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When delays become lethal: A case report of a rare and fatal complication in traumatic carotid cavernous fistula

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ABSTRACT

Carotid cavernous fistula (CCF) is a rare complication that can be easily missed after blunt traumatic brain injury. It is defined as an abnormal arteriovenous shunt between the carotid artery or its branches with the cavernous sinus. Most cases are traumatic in origin and although not typically life-threatening, carry a risk of serious complications. One of the most fatal complications is spontaneous intracerebral haemorrhage (ICH), occurring only 0.9-2.6% of cases. We report a rare case of a 27 yo female, who sustained traumatic brain and craniofacial injuries in a motor vehicle accident 25 d earlier. She presented with gradual loss of consciousness following ophthalmic symptoms that began 5 d prior to admission. Imaging confirmed the presence of ICH as a complication of traumatic CCF. This case highlights the diagnostic challenges of traumatic CCF and its potential complications. Diagnosis is challenging due to its rare occurrence with variable time interval for symptoms to develop (ranging from several hours to years after the initial injury), non-specific symptoms that may mimic other conditions, and delays in seeking care. Delayed diagnosis can be fatal, leading to visual loss, cerebral infarction, and haemorrhagic events. Rapid recognition and prompt management of traumatic CCF are crucial for preventing complications and achieving complete resolution with low rates of morbidity and mortality.

ABSTRAK

Carotid cavernous fistula (CCF) merupakan suatu kondisi langka yang dapat dengan mudah terlewatkan sebagai salah satu komplikasi pasca trauma tumpul kepala. CCF adalah kondisi adanya hubungan abnormal antara arteri karotis atau cabang-cabangnya dengan sinus kavernosus yang membentuk pintasan arteriovena. Sebagian besar etiologi dari kasus CCF adalah trauma dan pada umumnya tidak mengancam jiwa, meskipun tetap memiliki potensi komplikasi serius. Salah satu komplikasi paling fatal adalah perdarahan intraserebral spontan (*intracerebral haemorrhage* / ICH) yang terjadi hanya pada 0,9–2,6% kasus. Perempuan, 27 tahun, mengalami cedera otak dan trauma kraniofasial akibat kecelakaan lalu lintas 25 hari sebelumnya, datang ke Instalasi Gawat Darurat dengan penurunan kesadaran progresif setelah mengalami gejala oftalmik 5 hari sebelumnya. Pemeriksaan radiologi mengkonfirmasi adanya traumatik CCF disertai ICH sebagai salah satu komplikasi CCF tersebut. Kasus ini menekankan tantangan diagnostik pada kasus traumatik CCF dan potensi komplikasi yang menyertainya. Tantangan diagnostik tersebut meliputi: kejadian CCF yang jarang; dengan interval waktu munculnya gejala bervariasi—dari beberapa jam hingga bertahun-tahun setelah cedera awal; gejalanya juga dapat menyerupai diagnosis lain; serta keterlambatan pasien dalam mengenal tanda-tanda CCF. Keterlambatan dalam diagnosis akan berakibat fatal dan berujung pada komplikasi mulai dari kehilangan penglihatan, infark serebral, hingga perdarahan intrakranial. Hal ini menyebabkan pengenalan dini pada traumatik CCF dan penatalaksanaan yang tepat merupakan komponen yang sangat krusial dalam mencegah terjadinya komplikasi, morbiditas dan mortalitas.

Keywords:
carotid cavernous fistula;
intracerebral hemorrhage;
trauma;
complications

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INTRODUCTION

Carotid cavernous fistula (CCF) is a rare vascular abnormality that arises from an abnormal arteriovenous shunt occurring between the carotid artery or its branches with cavernous sinus.[1,2] The majority of CCF is traumatic in origin, accounts for 75% of all CCF cases, and was developed in only 0.2% of patients with traumatic brain and craniofacial injuries. It occurs more frequently in basilar skull fractures, with an incidence up to 4%.[3-5] Although most CCF is not life-threatening, it can cause visual problems and carry potential 8.4% of haemorrhagic presentations, including epistaxis, subarachnoid (SAH) and intracerebral haemorrhage (ICH). Spontaneous ICH occurs only in 0.9 – 2.6% cases.[3,6] It usually presents with a variety of symptoms including headache, blurred vision, diplopia, proptosis, chemosis, and all of these symptoms are very similar to those of a skull base fracture and orbital injury following trauma. Due to this issue and the incidence of CCF is uncommon, the diagnosis of CCF may be difficult and delayed.[3,7]

Prompt management can resolve symptoms and prevent development of disabling sequelae. Therefore, even though the incidence is quite rare, rapid recognition of the CCF diagnosis and timely management are critical to avoid morbidity and mortality.[3,8,9] In this article, we reported a rare and fatal case of spontaneous ICH as a complication of traumatic CCF, and then we highlight the diagnostic challenge of CCF and provide a review of its potential fatal complications.

