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RECURRENT ODONTOGENIC KERATOCYST. A CASE REPORT

Mahendra R. Patait[1]*, Shriya R. Maknikar[2], Ashwin Thakre[3], Kartikee V. Bhoir[4], Vidya Rodge[5]

[1]*HOD, Department of OMDR, SMBT dental college and hospital
[2]3rd year post graduate, Department of OMDR, SMBT dental college and hospital
[3]Reader, Department of Oral and Maxillofacial surgery. SMBT dental college and hospital
[4]3rd year post graduate, Department of OMDR, SMBT dental college and hospital
[5]2nd year post graduate, Department of Oral Pathology, SMBT dental college and hospital,

***Corresponding Author:** Prof. Dr. Mahendra R. Patait (BDS, MDS, PHD, MBA)
*HOD, Department of OMDR, SMBT dental college and hospital

Abstract
Odontogenic keratocysts (OKCs) are benign cystic lesions originating from the dental lamina remnants, predominantly localized in the mandible's ramus and posterior body. Despite their benign nature, OKCs pose diagnostic and management challenges due to their potential for rapid growth and high recurrence rates. In this report, we present a case study of a 25-year-old male patient in whom a well-defined radiolucency with sclerotic borders was incidentally detected during pantomogram imaging. Subsequent histological examination and biopsy of the cyst wall revealed characteristic features consistent with mandibular body OKC, characterized by a cyst lined with parakeratinized stratified squamous epithelium. This case highlights the importance of meticulous radiographic assessment and histopathological analysis for accurate diagnosis and appropriate management of OKCs. Our findings contribute to the understanding and clinical management of this challenging entity in oral pathology.

Key Words: Odontogenic keratocysts, (OKCs), benign cystic lesions ,Cone Beam CT

INTRODUCTION

Odontogenic keratocyst (OKC), as classified by the World Health Organisation (WHO), is characterized as a benign uni- or multicystic intraosseous tumor of odontogenic origin, featuring a characteristic lining of parakeratinized stratified squamous epithelium and demonstrating potential for aggressive, infiltrative behaviour (WHO, [1]). Among odontogenic cysts, OKC represents the most prevalent type, characterized by its aggressive benign nature, cystic formation, and odontogenic origin, devoid of inflammatory features, while predominantly developing intraosseously. OKC typically exhibits a slow growth pattern, along with infiltrative biological behaviour, bone resorption capability, tooth displacement, and a notable recurrence rate post-surgical intervention. Clinically, OKC often presents as an expansible, solitary, lucent lesion with a smooth and frequently scalloped border, predominantly arising around the crown of the third molar in the posterior jaw, potentially leading to malocclusion through adjacent tooth displacement [2].

The prevalence of OKC accounts for approximately 7.8% of all jaw cysts, with incidence rates varying between 4% and 16.5%. Although OKC may affect individuals across all age groups, its highest

incidence is observed in the second and fourth decades of life. Furthermore, males exhibit a higher susceptibility to OKC, with a male-to-female ratio of 1.5:1, and it is more commonly observed in the White population. Mandibular localization is predominant, accounting for 66.9% of cases, occurring twice as frequently as in the maxilla. OKC typically manifests at the mandibular angle and ascends towards the root region, with a prevalence ranging between 69% and 83%. [3] Notably, it can infiltrate the mandibular body and ascending ramus, often adjacent to the mandibular third molar. However, solitary lesions are more common, unless in association with Gorlin-Goltz syndrome [4]

Patient Report

A 25-year-old patient had reported to the Out-Patient Department of the Department of Oral Medicine and Radiology with the chief complaint of dirty deposits on his teeth and he wanted to get his oral prophylaxis done.

The medical history and family history were non-contributory. The patient gives a history of enucleation of cysts in the mandibular anterior region 7 years ago. He also gives a history of surgical extraction with mandibular left posterior teeth 7 years ago under Local anaesthesia and without any complications or drug history. On Clinical examination, no relevant abnormality was detected. Intraoral examination revealed pit and fissure caries with 47, (Fig. 2) grossly decayed with 44, missing teeth with 34 and 36, and Miller class II recession with 33. Lymph nodes were non-palpable. Oral hygiene was fair with Grade I stains and calculus. The patient was referred to the Department of Periodontology for oral prophylaxis where he was advised Implant with 44, 34, 36 and an Orthopantomogram. The orthopantomogram revealed a well-defined radiolucency around the horizontally impacted 48. (Fig. 3)

INVESTIGATIONS

Panoramic radiographs unveiled a well-defined radiolucency enveloping the horizontally impacted tooth 48, with distinct sclerotic borders, extending from the distal aspect of tooth 47 to encompass approximately two-thirds of the mandibular ramus.(Fig. 3)

To meticulously delineate the lesion's extent and characteristics, cone beam computed tomographic (CBCT) scans were advised. The CBCT imaging revealed a substantial radiolucent lesion measuring approximately 23.56 mm in width, 9.93 mm in height, and 25.4 mm in depth extending from the periapical region of 48 to the upper part of the ramus. Perforation is not noted, but thinning of the lingual cortical plate can be seen (Fig. 4, Fig. 5, Fig. 6)

In tandem with radiographic evaluations, routine laboratory investigations were conducted, all yielding results within normal limits. Following the radiographic assessments, an incisional biopsy was meticulously performed to procure tissue samples for subsequent histopathological scrutiny.

