

Persistent Post-Traumatic Orbital Cerebrospinal Fluid Fistula with Multiple Episodes of Meningitis: Systematic Case Review and Case Report

CASE REPORT

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Abstract

Background: Orbital cerebrospinal fluid (CSF) fistulas are very rare. Most of these fistulas spontaneously heal within a couple of days and usually do not require surgical repair. Furthermore, they present low rates of complications.

Case presentation: We report a case of a 15-year-old boy that suffered a penetrating head trauma and developed multiple episodes of meningitis due to a persistent orbital and nasal CSF fistula. He underwent multiple different procedures to correct the dural breach: sphenoidal endoscopic approach, and three craniotomies, the last one succeeded with the placement of autologous grafts and biological glue. A lumbar-subarachnoidal peritoneal shunt was placed to aid for 5 days. The CSF fistula ceased to drain and he recovered of the meningitis with additional standard antibiotics.

Review: A systematic review in MedLine, Embase and Lilacs, identified 15 reported cases of persistent orbital CSF fistulas post-trauma. Traffic accidents were the most common mechanism of trauma. Most cases were reported in young boys and the main clinical manifestation was persistent "oculorrhea". The CSF leaks were traced with CT-scans and were predominantly managed surgically. No fatal outcomes were reported.

Conclusion: The reported cases raise awareness for persistent orbital CSF fistulas. Early diagnosis and adequate management is crucial to prevent complications, which are rare, but can be highly morbid and fatal.

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Keywords

CSF Fistula; CSF Leak; TBI; Complications; Meningitis; Adolescent; Oculorrhea.

Introduction

Cerebrospinal fluid (CSF) fistulas are a well-known complication of head trauma [1]. These fistulas form communications between the subarachnoid space and an internal or external cavity. They usually are a result of fractures and dural tearing in the skull base [2], causing CSF to leak through the nasal, middle ear or orbital cavity [18]. Nasal and middle ear fistulas are a lot more common and are justified by the physical features inherent to these cavities [1, 18, 7]. Orbital CSF fistulas are very rare [2, 16] and manifest as orbitocele, blepharocoele [7], chemosis, or excessive tearing (“oculorrhea”) [13, 16]. Most of these fistulas spontaneously heal within a couple of days and usually do not require surgical repair. Furthermore, they present low rates of complications [6, 11]. However, rare cases of persisting fistulas are reported, which require surgical management with dural reconstruction, especially when associated to complications.

We report a case of a teenage boy, who developed posttraumatic orbital and nasal CSF fistulas. These were only diagnosed after the presentation of multiple episodes of bacterial meningitis and delayed manifestations of oculorrhea and rhinorrhea. The boy underwent multiple surgical procedures for dural repair.

Case Report

A 15-year-old male suffered a penetrating head trauma while hunting in the woods. As he fired his old shotgun, the handle expelled a thin piece of wood right through the epicanthic fold of his left eye (in a left to right direction). He did not lose consciousness. His brother removed the piece of wood and took him to an emergency unit for medical evaluation. The patient had an unremarkable medical history and he was not taking any prescription medications.

He presented no signs of respiratory distress, his blood pressure was normal; pulse was stable, and

no signs of external bleeding. He was alert and oriented, times three (to person, place, and time), cooperative, and obtained a score of 15 on the Glasgow coma scale. His left eye presented conjunctival hemorrhage and a discretely swollen periorbital area. His pupils were isochoric with a reduced direct photomotor reflex on the right eye and consensual photomotor reflex in the left eye. The patient referred blindness in the right eye. His initial CT scans showed evidence of a short linear fracture in the medial wall of the left orbital cavity and through the ethmoidal bone (**Figure 1**). There were no intracranial hemorrhages and no signs of dural tear or other fractures. The patient stayed hospitalized for observation for 5 days. He remained clinically stable and was discharged with no complains and a good performance status of 0, except for his right eye blindness.

Figure 1: CT scan in axial cuts, which evidences the fracture and tracks the path of the penetrating wooden foreign body. In A, you can see the track and the edema caused by the penetrating injury. In B, an arrow demonstrates the path the foreign body traced through the medial wall of the left orbital cavity and nasal cavity, crossing the middle line through the ethmoidal cells penetrating in the skull base at the parasellar region of the sphenoid causing a lesion to the contralateral optic nerve.

