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Case report

Nature of Head & Neck Tumours

Carotid cavernous fistula: A rare complication of maxillofacial trauma

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Abstract

Introduction

Carotid cavernous fistula (CCF) is an abnormal communication between the internal carotid artery and the cavernous sinus. It is a rare complication caused by maxillofacial trauma and can lead to amaurosis. This paper describes two cases of young men with late CCF diagnosis.

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The first patient was 23 years old and had a basilar skull and he also had paralysis on the right side of his face. The diagnosis of CCF for this patient occurred three weeks after the onset of symptoms through an arteriography. Embolization treatment was then performed. The second patient was 21 years old and had a LeFort ll fracture on the right side of his face. He showed symptoms of CCF and was also treated by embolization. The classical clinical symptoms of chemosis, paralytic mydriasis, absence of consensus and photomotor reflex, restriction of eye mobility, proptosis, thrill and murmur were present in both cases.

Conclusion

This paper emphasizes the need for early diagnosis and prompt treatment to prevent loss of visual function in patients with carotid cavernous fistula.

Introduction

Carotid cavernous fistula (CCF) is the abnormal communication between the internal carotid artery and the cavernous sinus. Craniofacial trauma is a major factor for this complication. Early diagnosis of this rare vascular complication can preserve vision. Despite its description being well established in literature CCF resulting from maxillofacial trauma it is still responsible for amaurosis in some cases[1]. Although the treatment of this condition exceeds the qualification of the maxilla-facial surgeon, understanding its mechanisms is necessary to insure early diagnoses and to achieve the best prognosis[2].

The objective of this paper was to analyse two cases treated at the Centre of Cranio-Maxillofacial Surgery in the Department of Otolaryngology Head and Neck at Unicamp and to review the literature regarding the diagnosis and treatment of these fistulas.

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Case 1

A male patient aged 23 who was a victim of a motorcycle accident was hospitalized in the Intensive Care Unit (ICU) for 12 days. Two months later the patient suffered from otorrhagia on the right side, facial paralysis on the left side, swelling in the eye and loss of visual acuity. During the next 20 days the right eye developed progressive proptosis. The patient consistently complained about headaches and dysacousis with purulent discharge out of the right ear.

During physical examination we found a tracheotomy scar, bilateral crepitus in the temporomandibular joint, grade IV House Brackmann facial paralysis on the left side and perforation of the tympanic membrane with purulent discharge. There was no palpable fracture found on the face of the patient.

In the right eye we observed intense chemosis with exposure of lower tarsal conjunctiva, paralytic mydriasis, absence of consensus, absence photomotor reflex, restriction of mobility in all directions, proptosis, fremitus and murmur (Figure 1).

Computerized tomography of the base of the skull showed a fracture involving the right temporal region, parallel to the carotid canal that reached the clivus and the sphenoid body (Figure 2). No signs of fracture in the left temporal bone were identified.

The patient was then referred for an arteriography and possible embolization of the suspected carotid cavernous fistula. Once the CCF was confirmed, embolization was attempted but was not effective due to the high fistula flow. Two weeks later the fistula was completely closed using detachable balloon embolization and synthetic polymer, which resulted in occlusion of the right cavernous sinus.

A control angiography was performed six months later and revealed the absence of fistula. During the following nine months all symptoms disappeared except visual loss. An otoscopy showed that the right tympanic membrane was intact. The patient also maintained grade II left facial paralysis (Figure 3).

Case 2

A male patient, aged 21, was a motorcycle accident victim with a Le Fort II facial fracture that required a nine day stay in the ICU. In the interval of two and a half months, he developed proptosis with progressive visual loss in the right eye and he was sent to us for assistance.

During the physical examination we observed chemosis of the right eye, paralytic mydriasis, an absence of consensus and photomotor reflex, restriction of its mobility, fremitus and ocular murmur.

No evident changes to the base of the skull were present on the computerized tomography but, it did show osteosynthesis plates in the fronto-zygomatic suture and in the right inferior orbital rim.

