

ISSN: 2340-3438

Edita: Sociedad Gallega de
Otorrinolaringología.

Periodicidad: continuada.

Web: [www: sgorl.org/revista](http://www.sgorl.org/revista)

Correo electrónico:

actaorlgallega@gmail.com

SGORL PCF
Sociedad Gallega de Otorrinolaringología
y Patología Cervicofacial



Acta Otorrinolaringológica

Caso Clínico

Surgical management of traumatic cerebrospinal fluid fistula in a child with recurrent meningitis

Tratamento cirúrgico de fistula traumática de líquido cefalorraquidiano em criança com meningite recorrente

Lília Ferraria, Sílvia Alves, Nélia Ferraria, Mário Santos, Maria Helena
Rosa, Luis Antunes

Hospital Garcia de Orta

Recibido: 7/4/2018 Aceptado: 27/5/2018

Abstract

Introduction: The diagnosis and management of pediatric post-traumatic CSF leak can be challenging. Untreated leaks can be manifested by intermittent rhinorrhea or recurrent meningitis, which can be the only form of presentation. To report a case of a child with recurrent meningitis in which cerebrospinal fluid (CSF) leak diagnosis was established, treated by an extracranial approach.

Clinical Case: A 15-year-old girl had a past medical history of a fall from the second floor at the age of 8, that resulted in traumatic frontal sinus fracture. She had her first episode of pneumococcal meningitis at the age of 14 that lead to the diagnosis of post-traumatic CSF fistula. External surgery was performed to repair the fistula, with no evidence of CSF rhinorrhea on post-operative examination. She was readmitted at the age of 15-years-old for a new episode of pneumococcal meningitis. The high resolution computerized tomography scan revealed an anterior and posterior wall defect of the right frontal sinus. An extracranial approach was performed, with visualization of CSF trickling through the defect. Dural regeneration matrix and fascial graft were positioned on the defect and fibrin glue was used. Frontal sinus obliteration was performed using hydroxyapatite, with success.

Correspondencia: Lília Ferraria

Hospital Garcia de Orta

Correo electrónico: liliaferraria@gmail.com

Discussion: The basic principles of pediatric CSF fistulas repair, such as an adequate exposure of the defect, do not differ from those of the adults. The successful repair of CSF leaks results of a preoperative correct identification of the fistula and bone defect site, an accurate knowledge of reconstruction techniques and their judicious application in the procedure planning.

Keywords: Cerebrospinal fluid leaks, Recurrent meningitis, Pediatric trauma, Skull base defects, Extracranial approach

Resumo

Introdução: O diagnóstico e tratamento de fístulas de líquido cefalorraquiano (LCR) pós-traumáticas em crianças pode ser desafiador. Fístulas não tratadas podem-se manifestar por rinorreia intermitente ou meningite recorrente, que pode ser a única forma de apresentação. Este artigo tem como objetivo relatar um caso de uma criança com meningite recorrente em que o diagnóstico de fistula de LCR foi estabelecido e tratado por uma abordagem extracraniana.

Caso clínico: Adolescente de 15 anos com antecedentes de queda do segundo andar aos 8 anos de idade, de que resultou fratura traumática do seio frontal. O primeiro episódio de meningite pneumocócica observou-se aos 14 anos, o que levou ao diagnóstico de fistula de LCR pós-traumática. Foi submetida a intervenção cirúrgica extracraniana para correção da fistula, sem evidência de rinorráquia na avaliação pós-operatória. Aos 15 anos foi readmitida por um novo episódio de meningite pneumocócica. A tomografia computadorizada de alta resolução revelou um defeito da parede anterior e posterior do seio frontal direito. Foi realizada uma abordagem extracraniana, com visualização da fistula de LCR. Foi colocado matriz de regeneração da duramáter e fascia liofilizada no defeito e utilizado cola de fibrina. Obliterou-se com sucesso o seio frontal com hidroxiapatite.

