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# **Carotid Endarterectomy for Internal Carotid Artery Stenosis Associated with Persistent Primitive Hypoglossal Artery: Efficacy of Intraoperative Multi-modality Monitoring**

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## **Abstract**

Persistent primitive hypoglossal artery (PPHA) is a rare vascular anomaly and is usually asymptomatic. However, the PPHA may cause multi-territorial infarction when complicated by the internal carotid artery (ICA) stenosis. In this report, we describe a 73-year-old male who simultaneously developed cerebral infarction in both carotid and vertebrobasilar territories due to ICA stenosis associated with ipsilateral PPHA. The PPHA mainly provided blood flow to the vertebrobasilar territory in this case, because the bilateral vertebral arteries were markedly hypoplastic. He underwent carotid endarterectomy under internal shunting. Intraoperative multi-modality monitoring including angiography, motor evoked potential, and near infrared spectroscopy was very useful to avoid ischemic complications during surgery. Postoperative course was uneventful. It should be reminded that persistent carotid-basilar anastomosis can cause multi-territorial cerebral infarction mimicking cardiogenic embolism and may be a candidate for aggressive prophylactic intervention, when occlusive lesions develop in the carotid artery. It is very important to monitor hemodynamic and/or electrophysiological status in both carotid and vertebrobasilar territories to perform carotid endarterectomy safely in such cases.

## **Key words:**

persistent primitive hypoglossal artery, carotid stenosis, cerebral infarction, carotid endarterectomy, , intraoperative angiography

**Abbreviations:** CEA; carotidendarterectomy, CT; computed tomography, DWI; diffusion weighted imaging, ECG; electrocardiogram, GCS; Glasgow coma scale, ICA; internal carotid artery, MEP; motor evoked potential, MLF; medial longitudinal fasciculus, MRI; magnetic resonance imaging, NIRS; near infrared spectroscopy, PPHA; persistent primitive hypoglossal artery,

## **Introduction**

The primitive hypoglossal artery is one of the important embryonic anastomosis between the internal carotid artery and the longitudinal neural arteries, which later form the basilar artery. It normally regresses in the fetus, however, when it persists into adult life, it is called as persistent primitive hypoglossal artery (PPHA). The PPHA is usually observed incidentally on angiography. Previous reports have suggested that the PPHA may rarely cause multi-territorial cerebral infarction when complicated by stenosis of the ipsilateral internal carotid artery (ICA).

In this report, the authors describe a patient who presented cerebral infarction in both carotid and vertebrobasilar territories due to ICA stenosis associated with the ipsilateral PPHA. He safely underwent carotid endarterectomy under multi-modality monitoring to detect intraoperative critical ischemia in both carotid and verbrobasilar territories.

## **Case Report**

A 71-year-old male was treated with 100 mg/day of aspirin because of ICA stenosis for these 2 years. He suddenly developed consciousness disturbance and was admitted to our hospital. Neurological examinations on admission revealed consciousness disturbance (GCS=11), right oculomotor nerve palsy, left MLF syndrome, cortical blindness, and left hemiparesis. Diffusion-weighted MRI (DWI) on admission revealed acute cerebral infarction in the right frontal cortex, bilateral occipital lobe, bilateral cerebellar hemispheres, left pons and right midbrain (Fig. 1). Cerebral angiography and 3D-CT angiography demonstrated moderate stenosis of the right ICA (58% on NASCET criteria) associated with the ipsilateral PPHA (Fig. 2A). The bilateral vertebral arteries were markedly hypoplastic (Fig 2B), and the right PPHA mainly provided the blood flow to the vertebrobasilar territory. Precise cardiac examinations including 3D-CT angiography, Holter ECG, and transesophageal ultrasound revealed no cardiac source of embolism. Based

on these clinical presentation, the authors concluded that artery-to-artery embolism due to ICA stenosis caused multi-territorial cerebral infarction in both carotid and vertebrobasilar territories. He was medically treated and underwent rehabilitation. His neurological symptoms markedly improved except for cortical blindness. Thus, he could walk by himself with a cane 3 months after the onset.

