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Case Report

Endoscopic surgical approach to a rare bulla ethmoidalis mucocele presenting as proptosis

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ABSTRACT

Paranasal sinus mucoceles are expansile masses that form in the sinuses due to chronic blockage of their ostia, with occurrence in the ethmoid sinus being rare. In this case report, we present a patient diagnosed with a right bulla ethmoidalis mucocele. The patient presented with a short duration of painful right eye proptosis and diplopia. Examination revealed diplopia with restricted right eye movement, while nasoendoscopy was unremarkable. Radiological imaging identified a mucocele in the right anterior ethmoid. The mucocele was treated through marsupialization via a transnasal endoscopic approach, resulting in an improvement of the patient's symptoms. This case report highlights this rare occurrence and role of minimal invasive surgery in treatment.

Keywords: Bulla ethmoidalis, Mucocele, Proptosis, Image-guided surgery, CT navigation, Endoscopic sinus surgery, MRI unavailability

INTRODUCTION

A mucocele is a type of retention cyst, theorised to have been formed from the cystic degeneration of seromucinous gland, that usually develops due to obstructed outflow pathway in the exocrine system.[1,2] Mucoceles are relatively common in the anatomical region of the nose and paranasal sinuses, as these areas are line with pseudostratified columnar epithelium.[3]

It most commonly involves the frontal sinus (60-89%), followed by ethmoid (8-30%) and rarely maxillary and sphenoid sinuses.[2] Maxillary sinus mucoceles are rare in America and Europe, but various studies from Asia have reported their frequent occurrence, particularly following Caldwell-Luc surgery.[2] The occurrence of ethmoidal mucocele is uncommon and rarely reported. We present a case of right bulla ethmoidalis mucocele presenting with painful right eye proptosis.

CASE REPORT

A 46-year-old male with a history of hypertension, smoking, and surgically repaired cleft lip presented with a two-week history of right eye proptosis and one week of ocular pain and diplopia. Notably, there were no accompanying nasal symptoms or visual acuity impairment. Clinical examination revealed right eye exotropia and restricted extraocular movement in lateral and superior gaze, while nasoendoscopy was largely unremarkable except for a nasal floor defect consistent with prior cleft palate. Computed tomography (CT) imaging demonstrated a non-enhancing, hypodense lesion (2.5x2.3x2.7 cm) within the right anterior ethmoidal air cell. The lesion extended into the medial extraconal orbital space, causing bone thinning and erosion without clear demarcation from the medial rectus muscle or intraorbital optic nerve. No intracranial extension was observed. The patient underwent endoscopic sinus surgery under general anesthesia. Following an

uncinectomy, marsupialization of the mucocele was performed with drainage of thick, yellow mucus.

The integrity of the medial orbital wall was preserved, confirmed intraoperatively using the Stankiewicz maneuver. Postoperatively, the patient reported immediate pain relief, and at follow-up, all ocular symptoms had fully resolved.

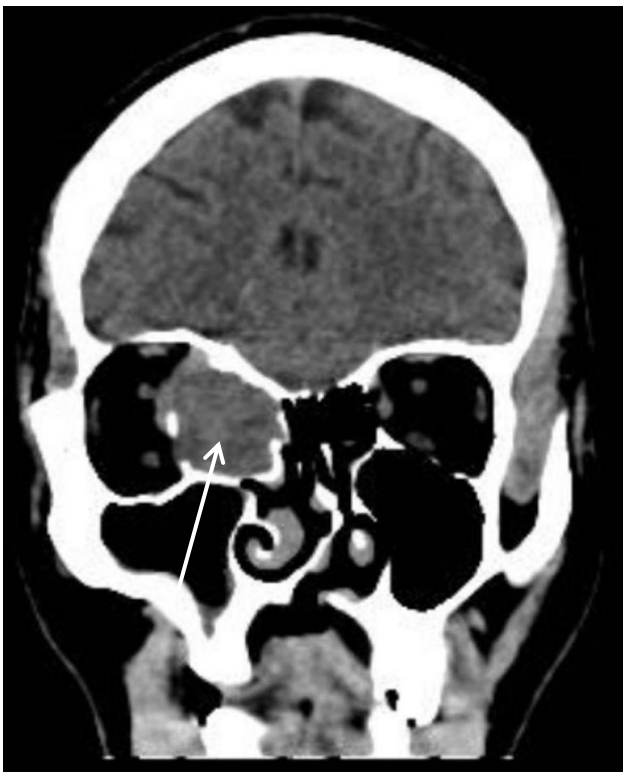

Figure 1: Coronal view shows a non-enhancing heterogenous hypodense lesion seen within the right anterior ethmoidal air cell (white arrow).

