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Odontogenic Keratocyst: A Case Series

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Authors' contributions

This work was carried out in collaboration between all authors. Author RK designed the study, performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript. All authors managed the analyses of the study. Author BS managed the literature searches. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

The maxillofacial region is affected by a more significant number of cysts than any other parts of the body. Odontogenic cysts are rare entities that appear in tooth-bearing areas. In this study, three cases of odontogenic keratocyst were collected over a period of 3 months in the year 2016. Clinical picture of these cases with regarding age, sex, anatomical distribution, radiological features, clinical and histological features were highlighted.

Keywords: OKC; KCOT; oral cancer; oral cavity; tumors.

1. INTRODUCTION

A cyst is defined as a pathological cavity having fluid, semi-fluid, or gaseous contents and which is not created by an accumulation of pus—Kramer 1974. Odontogenic cysts are the most common form of cystic lesions that affect the maxillofacial region. Odontogenic cysts originated from tooth germ structures (rests of Serres, epithelial rests of Malassez) and

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classified according to the stages of odontogenesis at which they occur. OKC is one of the rare developmental types of cyst classified apart from the dentigerous cyst. Odontogenic keratocyst tumour can develop during the first three stages of odontogenesis, that is, initial dental lamina development, cap and bell stages of enamel organ development, or maturation of epithelial and mesenchymal components. Keratocystic odontogenic tumour (KCOT) comprises a unique pathological entity characterised by aggressive or destructive behavior and propensity to recurrence. Usage of the term odontogenic keratocyst (OKC) had been under a lot of dispute from the time it was introduced in 1956. The World Health Organization (WHO) reclassified this lesion in 2005 as a KCOT and defined it as "a benign uni- or multicystic intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potentially aggressive, infiltrative behaviour and again renamed as odontogenic keratocyst. In this article we will be discussing a series of 3 cases of OKC with their clinical, radiological, histopathological features with various treatment modalities. Proper informed consent has been obtained from the patients [1,2,3,4,5,6].

2. CASE REPORT

2.1 Case 1

A 21 years male patient came with complaints of swelling and pus discharge in left mandible for the past 2 month. Swelling was initially smaller in size but it slowly increased in size to reach the present state. The patient experienced sour taste from pus discharge. No history of loss of sensation. On inspection, an intraoral swelling was evident relating to tooth #37 and measuring 0.5 cm x 1 cm in diameter. The colour of the overlying mucosa was normal. On palpation, afirm in consistency was felt. The panoramic view revealed a radiolucency measuring 2x3 cm in diameter and approaching the mandibular canal surrounded by radiopaque border. Excision was done and specimen was sent for histopathologic examination. Histopathological specimen reveals hyperchromatic nuclei with palisaded basal layer. Keratin deposits were observed in the cystic lumen. The picture was suggestive of OKC.

2.2 Case 2

20 year old female patient presented to our department Complaints of Painful swelling in left lower back tooth region for past two months. A patient was apparently normal before two months, then she noticed swelling in same region which was initially smaller in size gradually increased to attain the present state. Swelling was associated with pain which was of sharp pricking type, intermittent in nature, aggravates on consuming hot and cold beverages and relieved on hot fermentation. No extraoral swelling evident. On intraoral examination diffuse swelling was evident in relation to 34,35,36 which extends anteriorly from the distal aspect of 34 to the mesial aspect of 36 posteriorly. Inferiorly 2 mm short of buccal vestibule and superiorly marginal gingiva. Color of the swelling blends with normal mucosa. On palpation swelling was hard in consistency, and tender.opg reveals unilocular radiolucent lesion evident in the periapical region of 35 2X 2 approx extending from distal aspect of 34 to mesial aspect of 36 surrounded by hyperostotic border. The enuclunation was done along with tooth extraction. Histopathological examination specimen reveals of Odontogenic Keratocyst.

2.3 Case 3

16 year old male patient reported with a Complaints of swelling in the right cheek region. Also nasal obstruction and numbness in the upper alveolar region for 6 months. On extraoral examination diffuse swelling evident on the right cheek region which was 6 × 3 cm in size with ill defined borders. On palpation it was firm in consistency ,no warmth and non-tender. Intraorally a small swelling of size 1.5 × 1.0 cm present in the vestibule extending from mesial aspect of 21 to distal aspect of 15 and swelling of size 3 × 2 cm extending to the hard palate. OPG reveals a large cystic lesion with an erosion of anterior and medial wall and floor of the right maxilla concerning root of second premolar tooth. Para Nasal Sinus view reveals right maxillary haziness. Anterior rhinoscopy shows deviation of nasal septum towards left side and pushing of lateral nasal wall medially. FNAC—few macrophages seen. A combined endonasal and Caldwell-luc approach was done and cystic lesion removed. Biopsy sent for HPE and it reveals OKC.

