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Anterior Cerebral Artery Fusiform Aneurysm Attributable to Bilateral Persistent Primitive Olfactory Artery: Case Report

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Abstract

Persistent primitive olfactory arteries (PPOAs) are a rare variant of the anterior cerebral artery (ACA). Cerebral aneurysms may arise in the PPOA; most are saccular and on the unilateral PPOA. We report a 66-year-old male with bilateral PPOAs and a fusiform aneurysm on the left side detected at a health check-up. A brain magnetic resonance imaging (MRI) scan revealed a fusiform dilation in the proximal portion of the left ACA on a brain MRI. Good surgical results were obtained by combining trapping—and bonnet bypass surgery. Brain MRI and cerebral angiograms showed bilateral PPOAs and a fusiform aneurysm with the pearl-and-string sign in the proximal portion of the left PPOA. The aneurysm was trapped and a bonnet bypass using a radial artery (RA) graft was placed between the left superficial temporal artery and the distal portion of the left PPOA. The postoperative course was uneventful and 30 months after surgery he had no neurological symptoms; MRA showed no recurrence. In this patient, aneurysmal trapping and an A3-A3 bypass were an option, however, it would have placed an additional load on the right PPOA. Our decision to trap the aneurysm and perform bonnet bypass surgery using an RA graft led to success.

Keywords: anterior cerebral artery, fusiform aneurysm, persistent primitive olfactory artery

Introduction

Persistent primitive olfactory arteries (PPOAs) are a rare variant of the proximal anterior cerebral artery (ACA). In our patient, the residual medial olfactory artery failed to regress during embryological development and a hairpin turn remained.[1-3] The reported incidence of PPOA in the population is 0.14% in Japan,[3] 0.26% in Korea,[4] and 0.64% in Serbia;[5] the incidence of bilateral PPOAs is 0%-16.7%.[3-5]

Some patients with PPOA suffered olfactory disturbances,[4-7] suggesting that the arterial blood supply to the olfactory nerve was involved. Others,[3,4,8-11] reported that cerebral aneurysms are more likely to occur in individuals with PPOA. Most of these are saccular aneurysms in the unilateral PPOA; among 11 aneurysms 9 were saccular.[8]

We encountered a patient with bilateral PPOAs and a fusiform aneurysm on the left side; the combination of trapping and bypass surgery resulted in a good surgical outcome.

Case Report

This 66-year-old male with no relevant medical history underwent brain magnetic resonance imaging (MRI) at a health check-up. A fusiform dilation was detected in the proximal portion of the left ACA and he was referred to our hospital. He provided prior informed consent for the treatment and for publishing this report.

At the time of admission, he was alert and without neurological deficits. Brain MRA showed bilateral PPOAs; a fusiform cerebral aneurysm (6.5 × 7.3 mm) was observed in the proximal A1 segment of the left ACA. Internal carotid angiograms revealed the bilateral ACA to be PPOAs, and a fusiform aneurysm with the pearl-and-string sign was detected in the left proximal portion of the PPOA (Fig. 1).

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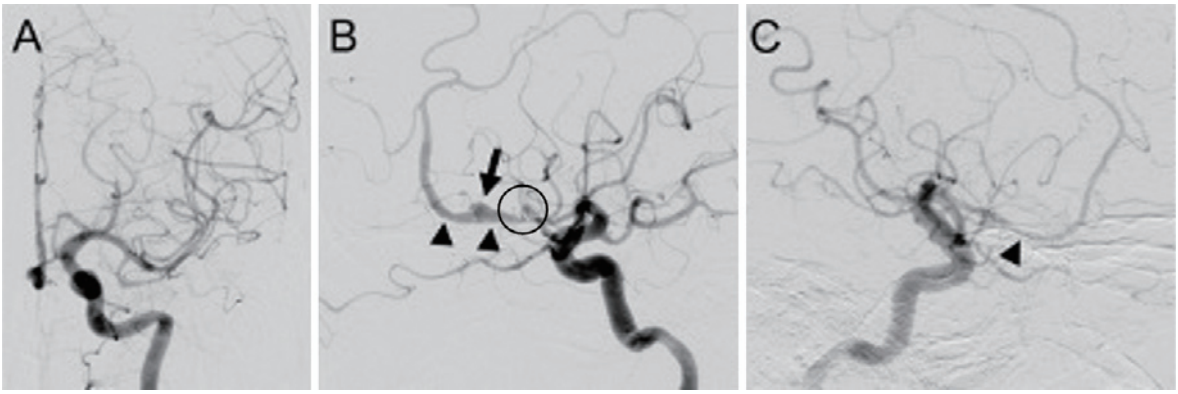

