A rare case report of complications in craniofacial injuries
Cavernous sinus-carotid fistula

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Abstract
Rationale: Cavernous sinus-carotid fistula (CCF) is a rare complication of craniofacial injuries in patients receiving oral and maxillofacial surgical care.

Patient concerns: A retrospective review of 15 patients with CCF records was conducted. In addition, we present a rare case of a 32-year-old Chinese woman with CCF receiving oral and maxillofacial surgical care.

Diagnoses: Digital subtraction angiography (DSA) confirmed a diagnosis of CCF.

Interventions: Endovascular surgery was performed via the femoral artery under general anesthesia without any complications.

Outcomes: The postoperative course was uneventful, and the subjective and objective ophthalmic symptoms had resolved.

Lessons: The symptoms of CCF may be delayed for several days or weeks. Thus, maxillofacial surgeons should be aware of this and avoid the untimely repair of facial fractures with potentially disastrous consequences. Interventional neuroradiologic approaches that involve the use of a detachable balloon have made it possible to care for patients with CCF without any complications.

Abbreviations: C.A. = carotid artery, CCF = cavernous sinus-carotid fistula, CT = computed tomography, DSA = digital subtraction angiography, ICA = intracavernous carotid artery, SCCF = spontaneous cavernous-carotid sinus fistula, TCCF = traumatic cavernous-carotid sinuses fistula.

Keywords: case report, CCF, craniofacial injuries, maxillofacial surgical care.

1. Introduction
Cavernous sinus-carotid fistula (CCF) is a rare complication of craniofacial injuries, occurring in only 0.17% to 0.27% of cases, according to data reported in the literature.\textsuperscript{[1-2]} Recent advancements, including new imaging techniques and improved surgical methods, have improved the diagnosis and treatment of CCF. However, its diagnosis and treatment are still major challenges for oral maxillofacial surgeons, because of the low incidence of CCF. Furthermore, untreated CCF may cause serious permanent neurologic and visual deficits or fatal epistaxis. This paper describes 15 patients diagnosed with CCF from 2000 to 2015.

2. Presentation of cases
2.1. Clinical data
This paper describes 12 Chinese men and 3 Chinese women diagnosed with CCF. There was a strong prevalence of young male patients aged 19 to 40 years, whereas the majority of the older patients were women (Table 1). The causes were a traffic accident (5 patients), beating (5 patients), falling (2 patients), and undefined (1 patient). In addition, there were 2 female patients with spontaneous cavernous-carotid sinus fistula (SCCF) without an identified cause. 14 patients presented with CCF on either side (5 right and 9 left), and 1 patient presented with CCF on both sides. Two patients displayed symptoms immediately after injury, and 13 patients displayed delayed symptoms, with the longest delay being 2 months after injury. The clinical findings were intracranial bruise (10 cases), pulsating exophthalmos (14 cases), conjunctival chemosis (15 cases), oculomotor palsy (1 case), decreased visual acuity (8 cases), headache and tinnitus (10 cases), intracranial hemorrhage and epistaxis (3 cases), diplopia (3 cases), and fundus oculi lesions (7 cases) (Table 2). Thirteen patients had cranio-maxillofacial fractures (3 cases on the temple, 3 cases on the frontal portion, 3 cases on the skull base, 1 case on the orbit, 1 case on the zygomatic bone, and 1 case on the Le Fort III of the maxilla), 1 patient had a mandible fracture. Four patients underwent common carotid artery (c. A), internal carotid artery (ICA), or external carotid artery (ECA) ligation. In addition, the ICA isolation method was used on 2 patients, and interventional therapy was used on another 2 patients. The results were satisfactory for all 8 patients. Another 7 patients did not undergo surgery and outcomes were incurable; only 1 patient displayed motor aphasia and center facial paralysis after left common c. A
and ICA ligation. Written informed consent was obtained from all patients.

