

# **Carotid Artery Stenting in a Patient With Internal Carotid Artery Stenosis and Ipsilateral Persistent Primitive Hypoglossal Artery Presenting With Transient Ischemia of the Vertebrobasilar System**

## **—Case Report—**

Kouhei NII, Hiroshi AIKAWA, Masanori TSUTSUMI, Masanari ONIZUKA, Hiroya NAKAU, Ritsurou INOUE, Yoshinori GO, and Kiyoshi KAZEKAWA

Department of Neurosurgery, Fukuoka University Chikushi Hospital, Chikushino, Fukuoka

### **Abstract**

**A 62-year-old man experienced transient episodes of vertigo associated with left upper extremity weakness. Cerebral angiography showed 75% right internal carotid artery (ICA) stenosis and divergence of a persistent primitive hypoglossal artery (PPHA) distal to the stenosis. The area of stenosis was at a high position and he had a past medical history of congestive heart failure, which contraindicated carotid endarterectomy (CEA). Therefore, carotid artery stenting (CAS) was performed with single distal balloon protection. The stenotic area was restored and he was discharged without suffering recurrent attacks. CAS may be an effective alternative treatment to CEA to prevent further ischemic attacks in the posterior circulation in patients with PPHA. CAS using simple embolic protection devices is possible if the distance between the distal end of the ICA stenosis and the origin of the PPHA is sufficiently long.**

Key words: transient ischemic attack of vertebrobasilar system, cervical internal carotid artery stenosis, persistent primitive hypoglossal artery, carotid artery stenting, embolic protection device

### **Introduction**

The persistent primitive hypoglossal artery (PPHA) is one type of embryonal carotid-vertebrobasilar anastomosis, and is the second most common carotid-basilar anastomosis after persistent primitive trigeminal artery, with an estimated incidence of 0.02–0.26%.<sup>5,12,19)</sup> PPHA is rarely associated with internal carotid artery (ICA) stenosis resulting in ischemic attacks involving the vertebrobasilar system, and is usually addressed by carotid endarterectomy (CEA).<sup>5,6,9,11,12,14–16,22)</sup> Carotid artery stenting (CAS) is an alternative to CEA in high-risk patients.<sup>20)</sup> We treated a patient with an ipsilateral PPHA who underwent CAS for symptomatic ICA stenosis.

### **Case Report**

A 62-year-old man experienced transient episodes of vertigo associated with left upper extremity weakness. He had a past medical history of hypertension and congestive heart failure managed adequately by medications. His familial history was unremarkable. Carotid duplex ultrasonography performed at another hospital revealed right cervical ICA stenosis, but no other significant abnor-

malities. However, magnetic resonance (MR) angiography showed right ICA stenosis and ipsilateral PPHA, and diffusion-weighted MR imaging showed a small high intensity area in the left cerebellar hemisphere. He was transferred to our institution for further evaluation and treatment.

Neurological and physical examination on admission disclosed no abnormalities except for vascular bruit at the right neck. We performed cerebral angiography to confirm these findings and to develop a treatment strategy. Cerebral angiography showed 75% right ICA stenosis and a PPHA branching distal to the stenotic site (Fig. 1). The distance between the distal end of the ICA stenosis and the origin of the PPHA was 18 mm. Left internal carotid angiography demonstrated no abnormalities, but cross circulation through the anterior communicating artery was not well developed. The bilateral posterior communicating arteries were not visualized. The right vertebral artery was absent and the left vertebral artery was hypoplastic (Fig. 2). Blood was supplied to the right anterior and middle cerebral artery and to the vertebrobasilar system via the stenotic right ICA, but no contralateral collateral circulation was visualized. Revascularization was performed to prevent possible future ischemic attacks and to avoid recurrent attacks. CEA was considered difficult because the stenotic area was at a high position, at the level of the



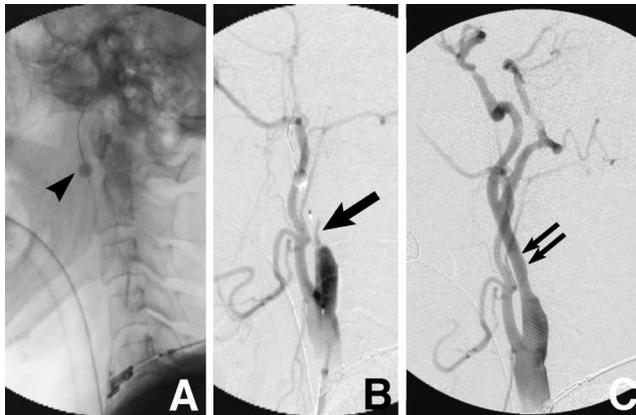
**Fig. 1** Preoperative right carotid angiogram showing severe internal carotid artery (ICA) stenosis (arrowhead) and branching of the persistent primitive hypoglossal artery (PPHA) (arrow). The distal end of the ICA stenosis did not extend to the origin of the PPHA.



**Fig. 2** Aortogram showing absence of the right vertebral artery (arrowhead) and hypoplasia of the left vertebral artery (arrow).

C2. In addition, congestive heart failure was a further risk factor for CEA.

