CASE REPORT

Orthokeratinized Odontogenic Cyst in Posterior Maxilla: A Rare Diagnosis

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Abstract

Aim: Aim of this case report is to discuss and identify a rare diagnosis yet seen commonly in oral and maxillofacial field and often misdiagnosed as odontogenic keratocyst (OKC).

Background: The term "odontogenic keratocyst" was first described by Philipsen in 1956. This was then modified by World Health Organization (WHO) in 2005 as "odontogenic unicystic" or "multicystic intraosseous tumor" with a distinctive parakeratinized stratified squamous epithelial lining and the propensity for aggressive and infiltrative activity. It has a male predilection and bimodal age distribution. Orthokeratinized OKC makes up to 2.1% of all the cystic lesions of the jaws.

Case description: A young female patient aged 18 years reported to our department with the chief complaint of pus like discharge from upper left back tooth region for the past 2 months. After all the clinical, radiological, and histopathological findings, the patient was then diagnosed with orthokeratinized odontogenic cyst (OOC) of posterior maxilla.

Conclusion: Orthokeratinized odontogenic cyst has clinical, histological, and biological characteristics that distinguish it from OKC, including better prognosis and decreased recurrence rate.

Clinical significance: It is important to distinguish between OKC and OOC as the latter shows less chances of recurrence and less scope for malignancy transformation.

Keywords: Maxillary, Odontogenic keratocyst.

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Introduction

A cyst is a pocket-like sac that can hold air, liquid, or other materials. Odontogenic keratocyst is a cyst of odontogenic origin lined by keratinizing epithelium. It is a cyst of aggressive nature and has high recurrence rate. Philipsen first coined the phrase "odontogenic keratocyst" in 1956. This was then modified by WHO in 2005 as "odontogenic unicystic" or "multicystic intraosseous tumor" with a distinctive parakeratinized stratified squamous epithelial lining and the propensity for aggressive and infiltrative activity.[1] It has a male predilection and bimodal age distribution. Orthokeratinized OKCs account for up to 2.1% of all jaw cystic lesions.[2] Most common sites of occurrence are ramus and mandibular molar region. The most common treatment modality considered is curettage followed by chemical cauterization due to its high recurrence incidences.

Herewith, we present a case report on OKC with orthokeratinized stratified squamous epithelial lining in posterior maxilla associated with an impacted third molar which was a radiohistopathological finding.

Clinical History

A young female patient aged 18 years reported to our department with the chief complaint of pus-like discharge from upper left back tooth region for the past 2 months. The patient was asymptomatic 2 months back after which she started experiencing pus-like discharge from upper left back tooth region which was intermittent and was associated with mild pain that aggravated only on chewing.

On examination, the patient did not have any extra oral finding such as swelling, paresthesia, tenderness or any form of discharge. Intra-orally, on inspection, the patient had mild swelling on palatal aspect with respect to the left maxillary second molar and gingival inflammation in the same region (Fig. 1). On palpation, all the inspector findings were confirmed and pus-like discharge was seen from the distal aspect of second molar on probing. After all the clinical examination, the patient was provisionally diagnosed with chronic periapical abscess with respect to second molar.

On the radiographic examination, orthopantomogram revealed a cystic cavity associated with the impacted third molar pushing the sinus lining (Fig. 2). Furthermore, a DentaScan was done that revealed a solitary, expansile, well-defined radio-opaque shadow of cystic cavity enveloping the impacted third molar infiltrating the maxillary sinus of same side with corticated borders (Fig. 3).

The patient was then planned for a fine needle aspirational biopsy for confirmatory diagnosis which revealed infected OKC of posterior maxilla (Fig. 4).

