# KERATOCYSTIC ODONTOGENIC TUMOR – TWO CASE REPORTS AND REVIEW OF LITERATURE

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## ABSTRACT

*Odontogenic keratocyst (OKC) was initially considered to be an epithelial developmental cyst of the jaws. It is commonly found in the mandible, and can become quite large due to its rapid growth, usually single although multiple odontogenic kerato¬cysts (OKCs) can occur as a component of nevoid basal cell carcinoma syndrome (NBCCS). Currently WHO reclassified this lesion from cyst to tumour underscores its aggressive nature and should motivate clinicians to manage the disease in a correspondingly aggressive manner and recommended the term Keratocystic odontogenic tumor (KCOT) for this lesion. Two case reports of KCOT are presented. In the first case the patient was a 45 year old female and location was right posterior maxillary region and in second case (27 year old male) it was found in right posterior mandibular area. The clinical, radiological, and histopathological features of this tumour are discussed.*

**Key words:** *Odontogenic keratocyst (OKC), Keratocystic odontogenic tumor (KCOT), Nevoid basal cell carcinoma syndrome (NBCCS).*

## INTRODUCTION

Odontogenic keratocysts (OKCs) are developmental odontogenic cysts of epithelial origin, first identified and described in 1876 and further characterized by Phillipsen in 1956.[1] The odontogenic keratocyst (OKC) is now designated by the World Health Organization (WHO) as a keratocystic odontogenic tumour (KCOT) and is defined as "a benign uni- or multicystic, intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behaviour.[2]

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## CASE 1

A 45 years old female patient reported to the Department of Oral Medicine and Radiology with the swelling on the right side of the face since two months. The past medical and dental history was unremarkable. On clinical examination intra-oral swelling in the right posterior maxillary region was seen extending from the muco-buccal fold of 15 anteriorly to the maxillary tuberosity posteriorly [Fig 1]. Expansion of the buccal plate was evident. The swelling was hard and non-fluctuant with diffuse margins. On aspiration straw coloured fluid was obtained. Orthopantamograph (OPG) showed radiolucency in 17,18 region destroying the tuberosity and zygomatic region with soft tissue shadow [Fig 2]. Surgical enucleation of the cyst was done. The enucleated specimen was sent for histopathologic examination in 10% formalin. On microscopic examination section showed odontogenic epithelial lining and connective tissue wall. The epithelial lining was stratified squamous epithelium of 6-8 cell layered thickness, showing detachment from the underlying collagenous capsule with palisading arrangement of nucleus in the basal layer. Histopathology was suggestive of keratocystic odontogenic tumour [Fig 3].

## CASE 2

A 27 year old male patient reported to the Department of oral medicine and radiology with the chief complaint of pain in right lower back tooth region for the past 2 months. Pain was associated with difficulty in mouth opening. The past medical and dental history was unremarkable. On clinical examination diffuse swelling was present on right side of the face at the angle of mandible. Swelling was 5 x 4 cm in size. It was firm in consistency and tender on palpation. Intraorally there was partially erupted 47. The distal half of 47 was covered with operculum and it was inflamed, edematous extending upto the middle of 46 on buccal side. Pus discharge was seen in relation to 47. Area of hyperkeratinisation was present along the right buccal mucosa in relation to 47 [Fig 4]. OPG revealed a well circumscribed radiolucency in the body and ramus of the right side of the mandible, thin fine septa were seen within the radiolucency. The borders were sclerotic and continuous [Fig 5]. Surgical enucleation of the cyst was done. The enucleated specimen was sent for histopathologic examination which showed stratified squamous epithelium of 5-8 cell layer thickness, with a corrugated parakeratin layer and flat epithelial connective tissue interface. Histopathology suggested keratocystic odontogenic tumour [Fig 6].

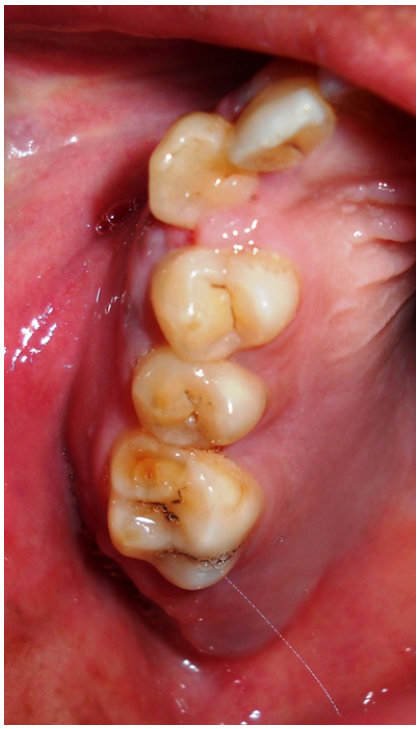

Fig 1: Intra oral photograph shows swelling in the right posterior maxillary region with expansion of cortical plates.

