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Isolated Haller Cell Mucocele: An Unusual Presentation

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Abstract

This article presents a rare and interesting case of isolated mucocoele of haller cell (Infraorbital ethmoid cell) presenting with extensive proptosis and visual disturbances in the absence of facial pain and symptoms of sinusitis. CT PNS with contrast revealed a heterogeneous non enhancing soft tissue lesion in the roof of the left maxillary sinus/floor of left orbit with severe thinning of the floor of the left orbit and not filling the maxillary sinus. This is the fourth reported case of isolated mucocoele of haller cell and the first case to present with significant proptosis and visual disturbances. Treatment opted was endonasal endoscopic surgical removal and restoring the appropriate orbital function and cosmesis. Early diagnosis and prompt surgical management can help avoid dangerous visual complications in these kind of rare presentations.

Keywords: Maxillary sinus mucocele; Proptosis; Haller cell mucocele; Infraorbital ethmoidal cell; Haller cell.

INTRODUCTION

Mucoceles are benign, locally expansile cyst-like lesions, lined by the mucoperiosteum of the involved paranasal sinus. More common in the fronto-ethmoidal region, they rarely involve the maxillary sinus. Antralmucoceles generally involve the lateral sinus wall first[2] andthe most common presentation is facial swelling.[3]

Migrating anterior or posterior ethmoidal air cells that pneumatize the roof of the maxillary sinus is termed as Haller cell with an incidence of 2–45% worldwide. This cell can rarely present with mucocele and mimics antralmucocele. Expansion of mucocele arising from posteriorly located haller cells when invading orbit can cause ophthalmological symptoms such as orbital edema, proptosis, diplopia, ptosis, visual or oculomotor disturbances, and pain in the eye.[8] Haller cells also have a strong association with dehiscence of the orbital floor (upto 60%).[6]

Our case was interesting as our patient neither had facial swelling nor signs of sinusitis and CT scan showed extension of lesion from roof of maxillary sinus towards orbit. And intraoperatively, a well defined calcified rim was identified surrounding the mucocele and it was found that superior wall of mucocele was extending to dehiscent orbital floor. These point towards gradually progressing mucocele arising from roof of maxillary cavity and extending to dehiscent orbital floor causing proptosis and diplopia which is quiet a rare presentation for a mucocele.

CASE

A 43 year old male presented with left orbital proptosis noticed by his collegues 2 months back and progressive left diplopia. An ophthalmology evaluation showed ill sustained pupillary reactions on both eyes (left > right) and diplopia in left

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inferior gaze. A screening MRI orbit showed a lesion in the left maxillary sinus and the patient was referred to otorhinolaryngology department for further evaluation and management. There was no past history of sinus trauma, infection or surgery. He was a poorly controlled diabetic and on oral hypoglycemic drugs and insulin. The patient presented to ENT OPD with evident proptosis but clinical examination and a nasal endoscopy showed no other nasal abnormalities or facial swelling.

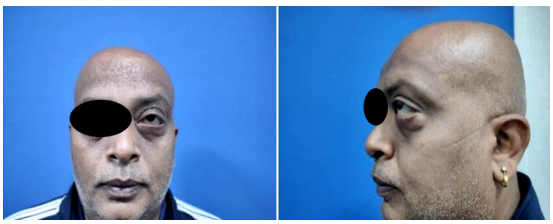


Fig. 1: Preoperative photograph of patient

A CT PNS with contrast revealed a heterogeneous,non enhancing soft tissue lesion measuring 3.1 x 3 x 2.4 cms with its epicenter in the floor of left orbit. There was severe thinning of the floor of the left orbit with displacement of the inferior rectus and proptosis. Closer attention showed ovoid lesion with a calcified rim in the upper 2/3rd of the maxillary sinus. A separate soft tissue density filled the lower part of the sinus (? Secretions). Rest of the paranasal sinuses appeared normal. In comparison to the previous MRI orbit done 2 months ago (lesion size 2.5*2.7*2.5 cm) present imaging showed gradual growth of the lesion. Radiologically a differential diagnosis of ameloblastoma /aneurysmal cyst was given.

After achieving euglycemic status we planned an endoscopic surgical management under general anesthesia. After decongesting the nose, a Diagnostic nasal endoscopy showed DNS to left, Bilateral hypertrophic inferior turbinates and a slight bulge in the medial wall of maxillary sinus. Left uncinectomy was performed. Mucocele identified and decompressed, the contents of cystic lesion was mucoid discharge. A well defined inferior calcified rim was identified and removed. Retained secretions of the maxillary sinus below the mucocele was drained out. Superior layer of mucocele wall was seperated from the dehiscent orbital floor. On table proptosis appeared to decrease and there was no prolapse of orbital contents into maxillary sinus. Anterior ethmoidectomy was done and medial wall of orbit appeared normal. As orbital floor appeared stable, no reconstruction surgery was required. Postoperative support was provided to the orbital floor using an inflated foleys catheter balloon and stabilized with a nasal pack. After pack removal 36 hours later left orbit retained its normal position, eye movements were normal and diplopia improved.

