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Multiple Unilateral Traumatic Carotid-Cavernous Sphenoid Sinus Fistulas with Associated Massive Epistaxis: A Consequence of Parkour

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ABSTRACT

Background: Traumatic carotid-cavernous fistulas (CCFs) present the clinician with diagnostic and surgical challenges. Extension of a CCF into the sphenoid sinus presents additional management difficulties. Endovascular interventions using various thrombogenic materials such as balloons, coils, or liquids are effective treatment strategies. Ideally, these techniques are used to obliterate the fistula while maintaining the patency of the parent artery.

Case Report: We present a rare case of traumatic carotid-cavernous sphenoid sinus fistulas complicated by multiple tears in the internal carotid artery with direct communication to the cavernous and sphenoid sinus. As a result, the patient developed massive epistaxis requiring emergent endovascular intervention. A total of 87 detachable coils were placed into the cavernous and sphenoid sinuses via transarterial and transvenous routes in a staged procedure, resulting in complete obliteration of the patient's multiple fistulas.

Conclusion: To our knowledge, this is the first reported case of multiple fistulous tears in the internal carotid artery with extension to the cavernous and sphenoid sinus. This report emphasizes the importance of early diagnosis and neurosurgical intervention.

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INTRODUCTION

Parkour is the discipline of free running originally developed in France in the early 19th century that involves moving from one point to another with maximum efficiency and speed and utilizing various acrobatic maneuvers to overcome obstacles. The sport has more recently been popularized in urban settings and is almost always practiced without protective gear. This is a case study of a young practitioner, or *traceur*, of parkour who sustained a traumatic brain injury that resulted in multiple unilateral carotid-cavernous fistulas (CCFs) extending into the sphenoid sinus. Traumatic CCFs occur in approximately 0.2% of closed head injuries[1] and present the clinician with both diagnostic and surgical challenges. Endovascular intervention with coils or other thrombogenic materials is the preferred method for treatment with the goal of obliterating the fistulas while maintaining patency of the internal carotid artery.

CASE REPORT

A 25-year-old *traceur* was performing the basic parkour maneuver known as *saut de détente*, leaping from rooftop to rooftop, when he plunged 3 stories onto a concrete alley. The patient was comatose when he arrived at the hospital with an enlarged, unreactive left pupil. Computed tomography (CT) scans revealed a ruptured spleen, multiple facial fractures, left basilar skull fractures, subarachnoid hemorrhage, and a left internal carotid intimal flap with contrast extravasation into the cavernous and sphenoid sinus. Additionally, the patient had a right internal carotid artery dissection and occlusion. Cerebral angiogram and treatment of the patient's presumed CCFs were initially delayed because of the patient's hemodynamic instability, elevated intracranial pressure, and subsequent pneumonia and sepsis. The presence of a left ocular bruit, as well as left-sided chemosis and proptosis, was noted during this time.

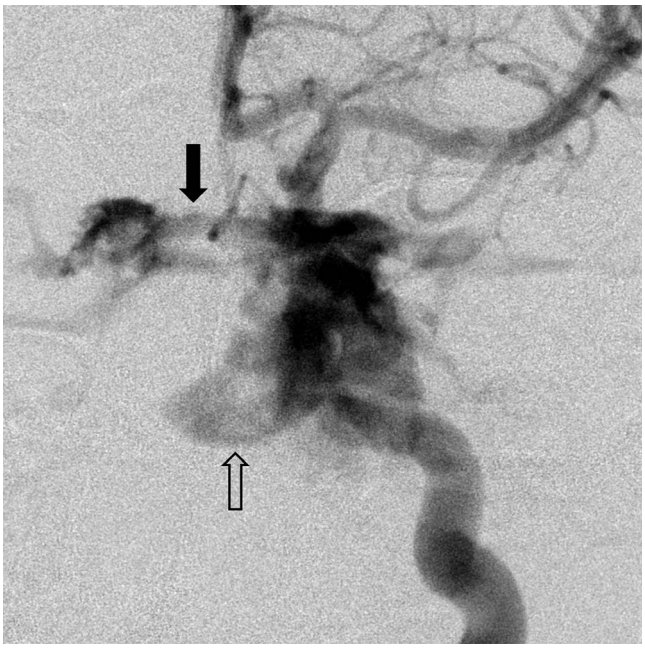

Figure 1. Preoperative anterior-posterior angiogram of the left interior carotid artery performed 10 days after the patient's injury reveals dense opacification of the left cavernous sinus indicative of a carotid-cavernous fistula. Additionally, contrast is shown filling the right cavernous sinus via the circular sinus (black arrow). A particularly rare finding is the opacification of the sphenoid sinus (outlined arrow).