At this timing, we considered that he could be the candidate for carotid endarterectomy to prevent further ischemic stroke, because he developed cerebral infarction in spite of medical treatment with aspirin. Intraoperative multi-modality monitoring was planned because the ipsilateral ICA supplied blood flow to both carotid and vertebrobasilar territories. Cerebral oxygenation status in the ipsilateral frontal lobe was continuously monitored using near infrared spectroscopy (NIRS, HEO-200; Omuron Co., Nagoya, Japan). Electrophysiological function in the motor tract was monitored using motor evoked potential (MEP, epoch XP; Axon Systems, NY, USA). Internal shunting was considered essential for him, because intraoperative cerebral ischemia might lead to cerebral infarction in the ipsilateral carotid and vertebrobasilar artery territories, resulting in disabling stroke. However, there was the possibility that the malpositioning of the internal shunt tube might interrupt the blood flow into either the ipsilateral carotid artery or PPHA. Therefore, we decided to employ intraoperative angiography to assess whether the internal shunt tube is correctly positioned during surgery or not.

Intraoperative angiography clearly demonstrated that the internal shunt tube was wedged in the right PPHA and did not supply the blood flow to the carotid territory when it was first inserted (Fig. 3A). Consecutive angiography showed that the internal shunt tube correctly supplied the blood flow to both carotid and vertebrobasilar territories when it was repositioned (Fig. 3B). Unstable, lipid-rich plaque was successfully removed (Fig. 4). No significant changes were observed on NIRS and MEP during surgery (data not shown).

Postoperative course was uneventful. Postoperative 3D-CT angiography demonstrated the resolution of ICA stenosis. The patient was discharged without any neurological deterioration. He has experienced no cerebrovascular events during about one year of follow-up period.

### **Discussion**

The persistent primitive hypoglossal artery (PPHA) is one of the rudimentary vessels forming the carotid-basilar anastomosis in the fetus. The embryologic development of the carotid and vertebrobasilar systems has been described by Paget, based on his extensive dissections of the 4-5 mm human embryo [1]. In the embryo, four vessels connect the primordial carotid artery with the longitudinal neural arteries in the 4-mm human embryo (30 days): the otic artery, the hypoglossal, and trigeminal and proatlantal artery. As the vertebral artery and basilar artery fuse at the 5 to 6-mm stage (around 40 days), anastomotic vessels gradually disappear: first the otic, and then the other three arteries. When it persists after birth, it is called as persistent primitive artery. Essential criteria for diagnosis of PPHA have initially been described by Lie [2] and later revised by Brismer [3]. According to their criteria, the PPHA fulfills the following findings: (1) the artery leaves the ICA as an extracranial branch at the level of C1-3; (2) the artery passes through the hypoglossal canal; and (3) the artery joins the caudal portion of the basilar artery. The PPHA usually represents an incidental finding in 0.027-0.26% of cerebral angiography [4]. In addition, persistent carotid-basilar anastomosis is often accompanied by hypoplasia or aplasia of the vertebral arteries [5].

The PPHA usually represents an incidental finding. However, it should be reminded that ICA stenosis associated with ipsilateral PPHA can be potential causes of cerebral infarction in both carotid and vertebrobasilar territories. In fact, previous reports have demonstrated that ICA stenosis associated with ipsilateral PPHA caused cerebral infarction in vertebrobasilar territory in three patients and in both carotid and vertebrobasilar territories in one. The latter is

very similar to the present case [5]. The PPHA has been believed to function as the pathway of artery-to-artery emboli into the vertebrobasilar territory in such cases [5-8]. Alternatively, other reports have presented two patients who developed watershed infarction in both carotid and vertebrobasilar territories due to tight ICA stenosis associated with ipsilateral PPHA. Hemodynamic insufficiency may provoke multi-territorial watershed infarction in these two cases, because the bilateral vertebral arteries were hypoplastic [9,10]. In the present case, therefore, right ICA stenosis caused artery-to-artery embolism in spite of medical treatments with aspirin, and multi-territorial infarction most likely occurred through the ipsilateral PPHA.

