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Case Study

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KERATOCYSTIC ODONTOGENIC TUMOR: A CASE REPORT

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ABSTRACT

The occurrence of jaw cysts constitute approximately 17% of all the intraoral pathologies. In routine clinical practice periapical cyst is the most encountered odontogenic cyst, followed by dentigerous and odontogenic keratocysts (OKCs). Odontogenic keratocysts (OKCs) of the jaws are developmental cysts arising from cell rests of the dental lamina (*cell rest of Serres*). The purpose of this case report is to study the etiopathogenesis and clinical manifestations of KCOTs, particularly in association with hypodontia. The aim of the paper is to understand all aspects of the entity including accurate diagnosis, treatment and prognosis of the lesion.

INTRODUCTION

In 2005, WHO defined keratocysts as benign, uni- or multi-cystic intraosseous neoplasms of odontogenic origin. These cysts have a characteristic thick, parakeratinized, stratified, squamous epithelial lining with a potentially aggressive and infiltrating ability.[1] In the same year, WHO, reclassified this lesion as benign keratocystic odontogenic tumor (KCOT) due to its high risk of recurrence, aggressive clinical course, mutations in the tumor suppressor gene (PTCH1), the occurrence of satellite cysts and the association with the Gorlin–Goltz syndrome.[2]

However, in year 2017, there was inadequate evidence to support this lesion as a neoplastic lesion therefore, the WHO released a revised classification of head and neck tumors in which KCOT was re-classified as the cyst instead of tumor, under the name of odontogenic keratocyst (OKC). The KCOTs has shown equal gender prevalence having peaks of occurrence in the second and fourth decades of life. They are commonly seen in the region of posterior mandible (65-70 % of cases), especially in the region of body and ascending ramus.[1,2]

CASE REPORT

A 32-year-old male patient came to the Department of Oral & Maxillofacial Pathology and Microbiology with a complaint of a swelling in right side of the face since 1 year and had no complaint of pain or discomfort (figure 1). The patient had no positive, associated medical, dental and family history. Intraoral examination revealed swelling extending from 44 to 46. The overlying mucosa was intact, non-ulcerated and showed no proliferation. On examination, displacement of 31, 41, 42 was observed, 43 and 48 were missing and all the associated teeth were vital. On extra-oral examination facial asymmetry was observed as mild diffuse swelling on right lower third of face, extending from, about 1cm below right commissure of mouth to the lower border of mandible. On palpation, no lymph nodes were palpable in association with the lesion (figure 2).

Figure 1: Extra oral examination.

Figure 2: Intraoral examination.

On radiographic examination, an OPG, revealed a well-defined radiolucency with corticated borders, crossing the midline, extending from the mesial aspect of 35 to the distal aspect of 47 mediolaterally and from the alveolar ridge region to the inferior border of the mandible supero-inferiorly. The 43 was seen impacted within the radiolucency. The radiolucency is seen pushing 33, 34, 41 and 42 distally and 44 mesially. The tooth 32 was missing. There was also loss of lamina dura wrt. 31, 33, 34, 41, 42, 44, 45, 46 and 47 (figure 3). The haematological evaluation showed no relevant imbalance of haematological parameters.

An incisional biopsy was performed under local anaesthesia and the mass of 1.5 x 3.0 x 0.7 cm in size was excised. This sample was then sent to Oral & Maxillofacial Pathology Department for histopathological examination.

Considering the patient history, clinical and radiographic examination a provisional diagnosis of dentigerous cyst was made but this differential diagnosis was still in question, as the dentigerous cyst is mostly seen in posterior aspect of mandible with impacted molar. A differential diagnosis of ameloblastoma was also made.

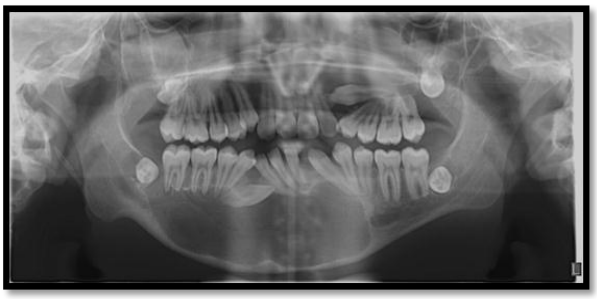

Figure 3: Orthopantogram.

Histopathological examination

The two bits of soft tissue specimen were excised from the lesion. These were creamish-brown in colour, firm in consistency & ovoid in shape which measured approximately about 1.5 x 3.0 x 0.7 cm in size (figure 5).

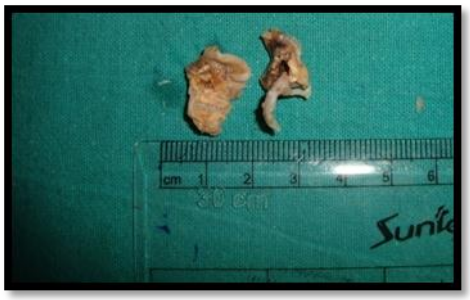

Figure 5: Grossing.

The H&E section, on evaluation, showed parakeratinized stratified squamous epithelial lining resting over the connective tissue stroma. The epithelial lining had uniform thickness consisting of 7 to 10 cells exhibiting corrugation. The epithelium–connective tissue interface was flat with no rete pegs formation. The basal cell layer was made of columnar cells that exhibit a characteristic palisaded pattern with polarized and intensely stained nuclei of uniform diameter. The fibro-cellular connective tissue stroma constitutes of mild inflammatory cell infiltrate mainly lymphocytes. The cyst wall also constitutes of spindle shaped fibroblasts and few blood capillaries.

