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Case Report

# Odontogenic Keratocyst – A Case Report of Silent Intruder

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## Abstract
Odontogenic Keratocyst, one of the commonly encountered cyst of odontogenic origin, was introduced by Philipsen in 1956. It is of considerable importance because of its potential for aggressive clinical behavior and recurrence. It is also found in association with Nevoid Basal Cell Carcinoma Syndrome.

**Keywords:** Odontogenic Keratocyst, PTCH gene, Nevoid Basal Cell Carcinoma Syndrome, Carnoy's solution

## INTRODUCTION

The Odontogenic Keratocyst (OKC) is one of the most aggressive cyst occuring in the oral cavity. It has a tendency to extend antero-posteriorly with minimal expansion of the jaw bones.[1] Its close association with PTCH gene mutation compelled this cyst to be considered as a neoplasm leading to its redesignation as keratocystic odontogenic tumour. However the recent evidences suggest it to be otherwise hence reconsidered as a cyst in the 2017 WHO classification.[1,2] Here we present a case of Odontogenic keratocyst of the right lower jaw, with an incidental radiolucent lesion on the contralateral side.

## CASE REPORT

A 48 year old female patient reported in private clinic with a chief complaint of pain and swelling in her right lower back tooth region for the past 11 months. The swelling was initially smaller in size which gradually increased to the present size. Pain was intermittent and recurrent in nature with no history of fever and trauma. No evidence of altered sensation was noted. On extraoral examination, the swelling extended antero-posteriorly from the middle of the body of right mandible till angle of the mandible and superio- inferiorly from a point approximately 2cm from the tragus of the right ear to the inferior border of the mandible. On palpation, the swelling was tender and hard in consistency with no evidence of bleeding or pus discharge.

Orthopantomogram revealed a well-defined radiolucent lesion with a sclerotic border of size 4cm x 2.5cm (approx.) in relation to apex of 47 and 48 extending up to the lower border of the mandible (Figure 1).

Incidentally a well-defined radiolucent lesion in relation to 34, 35, 36 & 37 measuring approximately 5cm x 3cm in size extending from the mid-root level to the lower border of mandible was noticed (Figure 1). However the patient didn't have any symptoms pertaining to it.

Intra-orally 46 was missing and buccal vestibule in relation to 47 & 48 was obliterated (Figure 2). The teeth were found to be vital. A provisional diagnosis of odontogenic cyst with a differential diagnosis of odontogenic tumour was made.

Excisional biopsy under local anesthesia was done by placing a vestibular incision on the right buccal region extending from retromolar region till the first molar. A window was created with micromotor, the cystic lining was removed and freshly prepared carnoy's solution was applied over the enucleated region. Closure with 3-0 silk was done. The excised tissue specimen was greyish brown in colour, soft to firm in consistency measuring about 2.2cm x 2.0cm x 1.2 cm in size and submitted for histopathologic examination. The H& E stained section showed a lining epithelium that

was parakeratinized stratified squamous type of 6 to 8 layers with prominent corrugated surface and characteristic palisading arrangement of basal cells (Figure 3). Surrounding connective tissue capsule was composed of dense collagen bundles and abundant chronic inflammatory cells predominantly of lymphocytes and plasma cells. Based on the histopathological findings, a diagnosis of Odontogenic Keratocyst of right mandible was made. The patient was not willing for any treatment on the left side of mandible.

