

CASE REPORT

An unusual Case of Bilateral Ethmoidal Mucopyocele

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ABSTRACT

Mucocele is defined as an epithelial-lined, mucus-containing benign sac completely filling the sinus and can be seen expanding to induce pressure effects on surrounding structures. Frontoethmoidal mucocele is by far the most common due to the complexity of its drainage, and incidence of maxillary mucocele is least.

A 20-year-old female presented with a chief complaint of protruding right eyeball, redness and watering from both eyes, persistent headache, and decreased vision in both eyes since 4 months, along with complaint of diplopia and bilateral nasal obstruction with marked nonaxial proptosis, chemosis, and downward globe displacement along with telecanthus and lateral dystopia in the right eye. Orbital movements were restricted in superolateral direction. Vision was markedly decreased. Fundus examination was normal on the left side, but right eye showed hyperemic optic disk and full optic cup with dilated blood vessels. Right corneal examination showed signs of exposure keratitis.

Contrast-enhanced computed tomography (CECT) of the paranasal sinus (PNS) revealed bilateral mucocele in ethmoid sinus with hypoplastic frontal and maxillary sinus with aplastic sphenoid sinuses, which is not a common presentation. Endoscopic-wide marsupialization of bilateral mucopyocele was performed under general anesthesia.

Keywords: Aplastic frontal sinus, Aplastic sphenoid sinus, Hypoplastic maxillary sinus, Mucocele, Paranasal sinus.

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INTRODUCTION

Mucocele was discovered by Langenback in 1819, and the term was introduced by Rollet in 1896. Histological features of mucocele were described by Onodi.¹ Mucocele is defined as an epithelial-lined, mucus-containing benign

sac completely filling the sinus and can be seen expanding to induce pressure effects on surrounding structures.² The capability to expand differentiates it from a blocked sinus cavity, which simply contains mucus.³ Infected mucoceles are called mucopyocele.

Frontoethmoidal mucocele is by far the most common due to the complexity of its drainage, and incidence of maxillary mucocele is least. Though the etiology of mucocele formation is not properly understood, the main factors are posttrauma after previous sinus surgery, chronic rhinosinusitis, nasal polyposis, tumors, and obstruction of sinus ostium. The incidence of mucocele is mostly unilateral. Also in children, cystic fibrosis leads to mucocele, possibly because of mucosal stasis formation.

Ophthalmologic symptoms are the most common; patients may also report rhinological or neurological complaints. The close proximity of mucoceles to the base of the skull and orbit makes early management a necessity. Frontoethmoidal mucocele most commonly presents with downward displacement and proptosis of the orbital globe and is associated with a mass in the nasal cavity and canthal region.⁴ The main objective of this case report is to report an unusual case presentation of bilateral ethmoidal mucocele in a patient having hypoplastic frontal and maxillary sinuses.

CASE REPORT

A 20-year-old female presented to the ear, nose, and throat outpatient department with a complaint of protruding right eyeball, redness and watering from both eyes, persistent headache, and decreased vision in both eyes since 4 months, which was markedly more on the right side (Fig. 1). Patient also complained of diplopia and bilateral nasal obstruction. The swelling of right orbital cavity was insidious, gradually progressive and painless. She gave history of trauma over right eyeball, nose, and supraorbital region 1 year back. The patient also complained of episodes of fever and burning micturition. There was no history of recent cough or epistaxis.

On anterior rhinoscopy, a bulge was present on both sides of middle meatus. The right side mass was larger in size than the left. X-ray of the paranasal sinus (PNS) showed left deviated nasal septum with hypoplastic maxillary sinus. Frontal sinuses appeared aplastic (Fig. 2).

