Radiology Case Reports 19 (2024) 146–149

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Ruptured posterior inferior cerebellar artery aneurysm associated with persistent primitive hypoglossal artery: A case report ☆

Hideaki Shigematsu, MD, PhD, Kazuma Yokota, MD, Akihiro Hirayama, MD, PhD, Takatoshi Sorimachi, MD, PhD*

Department of Neurosurgery, Tokai University School of Medicine, 143 Shimokasuya, Isehara, Kanagawa 259-1193, Japan

ARTICLE INFO

Article history:
Received 25 December 2022
Revised 29 September 2023
Accepted 30 September 2023

Keywords:
Transarterial embolization
Persistent primitive hypoglossal artery
Subarachnoid hemorrhage

ABSTRACT

Until now, 9 cases of an association of a posterior inferior cerebellar artery (PICA) aneurysm with the persistent primitive hypoglossal artery (PPHA) have been reported. We reported a case of a ruptured PICA aneurysm associated with the PPHA, which was successfully treated by intravascular embolization using inflation of an endovascular occlusion balloon in the proximal artery to stabilize the microcatheter tip. A 19-year-old woman presenting headache and mild consciousness disturbance was admitted to our hospital. Head computed tomography (CT) showed a subarachnoid hemorrhage in the interpeduncular cistern. Right common carotid angiography revealed an aneurysm with a maximum diameter of 3.7 mm at the proximal PICA, which was fed from the common carotid artery (CCA) through the internal carotid artery (ICA) and the PPHA and the vertebral artery. During coil embolization, a pulsatile fluctuation of the microcatheter tip caused by the minimum curvature of the proximal arterial route from the aorta to the PPHA made the continuation of the coil embolization difficult. Then, we inflated an occlusion balloon in the PPHA to stabilize the microcatheter tip, and the coil embolization was performed under the blank roadmap fluoroscopy. The aneurysm disappeared completely with the preservation of the PICA on the postoperative angiogram. A case of ruptured PICA aneurysm with proximal PPHA, which was successfully embolized with the assist of a proximal occlusion balloon inflation. When the pulsative movement of a microcatheter tip made coil embolization difficult, this technique could be useful.

☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

* Corresponding author:
E-mail address: shige@is.icc.u-tokai.ac.jp (H. Shigematsu).

https://doi.org/10.1016/j.radcr.2023.09.103

Introduction

Persistent primitive hypoglossal artery (PPHA), which is a persistent anastomosis of the internal carotid-vertebrobasilar artery system, is found in 0.027%-0.29% [1,2] of the general population. A reported rate of association of PPHA with vascular malformations or subarachnoid hemorrhage (SAH) was 27% [3]. During cerebral aneurysm embolization, a microcatheter tip is sometimes unstable to conduct the embolization when the arterial route to the aneurysm has minimum curvature. We report a case of successful embolization of a ruptured posterior inferior cerebellar artery (PICA) aneurysm associated with PPHA by using inflation of an endovascular occlusion balloon in the proximal artery to stabilize a fluctuating microcatheter tip due to a minimum curvature route to the aneurysm.

