ScientificScholar®
Knowledge is power
Publisher of Scientific Journals

www.surgicalneurologyint.com

Surgical Neurology International

Editor-in-Chief: Nancy E. Epstein, MD, Clinical Professor of Neurological Surgery, School of Medicine, State U. of NY at Stony Brook.

SNI: Infection

Editor
Ali Akhaddar, MD, IFAANS
Avicenne Military Hospital, Marrakech, Morocco

SNI
Open Access

Case Report

Bacterial meningitis caused by nontraumatic cerebrospinal fluid rhinorrhea with aqueductal stenosis: A case report

Saki Kotani, Yoshinobu Takahashi, Tamaki Morisako, Takumi Yamanaka, Naoya Hashimoito

Department of Neurosurgery, Kyoto Prefectural University Graduate School of Medical Science, Kyoto, Japan.

E-mail: Saki Kotani - saki-k@koto.kpu-m.ac.jp; *Yoshinobu Takahashi - yosinobuhokuto@yahoo.co.jp; Tamaki Morisako - morisako@koto.kpu-m.ac.jp; Takumi Yamanaka - tyamana@koto.kpu-m.ac.jp; Naoya Hashimoito - hashimotonaoya@me.com

***Corresponding author:**
Yoshinobu Takahashi,
Department of Neurosurgery,
Kyoto Prefectural University
Graduate School of Medical
Science, Kawaramachi Hirokoji,
Kyoto, Japan.

yosinobuhokuto@yahoo.co.jp

Received : 08 July 2022
Accepted : 27 August 2022
Published : 23 September 2022

DOI
10.25259/SNI_610_2022

Quick Response Code:

ABSTRACT

Background: Nontraumatic cerebrospinal fluid (CSF) rhinorrhea associated with aqueductal stenosis is rare. The resulting CSF leakage may cause bacterial meningitis, and appropriately timed surgical treatment should be considered.

Case Description: A 28-year-old woman with obstructive hydrocephalus secondary to aqueductal stenosis presented with intermittent nasal discharge. CSF rhinorrhea was suspected, but she refused surgery. During the course of conservative treatment, she developed meningitis. Exacerbation of hydrocephalus and CSF rhinorrhea was suspected, and the patient underwent endoscopic third ventriculostomy after recovery from meningitis. Postoperatively, ventricular size decreased and CSF leakage completely resolved. There was no recurrence of hydrocephalus or rhinorrhea.

Conclusion: Patients with intermittent CSF rhinorrhea due to exacerbation of hydrocephalus are at high risk for bacterial meningitis. Appropriately timed surgical treatment results in a favorable outcome.

Keywords: Aqueductal stenosis, Bacterial meningitis, Cerebrospinal fluid rhinorrhea, Endoscopic third ventriculostomy, Hydrocephalus

INTRODUCTION

Cerebrospinal fluid (CSF) rhinorrhea most commonly occurs following trauma or surgery, whereas nontraumatic CSF rhinorrhea associated with aqueductal stenosis is rare. Chronically increased intracranial pressure (ICP) can lead to the formation of a CSF fistula. The resulting CSF leakage may cause bacterial meningitis, and appropriately timed surgical treatment should be considered. Closure of the CSF fistula and CSF diversion may be effective, but the optimal treatment has not been established. Here, we report an adult case of bacterial meningitis secondary to nontraumatic CSF rhinorrhea with aqueductal stenosis.

CASE REPORT

A 28-year-old woman presented with headache and intermittent watery discharge from the left nostril. She had no history of head injury, head surgery, or obesity. Magnetic resonance imaging (MRI) of the head revealed obstructive hydrocephalus with aqueductal stenosis, ventricular

dilation with ballooning of the floor of the third ventricle, and CSF accumulation in the left sphenoid sinus and posterior ethmoid sinus. T2-weighted and fluid-attenuated inversion recovery (FLAIR) MRI revealed a hyperintense lesion in the tectal plate suggestive of a tectal glioma [Figure 1]. The lesion had been followed for 2 years without tumor progression. CSF rhinorrhea was suspected, but a CSF fistula was difficult to detect on head computed tomography (CT) and CT cisternography. The patient had refused surgery because she had no serious symptoms other than nasal discharge. During the course, temporary elevated white blood cell (WBC) had repeated, but she had been asymptomatic.

One day during follow-up in the outpatient clinic, she visited the emergency room due to headache and vomiting.

