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CASE REPORT - VASCULAR NEUROSURGERY - ANEURYSM

Serpentine aneurysm of the posterior cerebral artery treated by internal maxillary artery bypass followed by parent artery occlusion: a case report and literature review

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Abstract
Serpentine aneurysms of the posterior cerebral artery (PCA) treated by the internal maxillary artery (IMA) bypass are rare. Here, the authors report the case of a 34-year-old male patient who presented with a half-year history of gradual severe headache and right-sided limb monoparesis and paresthesia lasting for 1 week. Preoperative angiograms showed a serpentine aneurysm in the left distal PCA, which was treated with internal maxillary artery-radial artery-posterior cerebral artery (IMA-RA-PCA) bypass followed by parent artery occlusion (PAO). The postoperative course was uneventful; radiological images revealed that the aneurysm disappeared, and there was good graft patency and excellent perfusion of the distal PCA territories. To the authors' knowledge, this is the first and only case of distal PCA serpentine aneurysm to be treated by IMA-RA-PCA bypass followed by proximal PAO. These findings suggest that IMA bypass surgery is a good and feasible treatment option for serpentine aneurysms of the PCA that can preserve the parent artery. Moreover, the anatomic segments of the PCA and different treatment options available for PCA serpentine aneurysms are also discussed in this study.

Keywords Serpentine aneurysm · Posterior cerebral artery · Internal maxillary artery · Bypass

Introduction

Serpentine aneurysms of the posterior cerebral artery (PCA) are an extremely rare entity that is a subgroup of all intracranial serpentine aneurysms, which are defined by special radiological and pathological characteristics, including a serpentine channel through the heavily thrombosed aneurysm. Currently, the treatment options for serpentine PCA aneurysms include aggressive occlusion of the parent vessel, balloon/stent-assisted coiling, trapping with resection of the aneurysm, and bypass with proximal endovascular/surgical occlusion of the parent vessel or trapping. Although the bypass procedure is time-consuming and involves the possibility of occipital infraction during temporary clipping; as a last resort, this approach is still a useful and effective option associated with low rates of recurrence and ischemic events when used in selected patients in whom balloon test occlusion (BTO) fails. Here, we report a case in which serpentine PCA aneurysms were successively treated by IMA-RA-PCA bypass followed by proximal aneurysmal segment occlusion with a 4-year follow-up.

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Case report

History and examination

A 34-year-old man was referred to our institution with a half-year history of gradual severe headache and newly developing right-sided limb monoparesis and paresthesia for 1 week. Neurological examinations revealed that his visual acuity and visual fields were normal, the muscular strength of his right-sided limbs were grade IV, and his superficial sensation

was slightly impaired. Computed tomography (CT) showed a high-density irregular mass with compression of the left midbrain. Enhanced magnetic resonance (MR) demonstrated a 1.9 × 1.4-cm partially thrombosed aneurysm. Digital subtraction angiography (DSA) revealed a serpentine aneurysm on the left posterior cerebral artery (P2 + P3 according to Zeal and Rhoton [27]) and fusiform dilation of the left A1 segment of the anterior cerebral artery (ACA) Fig. 1. Additionally, vital arteries arising from the aneurysmal body and poorly developed collateral circulation were found. The patient underwent failed BTO.

Operation

Considering the patient's poorly developed collateral circulation, the failure of BTO and his normal visual condition, a combination of frontotemporal and subtemporal approaches was carried out, and an IMA-RA-PCA bypass was performed via end-to-end anastomosis (IMA to proximal radial artery) and end-to-side anastomosis (distal radial artery to distal aneurysm segment of the PCA) followed by parent artery occlusion (PAO) using a microsurgical clip. Intraoperative ultrasonic Doppler and indocyanine green video angiography showed good patency of the bypass graft. Routine neurophysiologic monitoring, including somatosensory evoked potentials and electroencephalography, were used.

Postoperative course and follow-up

The patient's postoperative course was uneventful without any neurological dysfunction, including no impairment in his visual acuity or visual field. His muscular strength had reached grade V before discharge. Postoperative cerebral angiography and CT showed good graft patency, complete obliteration of the aneurysm, and no further cerebral infarction. Follow-ups performed at 3 months and 4 years showed no neurological deficits.