Figure 2: Axial view shows there is involvement into the right medial rectus extraconal space which shows no clear plane of demarcation with the right medial rectus muscle (white arrow).

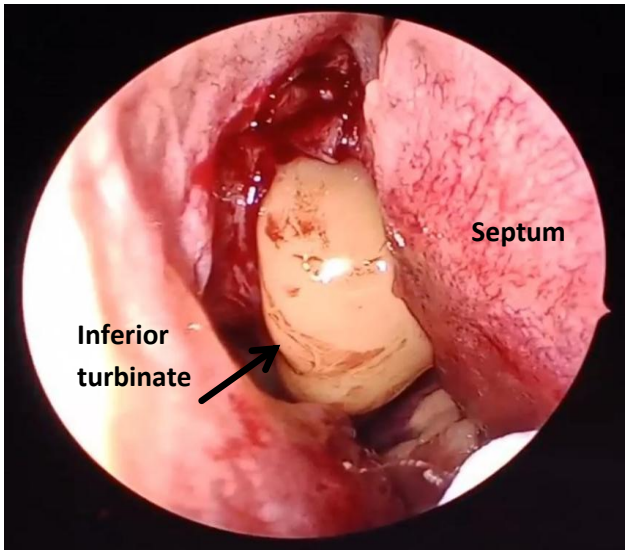

Figure 3: Pus (black arrow) seen draining from bulla ethmoidalis.

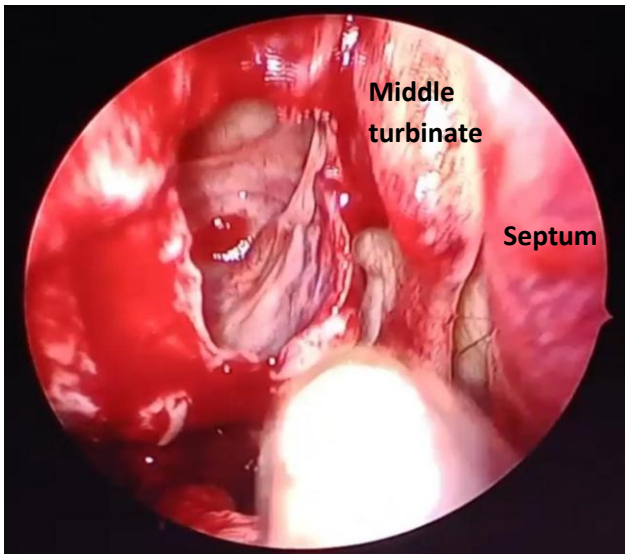

Figure 4: Shows upon drainage of pus.

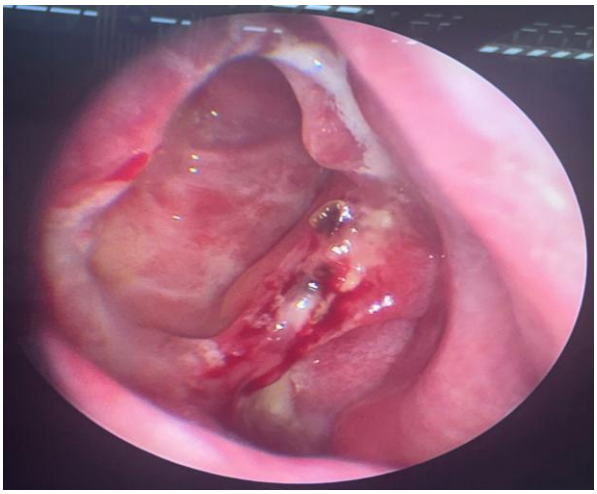

Figure 5: Shows 2 weeks post op.

DISCUSSION

Paranasal sinus mucoceles are mucus-filled, epithelium-lined sacs that appear as benign, expansile lesions within the sinus cavity. These mucoceles can develop at any age but are most frequently observed among individuals aged between 40 and 60 years old with slight male predilection.[5,6] The frontal sinuses are most frequently involved, accounting for 45-65% of cases, followed by the ethmoid sinuses (14-30%), maxillary sinuses (8-10%), and sphenoid sinuses (2-3%).[7,8] Mucoceles involving the bulla ethmoidalis, as seen in this case, are rarely documented in the literature.