As pointed out previously, ICA stenosis associated with the ipsilateral PPHA may be the candidate for aggressive prophylactic intervention, because ischemic stroke frequently results in more serious neurological deficits than usual [11,12]. Recent clinical trials have shown that carotid artery stenting can be the alternative for CEA in patients with ICA stenosis. However, the authors decided to perform CEA, because the distance between the distal end of carotid plaque and the origin of the PPHA was too short (approximately 3.5 cm) to position a protection device correctly in this case.

The authors planned to monitor hemodynamic and electrophysiological status in both carotid and vertebrobasilar territories, because the cross-clamping of the carotid artery was considered to cause critical ischemia in both territories due to hypoplastic vertebral arteries [7,13-15]. NIRS has been accepted as a useful monitoring device to non-invasively detect critical cerebral ischemia during carotid surgery [16,17]. However, NIRS is not suitable to monitor oxygenation state in the brainstem because of its optical principles. As previously reported, EEG has also been considered as useful modality to detect critical ischemia during CEA [9]. However, EEG can not detect intraoperative ischemia in the brainstem. On the other hands, recent studies have suggested that MEP can sensitively detect electrophysiological

dysfunction of the motor tract due to critical ischemia [18]. Therefore, multi-modality monitoring with NIRS and MEP may be the best choice in carotid surgery for patients with ICA stenosis associated with ipsilateral PPHA, although no significant changes were observed in the present case. We do not consider that intraoperative angiography is always needed in carotid surgery. However, intraoperative angiography, in this case, was very useful to prevent perioperative complications by correcting the position of internal shunt tube. Therefore, intraoperative angiography would be valuable during carotid surgery in the selected patients.

In conclusion, it should be reminded that persistent carotid-basilar anastomosis with ICA stenosis may cause multi-territorial cerebral infarction mimicking cardiogenic embolism. Multi-modality monitoring to detect cerebral ischemia in both carotid and vertebrobasilar territories during surgery would be useful to safely perform CEA for ICA stenosis associated with the ipsilateral PPHA.

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## **Figure legends**

### **Fig. 1**

Diffusion-weighted MR imaging on admission showed acute cerebral infarction over multiple vascular territories including the right frontal cortex, bilateral occipital lobes, bilateral cerebellar hemisphere, and brainstem.

### **Fig. 2**

A: Right common carotid angiogram (lateral view) showed the moderate stenosis of the origin of internal carotid artery (arrow) and the persistent primitive hypoglossal artery (PPHA) originates from the ICA and joins the basilar artery (arrow heads).

B: Three-dimensional computed tomography angiography (3D-CTA, anterior view) demonstrated that the right well-developed PPHA mainly provided blood flow to the vertebrobasilar territory and that the vertebral arteries were hypoplastic on both sides.

