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Obstructive Sleep Apnea Syndrome Revealing an Osteomeningeal Breach: A Case Report

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Abstract **Case Report**

Osteomeningeal breach is a discontinuity in the osteomeningeal layer that allows cerebrospinal fluid to flow into an air-filled cavity at the base of the skull. Its severity is associated with the risk of central nervous system infection. The majority of breaches are traumatic in origin, either accidental or iatrogenic. Spontaneous cerebrospinal fluid rhinorrhea is less commonly found. Obesity, often coupled to obstructive sleep apnea syndrome, can lead to hypercapnia, increasing the pressure of cerebrospinal fluid and thereby facilitating the development of osteomeningeal breaches. In cases of intracranial hypertension, the displacement of the pituitary gland can occur, and as a result, its cavity can be filled with cerebrospinal fluid, which characteristically describes an empty sella turcica syndrome. We present the case of a 52-year-old woman initially admitted for the management of sleep apnea syndrome, along with a history of chronic rhinorrhea. Analysis of the rhinorrhea fluid and craniofacial imaging revealed a cerebrospinal fluid leak through a breach in the right cribriform plate, associated with an empty sella turcica syndrome. The patient underwent surgical repair of the meningeal breach via an endonasal approach, leading to a successful resolution of cerebrospinal fluid rhinorrhea.

Keywords: Osteomeningeal Breach, Endoscopic Endonasal Surgery, Cerebrospinal Leak, CT, MRI, Rhinorrhea.

1. INTRODUCTION

The obstructive sleep apnea syndrome (OSAS) is a common condition affecting 2 to 5% of the adult population. It is characterized by obstructive apneas resulting from the closure of the upper airways, with obesity being a major risk factor. OSAS can lead to hypercapnia, potentially causing a chronic increase in cerebrospinal fluid pressure and an elevated risk of osteomeningeal breach. Additionally, individuals with a hypoplastic sellar diaphragm may experience intrasellar herniation of the subarachnoid space, leading to an empty sella turcica syndrome.

2. CASE REPORT

Our case involves a 52-year-old female patient with no notable medical history. The patient and her family report nocturnal snoring, with episodes of apnea, and morning headaches. Additionally, the patient complains of allergic rhinitis and presents intermittent episodes of rhinorrhea, with no improvement after medical treatment.

Upon physical examination, the patient presented in good overall condition, with a body mass index of 41 kg/m2 and blood pressure measuring 135/80 mmHg. Otorhinolaryngological assessment revealed rhinorrhea from both nasal cavities, slightly more pronounced on the right side. Nasofibroscopy has shown inflamed pale mucosa, with hypertrophy of the inferior turbinates. There was no significant septal deviation. Moreover, base of the tongue was broad and contributed to anteroposterior narrowing of the oropharyngeal lumen.

A CT scan of the skull base and sinus was done to better explore naso-sinusal cavities. It demonstrated a slight deviation of the nasal septum along with minor thickening of the maxillary sinus wall. Furthermore, it identified also a 3mm defect in the right cribriform plate (Fig 1).

Given this radiological finding, a sample of the rhinorrhea fluid was collected. This allowed for the detection, via immuno-electrophoresis, of beta-2-transferrin protein with a positive value of 22.5 mg/l, confirming a cerebrospinal fluid leak. Subsequent MRI imaging confirmed cerebrospinal leak through a basal frontal defect in the right ethmoid cribriform plate (Fig 2). Additionally, MRI revealed an empty sella turcica (Fig 3 and 4) with dilated and tortuous optic nerve sheaths (Fig 5). No evidence of encephalocele or

meningocoele was found. A lumbar puncture with pressure measure was performed, along with an ophthalmological examination, which were normal. Hormonal assessment of the pituitary axis (TSH, FSH, ACTH, and Prolactin) yielded normal results.

