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Case Report

Case report: regression of aspiration pneumonitis after nasal endoscopic repair of traumatic cerebrospinal fluid nasal leak

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Abstract: Aspiration pneumonitis is an inflammatory lung disease caused by the inhalation of oropharyngeal secretions colonized by pathogenic bacteria. Accurate diagnosis of aspiration pneumonitis can be challenging, and cerebrospinal fluid (CSF) rhinorrhea is often overlooked as a rare cause of aspiration. In this case report, we present the case of a 48-year-old male patient who experienced right-sided nasal flow of clear watery secretions for 6 months, accompanied by a dry cough as the major symptom. Through comprehensive assessment of clinical symptoms, sinus imaging, nasal endoscopy, and relevant laboratory testing, a presumptive diagnosis of traumatic cribriform plate fracture with CSF rhinorrhea was made. Chest imaging revealed flocculent ground glass shadows in the bilateral lungs. After ruling out viral pneumonia, nasal endoscopic repair of the skull base defect was performed. The patient's dry cough and rhinorrhea symptoms resolved within 1 week after surgery, and the pneumonia showed significant improvement and complete resolution within 2 weeks postoperatively. Despite the absence of characteristic symptoms and evident inhalation factors, chronic CSF rhinorrhea caused by the cribriform plate fracture was ultimately identified as the primary etiology of the patient's aspiration pneumonitis. This rare case highlights the importance of considering traumatic CSF rhinorrhea as an uncommon cause of aspiration, which can enhance physicians' awareness and focus on the less-common etiologies of aspiration. Such awareness can contribute to more accurate diagnosis and early operative intervention, particularly in the context of the coronavirus disease 2019 pandemic.

Keywords: CSF rhinorrhea, aspiration pneumonitis, endoscopic sinus surgery, skull base defect, COVID-19 era

Introduction

Cerebrospinal fluid (CSF) rhinorrhea refers to a CSF leak into the nasal cavity or sinuses due to defective dura mater and through sites such as a broken, ruptured, or thinned anterior or middle cranial fossa base, which may be caused by trauma or other congenital or spontaneous reasons [1]. These skull base injuries primarily originate from tumor invasion, head trauma, or intranasal or intracranial surgery [2]. Although some patients with CSF nasal leaks need only conservative treatment, others with persistent symptoms often require surgical intervention. Continuous CSF nasal leakage can lead to problematic symptoms such as headaches, nasal discharge, olfactory disorders, and visual disturbances [3]. Moreover, spinal fluid leakage from the intracranial space into the upper respiratory tract may lead to grave consequences due to the risk of ascending infection that may cause life-threatening fulminant meningitis [4]. Early repair and intervention are therefore vital.

The emergence and spread of coronavirus disease 2019 (COVID-19) was a global public health crisis that threatens healthcare, education, tourism, and business [5]. Patients with high nasal virus titers and the potential for aerosol production during intranasal instrumentation present a high risk of COVID transmission to medical personnel performing skull base surgeries [6]. The cuneiform plate lateral lamella is the thinnest bone of the anterior skull base and