On hospital day 10, the patient acutely developed massive epistaxis and became severely hypotensive, requiring implementation of a massive transfusion protocol. Two No. 24 French Foley catheters—as described by Fenn in 1968[2]—were used emergently to tamponade the bleeding through the sphenoid sinus, and the patient was taken to the hybrid neurosurgical intravascular suite for intervention.[3]

A No. 4 French micropuncture kit was used to access the right femoral artery, and a No. 6 French sheath was placed. A No. 6 French guide catheter (6 F MPC, Cordis Corporation) and a 0.035-in diameter guidewire were carefully navigated in a retrograde fashion through the aortic arch to selectively catheterize the cerebral vasculature. Angiography of the left internal carotid artery revealed a large CCF with strong opacification of the left cavernous sinus extending across the circular sinus to the right cavernous sinus (Figure 1). Extension of contrast into the sphenoid sinus was also seen. A microcatheter (Echelon 14, EV3, The Endovascular Company [now Covidien]) with a 0.014-in diameter microguidewire (X-Pedion 14, EV3, The Endovascular Company [now Covidien]) was then introduced, and a CCF with extension into the sphenoid sinus was catheterized. Superselective venography verified the appropriate placement of the microcatheter in the sphenoid sinus, and transcatheter coil embolization of the first identified fistulous tear was performed. Further exploration with the microcatheter uncovered 2 additional separate and distinct carotid-cavernous sphenoid sinus fistulas (CCSSFs) and a separate dissection and pseudoaneurysm. These CCSSFs and the pseudoaneurysm were also coil embolized. At this time, the patient had a total of 49 coils placed and an extended fluoroscopy time. Coils extended not only into the cavernous sinus but also into the sphenoid sinus. Although a fourth CCF was suspected secondary to continued filling of the cavernous sinus, the sphenoid sinus no longer filled, and therefore the ongoing risk of epistaxis was minimal (Figure 2). The surgeon felt that the benefit of continuing the intervention would be low, and the guidewires and catheters were withdrawn.

On hospital day 37, the patient returned to the operating room for further exploration of the CCFs (Figure 3). A CCF was again identified draining mostly in the posterior cavernous sinus and through the inferior and superior petrosal sinuses. A fourth fistulous tear was identified, and 2 detachable coils were deployed via the transarterial route described above. However, because of a limited fluoroscopic view secondary to the previous coil mass, no further intervention could be undertaken via an arterial route. Therefore, further coil implantation was performed via the transvenous approach. A No. 5 French guide catheter (5 F MPC, Cordis Corporation) with a 0.035-in diameter guidewire was advanced through the right femoral vein in an antegrade fashion to the inferior vena cava and eventually to the jugular vein and transverse sigmoid junction. A microcatheter (Echelon 14, EV3, The Endovascular Company [now Covidien]) was advanced into position at the posterior cavernous sinus followed by transcatheter coil embolization of the CCF. A total of 38 coils were deployed via the transvenous route. Angiography at this time revealed a marked reduction in flow through the CCF and a significant increase in redirection of flow to the left anterior circulation. The patient was discharged to an inpatient rehabilitation facility on hospital day 43. Approximately 4 months after his initial injury, a repeat cerebral angiogram demonstrated complete resolution of the patient's CCFs (Figure 4). At that time, the patient's neurologic deficits were limited to decreased visual acuity in his left eye, a left ptosis, mydriasis, and left oculomotor nerve palsy.