A 19-year-old woman who suffered from headaches and mild consciousness disturbance was admitted to our hospital. Head computed tomography (CT) showed SAH in the interpeduncular cistern (Fig. 1A). A right common carotid angiogram revealed an aneurysm with a maximum diameter of 3.7 mm in the proximal of PICA just distal to the junction with the vertebral artery, which was supplied by the internal carotid artery (ICA) through PPHA (Fig. 1B–F). Embolization of the aneurysm was planned with a single catheter technique using a guiding catheter and a microcatheter (Figs. 2 A and B). A microcatheter (SL-10 microcatheter, Stryker, Kalamazoo, MI) was introduced into the aneurysm through a guiding catheter (6Fr Fubuki 90 cm, Asahi Intecc, Aichi, Japan) located in the common carotid artery (CCA), the ICA and the PPHA. Fluctuation of the microcatheter tip in the aneurysm, which synchronized with the arterial pulsation, led to the failure of continuation of the procedure. To minimize the arterial movement of the microcatheter tip, we inflated an occlusion balloon (Scepter XC 4 mm × 10 cm occlusion balloon, Micro Vention TERUMO, Tustin, CA) in the PPHA, which fixed the microcatheter by compression of the microcatheter against the arterial wall (Fig. 2C). Heparin of 3000 units was administered intravenously, then active clotting time (ACT) was maintained for longer than 250 seconds. After the inflation of the occlusion balloon, the fluctuation of the microcatheter tip was remarkably reduced. Immediately after the introduction of the microcatheter into the aneurysm, the occlusion balloon was inflated, then framing using a coil (Target 360 soft 3 mm × 6 cm, Stryker, Kalamazoo, MI) without detachment was performed. Immediately after the occlusion balloon was deflated, the microcatheter tip became unstable. Then, the balloon was inflated again. To avoid the microcatheter movement for the safer procedure, we performed coil embolization during inflation of the occlusion balloon. While special attention was paid to keeping additional coils within the aneurysm-neck-side initial flame coil under roadmap guide fluoroscope, sufficient embolization using additional 5 coils ([Target 360 soft 3mm×6cm (Stryker), arget 360 ultra 2.5mm×4cm (Stryker), Target 360 nano 2mm×3cm (Stryker), Target 360 nano 1.5mm×2cm (Stryker), Target helical nano 1.5mm×2cm (Stryker]) was able to be performed. The inflation time of the occlusion balloon was 6 minutes and 26 seconds. Postembolization angiography revealed the disappearance of the aneurysm with the preservation of the PICA (Fig. 2D). Mag-

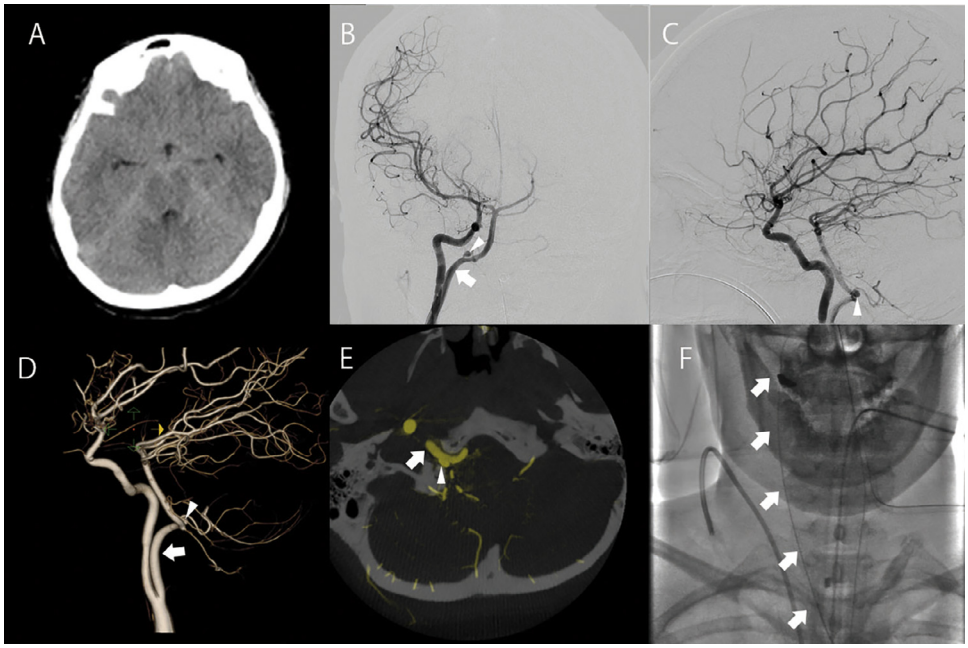

Fig. 1 – (A) A head computer tomogram showing thin subarachnoid hemorrhage in the interpeduncular cistern. (B) A frontal view of right internal carotid angiogram showing PPHA (a white arrow) and an aneurysm (an arrowhead) in the proximal to the PICA. (C) A lateral view of the right internal carotid angiogram. (D) A three-dimensional digital subtraction angiogram of right internal carotid artery (lateral view). (E) The maximum intensity projection axial image of a right internal carotid angiogram. (F) A image of the roentgen showing a guidewire through the CCA, ICA, and PPHA, which indicates minimum curvature of the artery. The white arrows indicate the guidewire. The CCA to the PPHA is almost straight.

