



## Case Report

## Contralateral traumatic carotid cavernous fistula after a craniomaxillofacial fracture

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## ARTICLE INFO

## Keywords:

Carotid-cavernous fistula  
Craniofacial fracture  
Trauma  
Angiography  
Case report

## ABSTRACT

**Introduction:** Carotid-cavernous fistula is an abnormal communication between the internal carotid artery, the external carotid artery or any of their branches and the cavernous sinus. This condition may occur spontaneously or after craniofacial trauma; in this case the fistula takes place on the same side as the craniofacial fracture and becomes symptomatic within a few weeks. The diagnosis is clinical; it must be evoked before any post-traumatic proptosis. Treatment must be started quickly to avoid visual or even vital complications.

**Case presentation:** We report the case of a 19 years old male patient who was admitted to the maxillofacial surgery department for osteosynthesis of a fracture of the left orbital roof after a traffic accident. The three-month's examination noted a right pulsatile proptosis with redness and decrease of the visual acuity. The cerebral MRI was in favor of a right sided direct CCF, which was confirmed by the arteriographie. The patient responded very well to embolization.

**Discussion:** Craniofacial trauma is a major cause of carotid cavernous fistula. When a patient has ophthalmic manifestations of vascular complications, early detection of CCF is important for preserving visual acuity. The diagnosis is mainly clinical based on the ophthalmological symptoms. CT and MRI scans show the indirect signs of the fistula. There are several types of invasive and non-invasive treatments. The evolution of the fistula is generally favorable and recurrence is not very frequent.

**Conclusion:** This case report is a documentation on an exceptional case of posttraumatic direct CCF occurring on the contralateral side of the skull base fracture.

### 1. Introduction

Carotid-cavernous fistula (CCF) is an abnormal shunt between the carotid artery, or one of its branches, and the cavernous sinus (CS), allowing the blood to flow from the carotid system to the CS. CCF can be either direct or indirect communication, depending on the etiology [1, 2]. Skull base fracture after a craniomaxillofacial trauma is responsible for 70%–90% of direct CCF [3]. CCF is manifested by neurological and ophthalmological symptoms that are often diagnosed at a late stage, which may lead to some severe complications.

Symptoms of direct CCF typically develop within a few days to a few weeks after craniomaxillofacial trauma, and mostly on the same side as a craniomaxillofacial fracture. We report an exceptional case of direct CCF taking place on the contralateral side of the craniomaxillofacial fracture.

### 2. Case report

Our work is a single case report and has been reported in line with the SCARE criteria [4]. It has been registered under the reference (researchregistry7071).

We report the case of a 19-year-old male patient who was the victim of a traffic accident (motorcyclist hit by a car) resulting in a craniofacial trauma with initial loss of consciousness. The patient was initially admitted in the intensive care unit for monitoring and medical treatment of a 3mm subdural frontal hematoma. He was then referred to the maxillofacial surgery department for management of his facial trauma.

Initially, the patient was stable and conscious. On clinical examination, the patient had bilateral periorbital oedema and ecchymosis. On the left side, we noted ptosis, a palpable bony step-off of the superior orbital rim and ophthalmoplegia. The visual acuity was rated at 8/10 on

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<https://doi.org/10.1016/j.amsu.2021.102761>

Received 29 July 2021; Received in revised form 17 August 2021; Accepted 22 August 2021

Available online 26 August 2021

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the left eye and 9/10 on the right eye, with diplopia and without any sensory or motor disorder on the right side. The craniofacial CT revealed a fracture of the frontal bone radiating to the supraorbital rim and to the roof of the left orbit. The patient underwent surgery for open reduction of his fracture through a coronal incision. The one week and one month's follow-ups revealed a persistent left ophthalmoplegia, with no significant abnormalities on the right side.

The patient presented to the ophthalmologic emergency room two months later with a right eye swelling and decreased visual acuity. The clinical examination revealed a visual acuity rated at 8/10 on the left eye and 6/10 on the right, a right eye redness, pulsatile proptosis and dilatation of the episcleral vessels, the intraocular pressure (IOP) was 16 mmHg on the left eye and 26 mmHg on the right. A facial MRI revealed the presence of a tortuous and dilated aspect of the Internal Carotid artery (ICA) in its intra-cavernous portion and of the homolateral superior ophthalmic vein (SOV), an enlarged aspect of the right CS, multiple dilated homolateral cortical veins and infiltration of the extraocular muscles (Fig. 1). In view of the clinical and radiographic signs suggestive of a CCF, the patient underwent cerebral angiography that confirmed the presence of a right CCF in the C3 segment of the ICA with no opacification of the overlying segments (Fig. 2 A). The right carotid territory was well covered by the left carotid and vertebrobasilar vascular axes (Fig. 2 B). Subsequently, 5 metal coils were placed to close the CCF. The final control showed a successful occlusion of the right ICA with slowing of venous drainage (Fig. 3). The one-month follow-up revealed a regression of the episcleral injection with improvement of visual acuity to 8/10 and of the IOP (16 mmHg) and persistence of the proptosis.

