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CASE REPORT - VASCULAR

Bilateral cavernous carotid aneurysms treated by two-stage extracranial-intracranial bypass followed by parent artery occlusion: case report and literature review

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Abstract Bilateral cavernous carotid aneurysms (CCAs) are often not amenable to neurosurgical clipping or endovascular coiling. Here, we report the case of a 50-year-old female who presented with a 1-year history of gradual severe headache. Preoperative angiograms revealed bilateral CCAs. Among these findings, the right giant CCA had been trapped after the external carotid artery-saphenous vein-middle cerebral artery (ECA-SV-MCA) bypass 8 years prior. Additionally, the left CCA was again trapped after the internal maxillary artery-radial artery-middle cerebral artery (IMA-RA-MCA) bypass, followed by parent artery occlusion (PAO), because of the enlargement of a 0.4-cm aneurysm to a 1.3-cm aneurysm during the 5th to 8th years following surgery. Postoperative radiologic findings proved that the aneurysms disappeared with good graft patency of the bilateral anastomoses and excellent filling of the bilateral MCA territories. This is the first case of bilateral CCAs treated with two stages of bilateral high-flow extracranial-intracranial (EC-IC) bypass, including an IMA-RA-MCA bypass.

Keywords Cavernous carotid aneurysms · Extracranial–Intracranial bypass · Internal maxillary artery · Enlargement · Bilateral bypass

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Introduction

A number of retrospective studies and case series have reported that cavernous carotid aneurysms (CCAs) account for 2–9% of all intracranial aneurysms and that bilateral CCAs, which were first reported by Sir Gilbert Blane in 1800 [1, 14], account for 6–11% of all CCAs [9, 16]. Most patients with CCAs are asymptomatic. Of those who present with symptomatic CCAs, most are over 50 years of age, female and Caucasian [3]. Idiopathic, traumatic, iatrogenic and mycotic aneurysms are common in the cavernous segment of the carotid artery [1]. Currently, the treatment paradigms for CCAs include the constructive strategies of flow diversion treatment, balloon/stent-assisted coiling and direct microsurgical clipping as well as the deconstructive Hunterian strategies via microsurgical clip placement/carotid ligation/endovascular PAO with or without revascularization [1, 17, 21]. Although the indirect method of giant CCA treatment is employed by endovascular coils or proximal internal carotid artery ligation, the contralateral CCA may be at increased risk of subsequent distention or enlargement when anterior cerebral crossover flow develops following carotid occlusion [12]. Herein, we report a case of symptomatic bilateral CCAs that were successively treated by an ECA-SV-MCA bypass and IMA-RA-MCA bypass followed by PAO during an 8-year interval.

Case report

History and Examination A 50-year-old female initially presented to our hospital with a 1-year history of gradual severe headache of the right forehead and orbital cavity. Neurologic examinations revealed normal visual acuities and visual fields without any cranial nerve palsy. Laboratory examinations were within normal limits. Computed tomography (CT) revealed a high-density, 2.5 × 3 cm non-uniform agglomeration next to the right side of the cavernous sinus and a 0.4 × 0.5 cm lesion on the left side, which had a slightly higher density (Fig. 1a). Digital subtraction angiography (DSA) showed a 3-cm giant aneurysm containing a thrombus on the right internal carotid artery (ICA) (C3 + C4 according to the Bouthillier Classification). Additionally, a well-developed right posterior communicating artery (PCoA) (Fig. 1b, c) and a 0.4-cm aneurysm on the left ICA (C4) were found (Fig. 1d, e).

