

# Persistent Primitive Hypoglossal Artery Associated with Cerebral Aneurysm

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## ABSTRACT

A persistent primitive hypoglossal artery (PPHA) is a rare vascular anomaly, which belongs to the group of carotid-basilar anastomosis that may occur in adults. The association of PPHA with cerebral aneurysms has also rarely reported. A review of the published cases of PPHA with cerebral aneurysms is made including the author's own case of 66 years-old woman with the left internal carotid artery aneurysm, which treated with surgical clipping. Key points concerning the definition, imaging, and clinical series data available for PPHA with cerebral aneurysm are highlighted. (*Kor J Cerebrovascular Surgery* 8:132-4, 2006)

**KEY WORDS** : Persistent primitive hypoglossal artery · Cerebral aneurysm · Hypoglossal canal

## Introduction

The persistent primitive hypoglossal artery (PPHA) is a rare remnant of one of the four embryonal carotid-basilar anastomoses.<sup>9)</sup> It is reported to occur in 0.02~0.26% of angiograms or of autopsy cases.<sup>13)</sup> More than a hundred cases of PPHA have been reported in the literature. Usually the PPHA is detected as a coincidental finding in neuroradiological examinations for cerebral aneurysm,<sup>7)12)</sup> cerebral ischemic disease,<sup>11)</sup> arteriovenous malformation,<sup>5)13)</sup> brain tumor<sup>4)</sup> and so on. Some were discovered in patients with neurological findings related to PPHA itself such as syncope.<sup>6)</sup> In this report, we describe a case of PPHA associated with the left ICA aneurysm and discuss the radiological findings and clinical implications of PPHA.

## Case Report

A 66-year-old woman had a 5-day history of headache and photophobia. On admission, she was alert with no deficit other than a left-sided ptosis with an associated fixed dilated pupil. A complete work-up showed that the patient did not have diabetes mellitus. There was no history of trauma, hypertension, heart disease, stroke, or cancer. Computed tomographic (CT) scan showed no subarachnoid hemorrhage. However, three-dimensional CT angiography revealed an aneurysm of the left internal carotid artery (ICA) with postero-lateral direction. Contrast-enhanced CT with a bony window, with an axial view, demonstrated the anomalous artery to pass through the right hypoglossal canal (Fig. 1A). Three-dimensional CT angiography revealed a PPHA on the right side, which entered the skull via the hypoglossal canal (Fig. 1B). A right ICA angiogram demonstrated a PPHA originating from the ICA at the C-2 vertebral level (Fig. 2). The right Pcomm was well-developed, but the left Pcomm was aplastic. Vertebral angiography showed the left vertebral artery was hypoplastic and provided blood flow to the posterior inferior cerebellar artery without connection to the basilar artery. The right vertebral artery was not detected. An Aneurysm was originated from the left supraclinoid ICA.

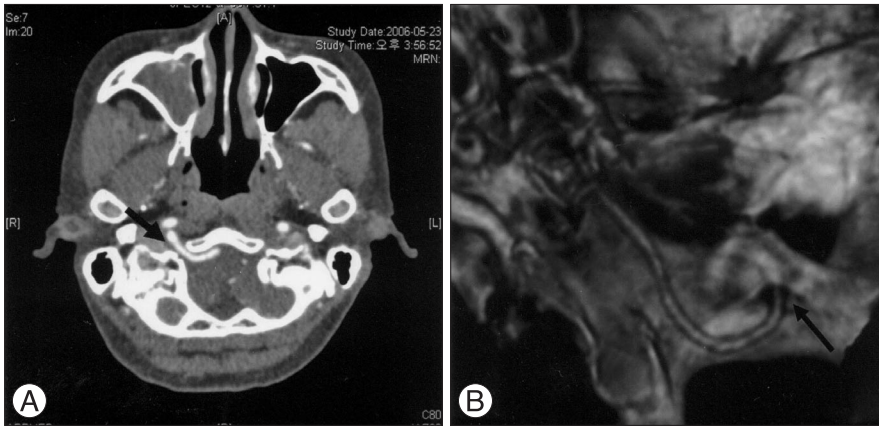
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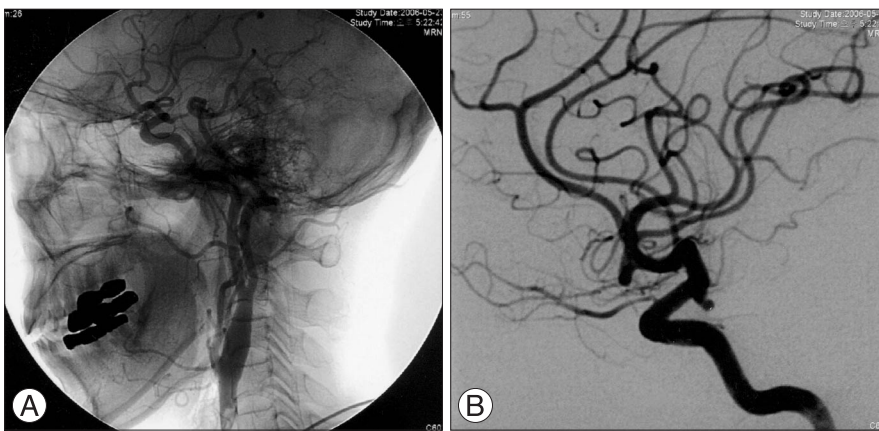
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**Fig. 1.** Contrast-enhanced CT with a bony window (A), with an axial view, demonstrates the anomalous artery (arrow) to pass through the right hypoglossal canal. Three-dimensional CT angiogram (B) shows a persistent primitive hypoglossal artery on the right side, which entered the skull via the hypoglossal canal (arrow).