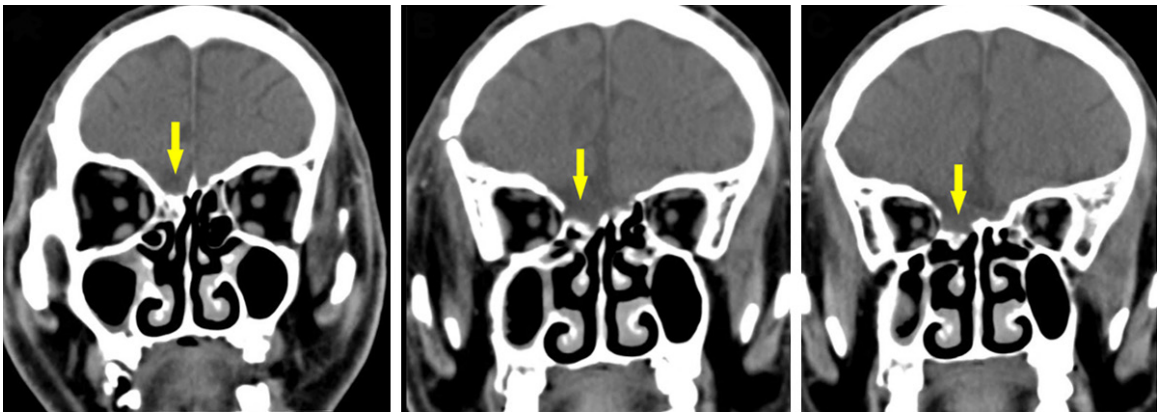

Figure 1. Preoperative CT images of sinus showing a defect in the left roof of the ethmoid sinuses (yellow arrow).

the most vulnerable part of the skull base [7]. Skull base fractures are typically caused by high-impact head trauma. Such fractures may be associated with severe intracranial complications, particularly CSF rhinorrhea, due to the inherent anatomic structure of the skull base [8]. Persistent CSF nasal leakage necessitates surgical treatment due to the risk of fulminant meningitis. In the COVID-19 era, endonasal endoscopic filling with nasal autogenous material is an effective treatment for CSF rhinorrhea, as skull base defects are localized [9].

Typically, the term "pneumonitis" refers to lung tissue inflammation unrelated to intracranial lesions and may manifest with symptoms such as wheezing, coughing, and shortness of breath [10]. Common etiologies include chemical exposure, infectious agents, inhalation, or radiation [11]. Aspiration pneumonitis is an infectious lung disorder caused by massive aspiration of the upper gastrointestinal or oropharyngeal contents. It is more common among people with risk factors such as dysphagia and disorder of consciousness [12]. However, due to the uncertain etiology and atypical clinical characteristics, the prompt diagnosis of aspiration pneumonitis can be challenging. Pulmonary involvement associated with CSF nasal leaks is rare; to our knowledge, only a few cases have been reported to date [13-15]. Here, we present a case of aspiration pneumonitis resulting from traumatic CSF rhinorrhea. The patient's CSF leakage was controlled after surgical repair by nasal endoscopy, leading to the resolution of most pneumonia symptoms and imaging features.

Case description

A 48-year-old man presented with a 6-month history of right-sided watery rhinorrhea. The spontaneous clear, salty nasal discharge stemmed from a closed head injury sustained in a traumatic motor vehicle accident 6 months earlier. After initial management involving bed rest and observation, the patient was discharged from a local hospital after 7 days. He denied any history of recurrent sinusitis, obstructive sleep apnea hypopnea syndrome, meningitis, or sinus surgery, and any recent symptoms of headaches or elevated intracranial pressure. Four months later, he developed a frequent dry cough due to rhinorrhea, which gradually worsened, specifically when recumbent, which led to poor sleep quality. Furthermore, the patient recently developed mild olfactory disorders. Despite receiving antibiotic treatment at the local community hospital, his symptoms did not significantly improve.

The patient sought further treatment at our outpatient department, where he underwent sinus computed tomography (CT) scan and nasal endoscopy. The sinus CT revealed a defect in the right roof of the ethmoid sinuses (**Figure 1**). Nasal endoscopy revealed slow-flow leak of clear fluid into the nasal cavity and posterior pharyngeal wall as well as white crusts in the nasal cavity (**Figure 2A**). This was combined with a positive β2-transferrin test and the presence of glucose in nasal secretions (4.2 mmol/L). The physician diagnosed the patient with a traumatic cribriform plate fracture accompanied by CSF rhinorrhea.