DISCUSSION

CCFs can be acquired or arise spontaneously. Barrow et al described an angiographic classification of CCFs that groups these lesions according to

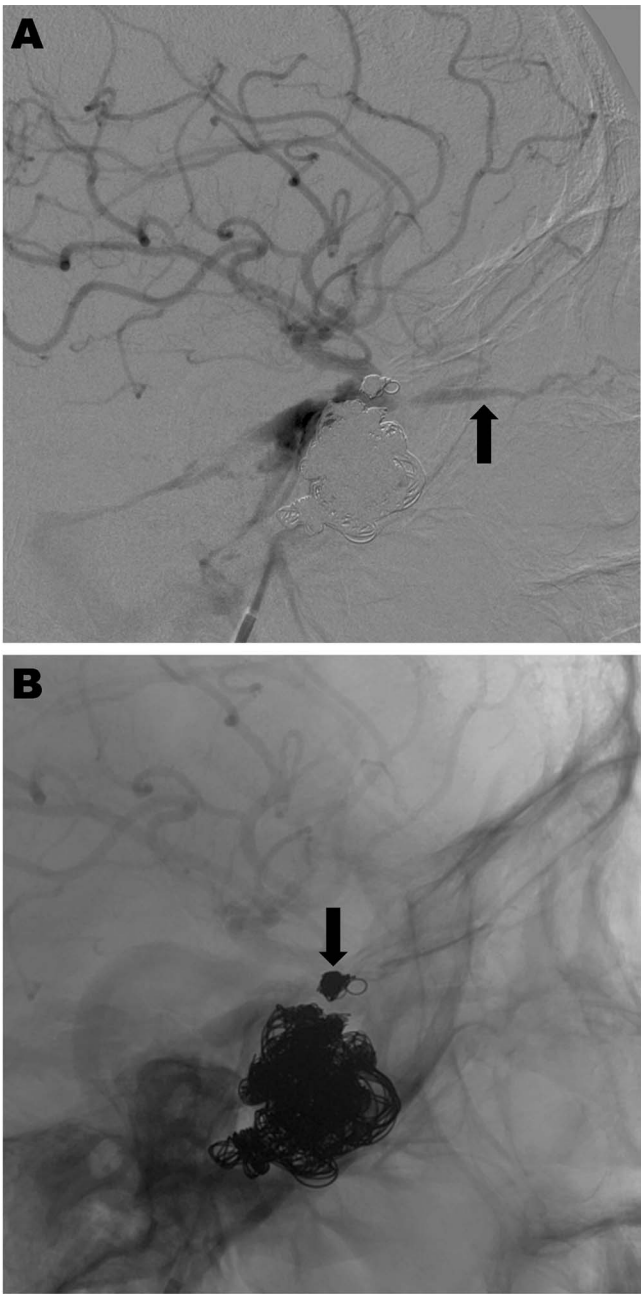

Figure 2. A: Postintervention lateral angiogram of the left interior carotid artery performed 10 days following the patient's injury reveals continued filling of the posterior cavernous sinus, as well as sluggish flow in the left superior ophthalmic vein (black arrow) following transcatheter implantation of 49 coils. No filling is shown in the sphenoid sinus. B: Note the separate coil mass (black arrow) in the dissection and pseudoaneurysm shown in the native view.

anatomic and hemodynamic characteristics.[4] Based on these criteria, the multiple CCSSFs demonstrated in this case report are classified as type A, direct fistulas, exhibiting high flow and high pressure across tears in the cavernous portion of the internal carotid

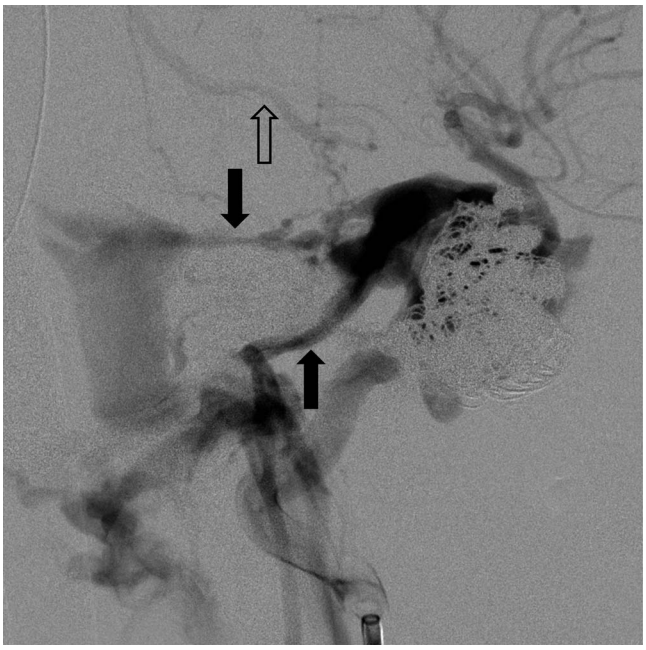

Figure 3. Lateral angiogram of the left interior carotid artery performed 27 days after initial coil implantation shows worsening carotid-cavernous fistulas with contrast filling the bilateral cavernous sinuses. Note the increased drainage to the superior and inferior petrosal sinuses (black arrows) as well as to venous collaterals filling the basal vein of Rosenthal (outlined arrow).

