

Extensive primary maxillary mucocele treated by combined external and endoscopic approaches

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SUMMARY

A mucocele is an epithelial lined mucus-containing sac completely filling a paranasal sinus cavity and capable of expansion. The common presentation of maxillary mucocele is nasal block, followed by cheek pain or pressure, nasal drainage and proptosis. Mucocele should be differentiated from a retention cyst by its histological appearance and imaging. Computed tomography scan and MRI are essential to confirm the diagnosis of mucocele, determine the location of the lesion, and evaluate possible involvement of the orbit or skull base. The management of mucoceles is by surgical removal of all their mucosal linings. Mucocele formation is usually due to an identifiable cause resulting in blockage of the common paranasal sinuses drainage. Primary mucocele in the maxilla is infrequently encountered. We presented a case of an extensive unilateral maxillary mucocele in a 36 years old lady without any predisposing factors, treated by combined endoscopic and Caldwell-Luc approaches. It is important for all clinicians to be aware that mucoceles may occur without any predisposing factors to avoid misdiagnosis.

INTRODUCTION

Maxillary mucocele may arise as a result of chronic nasal infection, previous trauma, allergy, sinonasal disease or prior sinus surgery.¹ But 64 % of the cause remains unknown.¹ Most reported origin of primary mucocele was the ethmoidal sinus, while secondary mucocele mostly from the maxillary sinus.² General occurrence of paranasal mucocele is commonest in frontal, followed by ethmoid, maxillary and sphenoid sinus.³ The incidence is reported commonly in 40 to 60 year old without any gender predilection.^{3,4} The frontonasal duct which is smaller as well as developmentally more variable, tends to be obstructed most as compared to the wider and consistent natural ostia of the maxilla.³ Primary maxillary sinus mucocele is a rare condition and unlike the normal development of mucocele which is due to obstruction of sinus ostia, has no predisposing factors. The common presentation of maxillary mucocele irrespective of type is nasal blockage, followed by cheek pain or pressure, nasal discharge and proptosis.^{1,2} Any patients having symptoms and signs suggestive of sinonasal conditions such as sinusitis should be referred to tertiary care for further expert evaluation which

can include nasoendoscopic examination and sinus computed tomography scan. Sinus computed tomography scan is especially indicated when a tumour is suspected or in patients that don't respond to conservative management.

CASE REPORT

A 36 years old lady with no underlying medical illness, presented with presented with left sided blood-stained nasal discharge and nasal blockage. There was no history of trauma and no symptoms of upper respiratory tract infection. She has no history of allergic rhinitis symptoms such as frequent sneezing or rhinorrhea. There was no facial pressure or anosmia. She did not experience any epiphora, double vision or orbital pain. There was no surgical history to the nose or paranasal sinuses. She was not on any medications. The nasoendoscopic examination revealed engorged inferior turbinate of left nasal cavity. The septum showed no deviation or spur. There was no bleeding point seen at the postnasal region, septum or turbinal area both anteriorly and posteriorly. Bilateral otoscopy was normal. The oral cavity and oropharynx were normal, and the neck examination was unremarkable. The blood-stained nasal discharge most probably originated from the dry nasal mucosa of the left side of nose due to narrow nasal space from the enlarged left inferior turbinate. She was started with intranasal steroid spray, oral antihistamine and alkaline nasal douching. She was seen again for a few times after the first consultation, with similar symptoms and examination findings. About 5 months after the initial visit, the patient complaint of left nasal blockage. She denied left facial pain, left epiphora or left protrusion of eye. There was no palatal or alveolar mass and she denied any recent loosening of tooth over the left upper alveolus. The examination showed narrowing of the left ostiomeatal complex with left turbinate engorgement abutting the septum. Otherwise the eyes and intra-oral examination were unremarkable. Computed tomography of paranasal sinuses (CTPNS) showed an expanded left maxillary sinus with isodense mass occupying the entire left maxillary sinus, measuring 5.0 x 3.7 x 3.8 cm in dimension, with small peripheral calcification (Figure 1A). The mass caused thinning and medial bowing of the medial wall of the left maxillary sinus, compressing the inferior and middle turbinates. Concurrently, the intramaxillary lesion