CASE

A 27 yo female was involved in a motor vehicle accident 25 d prior to her second admission. Following the accident, she was initially hospitalized for 14 d and diagnosed with a traumatic SAH, a basilar skull fracture with bilateral craniofacial injuries consistent with bilateral LeFort type II (FIGURE 1), and several closed fractures in her extremities (open fractures in the left tibia and fibula, closed fractures in the femur and radius). Her fractures were surgically repaired, her brain injury was managed conservatively according to standard protocols. Nimodipine was not administered due to the traumatic, rather than aneurysmal, nature of SAH, and monitoring for delayed vasospasm was performed, but none were observed at that time. After 14 d of hospitalization, she was discharged home in decent condition, completely conscious with Glasgow Coma Scale (GCS) of 15.

Her second insult occurred 11 d after discharge. She arrived at our Emergency Department (ED) with gradual loss of consciousness over the past two days. Her husband reported that she had been experiencing a worsening right-sided headache accompanied by progressively blurry vision, diplopia and bulging of her right eye for the past 5 d. She had been unable to open her right eye for the previous 2-3 d.

On this admission, physical examination revealed GCS score of E2V1M3, blood pressure was 158/70 mmHg with heart rate 62 bpm and respiratory rate 14 breath/min, afebrile. Oxygen saturation was maintained at 98% with non-rebreathing mask 15 litre per min before intubated to secure her airway. Neurologic examination revealed left-sided lateralization and ophthalmic examination showed right-sided proptosis, chemosis, eye redness with conjunctival injection and ocular bruits (FIGURE 2). Her pupil was equal, reactive to light and round at 4mm/4mm. Laboratory evaluations, including complete blood count, serum electrolytes, renal and liver function tests, coagulation profile, ware unremarkable. Urgent CT scan revealed a right frontotemporal ICH, along with brain oedema, trans-tentorial herniation, and proptosis of the right eyeball with engorgement of superior ophthalmic veins (FIGURE 3).

