Case Report

ODONTOGENIC KERATOCYST A Relapse that Refuses to Rest - A Case Report

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ABSTRACT:

Odontogenic Keratocyst (OKC) is a benign cystic neoplasm that can often be misinterpreted radiographically as a dentigerous, lateral periodontal, residual, or fissural cyst. Initially, it was thought to originate from primordial odontogenic epithelium and was therefore referred to as a primordial cyst. However, the true nature of OKC—now widely recognized as a Keratocystic Odontogenic Tumor (KCOT)—remains a matter of debate.

KEY WORDS: Keratocyst, Odontogenic Cyst, Keratocystic Odontogenic Tumor.

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INTRODUCTION:

Jaw cysts, particularly Odontogenic Keratocysts (OKCs), have long intrigued dental practitioners due to their ability to grow extensively before becoming clinically apparent. Unlike other jaw cysts, OKCs exhibit a strong tendency to recur after surgical excision. Their biological nature is still unclear, which led Philipsen in 2005 to rename them as Keratocystic Odontogenic Tumors (KCOTs). Despite this, the neoplastic potential of OKC continues to be debated among oral pathologists. The present article discusses a case of OKC, with an effort to better understand its biological behaviour [1].

CASE REPORT:

A 23-year-old male patient presented with mild pain and a progressively enlarging swelling seen in the anterior one-third region of the hard palate nearing to alveolar crest the maxilla, to our department (Figure 1). There were no specific aggravating or relieving factors for past one month. Investigations revealed a previously diagnosed and treated case of OKC performed and operated a year back. Further history indicated that he had been experiencing recurrent pain in the same area following cyst removal. Intraoral and radiographic findings of the patient showed a partially edentulous maxilla involving 12, 11, 21, 22, 23 teeth, without additional symptoms (Figure 2). Orthopantomogram scans revealed bone healing (Figure 3).

CBCT radiographic scans revealed a large expansile unilocular osteolytic lesion with ill-defined and irregular bucco-lingual cortical borders (Figure 4), extending in the regions from 11 till 23, which are seen with filled obturating material. The excised specimen, measured 3 × 2 cm, was friable, thin-walled, and contained cheesy white material.

Histopathological examination revealed a cystic cavity lined with para-keratinized stratified squamous epithelium with a corrugated surface (Figure 5). The basal cells displayed palisading, with nuclei

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Figure 1: Extra-oral appearance of the patient**.**

Figure 2:Intra-oral representation showing maxillary arch with missing teeth 12, 11, 21, 22, 23. Swelling extending 1X1 cms palatally and labially.

oriented away from the basement membrane. The epithelial-connective tissue interface was flat, occasionally showing splitting and absence of rete ridges. Capsular tissue appeared dense, with mild inflammatory infiltrates and satellite cysts noted at places. The patient was given a final diagnosis of recurrent odontogenic keratocyst. Surgical enucleation was performed successfully. A regular follow-up was done at regular intervals of 4 months for 1 year and no recurrence was reported.

DISCUSSION:

The earliest term used for OKC was *Cholesteatoma* (Hauer, 1926; Kostecka, 1929).

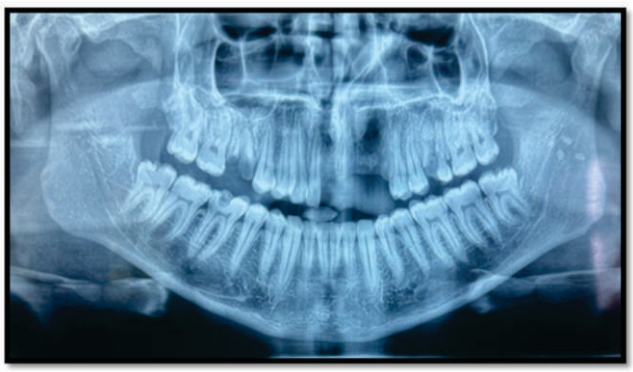

Figure 3: OPG representation of partially edentulous jaw.

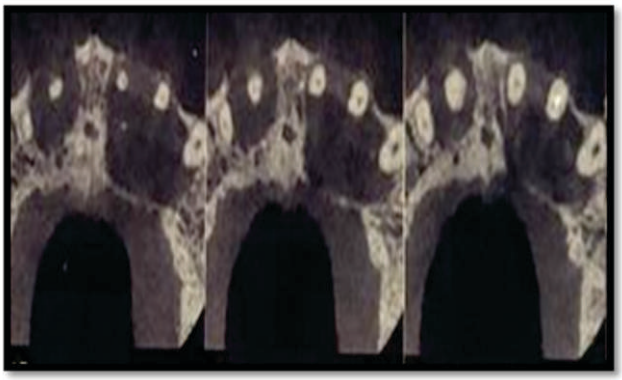

Figure 4: CBCT representation of a large osteolytic bone defect.

