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RESEARCH ARTICLE

Orthokeratinized Odontogenic Cyst: A case report with Clinicopathological review

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Abstract

Orthokeratinized Odontogenic Cyst (OOC) is a developmental cyst of odontogenic origin of jaw, which was formerly considered by the World Health Organization (WHO) as the uncommon orthokeratinized variant of Odontogenic Keratocyst (OKC), until the WHO in 2005, where it was separated from the Keratocystic Odontogenic Tumor (KCOT). It is an uncommon developmental cyst comprising of 0.4% of all odontogenic cyst and shows peculiar clinicopathologic aspects when compared with other developmental odontogenic lesions such as Dentigerous cyst and the Keratocystic Odontogenic Tumor. Clinically, it occurs as a solitary cyst, shows predilection for males and most often found in second to fifth decade. It mainly occurs in mandible, least recurrence was noted after surgical removal of the lesion. The purpose of this article is to present a case of OOC arising in the mandible and to discuss the review of literature.

INTRODUCTION

The Orthokeratinized Odontogenic Cyst (OOC), is a developmental odontogenic cyst comparatively uncommon type arising from cell rests of dental lamina [3, 12]. It was first reported by Schultz in 1927 as an orthokeratinized variant of the previously called Odontogenic Keratocyst (OKC), nowadays known as the Keratocystic Odontogenic Tumor , WHO 2005 [5, 14].

In 1981. Wright termed it as 'Odontogenic Keratocyst-Orthokeratinized variant' and specified its clinicopathological aspects assuring that OOC is an individual entity distinct from OKC due to its limited growth potential and lower recurrence [14]. In 1993, Vuhahula et al termed it as 'Jaw cyst with Orthokeratinization' and in 1998 Li et al nominate it as 'Orthokeratinized Odontogenic Cyst' [12, 13].

In this paper, we are reporting a clinical case of the OOC and review on its main clinicopathological aspects has been highlighted.

Case Report:

A-25 year old male patient reported to the department of Oral and Maxillofacial Surgery with the chief complaint of pain and swelling in his lower left back region of jaw and he also complaints of reduced mouth opening since last 3 days. The patient was taking medicines for pain, prescribed by a general practitioner. His past medical and past dental history was not relevant and his general physical status was good with all vital signs within normal limits. The extraoral examination revealed swelling over the left mandibular angle region which was tender and warm on

palpation (Fig.:1). On intraoral examination a diffuse swelling was present over 37 which was tender on palpation and also there was pus discharge from 37. 38 was not visible intraorally (Fig.:2). The orthopantomogram showed well-defined, unilocular radiolucent lesion involving posterior body and ramus of mandible. Lesion involved roots of 37 and an impacted 38 which was displaced up to inferior border of mandible (Fig.:3). All laboratory findings where within normal range. Based on history, clinical and radiological examination a provisional diagnosis of Dentigerous Cyst was made with a differential diagnosis of Keratocystic Odontogenic Tumor and Unicystic Ameloblastoma. As the size of the lesion was very large enucleation was done along with application of Carnoy's solution, open packing of the lesion was done with ribbon gauze soaked in povidone iodine solution, also extraction of 37 and 38 was done (Fig.:4). After initial healing obturator was given which will be adjusted according to healing (Fig.:5). The specimen enucleated was sent for histopathological examination (Fig.:6). The patient is on continued follow up and no fresh complaints have been noted.

Histopathologic examination revealed corrugated orthokeratinizing six to ten cells layer thick epithelium lining of cyst with distinct granular layer, basal cells are tall columnar. The connective tissue wall of the cyst is made up of fibrocartilage tissue which shows chronic inflammatory cells sub epithelially. Epithelial connective tissue interface was flat (Fig.:7). A final diagnosis of Orthokeratinized Odontogenic Cyst was made.

Discussion:

The term Odontogenic Keratocyst (OKC) was first used by Philipsen in 1956. Histologically, OKC are formed with a Stratified Squamous Epithelium that produces Orthokeratin (10%) , Parakeratin (83%) and both types of keratin (7%). Both Orthokeratinized and Parakeratinized variants shared common diagnosis as OKC, and many studies have been carried out over years showing that these two entities to be different from each other. Considering the behavior, recurrence rate, histopathological features and association with Nevoid Basal Cell Carcinoma Syndrome, the parakeratinized variant of OKC is reclassified as Keratocystic Odontogenic Tumor (KCOT) by WHO 2005, whereas orthokeratinized variant is considered as a different entity, Orthokeratinized odontogenic cyst (OOC) [15]. Wright in 1981 considered OOC as a separate variant other than histopathology was its reduced rate of recurrence [14].

The OOC is a developmental odontogenic cyst, it arises from cell rests of dental lamina. The incidence varies ranging from 5.2 to 16.8% or 3.3 to 12.2% of the cases previously classified as OKC to only 0.4 % of all odontogenic cysts [3, 15].

