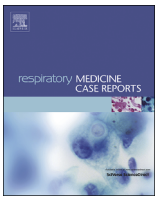
Respiratory Medicine Case Reports 42 (2023) 101814

Contents lists available at ScienceDirect

Respiratory Medicine Case Reports

journal homepage: www.elsevier.com/locate/rmcr



Case Report

A 58-year-Old non-smoking woman with intractable cough and rhinorrhea☆

Kyle Admire [a, *], Lauren Kring [a], A. Lukas Loschner [b]

[a] Department of Internal Medicine, Virginia Tech-Carilion, Roanoke, VA, USA
[b] Department of Pulmonary & Critical Care Medicine, Virginia Tech-Carilion, Roanoke, VA, USA

ARTICLE INFO

Handling Editor: AC Amit Chopra

Keywords:
Upper Airway Cough Syndrome
Cough
Pathology
Cerebrospinal fluid rhinorrhea
Pulmonary
Airway

ABSTRACT

Our case highlights an uncommon cause of CSF rhinorrhea. The patient was diagnosed with bacterial meningitis and appropriately treated after which, she developed unilateral rhinorrhea followed by non-productive cough. These symptoms were not responsive to multiple treatment regimens, and eventually imaging revealed a dehiscence in the ethmoid air sinus which was surgically repaired. We also performed a literature review on CSF rhinorrhea and provide insights into its evaluation.

1. Introduction

Generally, Upper Airway Cough Syndrome (UACS) is caused by Gastro-Esophageal Reflux Disease (GERD) or an irritant/allergic post-nasal drip. This case report demonstrates an unusual cause for UACS - mechanical dehiscence of the ethmoid air sinus due to inflammation from bacterial meningitis. CSF rhinorrhea is typically only seen after trauma, but our patient demonstrates that this condition can occur after multiple conditions. We also performed a literature review on CSF rhinorrhea and provide insights into its evaluation.

2. Case presentation

A 58-year-old non-smoking woman with a past medical history of hypertension and hyperlipidemia was admitted to the hospital with new-onset, severe headache, nausea, and vomiting. Upon arrival, she was febrile to 104®F but was mentating normally. During computed tomography (CT) of the head, her mentation acutely worsened, and she became unable to follow commands or converse appropriately. CT did not demonstrate evidence of acute processes that would explain her symptoms or evidence of increased intracranial pressure. She underwent a lumbar puncture which revealed very elevated WBCs with 94% neutrophils, high protein, and low glucose. She was empirically started on Vancomycin, ceftriaxone, ampicillin, and dexamethasone. Her mental status quickly improved, and she defervesced. Gram stain of her cerebrospinal fluid grew gram positive cocci in clusters & pairs and blood cultures grew streptococcus pneumoniae. She was discharged with a prolonged course of ceftriaxone.

☆ Only the named authors of this manuscript contributed to the content and writing of this manuscript. None of the authors received financial compensation from an external source in return for writing or publishing this paper.

* Corresponding author.
E-mail address: kjadmire@carilionclinic.org (K. Admire).

https://doi.org/10.1016/j.rmcr.2023.101814
Received 17 March 2022; Received in revised form 9 January 2023; Accepted 13 January 2023
Available online 11 February 2023

Fig. 1. Computed tomography demonstrating dehiscence of the superior posterior ethmoid air cell indicated by arrows. Panel A shows a coronal view. Panel B shows a sagittal view.

Shortly following discharge, she developed severe, intractable headaches and unilateral (right sided), salty-tasting rhinorrhea. A few months after the rhinorrhea began, she developed a sensation of nasal drainage running down the throat. This was followed by a progressive, persistent, non-productive cough. On initial examination, she was noted to have hyperemic and swollen nasal mucosa with thin secretions. She was evaluated by her PCP who trial inhaled corticosteroids with no improvement. She was then seen by allergist who prescribed nasal and oral corticosteroids as well as antimicrobial therapy, but her symptoms were not alleviated by these measures.

She underwent evaluation by allergy/immunology, otolaryngology, and pulmonology. Which did not reveal a history of known exposure to *Bordetella* pertussis, DVT/PE, mechanical ventilation, or respiratory illness requiring hospitalization, or gastroesophageal reflux symptoms. Chest radiography showed no acute cardiopulmonary process and no evidence of chronic pulmonary disease. Diagnostic rigid sinus endoscopy did not reveal obvious pathology in the nasal passages, nasopharynx, or middle meatus. The patient was continuing to have symptoms despite the initiation of proton-pump inhibitor, corticosteroid, and antihistamine therapy. CT of the sinuses was obtained which revealed dehiscence in the superior posterior ethmoid air cell with dependent fluid collection (Fig. 1). She was prescribed a humidifier for her bedroom which alleviated some of her nasal drainage. The patient was unable to provide a sample of nasal secretions for Beta-2 Transferrin testing. A diagnosis of upper airway cough syndrome due to cerebrospinal rhinorrhea after pneumococcal meningitis was made.

She underwent endoscopic repair of the right ethmoid sinus roof by otolaryngology and her rhinorrhea resolved immediately with the cough resolving shortly after. To date, the patient has had no further cough or other upper respiratory symptoms.

