

Bilateral Carotid Cavernous Fistula Caused by Head Trauma: A Case Report and Literature Review

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1. Abstract

1.1. Background

Carotid cavernous fistula (CCF) is a rare arteriovenous fistula that creates an abnormal communication between the cavernous sinus and the carotid artery. Bilateral CCF is very rare. Herein, we describe a man with bilateral ocular hyperemia and exophthalmos after a traumatic brain injury who was diagnosed with bilateral CCF based on computed tomography angiography.

1.2. Case Report

A 44-year-old man presented to the hospital because of bilateral ocular hyperemia and exophthalmos for 2 weeks after a traumatic brain injury, with aggravation of the left eye symptoms for 1 day. The patient had no special medical history. The left eyelid was swollen, the left bulbar conjunctiva was hyperemic and swollen, and the left eyeball was obviously protruding; the right eye appeared normal. The visual acuity was reduced in the left eye and normal in the right eye. Laboratory examination showed no obvious abnormality. Skull computed tomography angiography showed thickened and tortuous veins in the bilateral cavernous sinus areas that communicated with the bilateral internal carotid arteries, suggesting bilateral CCF; the venous changes were more obvious on the left side. The symptoms were markedly improved after embolization of the left fistula with a removable balloon.

1.3. Why Should an Emergency Physician Be Aware of This?

CCF is a rare condition with diverse symptoms and signs that vary between individuals. Therefore, CCF is easily missed or misdiagnosed. Many patients with CCF are not diagnosed and treated in a timely manner, which seriously endangers their health and even their life.

2. Introduction

Carotid cavernous fistula (CCF) is an acquired vascular lesion that comprises an abnormal communication between the cavernous sinus (CS) and the internal carotid artery (ICA), external carotid artery (ECA), or their branches (Figure 1). CCF occurs because of a skull base fracture that directly tears the internal carotid artery and its branches during craniocerebral injury, or a firearm injury that directly damages or indirectly contuses the arterial wall, resulting in abnormal communication between the artery and the CS [1-2]. The typical triad of CCF is pulsatile proptosis, ocular murmur, and conjunctival edema. CCF occurs in 1.0% to 2.5% of all patients with craniocerebral injury [3]. Bilateral CCF is rare, occurring in 1%–2% of patients with CCF [3-5]. If CCF is not treated in time, it can easily lead to a significant decline in visual acuity or even blindness [5]. Therefore, early diagnosis and timely treatment are beneficial to improve the prognosis of CCF. The gold standard for the diagnosis of CCF is digital subtraction angiography (DSA) [6]. The goal of CCF treatment is to occlude arteriovenous shunts

while maintaining the patency of the ICA [7]. Endovascular therapy is the preferred treatment for CCF. Herein, we report a case of a 44-year-old man with bilateral ocular hyperemia and exophthalmos after a traumatic brain injury, who was later diagnosed with bilateral CCF based on computed tomography angiography (CTA). We report the diagnosis and treatment and review the literature to deepen the understanding and provide guidance for the clinical diagnosis and treatment of CCF.

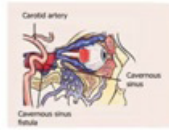


Figure 1: Schematic diagram of a carotid cavernous fistula.

3. Case Report

3.1. Chief Complaints

Bilateral ocular hyperemia and exophthalmos for 2 weeks after a traumatic brain injury, with symptom aggravation for 1 day.

3.2. History of Present Illness

The patient fell off his bicycle on December 16, 2020. The fall caused a head injury, with disturbance of consciousness, accompanied by nausea and vomiting (no further details were known). Following the initial injury, he received emergency treatment at the local hospital. Brain MRI showed no clear intracranial hematoma or intracranial vascular abnormalities (Figure 2). After receiving symptomatic and supportive treatment, he was sent home with instructions to remain under close observation. Two weeks later, he developed progressive ocular hyperemia, swelling, pain, and exophthalmos of both eyes, especially the left eye, and his visual acuity declined (Figure 2A). The patient visited the ophthalmology departments of local hospitals many times. Because the patient felt that his eye symptoms were worsening, he presented at the emergency department of our hospital seeking further treatment on January 21, 2021.

