



Endovascular treatment for unruptured aneurysm associated with persistent primitive trigeminal artery: a case report and literature review

Satoshi Murai¹ · Kenji Sugiu¹ · Tomohito Hishikawa¹ · Masafumi Hiramatsu¹ · Shingo Nishihira¹ · Naoya Kidani¹ · Yu Takahashi¹ · Isao Date¹

Received: 15 August 2018 / Accepted: 11 December 2018 / Published online: 19 December 2018
© Springer-Verlag GmbH Austria, part of Springer Nature 2018

Abstract

The persistent primitive trigeminal artery (PPTA) is the most common carotid-basilar anastomosis, and the incidence of cerebral aneurysms associated with the PPTA is approximately 4%. Since PPTA aneurysms often have a wide neck and other vascular anomalies, endovascular treatment using an adjunctive technique is the current first-line therapy. Here, we report a case of PPTA aneurysm treated by coil embolization with a stent-assisted technique. A detailed evaluation of the size and course of all vessels and collateral flow, including the Allcock test and balloon test occlusion, is necessary when deciding on the treatment strategy.

Keywords Persistent primitive trigeminal artery · Cerebral aneurysm · Coil embolization · Stent-assisted technique · Balloon test occlusion

Introduction

The persistent primitive trigeminal artery (PPTA) is the most common carotid-basilar anastomosis, and its prevalence has been reported to be between 0.1 and 0.6% [4]. A recent study reported that the incidence of cerebral aneurysms associated with PPTA was approximately 4%, similar to that of patients in the general population [3, 12]. Although endovascular treatment is currently the first-line therapy for PPTA aneurysms, these aneurysms usually have a wide neck and are often concomitant with other vascular anomalies [7, 14]. Therefore, a detailed assessment of their anatomy is necessary, including a preoperative Allcock test and balloon test occlusion (BTO). Here, we report a case of PPTA aneurysm successfully treated with stent-assisted coil embolization, and provide a literature review of endovascular treatment for PPTA aneurysms.

Case presentation

A 68-year-old woman with a 1-year history of left abducens nerve palsy was admitted to our institute for treatment of a large aneurysm in the left internal carotid artery (ICA)-PPTA bifurcation (Fig. 1a, b). The aneurysm measured 12.7 mm × 8.6 mm, and the diameter of its neck was 6.8 mm. The PPTA was classified as lateral type and Saltzman type 1. An Allcock test in the preoperative angiography revealed that although the basilar artery (BA) was hypoplastic, there was anterograde flow in the BA (Fig. 1c). Because the aneurysm had a wide neck and its neck completely rode on the PPTA, we decided on coil embolization with a stent-assisted technique (SAT) and double-catheter technique. One week before the procedure, 100 mg of aspirin and 75 mg of clopidogrel were administered to the patient. Under general anesthesia, a 7-Fr Roadmaster guiding catheter (Goodman, Aichi, Japan) was placed in the right ICA. An Excelsior XT-27 microcatheter (Stryker Neurovascular, Fremont, CA, USA) was advanced to the left P1 segment, and a Headway-17 microcatheter (MicroVention TERUMO, Tustin, CA, USA) was placed in the aneurysm. Then, a Neuroform 4.5 mm × 30 mm (Stryker Neurovascular, Fremont, CA, USA) was deployed from the PPTA to the ICA to preserve the PPTA (Fig. 1d). An Excelsior SL-10 microcatheter 45° (Stryker Neurovascular, Fremont, CA, USA) was placed in the aneurysm through the stent struts.