### 3. Discussion

Carotid-cavernous fistula is an abnormal shunt between the CS and the intracavernous segment of the ICA or one or more branches of either or both the ICA or external carotid artery [1,5].

CCFs can be classified by their cause (spontaneous or traumatic), haemodynamic behaviour (high flow or low flow), and anatomy or angioarchitecture (directly from the ICA, or indirectly from carotid artery branch vessels) [3,6]. In 1985, Barrow proposed a classification of CCFs into four types (A–D) depending on the arterial supply: Type A CCFs are direct, high-flow lesions connecting the ICA directly to the cavernous sinus. Type B, C, and D CCFs are all indirect, low-flow lesions that arise from meningeal branches of either the ICA or ECA [7]. In our case, the carotid-cavernous fistula was classified as Barrow's type A.

Direct CCFs represent 75%–90% of all CCFs. They occur in 0.2% of patients with craniocerebral trauma and in up to 3.8% of basilar skull fractures [2]. Other etiologies include rupture of an ICA aneurysm within the cavernous sinus, Ehlers–Danlos syndrome type IV, or iatrogenic interventions, including transarterial endovascular intervention, internal carotid endarterectomy, percutaneous treatment of trigeminal neuralgia, *trans*-sphenoidal resection of a pituitary tumour, and maxillofacial surgery [6,8].

The CS is a reticulated structure formed by an assembly of multiple thin-walled veins in which the ICA and multiple nerves pass through the vein network. When a direct CCF is formed, arterial blood enters the cavernous sinus at high pressure; it can cause blood flow reversal and interferes with normal venous return to the cavernous sinus [1], dilatation of the upstream venous network with arterialization is observed [9,10]. Such venous flow can affect the ophthalmic vein, leading to orbital congestion, and manifestations such as proptosis, ophthalmic fremitus or bruit and chemosis [1]. The perfusion pressure of the ophthalmic artery is simultaneously reduced, which causes retinal ischaemia and visual disturbances [1–3]. Additionally, increased pressure within the CS can lead to neurogenic strabismus or ophthalmoplegia [8]. The fistula can also affect the posterior draining system leading to cerebral complications such as ischemic or hemorrhagic stroke or cerebral thrombophlebitis ([11]), which explains the neurologic symptoms, such as confusion and expressive aphasia [12].

In Barrow's type A CCFs, the ICA might be directly torn either by a bony fracture or by shear forces during the traumatic incident ([6]). Since skull base fractures were not to be found in multiple post traumatic CCF cases, Helmke et al. proposed another theory: that there is a sudden increase in intraluminal pressure of the ICA with concurrent distal artery compression, which forces rupture of the vessel wall and results in a CCF ([13]). This particular theory could explain the presence of the CCF in the contralateral side to fracture in our case.

The most frequent clinical signs include proptosis (72%–98%), chemosis (55%–100%), orbital bruits (71%–80%), diplopia (88%), conjunctival injection (44%), abducens nerve palsy (49%) and ophthalmoplegia (23%–63%). Intracerebral or subarachnoid hemorrhage has been reported in 5% of CCFs [10,14–16]. Other presentations include progressive visual loss and blurry vision [1,6]. Symptoms typically develop within a few days to weeks after trauma, and mostly on the same side as a craniomaxillofacial fracture [1]. Zhu et al. reported a case of posttraumatic right CCF resulting in symptoms in the contralateral eye [3], and Shim et al. reported a case of delayed contralateral CCF that developed symptoms 7 months after the craniomaxillofacial fracture