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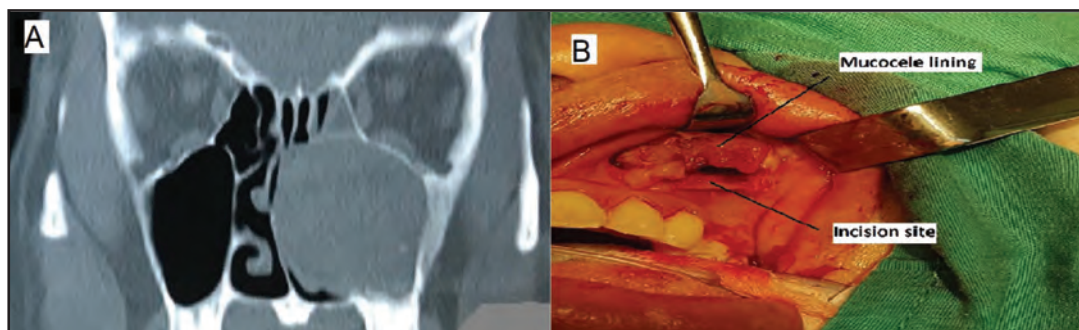


Fig. 1: The coronal view of sinus computed tomography scan showing heterogenous soft tissue density occupying the left maxillary sinus and extending into the left nasal cavity, with erosion of the medial bony wall of the maxillary sinus (A). The sublabial incision for left Caldwell Luc approach, superior to the canine fossa and extended laterally, showing part of the mucocele lining for complete removal (B)

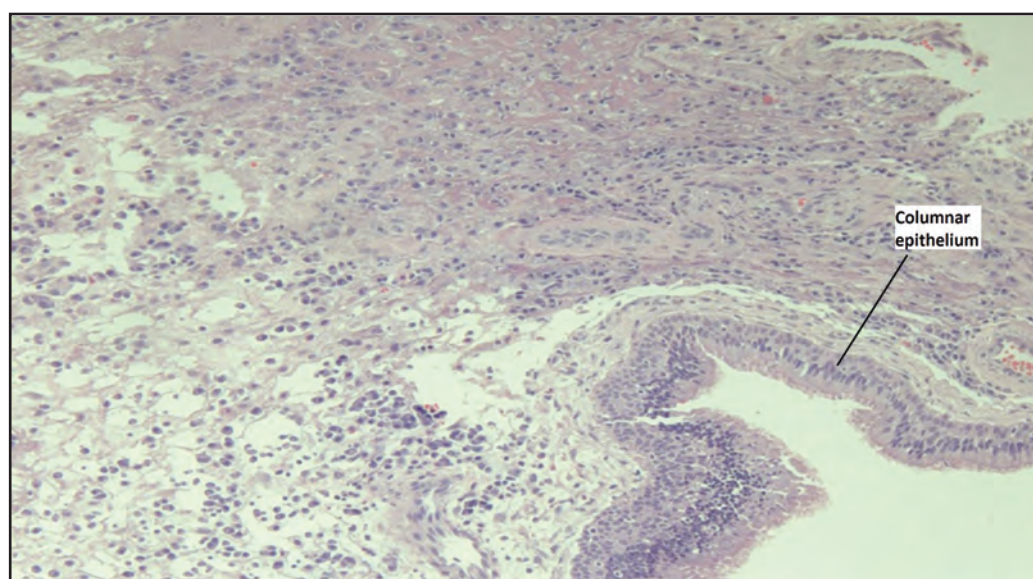


Fig. 2: Histopathological examination with hematoxylin and eosin staining (x 4 magnification) showing wall formed by hyaline connective tissue, with cystic areas lined by columnar epithelium

expanded through the medial wall to narrow the left nasal cavity due to the mass effect. The left ethmoidal sinuses showed mucosal thickening, while the frontal sinuses and sphenothmoidal recesses were patent. There was no left intraorbital, intracranial, nasopharyngeal or subcutaneous extension of the mass. She underwent left nasal middle meatal antrostomy and left Caldwell-Luc approach under general anaesthesia (Figure 1B). Intraoperatively, noted that both the left middle and inferior turbinate were medialized due to the left maxillary mass effect. On identification of the left maxillary ostium, it was noted that the antrum was widened but not draining the maxillary content as it was obliterated by the intramaxillary mass. The medial wall was thinned out and appeared soft. The left maxillary sinus ostium was further widened via middle meatal antrostomy, and the left maxillary sinus mass was visualized. It appeared as cystic looking, glistening, whitish greyish in colour, and fully occupying the whole maxillary sinus. After approaching the medial wall of maxillary sinus via middle meatal antrostomy, the mucocele ruptured and clear mucoicid content seen. The content was decompressed, the mucocele lining was