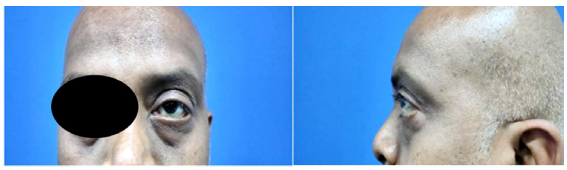
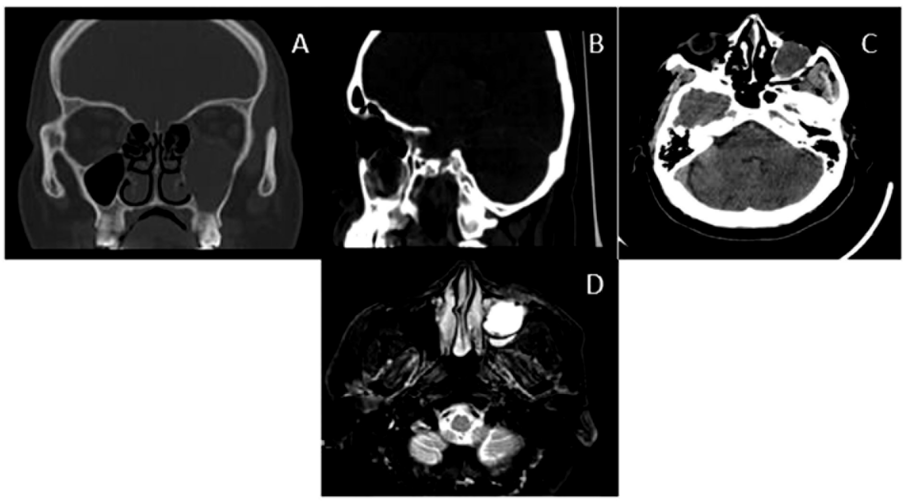


Fig. 2: Postoperative photograph of patient



A. Coronal CT PNS:Non enhancing soft tissue lesion with epicenter in the floor of left orbit with severe thinning of orbital floor
B. Sagittal CT PNS : Ovoid lesion with a calcified rim in the upper 2/3rd of the maxillary sinus
C. Axial CT PNS: Non enhancing soft tissue lesion extending into left orbit.
D. MRI Nose/Orbit: soft tissue lesion in left maxillary sinus

Histopathological examination was consistent with diagnosis of mucocele. Patient was followed up after 5 days and nasal endoscopy showed widely opened maxillary sinus , no remnant tissues noted. Within one week his left visual acuity and colour vision returned to normal with a visual aquity of 6/6 bilaterally and 17/17 bilaterally with the ishihara colour test plates. He also had complete resolution of diplopia and proptosis. 2 months postoperative period vision, extraocular movements and position of eyeball remains normal.

DISCUSSION

Maxillary sinus mucocoeles are uncommon but well defined entities. They may be confused with a rare entity of a Haller cell mucocoele. To the best of our knowledge, this is the fourth reported case of isolated mucocoeles of a haller cell and the first presenting with extensive proptosis and visual disturbances in the absence of facial pain and other symptoms of sinusitis.

Although uncommon, maxillary sinus mucoceles have distinct radiological findings. It usually fills the whole sinus and is nearly always associated with thinning of the anterolateral wall of the sinus, frequently with bulging of the medial wall and septum. There may be some diagnostic challenges, especially when extensive disease or anatomical variants are present.

Haller cell, a normal anatomical variant, when enlarged or infected can significantly constrict the posterior aspect of the ethmoidal infundibulum and maxillary ostium causing maxillary sinusitis & also predispose to an antralmucocoele. Inflammation of the Haller cell is common in pansinusitis, but an isolated mucocele of this cell is rare.They are usually located in the roof of the maxillary sinus. It shows a thin bony septum between the lesion and normal maxillary sinus. It can expand slowly erode the roof of maxillary sinus and extend into orbit.[5]

Diagnosis of Hallers cell is typically made by CT scan, as they cannot be identified by diagnostic nasal endoscopy because of their typical location lateral to the infundibulum. Easily seen on coronal PNS CT, they have been described as well defined, round, oval, or teardrop shaped, unilocular or multilocular radiolucencies with smooth borders that may or may not appear corticated, located medial to the infraorbital foramen.[8]

Haller cells also have a strong association with dehiscence of the orbital floor (upto 60%)[6], making the orbit more vulnerable. Expansion of a mucocele arising from such a Haller cell, when invading the orbit, can cause ophthalmological symptoms of proptosis, diplopia, ptosis, visual or occulomotor disturbances and pain in the eye. The case in point highlights both such findings.

Endonasal endoscopic approach is the preferred treatment in paranasal sinus mucoceles with low recurrence rate at or close to 0% and minimally invasive with a shorter postoperative recovery and less morbidity. Caldwell Luc approach is reserved for more extensive mucocoeles involving facial soft tissues and pterygomaxillary fossa.[7]

CONCLUSION

Though benign and slow growing in nature, early diagnosis and surgical management of Haller cell mucocele can help avoid dangerous visual complications. A prompt surgical intervention and proper ophthalmic follow up may negate the requirement of extensive reconstructive surgery even with advanced proptosis and orbital floor destruction. A high index of suspicion is needed to avoid missing such rare presentations.

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