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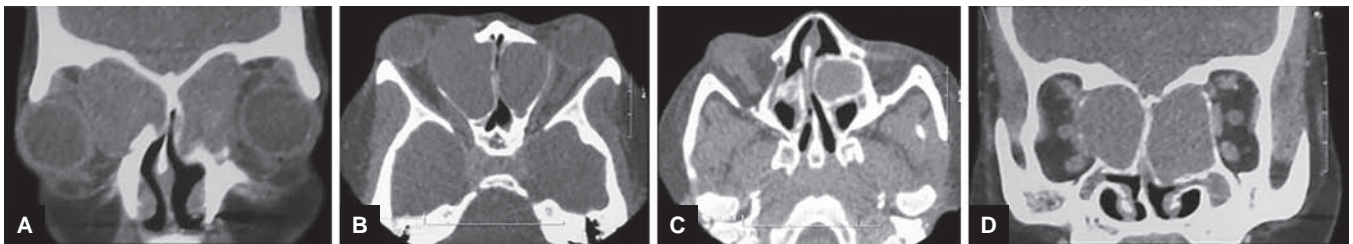
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Fig. 1: Patient



Fig. 2: X-ray paranasal sinus



Figs 3A to D: Computed tomography images

Diagnostic endoscopy of nasal cavity showed 1.5×1 cm size mass on the right side lateral to the medial turbinate and a 0.5×0.5 cm mass on the left side lateral to the medial turbinate. A needle aspiration from the left mass revealed a mucopurulent fluid with shrinkage of mass, showing that the mass was a cystic lesion rather than a soft tissue one.

Her ophthalmic examination revealed marked non-axial proptosis, chemosis, and downward globe displacement along with telecanthus (42 mm) and lateral dystopia in the right eye. Orbital movements were restricted in the right eye in superolateral direction. Pupillary reactions to light and accommodations were normal. Vision was markedly decreased, 6/24 in the right eye and 6/18 in the left eye. Her fundal examination revealed right eye hyperemic optic disk and full optic cup along with dilated blood vessels. Fundus examination of the left eye was normal. Corneal examination showed signs of exposure keratitis.

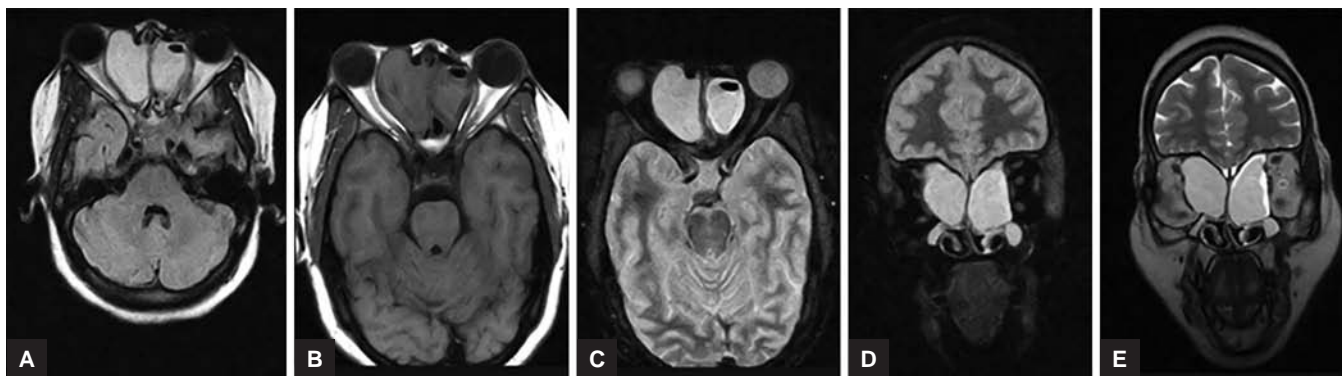
Contrast-enhanced computed tomography (CECT) of the PNS revealed expansion of bilateral ethmoid sinuses with isodense soft tissue filling the sinus cavities and resorption of ethmoid sinus septae – probably arising from middle ethmoid air cells. Air pocket was noted in the left ethmoid sinus along with lateral bulging of sinuses into the orbit (R>L) and nasal cavities (L>R). Erosion and remodeling of the medial walls of the orbits (R>L) were seen without infiltration of the medial rectus muscles. No intraconal extension was present. Extraconal

and preseptal extension into the right orbit along anterior aspect of medial wall of orbit was present (Figs 3A to D).

