

Case Report

Endovascular treatment of acute atherothrombotic internal carotid artery occlusion associated with persistent primitive hypoglossal artery

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ABSTRACT

Persistent primitive hypoglossal artery is a relatively rare anatomical variation and a type of persistent carotid–basilar anastomosis. Acute internal carotid artery occlusion associated with persistent primitive hypoglossal artery is rare, and atherothrombotic occlusion is extremely rare. We present a case of acute atherothrombotic internal carotid artery occlusion associated with persistent primitive hypoglossal artery that was successfully treated by endovascular treatment. A 70-year-old male with a history of left internal carotid artery stenosis was transferred to our hospital by ambulance because of abnormal behaviors and aphasia. He was diagnosed with cerebral infarction and left internal carotid artery occlusion. Left carotid angiography revealed the persistent primitive hypoglossal artery arising from the cervical internal carotid artery and complete internal carotid artery occlusion distal to the origin of the persistent primitive hypoglossal artery. Therefore, we performed endovascular treatment. Mechanical thrombectomy was performed under minimal flow arrest with consideration of brain ischemia causing coma. After additional balloon angioplasty, recanalization was achieved, and the patient's symptoms improved. During the 1.5-year follow-up period, no recurrence or restenosis was observed. This report provides evidence that atherosclerotic internal carotid artery stenosis associated with persistent primitive hypoglossal artery can occur even distal to the origin of the persistent primitive hypoglossal artery and that the lesion may become acutely occluded, leading to acute stroke. Endovascular treatment considering brain ischemia was effective in this case.

1. Introduction

Persistent primitive hypoglossal artery (PPHA) is a relatively rare anatomical variation and one of the persistent carotid–basilar anastomoses [1]. Acute internal carotid artery (ICA) occlusion associated with PPHA is rare [2]. In particular, acute atherothrombotic ICA occlusion distal to the PPHA origin is extremely rare and, to the best of our knowledge, it has not been reported previously. Herein, we present a case of acute atherothrombotic ICA occlusion associated with PPHA that was successfully treated by endovascular treatment (EVT).

2. Case report

A 70-year-old male was found walking with one shoe missing. He had aphasia, so he was transported by ambulance to our hospital. He had a medical history of hypertension, dyslipidemia, and hyperuricemia. He also had left ICA stenosis and was taking antiplatelet therapy (clopidogrel 75 mg daily), but the presence of persistent carotid–basilar anastomosis was not recognized (Fig. 1A). On emergency arrival, his consciousness was impaired and he has total aphasia and facial paralysis (National Institutes of Health Stroke Scale score 7/42). Magnetic

Abbreviations: BGC, balloon guide catheter; EVT, endovascular treatment; ICA, internal carotid artery; MRA, magnetic resonance angiography; PPHA, persistent primitive hypoglossal artery.

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resonance angiography (MRA) revealed left ICA occlusion (Fig. 1B). MRA also showed no obvious previous changes in the vertebrobasilar system, and the right vertebral artery and bilateral posterior communicating arteries were hypoplastic. Magnetic resonance imaging indicated only small infarcts (Fig. 1C). Therefore, we performed EVT under local anesthesia.

Left internal carotid angiography revealed the PPHA arising from the cervical ICA and complete ICA occlusion distal to the PPHA origin (Fig. 2A and B). As the ICA distal to the PPHA origin had a small vessel diameter and a steep branching angle, the balloon guide catheter (BGC; Optimo EPD 9 F, Tokai Medical Products, Kasugai, Aichi, Japan) was positioned proximal to the PPHA origin for intervention. We approached the occluded lesion, but it was too firm to cross. First, contact aspiration was performed using SOFIAFLOW 5 F (MicroVention, Tustin, CA, US). Balloon inflation of the BGC during retrieval caused the patient to enter a coma, which resolved immediately after deflation of the balloon. Recanalization was not achieved. Second, we successfully crossed the lesion and performed mechanical thrombectomy with a stent retriever (Embotrap III 5 × 37 mm; Johnson & Johnson, New Brunswick, NJ, US) and an aspiration catheter. BGC balloon inflation during retrieval was

started from the occluded lesion to minimize brain ischemia causing coma. A large amount of thrombus was retrieved, and recanalization was achieved. Residual stenosis remained, and the lesion became occluded once again. Therefore, we performed additional balloon angioplasty using the 2.5 mm × 9 mm Gateway balloon catheter (Stryker, Kalamazoo, MI, US). We also administered intravenous infusion of ozagrel sodium (a thromboxane A2 synthase inhibitor). Successful recanalization and adequate dilatation were achieved without stent placement and the patient's symptoms improved. Follow-up angiography 1 month after EVT demonstrated mild, but not progressive, stenosis at the left ICA distal to the PPHA origin (Fig. 2C). Cone-beam computed tomography also confirmed a persistent carotid–basilar anastomosis passing through the hypoglossal canal (i.e., PPHA) (Fig. 2D). Any other causes were ruled out, and we finally diagnosed atherothrombotic brain infarction due to acute ICA occlusion distal to the PPHA origin. During the 1.5-year follow-up period after EVT, no recurrence or restenosis occurred.

