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Images: Unilateral rhinorrhea in a patient starting autotitrating positive airway pressure therapy for obstructive sleep apnea

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ABSTRACT

We rerpot a case of a 65-year-old obese female who developed a unilateral nasal cerebrospinal fluid (CSF) leak after starting autotitrating positive airway pressure therapy for obstructive sleep apnea. The CSF leak was confirmed by beta-2 transferrin testing of the nasal fluid, as well as by identification of the leak through the anterior cribriform plate after administration of intrathecal fluorescein. The CSF leak was successfully repaired endoscopically, and autotitrating positive airway pressure was reinitiated one month postoperatively.
Keywords: obstructive sleep apnea; cerebrospinal fluid leak; rhinorrhea

INTRODUCTION

Rhinorrhea is a commonly reported symptom after initiating continuous positive airway pressure (CPAP) therapy for obstructive sleep apnea (OSA).[1] The differential diagnosis of rhinorrhea is broad, but one of the most serious causes is cerebrospinal fluid (CSF) leak. CSF leaks place patients at risk for meningitis and pneumocephalus,[2] and timely recognition and repair are important to prevent life-threatening sequelae. This case presentation describes a patient with OSA who developed a nasal CSF leak after starting autotitrating positive airway pressure (APAP), highlighting optimal diagnostic and therapeutic strategies for both CSF and perioperative OSA management.

REPORT OF CASE

A 65-year-old obese female with moderate OSA with a respiratory index of 20 on home sleep apnea testing was started on APAP 5-20 cm water with a nasal interface. She experienced symptomatic improvement with excellent APAP adherence. Six weeks after starting APAP, she developed right-sided clear rhinorrhea that worsened with bending forward. She denied new headaches, vision changes, recent head trauma or sinonasal surgery. Her pressure was adjusted to 12-20 cm water, because the reported apnea-hypopnea index from her device was 7.3 events an hour and 95th pressure was 12.2 cm water. Her rhinorrhea did not improve after 1 month of oral loratadine and topical fluticasone, so she was referred to an otolaryngologist. After an otolaryngologist's consultation, APAP was stopped, approximately 4 months after the onset of her rhinorrhea. Her nasal fluid tested positive for beta-2 transferrin, suggesting a CSF leak. Sinus computed tomography (CT) revealed no obvious skull base bony defect (Figure 1). The patient was taken to the operating room for nasal endoscopic exploration and repair of CSF leak, with intrathecal fluorescein to facilitate leak site localization. Lumbar opening pressure was normal at 20 mmHg. Fluorescein was identified leaking through a 2-mm defect in the right anterior cribriform plate (Figure 2). The defect was repaired successfully endoscopically with a free nasal septal mucosal graft. One month postoperatively, the free mucosal graft was healed into the skull base, and APAP 12-20 cm water was re-initiated. By 6 months postoperatively, there was no CSF leak recurrence.

DISCUSSION

Rhinorrhea is commonly encountered in patients with sleep-related breathing disorders,[1] and can occur after starting CPAP therapy.[3] Nasal CSF leaks are one cause of rhinorrhea and can occur spontaneously from idiopathic intracranial hypertension (IIH), skull base tumors, or traumatically from accidental or iatrogenic trauma.[2,4,5] Compared to other causes of rhinorrhea, nasal CSF leaks more commonly present unilaterally, and tend to be triggered by postural changes or straining.[2]

IIH often presents in obese females in their 30s-40s, but IIH-related CSF leaks tend to present later in their 50s.[4] Given these demographic factors, a significant proportion of patients with IIH and CSF leaks will have concurrent OSA.[2] A meta-analysis demonstrated CSF leaks being 4.73 times more likely in OSA compared to controls.[6] Additionally, another study on healthy subjects demonstrated increased intracranial pressure (ICP) after using CPAP of 12 cm H_2O.[7] There have also been case reports of patients with OSA developing CSF leaks after starting CPAP.[8,9] OSA can possibly increase ICP due to increases in hypercapnia leading to cerebral vasodilation.[10] Moreover, animal models have suggested that increases in positive end expiratory pressure can increase ICP due to transmission of pressure to the jugular and vertebral veins blocking cerebral venous return and increasing cerebral volume.[11] Together, these studies suggest having OSA and CPAP treatment can both contribute to increased ICP, placing patients at risk for CSF leaks, especially if they have premorbid skull base defects or IIH. An example of this relationship was highlighted by the patient in the current case report, who may have suffered from IIH that resulted in

a small cribriform plate bony defect and meningocele, which subsequently leaked after starting APAP. While her opening lumbar pressure was normal, that could have been due to her active nasal leaking.