Fig. 1 Preoperative cerebral angiograms show a persistent bilateral primitive olfactory artery (arrowheads). A fusiform aneurysm (6.5 × 7.3 mm, arrow) with proximal stenosis (circle) is detected in the proximal region of the left anterior cerebral artery (B).
A, B Left internal carotid artery, anteroposterior (A) and lateral view (B).
C Right internal carotid artery, lateral view.

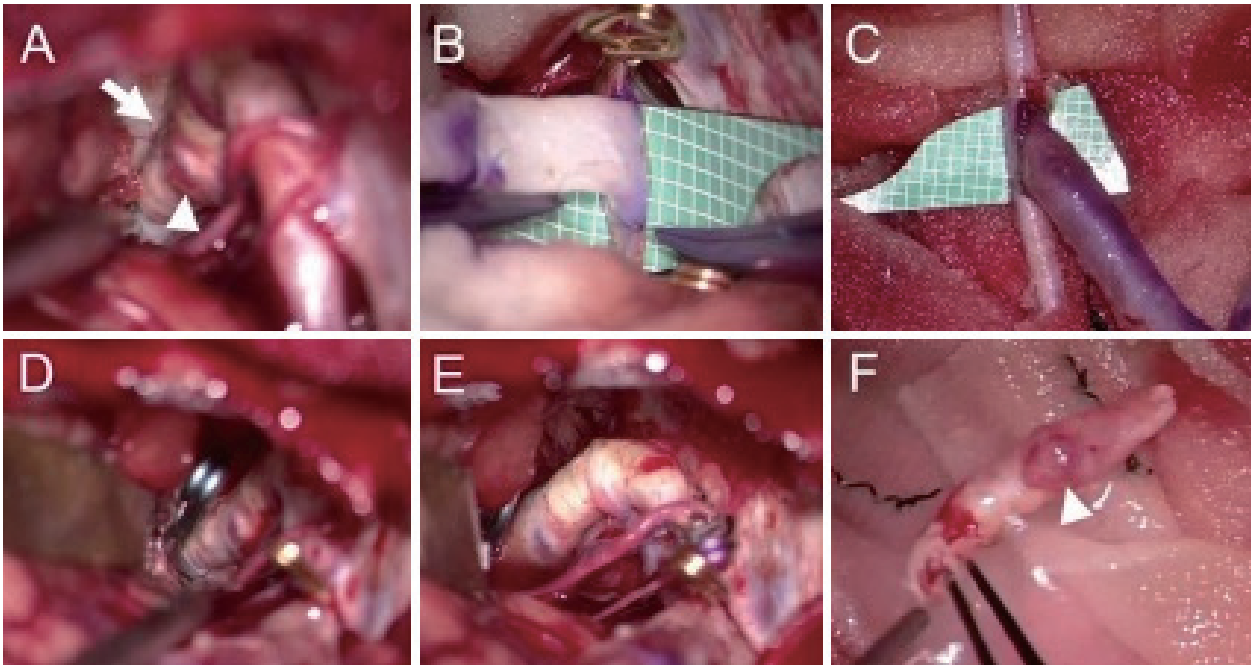

Fig. 2 Surgical views.
A Fusiform aneurysm (arrow). A part of the aneurysmal wall is extremely thin and bright red (arrowhead).
B Anastomosis to the distal side of the left A3 aneurysm with a radial artery graft and using 9-0 nylon sutures.
C The radial artery graft was anastomosed to the parietal branch of the left superficial temporal artery.
D, E The fusiform aneurysm was trapped (D, proximal clip; E, distal clip).
F Removal of the fusiform aneurysm. A part of the aneurysmal wall is extremely thin (arrowhead).

We explained to the patient that the natural history of cerebral aneurysms associated with bilateral PPOAs is unclear and presented him with treatment options that included conservative follow-up observation and surgical intervention and its associated risks. To trap the aneurysm, we planned a hemi-bonnet bypass using a radial artery (RA) graft between the left superficial temporal artery (STA) and the distal left portion of the ACA.

