



Case Report

# Coil embolization of subarachnoid hemorrhage with ruptured persistent primitive olfactory artery aneurysm

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## ABSTRACT

**Background:** Persistent primitive olfactory artery (PPOA) is a rare anomaly of the anterior cerebral artery. We experienced a rare case of subarachnoid hemorrhage caused by a ruptured saccular aneurysm of PPOA.

**Case Description:** A 72-year-old man was transported to our hospital with sudden headache. On examination, World Federation of Neurological Surgeons scale was Grade I, and computed tomography of the head showed subarachnoid hemorrhage in Fisher Group 3. Cerebral angiography showed left PPOA and a 4-mm saccular aneurysm at the hairpin turn. No other abnormalities causing bleeding were observed. Based on these findings, subarachnoid hemorrhage due to a ruptured PPOA aneurysm was diagnosed. As the patient had a ventilatory defect due to emphysema, direct approach to the lesion would have been difficult and an endovascular surgery was performed. Three coils were inserted into the aneurysm, and complete occlusion was achieved. Cerebral vasospasm was not observed, and the patient was discharged 1 month after surgery without any neurologic deficit.

**Conclusion:** Most aneurysms of the PPOA are formed at the hairpin turn, as observed in our patient; therefore, a hemodynamic mechanism may be involved in the etiology. To the best of our knowledge, there is no report on treatment using intra-aneurysmal coil embolization, indicating that ours was the first case. As the long-term outcome of intra-aneurysmal coil embolization for PPOA aneurysm is unknown, careful follow-up will be necessary in the future.

**Keywords:** Intra-aneurysmal coil embolization, Persistent primitive olfactory artery, Subarachnoid hemorrhage

## INTRODUCTION

Persistent primitive olfactory artery (PPOA) is a rare anomaly of the anterior cerebral arteries.<sup>[1]</sup> Three types PPOA have been reported:<sup>[3,6]</sup> PPOA Type 1 branches from the internal carotid artery terminal, heads toward the olfactory bulb, runs anteriorly along the olfactory track, sharply turns in the rear upper direction, and connects to the distal part of the anterior cerebral artery; PPOA Type 2 branches from the anterior cerebral artery as the orbitofrontal artery and becomes the ethmoidal artery after coursing through the cribriform plate; and PPOA Type 3 is the transition type of PPOA Type 1 and PPOA Type 2. Here, we report a rare case of subarachnoid hemorrhage caused by a ruptured saccular aneurysm of PPOA Type 1.

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## CASE DESCRIPTION

A 72-year-old man presented to our department with a sudden headache. Neurological examination on admission revealed a Glasgow coma scale score of 15 (E4V4M6). No neurological deficit was observed, and the World Federation of Neurological Surgeons score was Grade I. Computed tomography of the head revealed subarachnoid hemorrhage, classified as Fisher Group 3. Emergency cerebral angiography of the left internal carotid artery revealed that the left anterior cerebral artery coursed anteromedially and downward after division from the internal carotid artery, turned in an extreme acute angle, headed upwards, and ran posteriorly over the corpus callosum as a pericallosal artery. The anterior communicating artery was not visualized, and the Heubner artery branched from the A1 segment. Based on this characteristic course, we suspected it to be a PPOA [Figure 1a]. A 4-mm saccular aneurysm was found at the inward corner of the hairpin turn of the PPOA. No other abnormalities causing bleeding were observed [Figure 1b].

Based on the findings, we diagnosed the condition as subarachnoid hemorrhage due to a ruptured aneurysm of PPOA. The patient had emphysema before admission, and ventilatory defect developed after administration of general anesthesia. Therefore, a direct open approach by craniotomy was considered difficult, and endovascular surgery was performed.

