A Case of Carotid Artery Stenosis Associated with Persistent Primitive Hypoglossal Artery Treated by Carotid Artery Stenting with Double Distal Protection Using the Jail Technique

Yuichi Mochizuki,^{1,2} Takashi Higa,³ Yukiya Iwata,¹ and Takakazu Kawamata²

Objective: Ingenuity is required in treating carotid artery stenosis associated with persistent primitive hypoglossal artery (PPHA) because of anatomical singularity.

Case Presentation: A 69-year-old male presented with right thalamic infarction and was found to have marked stenosis of the right internal carotid artery (ICA). PPHA extending from a site distal to the stenosed area of the right ICA to the right vertebral artery was observed. Since there were few collateral channels for the posterior circulation, one FilterWire EZ (Boston Scientific, Natick, MA, USA) each was placed in the ICA and PPHA, and carotid artery stenting was performed.

Conclusion: The effectiveness of the double distal filter protection technique for ICA stenosis associated with PPHA and tips for the retrieval of the filter devices are reported.

Keywords ► persistent primitive hypoglossal artery, carotid artery stenting, double distal protection, closed-cell stent, filter device

Introduction

Persistent primitive hypoglossal artery (PPHA) is a persistent embryonic carotid vertebrobasilar artery anastomosis with a reported prevalence of 0.1%–0.25%.¹⁾ Since PPHA branches off the internal carotid artery (ICA), a therapeutic strategy different from usual carotid artery stenting (CAS) is required. In the case presented here, we performed CAS by placing one FilterWire EZ (Boston

Scientific, Natick, MA, USA) each in the ICA and PPHA for carotid artery stenosis associated with PPHA. The therapeutic strategy and tips for the retrieval of FilterWires are reported.

Case Presentation

The patient was a 69-year-old male who presented with left-sided numbness and was diagnosed with right thalamic infarction. Carotid artery ultrasonography showed 85% stenosis (European Carotid Surgery Trial [ECST]) and a marked increase in peak systolic velocity (PSV) at 368 cm/sec. Common carotid artery angiography revealed stenosis of the right ICA and a persistent primitive artery extending from the right ICA to the right vertebral artery (**Fig. 1A**). The posterior circulation was clearly visualized on right common carotid artery angiography (**Fig. 1B** and **1C**). The bilateral vertebral arteries were hypoplastic (**Fig. 1D** and **1E**). In source images of contrast-enhanced CT, the persistent primitive artery was confirmed to pass through the hypoglossal canal and was diagnosed as PPHA (**Fig. 2**). On 3D-CTA, the bilateral posterior communicating arteries were not delineated (**Fig. 2**).

¹Department of Neurosurgery, Higashi Saitama General Hospital, Saitama, Saitama, Japan

²Department of Neurosurgery, Tokyo Women's Medical University, Tokyo, Japan

³Department of Neurosurgery, Tokyo Rosai Hospital, Tokyo, Japan

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Corresponding author: Yuichi Mochizuki, Department of Neurosurgery, Higashi Saitama General Hospital, 517-5 Yoshino, Satte-shi, Saitama 340-0153, Japan

Email: mochiizuki.ns@gmail.com

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Fig. 1 Preoperative images. (A) Lateral view of common carotid artery angiography. Arrows indicate internal carotid artery. Dotted arrow indicates PPHA. Frontal (B) and lateral (C) view of common carotid artery angiography. Arrows indicate internal carotid artery. Dotted arrow indicates PPHA. Double arrow indicates basilar artery. Right brachiocephalic artery (D) and left subclavian artery (E) angiography. Arrow heads indicate vertebral arteries. PPHA: persistent primitive hypoglossal artery

After admission, the occurrence of extensive ischemia of the posterior circulation territory associated with the inadequate blood flow of the right ICA was considered possible due to the scarcity of collateral channels for the posterior circulation. Therefore, CAS was performed on the 2nd hospital day.