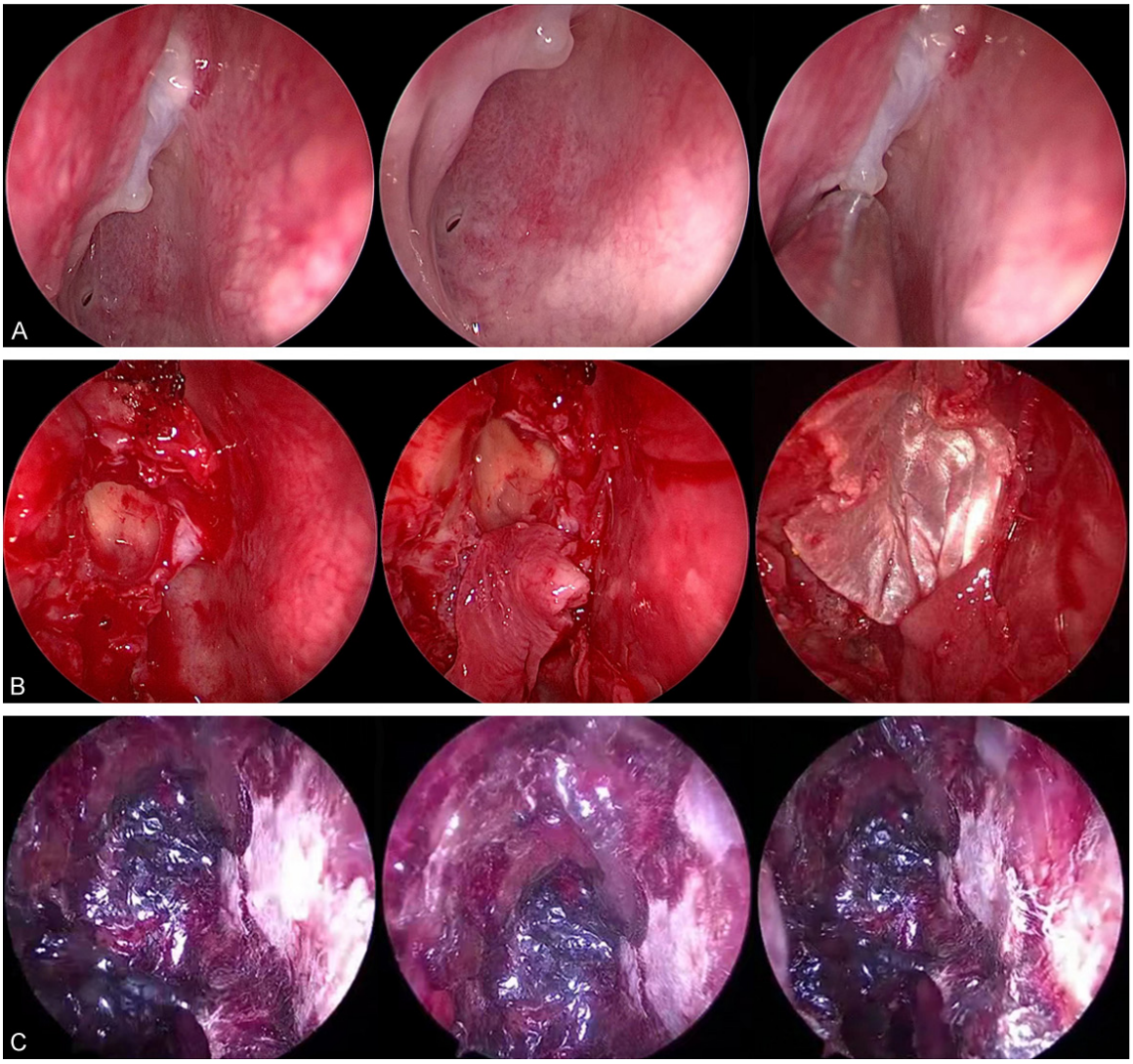

Figure 2. Endoscopic sinus surgery, 70° endoscopic view. A. Preoperative status; B. Intraoperative status; C. Status at 2 weeks after surgery.