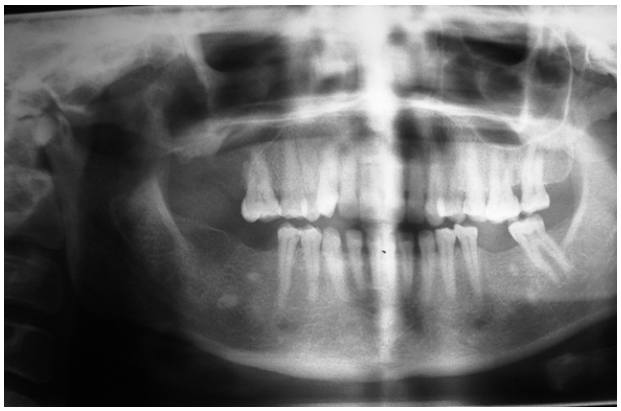

Fig 2: OPG showed radiolucency on the right sider maxillary posterior area destroying tuberosity and zygomatic region with soft tissue shadow.

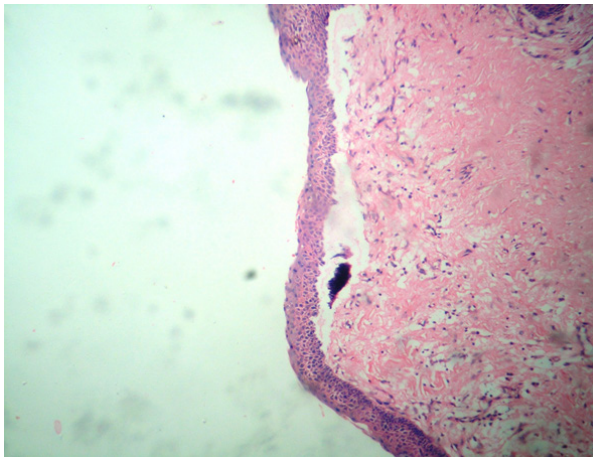

Fig 3: H&E, x10 view shows stratified squamous epithelium of 6-8 cell layered thickness, showing detachment from the underlying collagenous capsule with palisading arrangement of nucleus in the basal layer. Features suggestive of OKCT.

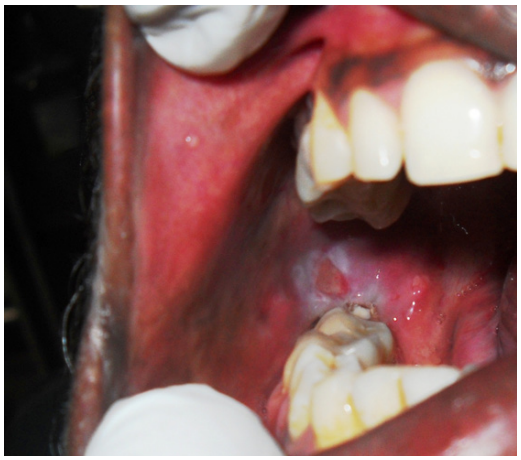

Fig 4: Intra oral photograph showing area of hyperkeratinisation present along the right buccal mucosa in relation to 47 region.

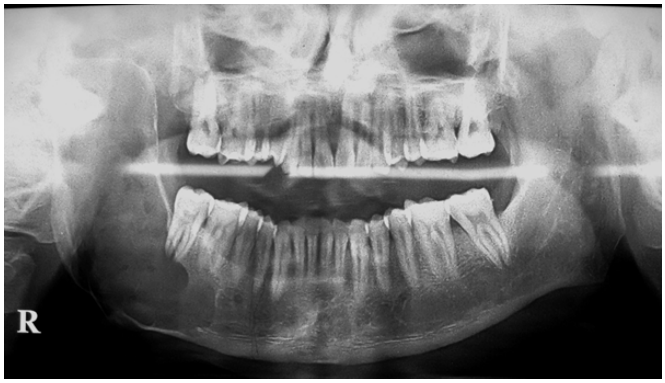


Fig 5: OPG revealed a well circumscribed radiolucency in the body and ramus of right side of the mandible, thin fine septa which are seen within the radiolucency.