Histopathological scrutiny of the biopsy specimen elucidated a cystic lumen peripherally lined by parakeratinized stratified squamous epithelium, characterized by corrugations. The basal layer of the epithelium showcased a palisaded arrangement accentuated by hyperchromatic nuclei. Furthermore, the connective tissue capsule encompassing the cyst evinced diffusely distributed moderate amounts of chronic inflammatory infiltrate and variably sized blood capillaries.

This integrative approach, amalgamating radiographic and histopathological findings, robustly substantiated the diagnosis of Odontogenic Keratocyst (OKC).

The following findings are highly suggestive of the diagnosis of OKC: (I) a cyst like radiolucency in the third molar region or mandibular ramus; (2) a diameter of more than 3 cm; (3) a unilocular cyst like radiolucency with scalloped margins; (4) a multilocular cyst; and (5) odourless, creamy or caseous contents [5]

TREATMENT

The patient was advised for surgical excision and biopsy (Fig. 7 Fig. 8). Careful enucleation of cyst was performed along with the extraction of 48 under local anaesthesia. Excised tissue was sent for histopathological investigation. Necessary prescriptions and postoperative instructions were given.

Discussion:

The characterization of odontogenic cysts featuring a lining of keratinized stratified squamous epithelium was initially documented in 1876 and later coined as Odontogenic Keratocysts (OKCs) by Phillipsen in 1956 [6]. The term "Odontogenic Keratocyst" was officially introduced by the World Health Organization (WHO) in 1992, referring to benign cysts of odontogenic origin exhibiting a specific histological appearance [7]. However, due to several factors including a high recurrence rate, aggressive clinical behaviour, mutations in the protein patched homolog tumor suppressor gene, the presence of satellite cysts, and its association with Gorlin-Goltz syndrome, this entity was reclassified as a benign keratocystic odontogenic tumor (KCOT) in the WHO classification of head and neck tumors in 2005 [1]

Subsequent revisions in the WHO classification saw a reversion of KCOT back to the cyst category under the designation OKC in the fourth edition, and it remains classified as such in the current fifth edition due to insufficient evidence to classify it as a neoplastic lesion .[8]

Differential Diagnosis

The diagnosis of OKC primarily relies on examining histopathological features. Typically, it presents with a delicate, easily breakable wall that can be challenging to remove from the bone intact. Inside the cyst, there may be a clear fluid resembling serum transudate or a cheesy substance. The thin fibrous wall generally lacks inflammation. Occasionally, small satellite cysts or clusters of odontogenic epithelium are visible within the fibrous wall. [9][10] Histologically, OKC may exhibit features reminiscent of myxoma, ameloblastoma, central giant cell granuloma, or other odontogenic cysts. Radiographically, OKC findings may resemble those of various other cysts or lesions, such as dentigerous cysts, residual cysts, or lateral periodontal cysts. OKC in the anterior midline of the maxillary region might be confused with nasopalatine duct cysts. Peripheral OKC within the gingival soft tissues is rare.[11]

The recurrence rate of OKC is reported to range between 7% and 28% [12]. To mitigate this high recurrence rate, surgical interventions have evolved over the years, with recommendations including marginal resection akin to the approach employed for unicystic ameloblastoma since the 1980s (18). Various surgical procedures have been proposed, including enucleation, enucleation combined with adjuvant therapies such as curettage, Carnoy's solution, cryotherapy, or peripheral ostectomy with Carnoy's solution, marsupialization, resection, or combinations thereof .

Histologically, the epithelial lining of OKCs is typically thin and homogeneous, with minimal to absent rete ridges [13] The basal cell layer exhibits a distinct palisading pattern of columnar or cuboidal cells, often transitioning directly into the thin stratum spinosum layer with intracellular edema and fewer than eight cell layers thick.

The stratum corneum is predominantly parakeratotic, although orthokeratotic features may also be observed. Inflammatory changes within the cyst wall are variable, with inflammation occasionally leading to the presence of rete pegs without parakeratinization. The contents of the cyst lumen vary widely, ranging from keratinaceous material to blood-filled spaces.