The chest CT scan revealed multiple flocculent ground glass shadows in the bilateral lungs. The patient had no COVID-19-associated epidemiologic history. Furthermore, COVID-19 was excluded after COVID-19 antibody testing of the peripheral blood and nucleic acid testing of oropharyngeal swabs were both negative. Moreover, tests for nine other respiratory pathogens and Epstein-Barr virus nucleic acid were also negative, which excluded the possibility of viral pneumonia. Additionally, the patient's pulmonary function tests demonstrated normal lung ventilation, and his serum allergen-specific immunoglobulin E test result was normal.

Based on these examination results, we initially concluded that the lung inflammation might have been caused by long-term CSF inhalation. Subsequently, the patient underwent endoscopic repair of the skull base defect using autologous nasal septal mucosal flaps and fascia lata (**Figure 2B**). The patient's dry cough was significantly relieved 1 week after the repair. Nasal endoscopy demonstrated recovery of the repaired area, with complete disappearance of CSF leak 2 weeks postoperatively (**Figure 2C**). Compared to the preoperative images (**Figure 3A**), pneumonitis appeared to persist 1 week postoperatively, mainly in the upper lobe of the right lung despite the disappearance of some multi-flocculated ground glass shadows (**Figure 3B**). Most of the pulmonary infiltrates disappeared 2 weeks postoper-

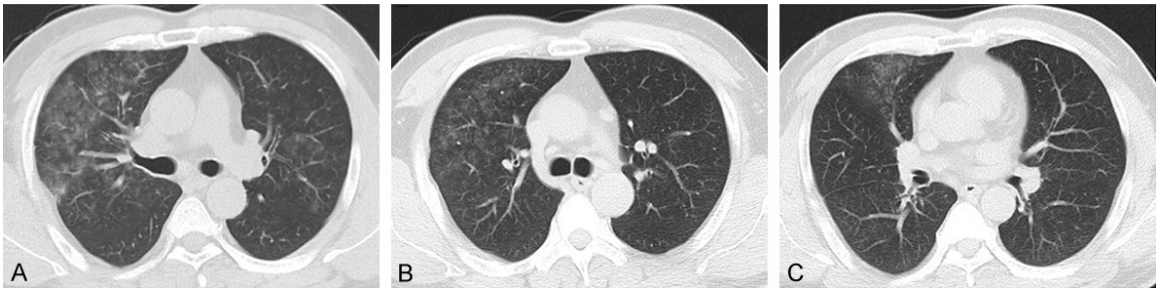

Figure 3. Chest CT images. A. Preoperative chest CT image; B. Chest CT image 1 week after surgery; C. Chest CT image 2 weeks after surgery.

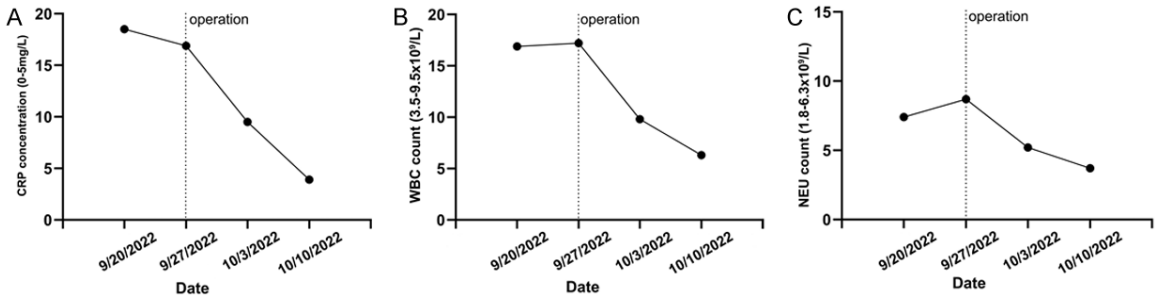

Figure 4. Postoperative changes in infection-associated inflammatory indicators. A. C-reactive protein (CRP) concentration; B. White blood cell (WBC) count; C. Neutrophil (NEU) count.

