

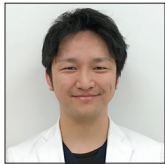
Technical Notes

Strategy of cerebral endovascular treatment for cervical internal carotid artery stenosis with a persistent primitive hypoglossal artery

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ABSTRACT

Background: Persistent primitive hypoglossal artery (PPHA) is a rare residual arterial anastomosis. We placed a CASPER stent using Spider FX as an embolic protection device (EPD) in a patient with internal carotid artery (ICA) stenosis and PPHA. There are no reports of carotid artery stenting (CAS) using a CASPER stent for ICA stenosis with PPHA. We report the EPD strategy used in this case and the usefulness and precautions of CASPER stent insertion for cervical ICA stenosis in association with PPHA.

Methods: A 9Fr sheath was placed in the right femoral artery and a 9Fr Branchor balloon guide catheter was guided to the common carotid artery. A Spider FX was placed proximal to the bifurcation of the ICA and the PPHA. A 10 mm × 20 mm CASPER stent was deployed at the site of the stricture with no postoperative ischemic complications.

Results: There was no intra-stent occlusion, stenosis, or plaque protrusion immediately after surgery, and no postoperative ischemic complications were observed.

Conclusion: CASPER stent deployment with the Spider FX in the ICA and PPHA bifurcation can be considered to be an effective treatment method for ICA stenosis associated with PPHA. However, care should be taken in selecting the appropriate EPDs and stents depending on the location of the stenosis and bifurcation of the PPHA.

Keywords: carotid artery stenting, carotid stenting, CASPER, persistent primitive hypoglossal artery

INTRODUCTION

Persistent primitive hypoglossal artery (PPHA) is a rare residual artery anastomosis.^[3,11,15] We used a Spider FX (Covidien, Mansfield, MA, USA) as an embolic protection device (EPD) to deploy a CASPER stent (Terumo, Tokyo, Japan) in a patient with internal carotid artery (ICA) stenosis associated with PPHA. There are only four previously reported cases of stent placement for ICA stenosis associated with PPHA.^[1,2,5-8,10,12-14,18] We report a strategy using a CASPER stent and Spider FX and compare it with the methods described in the previous reports.

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MATERIALS AND METHODS

A 76-year-old male patient with repeated episodes of fainting, apraxia, and right-sided weakness was diagnosed by magnetic resonance imaging and digital subtraction angiography with ICA stenosis and PPHA [Figure 1], for which he was scheduled to undergo carotid artery stenting (CAS). Dual antiplatelet therapy (DAPT) using aspirin and clopidogrel was administered 1 week before surgery. Using the VerifyNow System (Accriva Diagnostics, San Diego, CA, USA) to monitor antiplatelet therapy preoperatively, we determined that DAPT was effective based on test results of <550 Aspirin Reaction Units (ARUs) and <208 P2Y12 Reaction Units (PRU). Subsequently, surgery was performed under general anesthesia. A 9Fr sheath was placed in the right femoral artery and a 9Fr Branchor balloon guide

catheter (BGC) (Asahi Intecc, Aichi, Japan) was guided into the common carotid artery (CCA). A Spider FX was placed proximal to the bifurcation of the ICA and the PPHA, and a 10 mm × 20 mm CASPER stent was deployed in the stricture. After stent deployment, an RX-Genity balloon catheter for percutaneous transluminal angioplasty (7.0 mm × 40 mm, Kaneka Medics, Osaka, Japan) was used to dilate the stent at 6 atm for 5 s.

RESULTS

The results of preoperative VerifyNow System testing were 415 ARU and 159 PRU. The distance between the catheter tip of the Spider FX and the stent delivery system before stent placement was 6.5 mm [Figure 2a]. The CASPER stent was successfully deployed. However, the catheter tip of the