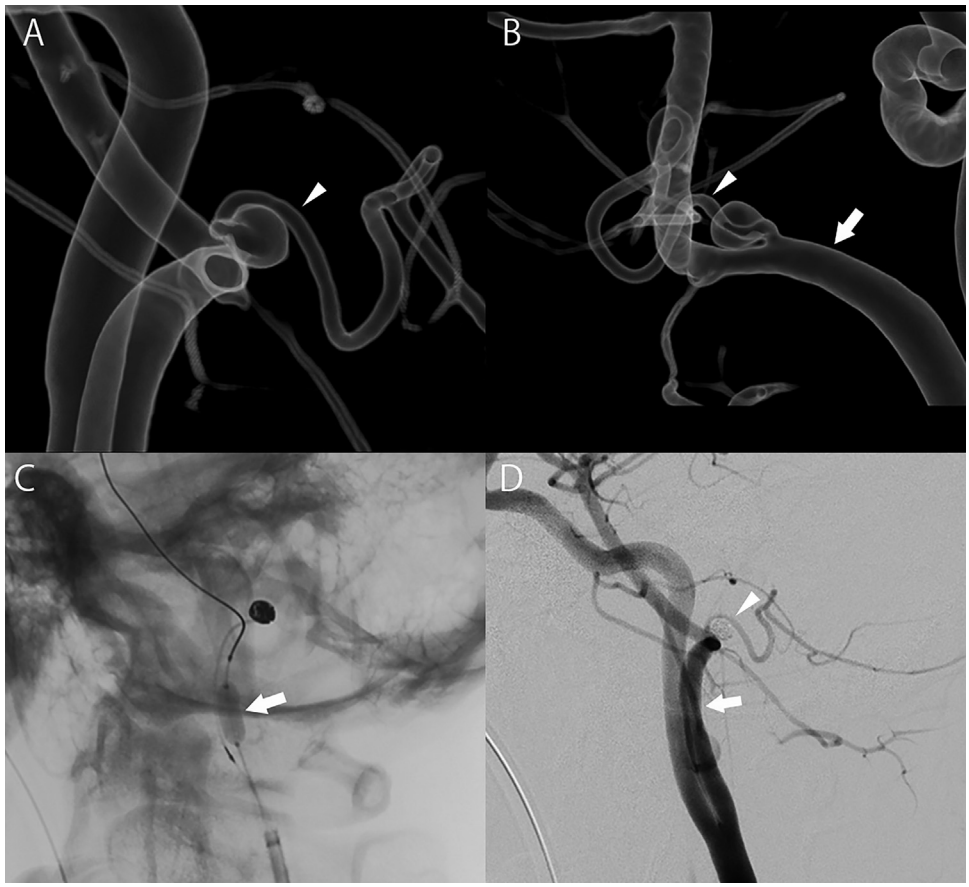

Fig. 2 – (A/B) A 3-dimensional digital subtraction angiogram of right common carotid artery (Working projection) An aneurysm with a maximum diameter of 3.7 mm is observed proximal to the PICA (white arrowhead). The white arrow indicates PPHA. (C) An image of the roentgen showing embolization is performed with the microcatheter stabilized while inflation of occlusion balloon in the proximal PPHA to stabilizing the microcatheter. (D) A postoperative right common carotid angiogram (lateral view) showing the disappearance of the aneurysm. The PICA (a white arrowhead) is well delineated. The white arrow indicates PPHA.

netic resonance (MR) imaging showed no cerebral infarction. After the operation, she completely recovered, and her postoperative course was uneventful. A follow-up MR angiogram 4-years after treatment showed no recurrence of the aneurysm.