This article is part of the Topical Collection on *Vascular Neurosurgery - Aneurysm*

✉ Kenji Sugiu
ksugiu@md.okayama-u.ac.jp

¹ Department of Neurological Surgery, Okayama University Graduate School of Medicine, Dentistry and Pharmaceutical Sciences, 2-5-1 Shikata-cho, Okayama 700-8558, Japan

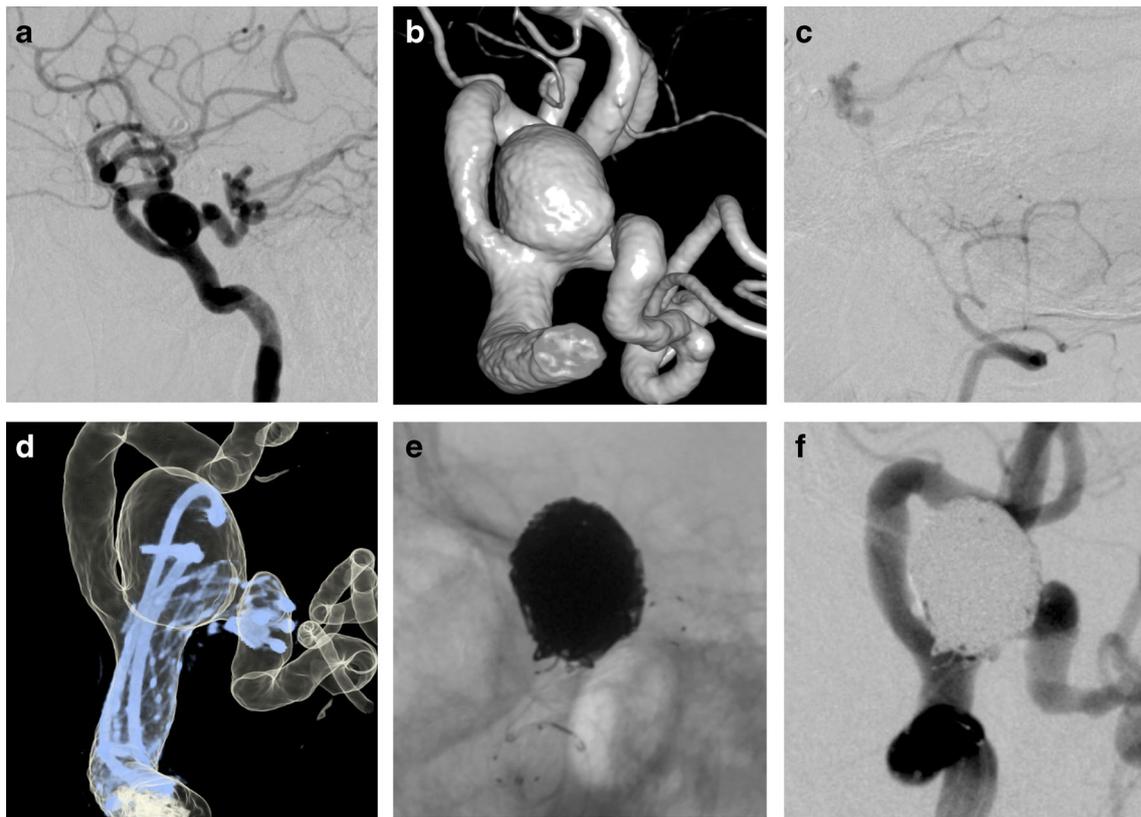


Fig. 1 Cerebral angiography and 3D digital subtraction angiography (DSA) show an internal carotid artery (ICA)-persistent primitive trigeminal artery (PPTA) bifurcation aneurysm with a broad neck, completely riding on the PPTA (a, b). An Allcock test shows antegrade flow through the hypoplastic basilar artery (BA) (c). After

a microcatheter is placed in the aneurysm, a 4.5 mm × 30 mm Neuroform is deployed to cover the neck. Next, another microcatheter is placed in the aneurysm through the stent struts (d). Using the double-catheter technique, coil embolization is performed and results in almost complete obliteration (e, f)

Using the double-catheter technique, coil embolization was performed mainly using HydroFrame and HydroSoft coils (MicroVention Terumo, Tustin, CA, USA), and the treatment resulted in almost complete obliteration (Fig. 1e, f). Although the abducens nerve palsy was not improved, there was no recurrence 1 year after the procedure.