3. DISCUSSION

The first description of OKC was published by Philipsen in 1956 for all the cysts that showed keratinization histological. Pindborg and Hansen (1963) suggested the histological criteria, which were confirmed by Browne in 1970 and 1971.

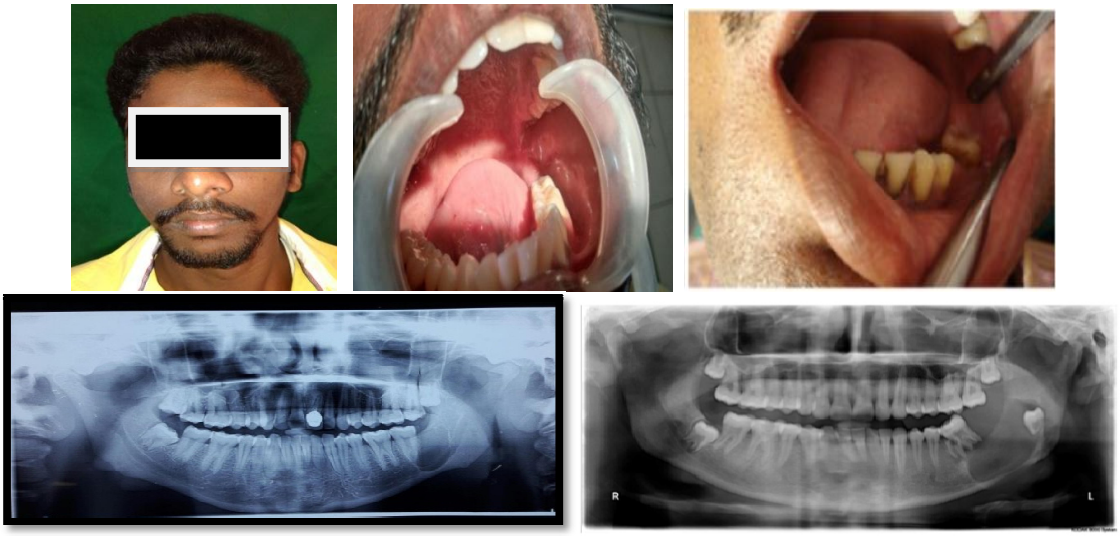

Fig. 1. Profile view; Figs. 2 and 3. Intraoral view of left buccal mucosa; Fig. 4. OPG

Toller (1967) suggested that the OKC should best be regarded as a benign cystic neoplasm, rather than an odontogenic cyst. In 1992, WHO reported that OKC was the preferred terminology for cysts with keratinized lining. In 2005, WHO reclassified intraosseous parakeratinized variant as a tumor. Cystic jaw lesions that are lined by orthokeratinized epithelium, therefore, do not form part of spectrum of KCOT, which remains a subject of controversy and is now recognized as an entirely different group of lesions termed orthokeratinized odontogenic cyst (OOC) [7].

There are few factors which led to re-characterization of the keratocyst as KCOT.

1. The KCOT exhibits locally destructive and highly recurrent behavior.
2. KCOTs are characterized by parakeratinized epithelium, in contrast to the orthokeratinized
   Variant seen in OOC. KCOT reveals budding of the basal layer into the connective tissue and frequent mitotic figures.
3. KCOTs are associated with inactivation of *PTCH*, the tumor suppressor gene.
4. Multiple KCOTs may present as one of the stigmata of the inherited NBCCS. It is also known as Gorlin syndrome [8].

OKC arises either from dental lamina or from its remnants. However, some cysts are thought to arise from basal cell proliferation of surface epithelium. Arising from the enamel organ before dental hard tissue formation is a theory that has very little evidence for justification. Identification of mutations in *PTCH* gene which is considered to be the mutated oncosuppressor gene in NBCCS has shed new dimensions to the pathogenesis of OKC. It reveals that syndromic patients as well as a minority of sporadic lesions show mutations in *PTCH* gene leading the trend towards the neoplastic nature of the lesion [1].

