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CASE STUDY

MAXILLARY SINUS MUCOCELE- AN UNUSUAL CLINICAL PRESENTATION

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ABSTRACT

Mucocele of paranasal sinuses is a benign, expansile cyst-like lesion filled with mucus and lined by the mucoperiosteum of the involved sinus. Mucoceles develop when the drainage of normal sinus secretion is obstructed, followed by mucous accumulation and expansion. Among the paranasal sinuses, the frontal sinus is most commonly involved. Mucoceles of the maxillary sinus usually present as an incidental finding, radiographically appearing as opacification of the involved sinus. Here, we present a case of mucocele of the right pmaxillary sinus with an unusual clinical presentation as an asymptomatic swelling of the palate and maxillary alveolus on the right side.

INTRODUCTION

A mucocele is an epithelium lined sac which is filled with mucous. (**Thiagarajan and Arjunan, 2012**) Paranasal sinus mucoceles are benign and expansible cyst-like lesions filled with mucus and lined by the mucoperiosteum of the involved sinus. The most significant local complication of sinusitis is the obstruction of the sinus ostium. Mucoceles develop due to this obstruction as the sinus mucosa continues to secrete mucus in the mucocele, which expands gradually, eventually resulting in thinning and destruction of the bony wall. (**Chang and Kang, 2010**) Frontal sinus is the most commonly involved sinus (89%), followed by the ethmoid (8%), sphenoid (2%) and maxillary (1%) sinuses. (**Chang and Kang, 2010**) As an antral lesion, a mucocele is the only cystic lesion that may cause expansion or erosion of the maxilla (**Han *et al.*, 1995**).

Case Report

A female patient aged 42 years reported to the Department of Oral Medicine and Radiology with the chief complaint of pain in her right upper jaw for the past 15 days. Patient's history revealed extractions on upper right back tooth region 12 years back due to dental caries. There was no past history of pain in the region. Pain developed 15 days ago which was gradual in onset and progression, mild, dull and continuous in nature which aggravated on bending the head down and there were no relieving factors. There was difficulty in breathing through the right nostril. About 5 days later, a swelling developed gradually on the right side of upper jaw and grew rapidly to the present size. Patient's medical, surgical, familial and personal histories were non-contributory and general physical examination revealed no abnormalities.

Intraoral examination revealed a solitary well-defineds swelling on the right side of hard palate and right alveolar ridge measuring approximately 5 x 4cm, oval in shape, extending from distal aspect of 15 to maxillary tuberosity anteroposteriorly and from midline of the palate to right buccal vestibule across the edentulous ridge mediolaterally. Mucosa over the swelling appeared bluish and stretched (Figure 1). On palpation, the swelling was non tender, soft, immobile and slightly compressible with no buckle cortical plate expansion. Teeth distal to the second premolar were missing in the first quadrant. Other findings include multiple carious teeth, talon's cusp irt 12 and 22, root stumps irt 28, 36, 45 and 46, missing teeth irt 23, 24, 37 and 47. Based on patient's history and clinical examination, a provisional diagnosis of residual cyst of the maxillary alveolus was considered and a differential diagnosis of adenoma of minor salivary gland of palate was given.

Patient was subjected for aspiration which yielded a brown colored fluid which was histopathologically reported as chronic nonspecific inflammation. Maxillary occlusal, panoramic and sinus view radiographs revealed pacification of the entire right maxillary sinus, suggestive of fluid accumulation, with extension of the cystic space into the alveolus and oral cavity (Figures 2, 3 and 4). Bone loss was evident in the alveolus lateral and inferior to the cystic swelling. Magnetic resonance

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imaging revealed a non-enhancing hyperintense collection on all sequences involving the entire right maxillary sinus with thinning of the walls of maxillary sinus.

