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Spontaneous cerebrospinal fluid rhinorrhea in a patient with elevated body mass index: A case report

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ABSTRACT

CSF rhinorrhoea is a rare clinical entity referring to the spontaneous leakage of CSF through the nasal passage. The primary cause of CSF rhinorrhoea can be either spontaneous, where the causes include a temporal bone anomaly, skull base defect, or dura mater defect, or non-spontaneous causes, including surgical or accidental trauma, tumors, and radiation therapy to the base of the skull. It is imperative to diagnose the condition in its early stages to avoid complications like meningitis, sepsis, and abscesses. Primary cases are uncommon and linked with increased BMI and intracranial hypertension (ICH). The beta-2 transferrin test is considered the gold standard for diagnosing a CSF leak; however, a clinical examination along with CT and MRI scans can also help with the diagnosis.

In this report, the patient is a 49-year-old female who presented with an elevated BMI. The patient came to the emergency department with the complaint of 'drooping' on the right side of the face, weakness on the same side, and a history of diabetes mellitus, hypertension, and schizoaffective disorder. CT and MRI of the patient revealed acute infarcts in the brain and findings in the temporal bone which indicated a CSF leak. Initially, conservative management was initiated, but later on, the development of subsequent symptoms warranted surgical management. Following radiological diagnosis using CT scans, the patient underwent the transsphenoidal technique to address the CSF leak.

This case highlights the complexities involved in diagnosing and managing CSF rhinorrhoea. Diagnostic assays, such as β-2 transferrin, along with imaging tests, are essential for accurate identification and treatment. Additionally, factors like obesity and coexisting health conditions can significantly influence treatment strategies and outcomes. However, the lack of comprehensive follow-up data and the reliance on a single case study limit the scope of the conclusions that can be drawn.

Keywords: cerebrospinal fluid rhinorrhoea, CSF, spontaneous cerebrospinal fluid rhinorrhoea

INTRODUCTION

A cerebrospinal fluid (CSF) rhinorrhoea develops when the integrity of the pressure barrier between the CNS and the external environment is disrupted. This presentation is described as the end result of both active and passive clearance of plasma at the choroid plexus [1]. The cause of CSF rhinorrhoea can be classified as spontaneous or non-spontaneous, each with distinct underlying causes. Congenital anatomical defects in the temporal bone, skull base, or dura mater are among the spontaneous causes, while non-congenital causes include surgical trauma, accidental trauma, tumors, and radiation therapy to the base of the skull [2,3]. Trauma is the main cause of CSF fistulas, both iatrogenic and non-iatrogenic, accounting for an estimated 80% to 90% of cases [4,5].

The current literature indicates that patients presenting with CSF rhinorrhoea require immediate surgical repair. Various techniques and materials may be adopted to achieve effective closure of the CSF fistula. In patients with high-flow CSF fistulas, improved outcomes have been reported with multi-layered, vascularised repairs, as this technique reduces the risk of postoperative CSF leaks. Conversely, patients with idiopathic intracranial hypertension require long-term management following surgical interventions to address the underlying disease process [6].

Early diagnosis and effective management are essential to prevent the life-threatening complications associated with CSF rhinorrhoea, including bacterial meningitis and brain abscesses. However, the available literature describes the challenges of-

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ten faced during the diagnosis of this condition. As depicted in previous case reports, patients presenting with unilateral clear nasal discharge accompanied by nonspecific headache symptoms should raise suspicion for CSF rhinorrhoea. Other prevalent symptoms include alterations in mental status, seizures, and meningitis [7,8]. This study includes a case of spontaneous CSF rhinorrhoea in a previously healthy 49-year-old obese female.

CASE PRESENTATION

A 49-year-old female patient with a history of diabetes mellitus, hypertension, and schizoaffective disorder presented to the emergency department with right facial asymmetry and body weakness on the right side. A comprehensive assessment of her current medical history was undertaken. At the time of admission, the patient was hypertensive (190/95 mmHg), and initial investigations showed a mean arterial pressure (MAP) of 141 mmHg, stable blood oxygen saturation (SO_2) of 97%, and a heart rate of 76 bpm. The patient also had a BMI greater than 25, which is a known risk factor.