Discussão: Os princípios básicos da correção de fístulas de LCR em idade pediátrica não diferem dos adultos. O tratamento bem sucedido das fístulas de LCR resulta de uma correta identificação pré-operatória do local da fistula e do defeito ósseo, de um conhecimento rigoroso das técnicas de reconstrução e da sua aplicação criteriosa no planeamento do procedimento.

Palavras-chave: Fístula de LCR, Meningite recorrente, Traumatismo pediátrico, Defeitos na base do crânio, Abordagem extracraniana

Introduction

A cerebrospinal fluid (CSF) fistula is a result of meningeal dural and arachnoid laceration with fistula formation, which may be spontaneous, secondary to a skull base fracture, related to surgical trauma or associated with pathology of the skull base and/or secondary to a high-pressure system.¹

The main cause of CSF fistula is secondary to a skull base fracture in 70%-80% of the cases and most commonly originate from the cribriform plate, the fovea ethmoidalis and the posterior wall of the frontal sinus or sphenoid sinus. CSF leaks appear in 6%-13% of all cranial traumas involving the anterior skull

base.¹ While penetrating head trauma is an obvious cause of both transient and persistent CSF fistula, most traumatic CSF leaks occur as a result of blunt trauma, because of the higher incidence of blunt head trauma overall. Posttraumatic cerebrospinal fluid (CSF) leaks in children are less common than in adults and occur in an incidence of 0,2–0,3% following childhood head trauma. This may be explained by the greater flexibility of skull bones that can lead to absorption of the traumatic shock.¹

Clinically, posttraumatic CSF leaks can be manifested by intermittent rhinorrhea or recurrent meningitis, which can be the only form of presentation.² Meningitis may develop acutely or even decades after the trauma.^{3,4} Overall, between 7% and 30% of all patients with posttraumatic CSF leakage will develop meningitis—this rate increases as the duration of CSF leakage increases.⁵

The diagnosis of a CSF leak after closed cranial trauma is based on the clinical evaluation, the immunofixation of beta-2-transferrin of the nasal drainage, the visual identification of a fistula and/or skull base defect (endoscopy), and its confirmation with a computerized tomography (CT) scan. The site of any defect should be defined using high-resolution coronal CT. This exam is the preferred test to detect defects at the cranial base.³

Most traumatic CSF leaks resolve spontaneously, the majority within the first 24–48 hours. Traumatic CSF leaks—as opposed to spontaneous ones—tend to stop spontaneously as a result of blood products and/or inflammatory adhesions at the site of the dural breach and associated skull fracture. Herniation of brain tissue into the traumatic defect may also play a role in leak cessation. However, persistent fistula do occur.⁵

Only a persistent CSF fistula becomes a significant management issue for the surgeon. The appropriate timing for surgical intervention in persistent posttraumatic CSF leakage is also controversial. What is clear is that many approaches can be successful, both intracranial and extracranial, depending on patient factors and the anatomy of the fistula.⁵ Recent studies indicate that the cumulative risk of developing meningitis within 10 years of a trauma is reduced from 85% to 7% when the surgery is successful. Consequently, there is general agreement that head-injured patients who present with enlarging pneumocephalus, persistent CSF leaks, or meningitis need urgent surgical repair.⁴

Clinical Case

A 15-year-old girl had a past medical history of a fall from the second floor at the age of 8, that resulted in a traumatic frontal sinus fracture. As there was no evidence of CSF rhinorrhea, she was not submitted to surgery at the time. Six years later, she was admitted to a pediatric hospital for an episode of pneumococcal meningitis and that leads to the diagnosis of CSF fistula due to the frontal sinus fracture. External surgery was performed in that hospital, to repair the fistula. No evidence of CSF rhinorrhea on post-operative period was reported.

She was readmitted one year later, at the age of 15 years old, for a new episode of pneumococcal meningitis. She was transferred to our hospital and the Otorhinolaryngology department was consulted. Clinically, there was no evidence of CSF rhinorrhea and through nasal endoscopy, we didn't localize the site of the leak. Therefore, a high resolution CT scan of the paranasal sinuses was requested. This exam revealed an

anterior and posterior wall defect of the right frontal sinus (Figure 1a and 1b). A fracture line in the posterior wall of frontal sinus created a communication with the intracranial space.