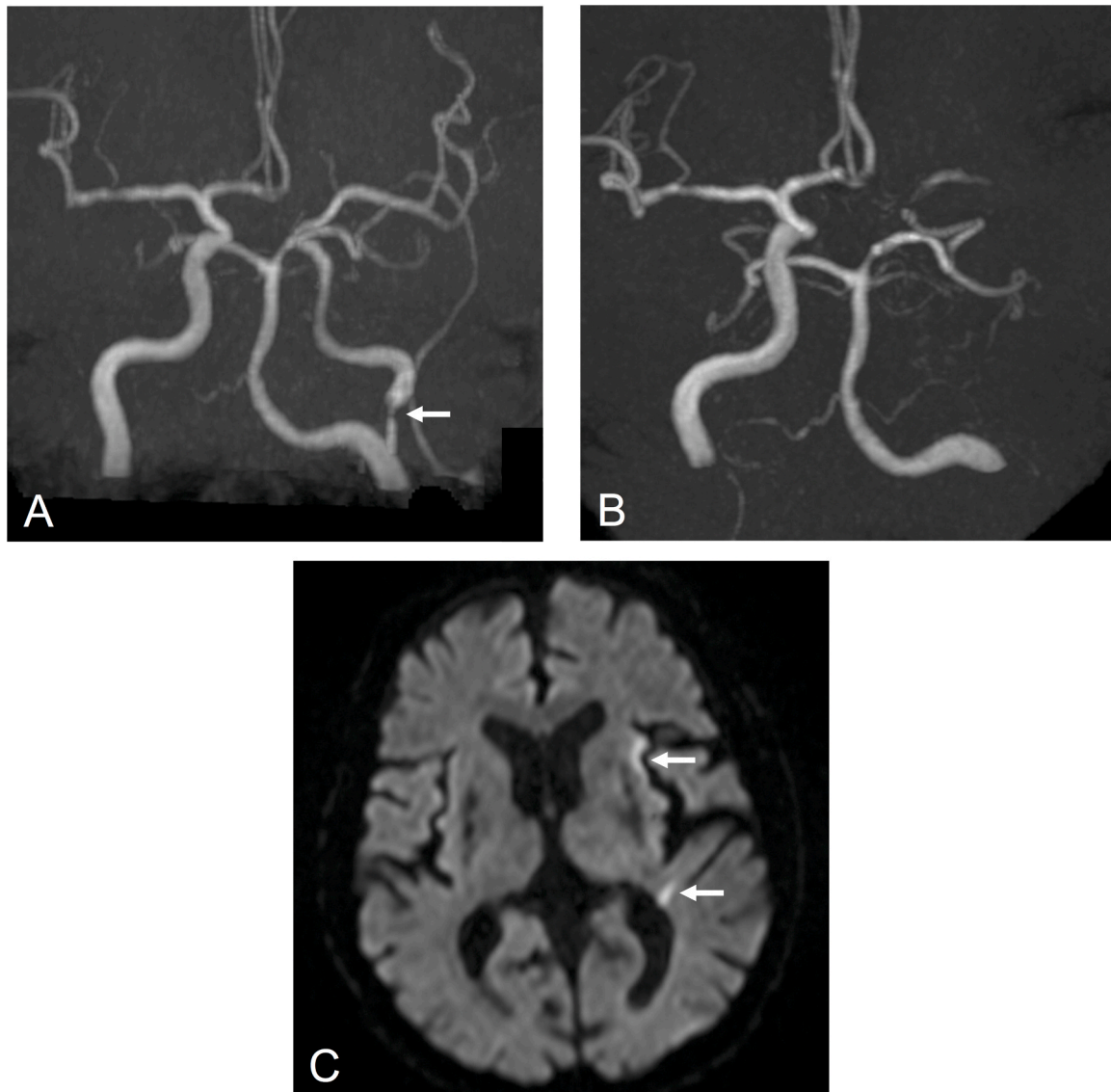


Fig. 1. Imaging findings in a case of acute atherothrombotic internal carotid artery occlusion associated with persistent primitive hypoglossal artery. (A) Magnetic resonance angiography 1 year before the onset of cerebral infarction showing stenosis at the left internal carotid artery (arrow). (B) Magnetic resonance angiography on admission revealing left internal carotid artery occlusion. (C) Magnetic resonance imaging indicating only small infarcts (arrows).