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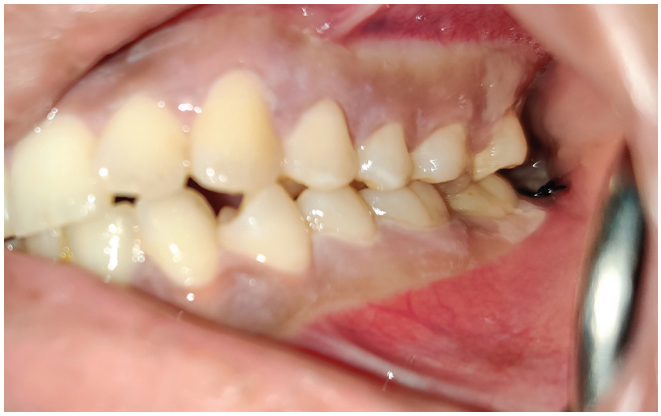


Fig. 1: Intraoral preoperative photograph

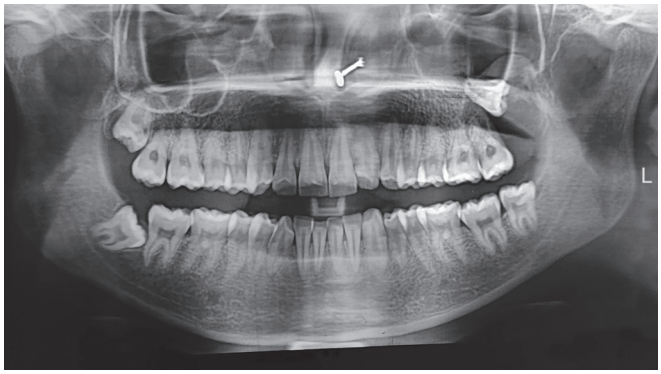


Fig. 2: Preoperative orthopantomogram

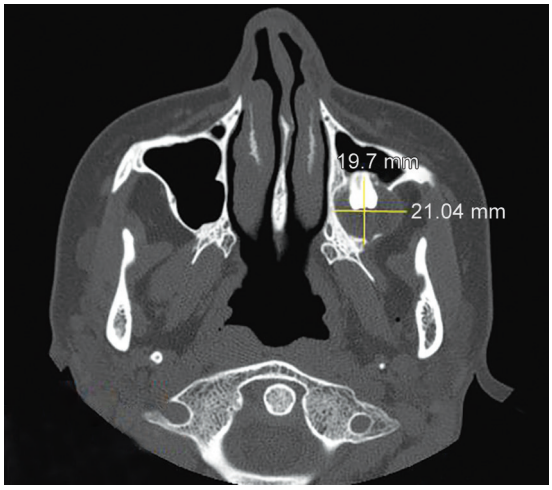


Fig. 3: Preoperative DentaScan depicting well defined cystic lining enclosing hard tissue

After all the clinical, radiological, and histopathological findings, the patient was then planned for curettage followed by chemical cauterization.

The patient was then informed and educated about the procedure and a written informed signed consent was taken for the same.

Under all aseptic conditions, the patient was prepared using 10% betadine solution and draped accordingly. An extra oral maxillary block was given using 2% lignocaine with adrenaline and up to 5-mL anesthetic agent was deposited. After checking all the subjective and objective signs and symptoms a buccal vestibular incision was given with respect to 17–18 region, and the cystic lesion was exposed. Using curette, whole of the cystic lining was excised in toto along with the involved impacted third molar (Fig. 5). Further, chemical cauterization was done using Modified Carnoy's solution for 3 minutes. After copious irrigation using normal saline, the primary closure was done using 3–0 vicryl suture in interrupted pattern.

The postoperative radiograph depicting posterior maxilla free from pathology and an intact maxillary sinus lining with satisfactory bone healing is shown in (Fig. 6).

The whole of the excised pathology was sent for histopathological examination, which revealed the cystic epithelium is stratified squamous orthokeratinized type, 6–9-cell layer thick, showing multiple keratin flecks. The overall histopathological report depicted orthokeratinized type infected OKC.

