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Rare Encounter: Odontogentic Keratocyst in The Posterior Maxilla: A Case Report

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Abstract

The odontogenic keratocyst (OKC) is a developmental cyst of odontogenic origin, known for its distinctive clinical and histopathological features. Despite being classified as benign, OKC demonstrates aggressive growth, local tissue infiltration, and a high recurrence rate, making it more difficult to manage than other odontogenic cysts. It most commonly affects the mandible, especially the posterior body and ramus, while maxillary involvement is rare.

This case report describes an uncommon presentation of OKC in the posterior maxilla. Initially misdiagnosed as a dentigerous cyst, the correct diagnosis was only established after a comprehensive diagnostic process, including clinical examination, radiographic imaging, and histopathological evaluation. The unusual location contributed to the diagnostic challenge, as OKC can mimic other cysts both clinically and radiographically.

This case underscores the importance of including OKC in the differential diagnosis of jaw cysts, especially in atypical locations. Early recognition is crucial due to the lesion's aggressive nature and potential for recurrence. Effective management involves thorough enucleation, close postoperative follow-up, and consideration of adjunctive therapies.

By documenting this rare maxillary presentation, the report contributes to the broader understanding of OKC and emphasizes the need for long-term monitoring to detect possible recurrences. Increased awareness of atypical presentations can aid in timely diagnosis and improve patient outcomes.

Keywords: Odontogenic Keratocyst, Posterior Maxilla, Adjuvant Therapies.

Introduction

Odontogenic keratocyst (OKC) is a distinctive form of developmental odontogenic cyst comprising 12% of the entire jaw cysts.[1] Philipsen coined the term 'odontogenic keratocyst' in the year 1956.World Health Organization classification 2005 reclassified the odontogenic keratocysts (OKCs) under benign odontogenic tumors. The new terminology given to this particular entity was keratocystic odontogenic tumor (KCOT). The evidence for reclassification was based on "aggressive growth", recurrence after treatment, the rare occurrence of a "solid" variant of OKC, and most importantly, mutations in the PTCH gene. The mutations in OKCs are not limited to PTCH, as mutations in CDKN2A, TP53, MCC, CADMI and FHIT have also been reported.[2] But the OKCs were reincorporated in the cyst classification because many papers showed that the PTCH gene mutation could be found in nonneoplastic lesions, including dentigerous cysts, and furthermore, many researchers suggested that resolution of the cyst after marsupialization was not compatible with a neoplastic process.[3] The etiology of odontogenic keratocyst (OKC) is believed to be associated with remnants of the dental lamina. A distinctive feature of this lesion is its tendency to spread along the cancellous bone spaces with minimal cortical expansion. Clinically, it is characterized by aggressive local destruction and a high recurrence rate, along with a propensity for multiplicity—particularly in patients with conditions such as Nevoid Basal Cell Carcinoma Syndrome, also known as Gorlin-Goltz syndrome. The most common site as per the occurrence rate of OKC is the mandible (73%) compared with 27% in the maxilla.[1] The present case report describes an unusual presentation of OKC in the maxillary posterior region in a 16 years female patient.

Case Report

An 16-year female patient reported to Department of Oral and Maxillofacial surgery, Sri Hasanamba Dental College and Hospital with the complaint of pain and pus discharge in her right upper back tooth region since 15 days.

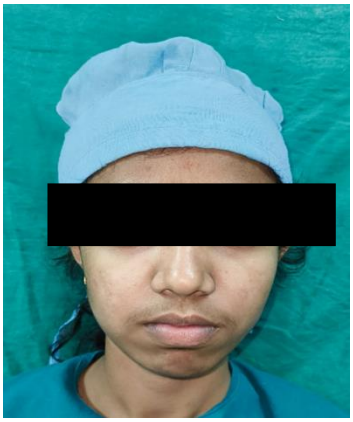

Figure 1: Extraoral photograph of patient

On extraoral examination, mild swelling was noted over the right middle 1/3rd of the face, 1cm away from the corner of the mouth extending till the lower border of ear lobule. Intraoral examination revealed a roughly oval, 1x1 cm swelling in the 17,18 region, obliteration of the labial vestibule with expansion of labial cortical bone. Swelling was soft to firm in consistency and perforation of labial cortical bone was suspected. Based on clinical manifestations, provisional diagnosis of "dentigerous cyst" was given. Radiographic investigations like orthopantamograph (OPG) and Cone beam computed tomography (CBCT) was performed,

