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RESEARCH ARTICLE

ODONTOGENIC KERATOCYST OF MANDIBLE – A CASE REPORT WITH REVIEW OF LITERATURE

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ABSTRACT

Odontogenic keratocyst is an odontogenic developmental cyst. It is derived from remnants of dental lamina. The term was first termed by Philipsen in 1956. OKC was renamed as keratocystic odontogenic tumor (KCOT) in the WHO classifications of head and neck tumors in 2005 due to its aggressive, high recurrence rates and specific histological characterstics and was reclassified into the cystic category in WHO classification of Head and Neck pathology (2017). They are unique odontogenic lesions that have the potential to behave aggressively, that can recur and can be associated with nevoid basal cell carcinoma syndrome. This article focuses on a case of 38 year old female patient diagnosed with odontogenic keratocyst in the left retromolar region and a brief review of literature.

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INTRODUCTION

The odontogenic cysts are derived from epithelium associated with the development of the dental apparatus. The type of epithelium can vary with most lesions having stratified squamous but some developmental or fissural cystsin the maxilla may have respiratory epithelium (Odell, 2017). There are various types of odontogenic cysts and various classifications have been put forward over the years. For the purpose of this article, we will be using the classification put forward by the WHO (World Health Organization) in 2017, wherein, odontogenic keratocyst is classified as a developmental odontogenic cyst.

CASE REPORT

A 38 year old female patient reported to the department of Oral Pathology and Microbiology with the chief complaint of swelling in the lower left back teeth region since 4 months. Patient was apparently normal four months back when she noticed a swelling which gradually increased to the present size. Patient gave no history of pain. Patient had visited the dentist 6-7 years for extraction of teeth in the same area. Extra-oral examination revealed no abnormality (Figure 1).

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Intra-oral examination revealed a swelling could on the left retromolar region of mandible which was oval in shape measuring 2cm × 2 cm approximately with smooth surface (Figure 2). The swelling was soft and fluctuant on palpation. Hard tissue examination showed mild stains, moderate calculus, missing 16, 35, 36, 37, 46, 47 and recession in relation to 26, 27. Orthopantomogram (Figure 3) shows a well-defined, unilocular radiolucent lesion in the edentulous left mandibular molar region. Based on clinical and radiographic features, a provisional diagnosis of radicular cyst was made, and an excisional biopsy was done after which the tissue was sent for histopathological examination. Macroscopically, multiple bits of tissue were received in 10% formalin which were brownish-gray in color, soft in consistency with the largest bit measuring 1.8 cm × 1.5 cm, 2 cm × 0.6 cm approximately and the remaining bits collectively measuring 2 cm × 2cm approximately (Figure 5). The tissues were taken under processing and routine hematoxylin and eosin staining was done. Histopathological examination revealed parakeratinized stratified squamous epithelium with underlying connective tissue capsule (Figure 6). The cystic lining was 6-8 cell layer thick with surface corrugation and basal cell showed palisading appearance (Figure 7). Connective tissue capsule showed dense bundles of collagen fibers interspersed with fibroblasts, chronic inflammatory infiltrate chiefly lymphocytes and plasma cells and endothelial lined blood vessels (Figure 8). Based on the overall histopathological features, final diagnosis of Odontogenic keratocyst was given.