Discussion

Pindborg and Hansen identified certain common features in 1962, including daughter cyst within the cystic wall has a consistent thickness, a keratinized design with a wavy surface, palisading of basal cells with hyperchromatic nuclei, and a keratinized design with a wavy surface.[3] The latter, however, was classified as a separate group by Wright in 1981, and is known as OOC. The WHO Working Group classified OKC as a tumor in 2005, and suggested the term keratocystic odontogenic tumor (KCOT) to distinguish the lesion from the orthokeratinizing variation, which is a separate disease and is still thought to be a developing odontogenic cyst.[4]

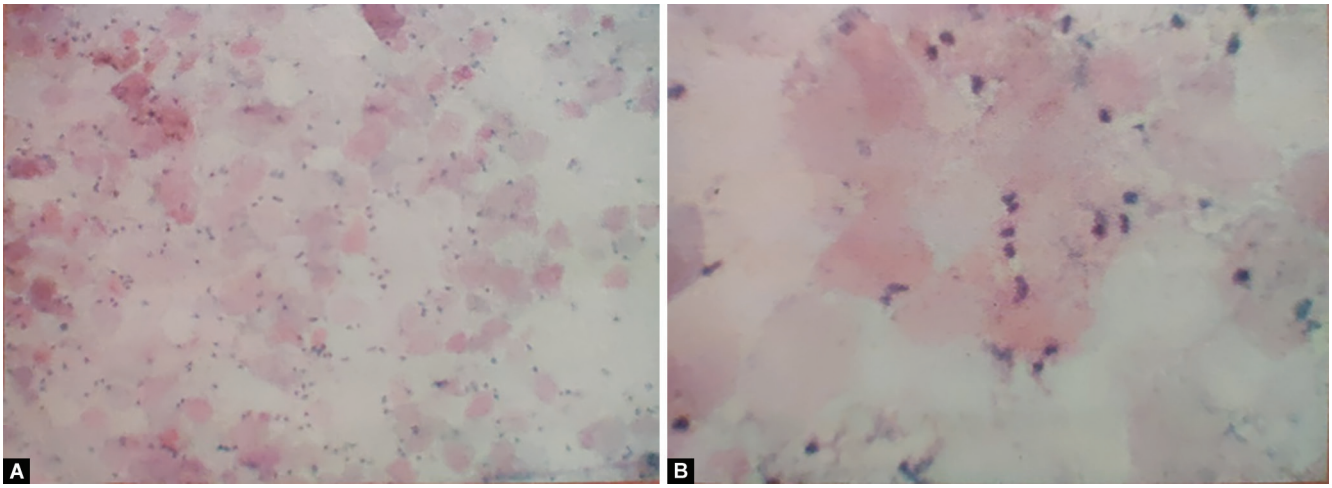
Orthokeratinized odontogenic cyst is a rare developing odontogenic cyst that accounts for between 5.2% and 16.8% of cases previously identified as OKC in the various previous case series.[5] In a thorough assessment of different populations, MacDonald et al.[6] and Li et al.[7] found that males are more frequently impacted than females, in contrast to our case. Clinically, both OKCs and OOCs have similar features as they are normally asymptomatic, and the most common presenting sign is a slow-growing expansile swelling occurring in the tooth bearing region, that is occasionally accompanied by pain and purulent discharge.[8]

Radiographically, in both OKC and OOC, a smooth solitary, mostly unilocular, and expansile lesion with corticated borders (until infected) is seen. Unlike OKC, most of the OCCs are associated with an impacted tooth and are generally diagnosed as a dentigerous cyst. These cysts have a diameter of 20–50 mm on average. The largest diameter of the lesion in our case was 19 mm.