The first MRI appeared to show a recent infarct within the superior frontal gyrus of the left hemisphere and a reduction in density of the M2 segment of the left middle cerebral artery (MCA) identified on FLAIR sequences. In consultation with the patient's family, it was ascertained that her symptoms had been present for more than 24 hours before admission. After the MRI, the patient's history was presented to neurologists, and, according to the acute stroke protocol, the patient was sent to the ICU for 24 hours before being transferred to the medical ward. Conservative medical treatment was initiated while monitoring the patient's status.

After four days, a CT scan showed an acute to subacute infarct in the left frontoparietal region. A new area of hypodensity was also observed in the left ganglion-capsular region, measuring 3 cm by 1.7 cm. Nonetheless, there was no evidence of a hemorrhagic transformation or mass effect. At that time, the patient was alert, sitting in bed, and able to recognize all family members; however, the patient was not oriented to time or place. Furthermore, there was an impairment in the patient's comprehension, along with aphasia, dysphagia, and cognitive and memory impairments.

Rehabilitation was initiated, and the patient was admitted to the rehabilitation center to begin an intensive program. Two weeks later, the patient began to experience a clear, watery nasal discharge alongside discomfort in the left ear. The patient was referred to an ear, nose, and throat (ENT) specialist, and CSF leakage was highly suspected. As a result, the patient was admitted to adult neurology for further observation and management, where an MRI of the brain showed a temporal bone defect. The neurology specialist recommended surgical treatment, specifically a craniotomy with the evacuation of the meningoencephalocele and a cranioplasty to address the skull-based defect. A duroplasty was also advised.

A second opinion was sought, which warranted a CT scan of the patient's sinus. The CT scan confirmed the diagnosis of CSF leakage. After discussing the findings, the patient and her family agreed to proceed with transsphenoidal surgery.

DISCUSSION

CSF rhinorrhoea results from abnormal communication between the subarachnoid space and a defect in the skull base, leading to the drainage of CSF through the nasal cavity. This condition has been categorized as either spontaneous or non-spontaneous. Spontaneous CSF rhinorrhoea, also referred to as non-traumatic CSF rhinorrhoea, is extremely rare, constituting only 4% of all reported cases [9-11]. This particular clinical presentation has been linked with increased intracranial pressure (ICH) and elevated BMI. In particular, idiopathic CSF rhinorrhoea has been frequently observed in middle-aged women who are overweight [12]. The current study also presented a case of spontaneous CSF rhinorrhoea in a 49-year-old female patient with an elevated BMI.

Although there are many case reports and series describing CSF rhinorrhoea, the pathophysiology of this presentation is still not well understood. In prior research, it is hypothesized that prolonged ICH may cause defects in a patient's skull to appear over time. These defects, paired with the continued ICH, can result in herniation of the patient's dura mater through the bony defects. Consequently, this weakens the dura mater and increases its susceptibility to tears and the formation of a dural-mucosal fistula [13].

Obesity has also been suggested as a prevalent cause of CSF rhinorrhoea due to its role in increasing intraabdominal pressure. This pressure elevates the diaphragm and increases both pleural and cardiac pressures. Badia et al. (2001) initially indicated that a relationship may exist between obese females and the risk of developing primary spontaneous CSF rhinorrhoea. In this 20-patient case series, all patients were clinically obese (BMI >30 kg/m²) and presented with spontaneous CSF rhinorrhoea [14]. Sugarman et al. (1995) substantiated this paradigm and proposed a mechanism for this relationship. They suggested that elevated BMI raises intraabdominal and intrathoracic pressure, increasing cardiac-filling pressure and impairing venous return from the

brain. This condition induces the ICH typically observed in patients with CSF rhinorrhoea [15]. In the case described in this report, the patient presented with an elevated BMI and complained of headaches exacerbated when bending over, potentially indicating ICH, although the patient had no signs of ICH on examination.