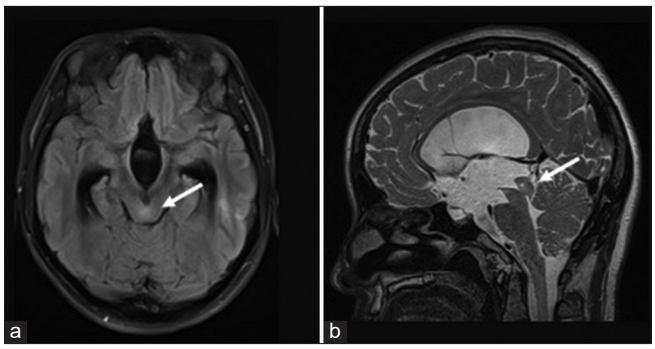

Figure 1: (a) Axial FLAIR MRI showed a hyperintense lesion in the tectal plate (white arrow). (b) Sagittal T2-weighted MRI showed the same lesion with aqueductal stenosis (white arrow head) and ventricular dilation with ballooning of the floor of the third ventricle.

She was initially alert and her body temperature was 37.2°C, but her level of consciousness deteriorated (Glasgow Coma Scale score: 5 [E2V2M1]) and body temperature increased to 39.4°C soon after admission. Nuchal rigidity was also observed. Laboratory findings showed a marked increase in WBC count ($2.01 \times 10^3/\mu L$) and neutrophil count (92.6%). CSF analysis revealed a markedly elevated cell count (20,310/μL), elevated protein level (2 mg/dL), and decreased CSF glucose (<5 mg/dL, serum glucose 123 mg/dL). CSF pressure was 9 cmH_2O. FLAIR MRI showed a hyperintense cerebral sulcus and hyperintense intraventricular lesions relative to CSF, suggesting meningitis and ventriculitis. T2-weighted MRI revealed increased CSF accumulation in the left sphenoid sinus and posterior ethmoid sinus. CSF pressure on admission was not high and there was no ventricular enlargement, but repeated ventricular enlargement and shrinkage had been observed over the past 2 years [Figure 2]. We considered this to be the reason for the transient remission of the symptom. The cause of the consciousness disorder was diagnosed as bacterial meningitis and ceftriaxone/vancomycin was soon administered intravenously. Three days later, the patient became alert. At this time, *Streptococcus pneumoniae* was detected in both blood and CSF cultures, and benzylpenicillin potassium (2.4 million U/day) was administered based on an antibiotic susceptibility test. After initiation of the antibiotic therapy, body temperature decreased and CSF analysis results gradually improved.

After recovery from meningitis, endoscopic third ventriculostomy (ETV) was performed for obstructive hydrocephalus with aqueductal stenosis. Biopsy of the tectal

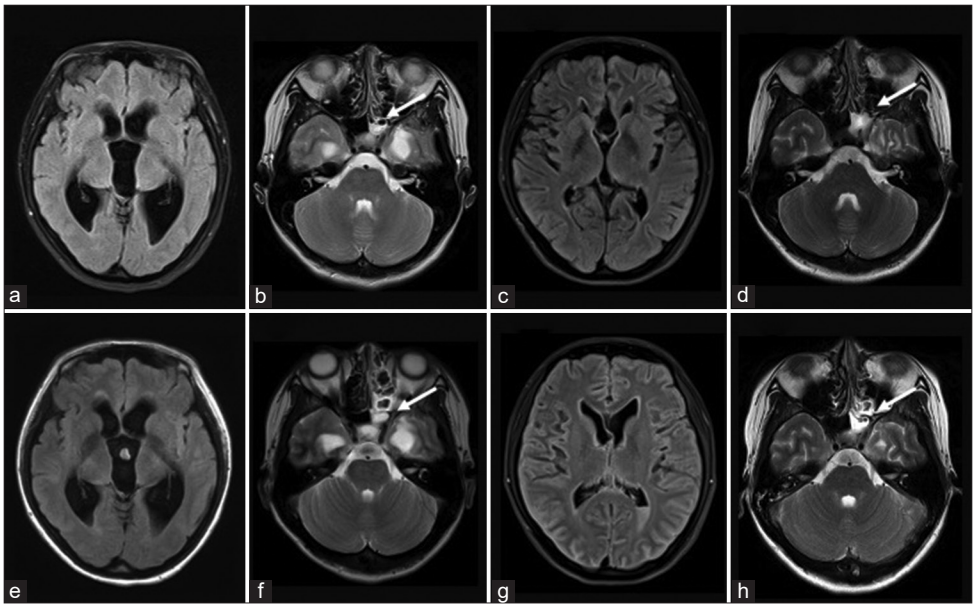

Figure 2: (a and b) First visit day, (c and d) 14 months later, (e and f) 19 months later, (g and h) 22months later (admission day). (a, c, e, g) Axial FLAIR MRI showed change of the size of ventricles. (b, d, f, h)Axial T2-weighted MRI showed CSF accumulation (white arrow).

plate lesion was not performed because a tumor could not be identified during surgery. CSF leakage completely resolved after surgery [Figure 3]. The patient was discharged to home and she has had no recurrence of hydrocephalus or rhinorrhea for 1 year.

