Rare Recurrence of Traumatic Carotid Cavernous Fistula after Parent Artery Occlusion—Report of Two Cases and Review of Literature

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Abstract

Keywords
► carotid cavernous fistula
► parent artery occlusion
► posttraumatic
► recurrence
► vasa vasorum

The authors present two cases of recurrence of traumatic carotid cavernous fistula (CCF) following complete exclusion by parent artery occlusion (PAO). In both cases, the fistula recurred through the development of indirect CCF and reconstitution of the occluded artery through the development of vasa vasorum or simple recanalization of the parent artery. The cavernous venous sac was patent in both cases. The patent cavernous sac along with the inflammatory or angiogenetic factors might have induced dural neovascularization leading to the development of indirect CCF. These factors along with ischemia of the arterial wall secondary to the steal phenomenon due to persistent shunt flow would have triggered the development of vasa vasorum. Thus the cavernous sac embolization may have to be considered in addition to PAO when PAO is planned as a therapeutic option for direct CCF.

Introduction

Carotid cavernous fistula (CCF) is an abnormal direct or indirect communication between the intracranial part of internal carotid artery (ICA) and or external carotid artery (ECA) with the cavernous sinus (CS). Based on the etiology, the direct CCF is classified as traumatic or spontaneous. Endovascular treatment of direct CCF includes several options such as embolization using balloons or coils, sealing of the rent using covered stents or newly introduced flow diverters.[1,2] Endovascular procedure has a high success rate of 88 to 98% with no to low complication rate.[3] Endovascular parent vessel occlusion (PAO) is an alternative option done in selected situations in which reconstruction of the artery is difficult.[1] The recurrence rate in the treatment of traumatic CCF varies between 10 and 15%, and this is primarily observed in cases treated with detachable balloon embolization. The failure may be attributed to premature balloon deflation, rupture, or migration.[1,3] However, there are only very few instances of recurrence of CCF following endovascular PAO.[4,5] We present two cases of CCF with recurrence following complete exclusion by PAO. The factors predisposing to this unusual occurrence are discussed.

Case Report

Case 1

A 55-year-old man was involved in a road traffic accident (RTA) and sustained a head injury, which required intensive case unit (ICU) admission for management. Computed tomography (CT) showed fractures involving the left temporal bone and left the orbital wall. The patient developed insidious onset redness and proptosis in the left eye 2 weeks after RTA, which was suspected to be CCF. Magnetic resonance imaging (MRI) showed the prominence of the left CS and bilateral superior ophthalmic veins (SOVs). Digital subtraction angiography (DSA) confirmed the presence of direct CCF in left cavernous ICA and a pseudoaneurysm of the left middle meningeal artery (MMA) (►Fig. 1A, B). He underwent endovascular intervention with PAO for left direct CCF and coil embolization of left MMA pse udoaneurysm (►Fig. 1C, D).
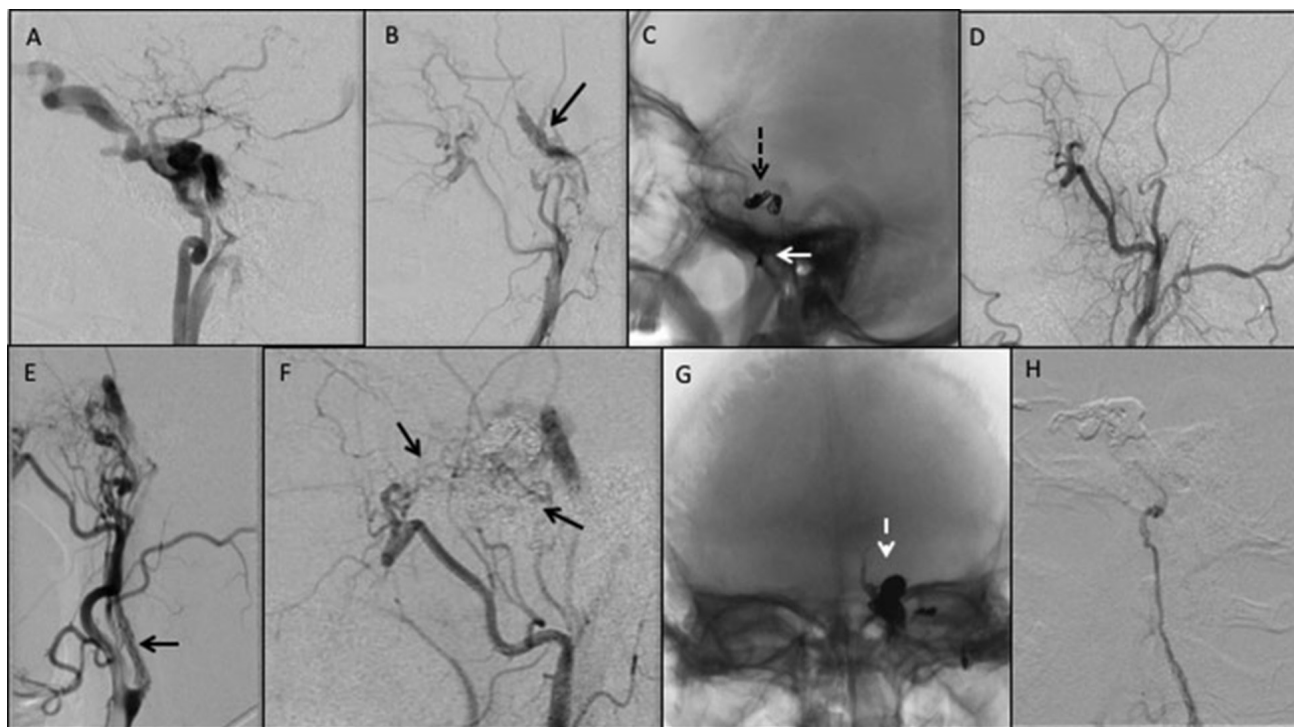
Fig. 1 Angiogram and intervention during the initial presentation (**A–D**). Left ICA angiogram (**A**) lateral view shows direct CCF with anterior, posterior, and cortical venous reflux. Left ECA angiogram (**B**) lateral view shows traumatic pseudoaneurysm of the MMA (black arrow). Radiograph of skull (**C**) AP view shows coil mass of PAO (dashed black arrow) and MMA pseudoaneurysm embolization in the left middle cranial fossa (white arrow). Left CCA angiogram postembolization (**D**) shows a complete cessation of the shunt and no opacification of the left ICA. Angiogram and intervention during the recurrence (**E, F**). Left CCA angiogram (**E**) lateral view shows recurrence of the left CCF with recanalization of the ICA via vaso vasorum (black arrow). Left ECA angiogram (**F**) shows distal IMA and MMA branches contributing to the fistula (black arrows). Radiograph of the skull (**G**) shows coil mass and Squid cast in the left parasellar region from transvenous embolization of the left cavernous sinus (dashed white arrow). Left CCA angiogram postembolization (**H**) shows complete cessation of the shunt.

