

Traumatic Carotid Cavernous Fistula with a Connection between the Supraclinoid Internal Carotid Artery and Cavernous Sinus via a Pseudoaneurysm Presenting with Delayed Life-threatening Epistaxis

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Traumatic carotid cavernous fistula (CCF) presents most commonly as a direct connection between the cavernous segment of internal carotid artery (ICA) and cavernous sinus (CS), and often accompanies basal skull fracture (BSF). The most frequently reported signs and symptoms are proptosis, chemosis, and bruit. Epistaxis is uncommon symptom of CCF. A 63-year-old man with a history of BSF due to blunt head trauma eight months previously was referred to our emergency room for massive epistaxis. Cerebral angiography demonstrated a high-flow CCF with a connection between the supraclinoid portion of left ICA and CS via a pseudoaneurysm, suggesting the life-threatening epistaxis had been caused by high-flow shunting, through the gap created by the BSF. After the complete obliteration of the pseudoaneurysm arising from the supraclinoid ICA by stent-assisted coil embolization, the CCF was no longer evident and epistaxis ceased. To the best of our knowledge, this is the first case of a traumatic CCF with a connection between the supraclinoid ICA and CS via a pseudoaneurysm, presenting with delayed life-threatening epistaxis.

Keywords: aneurysm, coil, fistula, trauma

Introduction

Traumatic carotid cavernous fistula (CCF) presents most commonly as a direct connection between the cavernous segment of internal carotid artery (ICA) and cavernous sinus (CS), forming a high-flow fistula. In these cases, the probability that the fistula heals spontaneously by thrombosis is low. Traumatic CCF often accompanies basal skull fracture (BSF), and has an incidence rate in this background of 3.8% of patients with BSF.¹⁾ The most frequently reported signs and symptoms are proptosis, chemosis, and bruit. Furthermore, as the condition progresses, it can lead to blindness and in rare cases can result in paralysis, unconsciousness, and even death.²⁾ We present a rare case of traumatic CCF with a connection

between the supraclinoid ICA and CS via a pseudoaneurysm that presented with delayed life-threatening epistaxis.

Case Report

A 63-year-old man was referred to our emergency room with massive epistaxis. Continuous bleeding of 500cc or more was observed, and despite nasal packing with epinephrine soaked gauze, it did not stop. He had a history of BSF, which had been diagnosed in our hospital eight months previously, caused by blunt head trauma. However, at that time, the patient denied any further examination or treatment and was transferred to another hospital. At this presentation, urgent computed tomography (CT)-angiography depicted a suspicious CCF without evidence of an acute intracranial lesion (Fig. 1). Consent was obtained from the patient's family for emergency cerebral angiography and possible endovascular intervention.

No obvious leakage of contrast medium was detected by angiography from either external carotid artery. Left ICA angiography showed a high-flow CCF with a connection between the supraclinoid ICA and CS via a pseudoaneurysm, measuring 7.2×6.5 mm, which was assumed, based on its location, to originate from the anterior choroidal artery. Cortical venous reflux was observed via the petrosal vein in the left posterior fossa (Fig. 2). Accordingly, we suspected the life-threatening epistaxis has been caused by high-flow shunting through a gap created by the BSF. Balloon test occlusion (BTO) was performed to assess the risk of stroke following possible total trapping of the ICA, but was not tolerated for 20 minutes with decreased visual acuity (Fig. 3). The balloon was positioned just proximal to the neck of pseudoaneurysm because it was located so close to the ICA terminus. Therefore, this poor tolerance to BTO might be resulted from steal phenomenon of the right ICA flow through the anterior communicating artery into CCF. Nonetheless, coil embolization of pseudoaneurysm was planned in order to block the fistula and preserve the parent artery. The patient was fully awake during the procedure. Systemic heparinization and antiplatelet premedication were not performed. After establishing optimal working angles, coil embolization was initiated. At first, the balloon-assisted technique was applied in order to prepare for a possible intraprocedural pseudoaneurysmal rupture and to

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Received: May 7, 2016; Accepted: August 18, 2016

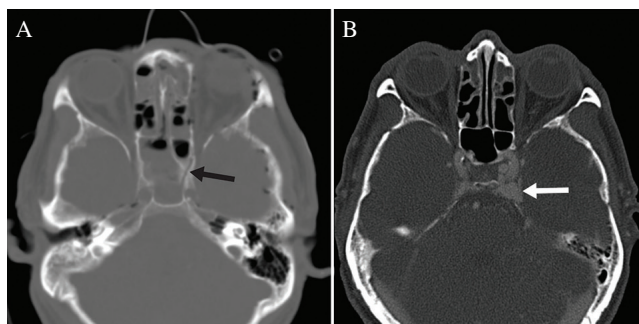


Fig. 1 (A) Bone window image of a brain CT scan after trauma showing a linear fracture (arrow) over the left side of the sellar floor. (B) Contrast enhanced CT-angiograph obtained 8 months later at readmission showing contrast leakage into the left CS (arrow), which was regarded suspicious for CCF.



