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An Aggressive Orthokeratinized Odontogenic Cyst of the Mandible: A Case Report.

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Abstract: *Orthokeratinized odontogenic cyst (OOC) is a developmental cyst that occurs in the jaw. it was initially defined by the world health organization as the orthokeratinized variant of odontogenic keratocyst (OKC). However, studies have shown that OOC has a peculiar clinicopathologic aspects when compared with other developmental odontogenic cysts, especially OKC's. The orthokeratinized odontogenic cyst is a distinct clinicopathologic entity and is histologically characterized by a thin, uniform, epithelial lining with orthokeratinization. Clinically, the orthokeratinized cyst is a single cyst, shows a predilection for males, and is most often found in the second to the fifth decade. It is not a dentigerous cyst but is often mistaken for a dentigerous cyst because of its association with unerrupted or impacted tooth in the posterior mandible. It exhibits lower clinical aggressiveness compared to its counterpart OKC. The purpose of the article is to present a case of large aggressive OOC in the anterior mandible and to highlight the importance of, distinguishing it from the more commonly occurring OKC or keratocystic odontogenic tumour (KCOT).*
Key Words: *Jaw cysts; Odontogenic cyst**;** Odontogenic keratocyst; Dentigerous cyst.*

I. Introduction

Orthokeratinized odontogenic cyst (OOC) is a developmental cyst that occurs mostly in the maxillary corpus, it was initially defined by the world health organization in 1992 as the uncommon orthokeratinized type of odontogenic keratocyst (OKC) (Sciubba JJ et al, 1999).

The lesion has been termed variously as an "orthokeratinized variant of OKC'' or a ''jaw cyst with orthokeratinization". The world health organization's new classification of the year 2005 for head and neck tumors has designated OKC as keratocystic odontogenic tumor (KCOT) and reclassified it as a neoplasm in view of its intrinsic growth potential and propensity to recur. According to this new classification, OOC should not be part of the spectrum of KCOT and should be distinguished from the latter (Wright JM, 1981; Philipson HP, 2005).

II. Case Report:-

A 25 year old systemically healthy female patient came up with the complaint of tenderness and slight swelling in the lower front jaw region, of duration of 1 month. On examination, it revealed to be a mild diffused swelling of approximate size 3 x 2 cm (Figure 1) over anterior mandible region, causing mild facial asymmetry. On palpation, swelling was firm with associated tenderness. Intra oral examination(Figure 2, 3) revealed the swelling extending from left premolar region to right lateral incisor, with obvious midline crossing. On palpation, mild buccal plate expansion & with multi lobular consistency of the bony plate was evident, also tenderness on palpation was present. There was no apparent overlying mucosal changes or signs of fluctuation. The associated teeth had variable degree of labio-lingual tipping of crown, but all teeth were found to be vital in subsequent vitality test.

Orthopantomogram (OPG) (Figure 4) revealed a well defined radiolucency with sclerotic and scalloped margins extending from periapical region of lower premolar to premolar region crossing arches, multi loculer in appearance, with thinning of lower border of mandible. Also associated impacted supernumery tooth like structure was noted in the superior periphery of the lesion (Figure 4).

Further Occlussal radiograph (Figure 5) revealed both buccal and lingual plate expansion with the lesion.

Her past medical history was of no relevance and general physical status was good. Lab findings included routine blood and biochemical investigations. Later to confirm the diagnosis, deep intrabony incisional biopsy was done, which revealed (Figure 6) it to be a orthokeratinized odontogenic cyst, with typical histopathological appearance.

Gradually the treatment planning for the patient was formulated, for the first phase intentional endodontic treatment of the associated teeth were undertaken and subsequently, under general anesthesia the patient was undertaken for cyst enucleation (Figure 7) and chemically cauterized with Carnoy's solution followed by primary closure.

Excised pathology was sent for histopathological examination, which further confirmed the incisional biopsy findings. The postoperative course was uneventful and there were no signs of recurrence after a periodic follow-up of 12 months and revealed (Figure 8) substantial healing of bony defect.

