



Case Report

Opening of unusual vascular collaterals leads to early recanalization of a giant intracavernous carotid artery aneurysm following common carotid artery occlusion: A Case report and literature review

Ana M. Castaño-Leon, Jose F. Alen, Alfonso Lagares

Department of Neurosurgery, Research Institute i+12-CIBERESP, Hospital Universitario 12 de Octubre, Universidad Complutense de Madrid, Avda Cordoba SN, Madrid, Spain.

E-mail: *Ana M. Castaño-Leon - ana.maria.castano.leon@gmail.com; Jose F. Alen - jafalen@gmail.com; Alfonso Lagares - algadoc@yahoo.com



*Corresponding author:

Ana M. Castaño-Leon,
Department of Neurosurgery,
Research Institute i+12-
CIBERESP, Hospital
Universitario 12 de Octubre,
Universidad Complutense de
Madrid, Avda Cordoba SN,
Madrid, 28041, Spain.

ana.maria.castano.leon@gmail.com

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ABSTRACT

Background: Parent artery occlusion (PAO) with or without bypass surgery is a feasible treatment for large intracavernous carotid artery (ICCA) aneurysms. The ideal occlusion site (internal or common carotid artery [CCA]) and ischemic complications after PAO have received special attention since the description of the technique. Unfrequently, some patients can also develop unusual external carotid artery-internal carotid artery collateral pathways distal to the ligation site that can explain the failure to aneurysm size reduction.

Case Description: We describe a rare case of delayed refilling of a large ICCA aneurysm partially thrombosed which early recanalized after surgical ligation of the cervical CCA through an unusual collateral pathway.

Conclusion: Based on our experience, we recommend periodic long-term follow-up neuroimaging, especially in those cases where potential collateral branches have not been clearly identified in the preoperative studies.

Keywords: Aneurysm, Collateral pathways, Intracavernous carotid artery, Parent artery occlusion

INTRODUCTION

Intracavernous carotid artery (ICCA) aneurysms account for 3–5% of all intracranial aneurysms.^[35] Due to its location, the risk of mortality and severe morbidity in case of aneurysm rupture is low^[42] and spontaneous thrombosis of the aneurysm has been reported.^[21] For that reason, any proposed treatment has to have lower risk of complications than their natural benign history.^[20] In general, aneurysms with extension to the subarachnoid space, erosion of the sphenoid bone, evidence of enlargement, or progressive symptomatic cases should be treated due to the risk of ophthalmoplegia, orbital pain, visual acuity decline, SAH, carotid-cavernous fistula, or thromboembolic events.^[22]

In most centers, due to the challenging exposition of ICCA, these aneurysms are considered not amenable for clipping, and their management usually consists in coiling or flow-diverter placement. Parent artery occlusion (PAO) is a feasible alternative in those cases with

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intra-aneurysm or internal carotid artery (ICA) thrombosis, none or very wide neck, age or medical condition of the patient, and after failure of endovascular techniques. There are a number of alternatives according to the location (ICA or common carotid artery [CCA]), timing (abrupt ligation or prolonged closure with temporal clamps),^[24,29] and the method for occlusion (surgical ligation^[13,18] or endovascular procedures).^[9,34,36,39] In addition, in those patients with moderate to high risk of ischemic complications, PAO should be preceded by revascularization techniques.^[15,16,23]

The reported mortality and morbidity rates of surgical PAO range from 6–20% to 4–12%, respectively.^[28,29,33] The main causes of worsening or death after PAO are cerebral ischemia, thromboembolism, intracerebral hemorrhage, and *de novo* or enlargement of contralateral brain aneurysms.^[3,12] On the other hand, refilling of the ICCA aneurysm is a complication unfrequently reported after PAO.

Here, we present a rare case of a partially thrombosed ICCA aneurysm which early recanalized after surgical ligation of the cervical CCA via an unusual collateral pathway.