artery. The multiple CCSSFs in the patient were likely the result of the adjacent basilar skull fracture and/or torsion on the cavernous portion of the internal carotid artery. Unlike spontaneous CCFs (types B, C, and D), type A CCFs are generally the result of trauma and rarely resolve spontaneously. We also noted the presence of a pseudoaneurysm during our intervention. Fatality resulting from massive subarachnoid hemorrhage associated with traumatic cavernous internal carotid artery pseudoaneurysms has been shown to be as high as 100%.[5]

The clinical presentation of CCFs can vary based on the size, location, and venous drainage patterns. The venous drainage of CCFs can be anterior, posterior, superior, or contralateral. Chemosis and rapid progression of proptosis are often associated with drainage via the superior ophthalmic vein. Indeed, in our case, the patient's initial CT angiogram and subsequent cerebral angiography showed marked retrograde flow through the left superior ophthalmic vein. The patient's left-sided chemosis, proptosis, and ocular bruit subsided shortly following coil embolization; however, the sequelae associated with cavernous sinus and intraocular hypertension included oculomotor nerve palsy and diminished visual acuity. These deficits persisted even after the

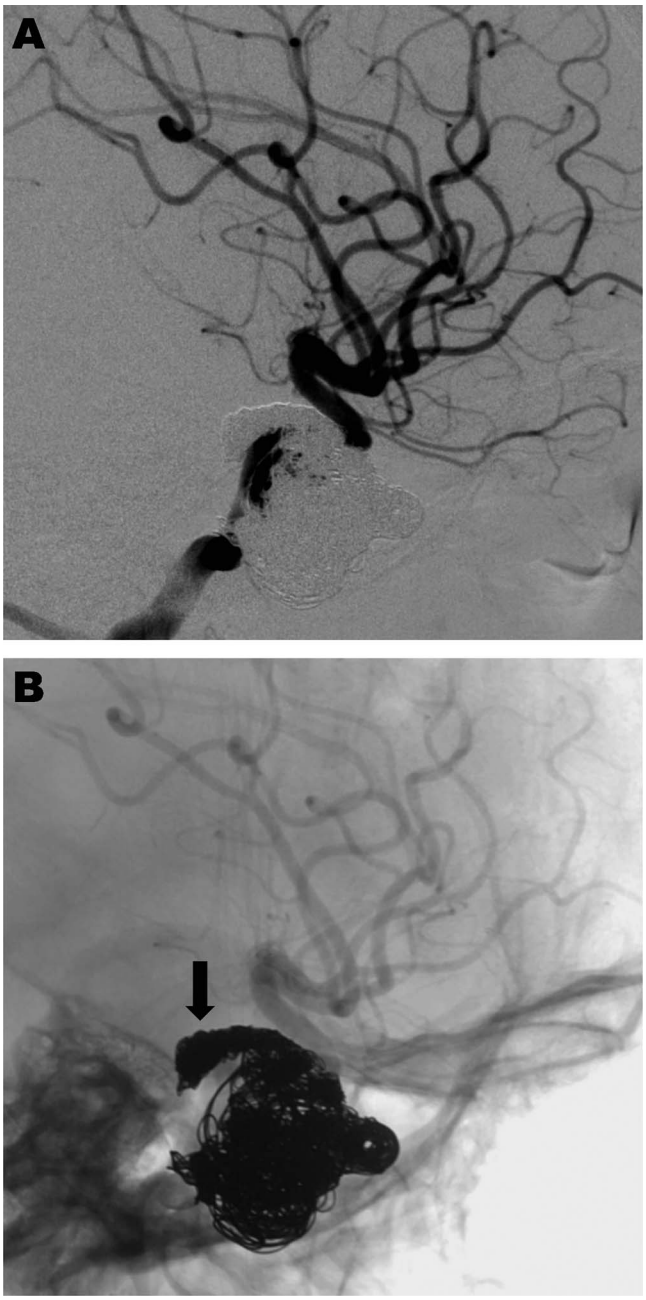

Figure 4. A: Postembolization angiography reveals a marked reduction in flow through the carotid-cavernous fistula and a significant increase in redirection of flow to the left anterior circulation. B: Note the coil mass in the posterior cavernous sinus (black arrow) placed during the transvenous embolization.