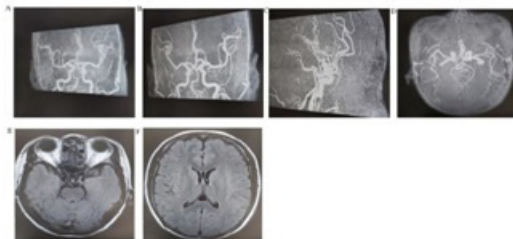


Figure 2: Changes at the time of injury seen on skull imaging. Skull CT showed no clear intracranial hematoma or vascular abnormalities. A–D: MRI findings of cranial vessels. E–F: MRI findings of the head.

3.3. Medical History

The patient reported no history of hypertension, diabetes, or drug intake.

3.4. Personal and Family History

The patient was married and had a healthy spouse and children. He

reported no history of exposure to an epidemic area or water from an epidemic area, no history of visiting prostitutes, and no family history of genetic diseases.

3.5. Physical Examination

At the time of presentation at our hospital, the patient had a temperature of 36.5°C, heart rate of 62 beats/min, respiratory rate of 18 breaths/min, and blood pressure of 132/88 mmHg. He was conscious, with rosy colored skin and mucous membranes. The left eyelid was swollen, the left bulbar conjunctiva was congested and swollen, and the left eyeball was obviously protruding; the right eye appeared normal with no obvious protrusion. There was no tremor in either eye. The visual acuity was decreased in the left eye and normal in the right eye. The bilateral pupils were equal in size and circular, with a diameter of about 3 mm, and normal pupillary light reflexes. No obvious abnormalities were detected on cardiopulmonary and abdominal examinations. The neck was soft, and the muscle strength and muscle tone of the limbs were normal. He had normal physiological reflexes and no pathological reflexes.

3.6. Laboratory Testing

No abnormalities were found on routine blood tests, coagulation function tests, and biochemical tests.

3.7. Imaging Examination

Skull CTA revealed thickened and tortuous veins in the bilateral CS areas that communicated with the bilateral ICA; these changes were more obvious on the left side. These findings were considered to indicate bilateral CCF (Figure 3).

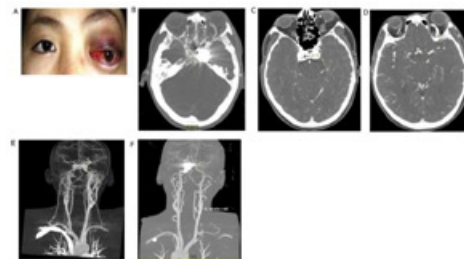


Figure 3: Changes in the eyeballs and intracranial blood vessels on CT performed 2 weeks after the injury. A: Progressive hyperemia in both eyes, accompanied by swelling and pain, especially in the left eye. B–F: Changes are seen in the cranial blood vessels on skull computed tomography angiography. There are thickened and tortuous veins in the bilateral cavernous sinus areas that communicate with the bilateral internal carotid arteries, with more obvious changes on the left side. Therefore, the patient was diagnosed with bilateral carotid cavernous fistulae.

3.8. Final Diagnosis

Bilateral CCF.

3.9. Treatment

After the left fistula was embolized with a detachable balloon, the left eyelid edema, bulbar conjunctival hyperemia and swelling, exophthalmos, and visual acuity were alleviated (Figure 4). At 14 days after treatment, the patient's symptoms were not significantly

relieved. DSA was performed again to examine the bilateral CCF. The left sinus was closed, but the right fistula was still visible. A detachable balloon was used to treat the right fistula. One month later, CTA revealed no carotid artery fistula.

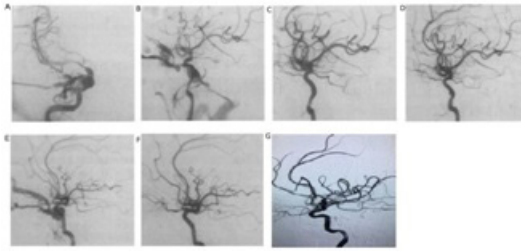


Figure 4: Changes in the bilateral cavernous fistulae on digital subtraction angiography performed before and after treatment. A, B: Right cavernous fistula before treatment. C: Right cavernous fistula after treatment. D: Right cavernous fistula at 1 month after treatment. E: Left cavernous fistula before treatment. F: Left cavernous fistula after treatment. G: Left cavernous fistula at 1 month after treatment.

