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RESEARCH ARTICLE

CALCIFYING EPITHELIAL ODONTOGENIC TUMOR- A RARE CASE IN THE MAXILLA

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ABSTRACT

Calcifying epithelial odontogenic tumor (CEOT) is an uncommon odontogenic tumor with well-known histopathological features. Most often located in the posterior mandible it is rarely seen in the maxilla. Appearing in the second to third decade, CEOT is a slow growing neoplasm with rare malignant potential. Even though some investigators advocate conservative approach as the cure, others consider radical surgical excision to evade recurrence or malignant transformation. We hereby present a rare case in a 28 year old female patient with the occurrence in the maxilla.

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INTRODUCTION

Calcifying epithelial odontogenic tumor (CEOT) is a benign, occasionally locally aggressive neoplasm of odontogenic origin (Pindborg *et al*., 1955), invading soft tissue and bone, forming 0.4-3% of all intra osseous tumors (Franklin *et al*., 1976 and Philipsen *et al*., 2000). First described by Pindborg in 1955, WHO has now classified this tumor as a rare benign odontogenic neoplasm, affecting mandibular impacted teeth in the posterior region and sometimes the maxilla (Kaushal *et al*., 2012 and Whitt *et al*., 2012). The tumor has a less recurrence rate in comparison to ameloblastoma with an overall good prognosis (Whitt *et al*., 2012). The article reports a rare case of CEOT of the maxilla.

Case Report

A 28 year old female patient reported to the department of Oral Medicine & Radiology with a painless slow growing swelling in the maxillary left posterior region since 4 months. She noticed a slight swelling intraorally which grew to the present size of 3cmx 3cm. The family and medical history were non-contributory.

On extra oral examination there was a diffuse swelling in the maxillary sinus region. The temperature over the swelling was normal with no lymphnode involvement. Intra oral examination revealed absence of maxillary left second premolar and an over-retained deciduous second molar. Maxillary left Canine and first premolar appeared to be grade one mobile. The overlying mucosa was normal with a slight obliteration of the buccal vestibule. Radiographic examination (orthopantamograph) showed a well-defined unilocular radiolucent lesion on the left side of the maxilla (Fig.1) extending anterio-posteriorly from the distal surface of first premolar to mesial surface of second molar and superior-inferiorly from floor of maxillary sinus to alveolar crest. The first premolar appears to be mesially inclined along with an overretained maxillary left deciduous second molar and mandibular left deciduous first molar. Depending on clinical and radiographic features a provisional diagnosis of benign odontogenic neoplasm such as CEOT was given with a differential diagnosis of odontogenic cyst, fibro-osseous lesion or a cement-ossifying fibroma. An incisional biopsy was performed and sent for routine histopathological examination.
Histopathologically, the tumor comprised of islands of odontogenic epithelium within the connective tissue. (Fig.2, 3) The cells show hyperchromatic nucleus with prominent inter cellular bridges. (Fig.4) The section was composed of abundant eosionphilic material which at places was undergoing calcification resembling 'Liesegang Rings' (Fig. 5) The tissue

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was stained with Congo Red which gave a pinkish-red stain to the eosinophilic amyloid like material (Fig. 6). Based on the above findings a final diagnosis of Calcifying Epithelial Odontogenic Tumor was made.

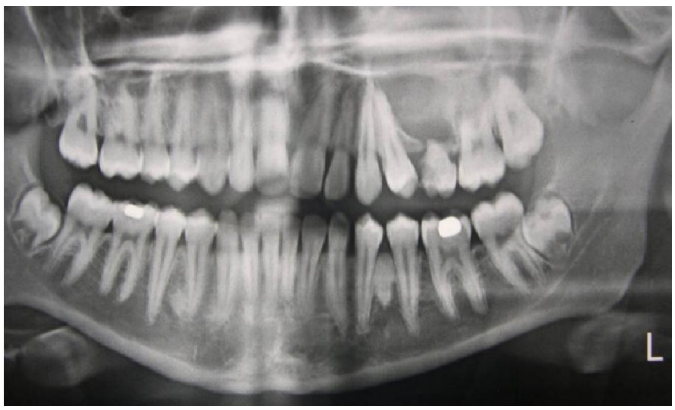

Fig. 1. Well-defined unilocular radiolucent lesion on the left side of the maxilla

