RESEARCH PAPER

Silent Shadows: A Rare Posterior Maxillary OKC Sparing the Sinus with an Adjacent Fibro-Osseous Enigma - A Case Report

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ABSTRACT

Background

Odontogenic keratocyst (OKC), recently reclassified as keratocystic odontogenic tumor (KCOT) and later returned to cyst status by the WHO, is a unique and locally aggressive developmental odontogenic cyst. This article explores its clinical, radiographic, and histopathological features, highlighting its high recurrence potential and association with nevoid basal cell carcinoma syndrome (NBCCS).

Case Presentation

This case report presents a rare posterior maxillary OKC sparing the sinus with an adjacent fibro-osseous enigma. OKCs often present asymptomatically, but can cause swelling, pain, and tooth displacement. Radiographically, they typically appear as well-defined radiolucencies, either unilocular or multilocular, often in the posterior mandible but rarest in maxilla less than 1% cases. Histologically, OKCs show a uniform epithelial lining with parakeratinized surface and palisaded basal cells.

Management

The management of OKC remains controversial, ranging from conservative approaches like marsupialization and enucleation to aggressive treatments involving resection. The article emphasizes the importance of long-term follow-up due to recurrence risks and discusses emerging adjuncts like Carnoy's solution and cryotherapy.

Conclusion

A comprehensive understanding of the lesion's behavior, diagnostic features, and management strategies is critical for effective treatment planning and minimizing recurrence.

Keywords: Odontogenic keratocyst, Posterior maxilla, Fibro-osseous lesion, Case report.

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INTRODUCTION

Odontogenic keratocyst (OKC) is a locally aggressive, tooth-derived cyst that arises from remnants of odontogenic epithelium in various regions of the jaw. It is the third most frequently encountered odontogenic cyst, representing approximately 12% of all maxillofacial cysts. OKCs are known for their high recurrence rates and potential for invasion into adjacent structures [1].

Previously recognized for years as the OKC in the WHO/IARC classification of head and neck pathology, this lesion was reclassified as a keratocystic odontogenic tumor (KCOT) from 2005 to 2017. It then returned to its previous nomenclature in 2017 after being reclassified as a cystic in the updated WHO/IARC classification. The OKC was reclassified as a non-neoplasm due to insufficient high quality evidence supporting its neoplastic nature, particularly regarding clonality however some pathologists still regard the OKC as a tumor based on the former classification, which has led to ongoing debate within the Head and Neck pathology community [2].

Maxillary OKCs are extremely rare with less than 1% cases documented. The present case study describes treatment and outcome of a solitary OKC in the posterior maxilla and highlights the value of interdisciplinary teamwork in diagnosing rare pathologies and achieving the optimal results [3].KCOTs are thought to originate from residual dental lamina or the basal layer of the oral epithelium. They exhibit a slight male predominance and typically present with a bimodal age distribution. The posterior mandible and ramus are the most common sites of involvement, while involvement of the anterior maxilla, Upper 3rd molar region, and maxillary sinus is uncommon [4].

Due to their non-specific clinical and radiographic features,OKC's may be mistaken for common cysts, potentially resulting in underdiagnosis and inadequate treatment, which can lead to unnecessary recurrences. Therefore, accurate diagnosis, appropriate surgical management, and long term follow-up are essential for a successful therapy [2].

CASE REPORT

A 35 years old male reported to the Dept of OMFS with complaints of pain and swelling in the right maxillary posterior region for the past 5 months. He had no deleterious habits and his family history and general health conditions were non-contributory.The patient had consulted a local dentist 3 months prior for simillar complaints of pain and swelling in the right posterior maxilla. An Intra oral periapical radiograph revealed a periapical infection associated with the right first and second molar. He was prescribed antibiotic and analgesic; However, the pain and discomfort persisted. Due to unresolved symptoms ,the patient was referred to our center for further evaluation and management.

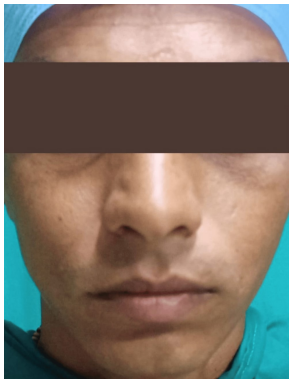
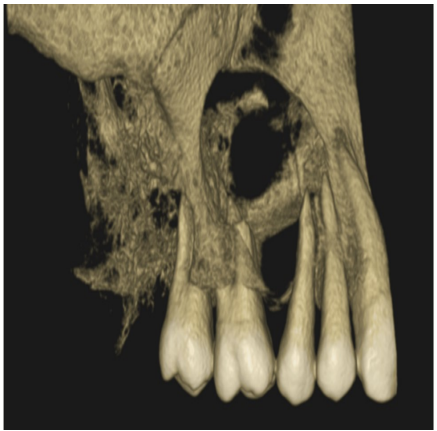


Figure 1: Preoperative profile of the patient : a 35-year-old male showing prominent swelling on the right side of the face.