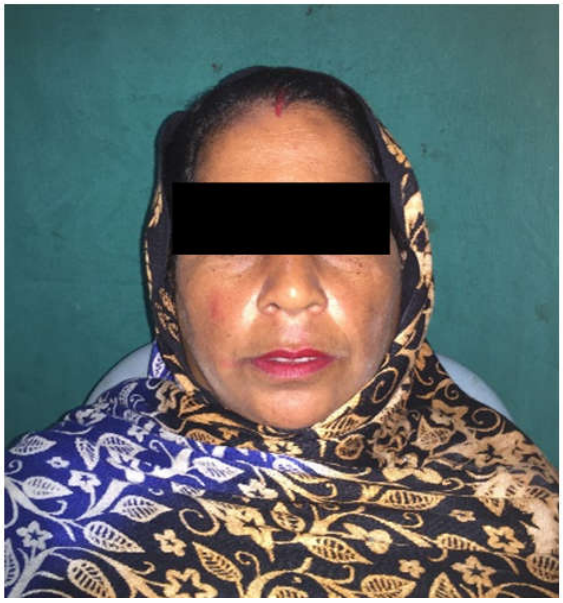

Figure 1. Extra-oral view revealing no gross facial asymmetry

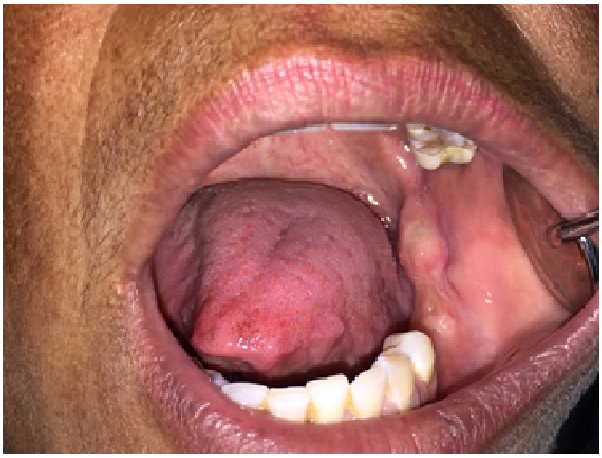

Figure 2. Intra-oral view revealing a solitary swelling in the left retromolar region

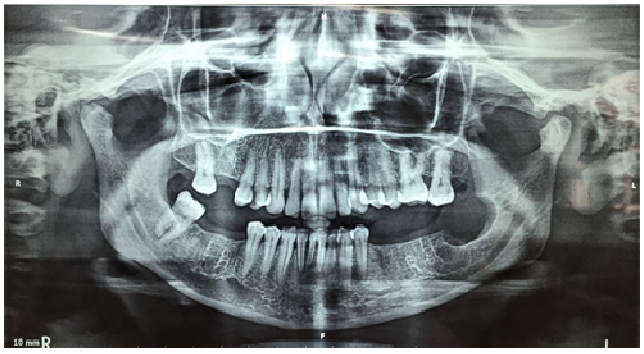

Figure 3. Orthopentomogram showing well-defined radiolucency

DISCUSSION

The odontogenic keratocyst is a developmental odontogenic cyst with a tendency to recur, characterized by a histological appearance of parakeratinized lining epithelium with palisaded ameloblast-like basal cells (Odell, 2017). It is a cyst derived from the remnants (rests) of the dental lamina (Rajendran, 2009). The term odontogenic keratocyst (OKC) was first coined by Philipsen (1956).