Mucocele formation is primarily driven by two factors: obstruction of the sinus outflow tract and an inflammatory response within the sinus.[6,9] Obstruction of the sinus ostia can be attributed to inflammation, trauma or tumor. Cytokines such as IL-1 and tumor necrosis factor, present in the epithelial lining of mucoceles, are believed to disrupt the balance between osteolytic and osteoblastic factors, favoring osteolysis and contributing to the surrounding bony erosion associated with mucocele expansion.[9,10] Besides inflammatory obstruction, primary causes include the cystic dilation of mucosal glands or the degeneration of polyps. Previous sinus surgery or facial trauma are common secondary causes of mucoceles, with the former often presenting as late as two decades after the procedure.[9] Obeso et al, reported that 29% of patients reported no identifiable risk factors for development of mucoceles.[11] In this case, the mucocele in the right bulla ethmoidalis was responsible for causing localized proptosis and restricted eye movement, which can mimic other orbital pathologies, such as orbital tumors or infections. The patient's symptoms, specifically the short duration of right painful proptosis, were consistent with the inflammatory nature of the mucocele, where pressure from the expanding mass likely affected the adjacent orbital structures, including the medial rectus muscle and the surrounding orbital fat.

Computed tomography (CT) remains the first choice of imaging in these cases as it provides detailed information on the bone structure. CT typically demonstrates an expansile, homogeneous mass without rim enhancement, unless there is an associated acute mucopyocele.[6,12] Given the non-enhancing nature of mucoceles on CT, neoplasms should be considered as a differential diagnosis, particularly when there is involvement of adjacent bony structures, with remodelling and areas of erosion.[12] Magnetic resonance imaging (MRI) will therefore be important for soft delineation in these types of cases. The radiological characteristics of mucoceles on MRI evolve with disease progression. In the early stages, mucoceles exhibit predominantly aqueous contents, resulting in hypointensity on T1 and hyperintensity on T2 images, while increased protein content over time causes hyperintensity on both T1 and T2 weighted sequences.[12,13] The CT scan was performed as part of preoperative assessment for this case which showed a non-enhancing heterogenous hypodense lesion seen within the right anterior ethmoidal air cell with involvement into the right medial extraconal space which shows no clear plane of demarcation with the right medial rectus muscle and and the intraorbital segment of right optic nerve as well as the medial aspect of sclera. Due to extended waiting times, MRI was not conducted; however, Image-guided surgery (IGS) was utilized to guide the procedure with precision. IGS has been demonstrated to mitigate complications typically associated with endoscopic sinus surgery, thereby improving surgical outcomes.[10,14,15,16]

Management of paranasal sinus mucoceles primarily involves surgical intervention, with the aim of decompressing the affected sinus and restoring normal sinus drainage.[7,8,10,17] Owing to advancements over the past two decades, endoscopic surgery has become the preferred technique, offering improved precision and outcomes.[7,8,17] Endoscopic sinus surgery have been shown to have minimal complications with immediate post-operative improvement in symptoms. However, mucoceles presenting with optic neuropathy necessitate both surgical drainage and infection control to alleviate symptoms effectively.[8] In this case, the patient underwent endoscopic sinus surgery via a transnasal approach, with marsupialization of the mucocele. This minimally invasive technique is preferred due to its ability to directly address the lesion, drain the mucus, and preserve the surrounding anatomical structures, including the orbital wall. The patient experienced immediate relief from eye pain following surgery, with complete resolution of ocular symptoms at the subsequent follow-up visit. Marsupialization remains the gold standard for treating mucoceles as it allows for adequate drainage while minimizing the risk of recurrence. This has been proven as early as 1997 by Har-el et al.[18] This technique not only offers a lower risk of postoperative complications but also preserves the integrity of adjacent structures, such as the orbital wall, which is critical in cases like this where the mucocele is in close proximity to the orbit and medial rectus muscle.

CONCLUSION

This case report highlights the rarity of a bulla ethmoidalis mucocele and underscores the importance of radiologic evaluation and surgical intervention in managing this condition. Endoscopic sinus surgery with marsupialization provides a minimally invasive, effective treatment option for mucoceles, ensuring symptom relief and preserving the function of the affected structures. The successful resolution of the patient's symptoms after surgery further supports the efficacy of this approach in treating ethmoidal mucoceles.

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