

3 YEARS LONG -TERM POSTTRAUMATIC CEREBROSPINAL FLUID FISTULA COMPLICATED WITH RECURRENT MENINGITIS

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ABSTRACT:

Most posttraumatic cerebrospinal fluid fistulas are accompanied by rhinorrhea and the physician can easily diagnose a fistula (leakage) in case of a traumatic event. Nevertheless, cases of fistulas occurring many years after the head injury have been reported in literature. We report the rare case of a 19-year-old female patient with a penetrating head injury and 3 years long-term posttraumatic cerebrospinal fluid fistula complicated with recurrent meningitis.

Key words: posttraumatic CSF fistula, recurrent meningitis, posttraumatic rinorrhea

INTRODUCTION

Posttraumatic cerebrospinal fluid leakage (CSF leakage) is a condition which occurs in up to 30% of the patients with skull base fractures [1, 2, 3]. In most cases, it is diagnosed within the first 24-48 hours after the head injury, before the occurrence of complications, the most common of which is posttraumatic meningitis. Although it has a rather low incidence rate, of only 0.38-2.03%, posttraumatic meningitis may have devastating consequences, with a mortality rate of up to 65% [4, 5, 6, 7, 8, 9, 10, 11, 12]. CSF fistula with late onset is a rare complication of head injuries, but it may occur as late as several years after the injury (even after 10 years), with several such cases mentioned in literature [13, 14, 15, 16, 17, 18, 19, 20, 21, 22]. We report the rare case of a female patient with frontal comminuted fracture suffered at the age of 11 (in 2007) and with the

occurrence of a posttraumatic CSF fistula 8 years later, which went undiagnosed and got complicated in the last 3 years by recurrent meningitis (between 2013 and 2015).

SHORT CASE REPORT

We report the case of a 19-year-old female patient who, 8 years ago, suffered a head injury with median penetrating frontal comminuted fracture (sleigh accident). Surgery was performed back then (2007), which consisted of craniectomy, yet the physical examination did not reveal any CSF fistula. 5 years after the surgical procedure, i.e. in 2012, the patient complained of recurrent episodes of bacterial meningitis with *Streptococcus pneumoniae*, which occurred every 6 months for 3 years and were treated in the infectious disease unit. Rhinorrhea was revealed during the latest

meningitis episode and the occurrence of a CSF fistula was suspected in the anterior cranial fossa. A computed tomography cisternography (CT cisternography) was performed, which revealed the contrast agent into the right nasal fossa through the anterior ethmoidal cells (fig. 1). Surgery was performed to close the fistula through the right free frontal bone flap and the dural defect was corrected using synthetic duramater. The patient's evolution was favorable, and the CSF fistula did not reoccur. The fracture line through the right paramedian ethmoid bone was evidenced during surgery.

DISCUSSIONS

Posttraumatic CSF fistula occurs in 2-3% of all head injuries [23] and in 10-30% of skull base fractures [24]. More than 50% of these become evident within the first 24-48 hours following the injury, and 70% during the first week [25, 26] which renders diagnosis setting relatively easy given the context of head injury. Nevertheless, CSF leakages have also been reported a few years after the head injury, in some cases even after more than 10 years [13, 14, 15, 16, 17, 18, 19, 20, 21, 22].

The first CSF rhinorrhea after a skull fracture which was correlated with was described in 1745 by Bidloo the Elder [27], and the first successful surgery of a CSF rhinorrhea occurred in 1926 and was performed by Dandy, who used autologus fascia lata [28].

CSF fistula is caused by an arachnoid and dura mater rupture usually associated with a bone defect (fracture line in the case of skull base fractures). The formation of an inflammatory dural scar with granulation tissue does not always provide sufficient protection against the paranasal sinuses bacteria, which may penetrate the intracranial compartment thus causing ascending bacterial meningitis [13, 17]. Moreover, due to the atrophy of this dural scar and to the small

injury, CSF rhinorrhea may occur later [16, 29] or may have an intermittent nature. The most common of these pathogens is *Streptococcus pneumoniae* which originates in the paranasal sinuses and upper airways, which is detected in 80-85% of the cases [2, 30]. Meningitis may promote inflammatory changes in the dural leak, thus leading to temporarily fistula arrest [31], which also happened in our patient, who suffered from recurrent bacterial meningitis every 6 months, more or less, for 3 years.