Obliteration of superior meatus and middle meatus with superior turbinates was not visualized, and remodeling of middle turbinates was seen. Nasal septum was not eroded. Extension of the soft tissue into bilateral frontoethmoid recesses, maxillary ostia, and infundibula was present. Hypoplastic frontal and maxillary sinuses and aplastic sphenoid sinus were seen. No bone thickening or hyperostosis or destruction was present, and only erosion/remodeling was seen. Maxillary and sphenoidal sinuses were also very small and had mucosal thickening. Ostiomeatal unit was obliterated.

Magnetic resonance imaging (MRI) revealed fluid-attenuated inversion recovery and T2 isohyperintense and T1 hypointense soft tissue filling the ethmoid, frontal, and maxillary sinuses, bulging into extraconal space of both orbits with lateral deviation of medial rectus muscles and right optic nerve. Left ethmoid sinus shows hypointense air pockets and T2 hyperintense rim suggestive of fluid inside the sinus (Figs 4A to E).

Fine-needle aspiration cytology from swelling showed few scattered inflammatory cells and macrophages along with mucoid material and abundant pus cells suggesting mucopyocele. Pus sent for culture and sensitivity from left cyst for bacterial and tubercular culture and sensitivity showed no growth on incubation (Fig. 5). Patient was planned for surgery and endoscopic-wide



Figs 4A to E: Magnetic resonance images

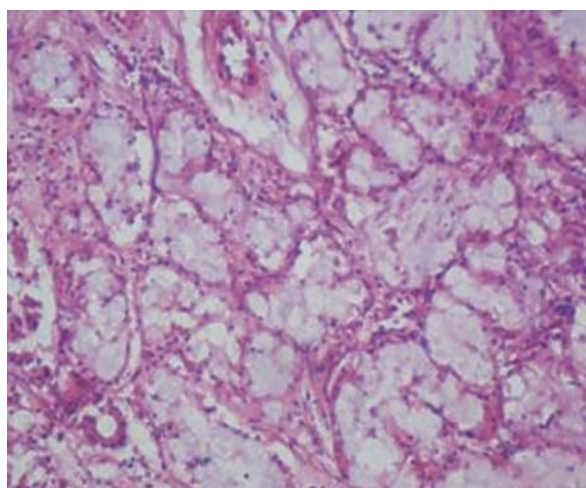


Fig. 5: Histopathology

marsupialization of bilateral mucopyocele was performed under general anesthesia.

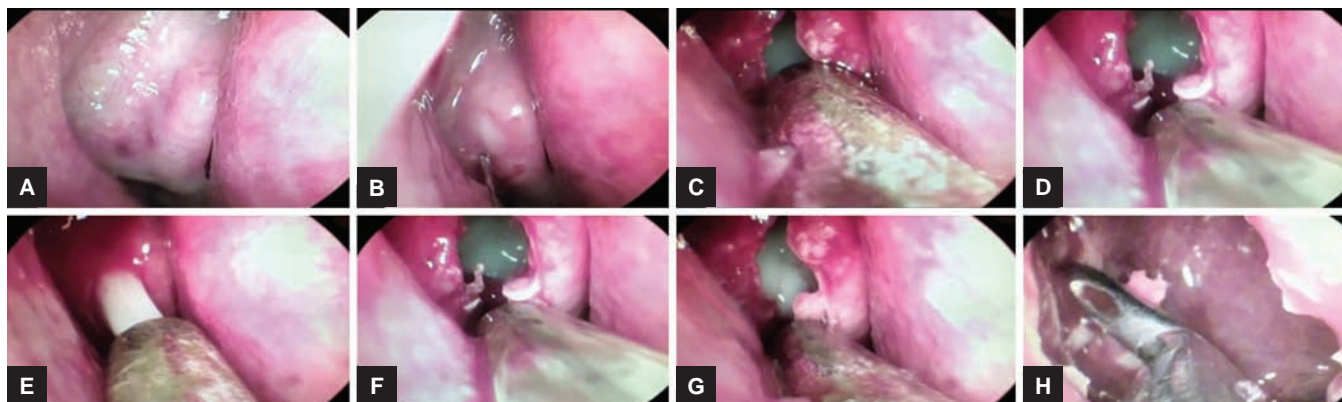
Intraoperatively, we found that the cysts extended up to the upper nasal cavity on both sides. The anteroinferior and mediolateral walls of the cysts were removed endoscopically after giving an incision and aspirating mucopus from both the sides. Wide marsupialization was done (Figs 6A to H). Bilateral ballotment of eyeballs was seen. Nasal cavity was packed after irrigation of mucopyocele.

