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Case Series

Odontogenic cysts in the maxillary sinus: A case series

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ABSTRACT

In this article, we present a series of three patients who reported to the department with different chief complaints. The patients underwent the necessary basic and advanced radiological investigations which showed presence of cysts along with impacted teeth in the maxillary sinus, following which all the patients were subjected to cyst enucleation under GA and the surgical specimens were sent for histopathological examination for definitive diagnosis.

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1. Introduction

Odontogenic cysts are a group of diverse lesions that range from small inconspicuous cysts found accidentally during routine examination to highly aggressive lesions that present tendency for malignant transformation. Of these, Odontogenic Keratocysts have been found to be the most aggressive with a recurrence rate of 16 to 30%.

OKC was first identified and described in 1876 and further classified by Phillipson in 1956. OKC presents with certain salient features that differentiates it from other odontogenic cysts. It behaves like a neoplasm with expansion of the cortical plates due to their growth potential and low protein content, which affects the cyst's osmotic pressure. Histologically the cyst lining may present as parakeratinised and orthokeratinised. The transformation in the parakeratinised epithelial lining owing to the growth factors and cytokines released by inflammatory infiltrates are responsible for the proliferative activities of the basal epithelium, making OKCs aggressive.

OKCs commonly occur in tooth bearing regions and the mandible is affected more common than the maxilla. The

anterior quadrant and the third molar region are the most common sites of origin in the maxilla.¹ Involvement of the maxillary sinus by OKC is unusual with <1% of cases reported in the literature.² Diagnosis of such lesions in the maxillary sinus is challenging and at times missed due to the overlapping of anatomical structures in conventional radiographs.

In this case report, we describe a rare case of OKC in relation to an ectopic maxillary third molar in the maxillary sinus of a 20-year old female patient.

2. Case Report 1

A 20-year-old female reported to the Department of Oral and Maxillofacial Surgery at Annoor Dental College and Hospital, Kerala, with a history of frequent nasal congestion for the past 6 months, following which the patient developed pain and pus discharge from the right maxillary third molar region since two months. The patient reported that she had taken antibiotics and analgesics occasionally for the same. The patient during the same time visited an Orthodontist who advised an OPG prior to starting orthodontic treatment. OPG revealed an impacted third molar in the right maxillary sinus and was advised to take

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CBCT for further investigation. CBCT gave an impression of an obliterated right maxillary sinus with an impacted 18. Since CBCT showed a large destructive lesion with resorption of the posterolateral wall of the maxillary sinus, a CT was further taken to assess the superoinferior extent of the lesion and to check for involvement of the orbital floor. CT evaluation showed that the orbital floor was intact, however, the lesion had involved the right lateral wall of nose causing resorption and the third molar was pushed into the right nasal cavity causing deviation of the nasal septum (Figure 1).

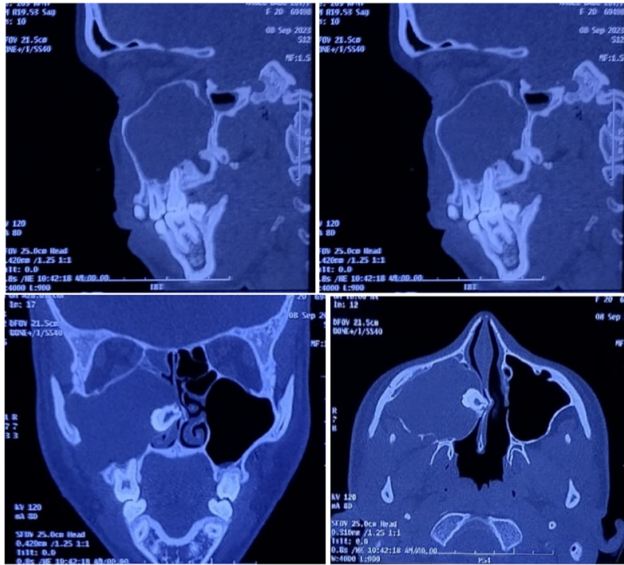


Figure 1: CT sections showing obliteration of right maxillary sinus with associated impacted tooth pushed to the septum of the nose

Following radiographic investigations, aspiration of the cystic lesion revealed pale yellow, viscous aspirate, suggestive of keratocyst. Therefore, the patient was planned for surgical enucleation of the cyst under GA using Caldwell Luc approach along with FESS by ENT surgeon, for exploration of the maxillary sinus and nasal antrostomy (Figure 2).

The oral cavity was sutured using 3-0 monocryl and betadine with metrogyl soaked ribbon gauze was packed into the maxillary sinus through the nose. Patient was extubated uneventfully and shifted to surgical ICU.