First operation Direct clipping followed by reconstruction of the right CCA was ruled out as primary therapy because the CCA was surrounded by bone. Flow diversion or other endovascular strategies seemed inappropriate because of the partially intraluminal thrombosis and progressive mass effect (Fig. 1a, b, c). Before the operation, Mata's test (press the ipsilateral carotid artery 20–30 min at a time for a week) was performed to examine the patient's tolerance of ICA occlusion. We decided to conduct an EC-IC bypass with an interposed SV graft followed by PAO through a frontotemporal transsylvian craniotomy. During that time, a 20-cm-long proximal SV was connected to a distal portion of the ECA starting from the lingual artery through an end-to-end anastomosis (Fig. 1f, g), and the distal SV was connected to the frontal branch of the M2 segment of the MCA through an end-to-side anastomosis (Fig. 1h, i). Ultrasonic Doppler was used to measure the SV graft blood flow of 110 ml/min. PAO was conducted by ICA ligation at the beginning of C1 and ICA clipping at the proximal origin of the ophthalmic artery to preserve the source of collateral flow by removing the clinoid process (Figs. 1h, i and 3f).

Postoperative course and follow-up The patient had a mild headache during the first 5 days after surgery. She recovered completely and was able to resume her regular activities ½ year later. Postoperative DSA revealed complete aneurysm occlusion and perfect MCA territory perfusion (Fig. 2a). Three-dimensional CT angiography (CTA) revealed sufficient bypass patency of the right ECA-SV-MCA without CCA recurrence during the 8-year follow-up (Figs. 2b and 3g). However, the diameter of the left CCA increased from