Several diagnostic mechanisms have been depicted in the current literature for identifying CSF rhinorrhoea. Nonetheless, the gold standard approach comprises the use of beta-2 transferrin or beta-2 trace protein, two biomarkers exclusively found in the CSF and perilymphatic fluid. The use of beta-2 trace protein for detecting CSF leakage was initially described in 1979 by Meurman et al. Since then, healthcare professionals have extensively employed this method for diagnosing CSF rhinorrhoea and skull-base cerebrospinal fluid fistulas. This approach remains the gold standard due to its reported sensitivity of 94% to 100% and specificity of 98% to 100% [16].

In the current case report, beta-2 transferrin testing was unavailable, necessitating reliance on comparative blood glucose concentrations between the draining fluid and blood. The presence of glucose in secretions strongly indicates CSF presence. However, this method is not recommended as a confirmatory test due to its low diagnostic specificity and sensitivity [17]. Furthermore, low sensitivity is observed in cases of bacterial contamination and low specificity in diabetic patients. Therefore, glucose detection in nasal discharge cannot serve as a unique diagnostic criterion for CSF leaks and should be corroborated with clinical symptoms and imaging studies.

While the beta-2 transferrin test is widely regarded as the gold standard for diagnosing cerebrospinal fluid (CSF) rhinorrhoea due to its high sensitivity and specificity, its unavailability in this case necessitated reliance on alternative diagnostic approaches. Here, the diagnosis was primarily established through a combination of clinical presentation, imaging findings, and comparative glucose analysis.

Clinical symptoms such as clear nasal discharge, positional headaches, and associated ear discomfort raised initial suspicions. These observations were corroborated by imaging studies, including high-resolution CT and MRI scans, which confirmed the presence of a temporal bone defect and a CSF fistula. Additionally, CT/MR cisternography was particularly critical in identifying the exact location and extent of the defect.

Although glucose concentration testing in nasal discharge lacks the precision of the beta-2 transferrin assay, its results, when interpreted alongside imaging and clinical findings, provided strong evidence for a CSF leak. This integrative diagnostic approach mitigated the limitations posed by the absence of the biomarker test, ensuring an accurate and timely diagnosis.

CSF leaks can be diagnosed through physical examination and via high-resolution computed tomography and magnetic resonance imaging (MRI) [18]. These imaging modalities are crucial, especially in instances where fractures or tumors are observed near the site of the suspected leak [9]. CT/MR cisternography, although invasive, is often considered the gold standard for diagnosing CSF leaks and determining their location and extent [19].

In the treatment of CSF rhinorrhoea, non-surgical management is initially recommended; however, when conservative approaches fail, surgical procedures become necessary to prevent serious and potentially fatal complications over time. Conservative management typically includes the prescription of acetazolamide, bed rest, and head elevation to reduce intracranial pressure.

Surgical procedures can be classified as endoscopic, extracranial-intracranial, or endoscopic-intracranial. The endoscopic approach is preferred due to its lower morbidity and higher success rates (90–100%) compared to the intracranial approach, which has a higher morbidity and lower success rate of 60–80% [9,20].

According to recent guidelines, the first-line treatment for CSF rhinorrhoea is endoscopic management of the defect, with extracranial approaches reserved for cases where endoscopic interventions fail. Additionally, therapies such as antibiotics, diuretics, lumbar drains, and head elevation may further reduce morbidity and mortality [9,21].

Research suggests that elevated BMI increases intracranial pressure (ICH), a key factor in the development of spontaneous CSF rhinorrhoea. This relationship can be attributed to increased intra-abdominal pressure associated with obesity, which indirectly elevates venous pressure and impairs venous outflow from the brain. The resulting ICH weakens the structural integrity of the skull base and dura mater over time, predisposing individuals to the formation of defects through which CSF may leak.

Additionally, adiposity-related systemic inflammation may exacerbate these structural vulnerabilities by impairing collagen synthesis and tissue repair. This mechanistic understanding aligns with clinical observations in obese patients, where symptoms such as headaches, particularly positional headaches, are indicative of elevated intracranial pressure. Including detailed studies or models to explore these relationships further would substantiate the findings and inform targeted management strategies for patients with elevated BMI.