The specimen was sent for histopathological examination, which showed a parakeratinised stratified squamous cystic lining, with the epithelium exhibiting a corrugated parakeratinised lining. Focal areas of the connective tissue showed dense chronic inflammatory cell infiltrate and the cystic lumen shows flakes of keratin. All the features confirming the diagnosis of parakeratinised odontogenic keratocyst.



Figure 2: Intraoral exposure of maxillary sinus and surgical specimen retrieved

3. Case Report 2

A 30-year old well-built healthy male was referred to the department of Oral and Maxillofacial surgery with a history of pus discharge from the maxillary left posterior region. An OPG taken at a dental clinic for the same chief complaint showed root resorption with respect to 25 and 26 and associated radiolucency, following which the patient was referred (Figure 3). A CBCT was taken to further investigate the lesion.

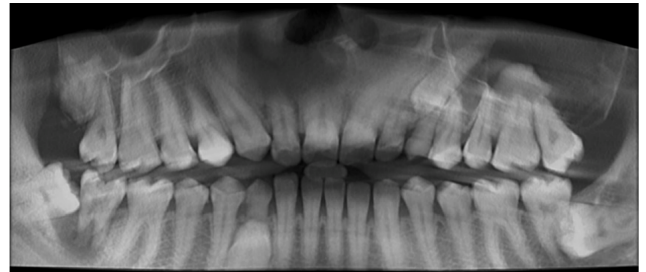


Figure 3: Reconstructed panoramic view

CBCT report showed a vertically impacted 23 arising just apical to the apex of 63 tooth and an inverted 28 impacted at the interdental region between 25 and 26 with its crown protruding into the sinus cavity. A well defined solitary lesion was also noted in the left maxillary sinus extending mesiodistally from the distal of 23 upto the maxillary tuberosity and superoinferiorly from the sinus floor upto the infraorbital region. There was also expansion and thinning of the bicortical plates with complete erosion of the maxillary tuberosity, with communication into the oral cavity. The lesion has also pushed the lateral wall of the nose medially (Figures 4 and 5).

Once the extent of the lesion was determined, the patient was operated under GA with cyst enucleation along with extraction of the impacted teeth (Figures 6, 7 and 8).

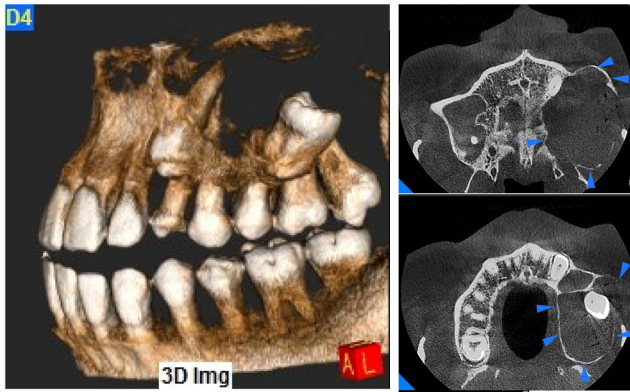


Figure 4: CBCT sections showing obliteration of left maxillary sinus with impacted teeth