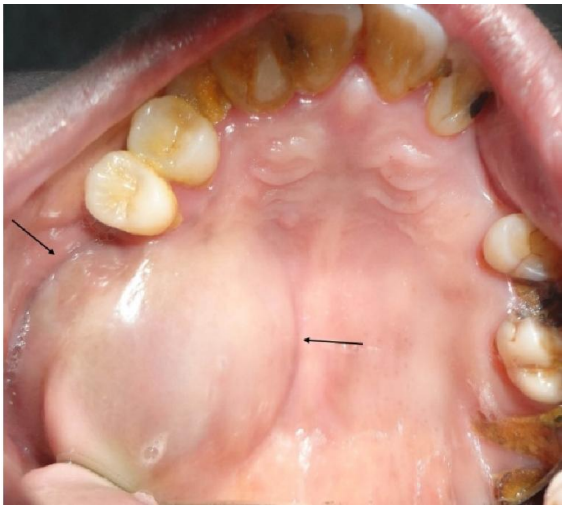

Figure 1. Intraoral photograph showing a well-defined swelling on the hard palate and alveolus on the right side

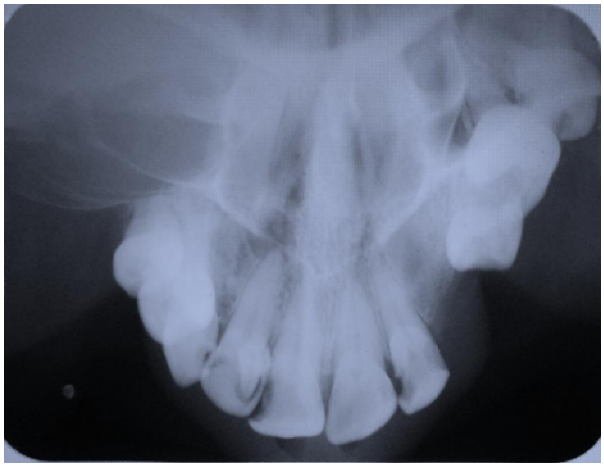

Figure 2. Maxillary occlusal radiograph showing opacification of right maxillary sinus with loss of bone and extension of cystic space into buccal cortex posterior to the second premolar

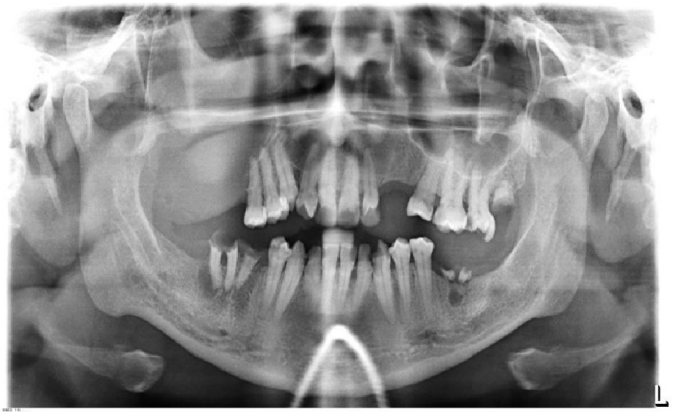

Figure 3. Panoramic radiograph showing opacification of the entire right maxillary sinus with extension of cystic space into the right alveolus

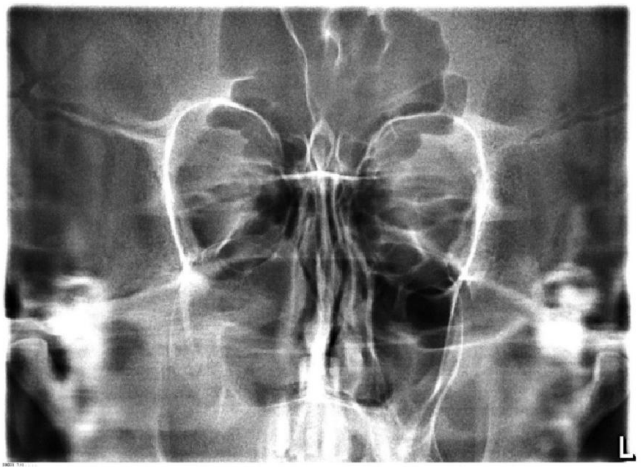

Figure 4. Sinus view showing opacification of right maxillary sinus

A focal hypointensity was noted in the basal area suggestive of sediments in the proteinaceous collection. (Figures 5 and 6) Patient was subjected for an incisional biopsy which was histopathologically reported as mucous collection with clot and normal tissue architecture. (Figure 7) Based on the history, clinical, radiographic and histopathological findings, a final diagnosis of mucocele of the right maxillary sinus was given. The patient was treated by functional endoscopic sinus surgery and the sinus lining removed. Patient was then reviewed at 6 months and 1 year post surgery which revealed no recurrence of the mucocele (Figure 8).