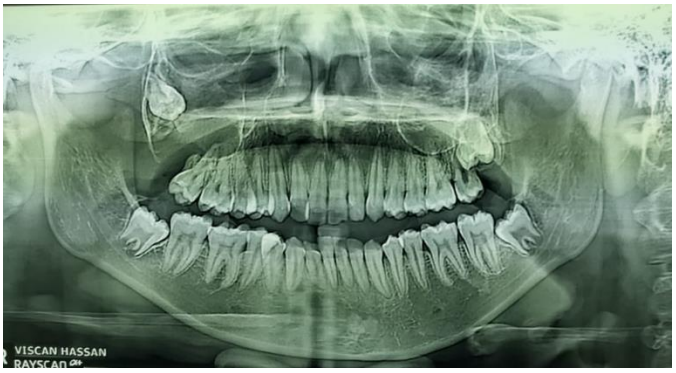

Figure 2: Orthopantomograph showing unilocular radiolucency in maxillary posterior region

Orthopantomagram (OPG) revealed well corticated radiolucency in the right maxillary posterior region associated with deeply impacted 18 (Figure 02). Radiolucency was approaching the posterior wall of maxillary sinus. and medial displacement of roots of 17 noted.

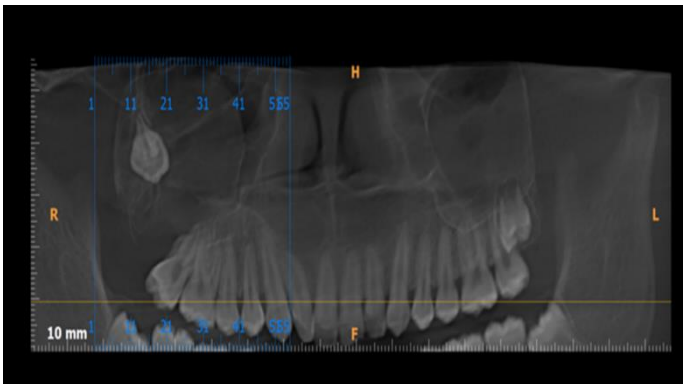

Figure 3: Derived Panaromic Image

Cone beam computed tomography (CBCT) images in cross-section and sagittal section and coronal sections revealed a fairly well demarcated, roughly oval shaped radiolucent lesion noted in the maxillary right posterior region associated with impacted 18.and the lesion is invading right maxillary sinus causing extreme elevation, thinning and possible minimal erosion of the sinus floor (Figure 03).

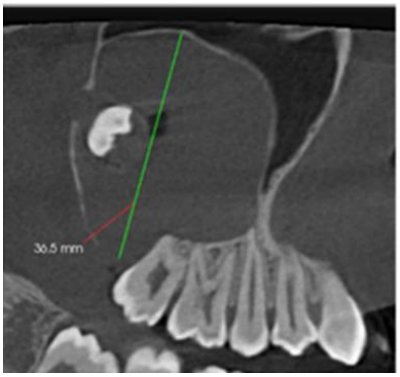

Figure 4 a: Superio-inferior

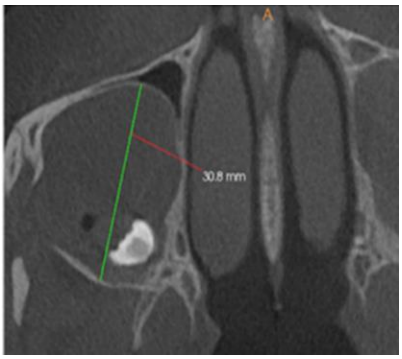

Figure 4 b: Anterio-posterior

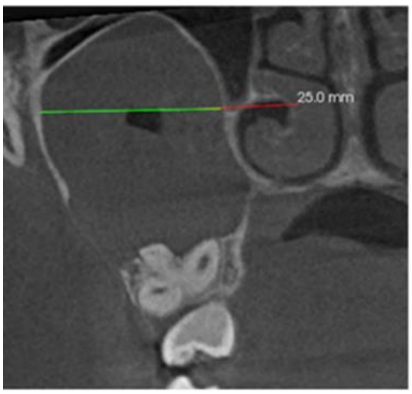

Figure 4 c: Medio-lateral

Mesio-distally, the lesion is extending from the roots of 15 till posterior wall of right maxillary sinus, its measures 36.5 mm superior-inferiorly (Fig.04 a),30.8 mm anterio-posteriorly (Fig .04 b) and 25 mm medio-laterally (Fig.04 c). Extreme displacement of 18 noted in disto-superior aspect and roots of 17 are tilted mesially, minimal expansion of buccal and palatal cortical plate noted.