delivered in piece meal using forceps and angled microdebrider. The surgery was completed using Caldwell-Luc approach to access the anterior and inferior maxillary walls. Bilateral noses were packed with merocel and removed after 48 hours. The patient was discharged well after nasal packing removal and appointment given a week after the surgery for nasal toileting. She was instructed to do regular nasal douching with saline to remove the nasal crusting and blood clot. On follow-up at the outpatient clinic, she remained asymptomatic without any post-operative complications including synechiae seen. The nasoendoscopic examination showed left maxillary sinus was clear from the disease, and the left inferior and middle turbinate were normal. The histopathological examination showed the mass was formed by hyaline connective tissue, with cystic areas lined by columnar epithelium consistent with a diagnosis of mucocele (Figure 2). On follow at 24 months post operatively, she was well and there was no medialization of the left lateral nasal wall. The maxillary sinus remained clear.

DISCUSSION

The differential diagnosis of paranasal sinuses benign cystic mass presenting in the nasal cavity includes retention cyst, mucocele, chronic rhinosinusitis with nasal polyps, mycetoma, maxillary mucosal cyst and odontogenic cyst.^{5,6} Occasionally, epidermoid cyst may also arise from the maxillary sinus.⁷ Mucocele should be differentiated from a retention cyst by its histological appearance and CT imaging.¹ Mucoceles grow under the periosteum, while retention cysts grow under the mucosa of the sinus. Thus, retention cysts are non-expanding, well circumscribed, mucosa covered masses, whereas mucoceles exhibit an osteolytic capacity with a tendency to expand along the least resistance route.

Plain paranasal sinuses X-ray does not play much role in diagnosing maxillary mucocele, unless for frontal sinus mucocele which readily can be seen.⁸ CT scan and, in selected cases, magnetic resonance imaging (MRI) are essential to support the clinical suspicion of mucocele, determine the location of the lesion, and evaluate possible involvement of the orbit or skull base.⁴ CT in the axial and direct coronal planes is the optimum modality in visualization of the bony expansion which occurs in mucocele formation. The CT scan appearance of a maxillary mucocele is an expansile mass with homogeneous substance that has an attenuation of 10 to 18 Hounsfield unit (HU). In contrast, long standing chronic mucoceles may have a larger protein content and the attenuation is higher (20–40 HU). On contrast study, there will be enhancement of the lining membrane of the mucocele. However, contrast enhancement is best reserved for MRI especially when secondary mucocele is suspected owing to sinonasal tumor.⁸

The management of mucoceles is by surgical removal of all their mucosal linings.⁹ Endoscopic approach is advocated for the removal of maxillary mucocele.¹⁰ Greatest advantages of this minimally invasive approach are reduced intraoperative pain, reduce postoperative cheek swelling and numbness and therefore shortened the hospital stay. However, an extensive maxillary mucocele may require a combined external and endoscopic approaches to ensure complete removal and prevent recurrence. Even though Caldwell Luc surgery is almost historical in the era of endoscopic sinus surgery, there are few conditions where the Caldwell Luc approach is useful as an access to the maxillary sinus; when the mucocele is located too far lateral in the maxillary sinus, severe thickening of medial bony wall of the maxillary sinus and when there is compartmentalization of the mucocele.^{2,10} Another indication for an external approach is when the mucocele is located at the anterior wall of the maxillary sinus or extended into the pterygomaxillary fossa.¹⁰ Planning of approach to maxillary mucocele prior to surgery is important in regards with the extension of the mucocele, surgeon

familiarity of the technique and facilities available. In our patient, we successfully removed the maxillary mucocele by combining both endoscopic and Caldwell Luc approach. In recurrent cases, endoscopic removal can be attempted by employing either a wide middle meatal antrostomy, if that has not been done, or a modified prelacrimar recess approach to ensure complete removal. Though rarely done, external approaches with marsupialization have been described.¹

CONCLUSIONS

It is important for all clinicians to be cognizant that mucoceles may occur without any predisposing factors to avoid misdiagnosis. Primary maxillary mucocele is a rare entity, where the exact aetiology is still unknown. Symptoms are mostly of mass effect in the maxillary sinus, together with symptoms related to its surrounding structures. Endoscopic approach is advocated but a combined approach of endoscopic and external are required for extensive lesion. As maxillary mucoceles can recur and in cases where histopathological examination has not excluded malignancy, regular tumour surveillance on follow-up is essential for early detection.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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