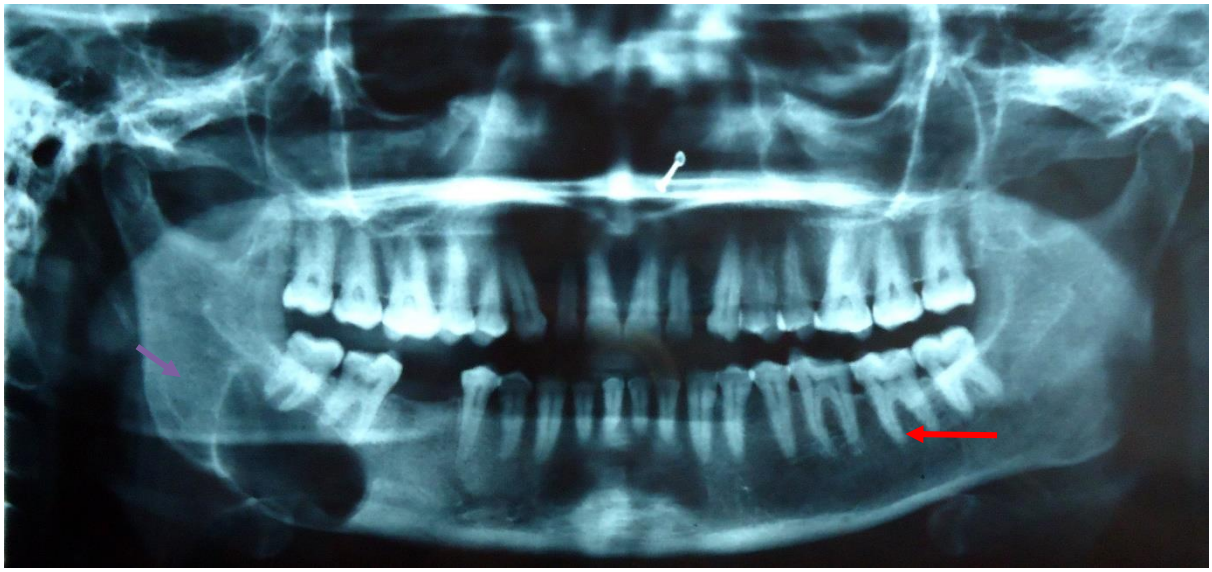

**Figure-1:** Orthopantomogram showing a well-defined unilocular radiolucency with sclerotic border in relation to 47 & 48 (Yellow colour arrow); Well-defined unilocular radiolucency in relation to 34,35,36 & 37 ( Red colour arrow).

