

A Case Report on Odontogenic Keratocyst of Right Mandible

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Abstract:- Odontogenic keratocysts (OKCs) are rare, aggressive cystic lesions originating from the dental lamina remnants and basal cells of the oral epithelium. Despite their typically asymptomatic presentation, OKCs may be incidentally discovered on dental radiographs or present with symptoms such as pain or swelling due to infection or bone expansion. This case report describes a 21-year-old male who presented with pain in the lower right molar region. This case highlights the diagnostic challenge of OKCs, which can mimic other odontogenic pathologies and underscores the importance of radiographic assessment for early identification and management of such lesions.

Keywords:- Odontogenic Keratocyst, Odontogenic Tumour, CBCT, MRI.

I. INTRODUCTION

The term "odontogenic keratocyst" was introduced by Philipsen in 1956, and in 1963, Pindborg and Hansen described its essential features. It is called a keratocyst because the cyst epithelium produces an abundance of keratin that fills the cyst lumen. Additionally, characteristics of the odontogenic keratocyst include flattening of the basement membrane and palisading of the basal epithelial cells, which resemble odontogenic epithelium. Toller (1967) proposed that OKC could be considered benign cystic neoplasms [1]. In 2005, the World Health Organization reclassified the odontogenic keratocyst (OKC) as a benign odontogenic tumor. This entity was renamed the keratocystic odontogenic tumor (KCOT). The rationale for this reclassification included its aggressive growth, tendency for recurrence after treatment, and, notably, mutations in the PTCH gene (protein patched homolog) [2].

OKC originates from remnants of the dental lamina and basal cells of the overlying epithelium. Although it is a rare cyst that accounts for about 19% of jaw cysts, it is often asymptomatic and may be discovered incidentally on dental radiographs. Symptoms, if they occur, are typically due to infection or bone expansion [3]. In this report, a case of large OKC of the mandible is described.

II. CASE REPORT

A 21-year male patient reported to Department of oral medicine and radiology with the complaint of pain in lower right back tooth region for 5 days. Pain was sudden in onset, dull aching, continuous, non-radiating, aggravated on opening mouth and relieved on medication. No associated swelling, history of fever or trauma, or any discharge reported. Patient's medical and past dental histories were non-contributory. Patient was moderately built and nourished with vital signs within normal limits.

Extra-orally no gross facial asymmetry noted, overlying skin appeared normal with no erythema or ulcerations [Figure 1]. Mouth opening appeared to be restricted to 25mm. On palpation there was no tenderness or local rise in temperature. Intra-orally on inspection, 48 was seen missing, overlying mucosa appeared normal [Figure 2]. On palpation retromolar region was tender, soft in consistency with discharge noted. Based on clinical manifestations, provisional diagnosis of Impacted teeth w.r.t 48 was given.



Fig 1: Shows Extraoral View with no Gross Facial Asymmetry



Fig 2: Shows Missing 48 with Normal Overlying Mucosa

Radiographic investigations like orthopantomogram (OPG) revealed the long axis of 48 is distally inclined to the long axis of 47. It also revealed a well-defined multilocular radiolucent lesion on right side of mandible of size approximately (25x10) mm with corticated borders , extending superiorly from sigmoid notch till the level of angle of the mandible inferiorly, antero- posteriorly from anterior aspect to mid portion of ramus [Figure 3]. The inferior most aspect of the radiolucent lesion appears to be in close approximation with roof of inferior alveolar canal.



Fig 3: Shows Impacted 48 with Multilocular Radiolucent Lesion

Cone Beam Computed Tomography (CBCT) revealed a well-defined multilocular radiolucentlesion on right mandible extending 1.5cm posteriorly from anterior border of ramus. Mild bucco- lingual expansion noted. Lingual cortical plate destruction and thinning of buccal cortical plate noted w.r.t 48 region [Figure 5]. Radiographic diagnosis of dentigerous cyst was given.