Surgical repair of the osteomeningeal defect was performed via an endonasal approach under general anesthesia, preceded by mucosal application of lidocaine and adrenaline for nasal cavity preparation. The defect was identified, and the surrounding mucosa was dissected, followed by bipolar coagulation to facilitate tissue retraction and reduce the defect size. Closure of the osteomeningeal defect was achieved using an abdominal fat graft reinforced with septal cartilage (Fig 6). Application of biological glue preceded the placement of a Silastic sheet to complete the procedure.

Clinical course was favorable, marked by the absence of rhinorrhea and signs of meningitis over a 12-month follow-up. Also, the patient underwent management of her obstructive sleep apnea syndrome (OSAS) in collaboration with pulmonologists. Continuous positive airway pressure (CPAP) ventilation was recommended, yielding positive outcomes in ameliorating snoring and nocturnal apneas.

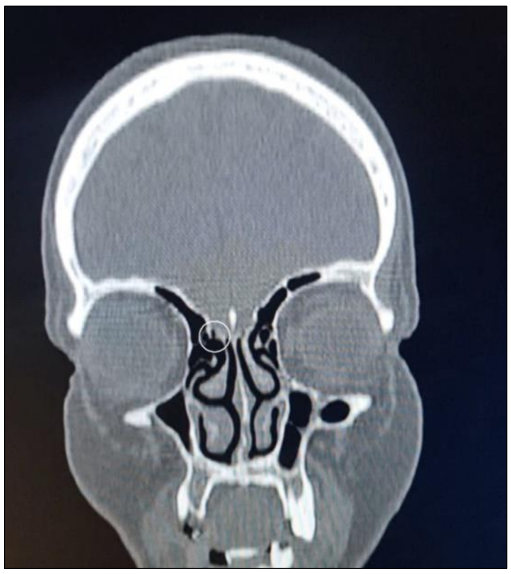

Figure 1: CT scan of the skull base and sinus: Defect in the right cribriform plate.

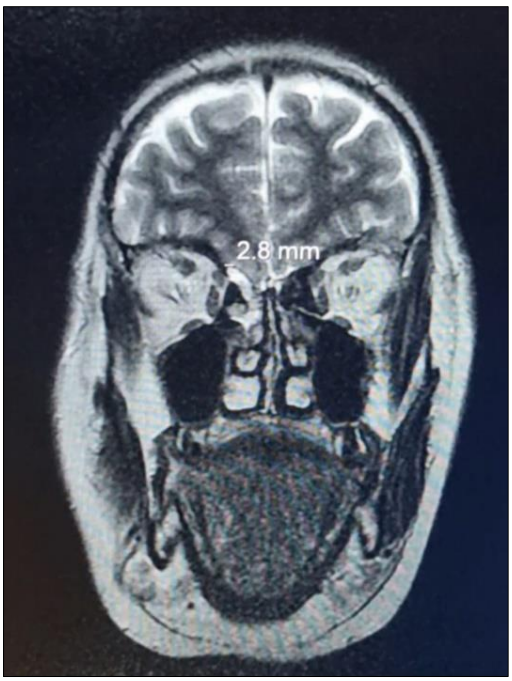

Figure 2: MRI confirmed cerebrospinal leak through a defect in the right ethmoid cribriform plate measuring 2.8 mm