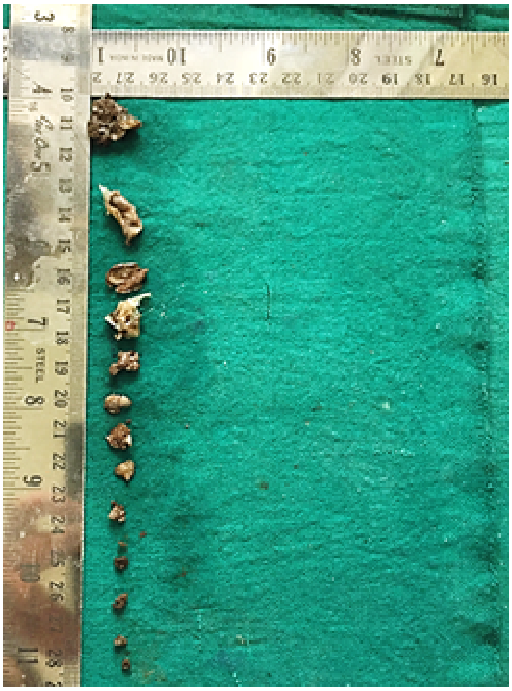

Figure 5. Grossing specimen

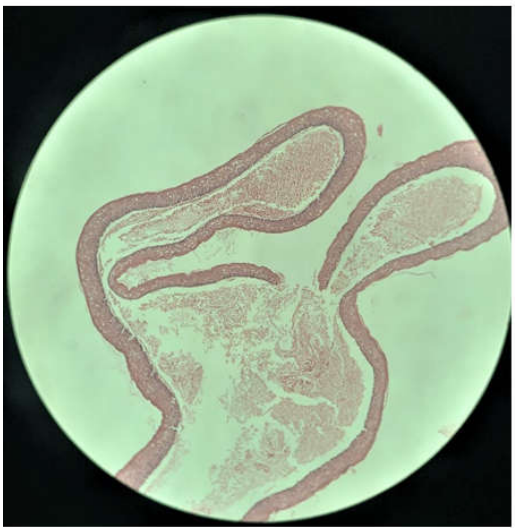

Figure 6. Photomicrograph at 10x revealing cystic lining and underlying stroma

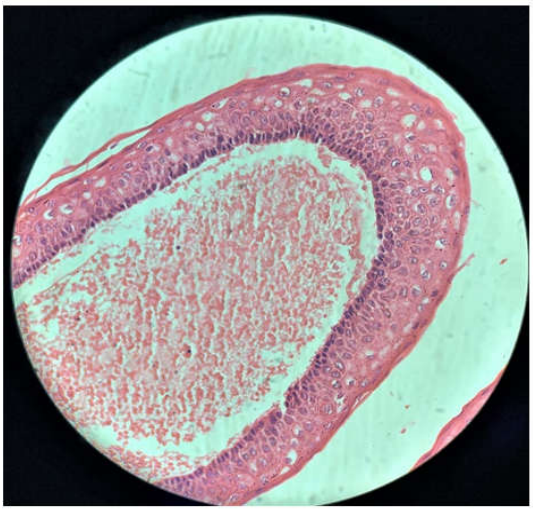

Figure 7. Photomicrograph at 40x of the cystic lining

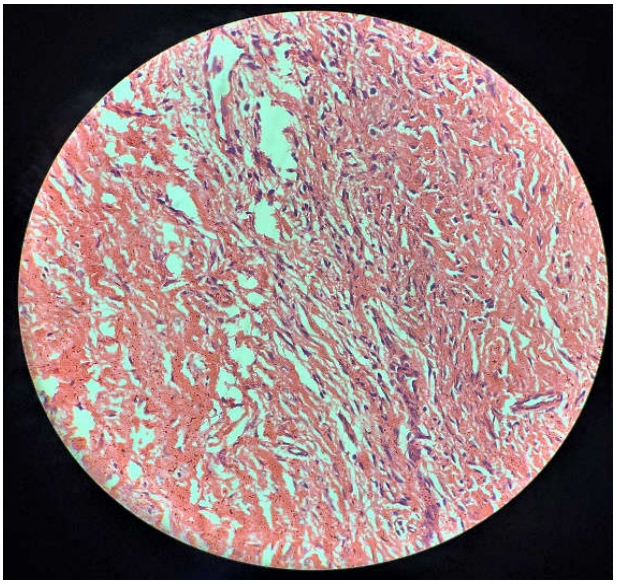

Figure 8. Photomicrograph at 40x shows the connective tissue stroma.

Pindborg and Hansen (1963) designated the term 'keratocyst' which was used to describe any jaw cyst in which keratin was formed to a large extent. Lucas (1972) made the point that emphasis that has been placed on keratinization is to some extent misleading; cysts of different types may all keratinize and if they do they are then liable to recur (Shear, 2008). Toller (1967) suggested that OKCs might be regarded as benign cystic neoplasms (Rajendran, 2009). According to WHO 1992 classification OKC is included in the odontogenic variety but according to WHO classification of benign tumors 2005 (Barnes *et al*. 2005) the odontogenic keratocyst is defined as benign keratocystic odontogenic tumor (KCOT) (Sumbh, 2017). KCOT was later reclassified as OKC under cyst of developmental origin (Soluk-Tekkesin, 2018). One of the characteristic features of the growth of this pathology is the tendency to grow along the cancellous channels with very little cortical expansion. Various theories of expansion of KCOT have been proposed to explain this. These include intraluminal hyperosmolality, active epithelial proliferation, collagenolytic activity of the cyst wall and synthesis of interleukin 1 and 6 by keratinocytes.[6,7]High proliferation rate, over expression of anti-apoptotic proteins (bcl-2) and expression of matrix metalloproteinase (MMPs 2 and 9) also favor growth and expansion of OKCs. Mutation in PTCH 1 ("patched") gene has also been considered as responsible for the pathogenesis of this cyst (Nayak, 2013). PTCH (Patched) is a tumor suppressor gene involved in both nevoid basal cell carcinoma syndrome and odontogenic keratocyst; mutations occur on chromosome 9q22.3–q31 (Cohen, 1999). Usually PTCH forms a receptor complex with the oncogene SMO for the SHH (Sonic hedgehog) ligand. SHH binding to PTCH releases the inhibition of growth signal transduction seen in PTCH binding to SMO. Thus the proliferating–stimulating effects of SMO are then predominant as seen in these conditions. The neoplastic capability and high recurrence have been attributed to a higher frequency of occurrence of proliferating nuclear antigen Ki67, p53 and bcl-2 positivity (Barnes, 2005). The odontogenic keratocyst can occur anywhere within the jaws, and examples within the gingival soft tissues have even been reported. As seen in the present case, approximately 65% to 75% of cases are seen in the mandible, with a predilection for the molar/ramus area (Gnepp, 2009).