Discussion

Serpentine aneurysm of the PCA is a rare and challenging lesion for clinical neurosurgeons because of its deep anatomic location, closeness to vital midbrain structures, and possession of a series of important perforators [2, 8, 9, 21, 24, 25]. Several authors have divided the PCA in different ways; previous reports include Yasarigil [26] (P1 to P5) and Zeal [27](P1 to P4). Zeal's classification is used most frequently. More recently, Seoane and Rhoton [21] provided another classification of the PCA that could be used to choose the best and most suitable approach for PCA aneurysm exposure according to the aneurysm location on angiographic imaging; their method divides the PCA into the following segments: S1 (anterior), S2 (middle), and S3 (posterior). Because it possesses a series of branches that originate

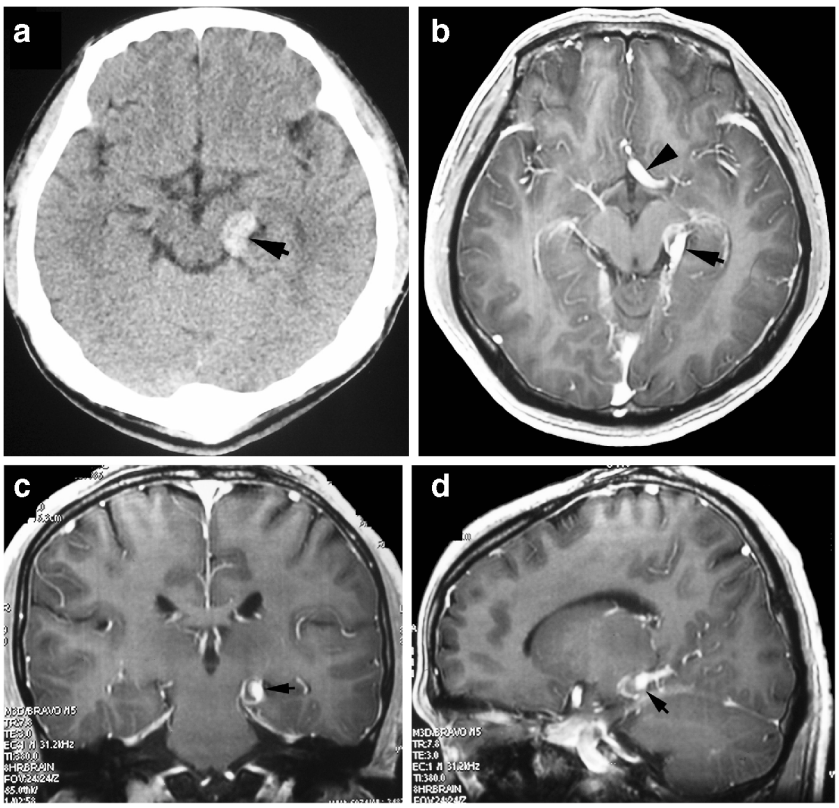

Fig. 1 Preoperative axial (**a**) CT and axial (**b**), coronal (**c**), and sagittal (**d**) contrast-enhanced MR images showing a vascular occupancy that compressed the cerebral peduncle (arrows) and fusiform dilation of the anterior cerebral artery (arrowhead, **b**)