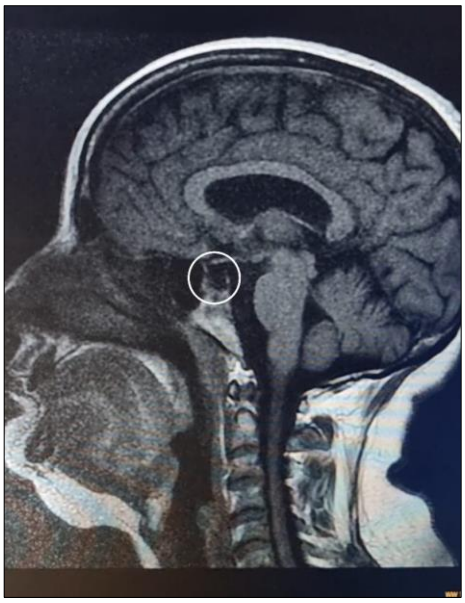
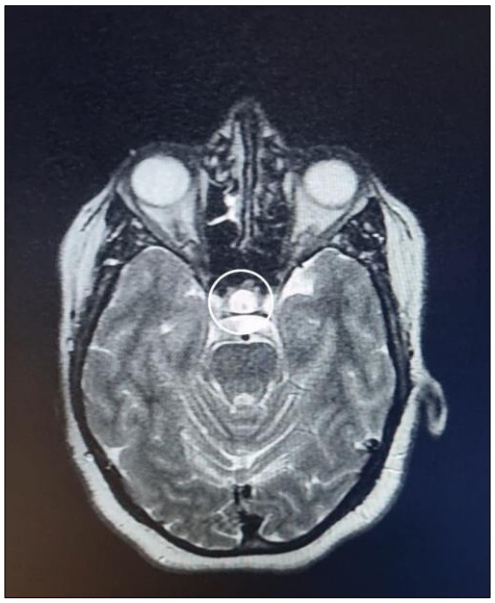

Figure 3 & 4: MRI showing an empty sella turcica; with Cerebrospinal fluid (CSF) filling its space.

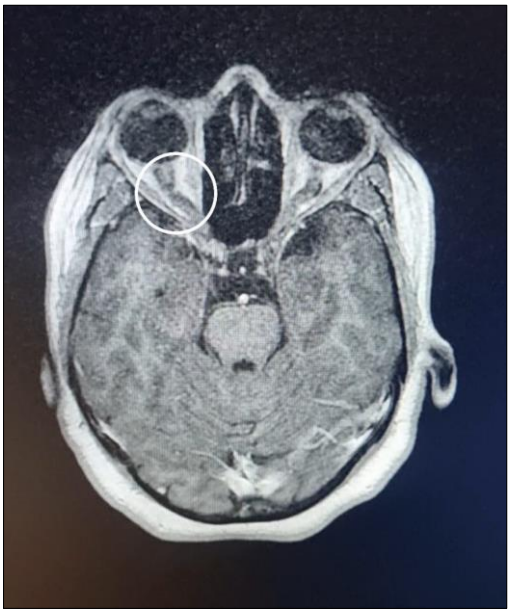

Figure 5: MRI showing with dilated and tortuous optic nerve sheaths: sign of intracranial hypertension.

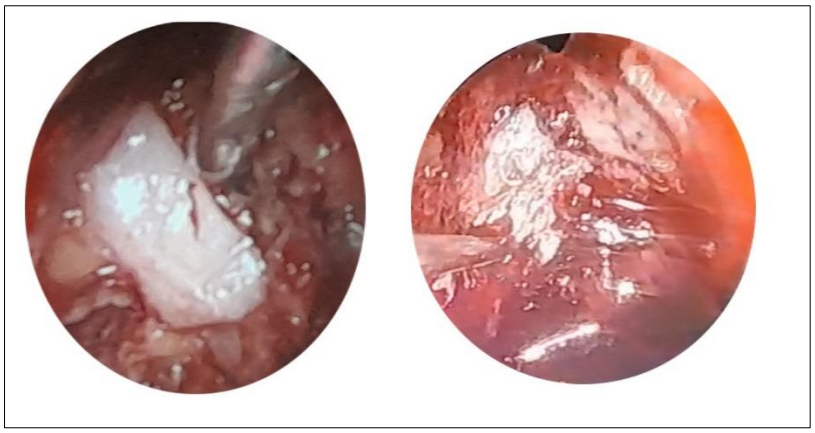

Figure 6: Repair of the breach through the transposition of a fat plug with cartilage reinforcement, secured with biological glue.