Overall features were suggestive of "Inflamed Keratocystic odontogenic tumor". (figure: 6-8)

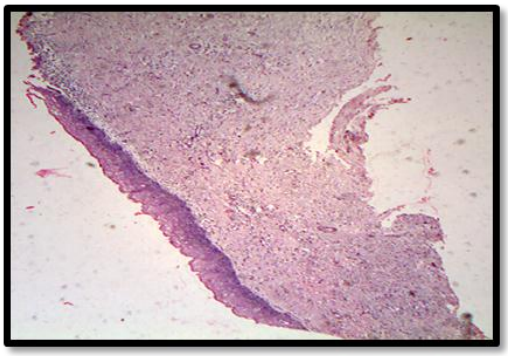

Figure 6: 4x: it shows parakeratinized stratified squamous epithelial lining and connective tissue wall.

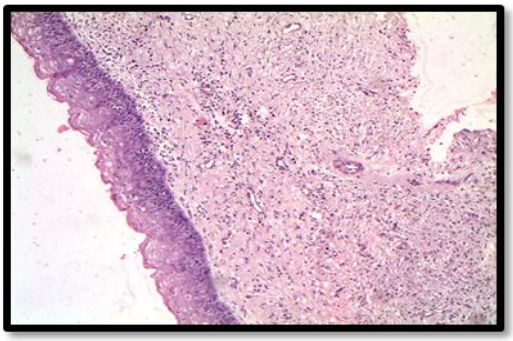

Figure 7: 10x: The epithelial lining is of uniform thickness and exhibits corrugation and without rete-ridges.

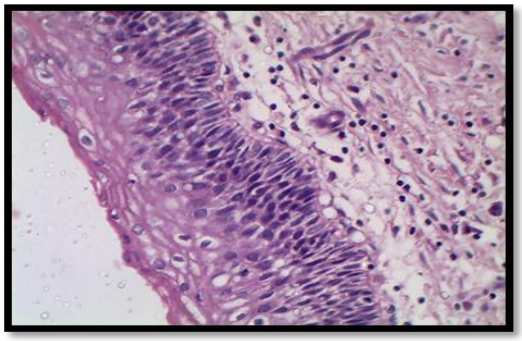

Figure 6: 40x: The basal layer has tall columnar cells with palisaded arrangement of the nuclei. The fibro-cellular stromal wall shows spindle shaped fibroblasts, few capillaries & mild chronic inflammatory infiltrates.

DISCUSSION

In 1992, the World Health Organization (WHO) categorized keratocysts as maxillary cysts of dysembryonic odontogenic origin. The well-known aggressive evolution of keratocysts, their histology, and new findings in genetics led the WHO in the year 2005 to reclassify these lesions as keratocystic odontogenic tumors (KCOTs).[1] It is widely accepted that KCOTs originate from odontogenic epithelium i.e. remnants of dental lamina. The proliferations of the basal cell layer of oral epithelium are also considered as possible sources of epithelial cells which may proliferate to form a KCOT. The aetiology of KCOTs is also strongly related to genetic factors particularly to mutation of tumor-suppressor PTCH gene, which is an important part of Sonic hedgehog (SHH) signalling pathway. The PTCH gene encodes PTCH transmembrane protein, which, together with SMO (smoothened), forms a receptor for SHH ligands and suppresses SMO mediated transcription of cellular proliferation genes.[4] Therefore, lack of PTCH function results in increased transcription of genes responsible for cell proliferation and leads in tumor formation.[5]

Various studies suggest that dysregulation of cell cycle and proliferation may also be important for KCOT pathogenesis. It is believed that KCOTs show increased cell proliferation rates and such phenomenon may be related to its aggressive growth. The KCOTs have equal prediction for both males and females in the second and fourth decades of life. They are more common in the posterior mandible especially in the posterior body and ascending ramus.[6] Clinically it manifests as a fast-growing aching tumefaction, with infiltration and expansion in the cortical bone. It may cause rhizolysis and dental dislocation. On radiography, a keratocyst appears as a uni- or multilocular radiotransparency. Alternatively, it can surround the crown of an impacted tooth, potentially being confused with a dentigerous cyst.[7]

Microscopically KCOT exhibits a uniform layer of parakeratotic stratified squamous epithelium. The epithelial lining is relatively thin, usually consisting of up to 6-8 layers, with characteristic flat connective tissue interface. The basal epithelial layer consists of palisaded cuboidal or columnar cells, which are frequently hyperchromatic.[1] The superficial layer is usually corrugated, consisting of flattened parakeratotic cells. The fibrous layer is thin and typically without inflammatory infiltrate. Within this part of KCOTs wall, proliferations of odontogenic epithelium and formation of microcysts may be observed.

KCOTs have thin and fragile walls therefore recurrence after the surgery is what makes surgical treatment considerably complicated compared to other cystic lesions of the jaws. Still, being a benign lesion without significant tendency for malignant transformation, routine use of radical surgery (such as resection of involved jaw) is questionable, both from medical and ethical point of view.[8] Therefore, it is not surprising that numerous adjunctive techniques have been developed for treatment of KCOTs. Establishing the balance between effective reduction of recurrence risk and selection of the least aggressive surgical procedure for each individual patient is a basic principle in treatment planning for these lesions.[9]

CONCLUSION

Due to the variation in clinical and radiographic appearances, KCOTs possess certain difficulties while making correct diagnosis for the clinicians. It is often falsely misinterpreted on radiographic evaluation as any other tumor/cyctic lesion. Hence, an accurate histopathological evaluation must be performed for accurate definitive diagnosis and appropriate management of the lesion.

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