Table 1 Summary of the literature on treated serpentine aneurysms of the PCA

Reference	Age/sex	Clinical presentations	Side	Segment of PCA	Treatment option	Aneurysm condition	Complications	Hemianopsia	Follow-up time (months)	Follow-up outcomes
1. Yu-Wei Hsu et al. [11] (2017)	49/F	Headache and LT lower limb paresthesia	RT	NA	Endovascular coil Endovascular coil	Disappeared	None	NA	5	Excellent
2. ZW Zhao et al. [28] (2008)	32/M	Headache	NA	NA	PAO	Disappeared	None	NA	NA	NA
3. Han Soo Chang et al. [1] (1986)	20/M	Hemiparesis	RT	NA	Trapping followed by STA-PCA bypass	Disappeared	Transient confusion, LT hemiparesis	Yes	3	Good
4. Akira Fukamachi et al. [7] (1982)	48/F	Headache, RT hemiparesis, RT homonymous hemianopsia, and disturbance of recent memory	LT	NA	Trapped and excised	Disappeared	LT oculomotor palsy	No	12	Improved
5. Youxiang Li et al. [17] (2008)	43/M	Headache and RT heminumbness	LT	P2	PAO	Disappeared	NA	NA	8	Excellent
	48/M	SAH	RT	P2	PAO	Disappeared	NA	NA	4	Excellent
	31/M	SAH	LT	P2	PAO	Disappeared	NA	NA	7	Excellent
	4/M	Headache	RT	P1	PAO	Disappeared	NA	NA	36	Excellent
6. Paul Hallacq et al. [10] (2002)	26/M	Headache and nausea	RT	P2	PAO	Disappeared	None	No	12	Excellent
7. Coley SC et al. [4] (2002)	43/M	Headache	RT	P2	Endovascular occlusion of the aneurysm	Shrink	None	No	6	Excellent
8. Kyu Chang Lee et al. [15] (1999)	18/F	Ptosis and headache	LT	P2	Trapping and aneurysmectomy	Disappeared	Temporary paresis of the left oculomotor nerve	No	NA	
9. Elisa F. Ciceri et al. [3] (2001)	21/F	NA	NA	P1	PAO	Disappeared	None	NA	6	Excellent
	12/M	NA	NA	P2a	PAO	Disappeared	None	NA	NA	Excellent
	58/M	NA	NA	P2-P3	PAO	Disappeared	PCA stroke and HHA	NA	NA	Excellent
	58/M	NA	NA	P3	PAO	Disappeared	None	NA	NA	Excellent
	57/M	NA	NA	P3	PAO	Disappeared	None	NA	NA	Excellent

LT left, *RT* right, *NA* not available, *PAO* parent artery occlusion, *STA* superficial temporal artery, *PCA* posterior cerebral artery, *P1* P1 segment of the PCA, *P2* P2 segment of the PCA, *P3* P3 segment of the PCA, *HHA* homonymous hemianopsia