CASE REPORT

A 63-year-old woman, with arterial hypertension as the only relevant medical history, presented with a sudden binocular diplopia. Right 6th nerve palsy was evidenced on physical examination at admission. AngioCT revealed a right ICCA aneurysm with signs of partial thrombosis without evidence of rupture [Figure 1a]. Patient's symptoms worsened to complete cavernous syndrome 5-days later, and a new AngioCT showed progression of the thrombosis with discrete mass effect [Figure 1b], so steroids and antiplatelet therapy were initiated. Angiography on admission confirmed the presence of a giant aneurysm of the anterior portion of the right ICCA [Figure 2a-c]. Since endovascular techniques seemed inappropriate due to the partially intraluminal thrombosis and very wide neck, PAO was considered as the optimal treatment choice. A balloon test occlusion (BTO) was performed to evaluate the preoperative

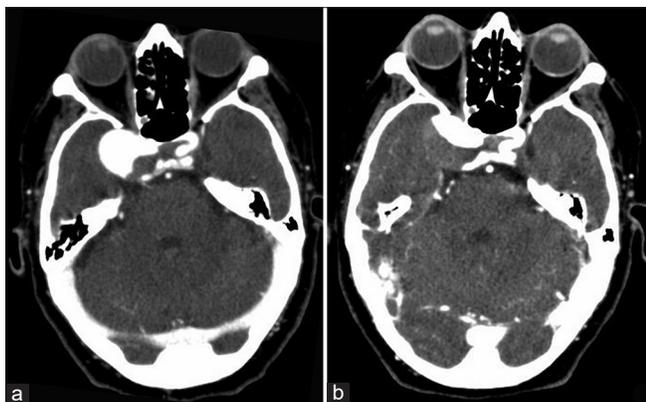


Figure 1: Admission AngioCT revealed a right intracavernous carotid artery (ICCA) aneurysm with signs of partial thrombosis without evidence of rupture (a). The patient experienced complete cavernous sinus syndrome in relation to progression of thrombosis of the ICCA aneurysm as a new AngioCT showed (b).

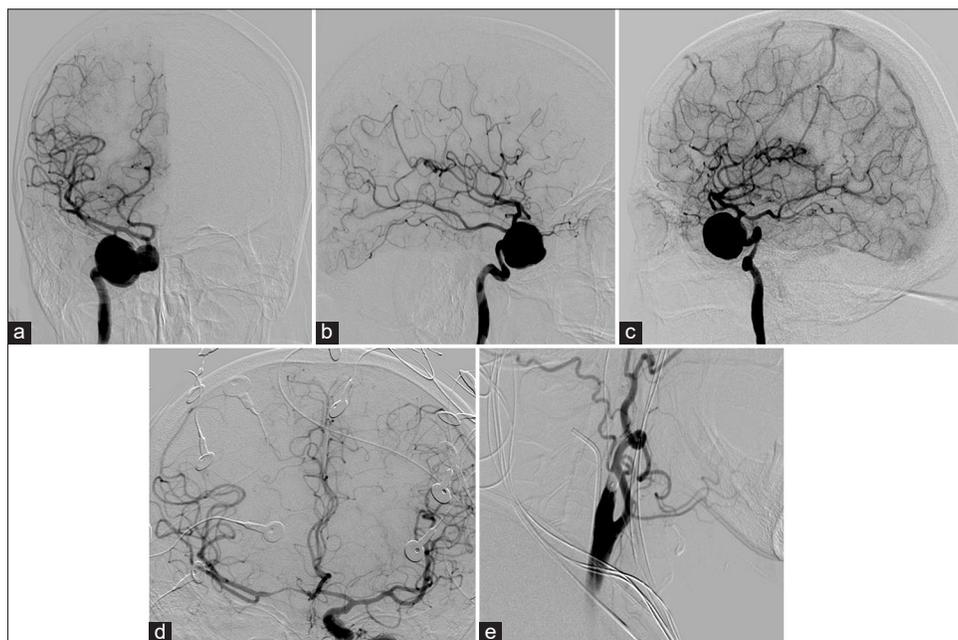


Figure 2: Right carotid angiography, anteroposterior (a) lateral (b) and oblique (c) projections, indicates a giant aneurysm (25 mm of main diameter, 9 mm neck width) at the cavernous segment of the right ICA. Left carotid angiography, anteroposterior projection, showing adequate collateral reserve via anterior communicating artery (d) and the aneurysm without filling of contrast after balloon occlusion at the proximal portion of the right ICA (e).

risk of ischemia without relevant findings and no evidence of retrograde filling of the aneurysm by anterior or posterior communicating arteries [Figure 2d and e]. Then, the patient underwent surgical ligation of the cervical CCA at the level of C6 with a 3/0-silk, and anticoagulation was started 3 days after surgery. Symptoms of cavernous sinus syndrome resolved with the exception of 6th nerve palsy during postoperative recovery. A cerebral magnetic resonance angiography (MRA) revealed decrease of the size of the aneurysm lumen and no ischemic lesions were detected [Figure 3a].