2.2. Report of a rare case receiving oral and maxillofacial surgical care

A 32-year-old Chinese female receiving oral and maxillofacial surgical care was involved in a motor vehicle accident 1 week prior and sustained a closed facial injury. Although she lost consciousness for less than 1 hour, there was no evidence of retrograde amnesia. Computed tomography (CT) disclosed a bilateral zygomatic fracture. In addition, CT scans did not reveal any fracture of the skull base. Upon examination, the patient regained consciousness. There were no significant motor or sensory deficits in the extremities. The patient presented with right ptosis, associating with conjunctival chemosis and periorbital edema. There was almost complete abducents and mild oculomotor palasy. The palpation of a zygomatic projection was suggestive of a linear fracture with no displacement. The nasal bones, mandible, and palate were intact. The jaw presented with an abnormal occlusion, and opened to 20 mm. Orthopanography revealed left LeFort II and right LeFort III maxilla fractures, and inferior displacement of the bilateral zygomatic arches. Other radiographs of the periorbital area were not suggestive of a fracture. From the findings described above, a diagnosis of concomitant zygomatic and maxilla fracture was made. The findings of the right eye were striking. Pulsatile proptosis and vascular engorgement of the bulbar conjunctiva were marked. The upper and lower lids were edematic, and the upper lid was ptotic. Movement was limited, except downward.

| Table 2 | The clinical features of patients with cavernous sinus-carotid fistula. |
|---------|---------------------------------------------------------------|
| Patient no. | Intracranial bruit | Pulsatile proptosis | Chemosis | Oculomotor palsy | Obstacle visual acuity | Intracranial hemorrhage epistaxis | Diplopia | Fundus oculi | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 1 | + | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 2 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 3 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 4 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 5 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 6 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 7 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 8 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 9 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 10 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 11 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 12 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 13 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 14 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |
| 15 | - | + | + | - | + | - | - | - | - | Vein dilatation A: V = 1:2 | Left optic papilla pale | A: V = 1:2~3 | Left optic papilla edema A: V = 1:2 | Vein dilatation | Left optic papilla edema | Left optic papilla pale A: V = 1:3 |

M = Male, F = Female, TCCF = Traumatic CCF, SCCF = spontaneous carotid-cavernous fistula, c. A. = carotid artery, wks = weeks, Fr = fracture.

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and nasally, suggesting fairly good superior oblique function. The pupils reacted equally and consensually to light. Visual acuity was unchanged at 20/20 in both eyes. A 6-mm proptosis was noted. Funduscopy disclosed an outward venous engorgement and several small pre-retinal hemorrhages below the disc. Tonometry indicated a mild elevation of the intraocular pressure. The visual fields were normal. An ophthalmologic consultation considered superior orbital fissure syndrome.

On the ninth day after admission, sudden bleeding from the mouth, nose, and trachea occurred, for which the patient received one unit of blood (400mL), while right anterior and posterior nasal packs were inserted. The patient was transferred to the Intensive Care Unit. The bleeding gradually stopped. Thereafter, the patient began to notice pulsatile noises coming from the right side of her head, followed by proptosis and an increase in the intraocular pressure. The patient presented with diplopia. During the next few days, visual acuity in the right eye was gradually reduced to 20/70. The pounding, roaring sound, which associated with an orbital frontal headache, became intolerable to the patient. Digital subtraction angiography (DSA) was underwent. Bilateral selective trans-femoral carotid angiograms confirmed the presence of a discrete right CCF. There was no significant steal or crossover into the left cavernous sinus. Cross-compression films revealed good filling from the opposite carotid system (Fig. 1A, B). A surgical approach was devised in order to occlude the fine fistula utilizing a coin. During the preliminary occlusion of the right ICA with a balloon, the patient suddenly experienced left arm and leg palsy, and the surgery was terminated. The surgeon believed this due to an ICA spasm, and recommended mata’s exercise. After a month, occlusion of the right traumatic CCF (TCCF) was performed. The cavernous sinus was reached through the right femoral artery by coaxial bouginage. Electroencephalography showed no abnormalities in the neurologic state of the patient during the surgery, and 2 coins were placed inside (Fig. 1C). The complete occlusion of the fistula was achieved, and the subjective or objective murmur disappeared. The carotid arteries continued to deliver a good blood supply into the intracranial circle that was previously excluded (Fig. 1D, E). Clinically, the pulsatile protosis, oculomotor palsy, and bruit immediately improved, whereas the abducens palsy and diplopia improved gradually. In addition, the restricted opening of the mouth recovered. Two months after the embolization, the subjective and objective ophthalmic symptoms were completely resolved, although right inferior hemianopsia persisted as a sequel of the CCF.