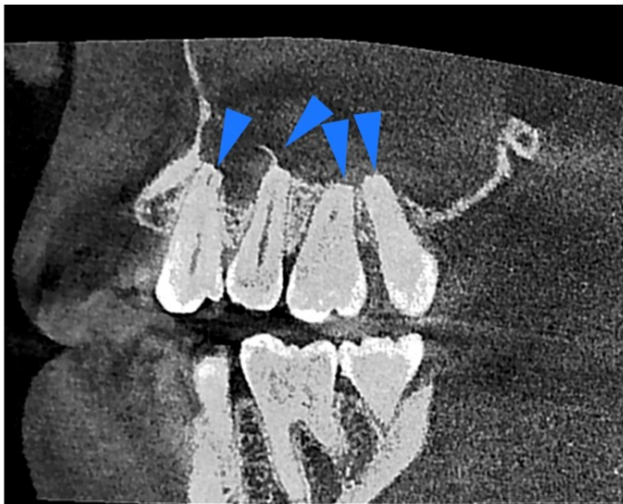


Figure 5: Resorption of root from 24-27

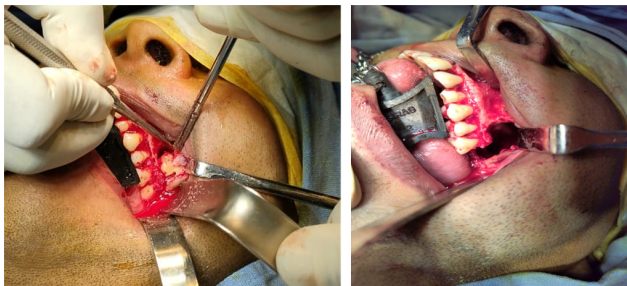


Figure 6: Intraoral exposure of maxillary sinus

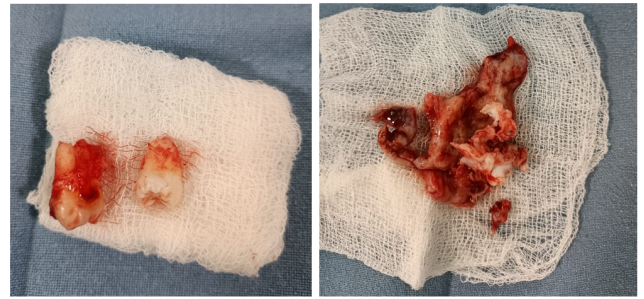


Figure 7: Impacted teeth removed from sinus and surgical specimen

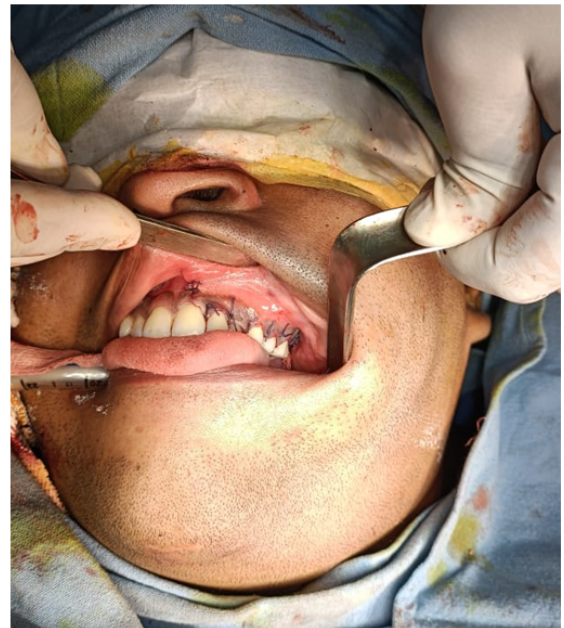


Figure 8: Surgical site closure

Intraoperatively, opening of the maxillary sinus revealed the presence of white cheesy viscous content that resembled keratin pearls which sprouted the differential diagnosis of an OKC. Once the sinus was cleared of the cyst along with the lining, the flap was sutured back except at the maxillary tuberosity due to lack of adjacent soft tissues. This opening was utilised to pack the sinus using iodoform ribbon gauze to facilitate healing and prevent oroantral communication as well. The patient has been kept on follow-up on a weekly basis to plan for closure of the site once the adjacent tissues are healthy and healed enough to withstand suturing.

The specimen retrieved intraoperatively was sent for histopathological examination, which revealed an orthokeratinised stratified squamous cystic lining epithelium in association with a fibrovascular connective tissue capsule. The lining epithelium consists of prominent granular cell layer. The connective tissue exhibited moderately dense chronic inflammatory cell infiltrate

and the associated lining epithelium showed hyperplastic changes with pseudo epitheliomatous hyperplasia. The section also showed keratin flakes.

Thus, a diagnosis of orthokeratinised odontogenic keratocyst was established.

4. Case Report 3

A 19-year old male patient with congenitally missing maxillary anterior teeth sought treatment for the same. During routine radiography, the presence of impacted 21, 22 and 23 was revealed with 23 impacted close to the zygomatic buttress. The OPG also revealed a radiopacity in the maxillary sinus following which a CBCT was taken. CBCT revealed the presence of multiple impacted teeth along with obliteration of the maxillary sinus by a well defined lesion with soft tissue density (Figure 9).

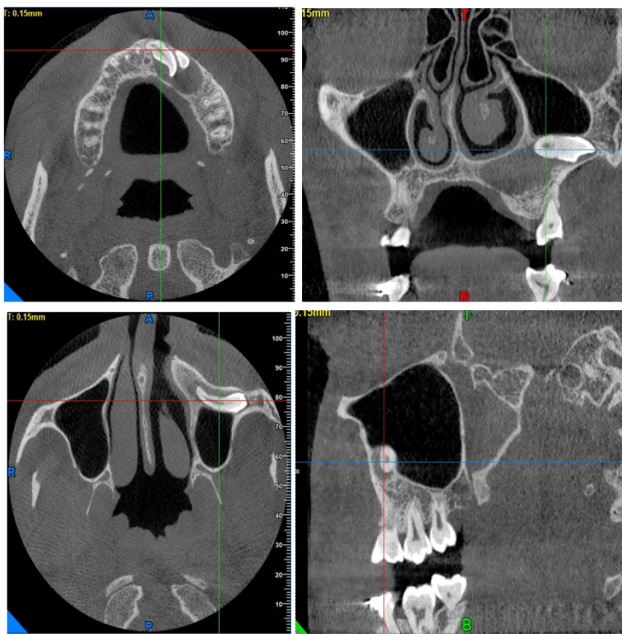


Figure 9: CBCT sections showing cyst in the left maxillary sinus with impacted teeth