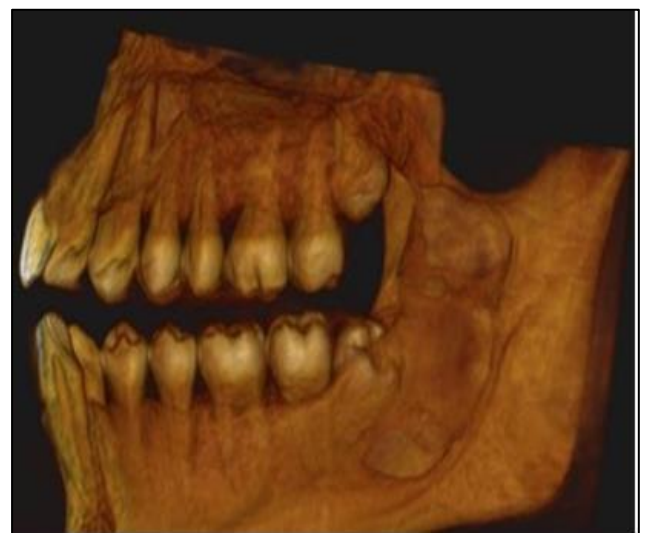


Fig 4: Shows Bone Destruction



Fig 5: Shows Thinning of Buccal and Destruction of Lingual Cortical Plates

On chair side investigation, Fine Needle Aspiration Cytology (FNAC) yielded cheesy material admixed with blood. A cyst enucleation was done followed by extraction of 48 and biopsy of the lesion was taken. On histopathological examination, a cystic cavity lined by parakeratinized stratified squamous epithelium and connective tissue capsule were seen. Basal layer showing palisaded columnar epithelial cells and connective tissue is fibrous with focal areas of inflammatory cells [Figure 6]. Histopathological findings were suggestive of OKC.

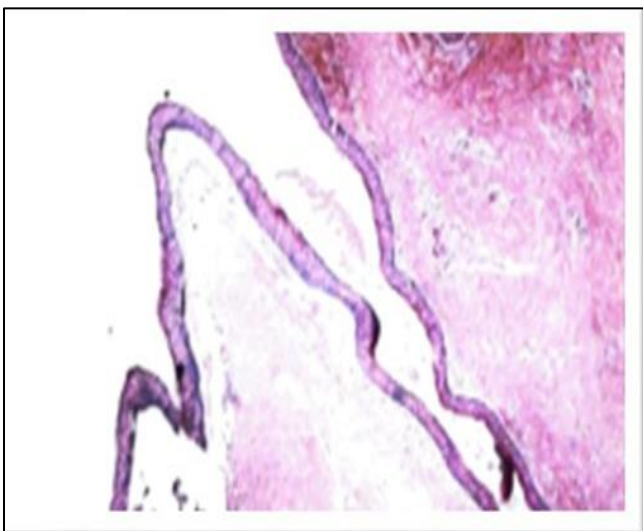


Fig 6: Shows Para Keratinized Stratified Squamous Epithelium with Palisaded Basal Layer

Hence, based on history, clinical and radiological examinations and aspiration cytology, final diagnosis of odontogenic keratocyst (OKC) was derived.

III. DISCUSSION

The term "odontogenic keratocyst" was introduced by Philipsen in 1956, and in 1963, Pindborg and Hansen outlined its key characteristics. It is called a keratocyst because the epithelium produces an abundance of keratin, which fills the cyst lumen [1]. However, it can mimic other jaw cysts in terms of clinical, radiographic, and histological features [7]. OKC represents 7.8% of all jaw cysts and can occur at any age, with peak incidence in the second and fourth decades of life. There is a tendency for it to occur more frequently in males. In the mandible, most cysts are found in the ramus. In the present case also cyst was found with impacted mandibular third molar extending into ramus. Mandibular cysts can cross the midline, while maxillary cysts may involve the sinus, nasal floor, premaxilla, and area around the maxillary third molars. Common symptoms include pain, soft tissue swelling, tooth displacement, drainage, and neurological signs such as lip or tooth paresthesia. [6] In the present patient had mild pain with no soft tissue swelling.

The presence of multiple OKCs is considered a key criterion for diagnosing Nevoid Basal Cell Carcinoma Syndrome (NBCCS). This autosomal dominant multi-systemic disorder is characterized by multiple nevoid basal cell carcinomas, multiple OKCs, palmar or plantar pits, calcifications of the falx cerebri, and skeletal abnormalities [9]. Multiple OKCs can also occur in other syndromes, including Noonan syndrome, Ehlers-Danlos syndrome, and orofacial-digital syndrome [8].

Investigations can include chairside procedures like fine needle aspiration cytology (FNAC), which typically reveals shiny straw-colored fluid, as well as biopsies (both excisional and incisional). Radiographic assessments may involve intraoral radiographs, orthopantomograms (OPG), cone beam computed tomography (CBCT), and MRI.