obliteration of the multiple fistulas. In addition to carotid sinus filling and superior ophthalmic vein drainage, the initial CT angiogram revealed sphenoid sinus extravasation, an ominous sign that can indicate impending arterial rupture into the nasopharynx. Instances of massive epistaxis in patients with CCFs have been reported to be approximately 3% with a mortality rate of 50%.[5,6] Key factors of survival in these cases are rapid control of the hemorrhage, transfusion, and immediate intervention. Frequently, a balloon test occlusion can be performed to establish the patency of collateral flow with the intent of endovascular occlusion of the affected internal carotid artery; however, this patient also had a right internal carotid artery occlusion, rendering deconstructive procedures undesirable.

The transarterial endovascular approach with detachable balloons is a well-established method for treating CCFs.[7,8] Embolization is achieved using latex balloons of different shapes and sizes that are inflated with either iodinated contrast or a polymerizing substance once they are placed through the fistula into the carotid sinus. The fistula must be large enough to accommodate a partially inflated balloon and small enough to ensure that the inflated balloon does not retract into the internal carotid artery. The use of multiple large balloons in the cavernous sinus can cause or worsen cranial nerve palsies, and incomplete occlusion of the fistula can produce a false aneurysm. Detachable balloons are now obsolete and no longer made for neurointervention.

Guglielmi detachable coils (GDCs) were introduced as a treatment technique for aneurysms that are difficult to treat surgically[9] and have been shown to be effective for treating CCFs.[7,10] This method of transarterial embolization was preferred in this case because of its improved ability to control placement through small fistulas and because of its ease of retrieval. The resulting coil mass artifact can be an obstacle for future endovascular treatment, and in our case with the exception of 2 coils, subsequent embolization after the initial treatment was via the transvenous route through the inferior petrosal sinus. Another endovascular option for entry into the cavernous sinus would have been via the superior ophthalmic vein. However, if all endovascular approaches are obscured by the previously implanted coil mass or are otherwise inaccessible, direct cannulation using fluoroscopic guidance through the orbit is another option that has been described as an effective means of accessing the cavernous sinus.[11] Our patient sustained multiple CCFs and required additional coiling through each fistula. The advantage of using GDCs is the ability of these pliable coils to fill and conform to the cavernous sinus. The disadvantages are time, cost, and radiation exposure. During our initial intervention, the maximum safe time under fluoroscopy was reached after almost 50 coils were placed.

Ethylene-vinyl alcohol copolymer (Onyx) is a relatively safe and effective embolization agent that has been used successfully in combination with

detachable coils or as a sole embolizing material in the treatment of CCFs.[12,13] However, one significant risk of Onyx embolization is reflux of the material into the parent vessel. For the treatment of CCFs, this risk can be minimized by the complete occlusion of the distal internal carotid artery with a balloon, slow injection, and careful balloon retrieval.[13] In our case, the risk of reflux may have been higher with the use of a liquid thrombogenic material because of the presence of multiple fistulas.

The use of covered stents is another method of treating CCFs that maximizes preservation of the parent artery. In select patients with an acceptable carotid siphon, the stent can be placed precisely over the fistula while maintaining the patency of the anterior choroidal artery and posterior communicating arteries. Covered stents such as the Jomed stent (Eminent Research Systems, Inc.) have been used off-label to treat similar lesions, and some case reports have shown them to be effective in the treatment of CCFs.[14-16] However, covered stents are not widely available at most institutions, including ours, and are not yet approved in the United States for this indication.

CONCLUSION

To our knowledge, this is the first reported case of multiple unilateral CCSSFs treated successfully with coil embolization. Early diagnosis and neurosurgical intervention are critically important. Cases of traumatic brain injury resulting in multiple CCSSFs with associated massive epistaxis are unusual. In this case, a good outcome was achieved through the use of transarterial and transvenous coil embolization.

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