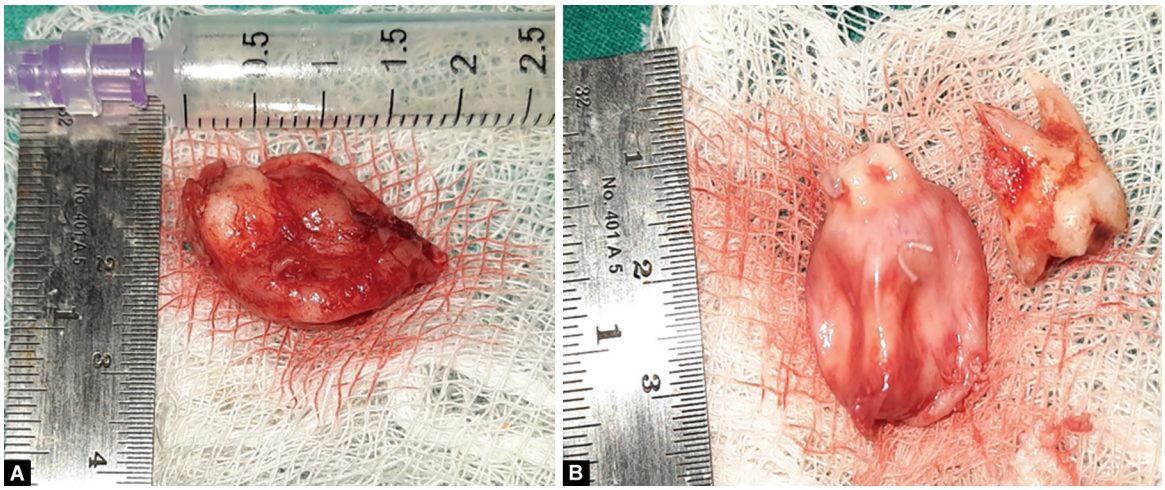
Histopathologically, OKC has a cystic lesion lined by varying thicknesses of parakeratinized type stratified epithelium with palisading of the basal cell layer, whereas OOC has a 4–8-cell layer orthokeratinized epithelium having prominent granular cell layer and flat to columnar basal cells without palisading and a capsular wall of fibrous connective tissue.

The rate of recurrence is about 0–4% after enucleation and chemical cauterization during a mean period of 6.4–7.8 years of regular follow-up. The recurrence rate is significantly lower than the 30% or higher recurrence rate linked with KCOT.[3]

Treatment of choice for this lesion is surgical excision followed by chemical cauterization using modified Carnoy's solution to prevent chances of recurrence.[5]



Figs 4A and B: Aspirational biopsy revealed neutrophils and lymphocytes along with multiple keratinocytes, giving an impression of infected OKC



Figs 5A and B: Surgical curettage in toto along with involved impacted third molar and infected second molar

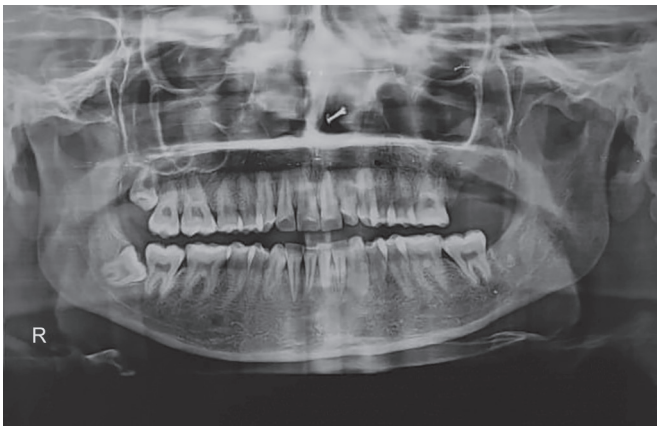


Fig. 6: Postoperative radiograph

As a result, OOC differs significantly from KCOTs in terms of clinical, pathologic, and behavioral aspects. It appears to be a rare but persistent collection of odontogenic developing cysts which can be classed as other known forms and so should be treated as a separate clinical entity.

Conclusion

Orthokeratinized odontogenic cyst has clinical, histological, and biological characteristics that distinguish it from OKC, including better prognosis and decreased recurrence rate. Various radiolucent pathology of the jaws, such as dentigerous cysts, ameloblastoma, and OKC, should all be examined in the evaluation of patients of OOC. When creating an effective treatment plan, consider the patient's age, the extent of the lesion, and the histological picture.

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