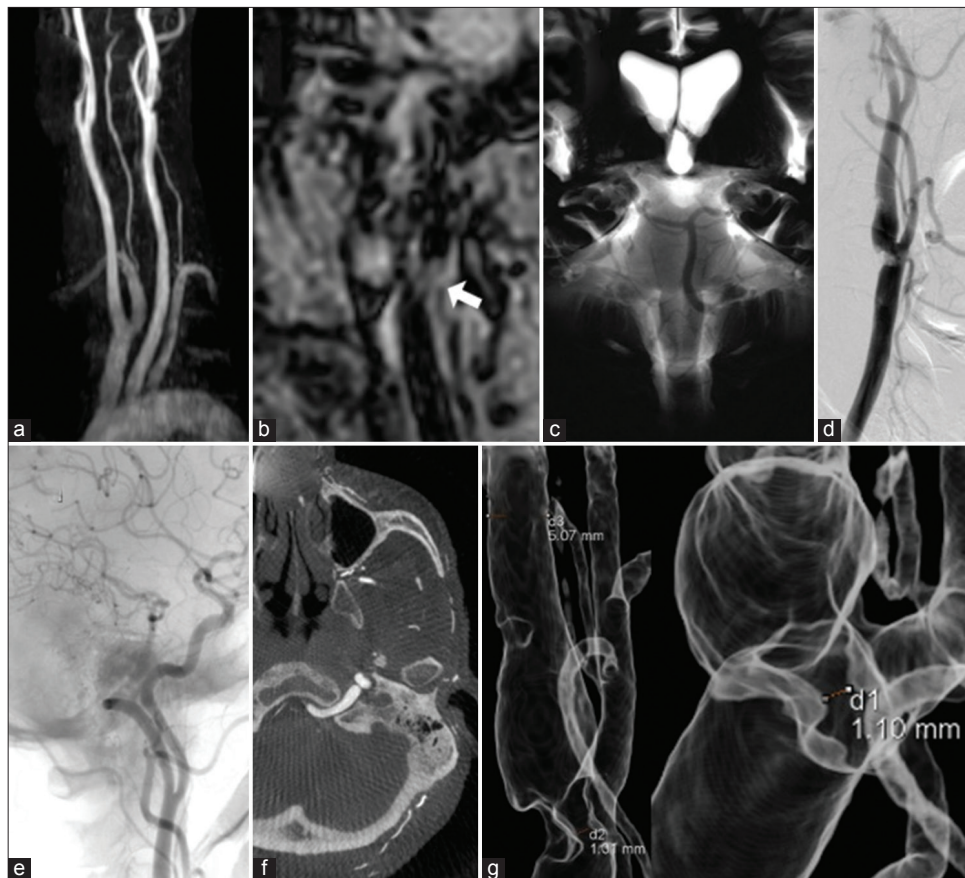


Figure 1: A 76-year-old male patient was brought to the emergency department due to repeated episodes of fainting, apraxia, and right-sided weakness. (a) Magnetic resonance angiography showed stenosis of the left internal carotid artery (ICA). (b) T1-weighted image black-blood magnetic resonance imaging (MRI) showed that the stenotic part had normal signal intensity (white arrow). (c) Basi-parallel anatomical scanning MRI revealed right vertebral artery hypoplasia. (d) Pre-procedure digital subtraction angiography (DSA) demonstrated two areas of stenosis, one immediately after the bifurcation and one distal to it. (e) Pre-procedure DSA of left common carotid angiography demonstrated the primitive hypoglossal artery distal to the stenosis of the ICA. (f) Pre-procedure three-dimensional rotational angiography (3D-RA) demonstrated the primitive hypoglossal artery penetrating the hypoglossal canal. (g) Pre-procedure 3D-RA showed that the stenotic part was about 1 mm in diameter.

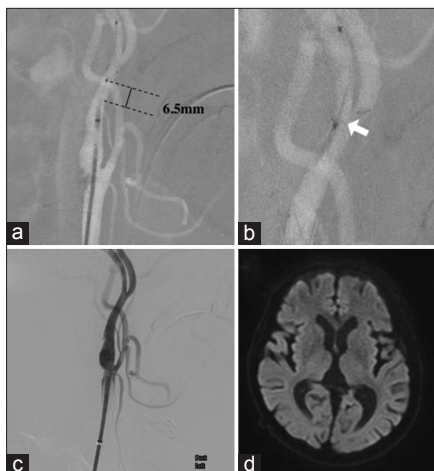


Figure 2: (a-c) Intraprocedure digital subtraction angiography. (a) The distance between the catheter tip of the Spider FX and the stent delivery system before stent placement was 6.5 mm. (b) The catheter tip of the CASPER stent delivery system got stuck in the Spider FX (white arrow), making its retrieval difficult. (c) No intra-stent occlusion, stenosis, or plaque protrusion was observed. (d) Postprocedure magnetic resonance imaging–diffusion-weighted imaging showed no ischemic complications.