The risk of meningitis after posttraumatic CSF leakage was reported to range between 12.5% and 50% [32]. Different studies also calculated the time elapsed from the injury to the occurrence of posttraumatic meningitis, which may range between 24 hours and several years [10, 11, 12, 18, 21, 33, 34, 35, 36]. Moreover, mean injury-infection intervals ranged between 1.84 days and 3.4 years [4, 6, 11, 10, 22, 33, 37]. In our case, the time elapsed was 8 years.

In its acute phase, i.e. immediately after the injury, rhinorrhea is the most common symptom. It is often unilateral and accompanied by cephalalgia if the fistula is associated with ascending meningitis or meningocele [38]. Rhinorrhea occurs most frequently in anterior skull base fractures, but it may also occur in temporal bones fractures when the CSF travels to the middle ear and from here through the Eustachian tube to the nasopharynx. From here it penetrates the nasal fossae. In its chronic phase, the patients may complain of cephalalgia, salty or metallic taste, hyposmia, intermittent nasal discharge or recurrent meningitis [39, 40, 41] like in the case of our patient.

MRI or high-resolution CT examination with coronal, axial and sagittal reconstructions and CT cisternography scanning of the skull base, including the paranasal sinuses, are the gold standard for preoperative location

detection of the CSF fistula. These two imaging techniques allow the detection of the location of the CSF fistula as they allow the viewing of the contrast substance traveling from the subdural area into the paranasal sinuses or into the nasopharynx. The success rate of CT cisternography in detecting and determining fistula location was reported to be 95.7% [42].

Since more than 85% of traumatic CSF rhinorrhea ceases when applying nonsurgical therapy such as bed rest, head elevation and CSF flow diversion [26, 41], most authors recommend nonsurgical therapy for 7-10 days [25, 43, 44], although the best CSF fistula management is still a matter of controversy.

Conservative treatment is aimed at decreasing leakage activity, of decreasing CSF pressure and of preventing a negative gradient between the intracranial and paranasal sinus cavities [41], which will allow the self-reparation of the dural

defect. The nonsurgical therapeutic approach was not considered in the case of our patient, since when complications of CSF fistula like meningitis occur, the surgical repairing of the fistula is the preferred therapy [24].

CONCLUSIONS

Despite its relatively low occurrence, CSF fistula occurring late after head injury may have a high rate of morbidity and mortality. Therefore, it is very important that CSF fistula be diagnosed as early as possible. This diagnosis should be taken into consideration in the case of recurrent meningitis in a patient with a history of head injury, even when the head injury dates back many years (sometimes even more than 10 years).

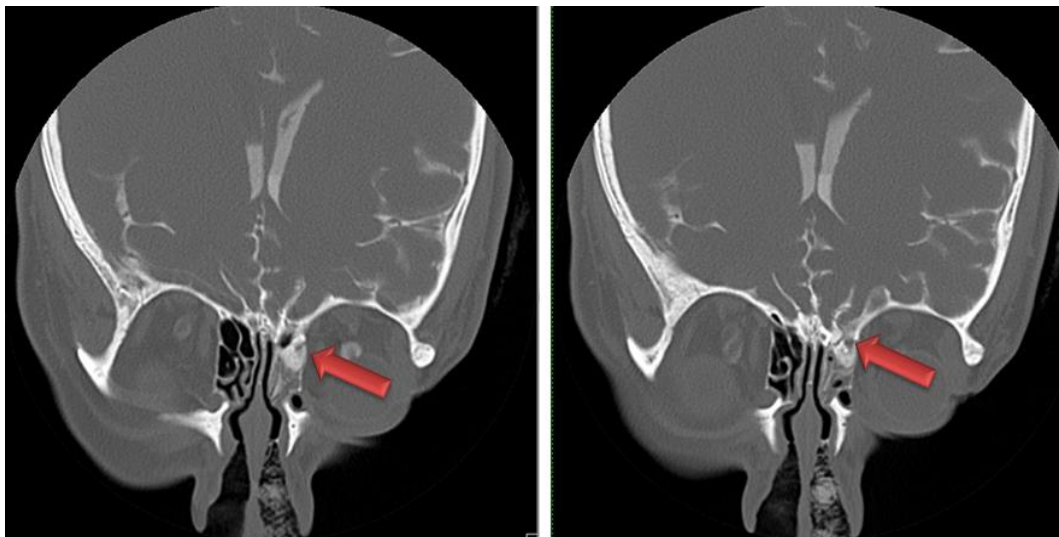


Figure 1. CT cisternography: it reveals the contrast agent travelling from the subdural area to the right nasal fossa (red arrow)

