

Idiopathic Nasal Septal Mucocele: A Case Report

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Article Info

Article Note:

Received: January 2024

Accepted: February 2024

Publish Online: March 2024

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Keywords:

Nasal Sepum;

Idiopathic;

Mucocele.

Abstract

Background: Septal mucocele is very uncommon. Those reported mostly have a prior history of trauma however; idiopathic septal mucocele is even rarer.

Case presentation: We report a case of idiopathic septal mucocele who presented with bilateral, progressive nasal obstruction. Clinical examination revealed bilateral, smooth bulge on the anterior cartilaginous septum, which on radiology indicated an expansile cystic mass. As it extended to the nasal spine superiorly, an open rhinoplasty approach was taken for its complete removal. The septal cartilage was absent inside the cyst.

Discussion: Histology showed tissue lined by respiratory epithelium with mucinous glands in the stroma is confirmative of mucocele. Two months post-surgery, the septal perforation had healed well with no evidence of recurrence.

Conclusion: Although rare, septal mucocele should also be considered as differential diagnosis of long-standing septal swelling.

Conflict of Interest: The authors declare no conflict of interest.

Please cite this article as: Gurung U, Bathwal R. Idiopathic Nasal Septal Mucocele: A Case Report. J Otorhinolaryngol Facial Plast Surg. 2024;10(1):1-5. <https://doi.org/10.22037/orlfps.v10i1.44880>

Introduction

Mucocele is a benign, mucus containing, expansile cystic lesion that can involve the paranasal sinuses, most often affecting the frontal (60%–65%), followed by ethmoid sinuses (20%–30%), and maxillary (10%), and sphenoid sinus (2%–3%) being less commonly affected (1, 2). Septal mucocele is a rare occurrence. Till date only 16 cases have been reported (3). Most of the cases have a prior history of trauma however; idiopathic septal mucocele is more rare. Here, we present a case of idiopathic septal mucocele and review the available literature to describe its etiology, clinical presentation, diagnosis and management.

Case Presentation

A 29-year-old male presented with three months of gradually progressive, painless bilateral nasal obstruction without any preceding trauma. This was not associated with nasal discharge or pain. Nasal vestibule examination revealed a smooth bulge

bilaterally on the anterior nasal septum completely occluding the nasal cavity. Its posterior limit confirmed on nasal endoscopy was till the anterior end of the middle turbinate. The mid nasal dorsum was widened. CT scan showed a well-defined craniocaudally, elongated 36x25x23 mm³ cystic expansile lesion in the anterosuperior aspect of nasal septum. There was bony remodeling in the perpendicular plate of ethmoid without involvement of the paranasal sinus (Figure 1). This lesion showed iso- to intermediate in T1 and high signal intensity in T2 in the cartilaginous septum without intracranial extension (Figure 2).

Fine needle aspiration was suggestive of mucocele. The patient underwent excision via open rhinoplasty and endoscopic approach with L-strut reconstruction of nasal septum under general anaesthesia (Figure 3). A 3 X 2.5 cm cyst containing mucoid material 2 cm posterior to the caudal end of cartilaginous septum reaching upto the nasal spine

<https://doi.org/10.22037/orlfps.v10i1.44880>

superiorly was completely excised (Figure 4). There was no obvious surgical plane between the septal mucosa and the mucocoele wall. Septal cartilage was absent within the cyst and the adjacent perpendicular plate of ethmoid posteriorly was found to be splayed. The remnant lower septal cartilage was used to make dorsal strut creating a L-strut, which was sutured to the upper lateral cartilage. Biopsy

showed tissue lined by respiratory epithelium with mucinous glands in the stroma confirmative of mucocoele (Figure 5). The septal perforation had healed well and the patient did not have saddle nose deformity till two months post-operatively. The patient then was lost for follow up as he had migrated abroad for work.



Figure 1. CT scan showing cystic expansile lesion in the anterosuperior aspect of nasal septum

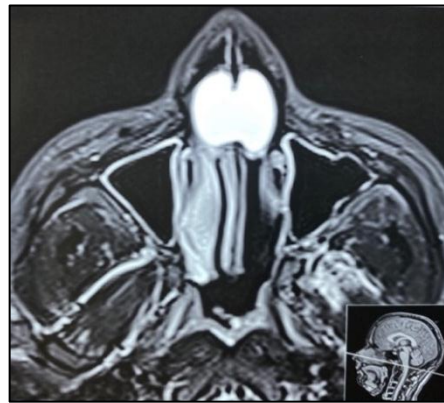


Figure 2. MRI showing Iso- to intermediate in T1 and high signal intensity in T2 in the cartilaginous septum without intracranial extension



Figure 3. Removal of the septal mucocoele by open rhinoplasty approach



Figure 4. Mucocoele along with the splayed perpendicular plate of ethmoid attached posteriorly.

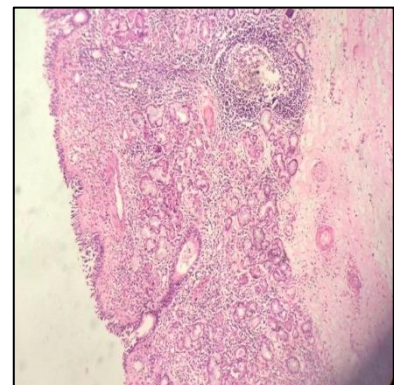


Figure 5. Biopsy showing tissue lined by respiratory epithelium with mucinous glands in the stroma

Discussion

Mucocoeles in the paranasal sinus develop due to mucosal entrapment or sinus outflow obstruction. With the increasing mucocoele content, they can induce bony remodeling and cause bony erosion by virtue of osteolysis (4). Mucocoeles can remain asymptomatic until they expand and cause pressure symptoms on the adjacent structures (5).