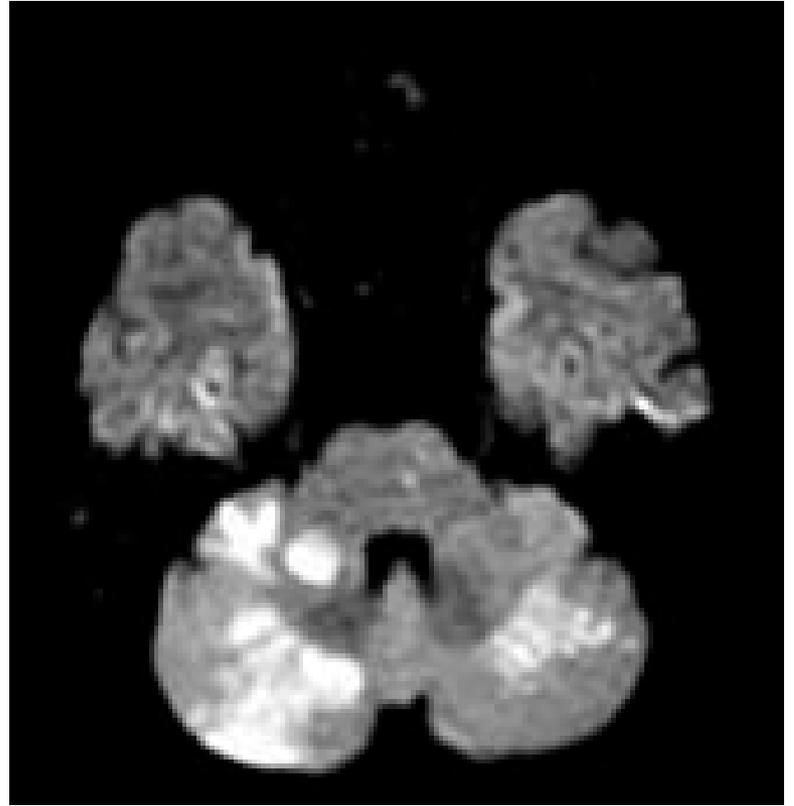
### **Fig. 3**

Intraoperative angiography showed that the distal end of the internal shunt tube (small arrows) was wedged into the PPHA (arrow) and the ICA was not opacified (A).

After re-positioning of the internal shunt tube, both ICA (arrowhead) and PPHA (arrow) were well opacified (B).

### **Fig. 4**

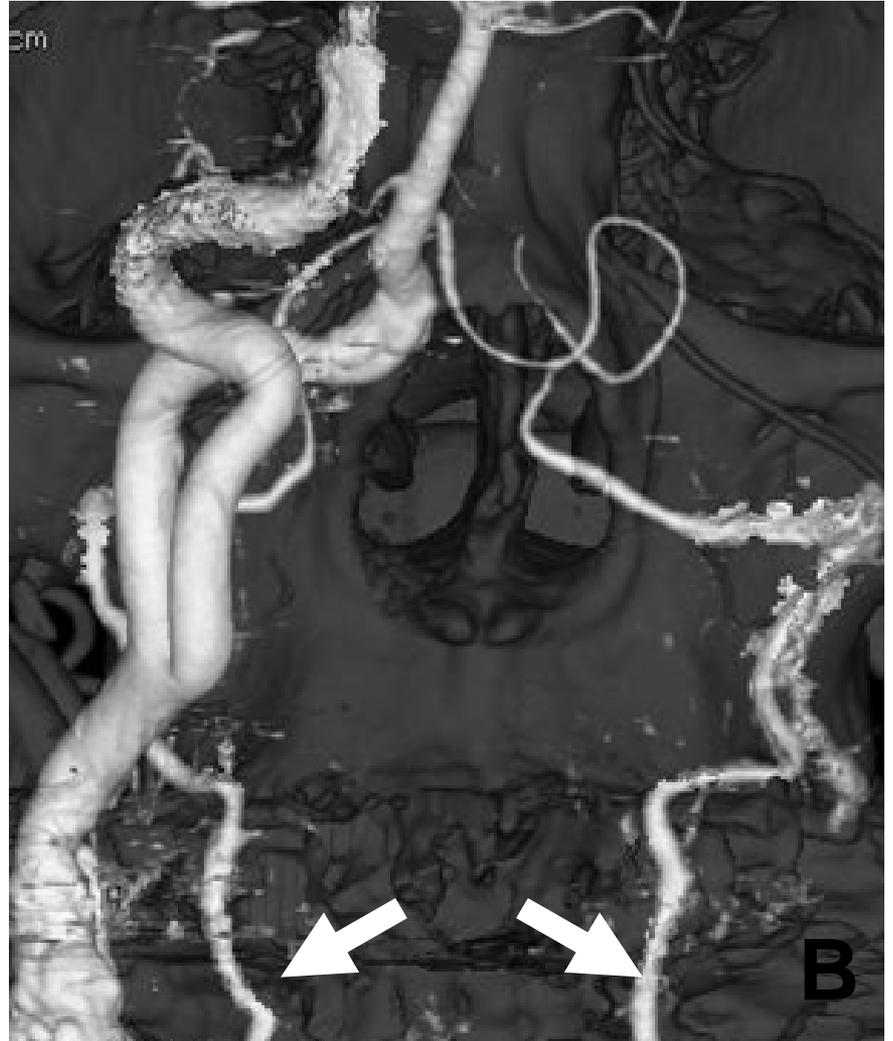
Intraoperative photograph showed the lipid-rich, unstable plaque with big ulcer in spite of moderate stenosis of the ICA.