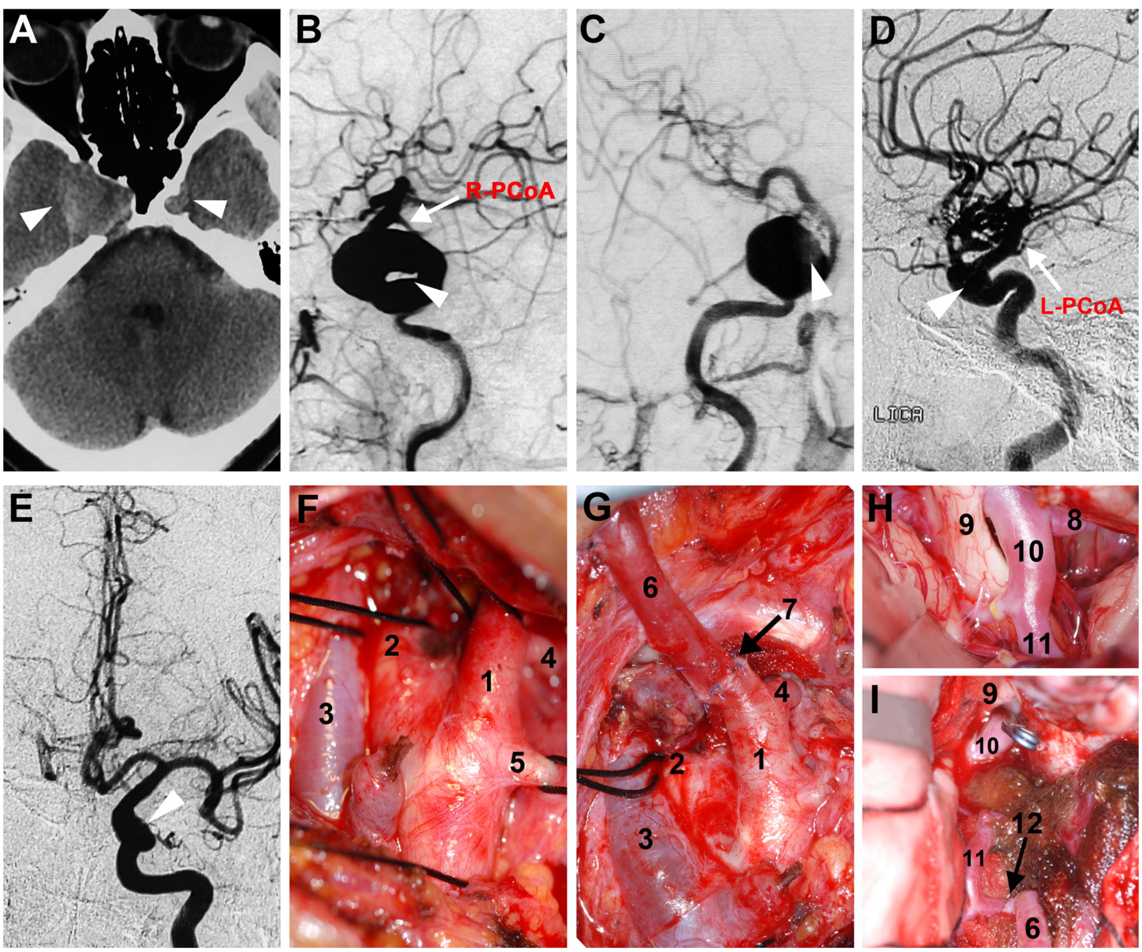

Fig. 1 Preoperative image examination and intraoperative exposure: **a** Axial CT. **b and c** Lateral (**b**) and front (**c**) position DSA. **d and e** Concurrent lateral (**d**) and front (**e**) position DSA. **f, g, h and i** 1, ECA; 2, ICA; 3, internal jugular vein; 4, lingual artery; 5, superior thyroid artery; 6, SV; 7, end-to-end anastomosis; 8, R-PCoA; 9, optic chiasma; 10, L-ICA; 11, L-MCA (M1); 12, end-to-side anastomosis