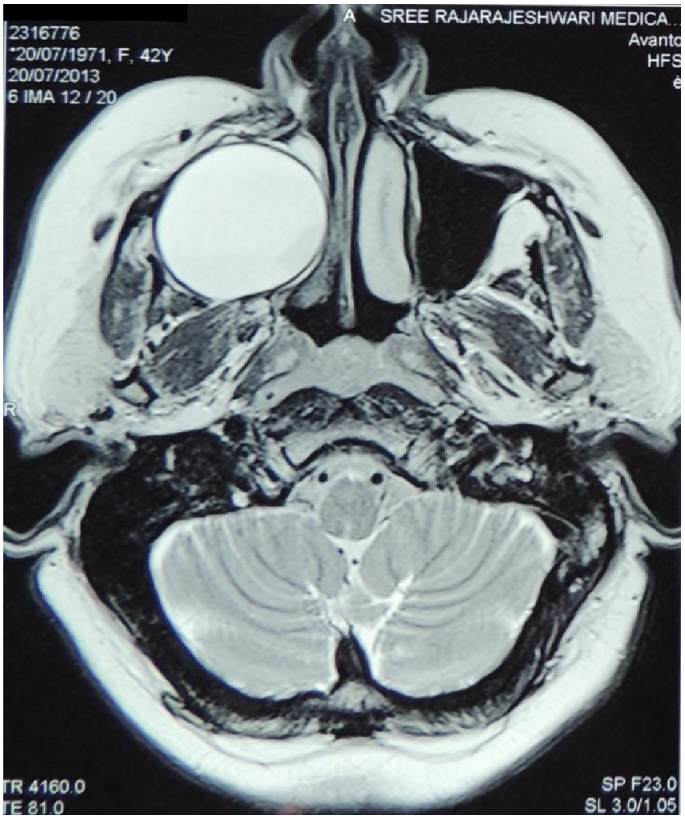

Figure 5. T2 weighted (MRI) axial section revealing a hyperintense collection involving right maxillary sinus

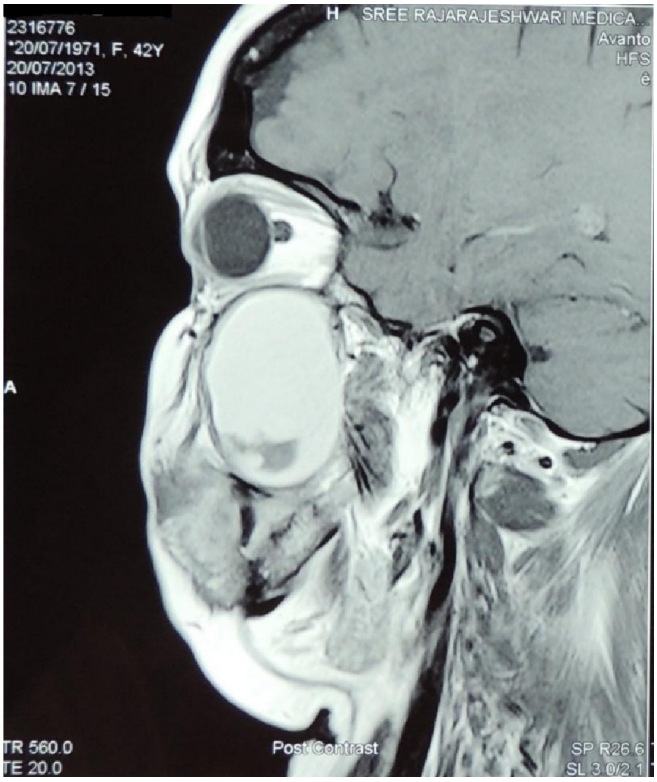

Figure 6. Post-contrast T1 weighted (MRI) sagittal section revealing a non-enhancing hyperintense collection involving right maxillary sinus