**Fig. 1**

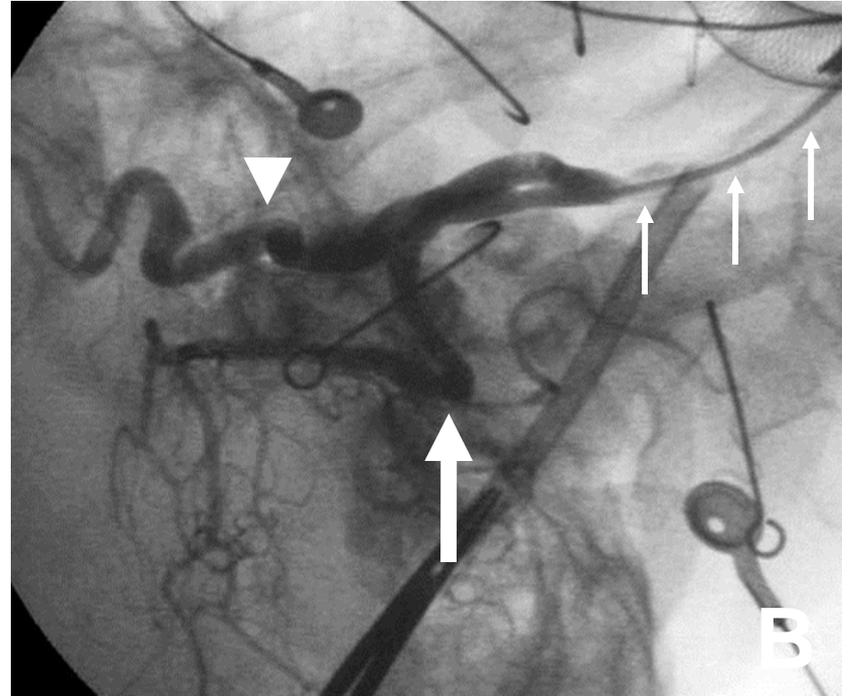
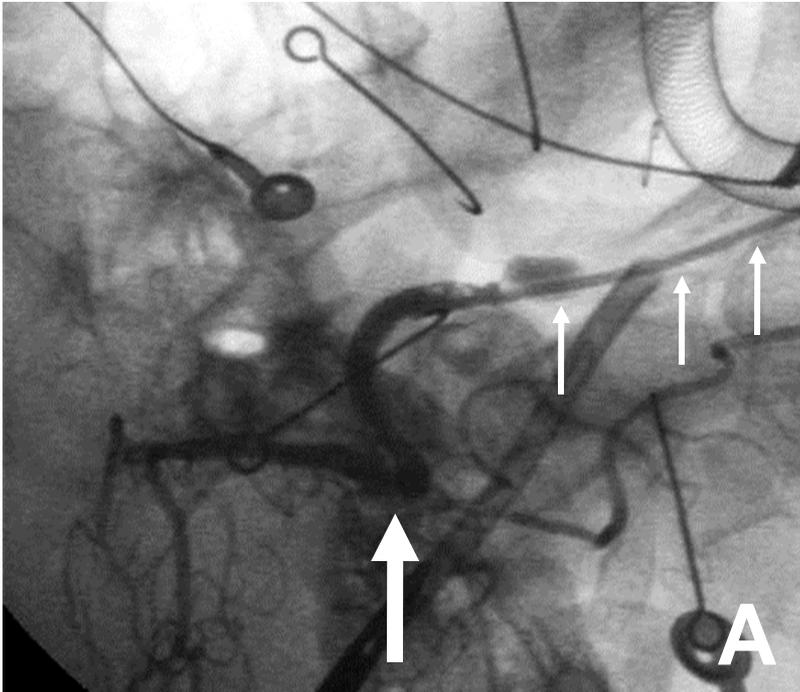