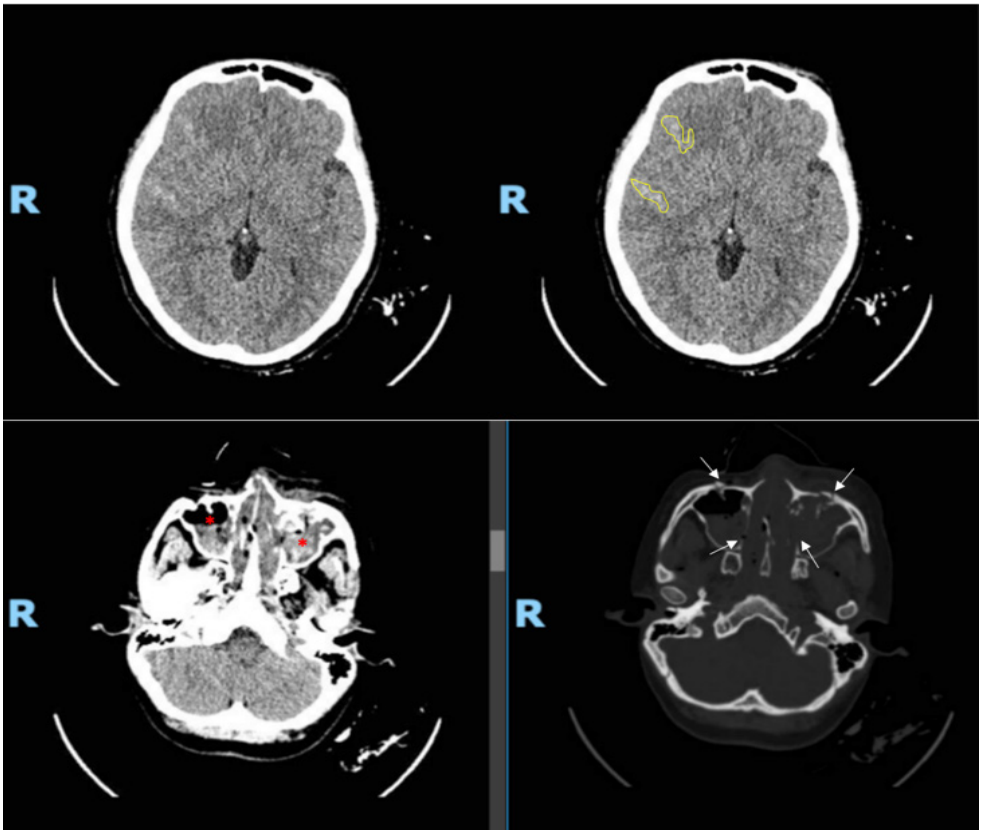

FIGURE 1. Previous head CT-scan after motor vehicle accident. Top image: right frontotemporal subarachnoid hemorrhage (yellow area); Bottom image: basilar skull fracture and maxillofacial injury (white arrow) with haemato-sinus maxillaries (red asterisk).

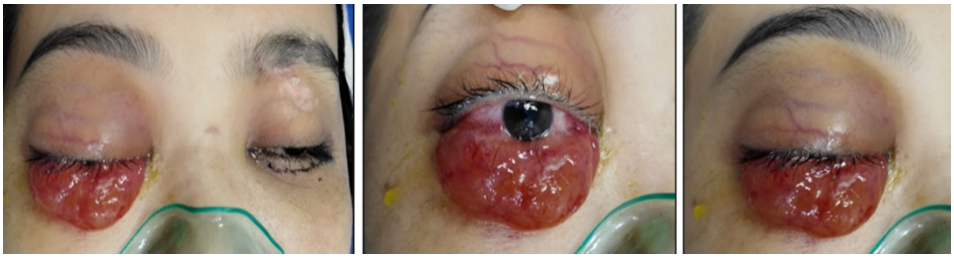

FIGURE 2. Right-sided proptosis, conjunctival injection and chemosis.

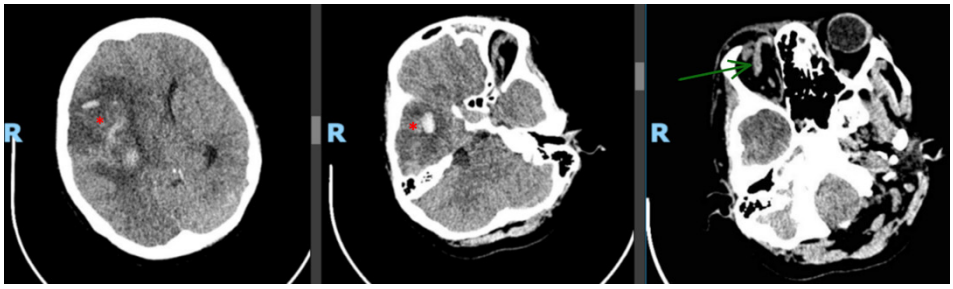

FIGURE 3. Urgent head CT-scan revealed new lesion of ICH at right frontotemporal, along with brain oedema (left and middle image, red asterisk); and engorgement and dilation of superior ophthalmic veins as indirect sign of carotid cavernous fistula (green arrow).

The combination of history of basilar skull fracture, Dandy's triad: proptosis, chemosis and ocular bruits and also engorgement of superior ophthalmic veins in head CT-scan supported the diagnosis of traumatic CCF with its rare complication of intracerebral haemorrhage. Mannitol had been used to manage her brain oedema and emergent surgical management had been arranged to treat her condition. Unfortunately, despite multiple attempts to maintain her condition stable, the patient's condition deteriorated and it was determined that she had brain death prior to surgery.

DISCUSSION

Carotid cavernous fistula (CCF) is a rare complication that can be easily missed complication following blunt traumatic brain injury. It is defined as the presence of an abnormal arteriovenous shunt occurring between the carotid artery or its branches with cavernous sinus.[3,8,9] Trauma accounts for approximately 75% of all CCF cases, with the remainder occurring spontaneously or iatrogenically. The incidence of CCF in traumatic brain and craniofacial injuries is only 0.2% of patients with male to female ratio 15:2, but occurs more frequently in basilar skull fractures, with an incidence up to 4% and is even higher at 8.3% in middle fossa fractures.[3–5,8]

Carotid cavernous fistula can be classified based on the hemodynamic properties, the etiology or the anatomy of the shunt (TABLE 2). The anatomy classification using the Barrow classification system is most commonly used (FIGURE 4), and the newest classification based on its venous-drainage system has also been developed by Thomas et al. to provide management approach using endovascular management.[4,9,10]