Extraoral examination revealed moderate to severe facial asymmetry due to enlargement of zygoma, infraorbital and lateral nasal regions. Redness and discomfort were also present on the affected side. No changes in the overlying skin were noted(Figure 1). Digital palpation showed no signs of fluctuation. Interincisal distance was adequate (45 mm). Intraorally, no soft tissue tumor or ulceration was observed. The affected vestibular region exhibited mild enlargement & tenderness upon palpation. A sinus tract was noted, discharging pale fluid with no signs of periodontal disease. The patient maintained adequate oral hygiene.Patients' blood parameters were within normal limits. Fine needle cytology (FNAC) was subsequently performed which revealed revealed pale fluid.

Radiological examination with cone beam computed tomography (CBCT) revealed a Well-defined Radiolucency (s/o- Lesion) extending from the distal surface of root of 13 to distal aspect of root of 16 (figure 2).External Root resorption was observed involving the mesiobuccal root of 16 and distal of 15. The lesion extended superoinferiorly from the floor of maxillary sinus to the crest of alveolar ridge. The radiolucent lesion measured approximately 23.1 mm superoinferiorly, 28.8mm buccopalatally, and 18.2 mm anteroposteriorly. Additionally, a mixed radiopaque and radiolucent area was seen extending from distal of 16 to beyond maxillary tuberosity, reaching upto the lateral pterygoid plate. This area measured 30.2 mmsuperoinferiorly, 26.1 mm buccopalatally and 23.7 mm anteroposteriorly with Partially obliterating maxillary sinus(figure 3).Expansion of both the buccal and palatal cortical plates was observed, resulting in thinning of the cortical bone and breach in the continuity of buccal cortical plate (figure 4).

Well-defined radiolucent lesion was observed extending from distal surface of root of 13 to distal aspect of root of 16, suggestive of cystic lesion. Additionally, a mixed radiopaque and radiolucent lesion was noted extending from distal aspect of 16 to beyond maxillary tuberosity, reaching upto lateral pterygoid plate, suggestive of a Fibro-osseous



lesion.

Figure 2: Three Dimensional reconstruction showing radiographic evaluation of cystic lesion in posterior maxilla.

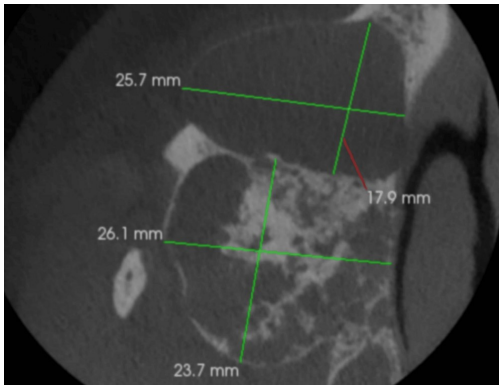


Figure 3: CBCT image showing axial dimensions of the cystic lesion with mixed radiolucent and radiopaque lesion in tuberosity suggestive of Fibro-osseous lesion.

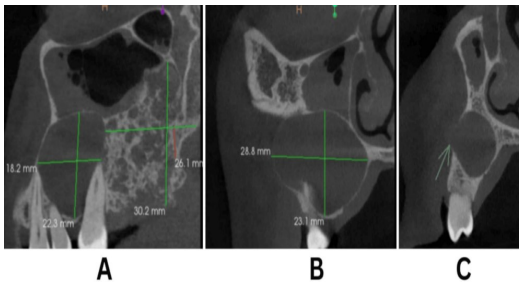


Figure 4: CBCT images showing Saggital and Coronal sections of the lesion.

A- Saggital section showing superoinferior and anteroposterior dimensions of cystic and suspected fibro-osseous lesion.
B- Coronal section showing superoinferior and mediolateral dimension of cystic lesion.
C- Coronal section showing breach in continuity of buccal cortical plate.

Based on clinical, radiographic and histopathological examination, a decision was made to perform cyst enucleation and curettage under general anaesthesia.

Under all aseptic condition, local infiltration was done in right maxillary vestibule using 2% Lignocaine with 1:80000 Adrenaline. A crevicular incision was made extending from the region of 14 to 17, along with releasing incision on either side. Full thickness mucoperiosteal flap was reflected, exposing the cystic site (figure 5).