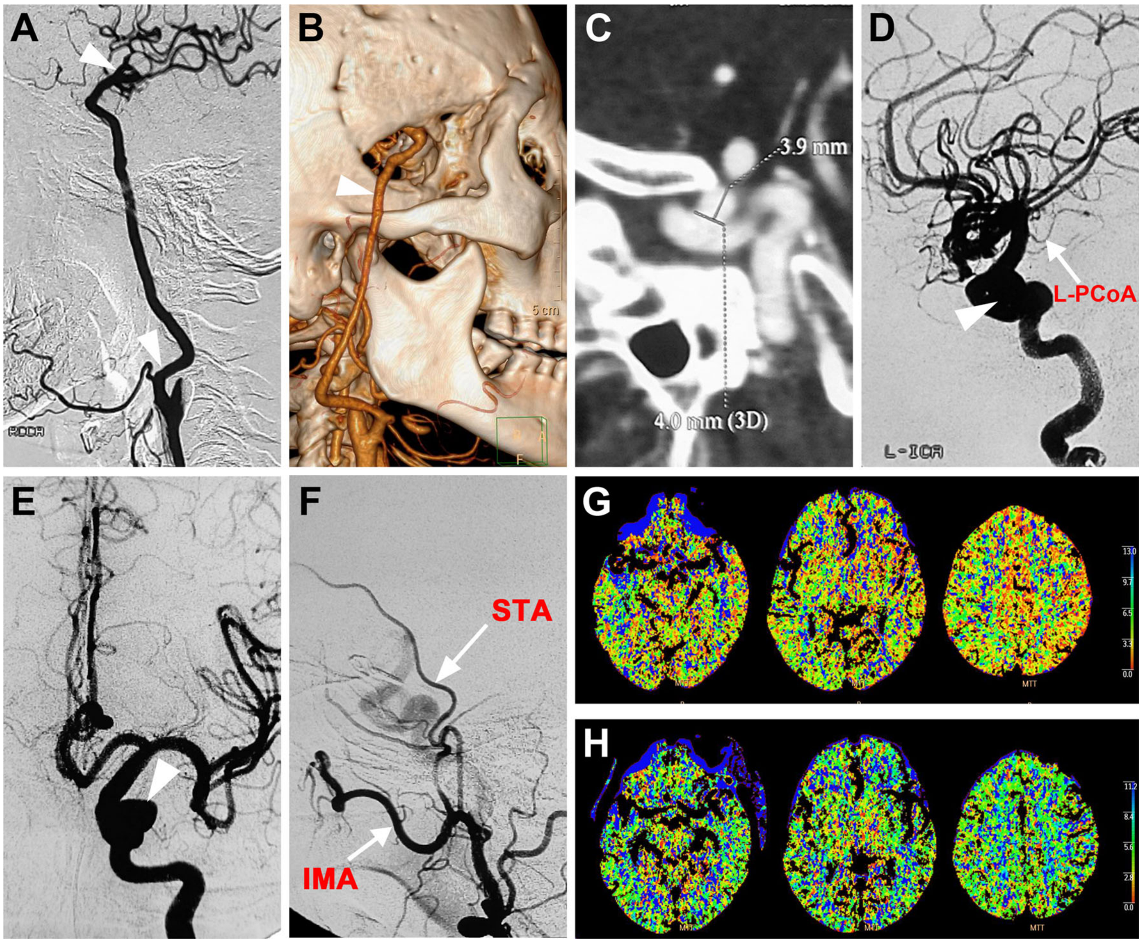

Fig. 2 Postoperative course: **a** and **b** Lateral position DSA (3 months) and CTA (5 years). **c** CTA (5 years). **d** and **e** Left ICA (8 years). **f** Left ECA. **g** CTP before the second operation. **h** CTP after the second operation. There was no significant difference in the right mean transit time (MTT) both between the left and right and before and after surgery

0.4 cm to 1.3 cm during the 5th to 8th years following surgery (Fig. 2c, d, e). The patient had suffered from hypertension for more than 10 years, although no neurologic symptoms appeared. Based on these radiologic findings, we proposed flow diversion treatment rather than EC-IC bypass because the pipeline embolization device could easily and effectively treat the left enlarged CCA. However, the patient and patient family members insisted on selecting craniotomy over flow diversion treatment because they had serious financial obstacles and strong confidence in the EC-IC bypass.

Second operation Considering the patency of the left PCoA on DSA images (Fig. 2d), the left IMA was much thicker than the ipsilateral superficial temporal artery (STA) (Fig. 2f), and the patient could tolerate balloon test occlusion (BTO) of the parent artery for over 30 min without discomfort. Thus, an IMA-RA-MCA bypass followed by PAO through a frontotemporal transsylvian craniotomy and zygomatic arch osteotomy was performed. An 8-cm-long RA was connected to the IMA through an end-to-end anastomosis (Fig. 3a), and the distal RA was connected to the frontal branch of M2 (MCA) through an end-to-side anastomosis (Fig. 3b). Intraoperative Doppler flowmetry (blood flow for the RA graft was 77 ml/min) and indocyanine green video angiography showed sufficient bypass patency (Fig. 3c, d). PAO was conducted using procedures identical to those of the first operation (Fig. 3c). The patient's postoperative course was uneventful without any neurologic dysfunction during the 6-month follow-up. Neither CT perfusion (CTP) nor CT demonstrated ischemic changes compared with preoperative findings (Figs. 2g, h and 3e). Postoperative CTA proved good graft patency of bilateral anastomoses and excellent filling of bilateral MCA territories (Fig. 3f, g).

Discussion

The location of the CCA is sometimes challenging. In fact, it is extremely important to distinguish the cavernous segment of the ICA from the ophthalmic segment (recognizing the dural ring) because an aneurysm located in the ophthalmic segment of the ICA can cause a subarachnoid hemorrhage