Table 1. Changes in blood biochemical indexes

Code	Indexes	Preoperative results	Postoperative results	Units
1	K^+	4.50	3.96	mmol/L
2	Na^+	144.63	143.22	mmol/L
3	Cl^-	103.17	103.78	mmol/L
4	Ca^{2+}	2.27	2.17	mmol/L
5	Mg^{2+}	0.81	0.91	mmol/L
6	Urea	3.77	4.97	mmol/L
7	Cr	65.00	61.00	μmol/L
8	ALT	24.80	23.80	U/L
9	GLU	5.49	4.37	μmol/L
10	AST/ALT	0.61	0.65	-

atively (**Figure 3C**). Additionally, infection-related inflammatory indicators, including C-reactive protein, white blood cells, and neutrophils, improved completely 2 weeks postoperatively (**Figure 4**). No significant abnormalities were found in the preoperative or 2-week postoperative results of blood biochemical indexes and urine routine tests (**Tables 1**, **2**). At the 6-month follow-up, the patient had no dry cough or CSF rhinorrhea (**Figure 5**).

Discussion

Aspiration pneumonitis is defined as an infectious lung lesion caused by the aspiration of large amounts of oropharyngeal secretions, gastric contents, and food, and is characterized by high incidence, insidious onset, and frequent neglect [1, 2]. Aspiration pneumonitis accounts for 5-15% of all cases of community-acquired pneumonitis [12]. Patients with aspiration pneumonitis had significantly increased hospitalization and 30-day mortality compared to patients with non-aspiration pneumonitis [16]. Therefore, the timely detection of inhalant-related factors is crucial for early diagnosis and therapy. One of the main reasons some patients are not initially definitively diagnosed with aspiration pneumonia is the similarity in clinical symptoms and chest CT imaging features to those of viral pneumonia. Common respiratory tract infection symptoms such as runny nose and dry cough may not initially raise sufficient concern from patients. Many imaging features of aspiration pneumonitis, such as ground glass shadows and multiple patches in the bilateral lungs, resemble those seen in most viral pneumonias, including COVID-19 [17].

Table 2. Routine urine test results

Code	Index	Preoperative results	Postoperative results	Units
1	Urine pH	4.8	4.9	-
2	Urine specific gravity	1.018	1.021	-
3	Urine protein	-	-	g/L
4	Urine glucose	-	-	mmol/L
5	Urine red blood cell	1	1	/μL
6	Urine white blood cell	2	1	/μL
7	Urine ketone bodies	-	-	-

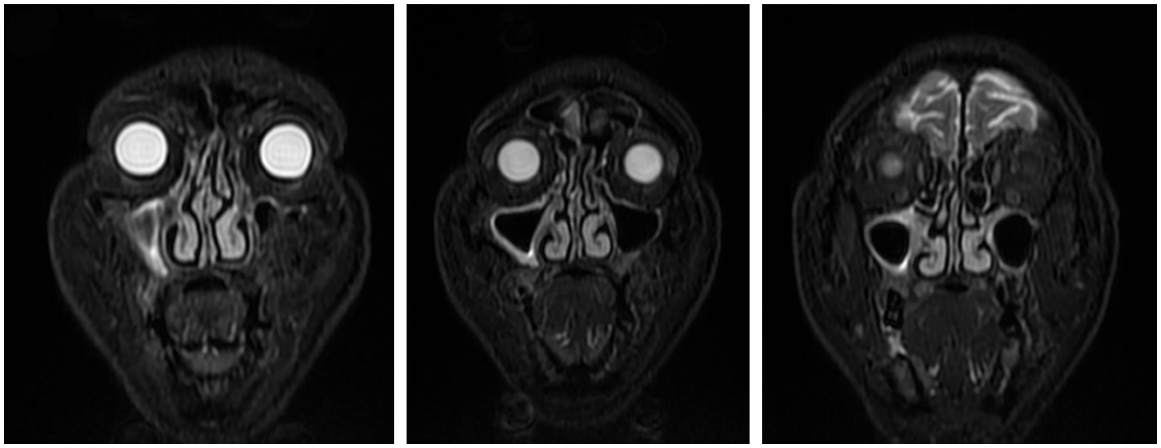

Figure 5. Sinus MRI images at 6 months after surgery.