OOC occurs predominantly in males with a ratio (M:F=2.59:1). Dong et al suggested a female predilection. OOC occurs in the third and fourth decades with an average age of 38.9 years. Clinically , it usually presents as a slow growing, asymptomatic jaw swelling, with or without pain with a size varying less than 1cm to 7cm. In our case patient was having pain, with reduced mouth opening. The mandible to maxilla ratio is 9.17:1, with a predilection for the mandibular molar and ramus region[11, 3, 6, 10].

Radiographically, OOC appears as a unilocular (87%) [11] or multilocular radiolucency often associated with an impacted teeth(60.8%) [11]. About two-third of the OOC are encountered in a lesion that appears radiographically like a Dentigerous cyst, as in the present case [9].

Histologically. OOC shows a thin, uniform epithelial lining four to eight cell layers thick and is composed of an orthokeratinized stratified squamous epithelium with a prominent granular cell layer. The basal cells are flat to cuboidal, without the palisading or polarization which are seen in KCOT. Immunohistochemical features shows positivity for K1, K10 and Loricrin. K4, K13 and K17 expressions are negative. Antiapoptotic marker bcl-2 absent in the basal layer, there is also decreased expression of ki-67 and p63 [11, 15, 3].

Differential diagnosis of the OOC includes Dentigerous cyst, Paradental cyst, Unicystic ameloblastoma, and KCOT [4].

Due to less aggressive behavior of OOC, the treatment of choice is enucleation along with removal of the involved teeth. Recurrence rate has rarely been noted 2.2% [4, 11].

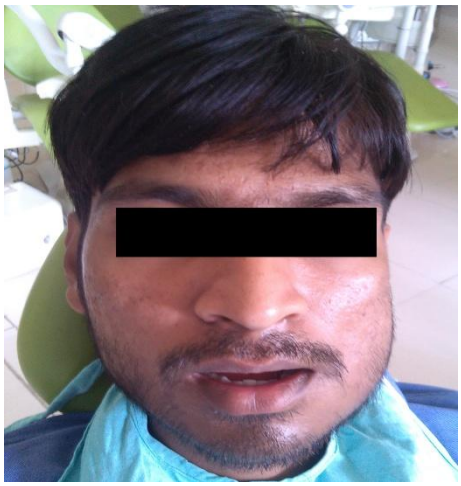

Fig.1: Extra-oral with mouth opening

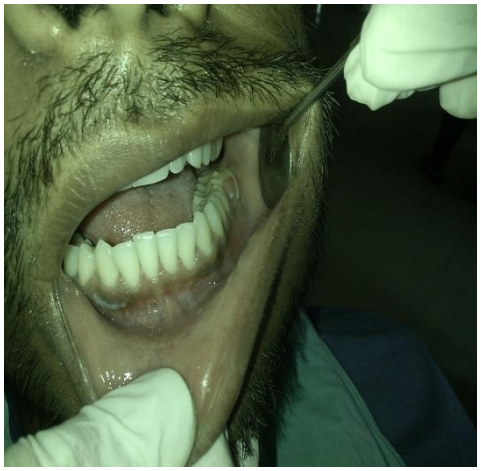

Fig.2: Intra-oral (pus discharge from 37)