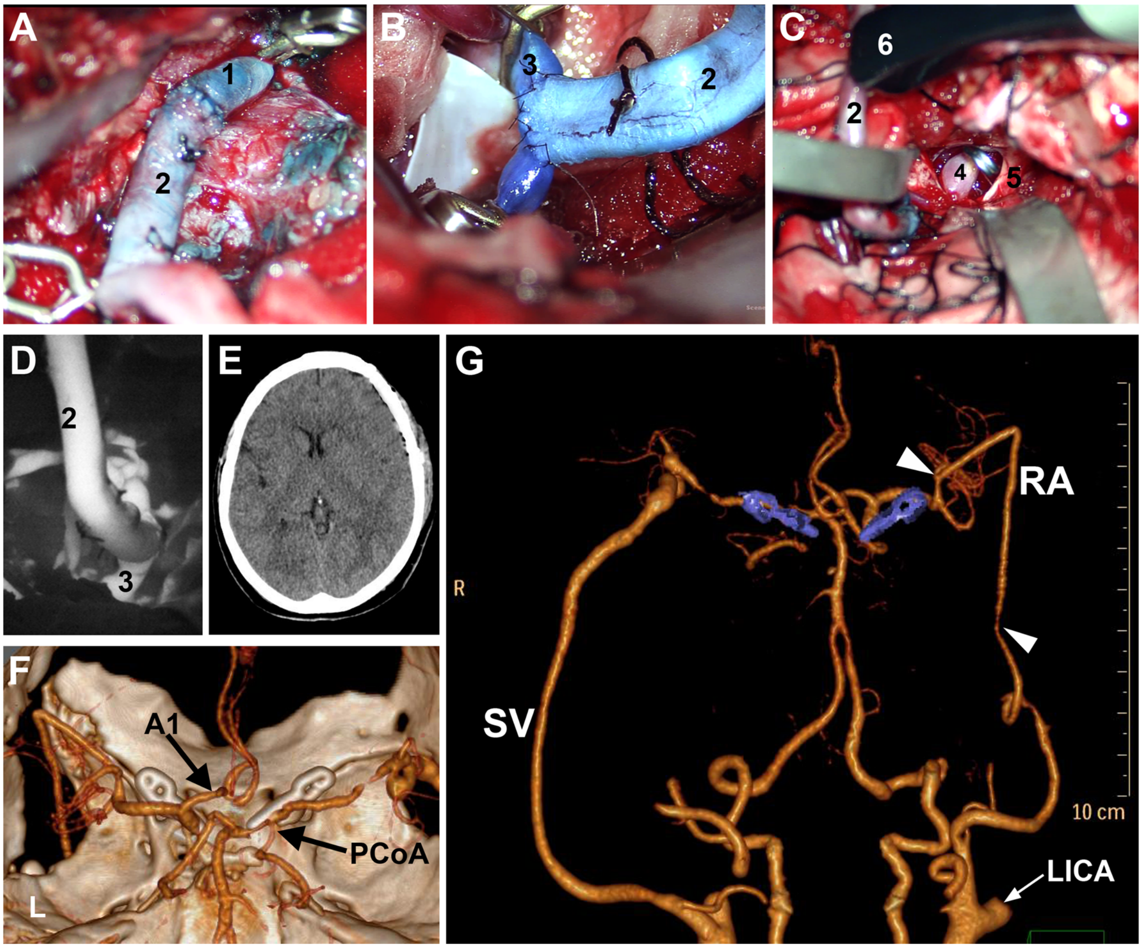

Fig. 3 Second operation and postoperative course: **a** End-to-end anastomosis. **b** End-to-side anastomosis. **c** Intraoperative Doppler flowmetry and ICA clipping. 1, IMA; 2, RA; 3, MCA (frontal branch of M2); 4, L-ICA; 5, optic chiasma; 6, Doppler flowmetry probe. **d** Indocyanine green video angiography; **e** CT scan performed 20 days after surgery; **f** CTA (20 days after surgery); **g** CTA (6 months after surgery)

(SAH), while an aneurysm located in the cavernous segment can cause a carotid-cavernous fistula in case of rupture [9]. This distinction is particularly difficult in the case of a giant aneurysm because a giant aneurysm originating from the cavernous segment of the ICA can pass the dural ring and become subarachnoid in location. Consequently, the decision of treatment for CCAs and the balance of risks/benefits must be evaluated carefully, since the risk of rupture is not the risk of a fatal bleeding, but the risk of a carotido-cavernous fistula that is not fatal, even if dangerous, for the involved eye [4, 9, 19].

