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Case Report

Delayed Carotid Cavernous Fistula after Craniomaxillofacial Trauma

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Received: February 03, 2020; **Published:** March 19, 2020

Abstract

The carotid-cavernous fistula (CCF) is an abnormal connection between the carotid artery and the cavernous sinus, which causes a sharp change in the direction and distribution of blood flow, that can have consequences such as severe alterations of the brain and orbits, or even death. CCF can develop after craniomaxillofacial trauma or spontaneously, however, it has also been associated with arterial hypertension and connective tissue diseases. Characteristic signs include pulsatile exophthalmos, murmur and venular dilation with chemosis (Dandy's triad). The purpose of this study is to present a clinical case of a direct traumatic carotidocavernous fistula, its diagnosis, management and complications.

Keywords: *Carotid Cavernous Fistula (CCF); Craniomaxillofacial Trauma*

Introduction

The carotid cavernous fistula (CCF) is an aberrant vascular communication between the internal carotid artery and the venous channels of the cavernous sinus of the sphenoid bone, causing an abrupt change in the direction and distribution of cerebral and orbital blood flow, which can cause severe sequelae [1]. This condition occurs more frequently in women, hypertensive or over 50 years patients. CCFs can be classified according to their etiology (traumatic or spontaneous), hemodynamic behavior (high and low flow) or angiographic evaluation (direct or indirect) [2]. Most CCFs after craniomaxillofacial trauma, spontaneously, or by pathologies of the cavernous sinus, and in a very small percentage, by iatrogenesis after procedures such as LeFort osteotomies, rhinoplasty, partial maxilectomies, reparations in fractures of the orbit floor, sphenoid sinus surgeries or nasopharyngeal biopsies [3]. The clinical features are closely related to the inversion of blood flow and stasis. Muscle thickening, edema of the orbital fat and injury of the cranial nerves associated with the venous sinus will result in an alteration of the photomotor reflex, chemosis, thickening and episcleral and conjunctival venous tortuosity, pulsatile glaucoma, optic disc edema and retinopathy of stasis and ischemia. Eyelid cyanosis, pulsatile proptosis and ophthalmoplegia are associated with ocular pain, together with decreased visual acuity and thrill [4]. Characteristic signs include pulsatile exophthalmos, murmur and venular dilation with chemosis (Dandy's triad) [5]. The time between symptoms and diagnosis ranges from 1 to 18 months, with an

average of 5 months, which increases the probability of severe sequelae [4]. Cerebral angiography is the diagnostic gold standard, allowing to identify the type, location and size of communication, and examining venous return and coexisting alterations, especially ischemic effects on the cerebral cortex [6]. Computed tomography scan and Doppler ultrasound are also acceptable alternatives, without showing superior results [7].

Purpose of the Study

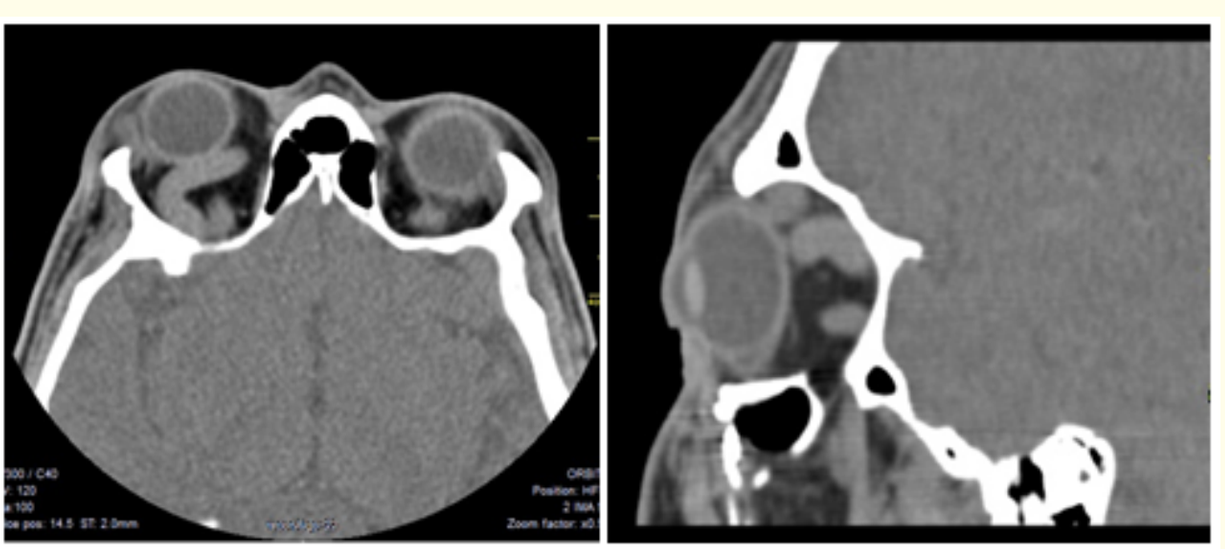
The purpose of this study is to present the clinical case of a direct traumatic carotidocavernous fistula, and how its management, diagnosis and treatment can affect the patient's life prognosis.

Case Report

A 47-year-old male patient was admitted to the Imbanaco Medical Center, Cali, Colombia, with a 1-day clinical picture consisting of polytrauma secondary to a traffic accident as a motorcycle driver. The patient did not report relevant medical-personal history. During the initial history, the patient was disoriented, denied diplopia and changes in visual acuity. Within the clinical examination, he presented severe edema in the right-side face, bilateral periorbital ecchymosis with 100% palpebral occlusion in the right eye, stigmas of bleeding in the nasal and oral region, reduced mouth opening quantified in 20 mm, anterior and posterior open bite, mobility of bone segments in the maxillary region and in the left mandibular body, ecchymosis in the floor of the mouth. The eye movements and pupils were not evaluable for edema. Tomographic evaluation shows a comminuted right malar fracture, displaced in a caudal and medial direction, compromising the lateral wall of the ipsilateral maxillary sinus, infraorbital rim and orbital floor. It also presents fracture of the right mandibular ramus and body, and fracture of the left mandibular parasymphysis (Figure 1). Mild craniocerebral trauma, multiple facial fractures, right clavicle fracture were diagnosed.