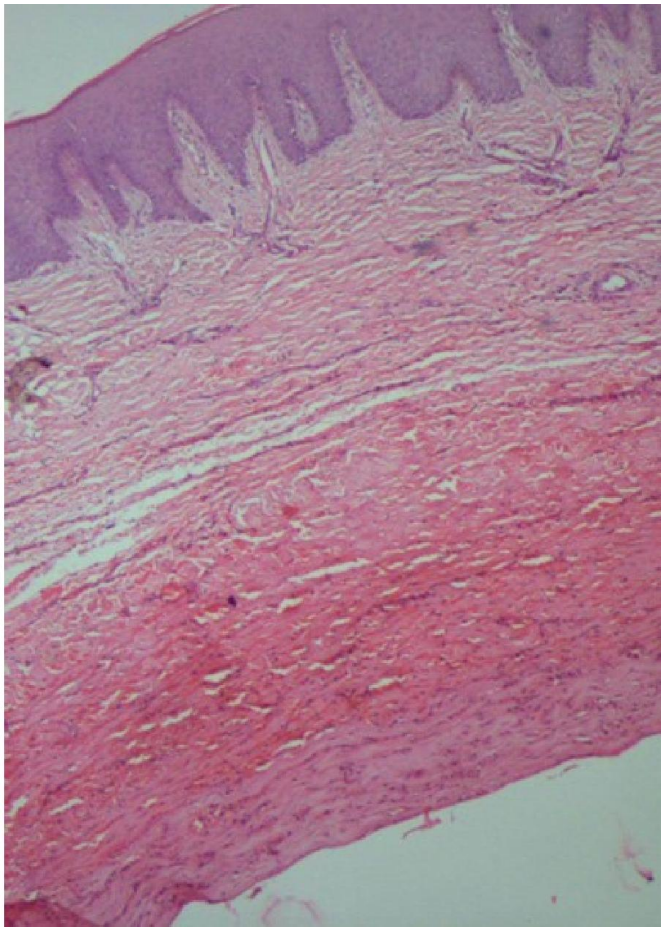

Figure 7. Photomicrograph (10X) showing stratified squamous epithelium which lines the underlying connective tissue with a pale eosinophilic area and extravasated red blood cells

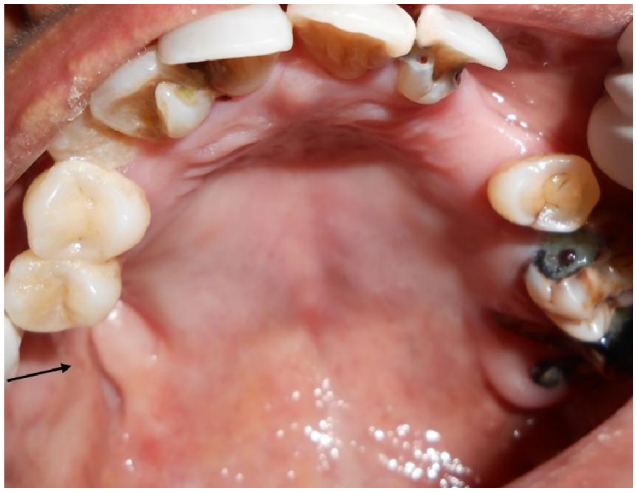

Figure 8. Post-operative photograph taken after 1 year showing no recurrence

DISCUSSION

Mucocele is defined as mucous filled cavities that can appear in the oral cavity, appendix, gall bladder, paranasal sinuses or lacrimal sac. The term mucocele is derived from the Latin words, mucus and coele (cavity). (**Rao *et al.*, 2012**) Mucocele of the frontal sinus was first described by Dezeimeris in 1725. (**Bhandary *et al.*, 2013**) Langeback first described paranasal sinus mucoceles in 1820. It was only in the early 1900s that mucoceles were given their name by Rolled and popularized by Gerber, who published 178 cases. (**Gendeh, 2014**) The reported incidence of these lesions is 1.4%-9.6%, occurring primarily during the third and fourth decades of life, and it is usually discovered during routine radiographic examinations. Generally, mucoceles are a self-limiting condition, with a rate of spontaneous regression and disappearance of 17.6%- 38%. (**Rosai, 2011**) Among the paranasal sinuses, the frontal sinus (89%) is most commonly involved followed by the ethmoid (8%), sphenoid sinuses (2%) and maxillary sinuses (1%). Fewer than 5% are bilateral and/or multiloculated. (**Marques *et al.*, 2011**) The incidence of occurrence of mucocele in the maxillary sinuses being low, our case is rare with an unusual intraoral presentation.