4. Discussion

CCF was first described by Cushing in 1907 [2] and is a rare arteriovenous fistula that comprises an abnormal communication between the CS and the ICA, ECA, or their branches (Figure 3). The direct transmission of high-pressure arterial blood to the CS and the draining veins leads to increased pressure in the CS and changes in the drainage pattern, resulting in a series of symptoms. The typical triad of symptoms in patients with CCF includes pulsatile proptosis, ocular murmur, and conjunctival edema, but some patients do not have the typical triad [8]. The symptoms and signs of CCF are diverse and vary among individuals. Some patients develop symptoms within 1 day of trauma, while many patients do not seek medical attention until several months later [9,10]. The present patient developed left eyelid edema, bulbar conjunctiva hyperemia and swelling, exophthalmos, and impaired vision 2 weeks after sustaining head trauma. Imaging examinations are very important for the diagnosis and treatment of CCF. The initial examinations are typically CT, CTA, MRI, and magnetic resonance angiography [11,12]. DSA can identify the fistula, draining veins, and collateral circulation compensation, and is the gold standard for the diagnosis of CCF. DSA is also an important reference for the formulation of treatment regimens [13,14].

The clinical manifestations of CCF are diverse. It is not difficult to diagnose CCF in patients with a clear history of head trauma and typical symptoms in combination with the imaging examination findings. However, for patients with atypical symptoms, especially those with spontaneous CCF, inexperienced physicians without high vigilance for such diseases may overlook the medical history, physical examination findings, and imaging examination findings; therefore, such patients are sometimes misdiagnosed as having conditions such as conjunctivitis or Graves' ophthalmopathy [15, 16], thus delaying the treatment. Patients with an acute transition from a white-eyed to red-eyed shunt, markedly elevated

intraocular pressure, and acute vision loss require immediate treatment [17,18]. The goal of therapy is to occlude the arteriovenous shunt while maintaining the patency of the ICA [17]. Endovascular therapy is a safe and effective option [19]; however, endovascular therapy should be carefully planned based on the angiographic features of each fistula. In some cases of bilateral CCF, adequate unilateral endovascular treatment can be used to eliminate bilateral fistulas [20]. In the present case, detachable balloons were used to embolize the left and right fistulae, resulting in marked relief of the clinical symptoms. One month later, CTA showed no fistula in the carotid artery. In summary, CCF is a rare arteriovenous fistula with various symptoms and signs and individual differences. Many patients with CCF are not diagnosed and treated in time, which seriously endangers their health and even their life. If there is a clear history of head trauma and gradual appearance of CS syndrome, CCF should be highly suspected. CT thin-slice scanning of the skull base can be used to detect skull base fractures, while MRI or magnetic resonance angiography can reveal large flow shadows in the CS area. Cerebral blood flow ultrasonography can assist in the diagnosis of CCF. The gold standard for the diagnosis of CCF is whole-brain DSA, which cannot only confirm the diagnosis, but also provides an accurate basis for selecting the treatment method. Angiography should be performed to understand the blood supply arteries and drainage veins of the lesions and identify the size and exact location of the fistula. A cross-angiography test can then reveal the circulatory compensation of the anterior and posterior communicating arteries, which informs the choice of the most appropriate surgical method. Endovascular therapy is the preferred option for CCF. CCF drainage modalities and treatment strategies are diverse. Each fistula presents a unique challenge depending on the etiology, angiographic features, and degree of feasibility; thus, it is necessary to carefully evaluate the approach and the choice of embolic materials [21].

References

1. Jozef C. Carotid-cavernous fistula from the perspective of an ophthalmologist A Review. *Cesk Slov Oftalmol.* 2020; 1: 1-8.
2. Cushing M. Consequences of cranial injuries. *NYJ Med.* 1907; 85: 7-10.
3. Preechawat P, Narmkerd P, Jiarakongmun P. Dural carotid cavernous sinus fistula: ocular characteristics, endovascular management and clinical outcome. *J Med Assoc Thai.* 2008; 91: 852-8.
4. Docherty G, Eslami M, Jiang K, Barton JS. Bilateral carotid cavernous sinus fistula: a case report and review of the literature. *J Neurol.* 2018; 265(3): 453-459.
5. Law G, Docherty G. Dural carotid cavernous fistula following hypertensive emergency. *BMJ Case Rep.* 2019; 12(9): e230823.
6. Botsford A, Shankar JJ. DSA-Dynavision in pretreatment planning for coil embolization of indirect carotid-cavernous fistula. *J Neurointerv Surg.* 2017; 9(4): 389-393.
7. Qi Y, Liu H, Jia J, Li J, Liu A, Jiang C. Endovascular treatment for