Symptoms of a progressive mass effect from CCAs are associated with compression of the cranial nerves, acute thrombosis, traumatic or spontaneous rupture, and osseous erosion of the sphenoid sinus, which can lead to fatal epistaxis [3, 18]. Other results of CCA rupture can include ocular ischemia due to the steal phenomenon or SAH when projecting intradurally or directly into the sinus at the time of rupture [3]. According to the International Study of Unruptured Intracranial Aneurysms, the 5-year cumulative rupture rate of unruptured CCAs was 0% for aneurysms <12 mm, 3% for 13–24 mm and 6.4% for aneurysms >25 mm [20]. However, the data did not stress the risk of increased volume for CCAs or symptomatic lesions. To ensure that the benefits of therapy outweigh the risks, it is recognised that there is no need to treat small (<12 mm) asymptomatic CCAs immediately. Exceptions include a gradual increase in size, projection of an aneurysm into the subarachnoid space, a developing thrombus or acute thrombosis within aneurysms and anatomical extension into the subarachnoid space or osseous erosion, which may otherwise result in SAH or fatal epistaxis [1, 3, 16, 20]. In this case, the right symptomatic giant CCA presented with an internal thrombus and obvious osseous erosion, while the left asymptomatic significant CCA had characteristics of an increase in size during monitoring, while the patient also suffered from hypertension.

Regarding the enlargement of the contralateral CCA after one-sided ICA occlusion for bilateral CCAs, Nishino et al. [12] reported that five of six (83.3%) patients who suffered from bilateral CCAs experienced this, and four underwent STA-MCA bypass surgery. The most likely cause for growth of the contralateral CCA is hemodynamic stress on the wall when the ipsilateral ICA is occluded. Additionally, spontaneous enlargement in CCAs can occur more easily in aneurysms arising from other locations, and congenital factors might also play a role [2]. As a result, periodic postoperative radiologic monitoring is essential for cases such as this.

Multiple treatment approaches for CCA that involve antithrombotics, endovascular therapy and surgical interventions have been proposed [3]. However, these treatments have failed to achieve effective conservative or endovascular management of bilateral CCAs [1, 7, 8]. Flow diversion treatment, such as a pipeline embolization device, is associated with a higher rate of symptom improvement, less need for future treatment and a lower rate of complications than the conventional endovascular procedures that are preferred for treatment for CCAs in the modern era. However, long-term follow-up is still needed to confirm this finding [1, 21]. Therefore, cerebral revascularization, particularly the EC-IC bypass, remains the preferable and feasible option for managing bilateral CCAs based on our practices. This case was treated with bypass techniques rather than with incomplete coiling and stents or flow diversion techniques, which may not definitively cure the aneurysm. Patients without good endovascular options can be referred easily for surgery when collaborative experts work together as a team. Appropriate selection of treatment modality will ensure that patients with complex aneurysms receive surgical treatments that will exclude the aneurysm and prevent unnecessary retreatments later for endovascular recurrence, compaction or rehemorrhage.

Only eight cases of bilateral revascularization followed by PAO have been reported in the literature [5, 6, 10, 11, 13, 18]. Nevertheless, none of these cases selected the IMA as the feeding artery (Table 1). When evaluating methods of how to manage the left enlarged CCA, preoperative DSA demonstrates a reduction in the caliber of the left PCoA compared with the preoperative evaluation 8 years earlier (Figs. 1d and 2d). This means that there is scant supply from the posterior circulation to the anterior, let alone supply from the left ICA to the bilateral anterior cerebral artery (ACA) with the anterior communicating artery to adequately compensate for the right hemisphere (Figs. 2e and 3f). Subsequently, BTO analysis was performed with CTP, which is always used to identify the risk for ischemic infarction after carotid occlusion [4, 18]. When considering sacrificing the ICA to treat an enlarged CCA, an STA-MCA bypass would be inadequate (Fig. 2f). As a result, the IMA-RA-MCA bypass, which is deemed to be an efficient intermediate/high-flow bypass, was performed followed by PAO [15]. In this case, it appeared appropriate to trap the bilateral CCAs in patients with headache symptoms due to cavernous sinus compression and persistent cranial neuropathy. The distal carotid clipping must be placed proximal to the ophthalmic artery to preserve the source of collateral flow through the removal of the clinoid process.