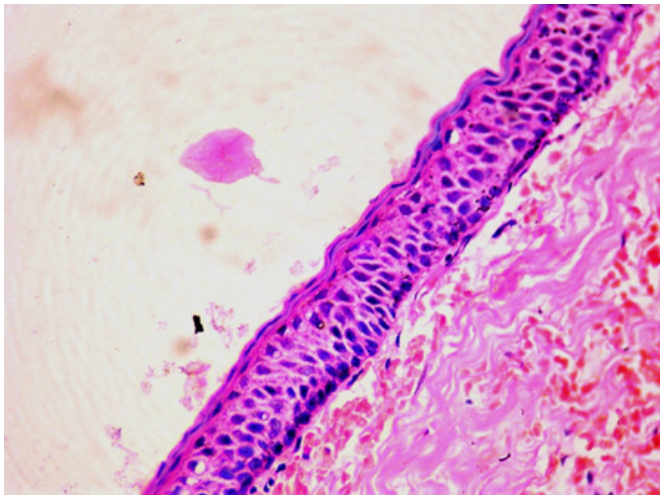

Fig 6: H&E, x40 view shows stratified squamous epithelium of 5-8 cell layer thickness, with a corrugated parakeratin layer and flat epithelial connective tissue interface suggestive of OKCT.

## DISCUSSION

Odontogenic Keratouystic tumors are relatively common developmental odontogenic cysts and account for 10-12% of all jaw cysts.[3,4] OKC shown to have bimodal age distribution case in this patient also distribution with first peak in 2nd and 3rd decade and the second peak in the 5th decade. Most studies reveal a slight male predominance. They are less common in maxilla than mandible with only 31.3% in maxilla.[5] Approximately three quarters of all OKCs occur in the body of the mandible, most commonly in the molar region and vertical ramus.[5,6] But when they occur in the maxilla they occur more commonly in canine region. Maxillary OKC tends to exhibit a unilocular, smooth, round border while mandibular ones had scalloped border.[7] The occurrence of OKC in maxilla is relatively rare and invasion of the maxillary sinus is unusual,[7] An OKC usually occurs as a single lesion. Multiple lesions are associated with the nevoid basal cell syndrome (Gorlin-Goltz syndrome).[3] In contrast with other odontogenic cysts, OKCs have a high recurrence rate, reportedly ranging from 13% to 60% (2-7). In approximately 50% of patients, the lesion is asymptomatic. In others, pain, swelling, expansion, drainage, and bone perforation are reported.[4,8] Buccal cortical plate expansion was noticed which is seen in only one third cases of maxillary cysts. Whereas in OKC the extension is more in anteroposterior direction and the pressure of the fluid is quite low and grows by extension rather than by expansion. The extension here is due to reasons like fingerlike projections from the cyst wall into the marrow spaces, and enlarges slowly but relentlessly along the path of least resistance.[5] Hence not much of cortical expansion is seen in the initial stages, and by the time it shows clinical swelling the lesion would have been quite huge. OKCs are usually discovered during the course of a routine radiographic examination and demonstrate a well-defined unilocular or multilocular radiolucency with smooth and often corticated margins which may simulate that of odentigerous, radicular, residual or a lateral periodontal cyst. Root resorption is seldom a feature.[9] Cawson et al in 2004 have considered OKCs as keratinizing cysts and have divided it into para-keratinised and ortho-keratinised linings. They have called para-keratinised cysts as odontogenic OKC or KCOT. Ortho-keratinised cysts have been called as orthokeratinised odontogenic cysts (OKOC).[5] Philipsen and Riechert have suggested that OKC should be considered as a benign tumour and hence be called as KCOT. Currently OKC has been reclassified by the WHO as a KCOT.[2] The recurrence rate associated with enucleation with adjunctive therapy such as cryosurgery and decompression (1-8 percent) is lower than that associated with enucleation alone (17-56 percent).[10] Postoperative follow-up with regular radiographic examination is important with OKCs because of the potential for recurrence. OKCs usually recur within five years after surgery,[11,12] but they can recur more than 15 years later.[13,14]

To conclude, althought initially it was considered to be a cyst because of its aggressive nature and high potential for recurrence, it has been rightly reclassified into tumour and termed by WHO as OKCT.

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