CASPER stent delivery system became lodged in the Spider FX, making retrieval difficult [Figure 2b]. The situation was resolved by repeatedly pulling the inner shaft of the CASPER stent delivery system while strongly pushing the delivery wire of the Spider FX. There was no intra-stent occlusion, stenosis, or plaque protrusion immediately after surgery [Figure 2c], and no postoperative ischemic complications were observed [Figure 2d]. The patient was discharged with a modified Rankin scale score of 0.

DISCUSSION

In addition to, persistent primitive trigeminal artery and persistent proatlantal artery, PPHA exists as a residual vascular anastomosis that connects the carotid and vertebral arteries.^[7] The reported incidence of PPHA is 0.02–0.26%, which is a relatively infrequent compared to other forms of residual arterial anastomosis.^[3,11,15]

Our patient had both ICA stenosis and PPHA and since the number of such case reports is small, there is no consensus regarding the appropriate treatment method. Treatment methods for ICA stenosis include carotid endarterectomy (CEA) and CAS.

In the SAPPHERE study, CEA was considered to carry a high risk in patients with heart disease, severe respiratory disease, contralateral carotid artery occlusion, contralateral laryngeal nerve palsy, previous direct neck surgery or neck

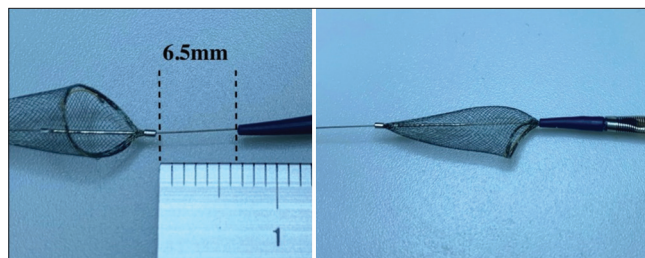


Figure 3: Reproduction of the intraoperative situation. The proximal loop of the Spider FX was stuck in the catheter tip of the CASPER stent delivery system, making it difficult to remove.

radiotherapy, and CEA restenosis with CEA, and CAS is considered to be a treatment option in such cases.^[16] CAS might also be selected for distal ICA lesions due to the difficulty of CEA at this site.^[9] CEA has been previously performed for ICA stenosis with PPHA.^[4] However, this case did not fall into the category of high-risk cases for CEA as defined in the SAPPHERE study, since stenosis was observed at a distal lesion in the ICA, it was predicted that CEA would be difficult, and hence, CAS was performed.

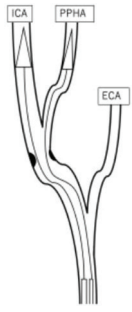

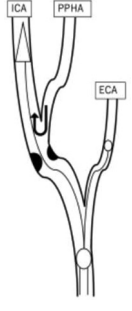
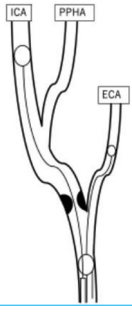
There are 12 reported cases of CAS for ICA stenosis accompanied by PPHA, including ours.^[1,2,5-8,10,12-14,18] However, in each case, the site of stenosis was different, as was the treatment method [Table 1]. When CAS is performed, proximal protection using a balloon guide catheter (BGC) has been used in several cases due to concerns about the dispersal of the plaque from the site of stenosis.^[5-8,12,18] In addition, distal protection using a Carotid GuardWire (Medtronic, Minneapolis, MN, USA) as an EPD to prevent intracranial plaque dispersal is also performed.^[10,12] However, since blood flow from the CCA is blocked during this procedure, it might be difficult in patients with low ischemic tolerance. In the present case, the contralateral vertebral artery was hypoplastic and the posterior circulatory system was predicted to be less resistant to ischemia. Distal filter (DF) type EPDs can be used to maintain intracranial blood flow, and there have been reports of DF type EPDs being used in patients with ICA stenosis and PPHA.^[1,13,14] Silva *et al.*^[13] and Ebiko *et al.*^[1] performed CAS without ischemic complications by implanting a DF type EPD in both the ICA and PPHA. The problem with this technique is that since the EPD is not coaxial with the stent, it might get stuck in between the stent and the vessel wall, making its retrieval difficult.

