



## AN UNUSUAL PRESENTATION OF ORTHOKERATINIZED ODONTOGENIC CYST (OOC)-A CASE REPORT AND REVIEW OF LITERATURE


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### ABSTRACT

Orthokeratinized Odontogenic Cyst (OOC) is a developmental cyst of odontogenic origin and was initially defined as the uncommon orthokeratinized variant of the Odontogenic Keratocyst (OKC), until the World Health Organization's (WHO) classification in 2005, where it was separated from the Keratocystic Odontogenic Tumor (KCOT). It is a relatively uncommon developmental cyst comprising of only 0.4% of all odontogenic cysts. It is rather mystifying that its radiographic features are similar to the dentigerous cyst and histological characteristics are similar to the odontogenic keratocyst; and it has inconsistent cytokeratin expression profiles overlapping with both the dentigerous cyst and odontogenic keratocyst as well as with the epidermis. It has a predilection for the posterior mandibular region. Here we report a rare case of OOC in an unusual presentation in anterior maxillary region.

**Key words:** Orthokeratinised Odontogenic Cyst, Developmental Cyst, Keratocystic Odontogenic Tumor, Dentigerous Cyst, Mandible.

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### INTRODUCTION

Orthokeratinized odontogenic cyst (OOC) is a developmental cyst that occurs in maxilla and mandible, It was initially defined by the World Health Organization (1992) as an uncommon orthokeratinized type of odontogenic keratocyst (OKC) [1]. It is a rare developmental odontogenic cyst arising from the cell rests of the dental lamina [2-4]. It was first described by schultz in 1927 as an orthokeratinized variant of the formerly called odontogenic keratocyst, which is known as the keratocystic odontogenic tumour in the recent days. In 1981 Wright defined it as an independent entity [2]. Since

then it has received various terminologies, such as “orthokeratinized variant of odontogenic keratocyst” or “orthokeratinized cyst of the mandible”. In 1988 Li et al. suggested the term “orthokeratinized odontogenic cyst,” which is the most accepted terminology till date [5]. World Health Organization's new classification (2005) for head and neck tumors had renamed OKC as Keratocystic Odontogenic Tumor (KCOT) and reclassified it as a neoplasm due to its intrinsic growth potential and propensity to recur. According to this new classification, OOC is not a part of the spectrum of KCOT and should be distinguished from the latter [1,4,6]. Radiographically the cyst appears to be a well-circumscribed, unilocular, or multilocular radiolucency that occasionally is associated with an unerupted tooth or with the root without causing resorption [7]. Here we present a case report of 45 year old

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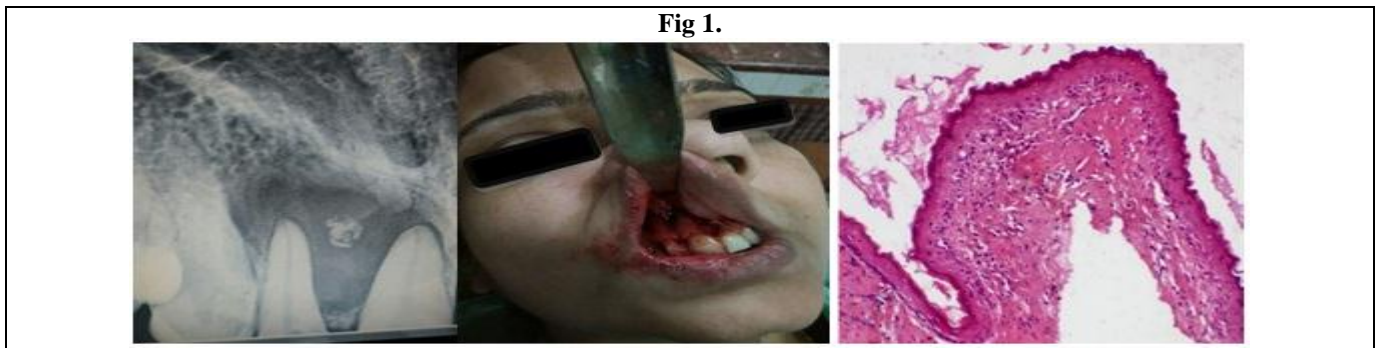
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female patient with a rare occurrence of OOC in maxillary anterior region.

**CASE REPORT**

A 45 year old female reported to Meenakshi Ammal Dental College, to the dept of Oral Medicine And Radiology, with a chief complaint of mobile discoloured teeth in the upper anterior region for past 2 months with no history of trauma. On intraoral examination, there was no obliteration of sulcus in relation to 11, 12 and the patient was asymptomatic. there was no pain on percussion in reation to 11,12. Pulp vitality test was performed in relation to 11,12,13,14,21,22 and the teeth 11,12 did not respond. The systemic review of the patient was non contributory. Based on the clinical finding, a provisional diagnosis of non vital teeth in relation to 11,12 was given. The patient was subjected for routine Intraoral Periapical Radiograph (IOPA) IRT 11,12 region (Fig.1 ). IOPA reveals normal enamel, dentin and pulp with a unilocular