REFERENCES:

1. Marentette LJ, Valentino J. Traumatic anterior fossa cerebrospinal fluid fistulae and craniofacial considerations. *Otolaryngol Clin North Am* 1991; 24:151-63.
2. Loew F, Pertuiset B, Chaumier EE, Jaksche H. Traumatic, spontaneous and postoperative CSF rhinorrhea. *Adv Tech Stand Neurosurg* 1984;11:169-207.
3. Jennett B, Teasdale G, Fry J, et al. Treatment for severe head injury. *J Neurol Neurosurg Psychiatry* 1980;43:289-295.
4. Baltas I, Tsoulfa S, Sakellariou P, et al. Posttraumatic meningitis: Bacteriology, hydrocephalus, and outcome. *Neurosurgery* 1994;35:422-427.
5. Taha JM, Haddad FS, Brown JA. Intracranial infection after missile injuries to the brain: Report of 30 cases from the Lebanese conflict. *Neurosurgery* 1991;29:864-868.
6. Wilson NW, Copeland B, Bastian JF. Posttraumatic meningitis in adolescents and children. *Pediatr Neurosurg* 1990-91;16:17-20.
7. Frazee RC, Mucha P, Farnell MB, Ebersold MJ. Meningitis after basilar skull fracture: Does antibiotic prophylaxis help? *Postgrad Med* 1988;83:267-274.
8. Lau YL, Kenna AP. Post-traumatic meningitis in children. *Injury* 1986;17:407-409.
9. Buckwold FJ, Hand R, Hansebout RR. Hospital-acquired bacterial meningitis in neurosurgical patients. *J Neurosurg* 1977;46:494-500.
10. Jones SR, Luby JP, Sanford JP. Bacterial meningitis complicating cranial-spinal trauma. *J Trauma* 1973;13:895-900.
11. Hand WL, Sanford JP. Posttraumatic bacterial meningitis. *Ann Intern Med* 1970;72:869-874.
12. Appelbaum E. Meningitis following trauma to the head and face. *JAMA* 1960;173:116-120.
13. Kim EJ. A Case of Pneumococcal Meningitis Associated with Posttraumatic Cerebrospinal Fluid Rhinorrhea. *J Korean Child Neurol Soc* 2006; 14: 358-352.
14. Kamochi H, Kusaka G, Ishikawa M, Ishikawa S, Tanaka Y. Late onset cerebrospinal fluid leakage associated with past head injury. *Neurol Med Chir (Tokyo)* 2013;53(4):217-220.
15. Salca HC, Danaïla L. Onset of uncomplicated cerebrospinal fluid fistula 27 years after head injury: case report, *Surg Neurol* 1997;47(2):132-133.
16. Passagia JG, Chirossel JP, Favre JJ. Surgical approaches to the anterior fossa and preservation of olfaction. In : Cohadon F (ed). *Advances and technical standard in neurosurgery*. Wein, Springer; 1999, 25: 197- 241.
17. Schick B, Weber R, Kahle G, Draf W, Lackmann GM. Late manifestations of traumatic lesions of the anterior skull base. *SkullBase Surg* 1997; 7: 77-83.
18. Crawford C, Kennedy N, Weir WR. Cerebrospinal fluid rhinorrhoea and Haemophilus influenzae meningitis 37 years after a head injury. *J Infect* 1994;28(1):93-97.
19. Stewart BT, Kaye AH. Delayed cerebrospinal fluid rhinorrhoea: a case report. *Aust N Z J Surg* 1992;62(10):818-820.
20. Pandya PM, Keogh AJ. Traumatic cerebrospinal fluid rhinorrhoea: a timely reminder. *Injury* 1991; 22(6):492.
21. Okada Jun, Tsuda T, Takasugi S, et al. Unusually late onset of cerebrospinal fluid rhinorrhea after head trauma. *Surg Neurol* 1991;35:213-217.
22. Linell EA, Robinson WL. Head injuries and meningitis. *J Neurol Psychiat* 1941;4:23-31.
23. Talamonti G, Fontana R, Villa F. "High risk" anterior basal skull fractures. Surgical treatment of 64 consecutive cases. *J Neurosurg Sci* 1995; 39: 191-197.
24. Yilmazlar S, E. Arslan, H. Kocaeli, et al. Cerebrospinal fluid leakage complicating skull base fractures: analysis of 81 cases. *Neurosurg Rev* 2006; 29: 64-71.
25. Prosser JD, Vender JR, Solares CA. Traumatic cerebrospinal fluid leaks. *Otolaryngol Clin North Am* 2011; 44 (4): 857-873.
26. Schlosser RJ, Bolger WE. Nasal cerebrospinal fluid leaks: critical review and surgical considerations. *Laryngoscope* 2004; 114 (2): 255-265.