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Figure 1: This illustration shows intense chemosis with exposure and edema of lower tarsal conjunctiva and proptosis of the right eye.

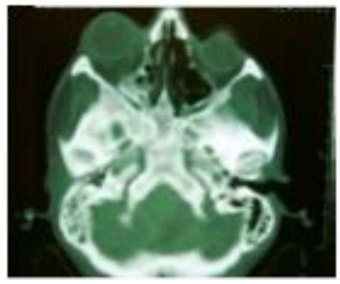

Figure 2: Computerized tomography in axial plane of the base of the skull shows a fracture involving the right temporal region, parallel to the carotid canal, reaching the clivus and the sphenoid body and proptosis of the right eye.

Figure 3: The nine months postoperative outcomes show great improvement of the trauma signs.

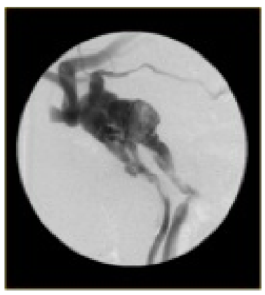

Figure 4: This image illustrates a communication of right internal carotid artery and cavernous sinus (CCF).

Figure 5: The radioscopy shows the good results after the embolization procedure.

Figure 6: The arteriography shows the good results after the embolization procedure, with good tissue perfusion.

Due to the suspicion of a CCF, the patient underwent an angiography to diagnose and to perform embolization which was successful on the first procedure (Figure 4, Figure 5 and Figure 6). After five months the patient had no further complications except for visual loss.

Two years after embolization, hyperemia and congestion of the right eye were observed (Figure 7). The patient suffered with headaches but had no proptosis. A new angiography was performed and the presence of a fistula was not found. One month later the subsequent physical examinations showed spontaneous improvement (Figure 8).

Discussion

The cavernous sinus was first described in 1732 by Ridley "as a circular sinus" and gained its current anatomical nomenclature, in the same year, after Winslow demonstrated the presence of trabeculae within the sinus of cadavers. In 1964 Dwight Parkinson described in detail the microanatomy and surgical treatment of carotid cavernous fistula. The initial results where discouraging[3].

The complexity of carotid cavernous fistulas is a result of their location within the unique anatomical structure of the body where an artery crosses a venous structure. Through the superior and inferior ophthalmic veins the cavernous sinus receives venous blood that is drained via the sphenoparietal sinus, superior petrosal sinus, basilar plexus and pterygoid plexus. When passing through the foramen lacerum the internal carotid enters into the cavernous sinus and in the posterior wall it is divided into three segments: 1) posterior ascending segment, 2) horizontal segment, the largest segment of the intracavernous carotid, 3) superior ascendant segment. The most accepted classification of CCF was described by Barrow[4] and is based on the communication between the carotid artery, its branches and the cavernous sinus.

This classification recognizes direct fistulas (type A), in which the internal carotid communicates directly with the sinus and indirect fistulas in which communication with the cavernous sinus occurs by dural branches.

Post traumatic fistulas occur in approximately 0.2% of traumatic brain injuries (TBI) and approximately 75% of these are caused by severe injuries such as vehicle accidents or penetrating traumas. They occur more often in young adult men because of their higher involvement in accidents. Spontaneous fistulas have a higher incidence in post-menopausal women, with no predominance in a particular race[6].

Direct fistulas are the most common and are caused by trauma in 70 to 90% of the time, mainly basilar skull fracture. They can also be iatrogenic (rhinoseptoplasties, endoscopic sinonasal surgeries, transsphenoidal hypophysectomy) or spontaneous by aneurysm rupture in the cavernous

Figure 7: This figure shows hyperemia and congestion of the right eye, without proptosis two years after the first procedure (embolization).

Figure 8: This figure shows the status of the patient one month after the suspicious relapse with a spontaneous improvement.
There was still a slightly hipermia of the right eye.

portion of the carotid or congenital malformation that break spontaneously due to vascular disease linked to collagen (e.g. Ehlers-Danlos syndrome, atheromatous disease or hypertension).

