

Bilateral carotid-cavernous fistula with spontaneous resolution

A case report and literature review

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Abstract

Rationale: Bilateral carotid-cavernous fistula (CCF) is rare and serious extra-ocular disease occurring in clinical which may result in severe complication. Unique manifestations and imaging examinations are important to the diagnosis.

Patient concerns: A case of bilateral carotid-cavernous fistula in an 60-year-old healthy man caused by a head injury is reported. Further clinical symptoms and signs and imaging examinations lead to the correct diagnosis.

Diagnoses: Computed tomography angiography of the brain aroused suspicion of bilateral CCF. On physical examination, intraocular pressure in the right eye was 35 mmHg, while the other eye was 56 mmHg.

Interventions: After diagnosis, the patient chose conservative treatment for some reasons.

Outcomes: The symptom of him had relieved in both eyes but no light perception in the right eye after two months telephone follow-up.

Lessons: Our case study demonstrated that a highly suspicion must be maintained when managing such patients to prevent serious consequences. At the same time, the early diagnosis and treatment of the disease have a critical relationship to the prognosis of patients, which should be paid attention to.

Abbreviation: CCF = carotid-cavernous fistula.

Keywords: bilateral carotid-cavernous fistula, intraocular pressure, trauma

1. Introduction

Carotid-cavernous fistula (CCF) refers to a series manifestations secondary to an abnormal communication between the internal carotid artery and the cavernous sinus or its branches and the cavernous sinus.^[1,2] The pathogenesis of CCF has been made clear.^[1] Regardless of people may compromise visual acuity and lead to brain ischemia. CCF is uncommon occurring in clinical especially bilateral CCF. Here, we report a case of bilateral CCF caused by a head injury.

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2. Case report

A 60-year-old healthy man was in a fall and received a head injury that result in subarachnoid hemorrhage and subdural hematoma 2 months earlier. The patient received conservative treatment and was discharged following the alleviation of the symptoms in the local hospital. However, the patient developed symptoms in both eyes 1 and a half months after the injury, such as blurred vision, swelling, tearing, and hyperemia. Few days later, the patient began to complain of eye pain, growing bilateral exophthalmos. When the symptoms became more severe, the patient was admitted to our hospital (Fig. 1). On examination, visual acuity and intraocular pressure in the right eye of the patient were hand motion and 35 mmHg, while the other eye were 20/25 and 56 mmHg. On physical examination, it revealed eyelid swelling, exophthalmos, severe chemosis, hyperemia, and limitation of eyeball movement in both eyes and reduction of the pupil light reflection in the right eye (Figs. 2 and 3). Blood tests, electrocardiogram, and chest radiographs did not show any



Figure 1. The appearance of the patient with carotid-cavernous fistula (CCF).

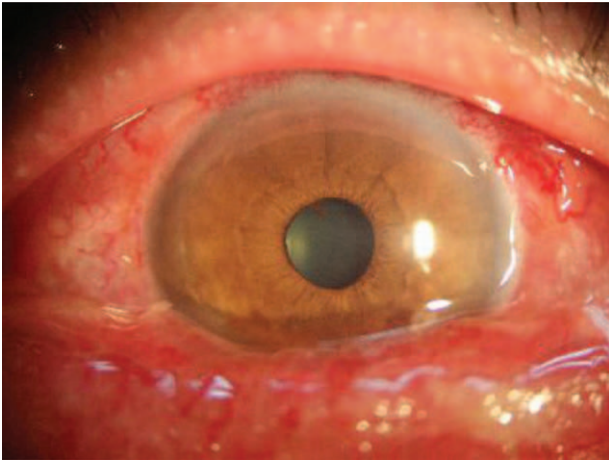


Figure 2. Slit lamp examination images of both eyes show conjunctival hyperemia, chemosis, and reduction of the pupil light reflection are seen in the right eye.

abnormality. On the contrary, computed tomography angiography revealed bilateral expansion of the cavernous sinuses and broadening of the superior ophthalmic vein without skull base fracture (Figs. 4 and 5). The result aroused suspicion of bilateral CCF. Since the diagnosis was clear, the patient was told to have a surgical treatment. However, the patient chose conservative treatment for some reasons. After 2 months telephone follow-up, the patient relieved the symptom in both eyes but no light perception in the right eye (Fig. 6).

3. Discussion

According to the etiology, a CCF may be divided into spontaneous and traumatic.^[1] Up to 70% to 90% of CCF occurs after trauma including blunt trauma to the skull.^[2] There is a report that CCF is more likely to develop after closed head injuries.^[3] Symptoms and signs of CCF consist of eyelid swelling, orbital bruit, orbital pain, pulsating exophthalmos, chemosis, hyperemia, pupillary dilatation, raised intraocular pressure, diplopia, ophthalmoplegia, and loss of visual acuity in clinical. It

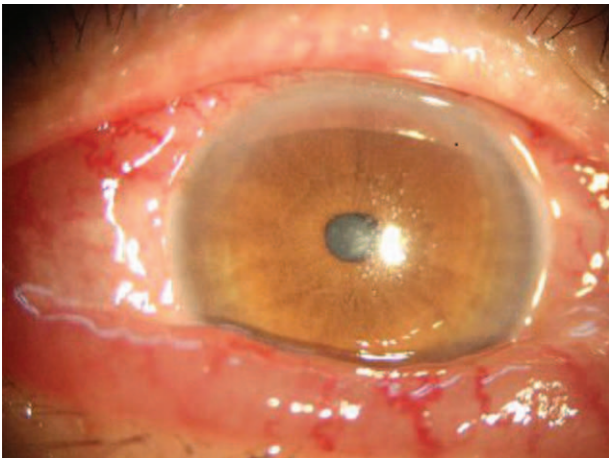


Figure 3. Slit lamp examination images of both eyes show conjunctival hyperemia, chemosis and the pupil light reflection are seen in the left eye.

