi and Lai (1963)\textsuperscript{11}</td>
<td>26, M</td>
<td>PHA-BA</td>
<td>conservative</td>
</tr>
<tr>
<td>Drakc (1968)\textsuperscript{8}</td>
<td>52, F</td>
<td>PHA-BA</td>
<td>wrapping</td>
</tr>
<tr>
<td>Huber and Rivoir (1974)\textsuperscript{9}</td>
<td>62, F</td>
<td>PHA-BA</td>
<td>unknown</td>
</tr>
<tr>
<td>Kodama et al. (1976)\textsuperscript{10}</td>
<td>34, F</td>
<td>PHA-BA</td>
<td>ligation</td>
</tr>
<tr>
<td>Bohnmalk and Story (1977)\textsuperscript{11}</td>
<td>44, F</td>
<td>PHA-BA</td>
<td>clipping</td>
</tr>
<tr>
<td>Bongartz et al. (1977)\textsuperscript{9}</td>
<td>28, F</td>
<td>PHA-BA</td>
<td>clipping</td>
</tr>
<tr>
<td>Waga et al. (1981)\textsuperscript{11,12}</td>
<td>54, F</td>
<td>PHA-BA</td>
<td>clipping</td>
</tr>
<tr>
<td>Murayama et al. (1985)\textsuperscript{10}</td>
<td>59, M</td>
<td>PHA-BA</td>
<td>clipping</td>
</tr>
<tr>
<td>Tsugu et al. (1990)\textsuperscript{11}</td>
<td>42, F</td>
<td>PHA-PICA</td>
<td>clipping</td>
</tr>
<tr>
<td>Present cases Case 1</td>
<td>33, M</td>
<td>PHA-PICA</td>
<td>clipping</td>
</tr>
<tr>
<td>Case 2</td>
<td>39, F</td>
<td>PHA-BA</td>
<td>clipping</td>
</tr>
</tbody>
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BA: basilar artery, PHA: primitive hypoglossal artery, PICA: posterior inferior cerebellar artery.

**References**

6) Huber P, Rivoir R: Aneurysm on a persistent left


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