For the above reasons, filter protection instead of distal balloon protection was judged to be appropriate. In the right common carotid artery, an 8 Fr Optimo (Tokai Medical Products, Aichi, Japan) was placed via the right femoral artery, and a 6 Fr FUBUKI (Asahi Intecc, Tokyo, Japan) was placed via the left femoral artery. A FilterWire EZ was deployed in PPHA from FUBUKI. A retrieval sheath was placed distally to the planned stenting site as a standby device. From the Optimo, another FilterWire EZ was guided to the distal part of the ICA and deployed (**Fig. 3A**). Pre-dilatation was performed using a Jackal RX 3.0 × 40 mm (Kaneka Medix, Osaka, Japan) at 8 atmospheres over 30 seconds. After a Carotid Wallstent 10 × 24 mm (Boston Scientific) was deployed from the ICA proximal to

Fig. 2 Preoperative images. 3D CT angiography and axial image of hypoglossal canal. PPHA passes through the hypoglossal canal. PPHA: persistent primitive hypoglossal artery

Fig. 3 Intraoperative images. **(A)** Each FilterWire EZ (Boston Scientific) is deployed at internal carotid artery (arrows) and PPHA (double arrows). **(B)** Postoperative angiography shows intact stent deployment. PPHA: persistent primitive hypoglossal artery

the bifurcation of PPHA to the common carotid artery, post-dilatation was performed using a Jackal RX 4.5 × 30 mm at 6 atmospheres over 30 seconds. The FilterWire EZ in PPHA was housed in the retrieval sheath, which had been placed close to it in advance, and retrieved through the space outside the Carotid Wallstent (Boston Scientific). There was no particular resistance in the retrieval, and no effect on the stent was noted (**Fig. 3B**). The FilterWire EZ in the ICA was retrieved by the usual procedure. While slow flow or no flow was not observed during the procedure, a small amount of debris was present in both of the retrieved filters. MRI (diffusion-weighted magnetic resonance imaging [DWI]) performed on the day after the procedure showed no distal embolism.

Discussion

There are two points in this report. One is that double distal protection using filter devices was necessary for anatomical reasons. The other is the retrieval of filter devices deployed outside the stent.

The prevalence of PPHA has been reported to be 0.1%–0.25%,¹⁾ and its relationship with carotid artery stenosis has been suggested.²⁾ Concerning its treatments, carotid endarterectomy (CEA) using a T-shaped shunt has been reported,³⁾ but there have also been reports that CAS was selected because CEA required shunt insertion at a high position.⁴⁾ There have been a few reports of CAS performed for carotid artery stenosis associated with PPHA,⁴⁾ and the therapeutic strategy varies widely depending

on whether the lesion is symptomatic or not, degree of development of the vertebral and posterior communicating arteries, distance from the distal end of the plaque to the bifurcation of PPHA, etc.

In our patient, the distance from the distal end of the plaque to the bifurcation of PPHA was short, and it was difficult to assure the placement of a distal protection device in the ICA proximally to the bifurcation of PPHA. In addition, as the disorder was symptomatic, and collateral channels were few, it was preferred to avoid balloon occlusion. We, therefore, selected, double distal protection using filter devices.

When double distal protection is used, one filter is jailed by the stent, and its retrieval poses a problem. Since it is difficult to pass the retrieval sheath outside the stent, it is necessary to place a catheter for retrieval of the filter distally to the planned site of stenting in advance (**Fig. 4A**). We used a closed-cell stent in consideration of 1) stent deformation after jailing of the catheter and 2) sticking of the catheter by the stent/squeezing out of thrombus during catheter removal. Similarly, double distal stenting using the jailing technique has been reported previously,⁵⁾ but this procedure differs from ours in that an open-cell stent was used. The procedure seems to have been completed without problem even with an open-cell stent, but, for the above reasons, and as soft plaque was suspected by the pre-operative examinations, we judged the use of a closed-cell stent to be safer. In our patient, thrombus was noted in the retrieved filters, but no distal embolism occurred, and the thrombus is considered to have been retrieved safely.

Fig. 4 Schema of jail technique. (A) Retrieval catheter (double arrow) is already put on distal side of stent (dotted arrow). Arrows indicate FilterWire. (B) Carotid Wallstent (Boston Scientific) is deployed. FilterWire EZ (Boston Scientific) and retrieval catheter are jailed. (C) FilterWire EZ is stored in retrieval catheter. (D) Retrieval catheter is withdrawn. PPHA: persistent primitive hypoglossal artery

Conclusion

The effectiveness of the double distal filter protection technique using filter devices and tips for their retrieval in a patient with symptomatic ICA stenosis associated with PPHA were reported.

Disclosure Statement

There are no conflicts of interest to disclose regarding this paper.

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