3. DISCUSSION

Osteomeningeal breach is a discontinuity in the osteomeningeal layer allowing cerebrospinal fluid (CSF) to flow into a facial hydroaeric cavity [1-16]. Osteomeningeal defects are categorized into secondary and primary groups [3-17]. Secondary post-traumatic etiologies regroup accidental and iatrogenic rhinorrhea. They occur at sites of meningeal or bone weakness. The literature underscores a marked male predominance, with an incidence peak between 30 and 50 years, largely attributed to post-traumatic etiologies. In contrast, primary rhinorrhea, accounting for only 3 to 4% of cases, more commonly localizes to the frontal and ethmoid sinuses, with rare occurrences in the sphenoidal sinus [4]. Primary rhinorrhea is classified into two types: normal CSF pressure rhinorrhea and high CSF pressure rhinorrhea. This one is occasionally associated with empty sella syndrome [4]. The arachnoid presents microdefects and micro herniations at its weak points. Due to fluctuations in CSF pressure and pulsations, these weak points may expand, leading to the development of defects, herniations, and the formation of arachnoid diverticula. Once in contact with the dura mater, these diverticula can result in its fenestration and the formation of meningeal defects [4].

Syndrome of sella turcica refers to a condition where there is herniation through the diaphragma sellae of the arachnoid into the suprasellar space, containing cerebrospinal fluid [5]. It is commonly observed in middle-aged, obese, and hypertensive females. This syndrome is an indicator of idiopathic intracranial hypertension [6]. Obesity, often associated with obstructive sleep apnea, can lead to hypercapnia, increasing cerebrospinal fluid pressure and predispose to osteomeningeal defects [7]. Clinical manifestations include frontal-orbital headaches present in about 50% of cases, along with visual disturbances. Furthermore, endocrine manifestations are frequent and can manifest as either hormone under secretion or over secretion syndrome [10].

In case of profuse clear rhinorrhea, the diagnosis of osteo-meningeal breach is evident, and the objective is to establish a precise topographic diagnosis. Conversely, when confronted with intermittent cerebrospinal rhinorrhea, the detection of beta-2-transferrin protein in nasal secretions is employed to confirm the diagnosis. Any clinical suspicion of cerebrospinal fluid breach, impose computed tomography (CT) and magnetic resonance imaging (MRI) to delineate the site of the defect, and identify the underlying etiology. Magnetic Resonance Imaging (MRI) is capable of identifying leaks not visualized by Computed Tomography (CT) scans [9-11]. Additionally, MRI can reveal indicators of underlying intracranial hypertension (ICH), including an empty sella turcica, optic nerve sheath dilatation, increased vertical tortuosity of the optic nerves, scleral flattening at optic nerve insertions, widened arachnoid spaces, Meckel's cave enlargement, and venous sinus stenosis [4-8].

Therapeutic management aims to close the defect typically using an interposition technique [12-16]. This procedure is usually conducted using an endoscopic endonasal approach. However, the neurosurgical approach (craniotomy) remains applicable in specific cases. This technique is often associated with increased morbidity. Consequently, endoscopic approaches are safer and have progressively replaced open and invasive techniques over the past three decades. The filling material may consist of septal mucosa from the middle or inferior turbinates, retroauricular or peri ombilical adipose tissue, temporalis or lata fascia, or even a neuropatch [14, 15]. This graft is applied over the breach and secured in place using biological glue or exogenous materials (such as a silastic sheet) if natural retention in the anatomical region concerned is not feasible [20-22].

4. CONCLUSION

Accurate diagnosis of OMB holds importance in both medical (for preventing infectious cerebral complications) and surgical (for determining the type and location of the breach to be addressed) contexts. This condition should be considered even in front of atypical presentations mimicking chronic rhinosinusitis or in association with other conditions such as obstructive sleep apnea syndrome (OSAS). Imaging techniques essentially computed tomography and MRI are employed to identify osteomeningeal breach. Endoscopic Surgery for CSF rhinorrhea is considered as the basic therapeutic technique, offering favorable outcomes while minimizing perioperative morbidity.

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