TABLE 1. Timeline of Clinical Events

Timeline	Events
First insult Day 0	Motor vehicle accident; diagnosed with: Moderate TBI (GCS 11) with traumatic SAH, basilar skull fracture and bilateral LeFort type II Multiple fractures (open fracture of left tibia fibula, closed fracture of left femur and radius)
Hospitalization of first insult Day 0-14	Conservative management of TBI Surgical fixation of fractures Discharged home fully conscious (GCS 15) in day 14
Onset of ocular symptoms Day 20	Onset of right-sided headache with progressive blurry vision, diplopia and bulging of the right eye
Gradual loss of consciousness Day 23	Worsening ocular symptoms, inability to open right eye, along with gradual loss of consciousness
Second insult (current admission) Day 25	ED presentation GCS 6 (E2V1M3), left-sided lateralization Right eye proptosis, chemosis, eye redness with conjunctival injection and ocular bruits Head CT-scan revealed a new right frontotemporal ICH with brain oedema, trans-tentorial herniation, and proptosis of the right eyeball with engorgement of superior ophthalmic veins Diagnosis of ICH with right traumatic CCF
Outcome Day 25	Clinical deterioration in ED, declared as brain death prior to surgical intervention

Traumatic CCF is exclusively type A, which is direct, high-flow shunts between the ICA and the cavernous sinus. The clinical symptoms from trauma to develop CCF range from within hours of trauma or may be delayed up to several years. In extreme cases, it may occur even after more than 10 years from the initial trauma.[4,6,8] The pathophysiologic changes responsible for its clinical symptoms are that the arteriovenous shunt to the cavernous sinus and the draining veins, leading to 'ophthalmic venous hypertension' and the eye symptoms appear.[5,7] All of these explanations are consistent with our case, as the patient suffered from traumatic brain and craniofacial injuries several weeks before the symptoms occurred, regardless of whether the patient was female, which is less common.

TABLE 2. Classification of CCF

Type	Fistula type based on the anatomy of the shunt (Barrow Classification)	Hemodynamic properties	Etiology
A	Direct – fistula connections between internal carotid artery (ICA) and the cavernous sinus	High-flow	trauma or rupture of aneurysm
B	Indirect – fistula connection between dural branches / meningeal artery of ICA and the cavernous sinus	Low-flow	Trauma or Rupture of Aneurysm
C	Indirect - fistula connection between dural branches / meningeal artery of external carotid artery (ECA) and the cavernous sinus	Low-flow	Spontaneous
D	Indirect - fistula connection between both dural branches / meningeal artery of ICA and ECA and the cavernous sinus	Low-flow	Spontaneous

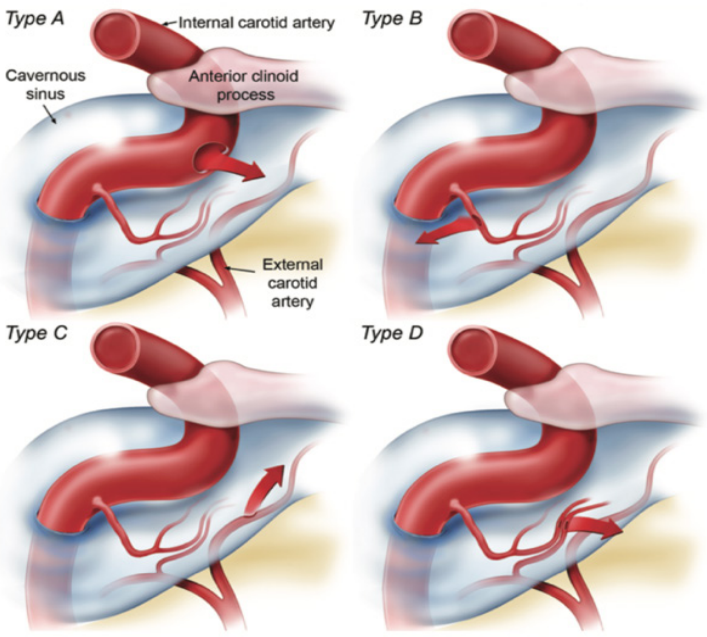

FIGURE 4. Barrow classification of CCFs based on the anatomy of the shunt (5)