III. Discussion:-

The orthokeratinized odontogenic cyst defined as an orthokeratinized variant of the odontogenic keratocyst was defined firstly by Wright in 1981 owing to it's different orthokeratinized histopathology and reduced likelihood to recur[2]. Although both the first two editions of the World Health Organization's histological classification of odontogenic tumors, recognized this entity as variant of OKC with orthokeratosis, the WHO's 2005 edition excluded it from its definition of a KCOT (Philipson HP, 2005). The 2005 edition reclassified the parakeratotic type as a Keratocystic Odontogenic Tumour and stated "Cystic jaw lesions that are lined by orthokeratinizing epithelium do not form part of the spectrum of a KCOT". Initially three histologic variants were recognized of OKC i.e. a parakeratinized variant, an orthokeratinized variant, and combination of the both. The less aggressive clinical behaviour and recurrence pattern of the orthokeratinized variant ultimately warranted the designation of the orthokeratinized variant as a separate entity, "Orthokeratinized Odontogenic Cyst" (Philipson HP, 2005; Crowley TE et al, 1992).

Vuhahula et al, in their study stated that reduced enamel epithelium involved in formation of dentigerous cyst that had completed its tooth-forming function had the capability to keratinize under appropriate stimuli, thus forming a true dentigerous cyst with keratinization (Dong Q et al, 2010). The incidence of KCOT is about eight times more than that of the OOC, thus OOC being the so called counter part of OKC is very rare occurrence of this keratocystic pathology. The clinical or radiographic features of OOC are not very distinctive, that can differentiate this entity from other inflammatory or developmental cysts found in jaws. OOCs are generally solitary asymptomatic lesions, occurring in the third to fourth decade and with a male predilection (Philipson HP, 2005). They are more commonly found in the mandible with an affinity for the posterior region and most often involve an unerrupted mandibular third molar tooth. The size can vary from less than 1 cm to large lesions greater than 7 cm in diameter. The lesion usually appears as a unilocular radiolucency, but occasionally multilocular lesions are also encountered similar to OKCs. About two thirds of OOCs are encountered in a lesion that appears clinically and radiographically as a dentigerous cyst with associated unerrupted or impacted tooth (Eryilmaz T et al, 2009). In the study of case series of OOC conducted by Qing Dong et al, 87% of the total cases of OOCs had unilocular radiolucencies. Whereas, multilocular lesions were found in 13% of total cases, also another important characteristic found to be involved with the pathology was that 50% of the total cases were found to be associated with an impacted tooth (Dong Q et al, 2010). OOCs are more commonly found in the mandible with an affinity for the posterior region and most often involve an unerrupted mandibular third molar tooth. The size can vary from less than 1 cm to large lesions greater than 7 cm in diameter (Eryilmaz T et al, 2009). The present case was found to be associated with a impacted Supernumery tooth with dimensionally large presentation of size more than 5cm.

Enucleation with curettage is the usual treatment for orthokeratinized odontogenic cysts. Recurrence has rarely been noted, and the reported recurrence rate is only 4% in OOC as compared to a high i.e. 28% in KCOTs (Mc Donald Jankowski DS, 2010).

The significant clinicopathologic differences between orthokeratinized and parakeratinized odontogenic cysts make it imperative that the orthokeratinized cyst be recognized as a distinct entity and accordingly treated so.

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Figure 1. Extra oral presentation of the patient.

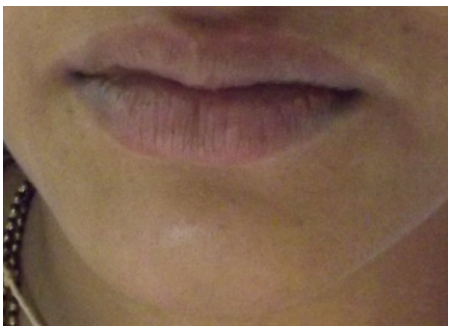

Figure 2. Noticable buccal plate expansion.

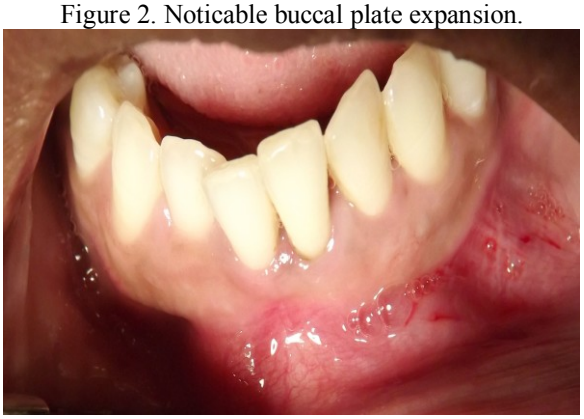

Figure 3. Lingual plate with no clinical expansion.

