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## INTERNATIONAL JOURNAL OF ADVANCED RESEARCH (IJAR)

Article DOI: 10.21474/IJAR01/15274

DOI URL: <http://dx.doi.org/10.21474/IJAR01/15274>



### RESEARCH ARTICLE

#### MULTILOCLAR APPEARANCE OF ORTHOKERATINIZED ODONTOGENIC CYST: A CASE REPORT

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#### Manuscript Info

##### Manuscript History

Received: 27 June 2022

Final Accepted: 30 July 2022

Published: August 2022

##### Key words:-

Developmental Odontogenic Cyst,  
Odontogenic Keratocyst,  
Orthokeratinized Odontogenic Cyst

#### Abstract

Orthokeratinized odontogenic cyst (OOC) was initially defined as the uncommon orthokeratinized type of odontogenic keratocyst (OKC) by the World Health Organization (WHO). Several studies have shown that OOC has different clinicopathologic aspects when compared with other developmental odontogenic cysts, especially OKC. So OOC now emerges to be a distinct entity, according to the 4th edition of the WHO Classification of Head and Neck Tumors which was published in January 2017. Clinically, it usually occurs as a single cyst with a unilocular radiographic appearance in posterior mandible with a predilection in males and mostly is seen between the third and fourth decades. In this case we present an OOC arise as a multilocular lesion causes thinning and expansion of the buccal cortical plate treated by enucleation and curettage.

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#### Introduction:-

In 1927, OOC was first described as an orthokeratinized type from OKC, which is called keratocystic odontogenic tumor (KCOT) by Schultzas. In 1992, WHO has defined OOC as the uncommon orthokeratinized type of OKC. In 2005, WHO redefined OKC as a neoplasm and termed it as KCOT because of its high recurrence rate, aggressive clinical behavior, association with nevoid basal cell carcinoma syndrome, tendency to multiplicity and mutations in the PTCH tumor suppressor gene.<sup>[1][2]</sup> In 2017, WHO reverted back to the original and well accepted terminology of OKC as many papers showed that the PTCH gene mutation could be found in non-neoplastic lesions, including dentigerous cysts.<sup>[1][3]</sup> In 2017, WHO classification of OOC was accepted as a separate entity for the first time. It differs both clinically and histopathologically from OKC.<sup>[1]</sup>

OOC may be presented as a swelling with or without pain which can reach a large size that causes cortical expansion. In most of the cases, it can be detected accidentally during a radiographic examination. Size of the lesion range from <1 cm to 7 cm.<sup>[4][5]</sup> It usually occurs as a single cyst with a unilocular radiographic appearance in posterior mandible with a predilection in males and mostly is seen between the third and fourth decades.<sup>[4][6]</sup>

Enucleation with curettage is the appropriate treatment of choice for OOCs. Only 4% of OOCs showed recurrence. In a study done by Crowley, Kaugars and Gunsolle recurrence rates of the OOC were 2.2%.<sup>[4][7][8]</sup>

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**Case Report:**

A 36 years old male patient came to the oral and maxillofacial department, faculty of dentistry, Ain Shams university. The patient was presented with pain in lower right jaw region. Intra-oral examination revealed mild swelling at the area of right angle of mandible with no facial asymmetry on extra oral examination.

Orthopantomogram was showed a well-defined multilocular radiolucency with corticated border in the angle of the mandible, associated with an impacted lower right wisdom tooth and extending from lower right second molar to the ascending ramus [Figure 1]. Computed tomography was showed an expansile osteolytic lesion with thinning and expansion of buccal cortical plate [Figure 2].

Patient reported free medical history. Differential diagnosis of OKC, dentigerous cyst, ameloblastoma and OOC was suggested, but ameloblastoma usually shows root resorption of the involved teeth. Aspiration was done and revealed cheesy white material. Incisional biopsy was decided as the first line of management to determine the further surgery.



**Figure 1:-** Orthopantomogram x-ray showing a well-defined multilocular radiolucency associated with an impacted right wisdom.