This case highlights that CSF rhinorrhea should be considered in OSA patients presenting with unilateral clear nasal drainage with or without headache. If a CSF leak is suspected, patients should stop CPAP to prevent pneumocephalus,[12] undergo nasal fluid beta-2 transferrin testing, and be referred to an otolaryngologist.[2] Beta-2 transferrin is highly sensitive and specific for CSF, and remains stable for at least 2 weeks while patients collect their nasal fluid, whether refrigerated or stored at room temperature.[5] Once CSF is confirmed, a sinus CT scan is obtained for leak site localization, then endoscopic repair is planned. When the defect is not apparent on CT, intrathecal fluorescein injected preoperatively can facilitate intraoperative leak site identification.[13] Endoscopic repair is then successful in 90%-97% of cases.[4]

One clinical dilemma postoperatively pertains to re-initiating CPAP for patients with OSA after nasal CSF leak repair. No studies have directly assessed this, but a recent international consensus statement showed that surgeons tend to delay CPAP initiation for 2-3 weeks postoperatively to prevent CSF leak recurrence and pneumocephalus.[2]

In conclusion, unilateral rhinorrhea should raise suspicion of a nasal CSF leak, for which there may be an association with OSA and CPAP use. Close collaboration with an otolaryngologist will ensure appropriate diagnosis and endoscopic management. To prevent pneumocephalus, patients with active CSF leaks should not use CPAP. Postoperatively, clinicians must weigh the benefits and risks of CPAP, but consider delaying at least 2-3 weeks.

ABBREVIATIONS

APAP, autotitrating positive airway pressure
CPAP, continuous positive airway pressure
CSF, cerebrospinal fluid
ICP, intracranial pressure
IIH, idiopathic intracranial hypertension
OSA, obstructive sleep apnea