In conclusion, the diagnostic and therapeutic management of OKCs remains multifaceted, necessitating a comprehensive understanding of its histopathological characteristics, clinical

behaviour, and treatment modalities. Further research into the molecular mechanisms underlying its aggressive nature and recurrence propensity is warranted to optimize patient outcomes and minimize the burden associated with this enigmatic odontogenic entity.

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IMAGE GALLERY

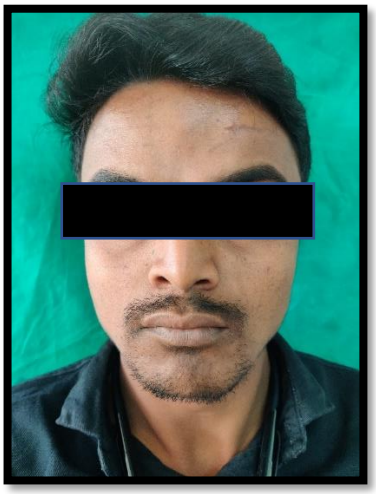

Fig 1

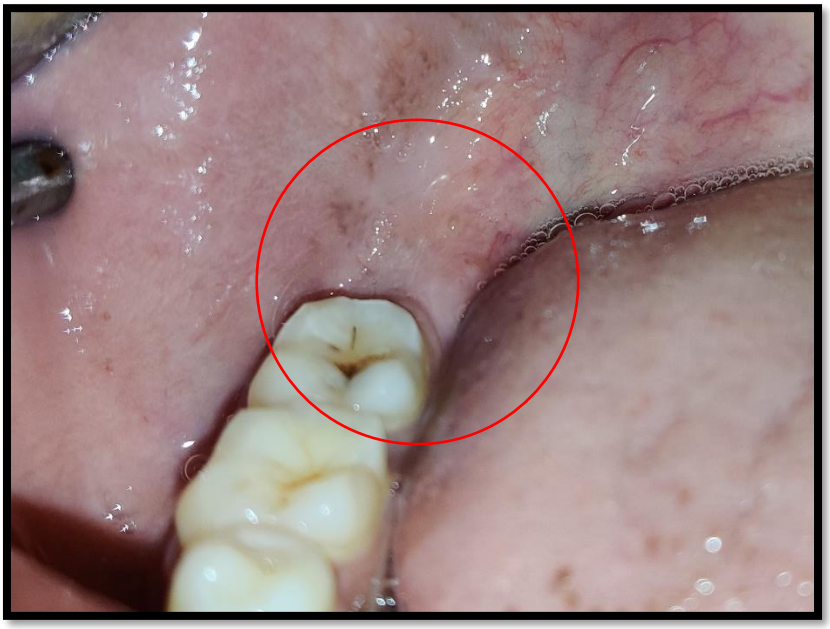

Fig 2 :- Intra oral presentation of 48 region showing no significant clinical Findings

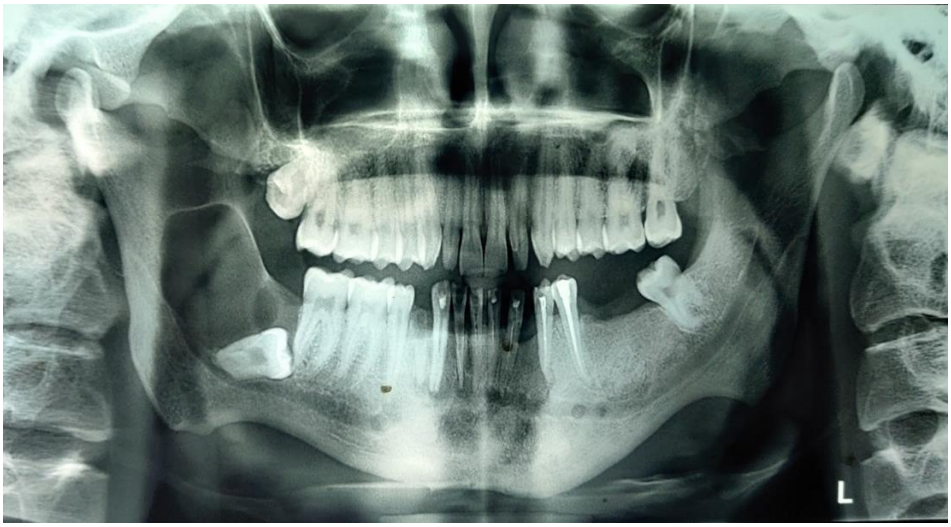

Fig 3 Orthopantomogram depicting impacted 48 enclosed with a well-defined radiolucency extending till ramus.