**Figure 2:-** CT (coronal cut) was showed an expansile lesion with thinning and expansion of buccal cortical plate.

The lesion was well encapsulated with thick lining which facilitate the excision of the lesion as one unit [Figure 3]. Thus, surgical enucleation of the lesion and curettage were done along with surgical removal of the impacted wisdom [Figure 4]. Gross examination of the excised specimen revealed a white cheesy material into the lumen. Microscopic examination of the specimen revealed an orthokeratinized stratified squamous epithelium of varying thickness.



**Figure 3:-** The lesion was well encapsulated with thick lining.





**Figure 4:-** Surgical enucleation of the lesion and curettage were done along with surgical removal of the impacted wisdom.

### Discussion:-

Orthokeratinized odontogenic cyst mostly is seen between third and fourth decades and has a male gender predilection.<sup>[9][10]</sup> In this case, 36-year-old male was reported with OOC. The main clinical feature of OOC is swelling associated with or without pain.<sup>[11]</sup> In the present case, patient had a mild swelling with pain in lower right angle region.

Radiographically, the OOC usually appear as a radiolucent unilocular lesion which frequently associated with impacted teeth with or without expansion.<sup>[4][6]</sup> In contrast to other studies, the lesion was large expansile multilocular radiolucent, which was associated with impacted wisdom with thinning and expansion of buccal cortical plate. Other radiolucent lesions of the jaw differentiated from the OOC are mainly odontogenic lesions such as dentigerous cyst, ameloblastoma and OKC. The OOC rarely appear as multilocular radiolucency in posterior mandible as the ameloblastoma and the OKC,<sup>[6]</sup> but OOC usually shows no root resorption of involved teeth, which is a characteristic feature of ameloblastoma.<sup>[9][12]</sup> OKC can be differentiated from OOC on various features like older age group, more antero-posterior extension without expansion, characteristic histopathological features different from OOC, parakeratinized layer, high recurrence rate, association with basal cell nervous syndrome.<sup>[13]</sup> OOC may appear as periapical lesions, so all radiolucent lesions should be sent for histopathological examination.<sup>[14][15]</sup>

Histologically, the cavity of OOC is lined by a uniform 4- to 9-cell layers thick stratified squamous epithelium, which showed a basal layer that exhibits palisade cuboidal or flat cells, with nuclear hyperchromatism. The intermediate layer is made up of polyhedral cells with eosinophilic cytoplasm with a thick superficial layer of orthokeratin. While OKC shows of 5- to 10-cell layers thick epithelium with the basal cells lined with an elongated nucleus with the presence of a characteristic superficial layer of parakeratin.<sup>[16][17]</sup> In this case, typical histologic features of OOC were present.

Very limited information is available about orthokeratinized odontogenic cyst due to very low incidence rate and due to lack of specific clinical and radiographic features. Due to the similarity between OOC and the dentigerous cyst, several studies have been done to identify if there is any relation between a dentigerous cyst and an OOC. These data shows that CK10 and CK17 expression is negative-to-weak in a dentigerous cyst, and moderate in OOC and OKC. These two cytokeratins are expressed in the keratinized epithelium. CK18 and CK19, expressed in the non-keratinized epithelium, are expressed in the dentigerous cyst and not in OOC or OKC. CK7 and CK13, normally expressed by the dental lamina and enamel organ, are weakly positive in a dentigerous cyst, while OOC and OKC show the expression of only CK13. This supports the point that OKC and OOC may be derived from the dental lamina.<sup>[18][19]</sup>

More research work is necessary in cases with OOC to find out etiopathogenesis, clinical and radiographic features of this lesion. OOC should be considered always in the differential diagnosis of all the radiolucent lesions involving impacted teeth.

The treatment of choice is surgical excision of lesion conservatively with removal of involved teeth. Long-term follow-up of the lesion showed no remarkable recurrence. In this case also enucleation with curettage of the lesion was done with removal of the impacted tooth.<sup>[20]</sup>

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