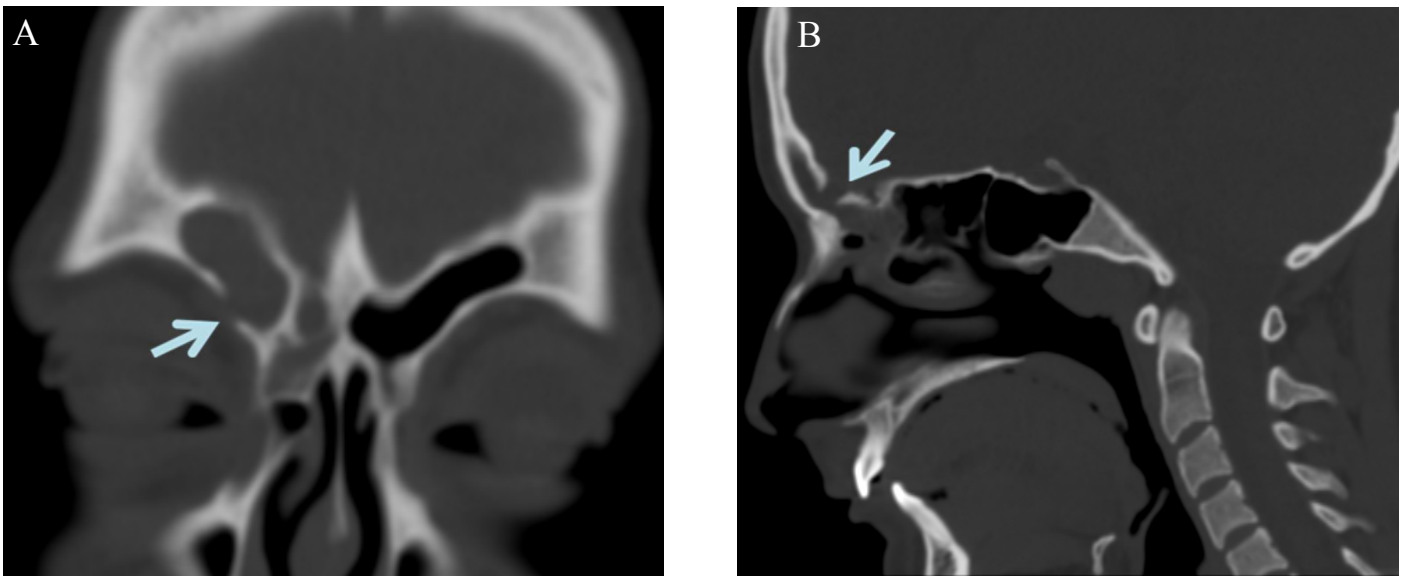


Figure 1a: Coronal CT scan shows an anterior wall defect of the right frontal sinus (arrow). Figure 1b: Sagittal CT scan shows a posterior wall defect of the right frontal sinus (arrow) that creates a communication with the intracranial space.

The child was treated by surgery under general anesthesia with endotracheal intubation. An extracranial approach via an eyebrow incision (Lynch-Howarth) was performed. The anterior wall of the frontal sinus was drilled with a cutting burr (Figure 2a). Meticulous attention was directed to the posterior table where a CSF trickling was visualized (Figure 2b). The mucosa and inner periosteum were removed with a diamond drill. The exposed dura on the posterior wall of the frontal sinus was covered by a layer of dural regeneration matrix and a synthetic fascial graft – onlay technique on the defect – with fibrin glue (Figure 3a). Separation of the frontal from the lower paranasal sinuses was achieved by placement of another synthetic fascial graft on the frontal recess for permanent occlusion. Frontal sinus obliteration was performed using hydroxyapatite cement (Figure 3b).