Moreover, the transsphenoidal approach was selected as the surgical intervention for addressing

the patient's CSF rhinorrhoea due to its minimally invasive nature and high success rates. Compared to traditional intracranial approaches, the transsphenoidal technique offers several advantages, including reduced morbidity, shorter recovery times, and fewer complications. This approach is especially suitable for cases involving anterior skull base defects, as it provides direct access to the site of the CSF leak with minimal disruption to surrounding structures.

In addition, recent studies have demonstrated success rates of up to 90–100% for the transsphenoidal approach, making it the first-line treatment for many patients with CSF leaks. The decision to avoid more invasive extracranial or intracranial methods was further supported by the patient's clinical presentation, imaging findings, and the absence of contraindications for endoscopic surgery. Additionally, the transsphenoidal approach is associated with improved postoperative outcomes, including a lower risk of infection and a faster return to normal activities. This choice underscores the importance of tailoring surgical interventions to the specific needs of the patient while incorporating the latest advancements in minimally invasive techniques.

Association of BMI with spontaneous CSF rhinorrhea

Several studies corroborate the strong association between elevated BMI and spontaneous cerebrospinal fluid (CSF) rhinorrhoea. Wen et al. (2019) reported that spontaneous CSF rhinorrhoea is more common in middle-aged women with a mean BMI of 33.1 kg/m², aligning with findings from this case report [22]. Similarly, Low et al. (2020) linked obesity with decreased bone mineral density in the cranial base, which predisposes patients to CSF leaks [23]. This underscores the mechanistic role of obesity-related intracranial pressure and bone thinning in the development of such leaks.

Complications of untreated CSF rhinorrhea

The importance of early diagnosis and surgical intervention is highlighted by Pandey et al. (2021), who described a patient presenting with recurrent spontaneous CSF rhinorrhoea that led to life-threatening meningoencephalitis. This case demonstrates the severe complications that can arise when CSF leaks are not promptly managed, including meningitis, seizures, and infections such as *Staphylococcus arlettae* [24].

Surgical management and outcomes

Studies by Kreatsoulas et al. (2020) and Kim-Orden et al. (2019) emphasize the efficacy of endoscopic surgical approaches. The endonasal endoscopic approach (EEA) was successful in over 90% of cases but highlighted the need for careful postoperative monitoring, especially in patients with elevated BMI, who are at higher risk for complications such as revision surgeries or CSF shunt placement [25,26].

Relevance to current case

The findings from these studies strengthen the significance of this case report by situating it within the broader context of spontaneous CSF rhinorrhoea cases linked to elevated BMI. Furthermore, the potential for severe complications, such as meningitis, underscores the necessity of prompt diagnosis and tailored surgical interventions [23].

Recommendations for follow-up

The current report underscores the importance of systematic follow-up in cases of CSF rhinorrhoea, particularly after surgical intervention. Monitoring the patient's recovery over an extended period is crucial for evaluating the efficacy of the treatment and identifying any potential complications, such as recurrence of CSF leakage, infections, or long-term neurological impairments [27]. A follow-up plan should include:

- **Regular imaging assessments:** Periodic CT or MRI scans to ensure the integrity of the surgical repair and detect any recurrent leaks.
- **Clinical evaluations:** Comprehensive assessments focusing on symptoms such as nasal discharge, headaches, or signs of infection.
- **Quality of life measures:** Evaluating the patient's overall well-being and any functional impairments post-surgery.
- **Documentation of long-term outcomes:** Collecting data on recurrence rates, patient satisfaction, and any complications to contribute to future clinical guidelines.

Integrating follow-up data into future case reports will provide a richer understanding of treatment success rates and help refine clinical practices for managing CSF rhinorrhoea [28].

Limitations

Some of the limitations associated with this case report include the lack of follow-up of the patient after surgery, especially because such follow-up is crucial for assessing the treatment protocols and the overall outcome of the condition. Additionally, relying on a single case limits the generalizability of the findings to other patients. The absence of certain diagnostic tools, such as beta-2 transferrin testing, may also have impacted the accuracy of the diagnosis. These limitations underscore the need for more

research studies to address these gaps in understanding and to improve the interpretation of results.