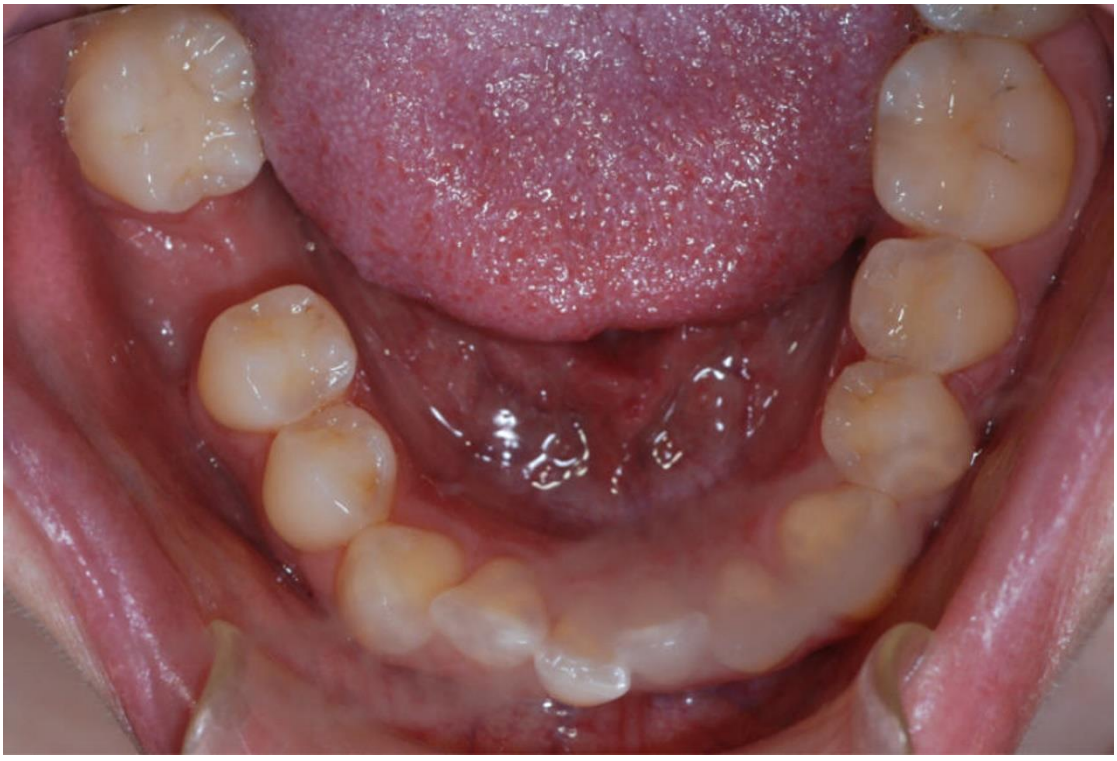

**Figure-2:** Intraoral photograph showing obliteration of buccal vestibule in relation to 47 & 48.

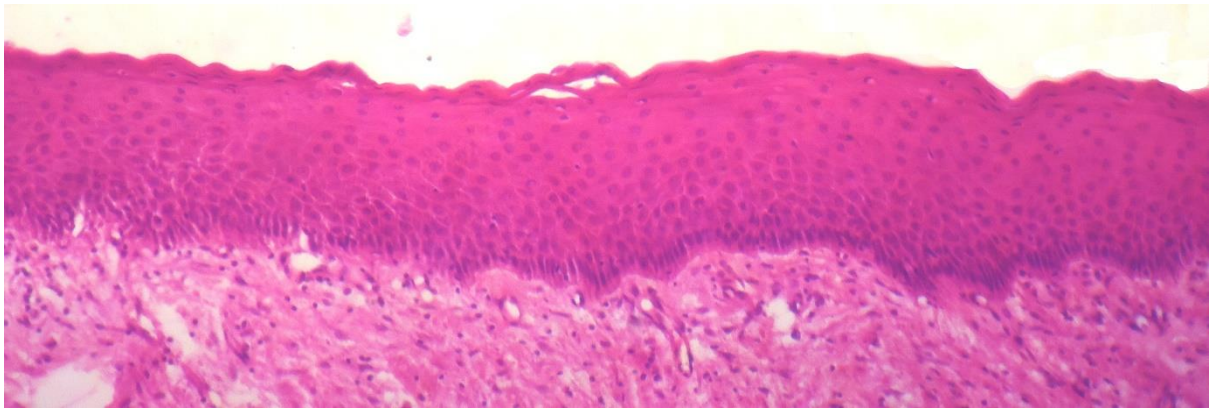

**Figure-3:** Histopathology showing parakeratinized stratified squamous epithelium with surface parakeratin corrugation and palisading basal cell layer (40 x magnifications)

# DISCUSSION

Odontogenic keratocysts (OKC) are developmental odontogenic cysts that are derived from either remnants of dental lamina or the basal cell layer of surface epithelium. It is named as keratocyst because the cystic lining produces keratin.[3] They show a bi-modal age distribution of 20-29 years and 40-59 years. Though OKC's may occur in any part of upper or lower jaw, the majority of cases occur in angle of mandible and ramus region.[3,4] OKC's occurring in maxillary region should be approached with caution due to the proximity of vital structures.[5]

OKC's can occur as single or multiple, the latter being more common in patients with Nevoid Basal Cell carcinoma syndrome (NBCCS). NBCCS (also known as Gorlin-Goltz syndrome) is an autosomal dominant multi systemic disease characterized by multiple nevoid basal cell carcinomas, multiple OKC's, palmar or plantar pits, calcification of falx cerebri and skeletal abnormalities like bifid, fused or splayed ribs.[6,7]

PTCH gene (Protein patched homolog) located on chromosome 9q22.3-q31, known for its tumour suppressor activity have been found to be mutated in both syndromic as well as sporadic cases. This lead to the controversial debate if OKC is to be considered as a tumour rather than a cyst.[6,8]

OKC's are unique in that despite its aggressive and extensive growth they often remain asymptomatic and are found as an incidental finding on radiographs. Some of the clinical manifestations include pain, swelling or drainage in symptomatic cases. Radiographically, they often present as a unilocular radiolucency with a well-developed sclerotic border with or without tooth involvement.[9]

The diagnosis of OKC is based on histopathological findings. They are characterized by a uniform epithelial layer of 6 to 10 cell thickness, prominent basal layer showing tombstone or picket fence appearance with absence of reteridges, corrugated parakeratin luminal layer. Connective tissue wall shows small islands of epithelium that may develop into small satellite or daughter cysts. Inflammatory cells are found in the connective tissue wall when the cyst is secondarily infected. Cholesterol crystals are also seen at the site of inflammation.[10]Lumen of keratocyst may be filled with clear yellow fluid to a semi solid cheesy material. The total protein concentration of the cystic fluid varies for each cyst. Toller suggested that, a total protein concentration of less than 4.0gram/100ml is indicative for OKC while total protein above 5.0gram is suggestive for radicular cyst, dentigerous cyst or fissural cyst.[11]

Orthokeratinized Odontogenic Cyst (OOC) is a rare, developmental odontogenic cyst which is considered to be a variant of Odontogenic Keratocyst (OKC) and often associated with syndromic cases. The epithelial lining of OOC lesions show 4 to 8 cell layers of thick, uniform orthokeratinized stratified squamous epithelium with a prominent granular cell layer. The basal cells are flat to cuboidal, without the palisading or polarization features. OOC has a better prognosis than OKC.[10]

Based on the size, location and extent of the lesion, the therapeutic intervention of OKC varies from enucleation, marsupialization to marginal or radical resection. Complete enucleation is often difficult because of the thin, friable

nature of the cystic lining. High recurrence rate of 30-60% observed in OKC could be due to residual cystic lining or could be a new cyst that has developed from the epithelial rests in the cystic wall.[7,8]

Carnoy's solution, a mixture of 60% ethanol, 30% chloroform, 10% glacial acetic acid and 1 gram ferric chloride, is applied into cyst cavity following enucleation to induce superficial tissue necrosis thus eliminating the residual lining.[7,12] In our case also, enucleation was followed by carnoy's solution. As the patient didn't accept for further treatment on the contralateral side, awareness was given and frequent follow up was adviced.

Due to high recurrence rate, post-operative follow up of OKC with annual radiograph is essential for atleast 5 years.[7]

# CONCLUSION

Most lesions encountered in the Oral and Maxillofacial region requires scrupulous diagnosis and treatment planning. This case report thereby reiterates the importance of thorough radiological and histopathological investigation which forms the base for successful management of the condition.

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