The patient underwent the same surgical procedure as the other two patients previously, cystic enucleation under GA with extraction of all the impacted teeth (Figure 10).

Histopathological examination of the surgical specimen showed a non-keratinised cystic lining epithelium associated with an inflamed hyperplastic connective tissue capsule, confirming the diagnosis of an inflamed dentigerous cyst.

5. Discussion

Odontogenic Keratocyst is a common odontogenic developmental cyst that accounts for 10-12% of all jaw cysts.³ In the mandible, they account for 65-83% of cases



Figure 10: Intraoperative pictures and extracted impacted teeth from the sinus

but their occurrence in the maxilla is rare, found to be less than 1% and usually associated with systemic syndromes.⁴

In the current case reports, the cysts involved almost the whole of the maxillary sinus, even extending medially upto the nasal septum. It was also associated with impacted teeth which were found to be pushed to the lateral wall of the nose, through the sinus. The majority of cases reported in literature have been found to be associated with symptoms but these cases have reported very vague symptoms such as that of sinusitis or not reported any symptoms until the lesion became too invasive.

Owing to its recurrence rate and potentially aggressive nature, OKC has been the theme of study for a long time. It has been suggested to originate from dental lamina remains⁵ and has also been considered a benign cystic neoplasm, due to its growth capacity and development characteristics related to the mutation of tumor suppressor gene, PTCH.⁶

OKC in the maxillary sinus is presumed to be arising from the entrapment of the odontogenic epithelium within the sinus. This occurs because of the close anatomic relationship between the dental lamina and developing antrum.⁷ Aggressive in nature, they have been found to penetrate into the surrounding soft tissues with expansion and perforation of the cortical bone.

Odontogenic keratocyst has two variants. Orthokeratinised and Parakeratinised. The rare orthokeratinised OKC has a lesser recurrence as compared to the parakeratinised variant. According to literature, the recurrence rate for parakeratinized OKCs was found to be 42.6%, but only 2.2% for orthokeratinized OKCs.⁸

The proclivity for parakeratinised OKCs to recur may be due to the presence of hypercellular epithelial lining with corrugations and a polarized layer of basal cells suggestive of a highly active epithelium.⁹

The primary aim of treatment is to achieve total eradication of the cyst along with the cystic lining, owing to its aggressiveness and recurrence rate. Various surgical options include marsupialisation, enucleation, enucleation with Carnoy's solution, enucleation with cryotherapy, curettage and resection. In one of the presented cases, surgical enucleation was combined with nasal anrostomy using FESS to ensure complete enucleation of the cyst lining as well as to facilitate easier monitoring during follow-up visits. This combined approach also facilitated closure of the intraoral surgical site thus reducing the possibility for development of oroantral communication.

Even with all the possible surgical options, there is no universally accepted treatment for OKC that ensures complete eradication.¹⁰ Enucleation alone has a recurrence rate of 17-56% and enucleation followed by Carnoy's application, has a recurrence of 1-8.7%.¹¹

The ectopic eruption of teeth in the non-dental sites is rare and one such site observed is the maxillary sinus. Though it's etiology remains unclear, some of the possible causes include trauma, infection, developmental anomalies and pathological conditions.¹² It has been suggested that the displacement of developing tooth buds by the growing odontogenic cysts results in ectopic eruptions.¹³

The presentation of an associated impacted teeth, it's location and involvement of the draining sinus made us to clinically consider it to be an infected dentigerous cyst, which was found to be true in only one of the cases. Hence it is important for the clinician to consider OKC as well in the differential diagnosis for such lesions when they occur in a younger patient.

6. Conclusion

OKC in the maxillary sinus is a rare occurrence which does not present with usual features as it's commonly encountered counterparts. Due to the potentially aggressive nature and recurrence rate of these cysts, we emphasize the significance of long term follow-up of these patients.

7. Declaration of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

8. Source of Funding

Nil.

9. Conflicts of Interest

There are no conflicts of interest.

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