Furthermore, the lack of definitive aspiration factors is another important reason. Patients with neurologic disorders, chronic degenerative diseases, or esophageal diseases are generally at a greater risk of aspiration. Moreover, as demonstrated in this case, directly observing a persistent CSF nasal leak is difficult. These factors delay the physician's understanding of the etiology of aspiration pneumonitis.

Currently, pulmonary infiltration caused by CSF rhinorrhea is under-recognized and rarely reported. Most cases present with nonspecific respiratory symptoms such as cough, fever, dyspnea, and shortness of breath. Nasal symptoms are frequently ignored or considered nonspecific or irrelevant. Our patient's symptoms, imaging findings, and inflammation-related indicators (due to pneumonia) rapidly improved after endoscopic surgical repair. Our findings demonstrate that traumatic CSF rhinorrhea is an important aspiration pneumonitis trigger. A previous study reported that nasal secretions could enter the lungs of both patients with cystic fibrosis and healthy individuals [18]. It is possible that bacteria from the nasal cavity and oropharynx can enter and remain in the lungs, which together with CSF elicits persistent local inflammatory responses. Persistent dry cough can increase intracranial pressure, thereby worsening CSF rhinorrhea and sustained aspiration. Additionally, persistent cough due to CSF nasal leakage tends to lead to gastric content reflux, which may also increase the risk of aspiration. Given our findings, further investigation that can directly link aspiration pneumonitis with CSF rhinorrhea is warranted.

When a patient presents with refractory pneumonia with chronic rhinorrhea, physicians should remain vigilant for the possibility of CSF rhinorrhea, especially if there is a prior induction event, such as head trauma. CSF rhinorrhea can be identified by measuring glucose or β2-transferrin, which is almost exclusively found in CSF [15]. Imaging procedures and otolaryngological examinations can also aid the diagnosis. It remains unclear whether CSF stimulates the lungs or whether inhaling bacteria-contaminated CSF causes infectious pneu-

monia, although both mechanisms may be involved. Ultimately, refractory pneumonia associated with CSF nasal leakage is treated with leakage control. Antibiotics may be beneficial to control the infection, but only temporarily. Some CSF nasal leaks recover spontaneously, but surgical repair is necessary for sustained leaks. Surgical delay should be avoided given the potential for nasal symptoms, aspiration pneumonia from untreated leaks, and the associated high risk of fulminant meningitis [1, 2]. Although rare, traumatic CSF rhinorrhea is one of the etiologies of massive aspiration. Timely and effective endoscopic repair of CSF rhinorrhea contributes to the avoidance of massive aspiration, leading to a lower incidence of aspiration pneumonia.

Conclusion

We present a symptomatic case of aspiration pneumonitis caused by CSF nasal leakage. While antibiotics can be used in the presence of pulmonary infiltrates, the effective treatment ultimately lies in repairing the CSF rhinorrhea. In the COVID-19 era, lung lesions due to CSF rhinorrhea may be detected incidentally by screening CT scans. Patients with CSF nasal leakage who also develop pulmonary symptoms and demonstrate specific findings, particularly ground glass shadows on chest CT, should be examined further for traumatic CSF rhinorrhea. Currently, pulmonary involvement in CSF rhinorrhea is often underdiagnosed. Although nasal endoscopic repair is the curative therapy, it is typically delayed because of the presence of pulmonary infiltration. Therefore, physicians should acknowledge that CSF rhinorrhea may be an important factor in refractory pulmonary infiltrates and that adequate and timely treatment is necessary to reduce complications.

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Disclosure of conflict of interest

None.

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