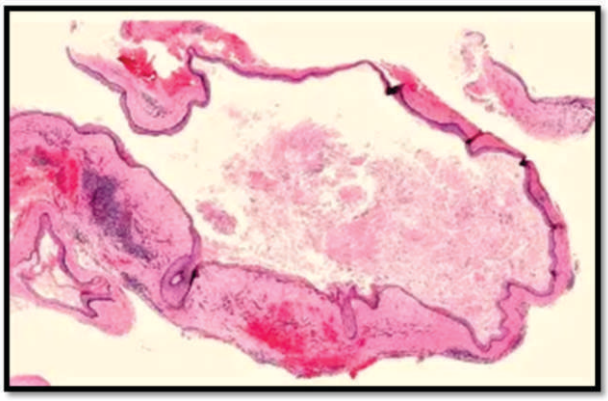

Figure 5: Histopathological findings revealing noninflamed fibrous cyst wall lined by thin uniform stratified squamous epithelium, prominent granular layer, thick luminal lamellated keratin, and cuboidal to flat basal layer.

Philipsen (1956) later introduced the term Odontogenic Keratocyst. Philipsen, Pindborg, and Hansen defined keratocysts as jaw cysts characterized by keratinization. Clinically, radiographically, and histologically, however, OKC may mimic other jaw cysts, making its classification complex[2-4].

Genetic factors are thought to play a role in its pathogenesis. OKCs are most common in the second and third decades, with a slight male predominance. The mandible, particularly the molar-ramus area, is the

most frequent site, as also observed in the present case. Symptoms may include swelling, pain, and discharge, with occasional paresthesia, although none were reported in this patient. Lesions may extend beyond 40 mm, often involving the ramus or inferior border of the mandible, while maxillary OKCs demonstrate more rapid expansion. [5]

Radiographically, OKCs may appear unilocular or multilocular, intraosseous, and often not associated with impacted teeth. Main in 1970, classified OKCs into four types: envelopmental, replacement, collateral, and extraneous. Of concern is their association with nevoid basal cell carcinoma syndrome (NBCCS), first described by Gorlin and Gotz. NBCCS is linked to mutations in the PTCH gene (chromosome 9q22.3) and presents with variable features, including multiple basal cell carcinomas, OKCs, skeletal abnormalities, and ocular manifestations[6,7].

Radiographic representation of OKC through-

OPG: They are typically seen as a solitary, radiolucent, unilocular, expansile lesion with smooth, corticated borders. These cortices are often scalloped around the roots of teeth. Three-quarters of lesions are located in the posterior mandible. When in the mandible, they typically grow along the length of the bone with minimal buccolingual expansion. In the maxilla, they expand into the maxillary sinus[8].

The appearance and location can vary. If associated with the crown of an unerupted/ impacted tooth, they can mimic a dentigerous cyst (DC). If associated with the roots of a non-vital tooth, they can mimic a radicular cyst (RC). If large enough, they will resorb the roots of adjacent teeth. They may occasionally appear septated, making the distinction from ameloblastoma difficult[9].

CT- Visualized as an expansile, cystic lesion with scalloped, well-corticated borders. Density of cystic contents varies with viscosity. Cortical breach suggests possible soft tissue involvement[9].

MRI- Odontogenic keratocysts typically demonstrate,
T1: high signal due to cholesterol and keratin contents**,**
T2: heterogeneous signal**,**
DWI/ADC: restricts due to presence of keratin**,**
T1 C+ (Gd)-peripheral enhancement, no enhancing nodular component (unlike ameloblastomas)**[10].**

The high recurrence rate of OKC is attributed to its friable epithelial lining, satellite cysts, and epithelial infoldings, making complete surgical removal challenging. Differentiating OKC from DC and orthokeratinized odontogenic cysts (OOC) is important, as OKCs exhibit more aggressive behaviour. Keratin profiling has been employed to highlight differences among these cysts**[7].** Both DC and OKC can be positioned pericoronally; but epithelial attachment of the DC tends to attach at the cemento-enamel junction of tooth. Radicular cyst is centered at the apices of non-vital teeth and does not scallop, do not exhibit septation, and is often more expandable. Ameloblastoma-tend to be multilocular, more expansile, with a textbook soap-bubble or honeycomb appearance, tendency for ameloblastoma to be more aggressive and cause more significant tooth resorption. Long-standing OKC may mimic ameloblastoma. Ameloblastic fibroma: in younger individuals, if the lesion is pericoronal to an impacted tooth. Simple bone cyst in younger individuals can be challenging to differentiate, but usually simple bone cysts exhibit minimal expansion, and spare the lamina dura as well as the periodontal ligament spaces, odontogenic keratocysts will tend to efface the lamina dura[11].