A 7 Fr Launcher (Medtronic, Minneapolis, MN, USA) was placed in the left internal carotid artery from the right femoral artery, while maintaining the activated clotting time >200 s under administration of systemic heparin, and 4.2 Fr ASAHI FUBUKI (ASAHI INTEC, Aichi) was guided to a C2 portion of the internal carotid artery as an intermediate catheter. From there, the Excelsior SL-10 reshaped J microcatheter (Stryker, Kalamazoo, MI, USA) was guided into the aneurysm with Transend EX platinum (Stryker, Kalamazoo, MI, USA). A cage was formed with Axium

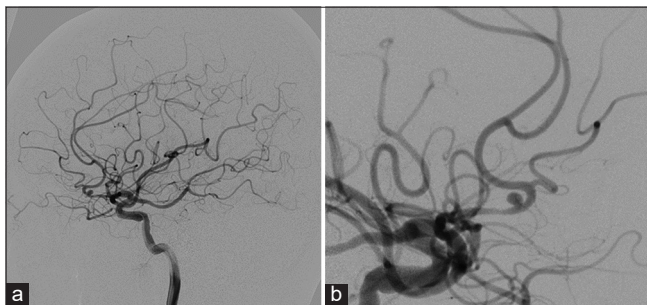
PRIME 3D (Medtronic, Minneapolis, MN, USA). Three coils were inserted into the aneurysm, and nearly complete occlusion was achieved [Figure 2a and b].

Postoperatively, the patient showed no symptomatic cerebral vasospasm and was discharged from the hospital 1 month after surgery without neurologic deficit. Cerebral angiography before discharge showed favorable embolization of the aneurysm.

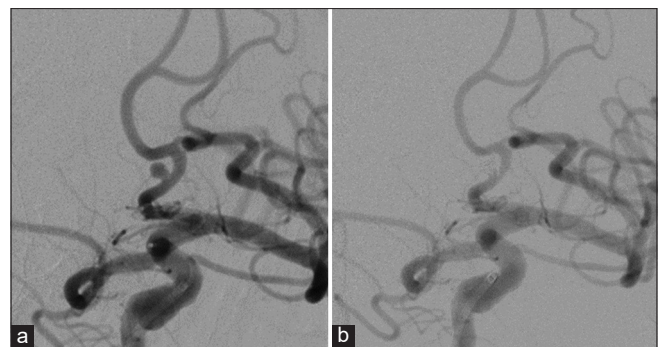
## DISCUSSION

Intracranial carotid arteries are embryologically classified into cranial and caudal divisions.<sup>[8]</sup> At 5 weeks of gestation, the cranial division becomes the primitive olfactory artery (POA), which is divided into the medial and lateral olfactory arteries. The POA runs in the cranial and ventral direction around the optic vesicle and supplies the olfactory area, nasal fossa, and nasal cavity.<sup>[8]</sup> The medial olfactory artery is differentiated into the anterior cerebral artery, while the lateral olfactory artery is differentiated into the lateral striate artery and anterior choroidal artery. Thereafter, the POA gradually regresses and becomes the Heubner recurrent artery. The proximal part of the anterior cerebral artery that runs the course of the POA without regression is called PPOA.<sup>[8]</sup> The frequency of PPOA is reported to be 0.14%.<sup>[12]</sup>

PPOA Type 1 branches from the internal carotid artery terminal, runs toward the olfactory bulb, runs anteriorly along the olfactory tract, sharply turns in the rear upper direction (hairpin turn), and is connected to the distal part of the anterior cerebral artery. PPOA Type 2 branches from the anterior cerebral artery as the orbitofrontal artery, runs into the nasal cavity through the cribriform plate, and becomes the ethmoidal artery.<sup>[4,6]</sup> PPOA Type 1 further has two patterns: one flowing in a wide area of the peripheral anterior cerebral artery and the other with a relatively localized supply to the frontal pole. In addition, Type 3 (PPOA is partially connected with the anterior cerebral artery), which is a hybrid between Type 1 and Type 2, has been reported.<sup>[3]</sup> In our patient, the



**Figure 1:** Preoperative left internal carotid angiograms showing anomalous arteries arising from the internal carotid arteries, which and run anteromedially along the frontal base and making a hairpin-pin bend, and a saccular aneurysm is seen on the hairpin-pin bend, lateral view (a) and oblique view (b).



**Figure 2:** (a) Preoperative angiograms revealing an aneurysm with oblique view (b) postoperative angiograms showing no filling of the aneurysm.