Nine months later, and without new neurological symptoms, signs of aneurysm growth were detected in a serial MRA [Figure 3b], and a new angiography was planned. Right CCA angiogram showed the therapeutic occlusion of the right ICA, but the right vertebral angiogram demonstrated proximal partial recanalization of the ICCA aneurysm. Collateral supply was explained from the external carotid artery (ECA) through the occipital artery with an inverse flow to the ICA [Figure 4a]. Therefore, surgical occlusion of the cervical ICA at the level of the ansa cervicalis of the hypoglossal nerve with a 0-silk was performed, and intraoperative Doppler ultrasound revealed no efferent flow in ICA. Postoperative new angiography confirmed the total occlusion of the aneurysm [Figure 4b]. After 6 years of follow-up, we have not detected refilling or thromboembolic events [Figure 5].

DISCUSSION

In ICCA aneurysms, due to the small number and caliber of branching vessels, PAO is a feasible option for treatment.^[7] The occlusion can be achieved by surgical ligation or by endovascular means. The treatment alternatives for symptomatic ICCA aneurysms with preservation of the parent artery include the obliteration of the aneurysm sac with coils^[15] or flow diversion. There are evidences suggesting that coiling can be conducted with a low complication

rate but the rate of recurrence is higher compared with PAO.^[38-40] New generation of flow-diverters stents has shown encouraging results but the safety due to frequent technical

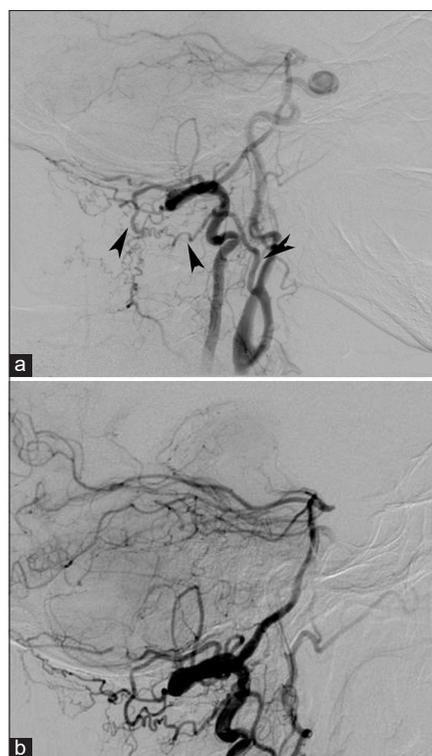


Figure 4: Nine months after common carotid artery ligation, a right vertebral angiography (lateral projection) showed partial recanalization of the intracavernous carotid artery (ICCA) aneurysm (a). Collateral supply was explained from external carotid artery through the occipital artery with an inverse flow to the internal carotid artery (ICA) (arrowheads). After cervical ICA surgical ligation, a new right vertebral angiography confirmed total occlusion of the ICCA aneurysm (b).

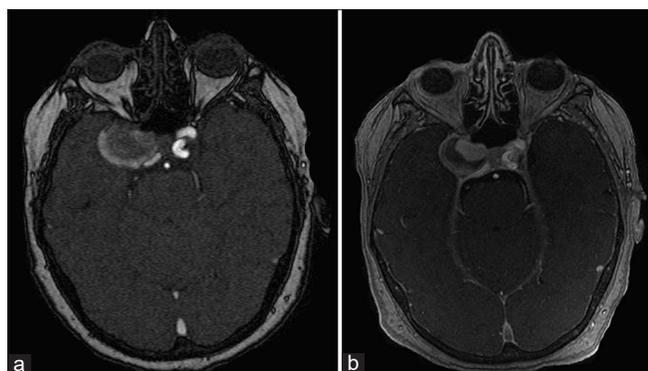


Figure 3: AngioMR (3D time of flight magnetic resonance sequence) showing reduction of the aneurysm size and minimal residual filling of the aneurysm immediately after the procedure (a). Eight months after the common carotid artery ligation, new AngioMR revealed signs of aneurysm recanalization (b).