Tonegawa *et al.*^[14] reported a case in which a DF-type EPD was placed at the bifurcation between the ICA and PPHA, resulting in successful CAS without ischemic complications. This method can be used even in patients with low ischemic tolerance. Using the same method, CAS was successfully performed in our case as well, without ischemic complications [Figure 2d]. However, there were some issues in the present case. During the deployment of

the CASPER stent, the tip of the catheter delivery system gradually moved distally. In addition, the catheter tip gets stuck in the Spider FX, making its removal difficult [Figure 2b]. The distance between the catheter tip and the

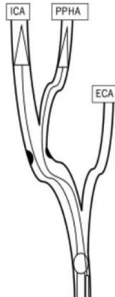
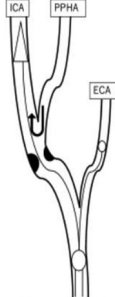


Spider FX was 6.5 mm before implantation [Figure 2a]. When the same system was retrospectively tested outside the vessel after the procedure, the proximal loop of the Spider FX entered the catheter tip, likely representing what

Table 1: Comparison of treatment methods in each report.

No	Authors	Figures	Guiding catheters	EPD	Stents	Remarks	Complications
1	Ebiko <i>et al.</i>		8Fr Optimo (Tokai Medical Products, Aichi, Japan)	FilterWire EZ (Boston Scientific, MA, USA)×2	Carotid WALLSTENT (Boston Scientific, CA, USA)	The common carotid artery was punctured directly under general anesthesia, and 8Fr and 4Fr sheaths were inserted. The surgical strategy was similar to that of cases No. 7 and No. 13.	None
2	Eller <i>et al.</i>		Balloon Guiding catheter (Details unknown)	N/A	Carotid WALLSTENT (Boston Scientific, CA, USA)	Proximal protection was achieved using a balloon-guided catheter, and the procedure was performed after confirming flow reversal.	None
5	Kaku <i>et al.</i>		Optimo (Tokai Medical Products, Aichi, Japan)	Carotid Guard Wire (Medtronic, Minneapolis, MN, USA) + FilterWire EZ (Boston Scientific, MA, USA)	Carotid WALLSTENT (Boston Scientific, CA, USA)	Blood flow to the common and external carotid arteries was blocked. Since contrast administration demonstrated blood flow from the PPHA side to the ICA side, the EPD was guided to the ICA, and the procedure was performed. The surgical strategy was similar to that of case No. 8.	None
6	Kanazawa <i>et al.</i>		9Fr Patlive (Clinical Supply, Gifu, Japan) +5Fr Guider soft-tip (Stryker, Kalamazoo, MN, USA)	Carotid Guard Wire (Medtronic, Minneapolis, MN, USA) to ICA + Hyper Form Occlusion Balloon (Medtronic, MN, USA) to ECA	Carotid WALLSTENT (Boston Scientific, CA, USA)	N/A	None

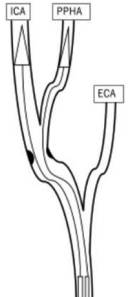
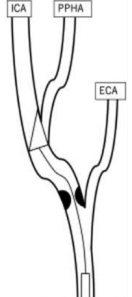
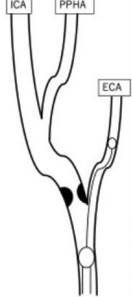
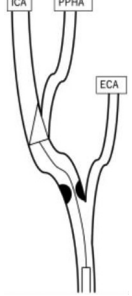
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Table 1: (Continued).