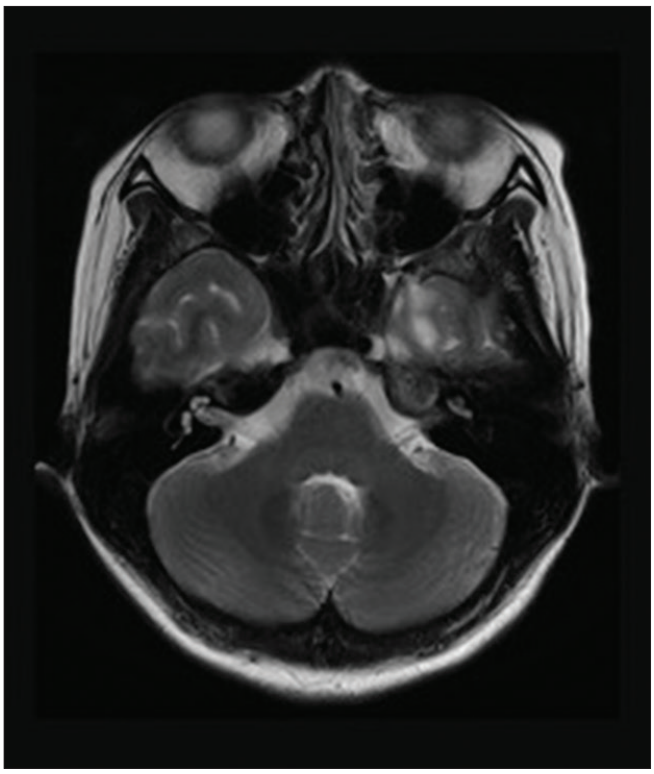

Figure 3: Postoperative magnetic resonance imaging (MRI). Axial T2-weighted MRI showed disappearance of cerebrospinal fluid leakage.

DISCUSSION

We encountered a case of bacterial meningitis resulting from nontraumatic CSF rhinorrhea with obstructive hydrocephalus secondary to aqueductal stenosis. The course of the patient suggested two important clinical lessons, as follows. First, chronically increased ICP can cause CSF rhinorrhea, and hydrocephalus has repeated remission and exacerbation according to CSF stagnation and leakage. Second, ETV can be effective for treating high-pressure CSF leakage and obstructive hydrocephalus with aqueductal stenosis.

Chronically increased ICP can cause CSF rhinorrhea. Thomson provided the first clear description and definition of spontaneous CSF rhinorrhea in 1899 and attempted to distinguish this condition from that caused by trauma.[17] Subsequently, Ommaya *et al.* introduced the term "nontraumatic CSF rhinorrhea" and classified this condition according to its mechanism.[14] CSF rhinorrhea most commonly occurs following trauma, and nontraumatic CSF rhinorrhea accounts for 3–4% of all cases.[10] Nontraumatic rhinorrhea is subdivided into "high-pressure leaks" and "normal pressure leaks:" high-pressure leaks include secondary to tumors or hydrocephalus, and normal pressure leaks include congenital anomalies or focal atrophy. In our case, the possibility of congenital anomalies is undeniable, but we possibly considered rhinorrhea caused by hydrocephalus resulting from aqueductal stenosis with suspect of tectal glioma because nasal discharge changed by progression of hydrocephalus. CSF pressure on admission was normal, this is because ICP decreased according to increased CSF leakage. CSF rhinorrhea associated with

Table 1: Cases of CSF rhinorrhea with aqueductal stenosis.