Treatment options range from enucleation and curettage to more extensive procedures such as en-bloc resection or hemimandibulectomy, depending on lesion size. Regular postoperative follow-up is essential due to the risk of recurrence. In the present case, surgical enucleation was performed, and the patient had been under follow-up for three months without signs of recurrence[5].

CONCLUSION:

OKC or KCOT, is an aggressive lesion with a high recurrence rate. Although believed to originate from dental lamina, its exact origin remains unclear. Radiographic examination is essential for OKC diagnosis, CBCT provides high-resolution, three-dimensional images of the jaw bones and surrounding tissues, particularly useful for: assessing lesion extent and relationship with adjacent structures, evaluating cortical expansion or perforation, detecting root resorption or displacement, guiding surgical planning. While surgical excision remains the treatment of choice, adjunctive techniques such as cryotherapy, decompression, and marsupialization are also being used to minimize recurrence.

Importantly, some OKCs have shown potential for malignant transformation into squamous cell carcinoma or basal cell carcinoma. Thus, recognizing the biological behaviour and prognosis of OKC is essential for effective management.

DECLARATION OF PATIENT CONSENT:

The authors certify that they have obtained all appropriate patient consent forms. In the form, the

patient(s)/guardian has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients/guardian 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.

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Conflicts of Interest
There are no conflicts of interest.

REFERENCES:

1. Philipsen HP. 'Keratocysticodontogenictumor,' in World Health Organization Classification of Tumours. Pathology and Genetics of tumours of the Head and Neck. 2005: pp.306-307
2. Nayak MT, Singh A, Singhvi A, Sharma R. Odontogenickeratocyst: What is in the name? J Nat SciBiol Med. 2013 Jul;4(2):282-5. doi: 10.4103/0976-9668.116968. PMID: 24082717; PMCID: PMC3783765.
3. Philipsen HP, Reichart PA. Classification of odontogenic tumours. A historical review. J Oral Pathol Med. 2006 Oct;35(9):525-9. doi: 10.1111/j.1600-0714.2006.00470.x. PMID: 16968232.
4. Pindborg JJ, Hansen J. Studies OnOdontogenic Cyst Epithelium. 2. Clinical and Roentgenologic Aspects Of Odontogenic Keratocysts. Acta Pathol Microbiol Scand. 1963;58:283-94. PMID: 14044666.
5. Shear M, Speight P. Cysts of the Oral and Maxillofacial Regions. 4th ed, Blackwell Munksgaard, 2007.DOI:10.1002/9780470759769
6. Main DMG. Epithelial jaw cysts: a clinic-pathological reappraisal. British Journal of Oral Surgery. 1970; 8(2): 114–25. https://www.bjoms.com/article/S0007-117X(70)80002-6/fulltext
7. Gorlin RJ, Goltz RW. Multiple nevoid basal-cell epithelioma, jaw cysts and bifid rib. A syndrome. N Engl J Med. 1960 May 5;262:908-12. doi: 10.1056/NEJM 196005052621803. PMID: 13851319.
8. Scholl RJ, Kellett HM, Neumann DP, Lurie AG. Cysts and cystic lesions of the mandible: clinical and radiologic-histopathologic review. Radiographics. 1999 Sep-Oct;19(5):1107-24. doi: 10.1148/radiographics. 19.5.g99se021107. PMID: 10489168.
9. Borghesi A, Nardi C, Giannitto C, Tironi A, Maroldi R, . DiBartolomeo F, Preda L. Odontogenickeratocyst: imaging features of a benign lesion with an aggressive behaviour. Insights Imaging. 2018 Oct;9(5):883-897. doi: 10.1007/s13244-018-0644-z. Epub 2018 Jul 31. PMID: 30066143; PMCID: PMC6206371.
10. Mahmood F. Mafee, Galdino E. Valvassori, Minerva Becker. Imaging of the Head and Neck. (2005) ISBN: 9781588900098
11. Gaillard F, Ranchod A, Yap J, et al. Odontogenic-keratocyst. Reference article, Radiopaedia. org (Accessed on 17 Sep 2025) https://doi.org/10.53347/rID-1762