After left frontotemporal and bilateral frontal craniotomy we identified the left A3 via the interhemispheric approach and traced the vessel to its proximal portion (Fig. 2 A). The aneurysmal wall was extremely thin and bright red, and a dissecting aneurysm was strongly suspected based on imaging and intraoperative findings. For the bonnet bypass, we harvested approximately 15 cm of the RA and anastomosed the graft to the left A3 portion on the

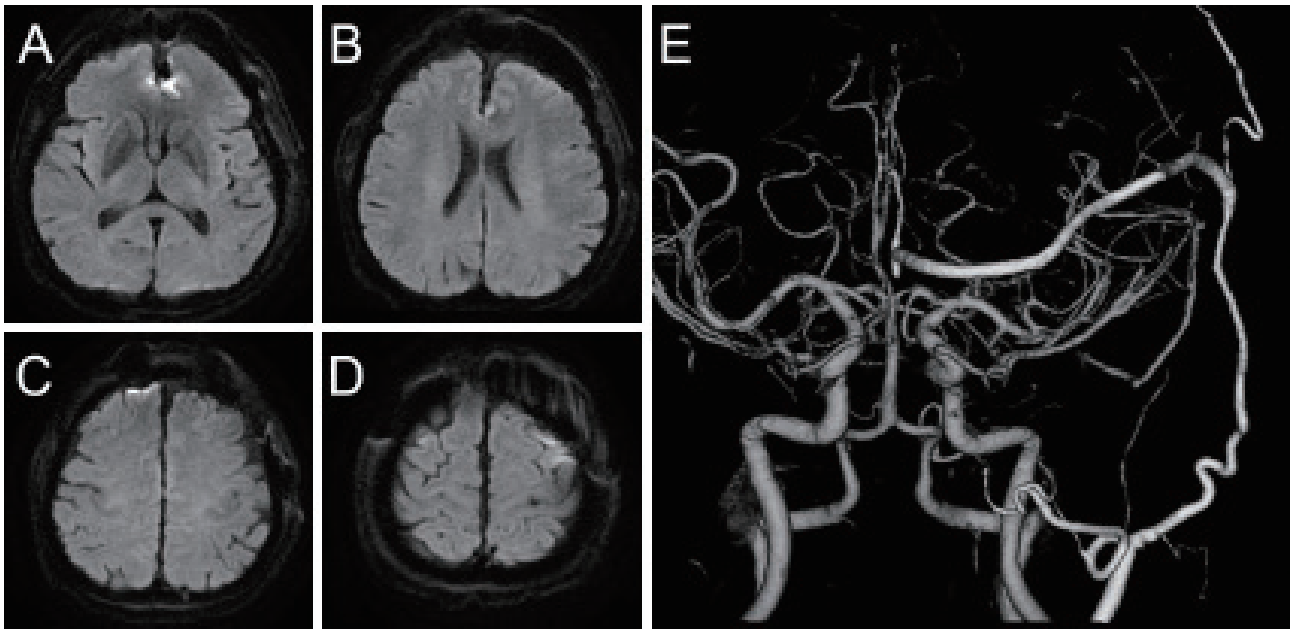

Fig. 3 Postoperative MRI scans.
A-D Diffusion-weighted images show no ischaemic lesions.
E Cerebral 3D-CT angiogram showing good patency of the bypass vessels and no lesions.
3D-CT: three dimensional-computed tomography; MRI: magnetic resonance imaging

distal side of the aneurysm (Fig. 2B). Then an RA graft was anastomosed to the parietal branch of the left STA (left STA-RA graft-left ACA hemi-bonnet bypass; Fig. 2C). Good blood flow was confirmed via intraoperative Doppler- and indocyanine-green video angiography, and the fusiform aneurysm was trapped and removed (Fig. 2D-F). There were no intraoperative changes in the monitored motor-evoked potential.

The postoperative course was uneventful; no ischaemic lesions were observed on the 1st postoperative day (Fig. 3 A-D). A cerebral three-dimensional-computed tomography angiogram obtained on the 7th day showed good patency of the bypass vessels and no lesions (Fig. 3E). The patient was discharged on the 15th day after surgery. At follow-up 3 years after surgery, there were no neurological symptoms, and MRA showed sufficient blood flow in the bypass vessels and no lesions.