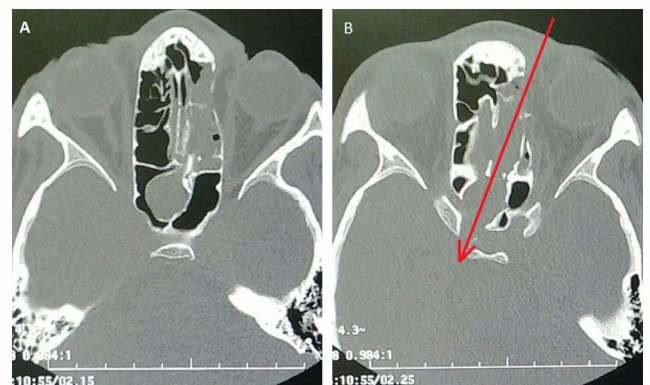
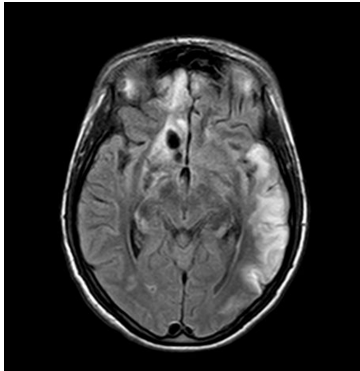


Figure 2: MRI in T1 weighted evidencing the left temporal meningoencephalitis.



Six days post-discharge, the patient returned to the emergency unit with headache, fever, photophobia, neck stiffness, seizures, and discrete intermittent rhinorrhea. The patient was diagnosed with bacterial meningitis and was transferred to the Hospital Universitário João de Barros Barreto Medical Center for infectious diseases. He began treatment with standard antibiotic therapy for bacterial meningitis. Despite the rapid diagnosis and institution of appropriate antibiotic treatment, he did not respond very well. He developed progressive right hemiparesis and impaired level of consciousness. His MRI scans showed a hyperintense area in the left temporal lobe, suggesting meningoencephalitis (**Figure 2**). Over the course of two weeks, he presented recurrent meningitis, which progressively disabled him.

A neurosurgical consult was requested to evaluate the persistent rhinorrhea and the recent appearance of a clear liquid drainage through the conjunctiva of the left eye. A post-traumatic CSF leak was suspected and the drained liquid was tested for beta-2-transferin. The results came back positive, confirming the CSF leak. For surgical planning, the patient also underwent a metrizamide CT cisternography that trailed the CSF fistulas towards the nasal and orbital cavity.

At first, the neurosurgical team in cooperation of the ENT team attempted a less invasive approach to correct the dural breach. Therefore, the patient

was submitted through an intranasal endoscopic transsphenoidal surgery and synthetic dural patches were placed around the fractured area. The procedure was uneventful, but did not show satisfactory results.

Given the persistence and associated infectious complications, the patient underwent surgical management for dural repair with placement of synthetic dural patches in attempt to cease the CSF leaks. The procedure was uneventful, but did not stop the CSF leaks. Postoperatively the patient worsened clinically and evolved with Broca's aphasia and meningoencephalitis together with an incisional empyema. Hence, he was taken back to the OR for surgical drainage. His neurological deficits kept worsening; therefore, a more radical approach was necessary to repair the dural tear.

The case was discussed in neurosurgical rounds and the board decided to do another transcranial approach as soon as he reached the minimal clinical conditions. The board suggested to retrieve autologous grafts of muscle, fascia lata and adipose tissue of the patient's left leg. These grafts added by surgical glue (N-Butyl-Cyanoacrylate) were used to line the anterior base of the skull bilaterally. The procedure was uneventful and post-operatively the patient ceased to drain CSF through the nasal and orbital cavity. A lumbar-subarachnoidal peritoneal shunt was placed to aid for 5 days. He continued on antibiotics and did not develop any new episode of meningitis after surgery. He was discharged with performance status 3, right eye blindness and right hemiparesis.