The most common presenting signs and symptoms of type A CCF are the sudden development of classic Dandy's triad: proptosis (72-98%), chemosis (55-100%) and orbital bruits (71-80%). Several articles have also stated that the presence of a frontal, temporal, or orbital bruit is the pathognomonic of the CCF. Other symptoms include diplopia (88%), headache (25-84%), blurry vision and ophthalmoplegia (23-63%). Cranial nerve deficits and tinnitus have also been reported. Due to its direct and high-flow type of CCF, traumatic type A CCF typically presents suddenly, progresses rapidly, and necessitates early diagnosis and prompt treatment before complications occur.[4,5,7,10,11]

Diagnosis of CCF is quite challenging, with differential diagnoses including non-penetrating eye injury, superior orbital fissure syndrome, orbital apex syndrome, and vascular lesion such as arteriovenous malformation or cavernous sinus thrombosis, which all may mimic the symptoms of CCF.[6,12] While the gold standard for diagnosis of CCF is cerebral angiography, patients typically undergo CT-scan, MRI, or CT angiography (CTA) / MR angiography (MRA) first. The sensitivity of CTA and MRA for diagnosis CCF is 87% and 80%, respectively.[3,5,9] Evidence of cavernous sinus enlargement, proptosis, extraocular muscle enlargement, superior ophthalmic vein dilation, or dilation of cortical vessels may be seen on CT or MRI, which are suggestive of CCF. Transcranial Doppler may also detect the presence of CCF with high sensitivity; one study reported the sensitivity was almost 95% when performed by an experienced operator.[3,10,12] Treatment options for traumatic type A CCF include endovascular therapy, stereotactic radiosurgery, and surgical management, with the primary goal is to occlude the fistula.[3,11]

Similarly, as described earlier, our patient met all the classic Dandy's triad, accompanied by headache, diplopia, and blurry vision. Head CT scan revealed ipsilateral proptosis and dilation of the superior ophthalmic vein, correlating with the side of her ophthalmic symptoms. Combined with a recent history of blunt traumatic brain and craniofacial injury, the clinical suspicion for CCF was high. Beyond the differential diagnoses that may mimic CCF, the diagnostic challenge in this case is the delay in seeking treatment. Notably, 5 d elapsed from the onset of ophthalmic symptoms to hospital admission, which contributed to the poor outcome.

Major complications of traumatic type A CCF are visual loss, cerebral infarction and haemorrhagic manifestations. The complications arise from venous hypertension that may lead into retinal ischemia, venous infarction of the brain stem and subsequent potential of haemorrhagic presentations, including epistaxis, SAH and ICH.[6,13-15] The incidence of haemorrhagic presentations is as low as 8.4%, and even more rare in spontaneous ICH only 0.9-2.6%. The prognosis for CCF is generally favorable with early detection and appropriate management, with morbidity and mortality rates of less than 10 % and 1%, respectively.[16,17] However, when present with ICH complication of traumatic CCF, the majority of patients require surgical evacuation and the morbidity and mortality rates rise significantly to over 80% and 10%, respectively.[4–6,9]

In our case, the patient presented with ICH, brain oedema and sign of herniation, culminating in mortality. Earlier recognition and intervention could have substantially altered this clinical trajectory. The patient might have had a favorable prognosis if the traumatic CCF had been diagnosed and treated before the onset of this rare haemorrhagic complication. This case serves as a powerful reminder of the

urgency in diagnosing CCF to prevent devastating outcomes, and highlights that any delay can be lethal.

CONCLUSION

In summary, this case underscores the importance of early recognition of traumatic CCF as a potential complication of blunt traumatic brain and craniofacial injury. Diagnosis is challenging due to its rare occurrence, variable onset (ranging from several hours to years after the initial injury), non-specific symptoms that may mimic other conditions, and delays in seeking care. The presence of classic Dandy's triad – proptosis, chemosis, and orbital bruits – particularly when accompanied by visual disturbance, ophthalmoplegia, diplopia, headache or tinnitus following head injury should prompt strong suspicion for CCF. Most traumatic CCFs are type A (direct and high-flow) lesions that develop abruptly and progress rapidly, with potential complications including visual loss, cerebral infarction, and hemorrhagic events. Delayed diagnosis and treatment significantly increase the risk of severe morbidity and mortality.

CONFLICT OF INTEREST

No conflict of interest.

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Written informed consent for publication of this case report and accompanying images was obtained from patient's legal guardian. Figure' s in this manuscript are reproduced with permission from Daniel L. Barrow. No external funding was received for this work.

AUTHORS CONTRIBUTIONS

All authors contributed equally to this article.

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