**Table 1:** Reported cases of persistent primitive olfactory artery aneurysm.

Author	Age	Sex	Aneurysm type	PPOA type	Ruptured/unruptured	Size	Treatment	Complication	Year
Yamaura <i>et al.</i>	59	F	Saccular	Type 1	Ruptured	7 mm	Clipping	Hyposmia	1979
Hanakita <i>et al.</i>	72	M	Saccular	Type 1	Unruptured	ND	Clipping	None	1988
Tsuji <i>et al.</i>	59	F	Saccular	Type 1	Ruptured	ND	Clipping	Anosmia	1995
Nozaki <i>et al.</i>	54	F	Saccular	Type 1	Unruptured	ND	Clipping	None	1998
Kaneko <i>et al.</i>	46	M	Fusiform	Type 1	Unruptured	ND	Trapping	Anosmia	1999
Yamamoto <i>et al.</i>	69	M	Saccular	Type 1	Unruptured	13.3 mm	Clipping	None	2009
Uchino <i>et al.</i>	61	F	Saccular	Type 1	Unruptured	ND	Observation	None	2011
Tahir <i>et al.</i>	ND	ND	Fusiform	Type 1	Unruptured	ND	Observation	None	2019
Present case	72	M	Saccular	Type 1	Ruptured	4 mm	Coil embolization	None	2020
Enomoto <i>et al.</i>	44	M	Saccular	Type 2	Ruptured	10 mm	Clipping	None	1986
Sato <i>et al.</i>	69	M	Saccular	Type 3	Unruptured	ND	Clipping	None	2015

ND: Not described

left anterior cerebral artery showed a characteristic hairpin turn, and its distal part flowed in a wide range of peripheral parts of the anterior cerebral artery. Thus, this case was considered to be PPOA Type 1.

Although residual PPOA itself does not cause symptoms, some patients have been reported to present with anosmia or an aneurysm as a complication.<sup>[4,6]</sup> A peripheral anterior cerebral aneurysm is relatively rare, accounting for approximately 5% of all cerebral aneurysms.<sup>[5]</sup> Aneurysms of the anterior cerebral artery frequently appear at the tip of the large flexion of the main artery.<sup>[5]</sup> According to the reported cases, aneurysms of PPOA occur at the hairpin turn and may be caused by hemodynamic factors.<sup>[7]</sup> Our patient also had an aneurysm at the hairpin turn, supporting the occurrence of the event by hemodynamic factors. In previously reported cases, aneurysms were reported to occur on the greater curvature side of the hairpin turn, while an aneurysm occurred on the lesser curvature side of the hairpin turn in this case, indicating it may not have been caused by typical hemodynamic stress.<sup>[1,2,4,7,9-14]</sup>

This patient underwent radical endovascular surgery. Due to the presence of a PPOA aneurysm at the hairpin turn without a branch, difficulty in the stable placement of the microcatheter in the aneurysm was considered. Therefore, an intermediate catheter was used to improve the stability of the microcatheter. Further, an aneurysm of the PPOA has a wide neck.<sup>[7]</sup> In our patient, the size of the aneurysm was 3 mm at the dome and 3 mm at the neck, with dome/neck aspect of 1.0, indicating a slightly wide neck. Therefore, the 3D coil was selected as the first coil. As a result, a cage could be formed without deviating the coil into the parent artery.

To the best of our knowledge, 11 patients with PPOA aneurysms have been reported, including our patient.<sup>[1,2,4,7,9-14]</sup> four with ruptured aneurysms and seven with unruptured aneurysms [Table 1]. The mean age was 60.5 years. There were 6 men and 4 women. Age and sex of one patient were

unknown. The aneurysms were saccular in nine patients and fusiform in two patients. Nine, one, and one patient had PPOA Types 1, 2, and 3, respectively. Clipping was performed in seven patients, trapping in one patient; and the treatment was not mentioned for two patients. There have been no reports of patients treated by intra-aneurysm coil embolization, indicating that ours was the first case. Of eight patients who underwent craniotomy, three (37.5%) developed postoperative smell disorder. As the PPOA runs parallel to the olfactory cord, it is difficult to preserve the olfactory cord by direct surgery depending on the location of the aneurysms.<sup>[4,9]</sup> In intra-aneurysmal coil embolization, the anatomical location of the olfactory cord does not need to be considered. Therefore, this procedure may be more advantageous than craniotomy in preserving the olfactory sense. However, PPOA aneurysm is a hemodynamically generated aneurysm, and the long-term treatment results, such as the incidence of coil compaction after intra-aneurysmal coil embolization and the occurrence of neovascular aneurysm, are unknown. Therefore, careful follow-up is important in the future.

## CONCLUSION

This is the first report of coil embolization for persistent primitive olfactory artery aneurysm.

## Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

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Nil.

## Conflicts of interest

There are no conflicts of interest.

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