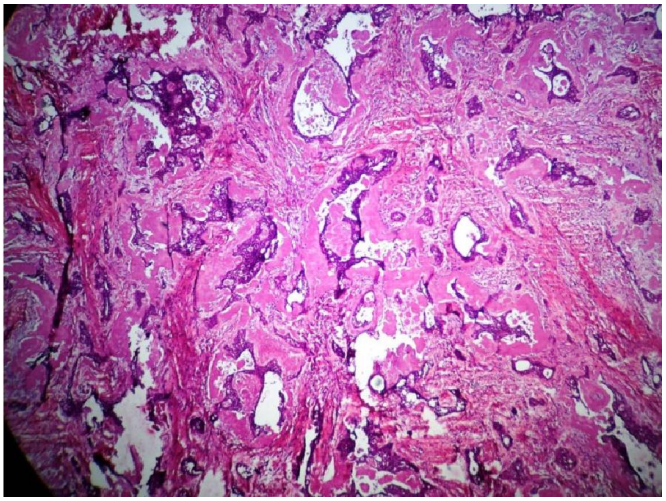

Fig. 2. Islands of odontogenic epithelium within the connective tissue. (H & E, 4x)

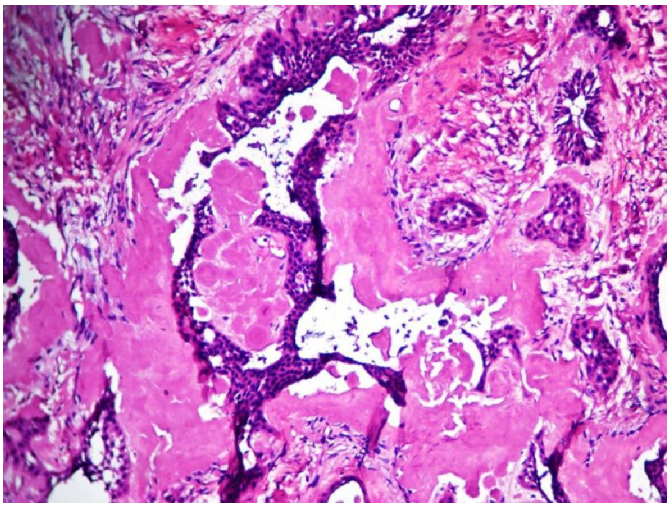

Fig. 3. Islands of odontogenic epithelium within the connective tissue. (H & E, 10x)

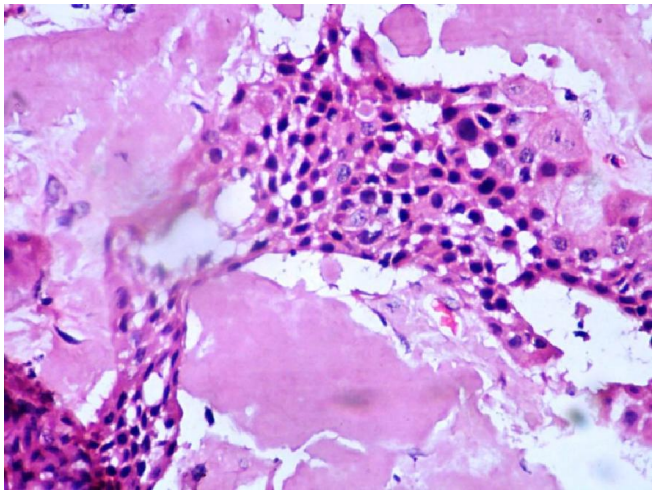

Fig. 4. Cells show hyperchromatic nucleus with prominent inter cellular bridges (H & E, 40x)

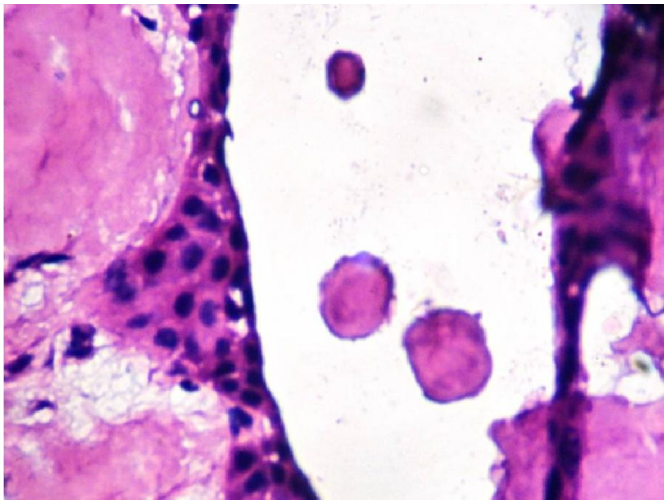

Fig. 5. Eosinophilic material undergoing calcification resembling Liesegang Rings (H & E, 40x)