Mucocoele of the nasal septum most often presents with insidious onset bilateral nasal obstruction and concurrent septal swelling usually affecting males in the fourth decade of life (2,4 - 12). On nasal endoscopy, it appears as submucosal mass in the nasal septum (2). Compared to other paranasal mucocoeles, septal mucocoeles present earlier as nasal obstruction is the main presentation (7). In a long standing

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case, there may be changes in the facial contour such as widening or mass on the nasal dorsum or on the medial canthus due to pressure effect namely on the nasal bone (6,13). Saddle nose deformity in the background of septal mucocele has also been reported probably due to instability of the nasal dorsum consequent to this locally destructive process(3). Other symptoms include visual changes, pain, recurrent infections secondary to extension to adjacent structures (4).

The etiology of septal mucocele is unknown. One plausible cause could be obstruction of pneumatized perpendicular plate of ethmoid which in itself is uncommon (5). Lei *et al.* observed pneumatization of perpendicular plate of ethmoid anteriorly starting from the frontal spinal junction at the anterior part of the perpendicular plate in close relation to the frontal sinus origin. Similarly, posteriorly, the pneumatization was found to originate from the sphenoidal spinal junction at the posterior part of the perpendicular plate (6). Fontes *et al.* attributed posterior pneumatization as a possible cause for the more posteriorly located septal mucocele (7). Similarly, in our case, possibility of anterior pneumatization pattern probably explains the anterior location of the septal mucocele alongside splaying of the anterior end of the perpendicular plate of ethmoid. The obstruction of the pneumatized septum could be triggered by trauma or inflammation (7).

The other explanation for its etiology is probable entrapment of nasal mucosa in the septum following any form of trauma, be it surgical or maxillofacial trauma (5, 7, 8). This has been contradicted by Yilmaz *et al.* who reported a case in a child with no predisposing factors (14). In our case also, there was no predisposing risk factor not even a septal deviation hence it was considered idiopathic. CT and MRI gives information about the size, location and relation to adjacent structures (2). Mucoceles appear as well defined, fluid-filled,

rounded lesion with bone remodeling on CT scans (5). This may be seen to involve the nasal root and perpendicular plate of ethmoid in case of septal mucocele (6). Magnetic resonance imaging helps rule out intracranial extension and differentiate from pathologies like meningocele, hemangioma, rhabdomyosarcoma, neuroblastoma (5). On MRI, mucoceles appear hypointense. With increase in protein concentration, which occurs in longstanding mucocele, it can appear hyperintense in both T1 and T2-weighted images. If infected, it may show rim enhancement. (2). CT in our case showed a well-defined cystic lesion with bone remodeling in the perpendicular plate of ethmoid. On MRI, it had iso- to intermediate in T1 and high signal intensity in T2 with no intracranial communication.

Surgical excision is the treatment of choice and is necessary to prevent intracranial or orbital complications due to its expansile nature. (2, 4) It could be either complete removal or marsupialization of the cyst via external or endoscopic approach depending on the size and location (4,5). Smaller lesion can be completely excised whereas larger lesions can be marsupialized if total removal is not possible (4). Endoscopic marsupialization is gaining popularity as it allows easy access to the lesion under good vision with consequent less complications and low recurrence rate (2, 9). However, there are variations to the surgical treatment reported in the literature. Septal mucopyocele has been excised completely endoscopically with resultant single wide cavity post-operatively (9). Endoscopic removal of the cyst along with adjacent dystrophic bone preserving the septal flaps has also been reported. Marsupialization in this case was deferred due to the bony remodeling (5).

Post-complications include bleeding, hematoma, mucosal tear, anosmia, septal perforation, vascular and skullbase injury with the risk being significantly low for endoscopic

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approach (5). In our case, a combined external rhinoplasty and endoscopic approach was taken for complete excision of the mucocele as there was evidence of bony remodeling but without skullbase erosion. The dorsal septal cartilage was deficient so due to the likeliness of saddle nose deformity in future as reported by Chou *et al.* (3), and marsupialization not adequately addressing the issue, the mucocele was excised in toto and the nasal dorsum reconstructed with the remnant septal cartilage. Except for the anticipated septal perforation, there were no other complications in our case.

On histopathology, the cysts contain mucoid fluid and are lined by pseudostratified ciliated columnar or cuboidal epithelium with inflammatory cell infiltrates. There may at times also be woven and lamellar bone (1,5). These classical findings tallied with the histopathology findings in our case. Septal mucocele needs to be differentiated from other midline nasal masses like nasal dermoid cyst, epidermoid cyst, nasal gliomas, encephaloceles, neuroblastoma, hemangioma, lymphangiomas, septal abscesses (6). These were ruled out on the basis of prior fine needle aspiration and radiological evaluation.

Conclusion

Nasal septal mucocele is rare condition with preceding trauma as the likely etiology. CT and MRI help in diagnosis, location, size and extent and to rule out extra nasal extension especially the skull base. Surgery is the treatment of choice with endoscopic marsupialization being the preferred modality with good outcome.

Acknowledgements

Not declared.

Patient Consent

Informed consent was taken from the patient for publication of this case report.

Conflict of Interest

The authors declared no conflicts of interest.

Financial Support

The authors declared that there was no financial support.

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