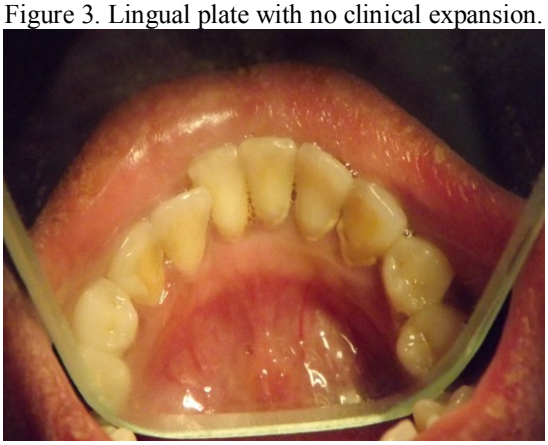

Figure 4. OPG showing a well defined radiolucency with scalloped and sclerotic margins over anterior region.

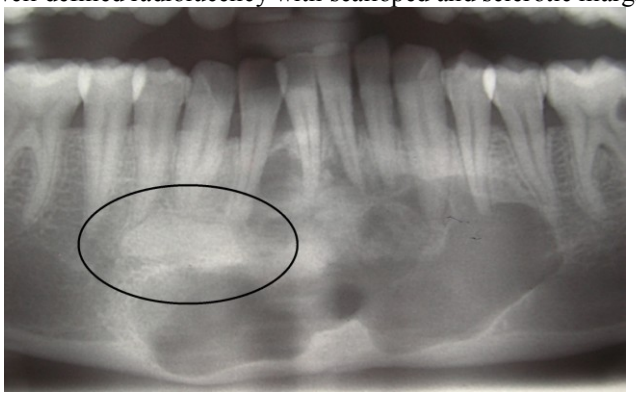

Figure 5. Occlussal view with apparent bicortical expansion.

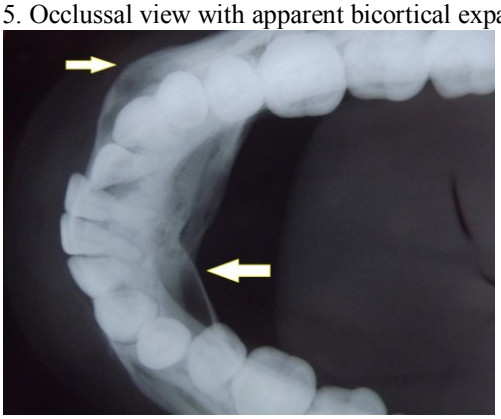

Figure 6. Orthokeratinized stratified squamous epithelium, with basal cell layer showing palisading arrangement and loss of retepegs with basement membrane.

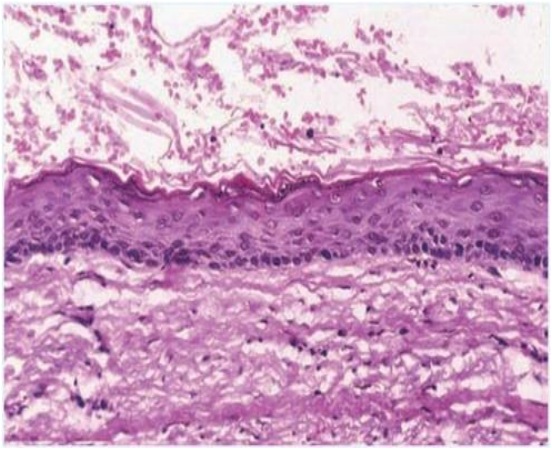

Figure 7. Enucleated Cystic Space.

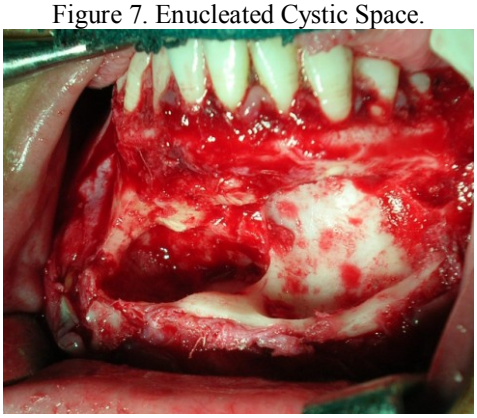

Figure 8. Healthy bony healing after 12 months.