Postoperatively, she had lumbar drain for 5 days, antibiotic therapy with ceftriaxone for 7 days and bedrest with elevated head. She was discharged from the hospital 5 days postoperatively and a regular follow-up was performed (Figure 4).

Examination 22 months after surgery, revealed a well-healed incision on the right eyebrow and no evidence of CSF rhinorrhea. She reported no further episodes of meningitis.

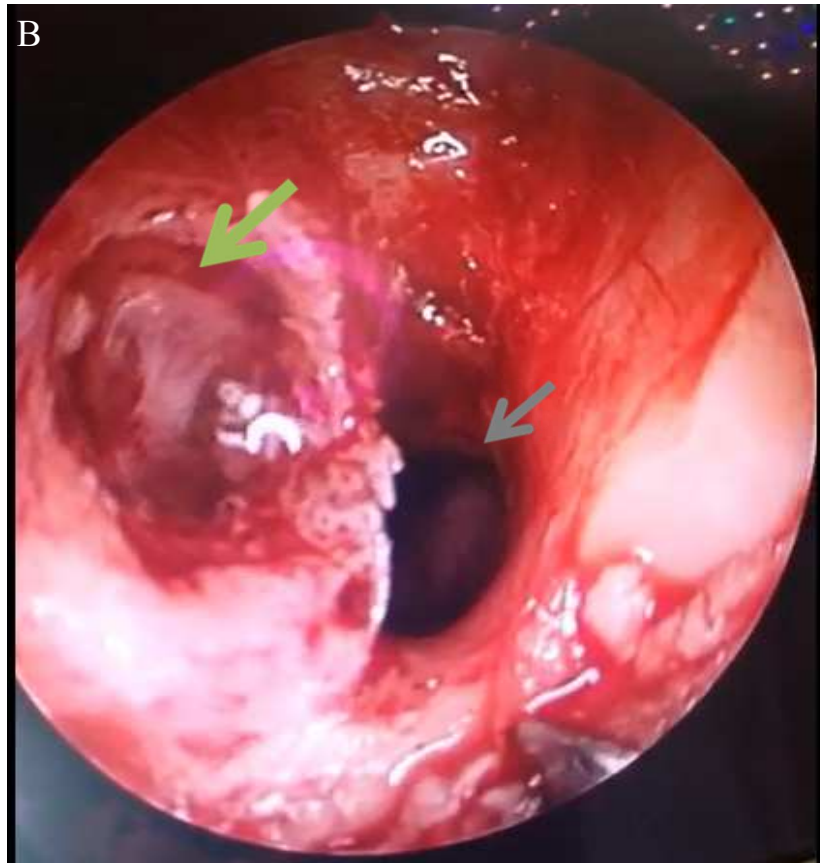


Figure 2a: An extracranial approach via an eyebrow incision (Lynch-Howarth). The anterior wall of the frontal sinus was drilled. Note the posterior table where a CSF trickling is visualized.
Figure 2b: Operative endoscopic view inside the frontal sinus – fistula site (green arrow) is identified with the CSF coming out from the posterior wall of the frontal sinus; frontal recess (grey arrow).