3. Discussion

3.1. Dissection

TCCF, a direct arteriovenous shunt, is caused by a tear in the intracavernous carotid artery (ICA) or one of its branches, leading to direct communication between the ICA and the cavernous sinus. The cavernous sinus, which localizes to the lateral part of the sphenoid bone, is a plexus surrounding the c. A in this specific area of the skull. The internal c. A. lies on the medial wall of the cavernous sinus. The cavernous sinus also contains cranial nerves III, IV, and VI, as well as the ophthalmic
and maxillary divisions of the trigeminal nerves.[13] These nerves pass forward through the cavernous sinus. The internal c. A. begins at the petrous apex after exiting from the carotid canal, and it ascends within the cavernous sinus slightly medial toward the posterior cavernous sinus and slightly medial toward the anterior clinoid process. It continues to run anteriorly along the carotid sulcus of the sphenoid bone, and then curves upward on the medial side of the anterior clinoid process. After it enters the cavernous sinus, it runs medially to cranial nerves III, IV, and VI, and laterally to the ophthalmic branch of cranial nerve V. The medial wall of the cavernous sinus is between the artery and pituitary gland. Finally, the internal c. A. penetrates the dura, running in posterior, medial, and superior directions.

There are 5 sites at which a fistula can occur. The incidence of a fistula at each of these sites, according to Debrun’s data on 54 patients with CCF is as follows: 1 case (2%) on the anterior ascending segment, 5 cases (9%) at the junction of the anterior ascending and horizontal segment, 22 cases (41%) on the horizontal segment, 15 cases (28%) at the junction of the horizontal and posterior ascending segment, and 11 cases (20%) on the posterior ascending segment.[8] In this case presented in this paper, the location of the fistula was on the horizontal segment. According to Nossek’s classification, TCCF’s can be divided into type I or type II.[9] Type I is characterized by a single tear in the intracavernous c. A, thus forming a high flow arteriovenous fistula. Type II is characterized by broken branches of the intracavernous c. A. Furthermore, the anterior cavernous sinus communicates with the superior and inferior ophthalmic veins. Arterialization of this system leads to local venous hypertension and the previously described eye signs posterior communications with the petrosal sinuses and basal venous plexus overlying the clivus have clinical significance in this condition.

3.2. Etiological factors

3.2.1. TCCF. Trauma is common to most medical conditions, and it follows orthognathic, plastic, and minor surgeries, such as phinoplasty.[6] Ext.Ethmoidosurgery,[7] Biopsy,[8] LeFort I osteotomy.[9] These cases are defined as traumatic, with males outnumbering females at a ratio of 2:1. They usually associate with a skull base, frontal or mid-facial fracture. For example, the intracavernous region of the arterial wall, which is surrounded by an irregular plexus of venous channels, can be torn or lacerated by a bony spicule or a shearing force (to which it is extremely susceptible, as it is fixed by the carotid canal posteriorly and by the dura anteriorly). Furthermore, its vasa vasmorum may be damaged, thus producing a dissecting aneurysm, which will eventually be ruptured into the cavernous sinus ensuing.[10]

3.2.2. SCCF. The causes of SCCF are more difficult to ascertain. SCCF is more common in older women with increased incidence during pregnancy; however, it rarely occurs in males. SCCF can be due to a pre-existing intracavernous carotid aneurysm, defect in the carotid wall, or traumatic rupture. The most common cause of SCCF is a rupture of a pre-existing subclinical carotid aneurysm. In 12% to 15% of cases, carotid aneurysms occurred bilaterally, which is probably due to a residual remnant of the primitive trigeminal artery.[11]