Discussion

This is the first reported case of stent-assisted coil embolization in which a stent was deployed in the PPTA to preserve the parent artery. Unlike other bifurcation-type aneurysms, PPTA aneurysms have several anatomical features, and a detailed assessment is thus necessary before treatment. The PPTA is divided into two groups, the medial and lateral types. In the medial type, the PPTA runs superior to the abducens nerve and pierces the dura toward the BA. In the lateral type, the PPTA runs inferolaterally to the abducens nerve and pierces the dura just medial to the trigeminal nerve. A large aneurysm may cause a mass effect in the cavernous sinus, such as

oculomotor and abducens nerve palsy [8]. Moreover, in the lateral type, the PPTA branches off a perforating artery to the pons, and it may therefore be possible for occlusion of the PPTA to cause an ischemic lesion in the brainstem [5, 10]. In our case, the PPTA was the lateral type and the patient presented with abducens nerve palsy. The abducens nerve palsy might have persisted after treatment because the aneurysm projection was superolateral, and the mass effect with coils to the abducens nerve persisted for a long time.

Saltzman's classification should also be considered for endovascular treatment. In Saltzman's type 1, the proximal BA and the posterior communicating artery (PcomA) may be hypoplastic. As the blood flow from the ICA to the PPTA is the main supply for the brainstem and cerebellar cortex, sacrificing the PPTA in endovascular treatment is not recommended. In Saltzman's type 2, the posterior cerebral artery receives blood flow from the PcomAs, and the BA receives antegrade flow through the VAs [8]. Internal trapping that includes the PPTA may be acceptable, although there remains a risk of brainstem infarction. In our case, the PPTA was

classified as Saltzman's type 1, and the BA was hypoplastic, so preservation of the PPTA was taken into consideration when choosing treatment.

Onizuka et al. reported that preoperative BTO is important when choosing the optimal methods for coil embolization [11]. The advantages of BTO are that it enables evaluation of whether or not (1) the BA is supplied by the antero- grade flow through the VAs, (2) there is enough collateral flow via the AcomA or the external carotid artery during ICA occlusion, and (3) if neck plasty under the balloon remodeling technique (BRT) is available. As described above, if there is no antero- grade flow in the BA, PPTA occlusion can cause brainstem infarction, resulting in severe neurological deterioration. Instead of BTO, the Matas and Allcock tests are also useful, as was true in our case.

Table 1 presents a summary of coil embolization for PPTA aneurysms since 2000 combined with our cases (the first case is shown as case 1, and the second case is not shown here) [1, 2, 5–7, 9, 11, 13, 15, 16]. Of these, seven (58%) cases required an adjunctive technique, such as BRT or SAT, because they include relatively wide-necked aneurysms (median 4.5 mm). PPTA occlusion was performed in three cases, all of which were Saltzman's type 2. Complete obliteration was achieved in eight (67%) cases immediately after coil embolization, and no periprocedural complications were observed. Cranial nerve symptoms were improved in all cases except ours.

We propose an endovascular treatment strategy for PPTA aneurysm based on the results of the Allcock test or BTO. In cases in which the posterior circulation mainly depends on the blood flow through the PPTA and there is no sufficient antero- grade flow in the BA during ICA occlusion, coil embolization with the BRT or SAT is recommended to preserve the PPTA (Fig. 2a). In contrast, in cases in which the Allcock test or BTO demonstrates that there is sufficient antero- grade flow in the BA through the VA and in which preservation of the PPTA is also thought to be difficult, internal trapping of the PPTA is a treatment option (Fig. 2b). There remains the risk of brainstem infarction, however, due to the occlusion of branches for the pons, especially in the lateral type. For treatment of wide-necked aneurysm, we initially chose the BRT, especially in ruptured cases because stent placement requires long-term dual antiplatelet therapy, which increases the risk of hemorrhagic complications. The SAT is favorable, however, in cases in which the aneurysmal neck completely rides on the PPTA (as in our case) and in cases where neck plasty with an occlusion balloon are thought to be difficult. Flow diverter placement is a potential alternative strategy. It can reduce the mass effect immediately, which can decompress cranial nerves earlier in cases with cranial nerve palsy.