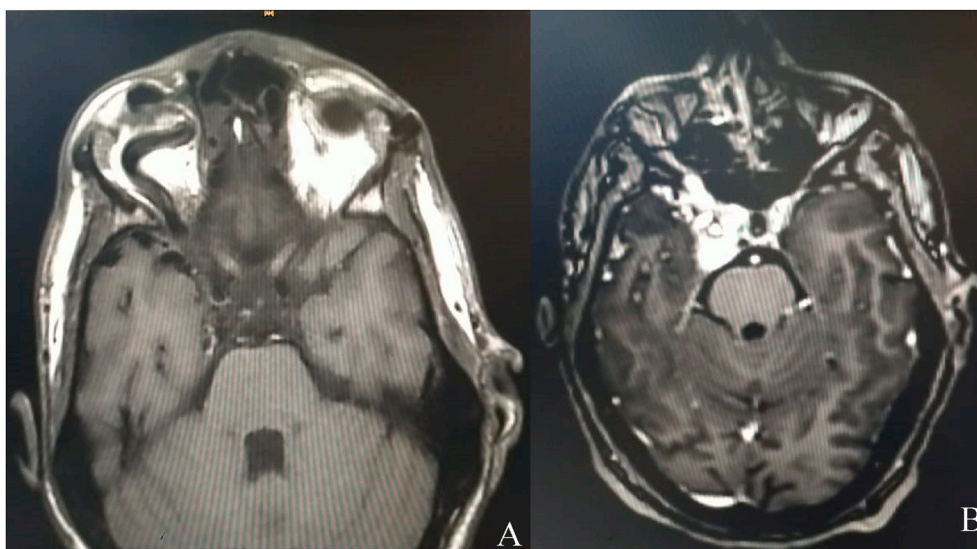
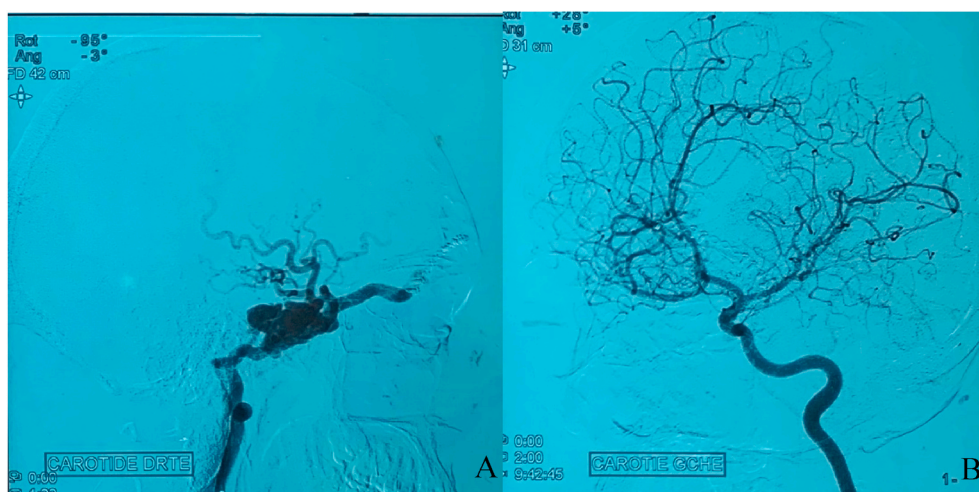


Fig. 1. Cerebral MRI showing: (A) a right exophthalmos with a dilatation and a tortuous aspect of the right ophthalmic, (B) enlargement of the right cavernous sinus.



**Fig. 2.** Preoperative cerebral angiography the opacification of the right ICA (A) in comparison with the left ICA (B) confirming the right CCF (arrows).



**Fig. 3.** Postoperative angiography showing the placement of coils to close the right carotid-cavernous fistula.

[1].

Once CCF is suspected, the best initial tests to perform are computed tomography (CT) or magnetic resonance imaging (MRI) since they are non-invasive, convenient and quickly accessible. Radiological signs such as enlargement and convexity of the lateral wall of the cavernous sinus, proptosis, enlargement of the extraocular muscles and periorbital fat, dilation of the SOV and the cortical or leptomeningeal vessels, as well as associated skull fractures are suggestive of CCF [3,6,8]. However, the absence of abnormalities does not exclude the diagnosis of CCF. These findings are suggestive but not specific for a CCF, a suspicion of CCF still requires cerebral angiography, which remains the gold standard and can be both diagnostic and therapeutic. Angiographically, high-flow fistulas show rapid filling of the cavernous sinus through the fistula with minimal or no filling of the intracranial vasculature [8].