**A**

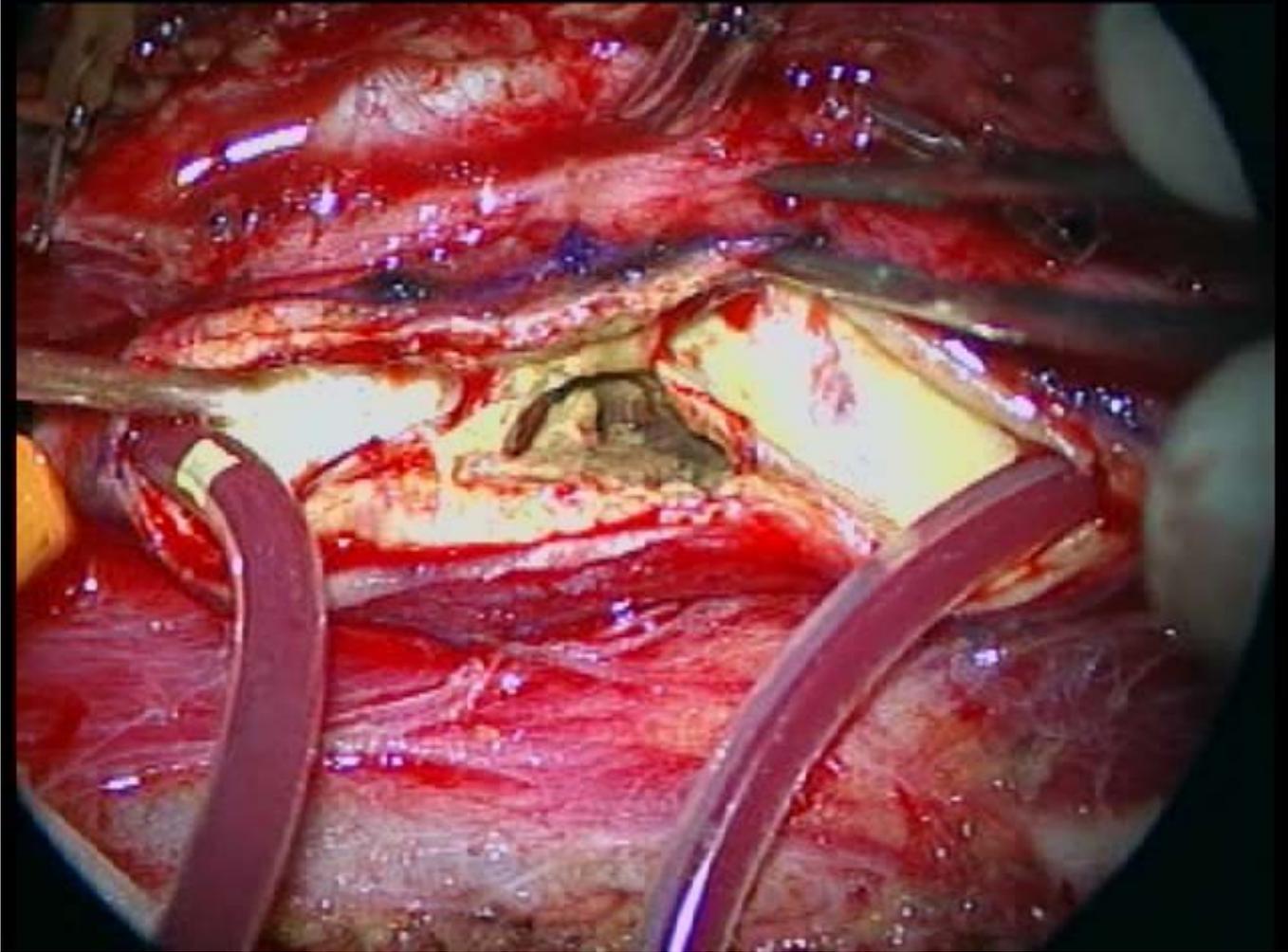


**B**

**Fig. 2**



**Fig. 3**



**Fig. 4**