Figure 5 a: Cheesy white aspirate

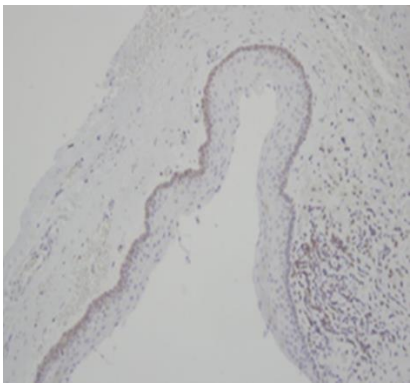

Figure 5b: Histopathologic features showing Stratified squamous epithelium with 6-8 cell layer thickness

Cystic aspiration yielded cheesy white fluid, and incisional biopsy revealed fragments of tissue lined by stratified squamous epithelium with surface ulceration and keratin debris, sub epithelium shows dense chronic inflammatory cell infiltrates with congested blood vessels suggestive of Odontogenic keratocyst.

Based on history, clinical and radiological examinations and aspiration cytology, provisional diagnosis of odontogenic keratocyst was derived. A surgical enucleation of the cyst was planned and a written consent was taken from the patient. Post-excision, chemical cauterization with Carnoys solution was done. Satisfactory healing was noted. Because of high reoccurrence of OKC periodic follow-up for next 5 years was advised

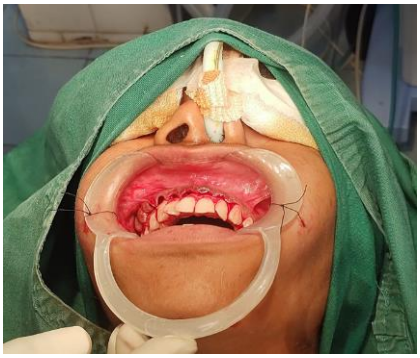

Figure 6 a: Crevicular incision placed from 23 to 28

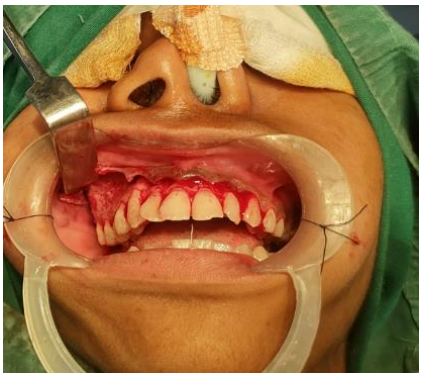

Figure 6b: Full thickness mucoperiosteal flap reflected

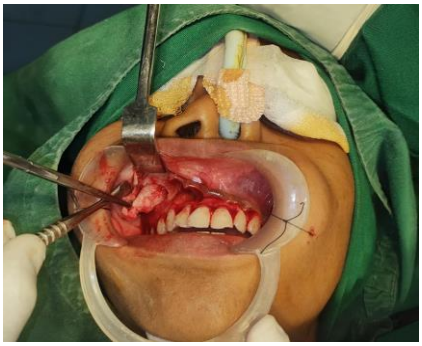

Figure 6c: Lateral bony window created and cystic lining exposed, removed in toto followed by extraction of 18

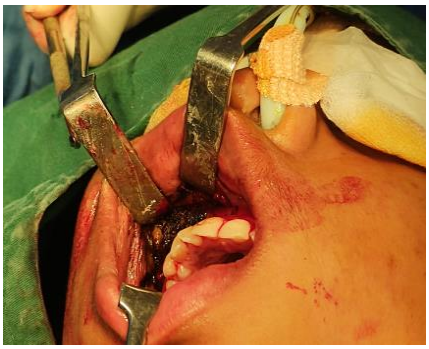

Figure 6d: Chemical cauterization done by modified carnoy's solution.