Ostrofsky found 'epithelial residues' in the retromolar regions and discussed their possible relationship to the formation of KCOTs. The fact that they are consistently found in recurrent keratocyst extending in the ascending ramus and in a considerable number of primary KCOTs in the same region, warrants consideration of an origin different from the dental lamina. The reason why one or two of these microcysts or epithelial islands begin to grow is not clear. In some patients they probably remain dormant for a long period (peak incidence of OKCs in 5th to 6th decade) or will never produce clinically significant cysts [9].

Odontogenic kerato cyst more common in males than females and occurs over a wide age range and is typically diagnosed during the second to fourth decade. KCOT has a predilection for occurring in the mandible (75.58%) as compared to maxilla as reported by many studies carried out in the past. In mandible, majority of cysts occur in ramus-third molar area, followed by the first and second molar and then the anterior mandible. In maxilla, the most common site is

third molar area followed by cuspid region. In the first and second case reported here, the lesion was located in the posterior ramus and in the third case, although located in Maxillary anterior region which are considered to be unusual [3].

In a review of 256 patients by Myoung et al in 2001, 118 of 256 patients had swelling (46.1%) at first admission, while 51 patients reported with pain (19.9%), and 42 patients (16.4%) had swelling and pain simultaneously. Purulent discharge was evident in 17 patients (6.6 %), while discomfort was evident in 12 patients (4.7%) and paresthesia in two patients (0.8%). About 14 patients had no symptoms (5.5%), i.e. lesions were found accidentally during radiographs. A similar finding was seen in both the cases reported here [10].

In the first & third case report, the patient gave a presenting complain of painless swelling and detected only after it was seen on the radiograph.and in second case patient complaints of swelling since two months and pain since 1 month suggesting the possibility of secondary infection. No evidence of extraoral swelling in first & second patient [11].

On radiographic examination, the KCOT cannot be distinguished from other intrabony cysts. In the mandible, the epicenter is commonly located superior to the inferior alveolar nerve canal. It usually shows evidence of a cortical border with a scalloped outline which represents variations in the growth pattern of the cyst. An important characteristic of KCOT is its propensity to grow along the internal aspect of jaws, causing minimal expansion. Radiographically, KCOTs may present as unilocular or multilocular radiolucencies with a well-defined peripheral rim. Larger keratocystic odontogenic tumour frequently mimic other pathologic entities, such as the dentigerous cyst, lateral periodontal cyst and the ameloblastoma (multilocular). Smaller KCOTs usually appear as asymptomatic unilocular radiolucencies with corticated borders [7].

The keratocystic odontogenic tumor wall is usually rather thin unless there has been superimposed inflammation. The lining epithelium is highly characteristic and is composed of:

- A parakeratinized surface which is typically corrugated, rippled or wrinkled
- A remarkable uniformity of thickness of epithelium, usually ranging from 6 to 10 cells thick [12].
- A prominent palisaded, polarized basal cell layer of cells often described as having ‘picket fence’ or ‘tomb-stone’ appearance [13].

Numerous surgical modalities have been suggested for the treatment of KCOTs, including enucleation with primary closure, enucleation with open packing and resection with or without loss of jaw continuity. The treatment depends on several factors, such as age, location and size of lesion, and whether the lesion is primary or recurrent. Total enucleation with or without “peripheral ostectomy" is a treatment of choice for most KCOTs unless lesion is recurrent or has significantly invaded soft tissue [4].

M Young et al. and Brannon, suggested that epithelial remnants or residual tissues are ostensibly prime potentiators of recurrence and for this reason, chemical cautery after enucleation, aggressive curettage of bony walls, cryotherapy modalities, peripheral ostectomy with a bone bur or even radical resection of involved jaw have been advocated as means of treatment for lowering the recurrence by removing the epithelium. Our cases were treated by enucleation along with extraction of associated teeth. The patients have been under follow-up since last one year without any complications [10].

4. CONCLUSION

Any unilocular or multilocular lesions of either jaw irrespective of the internal structure and type of borders should include a differential diagnosis of OKC as the typical features of OKC may not always be present. Postoperative follow-up of minimum 5 years is essential following surgical management, considering the high recurrence rate of these lesions [7].

CONSENT

As per international standard or university standard, patient’s written consent has been collected and preserved by the authors.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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