PAO was performed using detachable coils, and an immediate postintervention check angiogram revealed complete cessation of the shunt (►**Fig. 1C, D**). He was asymptomatic for the next 5 months when his symptoms of left eye proptosis and redness recurred. Check DSA revealed recurrence of the CCF on the left side (►**Fig. 1E, F**). The arterial feeders were from distal branches of left internal maxillary (IMA), clival branches of left ascending pharyngeal arteries, and left MMA (►**Fig. 1F**). Minimal supply from distal branches of right IMA and meningohypophyseal branches of right ICA was also noted. The left ICA showed recanalization through vasa vasorum, which also contributed to the fistula (►**Fig. 1E**). Transvenous embolization of the left CS was performed with coils and Squid 18 liquid embolic device (Emboflu), achieving complete obliteration of the sac and cessation of the shunt (►**Fig. 1G**). Check angiogram done 5 days postintervention showed no evidence of residual fistula. The patient remained asymptomatic until the last clinical follow-up at 2 years.

Case 2

A 25-year-old man was involved in an RTA and sustained maxillofacial and head injuries. One day later, he developed pulsatile proptosis and chemosis in the right eye. CT scan showed prominent bilateral SOV and mandibular fracture, and DSA examination revealed a right direct CCF (►**Fig. 2A**). He underwent detachable balloon embolization of the venous sac; however, because of the protrusion of the balloon

through the rent into the parent artery and resultant flow compromise, PAO was considered to avoid thromboembolic complications. PAO was performed using detachable coils (►**Fig. 2B–D**). Postprocedure period, he had good symptomatic improvement with subsequent resolution of his proptosis and chemosis. On 17th postprocedure day, he developed recurrence of right eye proptosis and chemosis that gradually progressed. Follow-up DSA showed recurrence of his right CCF with supply anterogradely from the recanalized right ICA as well as from the terminal branches of bilateral IMA (►**Fig. 2E, F**). Transvenous coiling of the right CS was done resulting in complete cessation of the shunt (►**Fig. 2G, H**).

Discussion

The mainstay of treatment of CCF is by endovascular approaches, and the choice of endovascular treatment is dependent on various factors such as the angioarchitecture of the fistula, the size of the rent, degree of the shunt, operator's preference, and institutional experience.