Discussion

As for aneurysms associated with PPHA, 53% of the reported aneurysm was located in the posterior circulation, and the PPHA-basilar artery (31.4%) was the most common location [4]. Ten cases of ruptured PICA aneurysms associated with PPHA have been reported, including the present case (Table 1) [5–13]. Five males and 5 females with a mean age of 28.7±14. years (mean± standard deviation[SD]). The mean maximum diameter of the aneurysms was 5.9±2.9 mm (mean± SD). The aneurysms were treated by clipping in 5 patients, coiling in 3, both clipping and coiling in one, the remaining one underwent conservative treatment because of her poor clinical condition.

A mechanism of the pulsative movement of the microcatheter tip in the present patient was suspected as the followings: Arterial pulsation was directly transmitted from the aorta to the PPHA because of the minimum curvature of the approach route from the aorta through the CCA and the ICA to the PPHA due to anatomical characteristics of the PPHA and the patient's young age; A small resistance between the arterial wall and the microcatheter due to the straightness of the approach route also caused the movement of the microcatheter. Usually, during embolization of cerebral aneurysms, the guiding catheter and microcatheter are stabilized by the resistance between the arterial wall at the bending sites and the catheter. A case of a ruptured PICA aneurysm associated with PPHA, in which endovascular embolization was terminated before completion because of the pulsative movement of the microcatheter tip, has been reported. The patient underwent clipping of the aneurysm in the chronic period [7]. Using balloon-guiding catheters to control the blood flow during coil embolization is a well-none procedure [14]. To the best of our knowledge, this case report is the first to document inflation of an endovascular occlusion balloon in a proximal artery to stabilize a microcatheter during coil embolization. The use of an inflated endovascular occlusion balloon in a proximal artery led to blood flow control as well as microcatheter stabilization by direct compression of the microcatheter against the arterial

Table 1 – Cases of persistent primitive hypoglossal artery - posterior inferior cerebellar artery ruptured aneurysm.

Author	Age/sex	WFNS grade	Aneurysm size	Premature rupture	Treatment	mRS at discharge
Tsugu et al. [6]	42/F	I	4 mm	No	clipping	1
Yamamoto et al. [9]	33/M	III	8 mm	No	clipping	6
Duffill et al. [13]	10/M	II	10 mm	Yes	clipping	0
Huynh-Le et al. [10]	47/M	I	n.a	No	clipping	0
Kobayashi et al. [12]	49/F	I	n.a	No	clipping	0
De Blasi et al. [5]	22/M	I	8.5 mm	No	coiling	n.a
Ohta, et al. [11]	42/F	V	5.7 mm	No	coiling	0
Varvari et al. [8]	15/F	V	n.a		-	6
Ishi et al. [7]	38/F	I	1.8 mm	Yes	clipping coiling	2
Present case	19/M	II	3.7 mm	No	coiling	0

WFNS, World Federation of Neurosurgical Societies; mRS, modified Rankin Scale.

wall. The difficulty of confirmation of the aneurysmal configuration and the parent artery using contrast medium injection under fluoroscopy is a major problem of this technique. A precise first flaming coil placement around an aneurysmal neck is key to overcome this shortcoming. Then the following coils should be placed within the flaming coil around the aneurysmal neck under blank load map fluoroscopy to preserve the parent artery patency.

Conclusion

We report a case of a ruptured PICA aneurysm associated with PPHA. In case of pulsative fluctuation of a microcatheter tip due to minimum curvature of the proximal arterial route, inflation of an occlusion balloon in the proximal artery could lead to stabilizing the microcatheter tip by both compressions of the microcatheter against the arterial wall and arterial flow control.

Patient consent

Informed consent was obtained from the family of the patient described in this report. The patient was a minor at admission, therefore, informed consent was obtained from her family.