The postoperative period was uneventful, and there was marked relief from proptosis of right eye. On the 3rd postoperative day, the patient showed marked visual improvement, with visual acuity of 6/18 in the right eye and 6/9 in the left eye with rapid resolution of proptosis and improved eye movements. Fundus examination showed marked reduction in optic nerve pressure. On the 5th postoperative day, the patient had striking reduction in telecanthus and lateral dystocia from preoperative condition (Figs 7A to D).

Histopathology of tissue was consistent with the diagnosis of mucopyocele. Patient was called for nasal endoscopy 2 weeks later, which showed widely open sinus cavity and healthy mucosa inside cyst.

DISCUSSION

Mucocele is a benign, mucus-filled, epithelial-lined sac occurring in PNS. These expand gradually and present with pressure symptoms varying from nasal obstruction to ophthalmological symptoms. Sinus ostium stenosis is more commonly seen in the frontal sinus⁵ where 2 mm narrow duct passes through the ethmoid bone for a distance of 5 to 10 mm very close to the ethmoid sinuses. Frontal sinus is most commonly involved, usually in association with anterior ethmoid (64%), followed by maxillary sinus, sphenoid, and posterior ethmoid sinuses.



Figs 6A to H: Intraoperative pictures



Figs 7A to D: Postoperative day 5

According to the pathogenic mechanism, primary mucocele develops as a retention cyst of sinus epithelium mucus glands and secondary mucoceles are formed by obstruction of sinus ostium, from masses which extend inside the sinus. Histological examination reveals bone resorption and remodeling. It may be a sequela to stimulation of lymphocytes and monocytes, which further promotes cytokine production from the fibroblasts of sinus lining. It results in expansion of mucocele.

Clinical presentation of the mucoceles varies from asymptomatic to severe headache and disturbance in vision. Proptosis and diplopia are the most common complaints. On physical examination, periorbital tenderness, chemosis and edema, diplopia, and restricted extraocular movements can be seen.

A case with bilateral ethmoidal mucocele, with aplastic sphenoid sinuses and hypoplastic maxillary and frontal sinuses, has not been reported in the medical literature till date to the best of our knowledge, thus making this a rare case presentation.

Differential diagnosis includes orbital abscess, dacryocystitis, pneumosinus dilatans, frontal sinus tumors like osteomas, meningocoele, exophthalmos, orbital tumors, etc.⁶ Surgical drainage and marsupialization is the treatment of choice. Its goal is to drain the mucocele and ventilate the sinus and eradicate the mucocele so that recurrence is not present. Patients with frontal sinus mucocele are also treated with an external surgical approach. Endoscopic marsupialization is the treatment of choice for ethmoidofrontal, maxillary, sphenoidal, and ethmoidal mucoceles.

CONCLUSION

A rare presentation of bilateral ethmoid mucocele should be kept in mind if a patient presents with similar complaints. Endoscopic marsupialization is the treatment of choice in such cases, and timely intervention prevents loss of visual acuity and cosmetic morbidity.

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