The recurrence rate following treatment of direct CCF has been very low, as shown by various studies.[3] Most of the recurrence occurred with the use of the detachable balloons, caused by premature deflation or rupture and occurring within days of treatment. Recurrence after ICA occlusion/PAO has been seen due to reverse filling of the fistula through the distal ICA.[5] Studies have shown recurrence
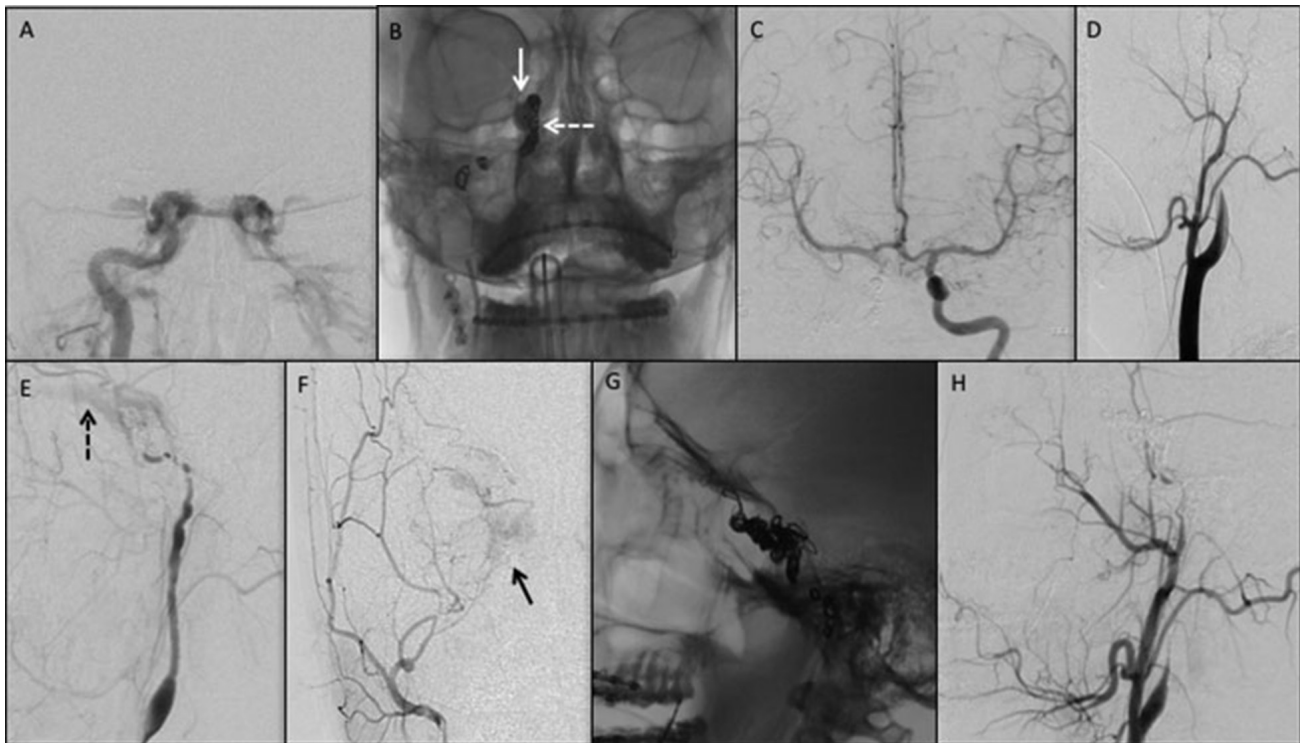
Fig. 2 Angiogram and intervention during initial presentation (**A–D**). Right ICA angiogram (**A**) AP view shows direct CCF with anterior and posteroinferior venous drainage. Radiograph of skull (**B**) AP view shows detachable balloon in the cavernous sinus (white arrow) with coil mass of PAO in the right para sellar region (dashed white arrow). Left ICA angiogram postembolization (**C**) AP view and right CCA angiogram (**D**) lateral view show a complete cessation of the shunt. Angiogram and intervention during the recurrence (**E, F**). Right CCA angiogram (**E**) lateral view shows recurrence of the left CCF (dashed black arrow) with recanalization of the ICA. Right ECA angiogram (**F**) AP view shows distal IMA branches contributing to the fistula (black arrow). Radiograph of the skull (**G**) lateral view shows coil mass in the right paraseller region from transvenous embolization of the right cavernous sinus. Right CCA angiogram (**H**) shows a complete cessation of the shunt and no opacification of the right ICA.

after coil embolization due to less coil packing density[6] and after covered stent placement due to endoleak caused by a mismatch in diameter and length between the graft and the fistula.[7]

Other rare causes of recurrence of CCF after successful treatment include the development of indirect CCF and recanalization of the occluded parent artery through vasa vasorum. Indirect CCF has been reported following parent artery occlusion, balloon embolization, and covered stent placement for direct CCF (►**Table 1**). The authors have attributed this phenomenon to the angiogenesis triggered by angiogenic factors such as vascular endothelial growth factor (VEGF), which are expressed due to venous hypertension in a high flow CCF or during healing of lacerated dura/CS wall.[4,8–10] This hypothesis is supported by the results from animal studies, which showed increased VEGF expression promoted by hypoperfusion, sinus hypertension, and sinus thrombosis, leading to the development of dural arteriovenous fistula in rat models.[8]