There are several theories about the origin and development of maxillary sinus mucoceles, including chronic infection, allergic sinonasal diseases, trauma and previous surgery. Mucoceles are thought to arise as a consequence of obstruction plus inflammation. Three main theories of pathogenesis are found in the literature: pressure erosion, cystic degeneration of glandular tissue and active bone resorption and regeneration. (**Marques *et al.*, 2011; Lund, 2008**) **Fu *et al.*** classify paranasal sinus mucoceles as primary and secondary, based on their anatomic and invasive characteristics. The proposed mechanisms for primary mucocele formation are the inflammatory blockage of mucus drainage, secretory duct obstruction and cystic degeneration of polyps. Furthermore, it is suggested that the retention of residual mucosa in the wound and long-term contact of tissue fluid could also lead to the formation of secondary mucoceles. (**Rosai, 2011**) There may be a considerable time lag between the initiating factor and the clinical presentation with the mucocele. In the case of surgery

or trauma this is an average of 23 years, whereas following an acute infective episode the mean time for presentation is 22 months (**Marques *et al.*, 2011**).

The symptoms commonly associated with the presence of paranasal sinus mucoceles are related to their expansion, usually through the least resistant path, and subsequent pressure on surrounding anatomic structures. (**Rosai, 2011**) Symptoms most frequently encountered are frontal pressure, headaches, facial swelling, or visual disturbances. (**Liu *et al.*, 2013**) The most common presenting symptom for a maxillary sinus mucocele is facial swelling with infra-orbital anaesthesia. Medial extension of the maxillary mucocele into the nasal cavity can push the inferior turbinate and cause nasal obstruction. When extending to the orbit, it may cause displacement of the eye, proptosis, ptosis, double vision and epiphora. Infrequently, as seen in our case, inferior expansion into the palatal and alveolar bone can cause palatal swelling and mobility of teeth. (**Chang and Kang, 2010**) Without bony erosion, the mucoceles must be differentiated from chronic sinusitis, retention cysts, and extensive polyposis. Once bone destruction has occurred, differential diagnosis should include malignant conditions, such as adenoid cystic carcinoma, squamous cell carcinoma, undifferentiated carcinoma, plasmacytoma, rhabdomyosarcoma, lymphoma, schwannoma, and tumors of dental origin, and a biopsy is required to confirm the diagnosis (**Sreedharan *et al.*, 2011**). Traditionally, the recommended treatment is Caldwell-Luc technique with total removal of the sinusal mucous membrane and inferior antrostomy which ensured ventilation and drainage of the sinus. (**Chang *et al.*, 2010**) With the increasing popularity of minimally invasive surgery, functional endoscopic sinus surgery has become the standard procedure. (**Lee, 2010**) More recently, the technique was almost completely replaced by endoscopic marsupialization which is effective with very low recurrence rate at or close to 0% and minimally invasive with a shorter postoperative recovery and less morbidity. (**Chang and Kang, 2010**) As with all mucoceles, long-term observation is necessary in maxillary sinus mucoceles to ensure that recurrence has not occurred (**Marks *et al.*, 1997**).

Conclusion

Mucoceles of the maxillary sinus usually present as an incidental finding, rarely producing symptoms. In such cases, routine radiographs with advanced imaging modalities like computed tomography and magnetic resonance imaging helps in accurately diagnosing the condition. Usually a wide endoscopic endonasal marsupialization of paranasal sinus mucocele is safe and less invasive than external approaches. Marsupialization with establishment of ostial drainage relieves the symptoms of the mucocele and also prevents the re-accumulation of mucus. Thus, a clear understanding of paranasal sinus mucoceles is of utmost importance in early diagnosis and rapid surgical intervention.

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