radiolucency in between the apical portion of 11,12 surrounded by a scalloped margin with centered calcified material within the radiolucency. Based on this a radiographic diagnosis of benign odontogenic tumour was made with differential diagnosis of calcifying epithelial odontogeniccyst, radicular cyst ,ameloblastic fibro odontoma, orthokeratinized odontogenic cyst were considered. The patient was subjected for routine blood investigations which were within the normal limits. Excisional biopsy was performed (Fig.1) and the specimen was sent for histopathological examination. On microscopic examination, (Fig.1) an orthokeratinized stratified squamous epithelial lining and a fibrous connective tissue wall. The epithelium was of 4-6 layer thickness with surface corrugation and prominent granular layer. The basal cells were cuboidal. Based on this a histopathological diagnosis of orthokeratinized odontogenic cyst was made.



**Table 1. Keratin profile in OOC, KCOT & their localization within epithelium**

Keratin profile	Expression within epithelium	OOC	KCOT
K4	Superficial layer	- *	+*
K13	Superficial layer	-	+
K17	Basal & suprabasal	-	+
K19	Basal & suprabasal	-	+
K1	Suprabasal layer	+	-
K10	Suprabasal layer	+	-
LOR	Superficial layer	+	-

\* positive : - (+), negative: - (-), K:-Keratin, LOR:-Loricrin

**DISCUSSION**

OOC is a rare developmental jaw cyst that occurs twice as frequently in the posterior region of mandible than the maxilla. It is predominantly seen in young adults, with a male to female ratio of 2:1. The size can vary from lesser than 1 cm to greater than 7 cm in diameter [8]. OOC appears clinically and radiographically representing dentigerous cyst as they most often involve an unerupted mandibular third molar [9]. Swelling is the most frequent

symptom and is accompanied with pain although in most of the cases, the lesion is asymptomatic [gonazel] Large lesions can cause cortical expansion [10]. Radiographically, most of the cases appear as unilocular lesion with well-defined margins. Dong Q et al in their study of 61 cases, found radiographic data in 54 cases of OOC [11]. Out of 54 cases, 47 cases showed unilocular radiolucency and 7 cases showed multilocular radiolucency [1]. Li et al in their study of 15 cases found that 14 cases

were radiographically unilocular and only one case was multilocular [12]. The lesions with similar clinical and radiographic presentation should be considered in the differential diagnosis such as dentigerous cyst, KCOT, ameloblastoma in particular Unicystic ameloblastoma. Histopathologically all cystic lesions show cystic lumen, lining epithelium and connective tissue wall. The cystic cavity is lined by a regular stratified squamous epithelium, usually thin and uniform about 4- to 9-cell layers thick. This epithelium presents a defined basal layer that exhibits palisaded cuboidal or flat cells, with nuclear hyperchromatism, an intermediate layer of polyhedral cells with eosinophilic cytoplasm, and a thick superficial layer of orthokeratin. This entity must be differentiated from the KCOT that shows a regular epithelium of 5- to 10-cell layers thick with the basal cells lined with an elongated nucleus and the presence of a characteristic superficial corrugated layer of parakeratin. While making the diagnosis of any cystic lesion, content of cystic lumen, configuration of lining epithelium and cystic wall should be taken into consideration. Histopathologically, KCOT shows more resemblance to OOC. Enucleation with curettage is the usual treatment for orthokeratinized odontogenic cysts. Recurrence has been noted very rarely and the reported recurrence rate is only 4% in OOC as compared to high i.e. 28% in KCOTs<sup>5</sup>. The following table illustrates the keratin profile expression within the epithelium in KCOT and OOC (TABLE:1)

The less significant expression of p63 in OOCs compared to KCOTs suggests a lower proliferative and self-renewal potential for OOCs which may explain the different clinical behaviors between OOCs and OKCs. Cell proliferation molecules and related factors including K67, proliferating cell nuclear antigen, p53 and argyrophilic nucleolar organizer regions had been used to indicate

biological behavior of odontogenic cysts and tumors. Compared with KCOTs, expression level of Ki-67 and p63 are significantly lower in OOCs, suggesting a lower proliferative activity [12, 13]. Enucleation with curettage is the usual treatment for orthokeratinized odontogenic cysts. Prognosis is good with very low recurrence rate. KCOTs are treated similarly to other odontogenic cysts, that is, by enucleation and curettage and aggressive type are treated with bone resection and curettage [8].

## CONCLUSION

Orthokeratinized odontogenic cyst is an independent clinical and pathological entity with biological behaviour, management & prognosis different from that of keratocystic odontogenic tumour (KCOT). Thus this review shows the importance in diagnosis of OOC and KCOT as both the lesions look similar but with different biological behavior and there is more tendency of clinical misdiagnosis which in turn could affect the prognosis of the patient.

## STATEMENT OF HUMAN AND ANIMAL RIGHTS

All procedures performed in human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animals performed by any of the authors.

## ACKNOWLEDGMENTS

Nil

## CONFLICT OF INTEREST

None.

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