3. Discussion

Cerebrospinal fluid (CSF) rhinorrhea is an exceedingly uncommon condition which has the potential for significant morbidity and mortality due to the inherent disruption in the barrier between the nasal cavity and the cranial fossae [1].

Etiologies of CSF leaks are traditionally grouped into categories of traumatic, iatrogenic, congenital, tumor-related, or spontaneous [1,2]. The vast majority (> 90%) of CSF rhinorrhea cases are due to either blunt or penetrative head trauma with most of these patients presenting with CSF leak within 48 hours of injury. Iatrogenic CSF leaks can be seen after any surgical manipulation near the skull base. Typically, iatrogenic causes will present with a CSF leak a few days following the procedure. Congenital causes of CSF rhinorrhea are almost exclusively seen in the pediatric population and generally are the result of embryologic defects such as patent foramen cecum or fonticulus frontalis. Tumor-related CSF leaks are associated with locally aggressive lesions which erode the bony structures of the anterior cranial fossa which results in inflammation and subsequent destruction of the dura. Resections of these tumors can cause immediate CSF leakage which is in contrast to other iatrogenic causes, but is generally repaired at the time of resection.

Spontaneous CSF rhinorrhea occurs in patients without one of the previously mentioned causes as a preceding etiology. While previously thought to be idiopathic, there is recent evidence which supports the theory that CSF rhinorrhea is related to a secondary intracranial process. This primarily seems to be due to conditions with increased intracranial pressure [3]. This is the case with our patient who presented with streptococcus pneumoniae meningitis which resulted in increased intracranial pressure. Current evidence suggests that elevated intracranial pressure causes pressure on the anterior skull base which results in bone remodeling, thinning, and finally, defect formation.

Presenting symptoms of CSF rhinorrhea include clear, watery nasal discharge which is usually unilateral. CSF leak should be considered primarily in patients who have had trauma, recent surgical intervention, known intracranial or intranasal tumor. Additionally, patients who have symptoms which may suggest increased intracranial pressure such as headache or visual disturbances, or recent or recurrent episodes of meningitis may warrant further investigation for CSF leak.

Beta-2 Transferrin assay is currently the best available test for identifying the presence of CSF in sinonasal fluid [4,5]. Unfortunately, our patient was not able to provide a sample of fluid for this test to be performed. High-resolution CT is the imaging modality of choice for suspected CSF rhinorrhea as it can show evidence of skull base defect, anatomic abnormalities, or neoplasms which may be eroding the bony structures.

Treatment of CSF rhinorrhea is primarily based on the etiology. Experts recommend conservative management following accidental trauma as the majority of immediate-onset CSF leaks will spontaneously resolve. Other types of CSF leaks such as traumatic with delayed onset, iatrogenic, tumor-associated, and spontaneous often require procedural closure [6]. Surgical management can be done in a variety of ways, but more recently there is a favoring of endoscopic techniques for these closures [7].

4. Conclusions

1. CSF leak is a common complication of trauma and typically has an onset less than 48 hours after the injury
2. Patients who present with rhinorrhea after an episode of meningitis should have careful history and exam performed to determine if CSF leak is a possible cause
3. Beta-2 Transferrin assay is the best test for CSF rhinorrhea, but will not lateralize the defect

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

References

[1] A. Abuabara, Cerebrospinal fluid rhinorrhoea: diagnosis and management, Med. Oral Patol. Oral Cir. Bucal 12 (5) (2007) E397–E400 Published 2007 Sep. 1.
[2] E. Marchiano, E.T. Carniol, D.E. Guzman, M.D. Raikundalia, S. Baredes, J.A. Eloy, An analysis of patients treated for cerebrospinal fluid rhinorrhea in the United States from 2002 to 2010, J Neurol. Surg. B Skull Base 78 (1) (2017 Feb) 18–23.
[3] S.M. Lieberman, S. Chen, D. Jethanamest, R.R. Casiano, Spontaneous CSF rhinorrhea: prevalence of multiple simultaneous skull base defects, Am. J. Rhinol. Allergy 29 (1) (2015 Jan-Feb) 77–81.
[4] R.G. Ryall, M.K. Peacock, D.A. Simpson, Usefulness of beta 2-transferrin assay in the detection of cerebrospinal fluid leaks following head injury, J. Neurosurg. 77 (5) (1992 Nov) 737–739.
[5] G.M. Oakley, J.A. Alt, R.J. Schlosser, R.J. Harvey, R.R. Orlandi, Diagnosis of cerebrospinal fluid rhinorrhea: an evidence-based review with recommendations, Int. Forum. Allergy Rhinol. 6 (1) (2016 Jan) 8–16.
[6] A.S. DeConde, J.D. Suh, V.R. Ramakrishnan, Treatment of cerebrospinal fluid rhinorrhea, Curr. Opin. Otolaryngol. Head Neck Surg. 23 (1) (2015 Feb) 59–64.
[7] E.E. Dodson, C.W. Gross, J.L. Swerdloff, et al., Transnasal endoscopic repair of cerebrospinal fluid rhinorrhea and skull base defects: a review of twenty-nine cases, Otolaryngol. Head Neck Surg. 111 (5) (1994 Nov) 600–605.