**Fig. 2.** A right ICA angiogram (A) demonstrates a persistent primitive hypoglossal artery originating from the ICA at the C-2 vertebral level. A left ICA angiogram (B) reveals an aneurysm of the left internal carotid artery.

Clipping of the aneurysm was performed by a pterional approach because of decompression of aneurysmal mass effect to third cranial nerve. Intraoperatively, the anterior choroidal artery was seen, but the posterior communicating artery could not be identified. The aneurysm was successfully clipped and the anterior choroidal artery was preserved. The patient made a full recovery including complete resolution of her third cranial nerve palsy and was discharged.

## Discussion

A PPHA is one of the persistent embryonic carotid-basilar anastomoses.<sup>9)</sup> It is rarely encountered and is usually found incidentally at the time of cerebral angiography, and the estimated incidence has been reported to be 0.025%.<sup>13)</sup> The criteria for diagnosis of PPHA are that the artery arises from the cervical ICA at the levels of C-1 to C-3, the artery passes through the hypoglossal canal to the posterior cranial fossa, the basilar artery is filled only by the distal part of the

junction with the anastomosis, and angiography indicates deficiency or absence of the posterior communicating artery.<sup>2)</sup> When a PPHA is present, usually the vertebral artery is either absent on the ipsilateral side and hypoplastic on the opposite side, or hypoplastic on both sides.<sup>10)</sup> Our case satisfied these criteria. In addition, 3D-CT angiography demonstrated that the artery passed through the hypoglossal canal which confirmed the diagnosis. In this case, CT scan demonstrated the right hypoglossal canal to be larger than that on the left side. There were no findings suggesting any bony destruction of the hypoglossal canal. In the present case a CT scan and 3D-CT angiography were done, and these findings directly demonstrated the PPHA to pass through the large hypoglossal canal, which was the most reliable finding for the diagnosis of PPHA.

Patients with PPHA may have various associated lesions. Like other persistent primitive arteries, the main coexisting lesions are cerebral aneurysm and occlusive cerebrovascular disease.<sup>7)(11)(12)</sup> The clinical features of 134 patients with PPHA associated with intra- or extracranial lesions showed that

cerebral aneurysms were the most frequent association (26.9%), followed by occlusive cerebrovascular disease (20.9%), brain tumors (9.7%), and arteriovenous malformation (3.0%).<sup>13)</sup> These high incidences may be explained by the contribution of congenital factors. The fragility of the vascular wall and/or hemodynamic stress due to the presence of a PPHA are reportedly related to aneurysm formation. In the literature review, the patients were between 30 and 59 years of age (mean 45.6 years) in 65.8% of these cases, indicating that the age of patients with PPHA is lower than that usually seen in patients with subarachnoid hemorrhage.<sup>8)</sup> With respect to the location of these aneurysms, 31.4% were located at the PPHA-basilar artery junction and 53% in the posterior circulation. In cases in which anterior circulation aneurysms were present, the distal anterior cerebral artery and the middle cerebral artery were involved in 13.7% and 11.8% of cases, respectively; in only 5.9% of cases was the anterior communicating artery involved and in no reported case was the ICA-posterior communicating artery involved. In cases in which there was a PPHA, many aneurysms were located in the posterior circulation. The PPHA is considered to have some association with aneurysms arising in the posterior circulation. In our patient the PPHA was thought to have been unrelated to development of the aneurysm.

## Conclusions

The PPHA represents a rare carotid-basilar anastomosis. The existence of PPHA has been related with increased incidence of cerebral aneurysms. CT angiography provides excellent anatomic localization of PPHA in its parts and depicts clearly its entrance to the hypoglossal canal.

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