Discussion

PPOAs and cerebral aneurysms

Aneurysms have been reported in individuals with PPOA.[3,4,9,11] They tend to be located at the characteristic hairpin bend[3,6-13] but can also be found at the proximal segment of the PPOA.[14,15] The PPOA is classified into 3 types.[1,2,5,6,8,14-16] Type 1 arises from the ICA and runs anteriorly along the olfactory tract. It makes a hairpin turn and supplies the distal ACA area. Type 2 arises from the normal ACA and passes through the cribriform plate to supply the nasal cavity as the ethmoidal artery. Type 3 is the transitional type between types 1 and 2.[14] Of these 3 types, type 1 is the most likely to elicit cerebral aneurysms;[3,6,7,9,13] among 11 PPOAs with aneurysm, 9 were of type 1, one was type 2, and one was type 3.[8] Ours was type 1.

Among 4 reported patients with bilateral PPOAs[3,4,6,17] plus ours in which the type could be determined, all were type 1. To the best of our knowledge, there has been only one earlier report of a patient with bilateral PPOAs and aneurysms.[6] The patient was a 54-year-old woman with a 3-year history of left-sided anosmia; the bilateral PPOAs were type 1. A saccular wide-necked aneurysm found in the hairpin curve of the left PPOA was clipped via an interhemispheric approach.

Although the relationship between PPOAs and aneurysms is unknown, haemodynamic stress,[3,7,8,16] increased haemodynamic stress resulting from hypoplasia of the contralateral A1 segment, or structural weakness and congenital fragility of the arterial wall may be involved.[6,7,9,14,15] In our patient we observed hypoplasia on the right side, suggesting that strong haemodynamic stress on the affected left side may have precipitated the development of the fusiform aneurysm.

Treatment strategy

Earlier patients with PPOA aneurysms were managed by observation, open microvascular techniques via craniotomy and endovascular treatment. Among 11 patients with PPOA-associated aneurysms (saccular: n = 9, fusiform: n = 2), 7 underwent clipping, one each aneurysmal trapping or coiling and 2 were observed.[8]

Kinachtchouk et al.[18] treated a PPOA aneurysm by the endovascular placement of a flow-diversion stent. Of 8 patients who underwent craniotomy, 3 developed a postoperative smelling disorder.[8] As the PPOA runs parallel to the olfactory cord, depending on the aneurysmal site, it may be difficult to preserve the cord by direct surgery.[8,15] Intravascular treatment does not need to consider the anatomical location of the olfactory tract, consequently it may be more advantageous than craniotomy. However, since haemodynamics may result in the development of PPOA aneurysms, coil compaction and aneurysmal recurrence must be considered.[8]

Our patient's fusiform aneurysm was located in the left hairpin turn of the PPOA. Although we considered aneurysmal trapping and placing an A3-A3 bypass, the procedure risks haemodynamic stress, placing an additional load on the contralateral hypoplastic PPOA and thereby increasing the risk of aneurysm development. Therefore, we performed trapping plus hemi-bonnet bypass surgery using an RA graft. The usefulness of STA-RA graft-ACA has been reported.[19-23] Others[24-26] used a contralateral STA as a graft for bonnet bypass surgery between the STA and ACA. Contralateral STA grafting may be an alternative option because it avoids distant graft harvesting and associated complications.[25,26] However, grafting decisions are dictated by the development, length, and course of the STA branches.[26]

In this patient, bypass surgery was performed due to concerns about ischaemia of the trapped vessel, but if there is abundant collateral circulation, simple trapping of the affected PPOA without bypass surgery is a viable option. It has been reported that fusiform aneurysms due to right A2 dissection can be treated with resection alone without bypass surgery.[27] Regarding balloon occlusion tests to determine the necessity of bypass surgery, Isozaki et al.[28] reported that in a patient with a fusiform aneurysm on the posterior cerebral artery, a super-selective balloon occlusion test of a posterior communicating artery was useful for developing their surgical plan. The necessity of combined bypass surgery should be carefully judged for each patient.

Conclusion

We encountered a rare case of a fusiform aneurysm in the proximal portion of the PPOA. The patient was successfully treated by aneurysm trapping and a STA-RA graft-distal PPOA hemi-bonnet bypass.

Conflicts of Interest Disclosure

The authors declare they have no conflicts of interest and no commercial relationships and received no support from pharmaceutical or other companies. All authors are members of The Japan Neurosurgical Society (JNS) and have completed the Self-reported Conflicts of Interest Disclosure Statement Forms available on the JNS website.

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