Case	Reference	Age[a]/sex	Site of fistula	Treatment approach	
				First approach	Second approach
1	Nishikawa *et al.* (1972)[13]	38/F	Ethmoid sinus	Surgical repair, LPS	(None)
2	Little and MacCarty (1976)[9]	22/M	Frontal sinus	VPS	Surgical repair
3	Ikeda *et al.* (1978)[5]	22/F	Frontal sinus	VPS	Surgical repair
4	Cabezudo *et al.* (1981)[3]	29/M	Frontal sinus	VAS	Surgical repair
5	Jooma and Grant (1983)[7]	12/M	Cribriform plate	VPS	Surgical repair
6	Tokuno *et al.* (1995)[18]	10/F	Sphenoid sinus	Surgical repair	VPS
7	Muzumdar *et al.* (2003)[12]	30/M	Cribriform plate	VPS	(None)
8	Barazi *et al.* (2008)[2]	28/M	Frontal sinus	ETV	Surgical repair
9	Kansal *et al.* (2010)[8]	19/M	Cribriform plate	VPS	Endoscopic repair
10	Yoshimura *et al.* (2012)[20]	28/F	Cribriform plate	Ventricular drainage + surgical repair	ETV
11	Apra *et al.* (2019)[1]	27/F	Frontal sinus, ethmoid sinus	Ventriculocisternostomy + surgical repair	(None)
12	Tang *et al.* (2020)[16]	25/M	Frontal sinus, sphenoid sinus	ETV+lumbar drain	LPS
13	Present case	28/F	Frontal sinus, sphenoid sinus	ETV	(None)

M: Male, F: Female, [a]: Years, VPS: Ventriculoperitoneal shunt, LPS: Lumboperitoneal shunt, ETV: Endoscopic third ventriculostomy

aqueductal stenosis is rare, with only 12 reported cases to date [Table 1].[1-3,5,7-9,12,13,16,18,20]

CSF fistulas commonly occur in anatomically fragile areas such as the ethmoid sinus, cribriform plate, and sphenoid sinus.[12,14,15] Persistently elevated ICP secondary to aqueductal stenosis may cause bone erosion in fragile areas, and formation of a direct communication between the lateral ventricle and frontal sinus may cause CSF rhinorrhea.[12] However, ICP has been reported to decrease with increased CSF leakage, leading to transient disappearance of elevated ICP.[12] CSF stagnation secondary to aqueductal stenosis results in ventricular enlargement, and this might have caused repeated remission and exacerbation of hydrocephalus in our patient. Intermittent CSF rhinorrhea is caused by repeated remission and exacerbation of hydrocephalus and also repeated temporary elevated WBC suggests inflammation following rhinorrhea. The high risk of bacterial meningitis suggests that early surgical treatment should be considered in such cases.

ETV can be effective for treating high-pressure CSF leakage and obstructive hydrocephalus with aqueductal stenosis. Management of CSF rhinorrhea is variable and has not been clearly established [Table 1]. Closure of a CSF fistula has been performed to treat CSF rhinorrhea, but there is a higher rate of recurrence after surgical closure (25–87%) than in cases of CSF leakage of other etiologies (10%).[11] Once the CSF fistula is closed, ICP tends to increase because CSF diversion into the nasal cavity no longer occurs.[16] Furthermore, there are reports of tension pneumocephalus after ventriculoperitoneal shunt insertion. There have also been recent reports of successful fistula repair and CSF diversion.[4,6,11,19] However, in cases, in which CSF fistulas are not detected, it is difficult to perform closure. We successfully treated CSF rhinorrhea with ETV, but there are only a few other reports of success using this technique.[2,6,19] However, ETV for nontraumatic CSF rhinorrhea with aqueductal stenosis might have two limitations: the ETV increases the CSF flow fistula as it restores CSF circulation in basal cisterns and the CSF resorption might be limited by the episode of meningitis. Thus, further studies are necessary and we will continue with follow-up observation of our patient.

CONCLUSION

We report a case of bacterial meningitis secondary to nontraumatic CSF rhinorrhea with hydrocephalus, in which a favorable outcome was achieved with ETV. Chronically increased ICP can cause CSF rhinorrhea, which may lead to improvement in hydrocephalus when ICP decreases with increased CSF leakage. Appropriately scheduled surgery is necessary due to the high risk of meningitis. ETV can also be an effective treatment approach in cases of high-pressure CSF leakage and obstructive hydrocephalus with aqueductal stenosis.

