



Delayed Posttraumatic Blepharocoele: A Rare Case Report with Review of the Literature

Blefarocoele pós-traumática tardia: relato de caso raro com revisão da literatura

Amey P. Patankar¹ Shivani Chaudhary²

¹ Department of Neurosurgery, Baroda Medical College and SSG Hospital, Vadodara, Gujarat, India

² Department of Surgery, Baroda Medical College and SSG Hospital, Vadodara, Gujarat, India

Address for correspondence Amey P. Patankar, MBBS, MS, MCh, 703 Rajarshi Darshan Tower, Near Jalaram Mandir, Karelilbag, Vadodara, Gujarat, 390018, India (e-mail: docapp@icloud.com).

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Abstract

Though posttraumatic cerebrospinal fluid (CSF) rhinorrhea and otorrhea are fairly common, blepharocoele and blepharocoele are rare, with only 15 cases reported to date.

Keywords

- blepharocoele
- craniopalpebral fistula
- orbital encephalocoele
- blepharocoele-phalocoele

A 29-year-old female patient presented with a complaint of swelling of the right eyelid that had begun three months before. The patient had sustained a head injury 24 years prior to presentation.

Imaging studies revealed the presence of a craniopalpebral CSF fistula. The patient underwent successful surgical repair of the fistula with craniotomy and duroplasty by autologous fascia lata graft.

Delayed development of blepharocoele 24 years after trauma is unusual, and, to our knowledge, the case herein reported is the first one in the literature.

Resumo

Embora a rinorreia e a otorreia pós-traumática do líquido cefalorraquidiano (LCR) sejam bastante comuns, a blefarocoele e a blefarocoele são raras, com apenas 15 casos relatados até o momento.

Paciente do sexo feminino, 29 anos, apresentou queixa de edema em pálpebra direita com início há três meses. O paciente havia sofrido um traumatismo cranioencefálico 24 anos antes da apresentação.

Os exames de imagem revelaram a presença de fístula líquórica craniopalpebral. O paciente foi submetido com sucesso ao reparo cirúrgico da fístula com craniotomia e duroplastia com enxerto autólogo de fâscia lata.

O atraso no desenvolvimento de blefarocoele 24 anos após o trauma é incomum e, até onde sabemos, o caso aqui relatado é o primeiro na literatura.

Palavras-chave

- blefarocoele
- fístula craniopalpebral
- encefalocoele orbital
- blefarocoele

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Introduction

Blepharocoele is herniation of cerebrospinal fluid (CSF) into the eyelid. It is a rare condition, with only 15 cases reported to date.¹⁻¹⁵ It is usually traumatic in origin,¹⁻¹³ and very rarely congenital, without any history of trauma.^{14,15} Traumatic blepharocoele develops as a result of the breaching of an orbital bone fracture in the dura, leading to the formation of a craniopalpebral CSF fistula.

Case Presentation

A 29-year-old female presented with a complaint of swelling in the right eyelid that had begun three months before. The swelling appeared in the morning, upon waking up from sleep (►Fig. 1), and it gradually subsided as the day passed (►Fig. 2), only to reappear the next morning. The swelling was soft, with positive transillumination. The patient did not have any recent history of trauma or surgical procedures but had sustained a head injury 24 years before at the age of 5 years, for which she was managed conservatively. No radiological investigations were performed at the time of the injury.

A magnetic resonance imaging (MRI) scan of the orbit revealed a craniopalpebral CSF fistula in the right orbit (►Fig. 3). A computed tomography (CT) scan of the orbit showed a defect on the roof of the right orbit with irregular raised margins, suggestive of an old fracture (►Fig. 4).

Fundus examination and CSF manometry were performed to rule out raised intracranial pressure. The opening pressure

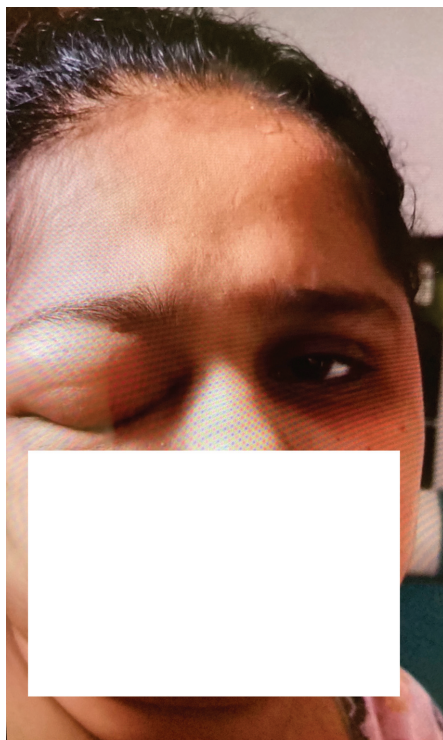


Fig. 1 Preoperative photograph of the patient in the morning showing right eyelid swelling.



Fig. 2 Preoperative photograph of the patient in the evening showing complete resolution of the right eyelid swelling.

of the CSF was of 12 cm of water, and the fundus examination was normal, without any signs of papilledema.