- spontaneous carotid cavernous sinus fistula caused by fibromuscular dysplasia: A rare case report and literature review. *Clin Neurol Neurosurg.* 2020; 195: 105882.
8. Gorner AT, Luong M. Carotid cavernous fistula presenting as common red eye. *CJEM.* 2021; 23(1): 123-125.
 9. Hu WY, Hudon M. Traumatic carotid-cavernous fistula. *Can J Neurol Sci.* 2000; 27(1): 71-2.
 10. Agudelo-Arrieta M, Vergara-Garcia D, Madrinan-Navia H, Palmera-Pineda H, Vergara-Dagobeth E, Riveros WM. Concurrent carotid-cavernous fistula and cervical internal carotid artery pseudoaneurysm due to a gunshot injury: A case report. *Neurochirurgie.* 2022; 68(1): 133-136.
 11. Timol N, Amod K, Harrichandparsad R, Duncan R, Reddy T. Imaging findings and outcomes in patients with carotid cavernous fistula at Inkosi Albert Luthuli Central Hospital in Durban. *SA J Radiol.* 2018; 22(1):1264.
 12. Chen CC, Chang PC, Shy CG, Chen WS, Hung HC. CT angiography and MR angiography in the evaluation of carotid cavernous sinus fistula prior to embolization: a comparison of techniques. *AJNR Am J Neuroradiol.* 2005; 26(9): 2349-56.
 13. Ishibashi T, Maruyama F, Kan I, Sano T, Murayama Y. Four-dimensional digital subtraction angiography for exploration of intraosseous arteriovenous fistula in the sphenoid bone. *Surg Neurol Int.* 2021; 12: 85.
 14. Kohyama S, Ishihara S, Yamane F, Kanazawa R, Ishihara H, Suzuki M. Three-dimensional digital subtraction angiography for endovascular treatment of direct carotid-cavernous fistula -case report. *Neurol Med Chir (Tokyo).* 2010; 50(5): 404-6.
 15. Lee H, Yoon S, Baek S. Diagnostic clues of IOP pulsation on applanation tonometry in carotid-cavernous fistula patients. *BMC Ophthalmol.* 2022; 22(1): 37.
 16. Xia Y. A red eye induced by a spontaneous carotid cavernous fistula. *Am J Emerg Med.* 2018; 36(12): 2336.e1-2336.e2.
 17. Shaibani A, Rohany M, Parkinson R, Hopkins JK, Batjer HH, Dabus G. Primary treatment of an indirect carotid cavernous fistula by injection of N-butyl cyanoacrylate in the dural wall of the cavernous sinus. *Surg Neurol.* 2007; 67(4): 403-8.
 18. Holland LJ, Mitchell Ranzcr K, Harrison JD, Brauchli D, Wong Y, Sullivan TJ. Endovascular treatment of carotid-cavernous sinus fistulas: ophthalmic and visual outcomes. *Orbit.* 2019; 38(4): 290-299.
 19. Aguiar GB, Silva JM, Paiva AL, Jory M, Conti ML, Veiga JC. Endovascular treatment of carotid-cavernous vascular lesions. *Rev Col Bras Cir.* 2017; 44(1): 46-53.
 20. Phadke RV, Kumar S, Sawlani V, Mazumdar B, Gujral RB. Traumatic carotid cavernous fistula: anatomical variations and their treatment by detachable balloons. *Australas Radiol.* 1998; 42(1):1-5.
 21. Kumar S, Phadke RV, Mazumdar B, Roy S, Gujral RB. Double traumatic carotid-cavernous fistula and its treatment by detachable balloons. *Neuroradiology.* 1992; 34(6): 532-3.
 22. Gasparian SA, Chalam KV. Successful repair of spontaneous indirect bilateral carotid-cavernous fistula with coil embolization. *J Surg Case Rep.* 2021; 2021: 140.
 23. Ekinci B, Koktekir E, Kal A, Karalezli A. Proptosis, congestion, and secondary glaucoma due to carotid-cavernous fistula after embolization. *J Craniofac Surg.* 2011; 22(5): 1963-5.