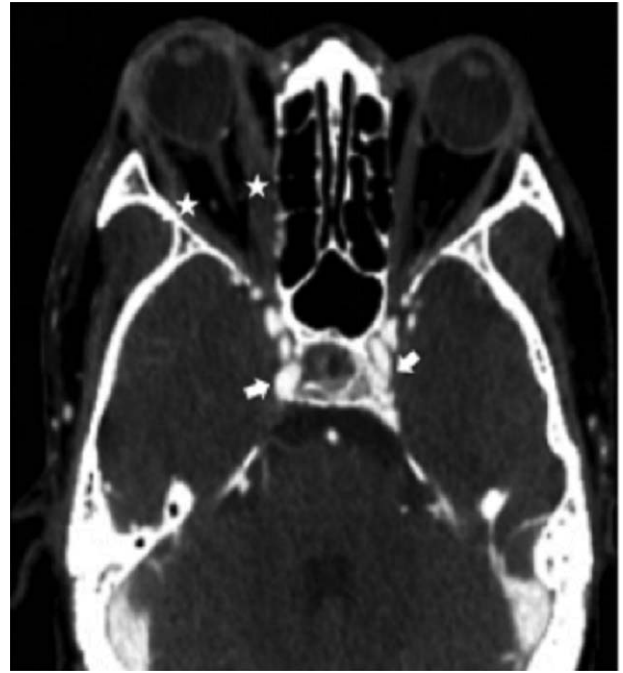


Figure 4. Computed tomography angiography (CTA) shows more obvious extraocular and intraocular muscle enlargement on the right (white stars), and confirms the connection between the internal carotid artery and the cavernous sinus on both sides (white arrows).

is also characterized by the following extra-ocular symptoms: cranial nerve palsy, pulsatile tinnitus, epistaxis, intracranial bruit, brain ischemia, and intracranial hemorrhage.^[2-4] CCF is often



Figure 5. Computed tomography angiography (CTA) shows a dilated superior ophthalmic vein in both eyes (white arrows).



Figure 6. The picture of the appearance of the patient after 2 months telephone follow-up taken by himself transferred by social software.

followed by ocular symptoms as the first clinical spectrum, which is often missed or misdiagnosed with orbital tumors, orbital cellulitis, thyroid eye disease, and inflammatory pseudotumor. Closer examination such as slit lamp examination, ultrasonography, and computed tomography could distinguish differences.

Actually, we highly suspected the ocular manifestation was secondary to head injury due to the history of trauma, disease duration, the onset of urgency, symptom characteristics, and imaging examination. First of all, the patient showed severe conjunctiva edema what mostly because of a fistula led to a rise in pressure in the superior ophthalmic vein, causing the ophthalmic veins to engorge and intraocular pressure to elevation and the orbit of edema; second, the ophthalmic artery pressure decreased during carotid sinus fistula, causing ischemia and hypoxia of the eye ball which aggravates the symptoms and signs; and last, the

elevated intraocular pressure may led to secondary glaucoma and result in decreased visual acuity.^[4] The above 3 mechanisms interact to aggravate ocular symptoms of the patient. The clinical spectrum of CCF could be relentless, demanding a surgical therapy. Although there is a possibility of spontaneous closure, detachable balloon catheterization has become the preferred method of treatment of CCF because of its small trauma, good effect, high cure rate, and low disability rate.^[5]

Because of limited understanding of this disease, there is an improper diagnosis, which leads to the blindness of treatment. If diagnosed in time, the disease cures will increase. If the delay in diagnosis, it will result in poor prognosis of patients with poor vision. Therefore, our ophthalmologist should pay high attention to CCF.

References

- [1] Barrow DL, Spector RH, Braun IF, et al. Classification and treatment of spontaneous carotid-cavernous sinus fistulas. *J Neurosurg* 1985;62: 248–56.
- [2] Chaudhry IA, Elkhamry SM, Alrashed W, et al. Carotid cavernous fistula: ophthalmological implications. *Middle East Afr J Ophthalmol* 2009; 16:57–63.
- [3] Kaplan JB, Bodhit AN, Falgiani ML. Communicating carotid-cavernous sinus fistula following minor head trauma. *Int J Emerg Med* 2011;5:1–5.
- [4] Pülhorn H, Chandran A, Nahser H, et al. Case report: traumatic carotid-cavernous fistula. *J Trauma Nurs* 2016;23:42–4.
- [5] Xiao-Quan Xu , Sheng Liu , Qing-Quan Zu , et al. Follow-up of 58 traumatic carotid-cavernous fistulas after endovascular detachable-balloon embolization at a single center. *J Clin Neurol* 2013;9:83–90.