The patient underwent surgical repair of the craniopalpebral CSF fistula under general anesthesia in the supine position. A right frontal craniotomy with an intradural sub-frontal approach revealed that the gliotic brain tissue was adherent to the bony defect of the roof of the orbit. The gliotic brain tissue was separated from the bony defect, and the margins of the defect were defined. The irregular and raised bony edges of the roof of the orbit were flattened. The defect was repaired by autologous fascia lata graft, which was

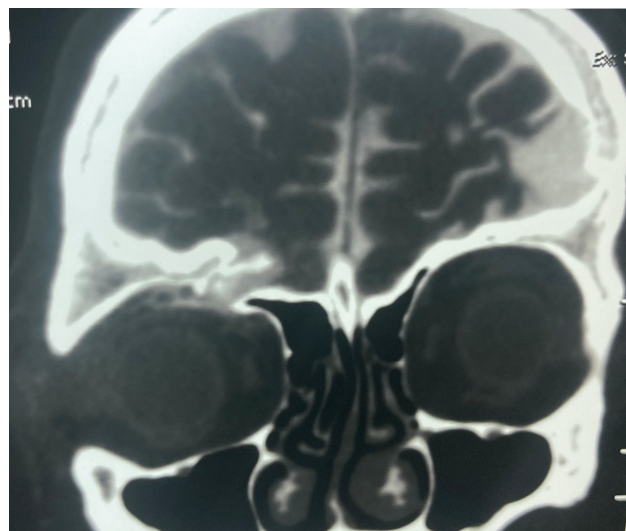


Fig. 3 T2-weighted magnetic resonance imaging scan of the brain in coronal view showing the craniopalpebral fistula.



Fig. 4 Computed tomography scan of the brain and orbit showing the bony defect with irregular margins on the roof of the orbit.

anchored to the dura of the skull base by 2 stitches of 4-0 Vicryl suture (Ethicon, Inc., Raritan, NJ, United States). The postoperative course was uneventful, and in the early morning of the first postoperative day the eyelid swelling had disappeared (→**Fig. 5**). On the follow-up after 6 months, the patient remained asymptomatic (→**Fig. 6**)

Discussion

Cerebrospinal fluid fistulas complicate ~ 2% of all head injuries and ~ 12% to 30% of all skull base fractures;¹¹ CSF



Fig. 5 Postoperative photograph of the patient in the morning of first postoperative day showing no eyelid swelling.



Fig. 6 Postoperative photograph of the patient six months after surgery showing no eyelid swelling.

rhinorrhea and otorrhea are the most common forms of CSF fistulas after a head injury.

Leakage of the CSF into the orbit (orbital encephalocele) is rare, and it can manifest as pseudolacrimation (CSF oculor-rhea).^{16–19} The rarity of craniopalpebral fistulas is due to the fact that the orbital walls are thicker and less fragile as compared with the thin bones of the frontal and ethmoidal sinuses and the cribriform plates.⁴ Craniopalpebral fistulas are more commonly reported in children,^{1,5,6,8,10,11,13,20} probably because their orbital walls are thinner. Additionally, frontal sinus agenesis has been hypothesized as one of the factors for the development of blepharoenkephalocele.^{2,9} Absence of the frontal sinus may enable the direct passage of CSF into the upper eyelid following a head injury. In the case herein reported, the frontal sinus was well developed (→**Fig. 4**).

The patient had right eyelid swelling, which was more intense the morning and gradually subsided as the day progressed. This is because the intracranial CSF pressure rises during the night in supine position, leading to more CSF egress into the eyelid through the fistulous tract. During the day, because of the upright posture, the decrease in intracranial pressure caused an outflow of CSF from the eyelid, leading to the disappearance of the swelling.

How the head injury sustained 24 years before led to blepharoenkephalocele is a matter of conjecture. It is unlikely that the defect was congenital, because its margins were irregular, rough and with raised edges, suggestive of callus formation after trauma. It is possible that the patient had sustained a fracture of the thin orbital roof at the time of the head injury, with entrapment of brain matter into the fracture. The pulsatile brain matter gradually eroded the periorbita,

leading to leakage of CSF into the upper eyelid. The case herein reported may be considered one of a small “internal growing skull fracture”, which was not detected for 24 long years, as it was hidden from the external environment and not causing any orbital compression.

Diagnosis in this case was difficult, because craniopalpebral fistulas usually present within one to three months of trauma. Aspiration of CSF from the eyelid in such cases is strictly contraindicated, as it may cause meningitis. The disappearance of the swelling during the daytime and the history of trauma, though remote, provided a clue to the diagnosis, which was confirmed by imaging studies. The CSF manometry and fundus examination ruled out benign intracranial hypertension.

Though successful healing of the fistula by conservative means has been reported,^{2,7,12} the treatment of this condition is almost always surgical, with repair of the fistula by pericranial or fascia lata grafts. Additional skull base repair by titanium mesh may be required in cases of large skull base defects.

Conclusion

Blepharocoele, or craniopalpebral fistula, is rare, with only 15 cases reported to date. Cranioorbital and craniopalpebral fistulas should be suspected in patients with orbital fractures, in whom the posttraumatic orbital swelling or proptosis fails to resolve in two to three weeks. To the best of our knowledge, delayed presentation of blepharocoele, 24 years after a head injury, has not been reported to date, this being the first such case.

Conflict of Interests

The authors have no conflict of interests to declare.

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