Table 1 PPTA aneurysms treated with coil embolization since 2000

Author, year	Age, sex	Symptom	Location	Saltzman's type	Evaluation of collaterals	Maximum size (mm)	Neck diameter (mm)	Treatment	Results	Complications	Follow-up	Clinical outcome
Mohammed, 2002	58, F	SAH	ICA-PPTA	2	Not described	5	4.5	SAT	CO	None	CO, 9M	–
Li, 2004	51, F	Epistaxis	ICA-PPTA	2	Not described	Not described	Not described	PPTA occlusion	DF	None	Not described	Improved
Zhao, 2005	28, F	Headache	ICA-PPTA	2	Not described	3.2	4	SAT	DF	None	CO, 6M	Improved
Onizuka, 2006	17, F	CN6 palsy	ICA-PPTA	2	BTO	Not described	Not described	IT	CO	None	Not described	Improved
Sherkat, 2009	64, F	SAH	ICA-PPTA	2	Not described	8	Not described	Simple	DF	None	DF, 4W	–
Aguiar, 2011	53, F	SAH	BA-PPTA	1	Not described	4	4	BRT	CO	None	Not described	–
Kai, 2011	71, F	SAH	PPTA trunk	1	Allcock test	7	4.8	BRT	CO	None	Not described	–
Yoshida, 2011	60, F	CCF, CN6 palsy	PPTA trunk	1	Allcock test	6	Narrow	Simple	CO	None	CO, 6M	Improved
Ladner, 2013	66, F	TN	PPTA trunk	2	Not described	7	2.8	Simple	CO	None	CO, 6M	Improved
Ajeet, 2016	44, M	Headache	trunk	2	Not described	9	Not described	Dual SAT, IT	CO	None	CO, 6M	Improved

Table 1 (continued)

Author, year	Age, sex	Symptom	Location	Saltzman's type	Evaluation of collaterals	Maximum size (mm)	Neck diameter (mm)	Treatment	Results	Complications	Follow-up	Clinical outcome
			PPTA trunk									
Our case 1	68, F	CN6 palsy	ICA-PPTA 1	1	Allcock test	12.7	5.4	SAT	NR	None	CO, 1Y	Not improved
Our case 2	79, F	CN6 palsy	ICA-PPTA 1	1	BTO	10.1	4.5	BRT	CO	None	CO, 3M	Improved

BRT balloon remodeling technique, *BTO* balloon test occlusion, *CN* cranial nerve, *CO* complete occlusion, *DF* dome filling, *ICA* internal carotid artery, *IT* internal trapping, *NR* neck remnant, *PPTA* persistent primitive trigeminal artery, *SAT* stent-assisted technique, *SAH* subarachnoid hemorrhage, *TV* trigeminal neuralgia

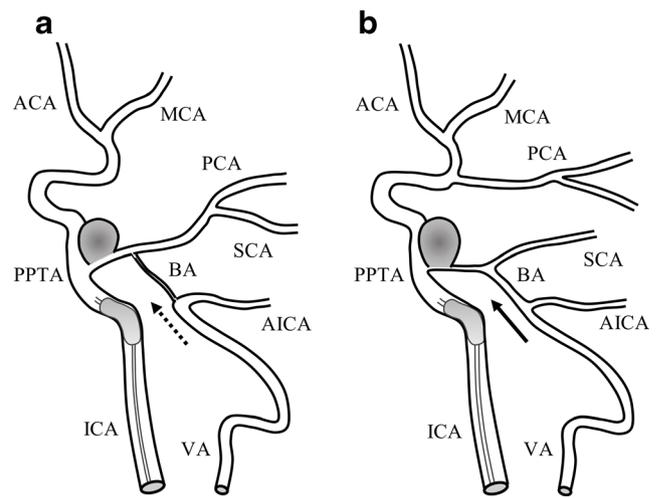


Fig. 2 In Saltzman's type 1, the posterior circulation mainly depends on the blood flow through the PPTA, and there is no sufficient flow in the BA during ICA occlusion (dotted arrow: insufficient flow) (a). In Saltzman's type 2, the Allcock test or BTO demonstrates that there is sufficient antegrade flow in the BA through the VA (black arrow: sufficient flow) (b)

Conclusion

We reported our experience and treatment strategy of coil embolization for PPTA aneurysms. These aneurysms include anatomical features, and a detailed evaluation of the size and course of all vessels and collateral flows, including the Allcock test and BTO, is thus needed when choosing the optimal treatment strategy.