Radiographically, the majority of odontogenic keratocysts (OKCs) are unilocular (40%), characterized by a well-defined peripheral rim. Scalloping of the borders is a common observation, indicating variations in the cyst's growth pattern. Multilocular radiolucent OKCs (20%) can also be seen, typically presenting as a central cavity with satellite cysts, especially in the area of the third mandibular molar, where they may be mistaken for ameloblastoma on radiographs [1]. Often found in association with impacted teeth in 25-40% of cases. [6]

The types of OKCs include: (a) Replacement type: cysts that develop in place of normal teeth; (b) Extranous type: cysts that occur in the ascending ramus, away from the teeth; (c) Collateral type: cysts located adjacent to the roots of teeth; (d) Envelopmental type: cysts that surround an adjacent unerupted tooth [Figure 7]. [6]

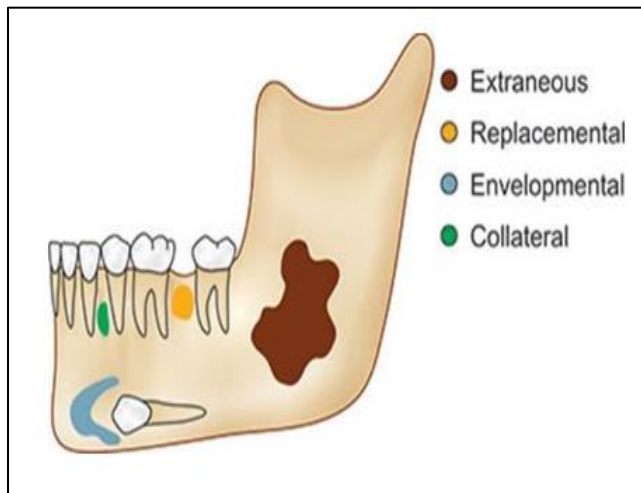


Fig 7: Types of Odontogenic Keratocyst

Histologically, the lining epithelium is distinctive, featuring a parakeratinized surface that is typically corrugated, rippled, or wrinkled. The epithelium exhibits a notable uniform thickness, usually between 6 to 10 cells. The basal layer of cells is prominently palisaded and polarized, often referred to as having a "picket fence" or "tombstone" appearance. These cysts are lined with stratified squamous epithelium, and no rete ridges are present [1]. Small satellite cysts, cords, or islands of odontogenic epithelium can be found within the fibrous wall [4]. The lumen of the keratocyst may contain a thin, straw-colored fluid or a thicker, creamy substance [1].

The treatment of OKCs is determined by factors such as the patient's age, the size and location of the cyst, soft tissue involvement, and the histological variant of the lesion. In 1985, Eyre and Zarewska outlined the following treatment options for OKC/KOT: (A) Enucleation: (a) with primary closure, (b) with packing, (c) with chemical fixation, (d) with cryosurgery; (B) Marsupialization: (a) alone or (b) followed by enucleation; (C) Resection. [5]. In the present case enucleation of cyst along with extraction of impacted 48 was done.

The recurrence rate of OKCs has ranged from 2.5% to 62%. This wide variation may be influenced by the length of the follow-up period and the treatment methods employed, particularly in patients with nevoid basal cell carcinoma syndrome [5]. While many OKCs tend to recur within five years of the initial surgery [4]. Some potential causes of recurrence include incomplete removal of the cystic lining, the thin and fragile nature of the epithelial lining, increased cell proliferative activity within the epithelium, adherence to adjacent soft tissue, and the growth of new OKCs from satellite cysts/ daughter cysts/ remnants/ or cell rests, etc. [6]. Aside from the tendency for recurrences, the overall prognosis for most odontogenic keratocysts is favorable. [4]

IV. CONCLUSION

The odontogenic keratocyst (OKC) is a unique developmental cyst that presents diagnostic and therapeutic challenges due to its aggressive behavior, high recurrence rate, and association with syndromes such as nevoid basal cell carcinoma syndrome. This case of a young male patient highlights the importance of early detection and appropriate treatment planning. Complete surgical excision, close follow-up, and consideration of histopathological features are essential for minimizing recurrence. This report underscores the necessity for clinicians to remain vigilant and adopt an individualized approach to managing OKC, particularly in younger patients, to ensure optimal outcomes.

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