Declaration of the patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Apra C, Penet N, Froelich S. Cerebrospinal fluid fistula in the frontal sinus secondary to obstructive hydrocephalus. World Neurosurg 2019;131:19-20.
2. Barazi S, Choo M, Martin A. CSF rhinorrhea due to a pulsion diverticulum of the frontal horn. Br J Neurosurg 2008;22:580-1.
3. Cabezudo JM, Vaquero J, García-de-Sola R, Areitio E, Martinez R. Direct communication between the lateral ventricle and the frontal sinus as the cause of CSF rhinorrhea in aqueductal stenosis. Acta Neurochir (Wien) 1981;57:95-8.
4. Carrau RL, Snyderman CH, Kassam AB. The management of cerebrospinal fluid leaks in patients at risk for high-pressure hydrocephalus. Laryngoscope 2005;115:205-12.
5. Ikeda K, Nakano M, Tani E. Tension pneumocephalus complicating ventriculoperitoneal shunt for cerebrospinal fluid rhinorrhoea: Case report. J Neurol Neurosurg Psychiatry 1978;41:319-22.
6. Ishiwata Y, Shinozawa T, Ichikawa T, Kojima Y, Inada Y. Efficacy of percutaneous lumboperitonial shunt for management of cerebrospinal fluid leakage. Neurol Med Chir (Tokyo) 1985;25:923-7.
7. Jooma R, Grant DN. Cerebrospinal fluid rhinorrhea and intraventricular pneumocephalus due to iPntermittent shunt obstruction. Surg Neurol 1983;20:231-4.
8. Kansal R, Mahore A, Goel A. Cerebrospinal fluid rhinorrhea after ventriculoperitoneal shunt in a patient with tectal plate glioma. J Clin Neurosci 2010;17:532-3.
9. Little JR, MacCarty CS. Tension pneumocephalus after insertion of ventriculoperitoneal shunt for aqueductal stenosis. J Neurosurg 1976;44:383-5.
10. Loew F, Pertuiset B, Chaumier EE, Jaksche H. Traumatic, spontaneous and postoperative CSF rhinorrhea. Adv Tech Stand Neurosurg 1984;11:169-207.
11. Martínez-Capoccioni G, Serramito-García R, Martín-Bailón M, García-Allut A, Martín-Martín C. Spontaneous cerebrospinal fluid leaks in the anterior skull base secondary to idiopathic intracranial hypertension. Eur Arch Otorhinolaryngol 2017;274:2175-81.

12. Muzumdar D, Nadkarni T, Goel A. Spontaneous cerebrospinal fluid rhinorrhea as a presenting symptom of aqueductal stenosis case report. Neurol Med Chir (Tokyo) 2003;43:626-9.
13. Nishikawa M, Karasawa J, Kamada K. Non traumatic CSF rhinorrhea associated with obstruction of the aqueduct. Brain and Nerve (Tokyo) 1972;24:173-7.
14. Ommaya AK, Di Chiro G, Baldwin M, Pennybacker JB. Non-traumatic cerebrospinal fluid rhinorrhoea. J Neurol Neurosurg Psychiatry 1968;31:214-25.
15. Shetty PG, Shroff MM, Fatterpekar GM, Sahani DV, Kirtane MV. A retrospective analysis of spontaneous sphenoid sinus fistula: MR and CT findings. AJNR Am J Neuroradiol 2000;21:337-42.
16. Tang C, Zhu J, Feng K, Yang J, Cong Z, Cai X, *et al.* Successful treatment of spontaneous cerebrospinal fluid rhinorrhea with endoscopic third ventriculostomy and lumboperitoneal shunt: A case report. Front Neurosci 2020;14:57.
17. Thomson St. C. The Cerebro-Spinal Fluid: Its Spontaneous Escape from the Nose. London: Cassell; 1899.
18. Tokuno T, Ban S, Nakazawa K, Yoshida S, Matsumoto S, Shingu T, *et al.* Non-traumatic cerebrospinal fluid rhinorrhea associated with hydrocephalus: A case report. No Shinkei Geka 1995;23:265-9.
19. Wise SK, Schlosser RJ. Evaluation of spontaneous nasal cerebrospinal fluid leaks. Curr Opin Otolaryngol Head Neck Surg 2007;15:28-34.
20. Yoshimura M, Matsusaka Y, Terada A, Ishiguro T, Nakajima H, Yamanaka K, *et al.* Spontaneous cerebrospinal fluid rhinorrhea associated with long-standing overt ventriculomegaly in adults (LOVA). No Shinkei Geka 2012;40:897-902.

How to cite this article: Kotani S, Takahashi Y, Morisako T, Yamanaka T, Hashimoito N. Bacterial meningitis caused by nontraumatic cerebrospinal fluid rhinorrhea with aqueductal stenosis: A case report. Surg Neurol Int 2022;13:439.