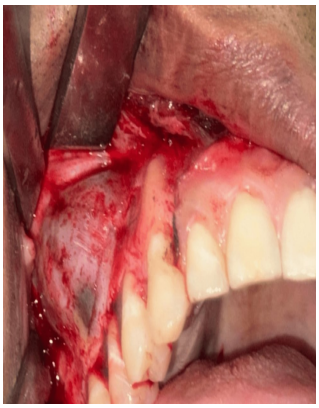


Figure 5: Exposure of the flap: Incision given, followed by exposure of the lesion.

Thin buccal cortical plate was removed to gain access to the cystic lining. Complete cystic lining was removed followed by curettage and thorough irrigation was done using 10% betadine solution and normal saline(figure 6).

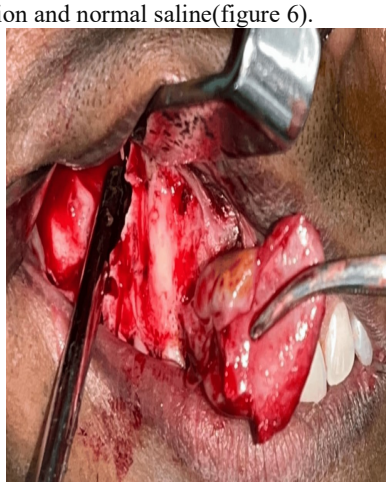


Figure 6: Enucleated cystic lesion in total followed by complete curettage of surrounding bone.

The diseased bone was removed with the help of a bone rounger. The cystic cavity was treated with hydrogen peroxide and 5 Floro-Uracil using cotton peanuts to ensure complete removal of the cystic lining. Carnoy's solution was not used in this case due to its deeper penetration in maxilla and the associated risk of injury to vital structures in close proximity. The hollow cavity was then packed with AB-

GEL, followed by closure using 3-0 resorbable sutures.(figure 7)

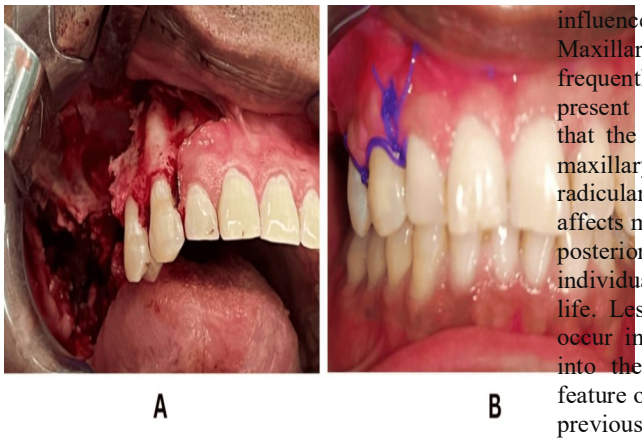


Figure 7: Post enucleation intra-oral images.
A-Diseased bone removed and surgical site treated with Hydrogen peroxide and 5-Floro Uracil for chemical cauterization.
B-Surgical site packed with resorbable gel foam followed by closure with bioresorbable sutures.

The excised lesion was sent for histopathological examination, which revealed an ODONTOGENIC KERATOCYST OF POSTERIOR MAXILLA. The patient was advised for regular follow-up at 1 week, 3 weeks and 3 months follow up to monitor for any recurrence(figure 8).

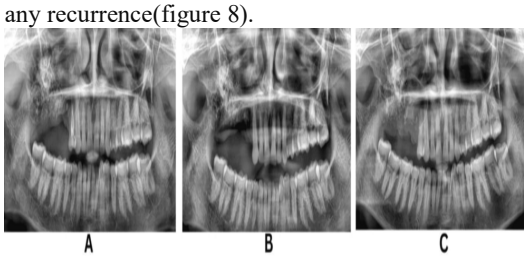


Figure 8: Radiographic images showing post-operative follow up.
A-1 month Postoperative follow-up image
B-3 months Postoperative follow-up image
C- 6 months Postoperative follow-up image

DISCUSSION

Since the odontogenic keratocyst (OKC) was identified and its histological criteria were established, this lesion has been the subject of extensive research and analysis due to its propensity for recurrence and aggressive behavior. It has been proposed that dental lamina remnants are the source of OKC. Because of its growth potential and developmental characteristics- linked to the mutation in the tumor suppressor gene, PTCH, which are present in sporadic cases and associated with basal cell nevus syndrome, some scholars argue that it should be classified as a non-malignant cystic tumour. It has also been shown that the development of OKC is influenced by intracystic fluid pressure [2].

Maxillary OKCs are uncommon and more frequently occur in the cuspid area. In the present case, radiographic imaging showed that the cyst base was located at the second maxillary molar, which is more indicative of a radicular cyst [3]. OKC's predominantly affects males, are more commonly found in the posterior mandible, and typically occur in individuals in their second and third decades of life. Less than one percentage of all OKC's occur in the upper jaw, often with extension into the maxillary sinus [5]. An intriguing feature of the cases we described is that, unlike previous reports of large OKC's likely originating in the maxillary sinus and spreading with an association to impacted teeth, the OKC's in our case was entirely contained in the maxillary alveolar bone or erupted teeth without any sinus involvement.