REFERENCES

[1] Brismar J. Persistent hypoglossal artery, diagnostic criteria. Report of a case. Acta Radiol Diagn (Stockh) 1976;17(2):160–6. doi:10.1177/028418517601700204.

[2] Uchino A, Saito N, Okada Y, Kozawa E, Nishi N, Mizukoshi W, et al. Persistent hypoglossal artery and its variants diagnosed by CT and MR angiography. Neuroradiology 2013;55(1):17–23. doi:10.1007/s00234-012-1074-0.

[3] Agnoli AL. Vascular anomalies and subarachnoid haemorrhage associated with persisting embryonic vessels. Acta Neurochir (Wien) 1982;60(3-4):183–99. doi:10.1007/bf01406306.

[4] Kanematsu M, Satoh K, Nakajima N, Hamazaki F, Nagahiro S. Ruptured aneurysm arising from a basilar artery fenestration and associated with a persistent primitive hypoglossal artery. Journal of Neurosurgery 2004;101(3):532–5.

[5] De Blasi R, Medicamento N, Chiumarullo L, Salvati A, Maghenzani M, Dicuonzo F, et al. A case of aneurysm on a persistent hypoglossal artery treated by endovascular coiling. Interv Neuroradiol 2009;15(2):175–8. doi:10.1177/159101990901500206.

[6] Tsugu A, Matsumae M, Ikeda A, Sato O. [A case report: persistent primitive hypoglossal artery aneurysm]. No Shinkei Geka 1990;18(1):95–100.

[7] Ishii K, Naito T, Kato T, TANEI T, TSUKAMOTO E, OKADA K, et al. Coil Embolization for Recurrent Aneurysm Associated with a Persistent Primitive Hypoglossal Artery after Clipping Surgery. Noukekannaichiryou 2020;5(3):143–8.

[8] Varvari I, Bos EM, Dinkelaar W, van Es AC, Can A, Hunfeld M, et al. Fatal Subarachnoid Hemorrhage from an Aneurysm of a Persistent Primitive Hypoglossal Artery: Case Series and Literature Overview. World Neurosurg 2018;117:285–91. doi:10.1016/j.wneu.2018.06.119.

[9] Yamamoto S, Sunada I, Matsuoka Y, Hakuba A, Nishimura S. Persistent primitive hypoglossal artery aneurysms–report of two cases. Neurol Med Chir (Tokyo) 1991;31(4):199–202. doi:10.2176/nmc.31.199.

[10] Huynh-Le P, Matsushima T, Muratani H, Hikita T, Hirokawa E. Persistent primitive hypoglossal artery associated with proximal posterior inferior cerebellar artery aneurysm. Surg Neurol 2004;62(6):546–51 discussion 51. doi:10.1016/j.surneu.2004.03.018.

[11] Ohta T, Hayashi H, Takahashi Y, Murao K. Coil embolization of a raptured aneurysm of the posterior inferior cerebellar artery associated with primitive hypoglossal artery. JNET: Journal of Neuroendovascular Therapy 2012;6(2):127–32.

[12] Kobayashi M, Akaji K, Tanizaki Y, Mihara B, Ohira T, Kawase T. Posterior inferior cerebellar artery aneurysm associated with persistent primitive hypoglossal artery. Neurol Med Chir (Tokyo) 2008;48(6):259–61. doi:10.2176/nmc.48.259.

[13] Duffill J, Lang DA, Dwyer GN. Subarachnoid haemorrhage in a child from an aneurysm of a persistent primitive hypoglossal artery. Br J Neurosurg 1996;10(6):607–10. doi:10.1080/02688699646952.

[14] Ohshima T, Miyachi S, Matsuo N, Kawaguchi R, Niwa A, Maejima R, et al. Efficacy of the proximal balloon flow control method for endovascular coil embolisation as a novel adjunctive technique: A retrospective analysis. Interv Neuroradiol 2018;24(4):375–8. doi:10.1177/1591019918763614.