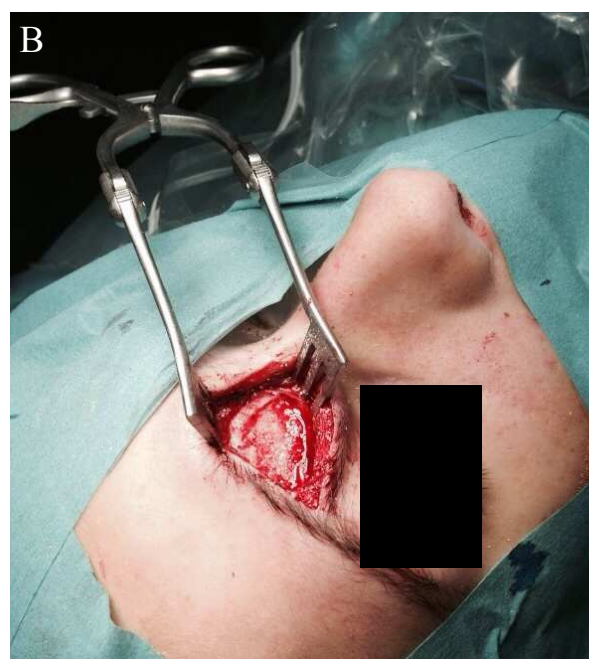


Figure 3a: The exposed dura on the posterior wall of the frontal sinus was covered by a layer of dural regeneration matrix and a synthetic fascial graft – onlay technique on the defect. Figure 3b: Frontal sinus obliteration using hydroxyapatite cement.

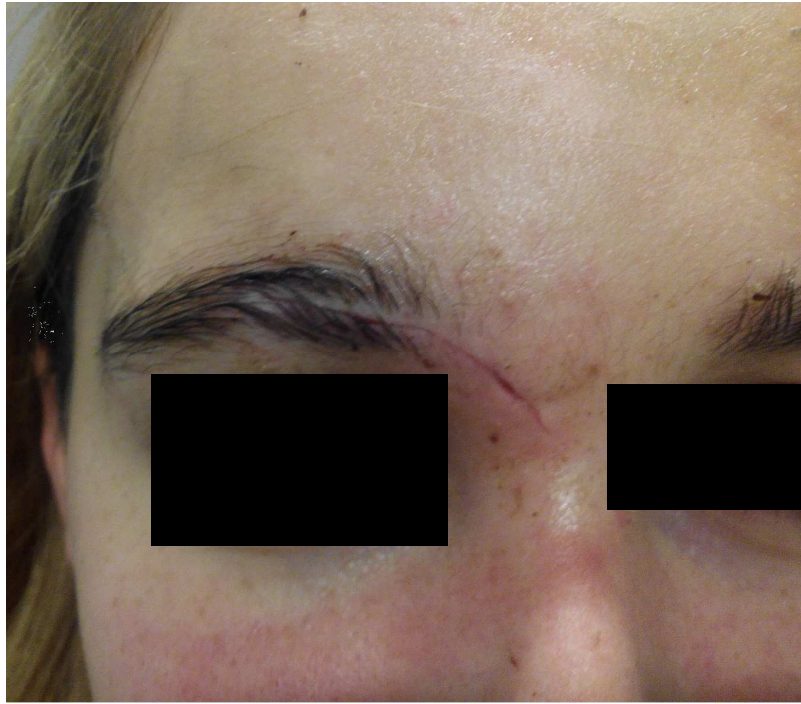


Figure 4: Post-operative image: a well-healed incision on the right eyebrow.

Discussion

Diagnosis of CSF fistula is often based on clinical suspicion when there is an unilateral rhinorrhea with a history of previous surgery or head trauma. Rarely, recurrent meningitis may be the only indication that an abnormal communication with the nasal cavity exists.⁶

If there is CSF rhinorrhea, diagnosis of a CSF fistula is accomplished after nasal inspection and performance of protein electrophoresis - Beta-2-transferrin test - in the nasal fluid, which demonstrates transferrin protein bands.⁷ This test is rapid and requires just few milliliters of liquid. It is the most sensitive laboratory test, when the rhinorrhea is active, for the presence of CSF in the nasal fluid - sensitivity of about 98%.^{1,8} The limitation of this test is that rhinorrhea must be present. It is also sometimes difficult to collect the fluid sample, especially from pediatric patients.¹ The dipstick test for glucose is not reliable, as both of these components can be seen in normal nasal and lacrimal secretions.⁸ In this case report, laboratory confirmation of the CSF fistula with Beta-2-transferrin was not possible to perform due to the absence of CSF rhinorrhea. The diagnosis of a CSF fistula was suspected based on clinical history of recurrent pneumococcal meningitis.