When compared to dural fistula, the size of direct lacerations direct fistula is

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proportionally greater and determines the difference of the endovascular treatment between them[7,8].

Direct CCFs have a different hemodynamic change compared to the dural fistulas. After the establishment of the arteriovenous fistula the blood pressure and flow are transmitted to the cavernous sinus. This causes reverse flow and venous dilation of the veins that drain into the sinus, as well as an increased flow in their usual draining veins. When they reverse their venous flow the ophthalmic veins cause engorgement and orbital congestion that prevents drainage of the aqueous humor and increases intraocular pressure and secondary glaucoma. The elevated intraocular pressure may compromise retina perfusion and result in decreased visual acuity, a surgical emergency. Eye oedema resulting from the impairment of venous drainage may lead to mechanical limitations of the extraocular muscles which may already have their function compromised by compression on the cranial nerves[10,11,12].

The intercavernous sinuses can transmit the same hemodynamic changes, to the other side resulting in a similar clinical picture.

The result of blood flow reversal and increased venous pressure in the ophthalmic veins can result in exophthalmos, ophthalmic fremitus, chemosis, proptosis, and diplopia. Loss of vision occurs in about 90% of direct CCFs and 20 to 30% of indirect CCFs.

Glaucoma and retinal perforation are symptoms of direct CCF and may appear days or weeks after the TBI. Ischemia involving cranial nerves III, IV, V and VI may be premature and evident due to mass effect on the cavernous sinus. This is different from the indirect CCF that has slow evolution and usually manifests with ocular hyperaemia and tortuous arterializations of the conjunctiva, without proptosis. After angiography 20 to 50% of indirect fistulas close spontaneously[13].

The diagnosis of Carotid Cavernous Fistulas is based on clinical history, physical examination and radiological examination.

The gold standard complementary diagnostic method is the selective digital arteriography of the carotid artery. This exam shows the size and estimates the damage of the fistula. The tests that have a complementary role in diagnosis are magnetic resonance, computed tomography angiography and Transcranial Doppler[14,15,16].

During differential diagnosis it is important to consider cerebral aneurysms, vascular malfunctions of the eye, orbital inflammations, retro-orbital cellulitis, exophthalmos secondary to thyroid disease, retrobulbar haemorrhage, neoplasm of lacrimal gland, cavernous sinus thembosis and vasculitis[17]. To ensure proper diagnosis we emphasize the importance of multidisciplinary examination of the eye function and periorbital condition.

Carotid Cavernous Fistula treatment has become more effective, safer and less invasive with the advent of new neuroimaging techniques.

The positive results obtained with endovascular techniques using detachable balloons have made embolization the treatment of choice for direct CCF, allowing occlusion of fistulas without ligation of the vessel holder (internal carotid artery) in 80% of cases. When the size of the fistula is too large or incompatible with the size of the balloon, the use of electrodetachable coils decreases the size of the hole allowing for the occlusion of the remaining fistula.[19]. Radiological examination of the skull is necessary after embolization to control the location of the balloon.

Liang found significant differences between the time of the first symptoms and embolization by comparing groups with and without complications[20]. Collaborating that with early detection and immediate intervention it is possible to accomplish full visual recovery with proper treatment[21,22].

Intravenous treatment has been recommended for indirect CCF. This is a safer treatment with less arterial thrombotic events, lower rate of treatment failure and easier implementation. The most common complications observed in the treatment of CCF by embolization are the migration of the balloon, formation of a false aneurysm, on the internal carotid (which usually recedes spontaneously), perforation of the superior ophthalmic vein, orbital haemorrhage and injury of the abducens and trochlear nerves. Post treatment recurrences and deaths have been reported[23].

Conclusion

Although rare, CCF is a serious complication that should be considered in the differential diagnosis of ocular proptosis and visual loss associated with maxillofacial trauma.

Consent

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

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