CONCLUSIONS

Patients who present with CSF rhinorrhoea should undergo early diagnosis and management to avoid potentially fatal complications such as meningitis, intracranial sepsis, and abscesses. Primary CSF leak is a rare clinical condition that is more commonly associated with an increased BMI and intracranial hypertension (ICH). Beta-2 transferrin testing is the most reliable method for identifying CSF in secretions; however, clinical examination combined with CT or MRI findings is sufficient to establish a diagnosis. Among the available surgical treatments for CSF leaks, a postoperative period without complications is usually indicative of a favorable outcome.

Patients' consent:
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REFERENCES

1. Wang W, Emmet E, Leyva A, Dennison JV, Dodson S. A rare case of spontaneous recurrent cerebrospinal fluid rhinorrhea. *Cureus*. 2019 Jan 14;11(1):e3883. doi: 10.7759/cureus.3883.
2. Hiremath S, Gautam A, Sasindran V, Therakathu J, Benjamin G. Cerebrospinal fluid rhinorrhea and otorrhea: A multimodality imaging approach. *Diagn Interv Imaging.* 2019;100(1):3-15. doi: 10.1016/j.diii.2018.05.003.
3. Vaishnavadevi G, Yoganandan D. Non spontaneous cerebrospinal fluid rhinorrhoea and subsequent bacterial meningitis: A case report. *J Clin Diagn Res.* 2022;16(8).
4. Pradhan B, Shreepali A. Endoscopic repair of CSF rhinorrhoea: Our experience. *Nepal J ENT Head Neck Surg.* 2013;4(2):26-8.
5. Zlab MK, Moore GF, Daly DT, Yonkers AJ. Cerebrospinal fluid rhinorrhea: A review of the literature. *Ear Nose Throat J.* 1992;71(7):314-7.
6. DeConde AS, Suh JD, Ramakrishnan VR. Treatment of cerebrospinal fluid rhinorrhea. *Curr Opin Otolaryngol Head Neck Surg.* 2015;23(1):59-64. doi: 10.1097/moo.0000000000000124.
7. Briner HR, Swift AC. CSF rhinorrhoea and the anterior skull base. In: Contemporary Rhinology: Science and Practice. Springer; 2023. p. 491-508.
8. Caulley L, Hopkins C, Lund VJ. Diagnostic algorithm for cerebrospinal fluid leak. In: CSF Rhinorrhea: Pathophysiology, Diagnosis and Skull Base Reconstruction. Springer; 2022. p. 73-84.
9. Boppana HK, Welch T, Calderon C. Spontaneous cerebrospinal fluid rhinorrhea: A case report. *Cureus*. 2020;12(7):e9401. doi: 10.7759/cureus.9401.
10. Ozdogan S, Gergin YE, Gergin S, Tatarli N, Hicdonmez T. Spontaneous rhinorrhea mimicking sinusitis. *Pan Afr Med J.* 2015 Feb 4;20:97. doi: 10.11604/pamj.2015.20.97.5748.
11. Peng D, Yang K, Wu C, Gao F, Sun W, Lu G. Ventriculoperitoneal shunt alone for cerebrospinal fluid rhinorrhea with neuroendocrine alterations in idiopathic intracranial hypertension: A case report and literature review. *Front Neurol.* 2022 Feb 10;13:809224. doi: 10.3389/fneur.2022.809224.
12. Quatre R, Attye A, Righini CA, Reyt E, Giai J, Schmerber S, et al. Spontaneous cerebrospinal fluid rhinorrhea: Association with body weight and imaging data. *J Neurol Surg B Skull Base.* 2017 Oct;78(5):419-424. doi: 10.1055/s-0037-1603731.
13. Lieberman SM, Chen S, Jethanamest D, Casiano RR. Spontaneous CSF rhinorrhea: Prevalence of multiple simultaneous skull base defects. *Am J Rhinol Allergy.* 2015;29(1):77-81. doi: 10.2500/ajra.2015.29.4121.