Discussion

Penetrating head injuries can cause serious damages even through a small wound entry [14, 17]. CSF fistulas occur when a penetrating head injury causes a skull fracture and breaches the underlying layers of meninges [4]. Despite this being a common complication, described in 15 to 20 % of head injuries

Table 1. Systematic review based on the MOOSE guidelines in MedLine, Embase and Lilacs using various combinations of the terms "orbital CSF fistula", "cranio-orbital fistula", "posttraumatic", "tbi". 36 studies were included for further analysis and 15 met the inclusion criteria for this review [3, 16, 13, 12, 20, 10, 22, 18, 2, 8, 23, 17, 7, 5, 1].

#	Author, Year	Gender/ Age*	Mechanism	Clinical manifestation	Complication	Diagnosis	Treatment
1	Bagolini, 1957	N/I/9m	Car accident	Oculorrhea + Hematoma of upper eyelid + Anisocoric	N/I	N/I	Surgical
2	Joshi, 1978	F/8m	Car accident	Rhinorrhea + Oculorrhea	-	Dextrostix test + CT scan	Transcranial surgery + Dural repair
3	Garza-Mercado, 1982	M/20	Assault	Oculorrhea + Edema of Eyelids + Ecchymosis + Limited EOM	N/I	N/I	Surgical
4	Sibony, 1985	M/27	Car accident	Orbitocele + Proptosis	Cerebral abscess + Phlebitis	Cisternography	Conservative
5	Dryden, 1986	M/4	Car accident	Oculorrhea for 2 years	Meningitis	Dextrostix test + CT scan	Transcranial surgery + Silastic implant
6	Till, 1987	M/14m	Stabbing wound	Oculorrhea	N/I	Dextrostix test + CT scan	Surgical
7	Salame, 2000	F/20	Sport injury	Oculorrhea	N/I	Physical exam + Glucose test	Conservative
8	Arslantas, 2003	N/I/3	Fall	Orbitocele	N/I	CT scan + MRI	Transcranial surgery + Dural repair
9	Civelek, 2006	M/7	Occupational accident	Proptosis + Diplopia + Periorbital abscess + Orbital cellulitis + Oculorrhea	N/I	N/I	Surgical
10	Twaij, 2009	M/3	Penetrating orbital injury	Orbitocele + Proptosis + Periorbital edema	Reduced visual acuity	MRI	Conservative
11	Pereira, 2011	F/7m	Gunshot injury	Luxation of globe + Oculorrhea + Ecchymosis + Eyelid hematoma	Blindness	CT scan + surgical exploration	Transcranial surgery + Sponge block + Organic glue
12	Chandra, 2013	F/4	Blunt head injury	Upper eyelid swelling	N/I	CT scan	Transcranial surgery + Dural repair
13	Pease, 2013	M/22	Car accident	Oculorrhea + Limited EOM	-	CT scan	Transcranial surgery + Skull base reconstruction
14	Borumandi, 2013	F/49	Fall	Eyelid swelling + Ecchymosis	N/I	CT scan + MRI	Conservative
15	Apkarian, 2014	M/34	Car accident	Periorbital swelling + Periorbital pain	N/I	CT scan + Beta-2-transferrin test	Conservative

*Age is described in years (y) or in months (m). N/I means "not informed". EOM means extra-ocular movements. Some reports only mentioned a surgical or conservative approach, not explaining in details the adopted treatment measures.

[15], orbital CSF fistulas are rare [21, 18]. A systematic review found 15 reported cases (**Table 1**). The rareness of this presentation is justified by protective anatomical characteristics, including: (a) the usually intact conjunctiva, unfavorable for the formation of a CSF exit. (b) the sinus walls, which are thinner than the orbital walls and, therefore, easier to fracture based on the principle of least resistance [16, 4]. (c) the orbital cavity, which is considered a virtual space with a zeroed internal pressure [18], main reason the orbital CSF leaks are asymptomatic or course to a spontaneous resolution [21].