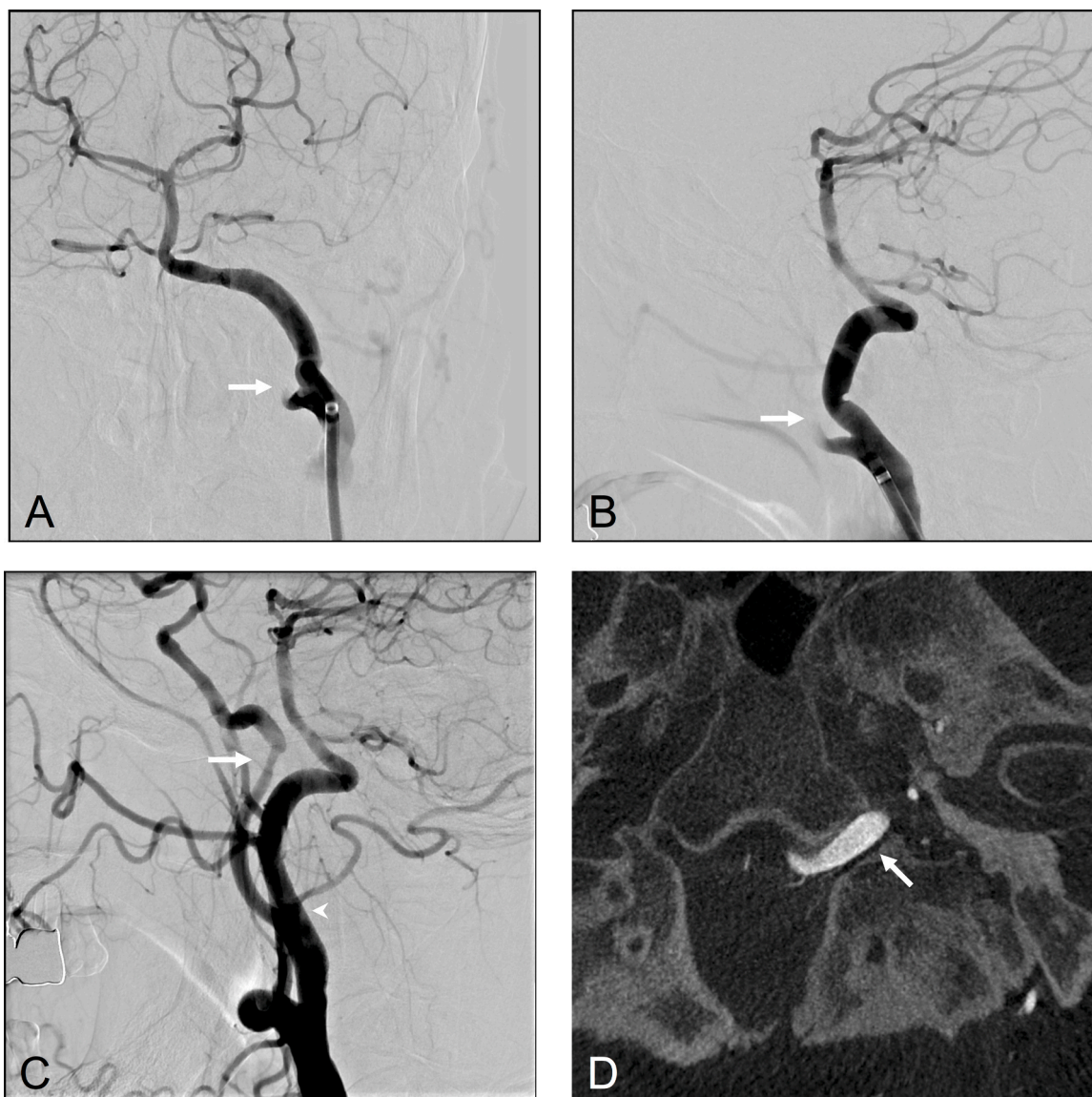


Fig. 2. Angiographic findings in endovascular treatment of acute atherothrombotic internal carotid artery occlusion associated with persistent primitive hypoglossal artery. (A, B) Pre-procedural angiography (A: anteroposterior view; B: lateral view) demonstrating complete left internal carotid artery occlusion distal to the origin of the persistent primitive hypoglossal artery (arrows). (C) Follow-up angiography 1 month after endovascular treatment showing mild left internal carotid artery stenosis (arrow). The arrowhead indicating the origin of the persistent primitive hypoglossal artery. (D) Cone-beam computed tomography confirming persistent carotid–basilar anastomosis passing through the hypoglossal canal (arrow).