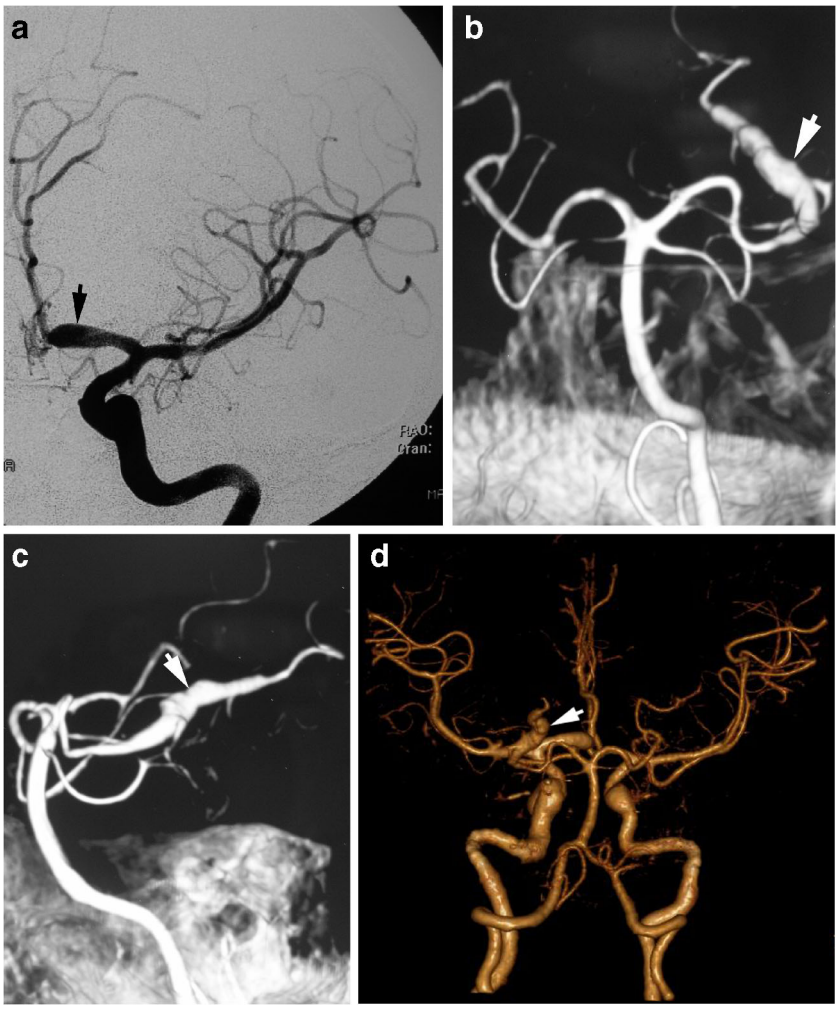

Fig. 2 Preoperative digital subtraction angiogram (**a**) showing fusiform dilation of the A1 segment of the anterior cerebral artery (arrowhead, **a**) and poor collateral circulation supplied by the ipsilateral ACA, MCA ,and PCoA. 3D digital subtraction angiograms (**b**, **c**) and 3D CT angiogram (**d**) confirmed the presence of a serpentine aneurysm of the posterior cerebral artery (arrow)

from different segments of the PCA and provide blood flow to many vital brain structures, such as the thalamus, cuneus, visual cortex, and parieto-occipital sulcus [21], performing parent-vessel sacrifice to treat PCA serpentine aneurysms may increase the possibility of infraction, which can lead to neurological deficits, including contralateral hemiparesis, homonymous hemianopsia, and hemihypesthesia [24]. Therefore, it is vitally important to maintain efficient blood flow to PCA territories. In the serpentine aneurysm of the distal PCA reported here, an IMA bypass [22] was performed, and the aneurysm was exposed via a pterional and subtemporal approach to remove the zygomatic arch and resect some of the temporal lobe.

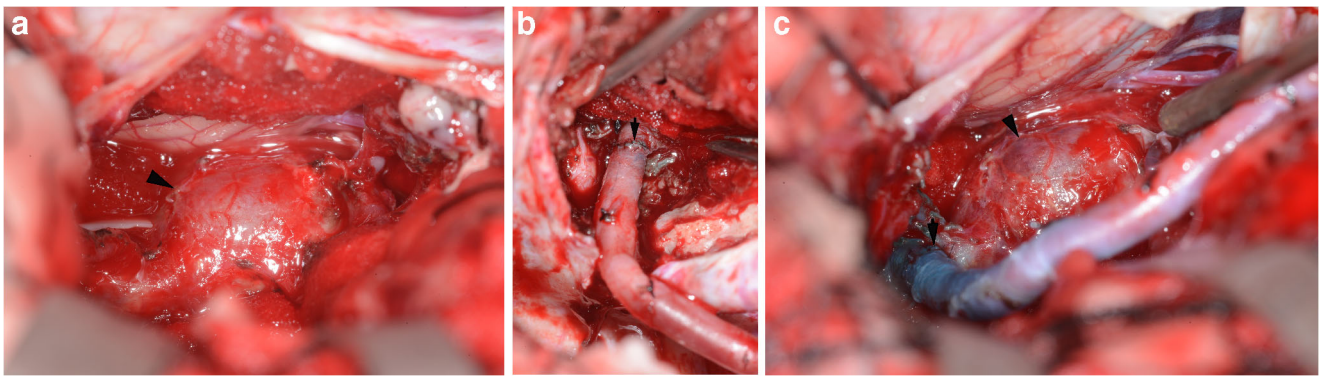

Fig. 3 Intraoperative photographs showing an aneurysm of the PCA (arrowhead, **a**, **c**), an end-to-end anastomosis site (arrow, **b**) between the proximal radial artery and the internal maxillary artery and an end-to-side anastomosis site (arrow, **c**) between the distal radial artery and the distal aneurysmal segment