Based on the speed of the penetrating head injury, these can be classified in high and low speed injuries [14]. This case describes an injury caused by a high speed and low temperature foreign object (wood), which caused the fracture in the left orbital cavity, crossed the ethmoidal sinus in the midline and immediately damaged the right optic nerve. Low temperature injuries are more prone to local infections, as reported in this case, different from high speed and high temperature injuries, such as gunshot wounds. Penetrating injuries by wooden foreign objects carry a higher risk of infection [17].

Penetrating wounds require thorough investigation and for this, the follow recommendations are important: never underestimate the internal damage; do not pull out the foreign object without medical support, such careless removal can induce bleeding and lead to fatal consequences [14]. The brother of our patient inadvertently removed the foreign object and could have worsened the damages of the injury. Moreover, the third recommendation is that no part of the object should remain in the injured tissue. The remnant of foreign objects in penetrating head injuries can lead to post-traumatic complications like meningitis and brain abscess. This case described a penetrating head trauma with a wooden foreign object which is a rare cause of orbital lesion [17]. Wooden foreign objects are hard to be seen by CT-scans and therefore difficult to evaluate [14].

Any patient with a head injury and excessive drainage through the orbital conjunctiva or excessive "tearing" should be suspected to have CSF oculo-rhea, especially when concomitant with rhinorrhea [13]. Nonetheless, these clinical presentations can go unrecognized [16], as most of the fistulas resolve spontaneously in 24 to 48 hours. The fistula closes by the adhesion of blood products and inflammatory cells at the site of the breach, herniation of the brain tissue into the traumatic defect [16], or by the proliferation of fibrous tissue or mucosa over the laceration [15].

When a post-traumatic fistula is suspected, CT-scans can be useful to detect the bone fractures that caused the CSF leak. To confirm the presence of the CSF leak, the output liquid can be tested for glucose levels above 30 mg/ml or for the presence of beta-2-transferrin. Some authors suggest to routinely test the lacrimal and nasal secretions of patients with anterior fossa fractures in high-energy traumas, especially in infants or young adults [13]. For surgical planning, some surgeons can request a CT-cisternography (metrizamide) to track the fistula [20].

After identifying the leak, the goals of therapy for fractures of the skull base include the prevention of intracranial infections, decompression of intracranial nerves, and if necessary, repair of vessel compression [15]. If the leak is not clinically evident and no other major injuries are present, prophylactic surgery is not recommended [19]. The administration of prophylactic antibiotic therapy remains controversial but helps to decrease the morbidity and mortality caused by skull base fractures [15]. Conservative treatment is the standard approach when the leak ceases spontaneously within a week, or when the fracture is small or not visible. This conservative approach includes measures to prevent intracranial hypertension and CSF drainage. If the orbital CSF leak does not fulfil the above-mentioned requirements for conservative treatment, a surgical repair can be conducted via a transcranial or an endona-

sal approach [15]. For the surgical plan, the dural breach and fistula need to be identified. The dural breach can be corrected with sponges, dural grafts, fibrin or organic glue, reinforced by sutures [17,15]. In case the dural tear is not identifiable, a layer of muscle plug and grafts of fascia can be placed over an extended suspected area to seal the fistula [9]. In our case, we lined the anterior base of the skull with surgical glue, dural graft, and autologous tissue from the fascia lata, muscle and adipose layer, which showed to be effective in closing the dural breach, leading to its cure.

Conclusion

It is important to early diagnose and treat persistent orbital CSF fistulas. These fistulas are diagnosed by a suggestive clinical presentation (excessive tearing), CT-scans, lacrimal secretion testing and by CT-cisternography with metrizamide. Associated complications are rare, but when present can be highly morbid. This case report demonstrates a different surgical strategy to approach persistent CSF fistulas. Therefore, we emphasize that the awareness and fundamental understanding of the lesion is essential to diagnose the condition for successful management.

Patient consent

The patient's legal guardian signed a consent form approving the submission of this case to the journal. The Ethics Board of the Ophir Loyola Hospital approved our report.

Acknowledgements

The authors would like to thank the research group "Grupo de Ensino e Pesquisa em Doenças Cerebrovasculares/UFGA" for the support in compiling this paper.

Conflicts of interest

The authors declare that they have no conflict of interest.

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