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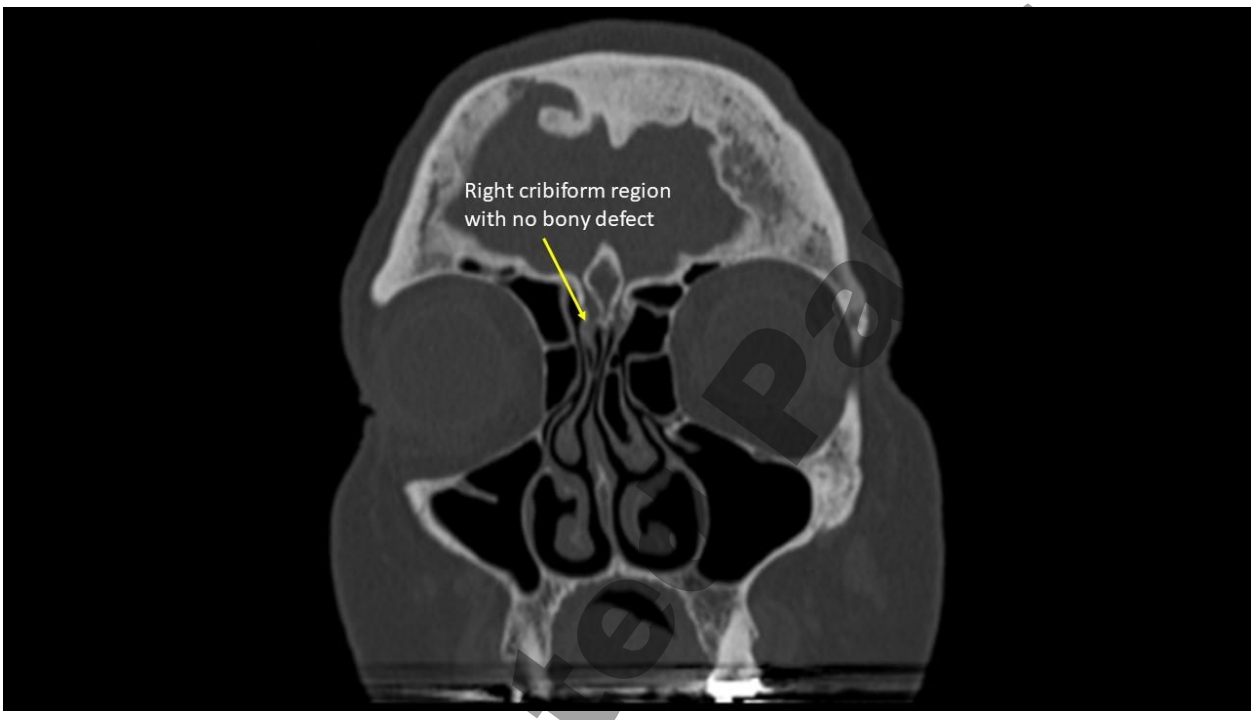
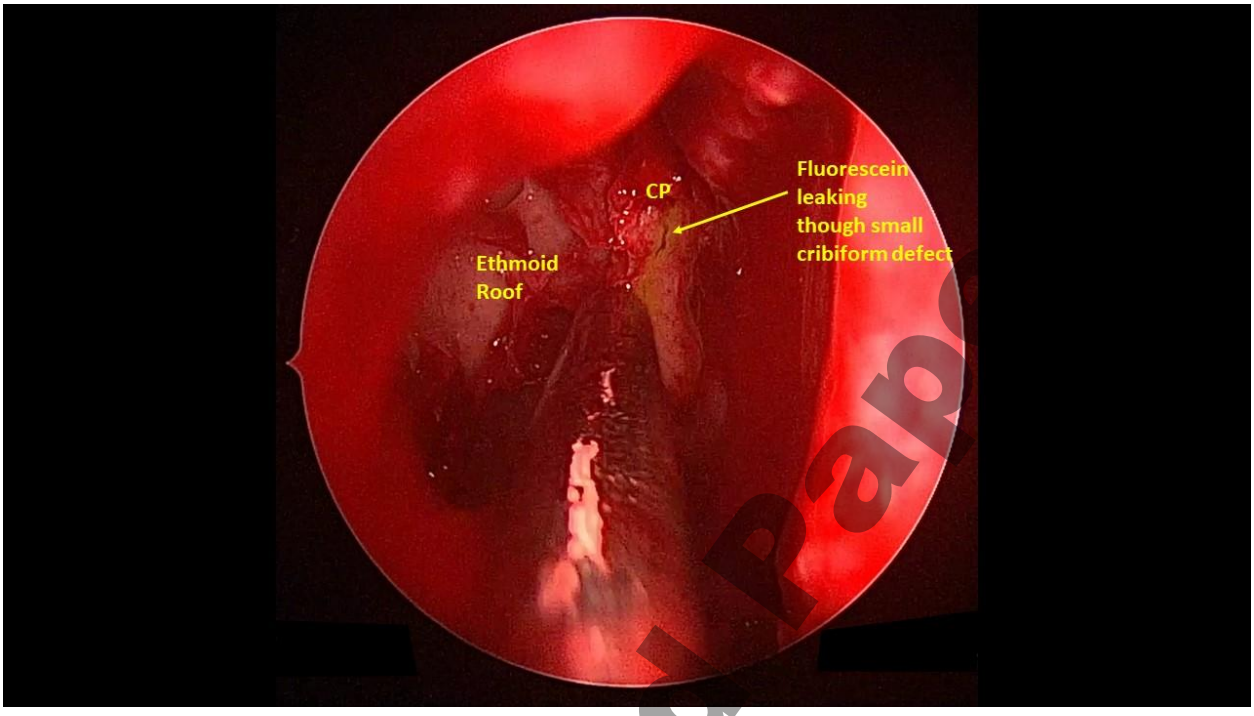
FIGURE TITLES AND LEGENDS

Figure 1: Coronal bone-window sinus computed tomography showing no overt defect in the ethmoid or cribriform regions of the anterior skull base.



In these situations, one option for localizing the CSF leak site is intrathecal fluorescein injected through a lumbar drain at the time of surgery.

Figure 2: Endonasal endoscopic view of the right ethmoid sinus region, highlighting the cribriform plate (CP) region and the small linear cribriform defect and resultant fluorescein-stained CSF leaking through the defect.



This allowed precise localization of the CSF leak site, which allowed for successful endoscopic repair.