Table 1 Nine cases of bilateral CCAs that underwent bilateral revascularization followed by PAO

	Reference	Age/sex	Diagnosis	Operation			Graft patency duration
				Left	Right	Time interval	
1	Ng et al. (2001) [11]	59/F	Left caroticocavernous fistula /right CCA	Common carotid artery-SV-MCA (M3) end-to-side bypass &	STA-MCA and ECA-SV-MCA (M3) bypass with ICA occlusion	2 days	6 months (B)
2	Ma et al. (2010) [10]	30/M	Fibromuscular dysplasia/bilateral CCAs	ECA-SV-MCA(M2) bypass with ICA ligation	STA-MCA bypass with ICA occlusion &	60 months	4 years (L) 9 years (R)
3	Rehman et al. (2010) [13]	11/M	Juvenile Paget's disease/bilateral CCAs	STA-MCA bypass with ICA occlusion &	STA-MCA bypass with ICA occlusion	3 months	1 year (B)
4	Gobble et al. (2012) [6]	-----	Bilateral CCAs	ECA-SV-MCA bypass	ECA-SV-MCA bypass	-----	One-sided occluded
5	Fujimura et al. (2014) [5]	28/F	Bilateral CCAs	STA-MCA(M4) and ECA-SV-MCA (M2) bypass with ICA ligation &	STA-MCA bypass with ICA ligation	10 months	11 months (L) 1 month (R)
6	Uozumi et al. (2015) [18] (for 6,7,8)	75/F	Bilateral CCAs	STA-MCA bypass with ICA ligation	ECA-RA-MCA bypass with ICA ligation &	31 months	13 months (L) 44 months (R)
7		38/M	Bilateral CCAs	ECA-RA-MCA bypass with internal trapping	ECA-RA-MCA bypass with ICA ligation &	26 months	70 months (L) 96 months (R)
8		44/M	Bilateral CCAs	ECA-RA-MCA bypass with ICA ligation &	ECA-RA-MCA bypass with ICA ligation	8 months	70 months (L) 62 months (R)
9	Our case	50/F	Bilateral CCAs	IMA-RA-MCA(M2) bypass with ICA ligation	ECA-SV-MCA(M2) bypass with ICA ligation &	96 months	6 months (L) 102 months (R)

Abbreviations: CCA, cavernous carotid aneurysm; F, female; M, male; L, left; R, right; B, bilateral; ECA, external carotid artery; SV, saphenous vein; MCA, middle cerebral artery; IMA, internal maxillary artery; RA, radial artery; STA, superficial temporal artery; &, the first side for operation

In summary, even if technically desirable from the surgical point of view in this case, decisions for bilateral CCAs should be balanced and individualized. Treatment approaches should consider the clinical presentation, size and enlargement of the aneurysm, etiology, developing thrombus or acute intraluminal thrombosis, osseous erosion, adequacy of the cross circulation and patient preference. To the best of our knowledge, this is the first case of bilateral CCAs that were successfully treated by two stages of high-flow EC-IC bypass, including an IMA-RA-MCA bypass followed by trapping. Nevertheless, long-term follow-up of outcomes is very important.

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Compliance with Ethical Standards The patient/next of kin/guardian has consented to submission of this case report to the journal.

Conflicts of interest None.

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