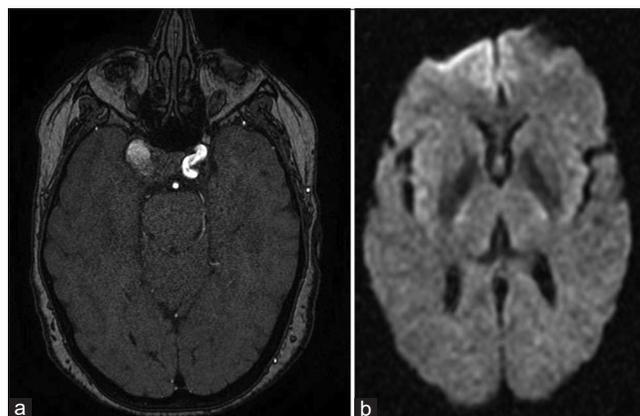


Figure 5: Last follow-up AngioMR showing the absence of flow in the right intracavernous carotid artery (a, 3D time of flight magnetic resonance sequence) and no evidences of ischemic lesions (b, diffusion-weighted imaging sequence).

complications (vasospasm, incomplete opening, and inability to place the flow-diverter due to vessel tortuosity) and long-term effectiveness is unknown.^[31,32] In our patient, we selected the surgical ligation because the selective occlusion of the proximal arterial lumen with coils has been associated with higher rate of recanalization.^[26] We also excluded aneurysm coiling and flow diversion due to the very wide neck of the aneurysm, arterial tortuosity, and the distortion of the intra-aneurysm thrombus by any endovascular treatment was not recommended. We also expected that the mass effect remains significant after endosaccular coiling.

Even when similar rates of success have been reported, there is controversy about the best site of PAO.^[13] Previous data suggested that occlusion of ICA maximally reduced the pressure and pulsation of the aneurysm minimizing the risk of persistent aneurysm refilling, and the rate of thromboembolism can be lower by decreasing the dead space between the occlusion site and the aneurysm.^[14] Nevertheless, we decided to occlude CCA because ischemic complications have been associated more often with ICA ligation.^[7,14,18,33] The described “safety” of CCA ligation is based on continued patency of the ICA either by anterograde filiform flow or retrograde ECA flow, which may minimize thromboembolic complications from clot formation in the ICA and maintain higher distal perfusion pressure during the perioperative period until collateral flow has been established.

Although ischemic and thromboembolic complications after PAO have received the attention, delayed refilling of ICCA aneurysm could happen. Conventionally, collateral pathway development is based on the proliferation of vasa vasorum induced by neoangiogenic factors after progressive atherothrombotic ICA occlusion.^[5,11,19] However, Numagami^[30] and Meguro^[25] described how an abrupt occlusion of ICA with coils might be also recanalized through vasa vasorum. One of the first descriptions of the opening of unusual collateral pathways was done by Pelz.^[10] He described in two patients with atherothrombotic occlusion of ICA how the ascending pharyngeal artery functioned as the collateral pathway that maintained the patency of proximally occluded ICAs. It was possible by the anatomic variation of the origin of the ascending pharyngeal artery from the ICA and the anastomoses of its muscular branches with branches originating from the occipital and vertebral arteries.

According to the previous data, two main types of recanalization after PAO may occur:

1. Retrograde recanalization: backflow of the ophthalmic artery,^[8,37] persistent trigeminal artery,^[6] posterior communicating artery, and bypass arteries.^[4] Since the risk of aneurysm rupture is believed to be lower after retrograde recanalization and there was no evidence of aneurysm filling by anterior or posterior communicating

arteries in the first angiogram, we did not consider to trap the aneurysm by distal occlusion.

2. Anterograde recanalization: vasa vasorum and collateral embryonal pathways between the ICA and ECA (in the case of CCA, meningohypophyseal branches, and inferior lateral trunk). Recently, Wang^[41] described three pathways of anterograde ICA supply after atherothrombotic CCA occlusion in 16 patients studied by transcranial Doppler, angiography, or MRA. In 8 of 16 patients, pathway 1 (ipsilateral vertebral artery [VA]-occipital artery-ECA-ICA) was noticed. In the rest of patients, ipsilateral thyrocervical trunk or costocervical trunk through ascending cervical artery or deep cervical artery-occipital artery (pathway 2) or contralateral ECA-contralateral superior thyroid artery-ipsilateral superior thyroid artery (pathway 3) explained retrograde flow in the ECA. According to the author, the basic driving force that leads to the formation of collateral circulation is the decrease in blood pressure in ICA after CCA occlusion. Then, blood flow in adjacent arteries may be drawn reversely to the distal segment (“ICA steal”).