No	Authors	Figures	Guiding catheters	EPD	Stents	Remarks	Complications
7	Mochizuki <i>et al.</i>		8Fr Optimo (Tokai Medical Products, Aichi, Japan) +6Fr FUBUKI (Asahi Intecc, Aichi, Japan)	FilterWire EZ (Boston Scientific, MA, USA)×2	Carotid WALLSTENT (Boston Scientific, CA, USA)	The surgical strategy was similar to that of cases No. 1 and No. 13.	None
8	Murai <i>et al.</i>		MOMA (Medtronic, MN, USA)	FilterWire EZ (Boston Scientific, MA, USA) to ICA	Carotid WALLSTENT (Boston Scientific, CA, USA)	Blood flow to the common and external carotid arteries was blocked. Since contrast administration revealed blood flow from the PPHA side to the ICA side, the EPD was guided to the ICA and the procedure was performed. The surgical strategy was similar to that of case No. 5.	None
10	Nii <i>et al.</i>		Guider softip (Stryker, Kalamazoo, MN, USA)	Carotid Guard Wire (Medtronic, Minneapolis, MN, USA)	Carotid WALLSTENT (Boston Scientific, CA, USA)	The surgical strategy was similar to case No. 12.	None
12	Ryu <i>et al.</i>		9Fr Optimo (Tokai Medical Products, Aichi, Japan)	Carotid Guard Wire (Medtronic, Minneapolis, MN, USA)	Carotid WALLSTENT (Boston Scientific, CA, USA)	The surgical strategy was similar to case No. 10.	None

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Table 1: (Continued).

No	Authors	Figures	Guiding catheters	EPD	Stents	Remarks	Complications
13	Silva <i>et al.</i>		6Fr Envoy (Codman Neurovascular, Miami Lakes, FL, USA) +5Fr Envoy	Emboshield (Abbott Vascular, Santa Clara, CA, USA)×2	Precise (Codis, Johnson& Johnson, Miami, FL, USA)	The surgical strategy was similar to that of cases No. 1 and No. 7.	None
14	Tonegawa <i>et al.</i>		6Fr FUBUKI (Asahi Intecc, Aichi, Japan)	Spider FX (Covidien, Mansfield, MA, USA)	Carotid WALLSTENT (Boston Scientific, CA, USA)	The surgical strategy was similar to that of the present case.	None
18	Zhang <i>et al.</i>		MOMA (Medtronic, MN, USA)	N/A	Carotid WALLSTENT (Boston Scientific, CA, USA)	N/A	None
	Present Case		9Fr Branchor (Asahi Intecc, Aichi, Japan)	Spider FX (Covidien, Mansfield, MA, USA)	CASPER stent (Terumo, Tokyo, Japan)	The surgical strategy was similar to that of case No. 14.	None

The number column lists the corresponding reference numbers. EPD: Embolic protection device, ECA: External carotid artery, N/A: Not applicable, PPHA: Persistent primitive hypoglossal artery, ICA: Internal carotid artery

had occurred inside the vessel [Figure 3]. In this case, the situation was resolved by repeatedly pulling the inner shaft of the CASPER stent while strongly pushing the delivery wire of the Spider FX, although caution should be exercised because surgical intervention might be required in cases of difficult removal.

Conventional carotid artery stents used in CAS include the open-cell PROTÉGÉ (Covidien, Mansfield, MA, USA), Precise (Cordis, Johnson and Johnson, Miami, FL, USA), and the closed-cell Carotid Wallstent (Boston Scientific, Santa Clara, CA, USA). The CASPER stent has a small mesh structure and a double-layer micromesh structure

and has been used in recent years since it has been shown to have fewer embolic complications than conventional stents.^[17]

In the present case, a CASPER stent was used to prevent plaque protrusion and plaque scattering during the procedure, and a Spider FX was used as the DF-type EPD. Consequently, no postoperative ischemic complications were observed following the procedure [Figure 2d]. However, if the distance between the catheter tip of the CASPER stent delivery system and the Spider FX is insufficient, the selection of a stent other than the CASPER stent should be considered. Particular attention should be paid when the distance between the tip of the CASPER stent delivery system and the Spider FX is expected to be <6.5 mm, as in the present case.

CONCLUSION

CASPER stent deployment with Spider FX in the ICA and PPHA bifurcation is considered to be an effective method for treating ICA stenosis associated with PPHA. However, care should be taken to ensure the appropriate selection of EPDs and stents, depending on the location of the stenosis and bifurcation of the PPHA.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The author(s) confirms that they have used Artificial Intelligence (AI)-Assisted Technology for assisting in the writing or editing of the manuscript or image creations.