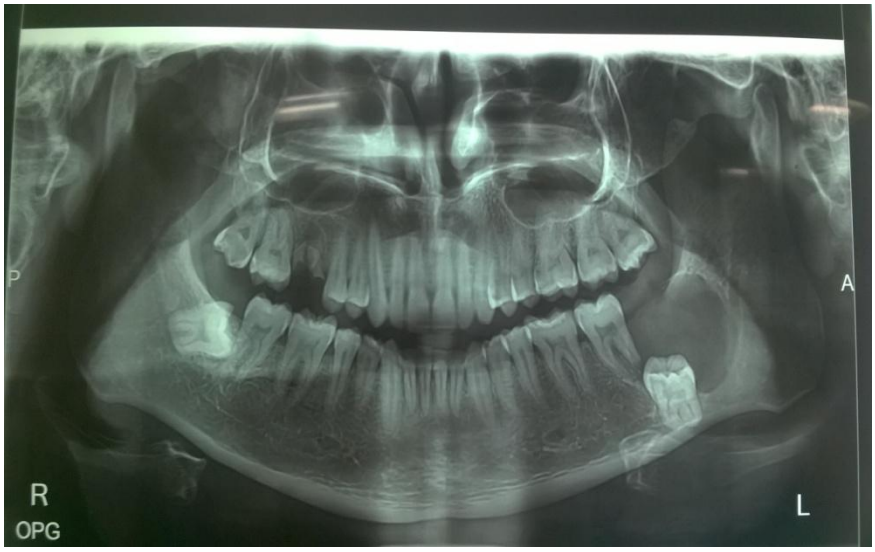

Fig.3: Preop OPG

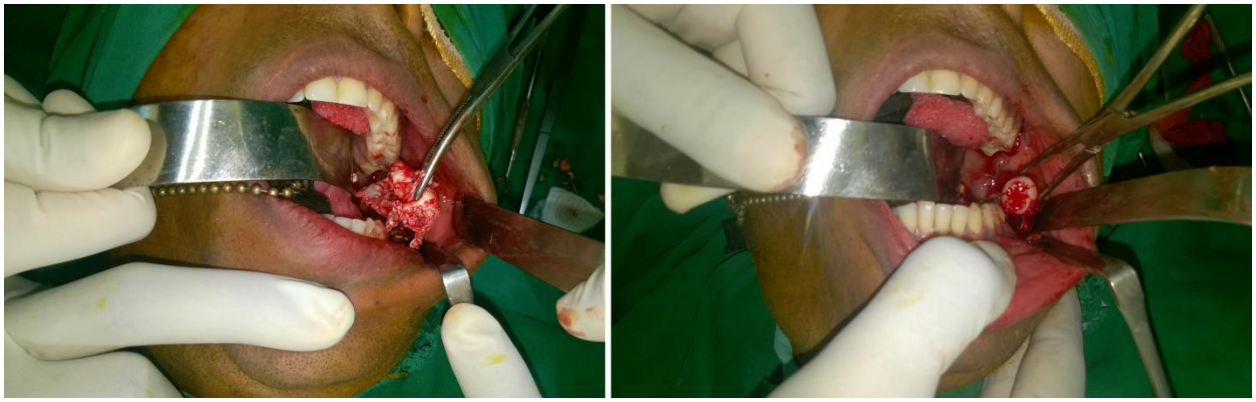

Fig.4: Intraop (cyst enucleation with extraction of 38)

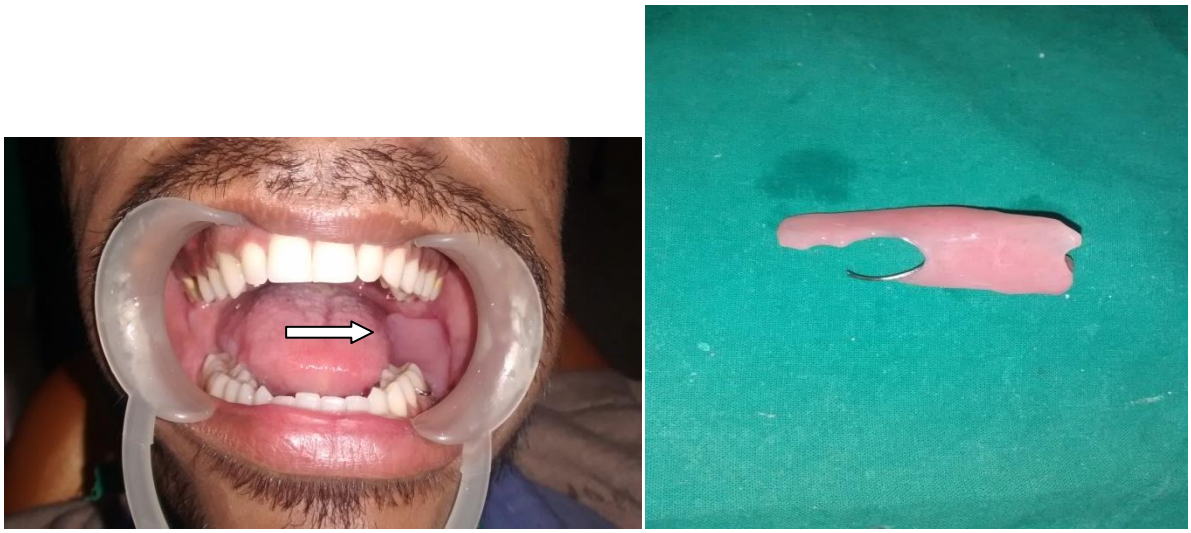

Fig.: 5 Obturator

Fig.: 6 Cystic content

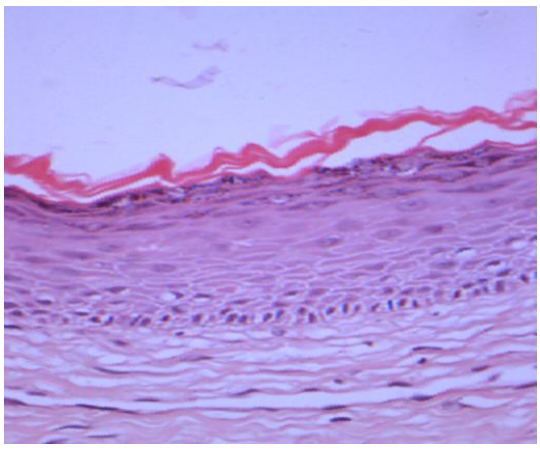

Fig.: 7 Histopathological slide

Conclusion:

To conclude that the Orthokeratinized Odontogenic Cyst (OOC) is an independent clinical and pathological variant of the Odontogenic Keratocyst (OKC) with a different prognosis. Hence, OOC should be considered in the

differential diagnosis of the radioleucent lesions of the jaw associated with an impacted tooth particularly those cases simulating as Dentigerous Cyst.

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