The site of the transdural fistula must be located before attempting repair. Radiographic exams - simple skull X rays - are ineffective. High-resolution CT in the axial and coronal planes is essential in identifying surgical anatomy and possible bony defects. Magnetic resonance imaging can be used as a noninvasive supplement to high-resolution CT in demonstrating herniated tissues.⁸ Contrast cisternography and intrathecal fluorescein dye diluted in CSF may be confirmatory, but the CSF leak must be active at the time of evaluation.¹ MRI cisternography is a method that does not require intrathecal injection of con-

trast and may become the method of choice for the evaluation of the CSF fistula.⁸ In this case report, the CSF leak was diagnosed through high-resolution CT scan of the paranasal sinus.

The majority of traumatic leaks resolve spontaneously within days of the injury.⁹ Although a surgical correction is required if persistent CSF rhinorrhea following acute trauma or recurrent meningitis are present. Repair is possible through intracranial, extracranial or endoscopic approaches. Each technique offers its own indications, advantages, and disadvantages depending on the site of leak.¹⁰

As the child's CSF leak was localized in the posterior wall of the frontal sinus, the endoscopic repair was not possible because of reduced access and visibility. Only small defects of the area, that can be seen bulging anteriorly when the frontal recess has been opened, can be dealt with endoscopically. An extracranial approach was performed, minimizing the risk of intracranial complications. A Lynch-Howarth incision with removal of the anterior wall of the frontal sinus allows access to the defect. The site of the defect can then be defined and repaired with fascia and fat to hold the graft. A great number of techniques and materials have been used for CSF leak occlusion, including autologous material as abdominal fat, nasal septum mucosa, bone, fascia lata and muscle grafts. The graft can be attached with fibrin glue, haemostatic sponges or vaseline gauzes. However, it appears that the location, size, techniques and materials used do not interfere directly with the success of the procedure. A critical aspect of the procedure is an adequate resection of the mucosa around the bone defect in order to permit the complete graft attachment.⁸ In this case report the graft consisted of dural regeneration matrix and a synthetic fascial graft fixed with fibrin glue. The graft can be positioned in an inlay form (between the dura and the skull base) or in an onlay form (on the nasal site of the leak). Indication for performing the onlay technique takes place when there is risk of damaging nerves and blood vessels during the dural detachment or during the intradural graft attachment.⁸ In this case, we performed the onlay technique. Some studies report that the form of graft positioning is not a critical factor in predicting success of the procedure.¹¹

Indications for obliterate the frontal sinus with fat or hidroxyapatite cement are traumatic disruption of the frontal recess, advanced suppuration with epidural abscess or meningitis, loss of significant portions of the anterior table of the frontal sinus, and failure of endoscopic approaches to satisfactorily communicate the frontal sinus and the nasal cavity.¹² In this case, the frontal sinus was obliterated with hydroxyapatite cement, that contoured the defect and filled the sinus.

Perioperative antibiotic was given and an intradural lumbar drain was put in place. Some authors still recommend the routine use of a lumbar drain, although recent studies suggest it should not be routinely performed.⁸ Activity resulting in increased intracranial pressure, such as straining, bending, lifting, and nose blowing, are avoided for several weeks after surgery.

Recurrent meningitis, especially pneumococcal, is usually caused by intracranial seeding of bacteria via a direct communication with an extracranial source. In an immunologically competent patient, recurrent meningitis should be considered a consequence of a transdural fistula until otherwise proven. The basic principles of pediatric CSF fistulas repair, such as an adequate exposure of the defect, do not differ from those of the adults. The successful repair of CSF leaks results of a preoperative correct identification of the fistula and bone defect site, as well as an accurate knowledge of reconstruction techniques and their judicious

application in the procedure planning. High-resolution CT in the axial and coronal planes is essential in defining bony anatomy and possible bony defects. The extracranial approach to anterior fossa CSF leaks can provide adequate exposure, virtually no central nervous system morbidity, and highly successful results.

Conflicts of interest: We have no conflicts of interest.

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