Surgical treatment consisted of open reduction of clavicular and facial fractures in a single surgical time; the latter were performed using osteosynthesis material including a right orbital floor mesh (Figure 2). The procedures were performed without intraoperative

complications. The hospitalization proceeded with adequate evolution, which is why discharge was decided, however, during his departure the patient refers a feeling of whistling noises in the right ear. Therefore, consultation is performed at the otolaryngology service, who diagnose post-traumatic tinnitus and request audiometry. Thus, the patient continues follow-up by outpatient consultation. At the third postoperative month, the patient presented a slight increase in tinnitus, without other alterations. In the fourth month, he started with symptoms of blurred vision and signs of proptosis and chemosis in the right orbit, so an assessment was requested by the optometry and ophthalmology services. The first service diagnosed presbyopia, the second one considered possible the presence of a foreign body in the right eye and began treatment with ophthalmic solution, but the symptoms became more acute, due to which the patient was referred to oculoplastic surgery. During the routine postoperative checkup appointment with the maxillofacial surgery service, an orbital tomography was requested, in which dilation of the ophthalmic vein, thickened of extra-ocular muscles and right unilateral proptosis were observed (Figure 3). With these findings, the patient was referred to the neurosurgery department, who requested an angiography and diagnosed a traumatic, high flow and direct unilateral right traumatic carotid fistula (Figure 4). Immediately, his hospitalization in the intensive care unit was indicated, due to the risk of rupture of the fistula. The patient was taken to the surgery room for embolization and placement of a coated stent, after which he presented a fistula rupture, followed by 2 cardiorespiratory arrests. The patient dies at the fifth postoperative month.



Discussion

The diagnosis and treatment of CCFs are undoubtedly beyond the competence of the maxillofacial surgeon. However, being a professional who constantly manages craniofacial trauma and invasive surgeries such as open reduction of orbital fractures or orthognathic surgery, it is useful to have knowledge of the signs and symptoms associated with this pathology. It is important to mention that the clinical picture presented by the patient can vary, taking into account that there are 2 types of CCF: high and low flow. Direct or high-flow fistulas occur between a main branch of the internal carotid artery and the cavernous sinus; they occur 3 times more and are associated with craniofacial injuries that cause tearing or transection of the muscular wall of the artery by shear forces [8]. Indirect or low-flow fistulas, which occur between the cavernous sinus and extra-dural branches of the internal carotid artery, the external carotid artery, or both, tend to be spontaneous or idiopathic and often unilateral [9]. The signs that we can find are proptosis, chemosis, episcleral congestion, orbital frustration, orbital/temporal murmur, ocular paralysis (VI cranial nerve, is the cranial nerve most affected by its location within the cavernous sinus), pulsatile exophthalmos, ptosis, intraocular pressure elevated, anterior segment ischemia, papilledema, optic nerve atrophy and trigeminal nerve involvement, however, subsequent CCFs may not show classic symptoms (exophthalmos, chemosis and fronto-orbital murmur) [8].

In this case, the patient barely started with the presentation of signs from the fourth postoperative month, however he did report the presence of tinnitus from the moment of discharge. Having undergone an open orbit fracture reduction surgery may have generated some signs such as proptosis, chemosis or exophthalmos during the immediate postoperative period, which adds an additional distraction factor. It should be noted that this case is not the first documented in which the first diagnostic impressions are not conclusive, because the condition is sometimes difficult to diagnose. Although the symptoms are usually pronounced, in some cases the patient may have mild or nonspecific characteristics such as red eyes, atypical glaucoma, diplopia, pulsatile tinnitus, temporary headache and ptosis [10].

In addition to the clinical examination, better diagnosis can be achieved by means of 3 imaging tests: cerebral arteriography, current gold standard in imaging diagnosis, can reveal the size, precise location of the CCF and also demonstrate the herniation of the cavern-

ous sinus to adjacent structures. In this case, it was the tool that helped confirm the diagnosis; computed tomography (that in this case, together with the clinical examination, helped to generate the diagnostic suspicion), demonstrates the presence of craniofacial fractures, asymmetric enhancement of the venous sinuses and dilation of ophthalmic veins; and finally the orbital color Doppler or the pulsed-wave Doppler ultrasound systems, are useful to increase the caliber and the inverted flow of the superior ophthalmic vein, so that it can be seen [1].

Today, modern endovascular techniques can be used to treat CCFs. Coils or liquid embolics can be delivered through intrasinusal catheterization to close the fistulas. Transarterial embolization with inflatable balloons or stents can be used to treat direct CCF. Surgical cut of the superior ophthalmic vein or direct cannulation of the cavernous sinus through percutaneous transorbital puncture can be performed when inadequate angiographic visualization limits standard endovascular approaches [11]. In this case, unfortunately, the stent placement procedure was insufficient, due to the cardiorespiratory arrest that occurred intraoperatively.

Conclusion

Cavernous carotid fistulas are a possible complication after trauma or craniomaxillofacial procedures; therefore, all related actors should know the signs and symptoms that can lead to a diagnosis as early as possible. Interdisciplinary management with neurosurgery services can avoid uninviting outcomes such as the one presented in this case. On the other hand, the proper use of new technologies today allows greater diagnostic clarity and more effective treatment.

Conflict of Interest

None declared.

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Volume 19 Issue 4 April 2020