Compliance with ethical standards

All patients consented to publication of this case report.

Conflict of interest The authors declare that they have no conflicts of interest.

References

1. Aguiar GB, Conti ML, Veiga JC, Jory M, Souza RB (2011) Basilar artery aneurysm at a persistent trigeminal artery junction. A case report and literature review. *Interv Neuroradiol* 17:343–346
2. Ajeet G, John D (2016) Dual stent-assisted coil embolization for fusiform aneurysm arising from persistent trigeminal artery. *Neurointervention* 11:131–134
3. Cloft HJ, Razack N, Kallmes DF (1999) Prevalence of cerebral aneurysms in patients with persistent primitive trigeminal artery. *J Neurosurg* 90:865–867
4. George AE, Lin JP, Morantz RA (1971) Intracranial aneurysm on a persistent primitive trigeminal artery. Case report. *J Neurosurg* 35: 601–604
5. Kai Y, Ohmori Y, Watanabe M, Morioka M, Hirano T, Kawano T, Sakurama T, Miura A, Kuratsu J (2011) Coil embolization of an aneurysm located at the trunk of the persistent primitive trigeminal artery. *Neurol Med Chir (Tokyo)* 51:361–364

6. Ladner TR, Ehtesham M, Davis BJ, Khan IS, Ghiassi M, Ghiassi M, Singer RJ (2014) Resolution of trigeminal neuralgia by coil embolization of a persistent primitive trigeminal artery aneurysm. *J Neurointerv Surg* 6:e22
7. Li MH, Li WB, Pan YP, Fang C, Wang W (2004) Persistent primitive trigeminal artery associated with aneurysm: report of two cases and review of the literature. *Acta Radiol* 45:664–668
8. Meckel S, Spittau B, McAuliffe W (2013) The persistent trigeminal artery: development, imaging anatomy, variants, and associated vascular pathologies. *Neuroradiology* 55:5–16
9. Mohammed M, Sandhu JS, Wakhloo AK (2002) Stent-assisted coil placement in a wide-necked persistent trigeminal artery aneurysm with jailing of the trigeminal artery: a case report. *AJNR Am J Neuroradiol* 23:437–441
10. Ohshiro S, Inoue T, Hamada Y, Matsuno H (1993) Branches of the persistent primitive trigeminal artery—an autopsy case. *Neurosurgery* 32:144–148
11. Onizuka M, Kazekawa K, Tsutsumi M, Kodama T, Aikawa H, Ikou M, Tomokiyo M, Matsubara S, Nii K, Tanaka A (2006) Hyperform remodeling balloon for the balloon occlusion test of persistent primitive trigeminal artery aneurysm—case report. *Neurol Med Chir (Tokyo)* 46:541–543
12. O'uchi E, O'uchi T (2010) Persistent primitive trigeminal arteries (PTA) and its variant (PTAV): analysis of 103 cases detected in 16,415 cases of MRA over 3 years. *Neuroradiology* 52:1111–1119
13. Sherkat S, Gazzeri R, Pantoli D, Fiume D, Tancredi A, Gazzeri G (2008) Endovascular treatment of primitive persistent trigeminal artery aneurysm associated with middle cerebral artery aneurysm. *Minim Invasive Neurosurg* 51:218–221
14. Vasović L, Jovanović I, Ugrešević S, Vlajković S, Jovanović P, Stojanović V (2012) Trigeminal artery: a review of normal and pathological features. *Childs Nerv Syst* 28:33–46
15. Yoshida M, Ezura M, Mino M (2011) Carotid-cavernous fistula caused by rupture of persistent primitive trigeminal artery trunk aneurysm—case report. *Neurol Med Chir (Tokyo)* 51:507–511
16. Zhao QP, Li TL, Duan CZ, Chen GZ (2005) Combined neuroform intracranial stent and bioactive matrix detachable coil for embolization of a broad-necked persistent primitive trigeminal artery aneurysm. A case report. *Interv Neuroradiol* 11:63–68