3.2.3. Clinical symptoms. The symptoms of CCF are generally monolateral, usually occurring at an average interval of 4 weeks after trauma, although this interval may range from a few hours to 1 year. Thus, the onset of symptoms may vary. The symptoms that are commonly associated with CCF include proptosis (94%), pulsating exophthalmia (40%), bruit (73%), orbital frontal headache and orbital pain (40%), chemosis (71%), extraocular palsy and diplopia (60%), loss of visual acuity (46%), and fifth nerve involvement (24%).[12-14] The pressure gradient resulting from the carotid-cavernous communication creates a retrograde flow into the ophthalmic veins. As a result, several symptoms may be noted. Firstly, pulsating exophthalmia may be evident, which can be eliminated by compressing the homolateral common c. A. Pulsation with an orbital headache and pain may also be felt in the affected temporal and parietal areas, as well as subjective or objective bruit. Its intensity is often higher on the fistula side, particularly at the level of the involved orbit, and dilation of superficial veins of the eye lids and forehead, chemosis, pulsating retinal veins, papilloedema, loss of visual acuity due to impaired retinal circulation and possibly, late optic atrophy, may also be evident. Secondly, diplopia, the most frequent symptom, is secondary to compression of the oculomotor nerve in the cavernous sinus, which maintains a close relationship with the c. A., and its involvement will result in a limitation of lateral gaze. The IIIrd and IVth nerves may also be affected, resulting in ophthalmoplegia. The ophthalmic and maxillary branches of the IVth nerve, when affected, will cause sensory disturbances, while the VIIth and VIIIth cranial nerves will become involved as the haematoma increases or the intraorbital pressure rises. Thirdly, ptosis is caused by the loss of sympathetic supply to Muller’s muscle or the disruption of the innervation of the levator palpebrae. Fourthly, disruption of the parasympathetic innervation may result in a fixed, dilated pupil. The latter, which associates with ptosis and anhydrosis, is suggestive of Horner’s syndrome.

3.2.4. Different diagnoses. Other conditions with similar symptoms include superior orbital fissure syndrome, orbital apex syndrome, orbital (retrobulbar) hematoma, and cavernous sinus thrombosis. Cavernous sinus thrombosis can occur in the absence of trauma. The exophthalmos related to CCF usually progresses for several days and then stabilizes. With orbital hematoma, there is a tendency toward the spontaneous resolution of exophthalmos. The loss of visual acuity with orbital hematoma is not common. The presence of orbital bruit is diagnostic, and it is characteristic of only CCF. The symptom that is most distressing to patients is hearing a hum, swish, or roar, which can be effectively suppressed by carotid compression. A recent history of dental treatment, facial infection with an elevated white blood cell count, or leukocytosis can be suggestive of cavernous sinus thrombosis. Furthermore, superior orbital fissure syndrome and orbital apex syndrome occur most frequently in conjunction with LeFort II and III maxilla fractures, which can involve the optic foramen and superior orbital fissure, thus disrupting the sensory and motor innervation to the orbit and adjacent structures. Direct orbital trauma without evidence of fracture and with all the symptoms discussed, except infection, which can be effectively suppressed by carotid compression. Plain and tomographic films indicating fractures of the sphenoid complex should lead the physician to a high index of suspicion for CCF. The single most important diagnostic study should be cerebral angiography when diffusion of the radiopaque material into the cavernous sinus during the filling phase is noted. This indicates the presence of a fistula; it also measures its size, determines the flow to the distal supraclinoid vessels, and most importantly indicates the competency of the circle of Willis and contralateral carotid circulation.[15,16]
3.2.5. Treatment. The treatment modalities for CCF involve dispelling the blood vessel in the cranium, allowing contraction of the projecting eye, rescuing eyesight, and preventing blood loss from the brain (i.e., hemorrhage). The treatment depends on the speed of blood flow through the fistula and its arterial supply, and the routes of venous drainage. The basic principle is to correct circulatory hemodynamics. However, there have been a few reports of spontaneous closure of such a fistula after accidental severe hypotensive episodes, and approximately 25% to 50% of dural CCFs will close spontaneously. The detachable balloon approach is considered by most neurosurgeons to be the best initial therapy. Surgical techniques are generally indicated when the ICA has been previously ligated or the balloon cannot be inserted into the cavernous sinus. If the detachable balloon approach fails or the balloon cannot be inserted into the cavernous sinus. If the detachable balloon approach fails or the fistula and ICA have been occluded but the fistula reoccurs, then surgery is the last resort. Surgery involves ligation or occlusion of the common, internal, and external carotid arteries or embolization of the intra-cavernous portion of the internal c. A. Although complications can arise, the success rate is 50%. In this study, one female patient who underwent common arterial ligation may arise in different patients. Furthermore, head injuries may be associated with a contralateral traumatic carotid cavernous fistula. A case report. Interv Neuroradiol 2007;13:287–93.

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