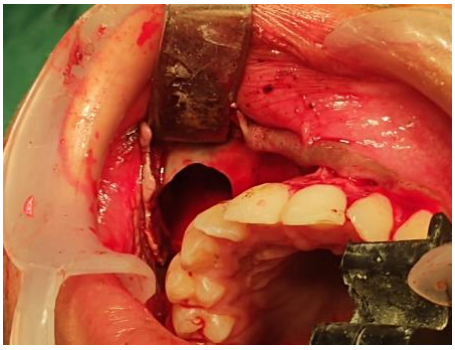

Figure 6e: Irrigation done with betadine and saline

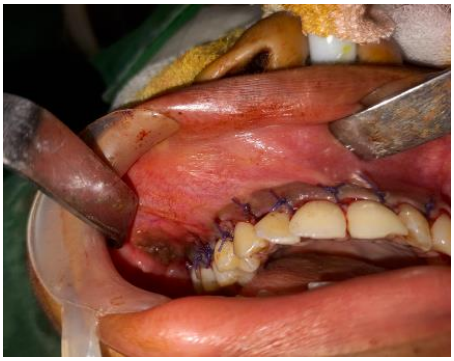

Figure 6f: Wound closure done with 3-0 vicryl

Discussion

OKCs are the third most common developmental jaw cysts. The term was broadly used for all keratin forming cysts during the 1950s. It was in 1956 that Phillipsen described OKC for epithelial developmental cysts,[1] main features of odontogenic keratocysts were described in 1963 by Pindborg and Hansen,[4] OKC demonstrates aggressive growth, local tissue infiltration, and a high recurrence rate, making it more difficult to manage than other odontogenic cysts.

It affects patients from the 1st to 9th decade of life with peak incidence in second to third decade and a second smaller peak is noticed in 50-70 years of age. It has slight male predominance.[2] Most of the cases of OKC affect the body of mandible, especially molar angle ramus region. Literature suggests that few cases of OKCs occur in maxilla, but in the maxilla, it is seen most commonly in the canine area, followed by third molar tuberosity and anterior maxilla. In most of the cases, it presents as a periapical lesion. Here, we present a case which has occurred in quite younger age, and the lesion was eroding maxillary sinus.

An important characteristic of OKC is its propensity to grow along the internal aspect of the jaws, causing minimal expansion. They are often asymptomatic and are detected on routine radiographic examination. In the present case, cortical expansion was noted and the lesion appeared to have grown labiolingually than mesiodistally.[2]

Radiographically, an odontogenic keratocyst (OKC) commonly presents as a well-defined radiolucent lesion that may be either unilocular or multilocular.[5] In this case, imaging studies, including computed tomography, demonstrated complete obliteration of the right maxillary sinus along with an ectopically positioned third molar within the sinus. Definitive diagnosis was established following surgical exploration, and histopathological examination revealed the characteristic features of an OKC.

Several treatment approaches for OKCs have been suggested in the literature. Conservative approach includes simple enucleation and marsupialisation. Aggressive approach includes chemical curettage with Carnoy's solution, peripheral osteotomy, and bone resection.[2]

Recurrence rate was observed that exclusively involved the parakeratinized variety,[4] The application of Carnoy's solution has been very effective in adjunct to peripheral osteotomy and enucleation in extensive lesions in reducing the recurrence rate. For our case too, freshly prepared Carnoy's solution was used post-enucleation and curettage. Even with Carnoy's solution application, a recurrence rate of 1-8.7% is reported and hence patient has been kept under periodic follow-up.[1]

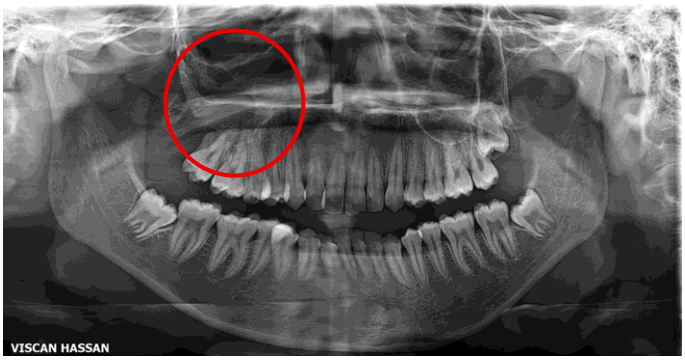

Figure 7: Six months post-operative OPG

Conclusion

OKCs have a high recurrence rate, necessitating thorough clinical and radiographic evaluation. Histopathological examination is essential for diagnosis. Given the aggressive nature and potential for recurrence, adjuvant therapy in addition to surgical intervention is recommended. Regular follow-up is crucial to detect recurrences early.

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