Fig. 2 Early (A) and late (B) arterial phase of a left ICA angiogram, anteroposterior (A) and lateral (B) projections, showing high-flow shunting from the distal ICA to the CS via a pseudoaneurysm (black arrows) arising from the supraclinoid ICA (white arrows). The arrowheads indicate cortical venous reflux in the left posterior fossa via the petrosal vein.

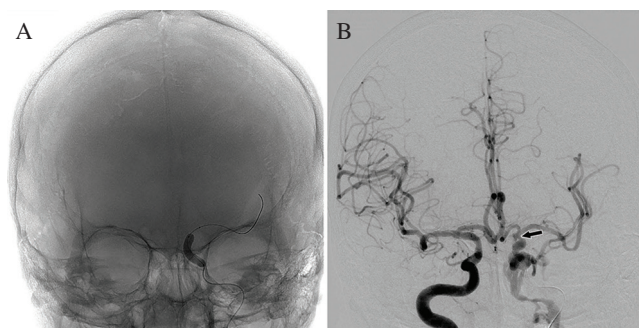


Fig. 3 Unsubtracted (A) and subtracted (B) images showing BTO, which was performed using a 4 × 15 mm balloon. Right ICA injection with complete occlusion of the left ICA by intraluminal balloon fills both hemispheres and shows more clearly the point of fistula (arrow).

achieve attenuated packing of the pseudoaneurysm. At the end of the procedure, a Neuroform stent was deployed in order to fill the coils in the neck of the pseudoaneurysm and divert the blood flow toward the aneurysm. The pseudoaneurysm was completely occluded by the employment of ten detachable coils and the fistula was completely blocked, although some coils protruded into the CS (Fig. 4).

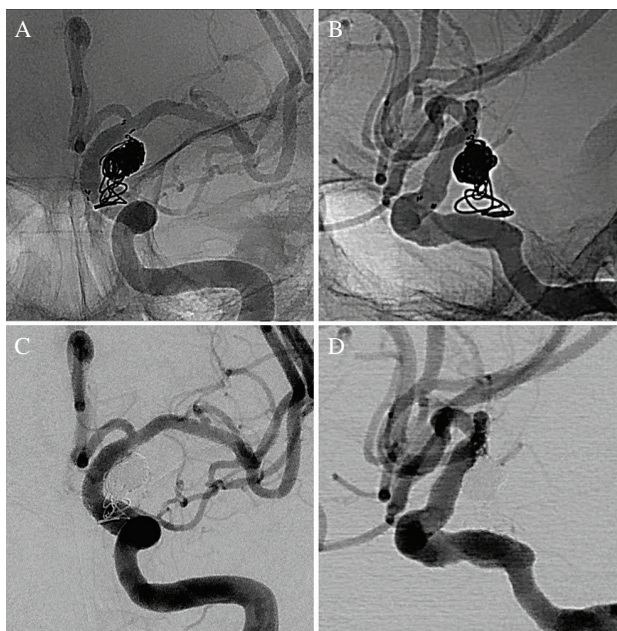


Fig. 4 Unsubtracted (A, B) and subtracted (C, D) images obtained immediately after successful stent-assisted coil embolization of the pseudoaneurysm. Anteroposterior (A, C) and lateral (B, D) views of the left ICA angiogram, revealing normal ICA patency, obliteration of the pseudoaneurysm, and complete blockage of the fistula.

Bleeding stopped immediately after the procedure, and the post-operative period was uneventful. He was discharged on the third post-operative day and remained asymptomatic at six months after discharge. Long-term follow-up to assess the coil compaction and recanalization is waiting to be done.

Discussion

Traumatic CCF usually presents as a direct connection between the cavernous segment of an ICA and the CS, forming a high-flow fistula. In other words, it is caused by a tear in the weak cavernous portion of the ICA. In our case, the CCF was formed by a connection between the supraclinoid portion of the left ICA and CS via a pseudoaneurysm, which was presumed, based on its location, to have originated from the branching site of the anterior choroidal artery. The following possible mechanism may explain the occurrence of a pseudoaneurysm of the intradural ICA after a nonpenetrating head injury and the delayed life-threatening epistaxis: Our patient had a BSF in the region of the left side of the sellar floor in proximity to the origin of the left anterior choroidal artery. The origin of the anterior choroidal artery was torn by overstretching after the movement of the brain during the impact of head injury with a simultaneous laceration of the dural covering of the CS, and the pseudoaneurysm arising from torn ICA gradually developed to fill the retrocarotid space and connected to the CS through the dural defect to form the high-flow CCF. This high-pressure shunting resulted in a large amount of bleeding through the gap created by BSF, and thus, led to the delayed life-threatening epistaxis.