It can occur at any age with peak incidence in the 2nd to 3rd decade of life and mean age being 32.3 years (Damm, 2002). Odontogenic keratocysts, like other jaw cysts, are symptomless until the bone is expanded or they become infected, both rare features in this cyst type.[1]The patient, in this case reported to the department with an asymptomatic swelling with no history of pain. Multiple OKCs can be seen in children which is a component of Nevoid Basal Cell Carcinoma Syndrome (Rajendran, 2009). It presents as a unilocular radiolucency with a well-defined peripheral rim, which stands true to the present case. Scalloping of border represents variation in growth pattern of cyst. Multilocular radiolucency, seen in 20% cases represents a central cavity having satellite cysts which is more frequent in larger lesions. It may contain crown of retained tooth within lumen; thus mimicking dentigerous cyst. Proximity to roots of adjacent teeth results in resorption or displacement (Rajendran, 2009). Large mandibular OKCs grow in a mesiodistal direction along the length of the bone, with minimal buccolingual expansion. An OKC located in the maxilla reveals expansion of the cortical bone, which can be seen as a bone deformity (Borghesi *et al*., 2018). Patients usually seek help from doctors if deformities in the bones are noticed or if spillage of cystic pus or a fistula is present (Hadziabdic, 2019).

On histopathology, the epithelium is distinctive with uniformly thin epithelial lining of six to eight cell layers and does not demonstrate rete ridges.[15]Parakeratinized surface is usually corrugated, rippled, or wrinkled with a prominent palisaded basal layer of cells showing tombstone or picket fence appearance. These characteristic features could be appreciated in the present case. In presence of inflammation, the epithelium loses its keratinized surface, may thicken & develop rete ridges or may ulcerate (Rajendran, 2009). Orthokeratinized odontogenic cyst (OOC) is an odontogenic cyst was initially termed as the uncommon orthokeratinized type of odontogenic keratocyst by the World Health Organization. It usually occurs in mandible (Sarvaiya *et al*., 2014). Variants of OKC that only produces orthokeratin acts different than most OKC. It is mostly found in dentigerous association around mandibular 3rd molar. It is less aggressive. It does not have hyperchromatic basal layer and are not associated with basal cell nevus carcinoma syndrome.[2]Orthokeratinized odontogenic cyst is now classified under developmental odontogenic cysts as an independent entity (Kamat, 2018). The treatment of choice is surgical enucleation with wide margin which may prove difficult if the cyst wall is thin & friable that can easily fragment. Perforation of cortical bone (especially in ramus) may complicate total removal. It has high recurrence rate of 13-60% (Rajendran, 2009). In presence of inflammation, there is a statistically significant increase in the proliferative activity of multilocular and multiple cysts, whereas there is no significant increase in proliferation seen in unilocular OKCs. This suggest that inflammation increases the neoplastic behavior. Therefore, it was suggested that aggressive treatment should be reserved for selective cases, in contrary to other authors, who believe that all OKCs behave as a tumor and should be treated aggressively.

Conclusion

Odontogenic keratocysts has been the subject of much debate over the years with respect to its origin, its growth, and treatment modalities. Most cases of OKCs are aggressive and

has a high recurrence rate. Thus, the clinical, radiographic, and histopathological correlations are essential for proper diagnosis, treatment and follow up.

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