In our opinion, some of the mechanisms that explain the lower risk of thromboembolism and ischemia after CCA occlusion are the same that influence the opening of unusual collaterals. The importance of the evaluation of proximal intracranial ICA branches during the planification of the site of PAO was described by Allen.^[2] He focused on the potential anastomosis between the ECA and branches of the petrous, cavernous, and lacerum portions of the ICA. These proximal branches can explain false negative BTO as the temporal occlusion is usually done at C1-2 level and sources for persistent filling and growth of ICA aneurysms. They recommend performing a second more proximal BTO before deciding where to interrupt the patent artery.

Even with the mentioned multiple sources recanalization of ICCA aneurysms after PAO is a rare complication. If we review the literature of previously reported cases, we identify only seven studies [Table 1]. Opposite to our case, in the vast majority, the collateral pathway was established after revascularization surgery with delayed proximal cervical ICA occlusion.^[1,17,27] We speculate that flow reduction in the right CCA after PAO led to potential anastomosis between the occipital artery of the ECA and meningeal branches from the right VA in our patient. However, we cannot confirm this mechanism because we did not perform any assessment of cerebral blood flow after ICA occlusion. An alternative explanation is the effect of a giant aneurysm in the parent artery flow that often is distorted/attenuated by the aneurysm and may have a favorable effect in the development of collateral pathways.^[24]

Table 1: Review of previous cases reporting recanalization of ICCA aneurysm after PAO.

Author, year	Number of patients	CCA aneurysm	Symptoms	Treatment	Collateral pathway	Time to revascularization	Treatment of the aneurysm refilling
Drake, 1994	1 (unknown)	Unknown	Unknown	Detachable balloon at ICA	Retrograde, persistent trigeminal artery	Unknown	Clipping of persistent trigeminal artery
Barnett, 1994	1 (72 years, man)	Right, CCA, large	Progressive abducens nerve palsy and oculomotor nerve palsy	BTO-clinical-, SPECT+ STA-MCA+ detachable balloon at petrous, and cervical ICA	Retrograde, bypass	2 months	None
Numagami, 1999	1 (69 years, Female)	Right, CCA, 27×20×22 mm	Blepharoptosis and diplopia	BTO-SPECT/SEP – detachable balloons at two points (proximal to aneurysm and within cervical ICA)	Retrograde, right ophthalmic artery (vasa vasorum)	6 months	None (revascularization only affected to the parent artery)
Li, 2004	50 years, Female	Right, CCA	Epistaxis	Parent artery occlusion with two detachable balloons	Retrograde, persistent trigeminal artery	4 months	Embolization of persistent trigeminal artery
	51 years, Female	Right, CCA	Eyelid drop	None	Retrograde, persistent trigeminal artery	At diagnosis	Retrograde, persistent trigeminal artery
Kagawa, 2007	1 (21 years, Female)	Right, CCA, 20×15 mm	Abducens nerve palsy	BTO-SPECT+ STA-MCA bypass+ cervical ICA ligation	Antegrade, right ECA, and VA	11 days	Endovascular coiling of aneurysm and recanalized ICA
Abe, 2011	8 (Unknown)	Unknown	Unknown	STA-MCA or RA-M2 depending of BTO-clinical+ ICA ligation	Antegrade from the ECA (5), backflow of RA-M2 graft (2), slack at the ligation site (1)	Unknown	Endovascular coiling of the parent artery
Nagatani, 2015	1 (67 years, Female)	Right, supraclinoid ICA, 18 mm	Reduced visual acuity	BTO-clinical+ ECA-ICA bypass+STA-MCA bypass+ cervical ligation ICA	Antegrade, anastomosis between anterior meningeal branching from VA and the APA from the ICA	4 years	None (patient denied to be treated)

SPECT: Single-photon emission computed tomography, SEP: Somatosensory evoked potentials, VA: Vertebral artery, ICA: Internal carotid artery, CCA: Cavernous carotid artery, APA: Ascending pharyngeal artery, RA: Radial artery

CONCLUSION

Delayed refilling of ICCA aneurysms based on unusual collateral pathway opening after PAO may occur. Periodic

long-term follow-up by neuroimaging is recommended, especially in those cases where potential collateral branches of have not been clearly identified in the preoperative studies.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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