REFERENCES

1. Ebiko Y, Yoshino Y, Ikota M, Watabe T, Uchiyama T, Soma N, Kusaka G. Carotid artery stenting with persistent primitive hypoglossal artery performed by carotid direct puncture and double distal filter protection: A case report. *Jpn J Stroke* 2021;43:540-5.
2. Eller JL, Jahshan S, Dumont TM, Kan P, Siddiqui AH. Tandem symptomatic internal carotid artery and persistent hypoglossal artery stenosis treated by endovascular stenting and flow reversal. *BMJ Case Rep* 2013;2013:bcr2012010578.
3. Fantini GA, Reilly LM, Stoney RJ. Persistent hypoglossal artery: Diagnostic and therapeutic considerations concerning carotid thromboendarterectomy. *J Vasc Surg* 1994;20:995-9.
4. Grego F, Stramanà R, Lepidi S, Antonello M, Bonvini S, Zaramella M, *et al.* Primitive proatlantal intersegmental artery and carotid endoarterectomy. *J Vasc Surg* 2004;39:691.
5. Kaku S, Nishimura K, Fuga M, Watanabe M, Iwamoto I, Murayama Y. Carotid artery stenting for symptomatic stenosis of the cervical carotid artery associated with persistent primitive hypoglossal artery: A case report. *J Neuroendovasc Ther* 2017;11:474-8.
6. Kanazawa R, Ishihara S, Okawara M, Ishihara H, Kohyama S, Yamane F. A successful treatment with carotid arterial stenting for symptomatic internal carotid artery severe stenosis with ipsilateral persistent primitive hypoglossal artery: Case report and review of the literature. *Minim Invasive Neurosurg* 2008;51:298-302.
7. Mochizuki Y, Higa T, Iwata Y, Kawamata T. 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. *J Neuroendovasc Ther* 2017;11:588-91.
8. Murai S, Kusaka N, Umakoshi M, Itami H, Otsuka S, Nishiura T, *et al.* Stenting for internal carotid artery stenosis associated with persistent primitive hypoglossal artery using proximal flow blockade and distal protection system: A technical case report and literature review. *J Stroke Cerebrovasc Dis* 2016;25:e98-102.
9. Nagata T, Mitsuhashi Y, Kawakami T, Tsuruta S, Inoue T, Ohata K. Selection of surgical procedures to treat carotid artery stenosis. *Surg Cerebral Stroke* 2019;47:266-71.
10. Nii K, Aikawa H, Tsutsumi M, Onizuka M, Nakau H, Inoue R, *et al.* 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. *Neurol Med Chir (Tokyo)* 2010;50:921-4.
11. Ouriel K, Green RM, DeWeese JA. Anomalous carotid-basilar anastomoses in cerebrovascular surgery. *J Vasc Surg* 1988;7:774-7.
12. Ryu B, Ishikawa T, Hashimoto K, Shimizu M, Yagi S, Shimizu T, *et al.* Internal carotid artery stenosis with persistent primitive hypoglossal artery treated with carotid artery stenting: A case report and literature review. *Neuroradiol J* 2016;29:115-21.
13. Silva CF, Hou SY, Kühn AL, Whitten RH, Wakhloo AK. Double embolic protection during carotid artery stenting with persistent hypoglossal artery. *BMJ Case Rep* 2013;2013:bcr2013010709.
14. Tonegawa T, Ohmori Y, Hitoshi Y, Yoshida K. Carotid artery stenting to treat internal carotid artery stenosis with persistent primitive hypoglossal artery: A case report. *Jpn J Stroke* 2020;42:275-9.
15. Vlychou M, Georganas M, Spanomichos G, Kanavaros P, Artinopoulos C, Zavras GM. Angiographic findings and clinical implications of persistent primitive hypoglossal artery. *BMC Med Imaging* 2003;3:2.
16. Yadav JS, Wholey MH, Kuntz RE, Fayad P, Katzen BT, Mishkel GJ, *et al.* Protected carotidartery stenting versus endarterectomy in high-risk patients. *N Engl J Med*

- 2004;351:1493-501.
17. Yamada K, Yoshimura S, Miura M, Kanamaru T, Shindo S, Uchida K, *et al.* Potential of new-generation double-layer micromesh stent for carotid artery stenting in patients with unstable plaque: A preliminary result using OFDI analysis. *World Neurosurg* 2017;105:321-6.
 18. Zhang L, Song G, Chen L, Jiao L, Chen Y, Wang Y. Concomitant asymptomatic internal carotid artery and persistent primitive

hypoglossal artery stenosis treated by endovascular stenting with proximal embolic protection. *J Vasc Surg* 2016;63:237-40.

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