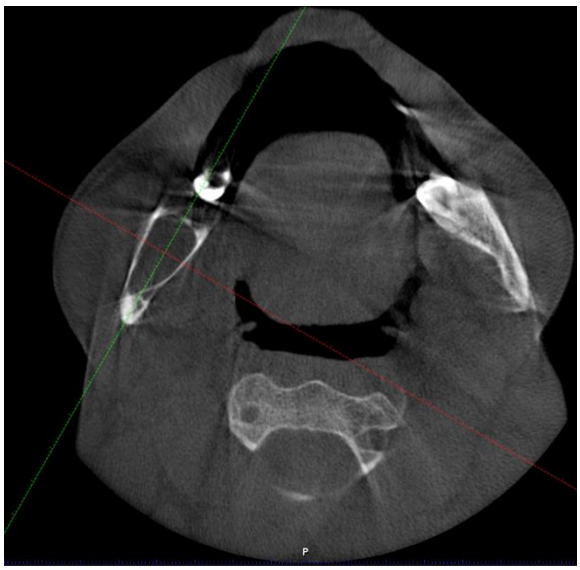

Fig. 4

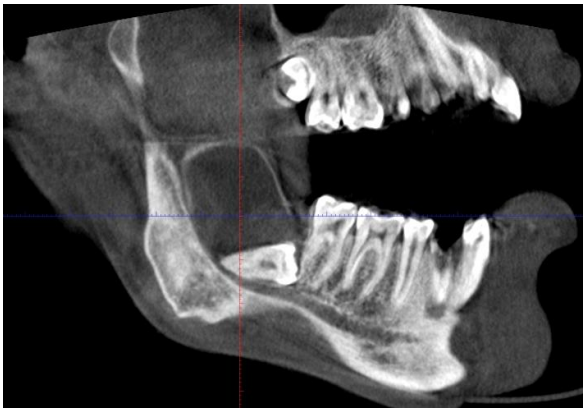

Fig. 5

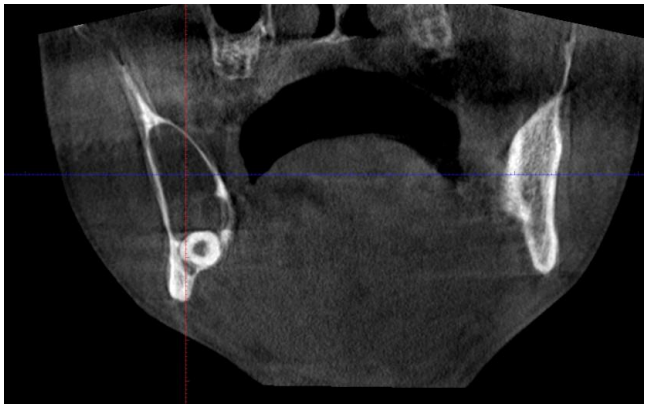

Fig. 6

Fig. 4, Fig 5, Fig 6: Depicting the cone beam computed images in the axial, sagittal and coronal sections respectively showing well defined radiolucency with an impacted 48

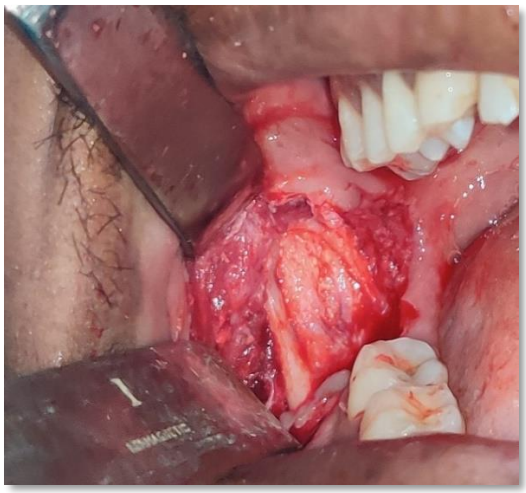

Fig. 7 The surgical site

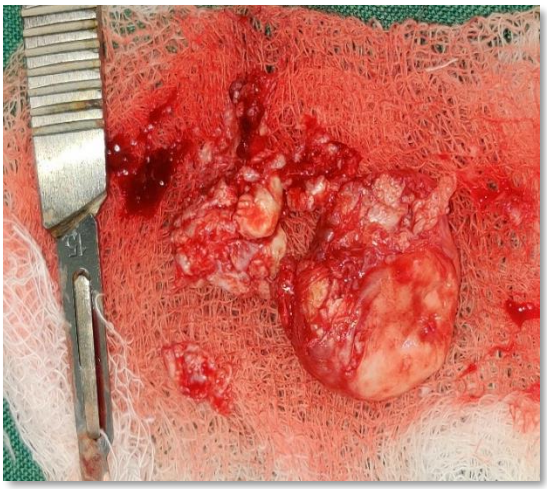

Fig. 8 The biopsy specimen