OKC has a somewhat unusual histological appearance compared to other odontogenic cysts. It features a well-polarized basal layer and a consistently thin cystic epithelial lining made up of 6-10 cell layers. The underlying connective tissue often tends to detach from this epithelial layer. The luminal surface is typically corrugated and exhibits para-keratinization [6]. Based on histology, OKC subtypes have been classified into parakeratotic and Orthokeratotic categories. However, Wright described the latter separately in 1981 as Orthokeratinised odontogenic cyst (OOC).The parakeratotic subtype is the most common (80%), highly recurrent, and significantly more aggressive than its counterpart. Individuals with Gorlin-Goltz syndrome have a higher likelihood of developing multiple cysts, along with anomalies in the teeth, skeleton, nervous system, and eyes. They are also more prone to recurrence. Since our patient did not exhibit any of these characteristics, further investigation was not deemed necessary [7].

The following are the histologic characteristics of the OOC: Orthokeratinized epithelium, cuboid cells with minimal tendency to polarize or palisade, less developed basal cell layer, keratin resembling that of the skin, Granular cells devoid of nuclei [8]. OKC often appears radiographically as a multilocular or unilocular radiolucency with smooth, scalloped, and typically corticated borders [9]. Since OKC involving the maxillary antrum is rare, a simple radiographic image may be misleading in such cases. Computed tomography (CT) can aid in diagnosis and preoperative planning by

revealing the full extent of these lesions [10]. Numerous cystic and neoplastic lesions, including dentigerous cysts, which present as a unilocular radiolucency associated with the crown of an impacted tooth and well-defined sclerotic margins- should be considered as the differential diagnosis for OKC. Although infection can change these characteristics [11]. Radiographically, OOC typically presents as a clearly defined, solitary or multilocular radiolucent lesion and is infrequently associated with an impacted tooth [12]. Other radiographic differential diagnosis include: Developmental lateral cyst, periapical cysts, Ameloblastoma, AOT, osseous cysts, CGCG, AV malformations, fibro-osseous lesions. Hence, microscopic examination is necessary for definitive diagnosis [13].

Based on the patients age, position and extent of pathology, various therapeutic modalities have been proposed, including Partsch I and II procedures, enucleation with carnoy's solution or cryotherapy, marginal, or segmental resection [9-14]. However, the optimal treatment apporoach remains a topic of debate. The use of Carnoy's solution which penetrates bone to a depth of approximately 1.54 mm [15]- is not recommended in cases involving the maxillary antrum due to the risk of damage to surrounding structures [16]. Resection may lead to operative complications and injury to adjacent anatomical features [17]. For complete removal of expansive pathologies in the upper jaw, particularly those involving delicate anatomical structures such as the antrum, nasal cavity, and orbital floor, radical approach like Weber-Ferguson incision, has been recommended. [18].Carnoy's solution, a mixture of chloroform, absolute ethanol, glacial acetic acid and ferric chloride has been suggested as an adjuvant therapy following marsupialization to address the issue of recurrence. However, A systematic review evaluating its effectiveness in the management of OKC's categorized the evidence as grade C [19]. In this case,Carnoy's solution was not used due to the potential risk of injury to adjacent nerves and vessels. In the event of an OKC recurrence, Carnoy's solution may be considered to reduce the need for another surgery. Radical approach involving resection and reconstruction was not recommended due to the patient's age and the low risk of malignant transformation.

Recurrence is common and usually occurs within the first five years following surgery; however, it can also happen after much longer intervals. Depending upon type of therapeutic modality, and the duration of follow-up, the reported recurrence rate range from 0% to around 62% [2,20].Recurrence rates are lower for resection and enucleation with bone curettage, and higher with simple enucleation alone [20] Since OKC's associated with teeth tend to have higher recurrence rates, special care should be given to the dentate region when enucleation is chosen as the treatment method [2,20].

CONCLUSION

OKCs are uncommon in the maxillary antrum, especially when associated with impacted maxillary third molars. Due to their aggressive nature, high recurrence rate, and similarity to other jaw cysts particularly Dentigerous cysts they should be included in possible diagnoses of maxillary radiolucencies. Accurate diagnosis relies on clinical, radiographic, surgical, and histopathological findings. Given diagnostic uncertainty, treatment should be guided by the worst-case prognosis. Conservative approaches like decompression and irrigation have become more common, but they carry a higher risk of recurrence, making long-term follow-up ideally for at least five years essential. Detecting recurrence in the maxillary antrum post-surgery remains particularly challenging.

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