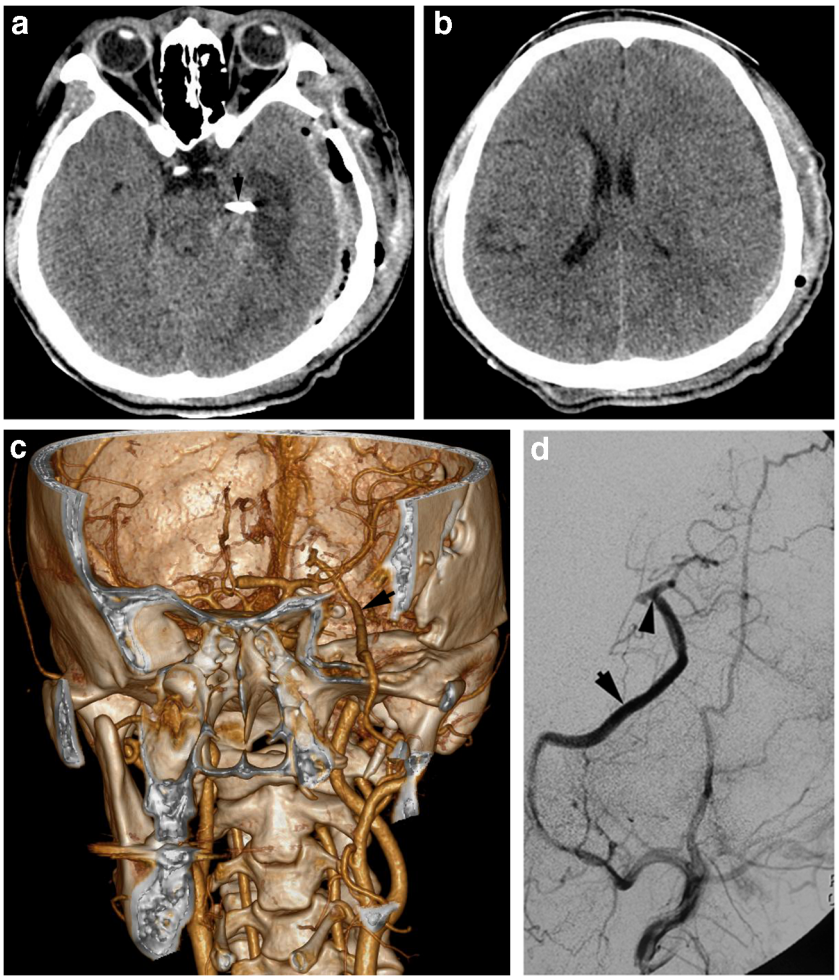

Fig. 4 Postoperative axial CT (**a**, **b**) showing no occipital infarction of the PCA territory and the microsurgical clip (arrow, **a**). 3D CT angiogram (**c**) and 2D digital subtraction angiogram (**d**) showing the patency of the graft vessel (arrow, **c**, **d**) and the distal anastomosis site (arrowhead, **d**)

The treatment of serpentine aneurysms of the distal PCA is both complex and challenging, and some of its complications are potentially lethal. Some authors [10, 12, 16, 19] have supported the opinion that occlusion of the PCA does not cause clinical deficits because the associated vascular network is well-developed [6] and provides collateral blood supplied from the terminal branches of the anterior and middle cerebral arteries. However, the regions supplied by the perforating

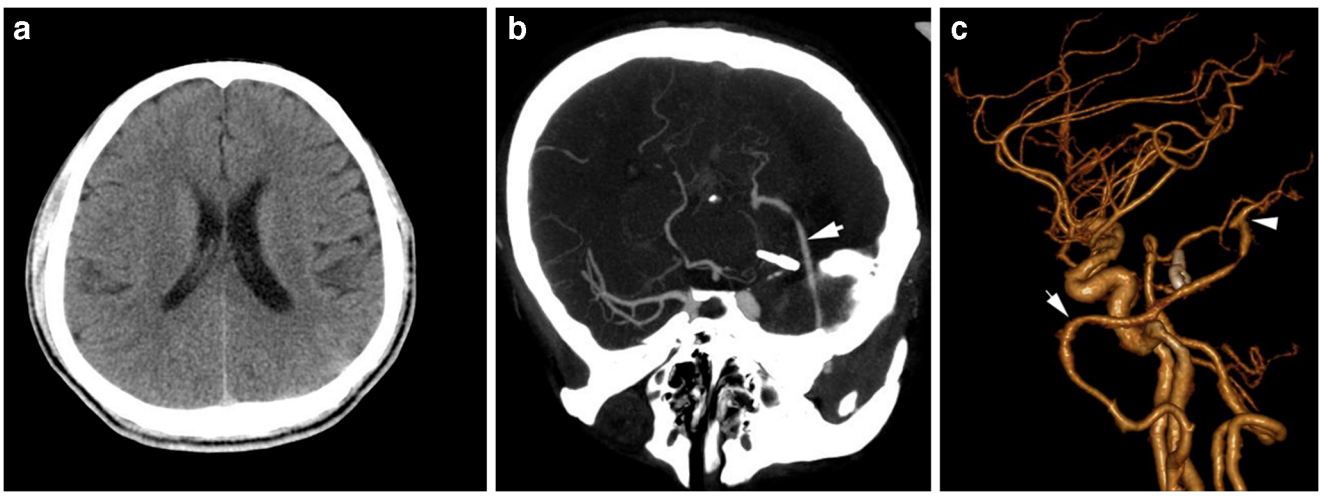

Fig. 5 Postoperative 3-month follow-up images. Axial CT (**a**) showing no occipital infraction of the PCA territory. CT angiograms (**b**, **c**) showing good patency in the graft artery (arrow) and the end-to-side anastomosis site (arrowhead, **c**)