27. Dagi FT. Management of cerebrospinal fluid leaks, in Schmidek HH, Roberts DW (eds): Schmidek & Sweet Operative Neurosurgical Techniques: Indications, Methods, and Results. Philadelphia: Saunders Elsevier; 2006, vol 1: 130–145.
28. Dandy WE. Pneumocephalus (intracranial pneumatocele or arocele). Arch Surg 1926; 12: 949–982.
29. Kim HS, Hur JW, Lee JW, Lee HK. Extraordinarily Long-Term Posttraumatic Cerebrospinal Fluid Fistula. J Korean Neurosurg Soc 2007; 42(5): 403–405.
30. Hoen B, Viel JF, Gerad A, Dureux JB, Canton P. Mortality in pneumococcal meningitis: A multivariate analysis of prognostic factors. Eur J Med 1993; 2: 28–32.
31. Naidich TP, Moran CJ. Precise anatomic localization of atraumatic sphenoidal CSF rhinorrhea by metrizamide CT cisternography. J Neurosurg 1980; 53: 222–228.
32. Eljamel MS, Foy PM. Post-traumatic CSF fistulae, the case for surgical repair. Br J Neurosurg 1990; 4: 479–483.
33. Lewin W. Gram-negative meningitis following head wounds with especial reference to infection with coliform bacilli. Br J Surg 1948; 35: 266–280.
34. Farrell VJ, Emby DJ. Meningitis following fractures of the paranasal sinuses: Accurate, non-invasive localization of the dural defect by direct coronal computed tomography. Surg Neurol 1993; 37: 378–382.
35. Galloway WH, Chambers W. Pneumococcal meningitis: Skull defects as an aetiological factor. Lancet 1953; 2: 68–70.
36. Matschke J, Tsokos M. Post-traumatic meningitis: histomorphological findings, postmortem microbiology and forensic implications. Forensic Sci Int 2001; 115: 199–205.
37. Jepson RP, Whitty CWM. Pneumococcal meningitis after head injury treated with intrathecal penicillin. Lancet 1946; 1: 228–232.
38. Psaltis AJ, Schlosser RJ, Banks CA, Yawn J, Soler ZM. A systematic review of the endoscopic repair of cerebrospinal fluid leaks. Otolaryngol Head Neck Surg 2012; 147: 196–203.
39. Kerr JT, Chu FW, Bayles SW. Cerebrospinal fluid rhinorrhea: diagnosis and management. Otolaryngol Clin North Am 2005; 38: 597–611.
40. Cassano M, Felippu A. Endoscopic treatment of cerebrospinal fluid leaks with the use of lower turbinate grafts: a retrospective review of 125 cases. Rhinology 2009; 47: 362–368.
41. Ziu M, Savage JG, Jimenez DF. Diagnosis and treatment of cerebrospinal fluid rhinorrhea following accidental traumatic anterior skull base fractures. Neurosurg Focus 2012; 32(6): E3.
42. Eljamel MS, Pidgeon CN, Toland J, Phillips JB, O'Dwyer AA. MRI cisternography, and the localization of CSF fistulae. Br J Neurosurg 1994; 8: 433–437.
43. Dalgic A, Okay HO, Gezici AR, Daglioglu E, Akdag R, Ergungor MF. An effective and less invasive treatment of post-traumatic cerebrospinal fluid fistula: closed lumbar drainage system. Minim Invasive Neurosurg 2008; 51 (3): 154–157.
44. Yeo NK, Cho GS, Kim CJ, Lim GC, Jang YJ, Lee BJ, Chung YS. The effectiveness of lumbar drainage in the conservative and surgical treatment of traumatic cerebrospinal fluid rhinorrhea. Acta Otolaryngol 2013; 133 (1): 82–90.