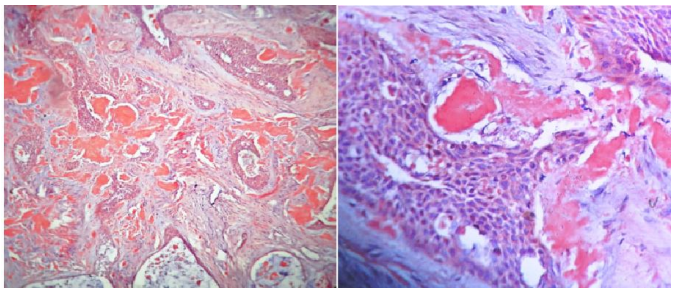

Fig. 6. Pinkish red stain of amyloid (Congo Red, 10x)

DISCUSSION

CEOT is an odontogenic epithelial tumor which is rare in occurence. It accounts for <1% of all odontogenic tumors (Shetty *et al*., 2014). According to WHO classification of 1992, CEOT is a "locally invasive epithelial neoplasm characterized by the development of intra-epithelial structures, probably of an amyloid-like nature, which may be calcified and which may be liberated as the cells breakdown." (More *et al*., 2015; Lim *et al*., 2005; Vinod *et al*., 2011 and Malik *et al*., 2014) It occurs more commonly in the mandible, and the

ratio of the occurrence in mandible to maxilla is 2:1. It has been seen that in the Asian population, the CEOT shows a predilection for the maxilla, whereas in the Western population it shows a higher mandibular prevalence (Rani *et al*., 2016 and Ng and Siar, 1996). It is most often located in the premolar-molar region of the mandible and may be associated with one or more impacted tooth (More *et al*. 2015 Lim *et al*., 2005; Müller *et al*., 2012; and Kaplan, 2001). The mean age of occurrence of this tumor is 40 years though it may range from 20 to 60 years and has no gender predilection. Majority of cases are intraosseous in nature with only about 6% of them arising in extraosseous locations (Rani *et al*., 2016 and Philipsen, 2000). The tumor may present as a painless, slow growing swelling (Shetty, 2014). Rarely, it may be associated with paresthesia (More *et al*., 2015; Lim *et al*., 2005 and Deboni *et al*., 2006). Histogenesis is uncertain, though it is believed to arise from stratum intermedium of dental lamina. This belief is based on the morphological resemblance of tumor cells to that stratum intermedium and a high activity of alkaline phosphatase and adenosine triphosphate in both these cells (Shetty, 2014). The radiographic appearance of CEOT varies, depending on the stage of development; it may either present as well-defined radiolucency, mixed radiolucent-radiopaque or a completely radio-opaque mass. It is seen that the lesion also concurrently erodes bone and thus, it often appears as mixed radiolucent-radiopaque mass with many small irregular trabeculae traversing the radiolucent area giving a characteristic "driven snow" appearance due to scattered flecks of calcification (More *et al*., 2015; Lim *et al*., 2005; Vinod *et al*., 2011; Kaushal *et al*. 2012; Tanimoto *et al*., 1988 and Wood *et al*., 1997). The rediolucency may be unilocular or multilocular radiolucency (Shetty *et al*., 2014). In our case, it was unilocular. The histologic findings of CEOT are well defined. The tumor consists of polyhedral cells arranged in masses, islands, sheets, rows, cords or strands in a scanty connective tissue stroma. The cells have well-defined borders and are pleomorphic. They have a prominent nucleoli and abundant finely granular cytoplasm which is filled with an eosinophilic "amyloid-like" material, which gradually becomes concentric calcified deposits, resembling psammoma bodies called the "Liesegang rings". This is considered as pathognomonic for this tumor. The round shaped eosinophilic amyloid material stains positive for Congo red and appears as an apple-green birefringence under a polarized microscope (More *et al*., 2015; Lim *et al*., 2005; Vinod *et al*., 2011 and Neville *et al*., 2008 and Lin *et al*. 2013). All these features were evident in our case. It is suggested that CEOT of the maxilla should be treated more aggressively as maxillary tumors grow more rapidly and are usually not well confined. Treatment, however, should be individualized for each case (More *et al*., 2015; Lim *et al*., 2005 and Vinod *et al*., 2011). The prognosis of the tumor will dictate the extension of the surgical margins. Hence, several prognostic factors have been proposed to estimate recurrence risk. Microscopically, less amyloid aggregation and foci of calcification have been suggested in association with more aggressive tumor behavior (Foroughi *et al*., 2015 and Sadeghi *et al*., 1982).

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