arteries or cortical branches are at risk of occipital infarction, as demonstrated by Drake [5], who showed that 17% (4/23) of patients with PCA occlusion had visual hemianopsia. In the modern era, BTO should be performed prior to microneurosurgery to evaluate the patient's tolerance to permanent sacrifice of the PCA. However, even if the patient passes the BTO, an irreversible infarction may occur postoperatively [13]. Therefore, it may be more important than was previously thought to preserve the parent artery in clinical practice, especially in patients in good preoperative condition. It is also difficult or unsuitable to clip-reconstruct serpentine aneurysms of the distal PCA when the entire vessel is involved [2], to clip-wrap such aneurysms given that the long segment of the artery could harbors disease [2], or to trap the aneurysm via resection when it possesses a series of perforators. Recently, endovascular techniques, such as balloon/stent-assisted coiling, have become popular as they are less invasive than open craniotomy. However, these procedures have a higher risk of recurrence and rehemorrhage [2] given that the remaining parent vessel is diseased and a long period of postoperative anticoagulation therapy is required; hence, their curative effect must be evaluated via long-term follow-up because over a prolonged period, complications could exceed the initial benefits of therapy. As a last resort, bypass followed by PAO may be considered in some selected candidates who lack collateral circulation, undergo a failed BTO, and have good preoperative vision and movement. Moreover, this procedure should not be performed with endovascular coiling of the parent artery to avoid postoperative complications related to anticoagulation therapy. The different treatment strategies available in PCA serpentine aneurysms are shown in Table 1.

Several bypass procedures for PCA aneurysms that involve the STA-posterior temporal artery, STA-PCA anastomosis, occipital artery-PCA anastomosis, grafting of extracranial great arteries to the PCA, and end-to-end anastomosis with the removal of the aneurysm have been proposed [14, 18, 20, 23]. To our knowledge, this is the first case of a PCA serpentine aneurysm to be treated by the IMA bypass followed by PAO. Although fusiform dilation of the A1 segment of the anterior cerebral artery (ACA) was observed, the decision was made to address the PCA aneurysm that had caused the patient's clinical presentations in order to improve the patient's quality of life and avoid fatal rupture of the aneurysm (Fig. 2). As a result, given our rich experience in cerebral revascularization, an IMA-RA-PCA bypass was performed as a standard intermediate/high-flow bypass, unlike other bypasses in which the donor vessel is the superficial temporal artery (STA) or occipital artery (OA) Fig. 3. In addition, this bypass procedure provided relatively higher blood perfusion to the distal cortical branches of the PCA, thus avoiding postoperative occipital infraction, as demonstrated in Figs. 4a, b, and 5a.

In summary, serpentine aneurysms of the distal PCA are rare. Treatment options include parent-vessel sacrifice, balloon/stent-assisted coiling, trapping with resection, and bypass, which should be considered in light of the results of BTO, the collateral circulation of the distal PCA territory, the condition of the patient, and patient preference. Preservation of the parent vessel is crucial for favorable treatment outcomes. To the best of our knowledge, this is the first case of a serpentine aneurysm of the distal PCA to be successfully treated by the IMA-RA-PCA bypass followed by PAO with long-term follow-up.

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Compliance with ethical standards

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

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee (Beijing Municipal Science and Technology Commission) and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Patient consent The patient has consented to the submission of the case report for submission to the journal.

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Comments

This is a case report of a serpentine aneurysm of the PCA treated by an internal maxillary PCA bypass using a radial artery interposition graft, and proximal artery occlusion. The authors are to be congratulated for tackling such a complicated aneurysm. This case is another example of the current role of bypass surgery in dealing with such complex aneurysms. The treatment options discussed are a nice addition and have an educational value to the reader.

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