3. Discussion

In this study, we present a rare case of acute atherothrombotic ICA occlusion associated with PPHA, which was successfully treated by EVT. PPHA is a type of persistent carotid–basilar anastomosis with a reported incidence of 0.027%–0.29% [1]. The diagnostic criteria for PPHA according to Brismar's revised criteria are as follows: (1) originating from the ICA as a large extracranial branch, (2) passing through the hypoglossal canal, and (3) having a basilar artery trunk originating from an anomalous artery (PPHA) [1]. The most significant of these criteria is the artery passing through the hypoglossal canal (Fig. 2D). In our case, the PPHA was not recognized before the onset of acute stroke, and its presence was not noticeable on maximum-intensity projection images of intracranial MRA (Fig. 1). It is important to have knowledge of persistent carotid–basilar anastomoses, such as PPHA, although there may be a limited likelihood of encountering them in the context of acute stroke.

Acute ICA occlusion associated with PPHA is rare, with only a few cases reported in the past [2]. With regard to the etiology of acute ICA

occlusion associated with PPHA, previous cases have been diagnosed as cardiogenic embolism [2]. Atherothrombotic ICA occlusion, as in our case, is extremely rare, and to our best knowledge, this phenomenon has not been previously reported. This report provides evidence that acute ICA occlusion associated with PPHA can occur even in atherothrombotic brain infarction (Fig. 1). There have been several case reports of patients with atherosclerotic ICA stenosis associated with PPHA who underwent revascularization (i.e., percutaneous transluminal angioplasty, endovascular stenting, carotid endarterectomy) [3–5]. With regard to the location of ICA stenosis associated with PPHA, stenoses proximal to the PPHA origin have been typically identified [3,5]. However, the PPHA origin has rarely been reported [3,4]. We did not identify any similar reports of atherosclerotic stenosis of the ICA distal to the PPHA origin, as in our case (Fig. 2). This report provides evidence that atherosclerotic ICA stenosis associated with PPHA can occur distal to the PPHA origin.

We performed EVT (mechanical thrombectomy and percutaneous transluminal angioplasty) for acute atherothrombotic occlusion of the ICA distal to the PPHA origin (Fig. 2). When performing EVT, several

technical issues were encountered that should be considered in future cases. First, anatomical findings associated with the PPHA should be considered. The BGC was placed proximal to the PPHA origin because the ICA distal to the PPHA origin was small in diameter and its branching angle was steep, although it would be ideal to insert the BGC into the ICA distal to the PPHA origin. Anatomical details regarding the association of the ICA with the PPHA are limited, which requires decisions to be made on a case-by-case basis. The same applies to the choice of device (aspiration catheter, stent retriever, or balloon catheter), and we would select smaller sizes than usual in such cases. Second, during the procedure, ICA- and PPHA-related ischemia should be considered. In our case, BGC balloon inflation caused coma. We considered this to be because the vertebral arteries and posterior communicating arteries were hypoplastic, and the PPHA played an important role in the perfusion of the posterior circulation (Fig. 1). Therefore, we should recognize that the PPHA is a principal vessel in the posterior circulation. In the present case, we performed EVT under minimal flow arrest in case of brain ischemia, which is another important consideration. We would like to emphasize that the most clinically important points are the procedural issues that must be considered in this particular intervention, such as anatomical variations and precautions to avoid brain ischemia.

4. Conclusion

We describe an extremely rare case of acute atherothrombotic occlusion of the ICA distal to the PPHA origin. This report provides evidence that atherosclerotic ICA stenosis associated with PPHA can occur even distal to the PPHA origin and that the lesion may become occluded, leading to acute stroke. EVT, considering the anatomical findings and brain ischemia associated with PPHA, was effective in our case.

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Ethical statement

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institution and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. The study was approved by the Ethics Committee of Nakamura Memorial Hospital (No. 2023120501), and informed consent was obtained from the patient.

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Declaration of Competing Interest

None.

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