14. Badia L, Loughran S, Lund V. Primary spontaneous cerebrospinal fluid rhinorrhea and obesity. *Am J Rhinol*. 2001;15(2):117-9. doi: 10.2500/105065801781543736.
15. Sugerman HJ, Felton WL 3rd, Salvant JB Jr, Sismanis A, Kellum JM. Effects of surgically induced weight loss on idiopathic intracranial hypertension in morbid obesity. *Neurology*. 1995;45(9):1655-9. doi: 10.1212/wnl.45.9.1655.
16. Marshall AH, Jones NS, Robertson IJ. An algorithm for the management of CSF rhinorrhoea illustrated by 36 cases. *Rhinology*. 1999;37(4):182-5.
17. Yadav YR, Parihar V, Janakiram N, Pande S, Bajaj J, Namdev H. Endoscopic management of cerebrospinal fluid rhinorrhea. *Asian J Neurosurg.* 2016;11(3):183-93. doi: 10.4103/1793-5482.145101.
18. Eljazzar R, Loewenstern J, Dai J, Shrivastava R, Iloreta A. Detection of CSF leaks: Is there a radiologic standard of care? A systematic review. *World Neurosurg.* 2019;127. doi: 10.1016/j.wneu.2019.01.299.
19. Vemuri NV, Karanam LSP, Manchikanti V, Dandamudi S, Puvvada SK, Vemuri VK. Imaging review of cerebrospinal fluid leaks. *Indian J Radiol Imaging.* 2017;27(4):441-6. doi: 10.4103/ijri.IJRI_380_16.
20. Younis M, Adly M, Yousry M, Zahran A, Elmoheen A. Spontaneous cerebrospinal fluid rhinorrhoea caused by sustained intracranial hypertension. *Cureus.* 2022 Apr 24;14(4):e24441. doi: 10.7759/cureus.24441.
21. Wang EW, Zanation AM, Gardner PA, Schwartz TH, Eloy JA, Adappa ND, et al. ICAR: Endoscopic skull-base surgery. *Int Forum Allergy Rhinol.* 2019 Jul;9(S3):S145-S365. doi: 10.1002/alr.22326.
22. Wen TY, Gendeh BS, Husain RAOC, Husain S, Jeevaratnam K. Spontaneous cerebrospinal fluid rhinorrhoea and its association with body mass index (BMI). *Bangladesh J Med Sci.* 2019;18(2):322.
23. Low CM, Kim DK, Smith AJ, Yin LX, Smith BM, Stokken JK, et al. Association of bone mineral density of the anterior cranial base, obesity, and spontaneous cerebrospinal fluid rhinorrhea. *Int Forum Allergy Rhinol.* 2021 Apr;11(4):804-806. doi: 10.1002/alr.22717.
24. Pandey S, Chakraborty R. A case of recurrent spontaneous CSF rhinorrhoea with meningoencephalitis. *Neuroscience*. 2021;5(4):124-8.
25. Kim-Orden N, Shen J, Or M, Hur K, Zada G, Wrobel B. Endoscopic endonasal repair of spontaneous cerebrospinal fluid leaks using multilayer composite graft and vascularized pedicled nasoseptal flap technique. *Allergy Rhinol* (Providence). 2019 Nov 13;10: 2152656719888622. doi: 10.1177/2152656719888622.
26. Kreatsoulas DC, Shah VS, Otto BA, Carrau RL, Prevedello DM, Hardesty DA. Surgical outcomes of the endonasal endoscopic approach within a standardized management protocol for repair of spontaneous cerebrospinal fluid rhinorrhea. *J Neurosurg.* 2020 Feb 28;134(3):780-786. doi: 10.3171/2019.12.JNS192891.
27. Cheng E, Grande D, Leonetti J. Management of spontaneous temporal bone cerebrospinal fluid leak: A 30-year experience. *Am J Otolaryngol.* 2019 Jan-Feb;40(1):97-100. doi: 10.1016/j.amjoto.2018.09.018.
28. Antipov V, Kalandari A. Spontannaya nazal'naya likvoreya: obzor literatury [Spontaneous nasal cerebrospinal fluid leak: literature review]. *Neyrokhirurgiya* [Neurosurgery]. 2024;26(1):137-47. (Russian).