There are only three reports of recanalization of occluded ICA through vasa vasorum following parent artery occlusion in the literature (►**Table 2**), in which all the cases were related to the PAO in giant cerebral aneurysms.[11–13] Vasa vasorum are small vessels located in the wall of the arteries responsible for microcirculation and providing nutrition. It is postulated that the development of vasa vasorum is induced or influenced by the ischemia, nutritional or metabolic demand of the vessel wall, and/or the organ supplied

by the occluded artery.[14] Single-photon emission computed tomography (SPECT) study had indeed shown that there are reduced cerebral blood flow and altered vasodilatory capacity in the ipsilateral brain, which could act as trigger to the development of vasa vasorum.[11]

We present two cases of recurrence following the endovascular treatment of traumatic CCF. In both the cases, the fistula recurred through the development of indirect CCF. In addition to the dural feeders, the reconstitution of the occluded artery was also observed through the development of vasa vasorum or simple recanalization of the parent artery. The cavernous venous sac was patent in both the cases: in the first patient, the sac was not embolized, and in the second patient, there was deflation of the detachable balloon deployed in the CS. In high-flow direct CCF, the dural arteries may contribute to the CCF that may not be recognized in the initial angiography. These latent fistulae become apparent when the ICA is occluded without the embolization of the CS sac. The cavernous sac then acts as a persistent sump, keeping the fistula patent following the PAO. However, a mere persistence of the dural shunts following PAO cannot explain the observations in our cases. In our case, the indirect CCF was in the proliferative phase, suggesting that dural neovascularization might have been induced by the inflammatory or angiogenetic factors.[4,8–10] These factors would have played a role in the development of vasa vasorum, leading to the recanalization of the occluded artery. Ischemia of the arterial

Table 1 Previous case reports or recurrence of CCF posttreatment

	Symptoms days after RTA	1st DSA	1st treatment	Recurrence (days)	Angioarchitecture	Final treatment
Terada et al (2002)[10]	180	Right traumatic CCF	Balloon embolization of the cavernous sac	150	Right type D indirect CCF fed by meningeal branches of bilateral ICA and right IMA	Transvenous coiling of the fistulous portion and intercavernous sinus
Lv et al (2008)[9]	330	Right traumatic CCF	5- × 19-mm covered stent	180	Right indirect CCF fed by bilateral MMA and accessory meningeal arteries	Transarterial (ECA) Onyx embolization of fistulas
Yoshino et al (2011)[4]	2–3	Right traumatic CCF	PAO with EC-IC bypass	10	Right indirect CCF fed by branches of accessory meningeal and infraorbital artery	Transvenous coiling of the cavernous sinus sac

Abbreviations: CCF, carotid cavernous fistula; DSA, digital subtraction angiography; ECA, external carotid artery; EC-IC bypass, external carotid internal carotid bypass; IMA, internal maxillary artery; MMA, middle meningeal artery; PAO, parent artery occlusion.

Table 2 Previous case reports or recanalization of ICA post parent artery occlusion

	Symptoms	1st DSA	1st treatment	Recurrence (days)	Angioarchitecture
Numagami et al (1999)[11]	Right ptosis and diplopia	Giant cavernous ICA an aneurysm	Parent artery occlusion with detachable balloon	180	Recanalization ICA by vaso vasorum
Meguro et al (2011)[12]	Left hemisphere stroke	Left ICA dissection	Left ICA occlusion with GDC coils	1,140	Recanalization ICA by vaso vasorum
Clarencon (2011)[13]	Headache and right 3rd cranial nerve palsy	Giant saccular aneurysm in right cavernous ICA	PAO	1,825	Recanalization ICA by vaso vasorum

Abbreviations: DSA, digital subtraction angiography; GDC, Guglielmi detachable coil; ICA, internal carotid artery; PAO, parent artery occlusion.

wall secondary to the steal phenomenon due to persistent shunt flow also would have triggered the development of vasa vasorum.[14]

Our observations suggest that the cavernous sac embolization may have to be considered in addition to PAO when PAO is planned as a therapeutic option for direct CCF. Coiling of the sac encourages thrombus formation within the cavernous sac that would eventually lead to complete obliteration when slow or persistent residual flow is present from ICA or dural ECA branches.

Contributions of Authors
- SK: Data collection, data analysis, data interpretation, manuscript preparation, critical revision
- VM: Manuscript preparation, critical revision
- JER: Manuscript preparation, critical revision
- SKK: Concept and design, data analysis, data interpretation, manuscript preparation, critical revision

Conflict of Interest
The authors have no personal or financial conflict of interest to disclose.

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