Consequently, CAS with single distal balloon protection was performed under general anesthesia. This was possible because the distance between the distal end of the ICA stenosis and the origin of the PPHA was sufficiently long. Using an 8F guiding catheter (GuiderSoftip; Boston Scientific, Natick, Mass., U.S.A.), a GuardWire plus System (Medtronic, Minneapolis, Minn., U.S.A.) was inserted as a distal-balloon protection device into the distal ICA at a site proximal to the origin of the PPHA (Fig. 3A). Temporary occlusion of the ICA and PPHA was achieved with 6-mm diameter inflation of the balloon and confirmed by stagnation of the contrast material (Fig. 3B). Based on the vessel diameter, the stenosis was predilated with a 4-mm diameter balloon (Symmetry; Boston Scientific). The balloon was inflated for 30 seconds at nominal pressure. A Wall-stent RP (fully opened 10 mm diameter/20 mm length; Boston Scientific) was introduced based on our estimation of the normal ICA diameter (Fig. 3C). Each step was per-



**Fig. 3** Intraoperative right carotid angiograms showing a distal balloon protection device inserted into the distal internal carotid artery (ICA) at a site proximal to the origin of the persistent primitive hypoglossal artery (PPHA) (A: arrowhead), temporary occlusion of the ICA and PPHA confirmed by stagnation of the contrast material (B: arrow), and the area of stenosis restored and the distal edge of the stent not extending to the origin of the PPHA (C: double arrows).

formed under distal blocking. The occlusion time was approximately 11 minutes. The stenosis was restored and the patient awoke from general anesthesia without neurological deficits.

His postoperative course was good. Postoperative imaging showed no thromboembolic lesions. He was discharged with no recurrent attack 5 days after the operation. Cerebral angiography obtained 3 months later revealed no significant restenosis of the lesion.

## Discussion

Most persistent primitive carotid-basilar anastomoses including PPHA are found coincidentally.<sup>16)</sup> PPHA may be associated with cerebral aneurysm, ischemic cerebrovascular disease, brain tumor, or cerebral arteriovenous malformation.<sup>21)</sup> The posterior circulation is maintained primarily by the PPHA because the posterior communicating and vertebral arteries tend to be hypoplastic or aplastic.<sup>16)</sup> Therefore, cerebral blood flow in the ipsilateral ICA associated with a PPHA may be greater than the contralateral flow, resulting in intimal injury and stenosis.<sup>22)</sup> The diagnosis in the present case was transient ischemic attack in the posterior circulation because diffusion-weighted MR imaging showed a small high intensity area in the left cerebellar hemisphere. Patients with ischemic attacks involving the vertebrobasilar system may have PPHA associated with ICA stenosis.<sup>4-6,8,9,11,12,14-16,22)</sup> CEA was performed to prevent possible future ischemia or to avoid recurrent attacks in 15 patients.<sup>2,3,5,6,9-12,14-18,22)</sup> One reported patient underwent CAS for ICA stenosis associated with an ipsilateral PPHA.<sup>8)</sup> If CEA is performed for ICA stenosis in patients with PPHA, a skin incision at a higher position and placement of a special shunt such as a Y-shaped shunt may be necessary.<sup>6,11,17)</sup> On the other hand, CAS requires minimal invasion of cervical tissue. Tem-

porary occlusion, which can be observed under direct fluoroscopy, is safer and easier, and reduces the occlusion time to several minutes. We suggest that CAS offers advantages over CEA because it can be performed in patients with a history of congestive heart failure indicating high risk for CEA.

Various embolic protection devices for CAS have been reported.<sup>1,7,13</sup> A modification of Parodi's method was used to treat a patient with ICA stenosis and a PPHA.<sup>8</sup> Retrograde flow in the PPHA was induced by temporarily blocking the distal ICA and the external and common carotid arteries. Usually, proximal protection devices should take priority over distal protection devices in the treatment of stenosis with vulnerable plaque or nearly occlusion.<sup>13</sup> In our patient, the distance between the distal end of the ICA stenosis and the origin of the PPHA was 18 mm, sufficient to allow CAS with a single distal blocking system. The preprocedural examinations demonstrated stable plaque. We think that the use of simple embolic protection devices will avoid possible complications that may occur due to complex manipulations. Distal filter protection devices may be acceptable. However, if the distal end of the filter device is located in the tortuous ICA or PPHA, stable placement may be difficult and balloon devices may offer advantages in such situations.<sup>1</sup> The distance between the balloon device and the distal end of stent device was shortest in our usable devices.

Reduction of cerebral perfusion is the inherent drawback to all occlusion-type protection devices.<sup>7,13</sup> Intolerance to balloon inflation for several minutes is much less common or problematic than might be predicted. In our patient, the cerebrovascular reserve capacity was adequate in preoperative evaluations. The external carotid artery was free with the distal embolic protection device. We consider that the minimum distal blocking time and the collateral circulation from external carotid artery prevented hemodynamic ischemia of the territories of the ICA and PPHA.

If the lesion extends to near or beyond the origin of the PPHA, other procedures such as CAS with complex embolic protection devices, percutaneous balloon angioplasty, or CEA are preferable. Future advances in endovascular techniques and devices will make treatment of complicated cases easier.

The successful treatment of the present patient with ICA stenosis and ipsilateral PPHA shows that CAS may be effective for preventing further ischemic attacks in the posterior circulation involving a PPHA. CAS using a simple embolic protection device is possible if the distance between the distal end of the ICA stenosis and the origin of the PPHA is sufficiently long.

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Address reprint requests to: Kiyoshi Kazekawa, M.D., Department of Neurosurgery, Fukuoka University Chikushi Hospital, 1-1-1 Zokumyoin, Chikushino, Fukuoka 818-8502, Japan.  
e-mail: kazekawa@xb3.so-net.ne.jp