A supraclinoid CCF caused by head trauma is rare. Just over ten cases have been disclosed by a systemic review of literature

Table 1 Summary of reported cases of posttraumatic intradural internal carotid artery-cavernous sinus fistula

Authors (Year)	Age (years)/ Sex	Presentation symptom related to CCF	Time from trauma to treatment	Associated pseudoaneurysm	Treatment	Outcome
Reddy et al. (1981) ⁸⁾	14/M	Proptosis, bruit, blindness	5 months	Yes	ICA ligation	Successful
Komiyama et al. (1991) ⁹⁾	42/M	Proptosis, chemosis, bruit	10 days	Yes	Transarterial balloon embolization (ICA preservation)	Poor due to pseudoaneurysm rupture
Masana et al. (1992) ¹⁰⁾	28/M	Proptosis, chemosis, bruit	4 months	Yes	Aneurysm neck clipping	Successful
Tytle et al. (1995) ¹¹⁾	46/F	Proptosis	15 years	Yes	Aneurysm neck clipping	Residual fistula from posterior circulation
Kinugasa et al. (1995) ¹²⁾	24/M	Proptosis, chemosis, bruit	1 month	No	Transvenous coiling	Complete occlusion
Fu et al. (2002) ¹³⁾	16/M	Chemosis	1 month	Yes	Aneurysm neck clipping	Successful
Weaver et al. (2003) ¹⁴⁾	42/M	Proptosis, chemosis	1 month	Yes	Transarterial endosaccular coiling	Complete occlusion
Oran et al. (2004) ¹⁵⁾	30/M	Proptosis, chemosis	2 years	Yes	Transarterial ICA trapping	Complete occlusion
Gandhi et al. (2009) ¹⁶⁾	14/M	Nonspecific	7 days	Yes	Transvenous coiling	Complete occlusion
Chen et al. (2009) ¹⁷⁾	37/M	Proptosis, chemosis, bruit	1 month	Yes	Transarterial endosaccular coiling	Complete occlusion
Zhao et al. (2012) ¹⁸⁾	40/M	Chemosis, bruit	2 months	Yes	Transarterial endosaccular BAC and Onyx embolization	Complete occlusion
Karanam et al. (2014) ¹⁹⁾	40/M	Proptosis, chemosis	4 days	Yes	Transarterial endosaccular coiling	Complete occlusion
Jinbo et al. (2015) ²⁰⁾	27/M	Blurred vision, bruit	7 months	Yes	Transarterial endosaccular coiling	Complete occlusion
Present case	63/M	Massive epistaxis	8 months	Yes	Transarterial endosaccular SAC	Complete occlusion

BAC: balloon assisted coiling, CCF: carotid cavernous fistula, F: female, ICA: internal carotid artery, M: male, SAC: stent assisted coiling.

in Pubmed (Table 1). All 13 patients except one were found to have a pseudoaneurysm connecting the intradural ICA and CS, like our case. Since 2000, endosaccular coiling of pseudoaneurysm via transarterial route was first attempted in most of the case including ours, and the results were excellent. Although the pseudoaneurysmal wall lacked the normal support structure and could be fragile, endosaccular coiling via transarterial route appears to be a feasible, effective and minimally invasive option for the treatment of traumatic CCF with a connection between the intradural ICA and CS via a pseudoaneurysm with a small ostia. Epistaxis is an uncommon symptom of CCF, and is usually accompanied by eye symptoms.³⁾ From Table 1, we found that all the patients were presented with eye symptoms, except our case. To the best of our knowledge, this is the first case of a traumatic CCF with a connection between the supraclinoid ICA and CS via a pseudoaneurysm to present with delayed life-threatening epistaxis, without eye symptoms.

Endovascular treatments of a pseudoaneurysm of the ICA include sacrifice or the preservation of the ICA. When BTO is well-tolerated, trapping or occlusion of the parent artery is options. However, 5%–22% of patients that tolerate BTO develop ischemic complications.⁴⁾ Therefore, preserving the patency of the carotid artery as much as possible is a more desirable goal. Coil embolization or stent-assisted embolization of ICA pseudoaneurysms have been used to preserve ICA patency. However, the lack of a true wall in these pseudoaneurysms and the absence of normal healthy tissue within the retrocarotid space may result in enlargement and/or rupture.⁵⁾ Graft-stents are being used more so to overcome these problems of treating ICA pseudoaneurysms.^{6,7)} However, the use of the stents intracranially is limited by their large profiles and inflexibilities, which make it difficult to negotiate tortuous vascular segments at the skull base. In the present case, we considered that the passage of a graft-stent

was not possible, because the pseudoaneurysm was too distally located. Furthermore, because eight months had elapsed since trauma, we consider fibrosis in the pseudoaneurysmal wall would have gradually progressed to form a relatively rigid wall, and thus, we chose stent-assisted coil embolization to occlude the aneurysm and preserve the parent artery.

In conclusion, to the best of our knowledge, this is the first case of a traumatic CCF with a direct connection between the supraclinoid ICA and CS via a pseudoaneurysm to present with delayed life-threatening epistaxis, without eye symptoms. Traumatic CCF should be considered in the differential diagnosis when a patient with a history of blunt head trauma sustains massive epistaxis.

Conflicts of Interest Disclosure

There is no conflict of interest with any financial organization regarding the material discussed in the manuscript.

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