Historically, CCFs were treated surgically by ligating the common carotid artery (CCA) or the cervical ICA proximal to the fistula and the intracranial ICA distal to the fistula, therefore trapping and occluding

the CCF [6,8]. They are no longer preferred due to their high risk of cerebral ischemic or embolic event [8] and to their mortality rate of 5% [6]. Endovascular treatment is the first line treatment modality for most CCFs, it is less invasive and carries a lower risk of cerebral infarction. The ideal approach depends on the arterial supply, the venous drainage, the speed of blood flow through the fistula, and the patency of the circle of Willis [17,18]. A transarterial approach via the ICA is most commonly used. The embolic material of choice, including metallic coils or liquid embolic agents is injected into the cavernous sinus through the microcatheter. Endovascular placement of covered or flow-diverting stents could be an alternative to embolization for the treatment of CCFs; they are deployed within the cavernous ICA redirecting blood flow away from the cavernous sinus [19,20]. Endovascular embolization offers a 90–100% cure rate [6,8,21], with a mortality rate <1% and a centre-dependent complication rate up to 20% [8]. Minor transient complications, including hematoma, facial pain, and ocular motor nerve palsies, occur in 1–30% of cases [17]. Reported complications include cerebral infarction, decreased visual acuity, diabetes insipidus, retroperitoneal hematoma, femoral vein thrombosis, ocular motor nerve palsies; trigeminal sensory neuropathy; significant IOP elevation and orbital hemorrhage. Other major complications include subarachnoid or intracerebral hemorrhage, sinus rupture, pulmonary emboli and extracranial extravasation of contrast ([6,8,22]). In cases in which endovascular treatment is not possible or is unsuccessful, open surgical intervention may be warranted; which may involve suturing, clipping, or trapping the fistula, packing the cavernous sinus to occlude the fistula, sealing the fistula with fascia and glue, ligating the ICA, or a combination of these procedures. Overall success rates using surgical intervention in the treatment of CCFs have been reported at between 31% and 79% [6].

If left untreated, CCF may lead to complications that could be life-threatening, such as varicose veins of the CS, intracranial hemorrhage or hypertension, post-traumatic pseudo aneurysm and massive epistaxis [22]. Ocular complications include increased IOP, glaucoma, retinal ischaemia or detachment [22]. Spontaneous resolution is observed in 5%–10% of CCF cases [17]. After complete obliteration of a CCF, resolution of preexisting symptoms is related to their duration and severity. Symptoms such as ocular bruits and pulsations typically disappear immediately [1], chemosis and proptosis generally resolve within hours to days [6,22]. Dilated conjunctival vessels and elevated IOP may take several weeks to months to return to normal [1]. In some cases of direct CCF proptosis and ophthalmoparesis may not disappear completely despite treatment [1,23]. Recurrence of CCFs due to recanalization postembolization is uncommon but can typically be treated by repeat embolization [6].

#### 4. Conclusion

Posttraumatic carotido-cavernous fistulas are a very rare but not exceptional complication of cranio-maxillofacial trauma. They can be invalidating and life threatening. Clinical and radiological findings are very suggestive but the final diagnosis is made by the arteriography, which can be both diagnostic and therapeutic. To improve the prognoses of patients with CCF, the endovascular treatment must be performed without any further delay. It is important to now that a CCF may occur on the contralateral side of a craniofacial fracture. This situation is very unusual but definitely merits our attention.

#### Sources of funding

The authors declared that this study has received no financial support.

#### Availability of supporting data

The data supporting this article are available for review by the Editor-in-Chief of this journal on request.

#### Ethical approval

Not applicable.

#### Consent for publication

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### Authors contributions

Elmrini Sanaa: Concept, data collection, writing the paper, reviewing following the journal criteria.

Razem Bahaa: Corresponding author, writing the paper, reviewing following the journal criteria.

Mahadi Azarak Annour: data collection, writing the paper.

Raiteb MOHAMED: data collection, writing the paper.

Elhamid Sami: writing the paper.

Baladi Oussama: writing the paper.

Faïçal Slimani: Correction of the paper and validation.

#### Registration of research studies

1. Name of the registry: researchregistry
2. Unique Identifying number or registration ID: researchregistry7071
3. Hyperlink to your specific registration (must be publicly accessible and will be checked): <https://www.researchregistry.com/browse-the-registry/#home/registrationdetails/611c06c62c46b0001ff3c012/>

#### Guarantor

Dr. Elmrini Sanaa.

#### Provenance and peer review

Not commissioned, externally peer-reviewed.

#### Declaration of competing interest

Authors of this article have no conflict or competing interests. All of

the authors approved the final version of the manuscript.

#### Acknowledgement

Not applicable.

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