Case Report

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CALCIFYING EPITHELIAL ODONTOGENIC TUMOR: A RARE ENTITY

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ABSTRACT:

The calcifying epithelial odontogenic tumor (CEOT) is a rare benign, locally invasive neoplasm which accounts for 1% of all odontogenic tumors. It was first described as a distinct entity by Jens J Pindborg in the year 1955, hence also termed as Pindborg's tumor. CEOT manifests in 3rd-6th decade of life with no specific gender predilection. Clinically it presents as a slow growing painless swelling with expansion of buccal cortex and sometimes may be associated with pain. The intraosseous variant of CEOT is more common, predominantly occurring in posterior mandible and it rarely manifests extraosseously. Radiographically it depicts varying features and is found to be associated with impacted tooth in 50% of cases. We report a case of CEOT in left posterior mandible that presented a perplexing enigma radiographically, noted as a multilocular radiolucency displacing impacted second molar and manifesting characteristic histological features of CEOT.

KEYWORDS: Odontogenic neoplasm, Pindborg tumor, mandible, impacted tooth

INTRODUCTION:

The calcifying epithelial odontogenic tumour (CEOT) also known as Pindborg tumor is considered a rare, benign, locally destructive epithelial odontogenic neoplasm.[1] CEOT accounts for 1% of all odontogenic tumours occurring in age group of 20 and 60 years of age, with no gender predilection.[1] These lesions mostly arise centrally within the jaws especially in the premolar-molar region of the mandible, although it may rarely manifest peripherally in the anterior maxillary or mandibular gingiva.[2] The tumor develops slowly as an asymptomatic, expansile mass of the jaw.[3-4] As the tumor grows and extends into adjacent structures, it may produce cortical expansion, tooth movement, and root resorption of associated teeth.[5] It has a varying radiographic behavior creating a dilemma for diagnosis.[6] In a study of 67 Pindborg tumors by Kaplan et al, the different patterns observed were mixed radiolucent and radiopaque pattern (65%), followed by the completely radiolucent pattern (32%) and the totally radiopaque pattern (3%).[7-8]

CASE DETAIL:

A 55 year old female patient reported to our department with a complaint of swelling in the left side of the lower jaw of 2 months duration. She had intermittent, dull-aching pain in the same region since

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1 month. On clinical examination, facial asymmetry was noted with diffuse swelling on left side of the face which was tender on palpation. Intraorally a well circumscribed swelling of approximately 4x3.5cm was seen in the left premolar-molar region (Figure-1).The swelling was covered by normal mucosa and was tender and hard in consistency on palpation with no signs of paresthesia. There was clinically missing second molar with mobility in first and third molar. A provisional diagnosis of ameloblastoma of left mandible was given considering the clinical findings and further patient was subjected to radiological investigations. The radiographic examination disclosed a well defined multilocular radiolucency on Orthopantomogram (OPG) with displaced second molar towards the lower border of mandible. Root resorption was evident in relation to second molar (Figure-2). A mandibular occlusal radiograph revealed expansion of the buccal cortical plate with intact lingual cortex. Based on the clinical and radiographic findings the differential diagnosis included dentigerous cyst at first, as it is commonly seen in posterior mandible associated with impacted tooth. Ameloblastoma was considered next, as it is more common in females in posterior mandible showing multilocular radiographic appearance with expansion and root resorption. Calcifying epithelial odontogenic tumor (CEOT) was next in the line as it has prevalence in this age and commonly presents as painless swelling in posterior mandible and demonstrates radiolucency in initial stages with impacted tooth that may be displaced, these features closely relates it with the present case. Ossifying fibroma and odontogenic myxoma were also included in differential diagnosis as they mostly occur in posterior mandible and may be associated with impacted tooth. Further, an incisional biopsy was performed and the tissue specimen was sent for histological examination, H& E stained sections showed closely packed sheets of polyhedral epithelial cells with multiple giant nuclei, distinct cell outline and prominent intercellular bridges. Homogenous eosinophilic material was noted in between these cells with calcifications in few areas suggestive of a histopathological diagnosis of CEOT (Figure-3). The treatment included hemimandibulectomy followed by surgical reconstruction using 2.5mm reconstruction plate under general anesthesia and arch-bar fixation was done for stabilization (Figure 4). Patient was under continuous surveillance and no recurrence is reported till date.

DISCUSSION:

The calcifying epithelial odontogenic tumour (CEOT) is considered by World health organization (WHO) as a benign, locally invasive epithelial odontogenic neoplasm.[1], also widely known by its eponymous term, Pindborg tumour.[2] CEOT accounts for 1% of all odontogenic tumours occurring in patients between 20 and 60 years of age, with a mean around 40 years and has no gender predilection.[1] Most of these lesions arise centrally within the jaws

especially in the premolar-molar region of the mandible. A rare peripheral variant that mostly occurs in the anterior maxillary or mandibular gingiva has also been noted in about 5% of cases.[2]

The origin of this tumor remains elusive, but several hypotheses have been proposed. It is postulated that intraosseous variant may originate from the stratum intermedium layer of the enamel organ while extraosseous variant may arise from the remnants of dental lamina or basal cells of gingival epithelium.[3-4] The tumor presents as a slow-growing, asymptomatic, expansile mass of the jaw. Peripheral lesions appear as firm, painless gingival swelling. Although the CEOT is a benign neoplasm, it shows variation in its behavior ranging from very mild to moderate invasiveness. As the tumor grows and infiltrates into adjacent structures, it may produce cortical expansion, tooth movement, and root resorption of associated teeth.[5]

CEOT presents with variable radiographic patterns based on its developmental phases.[6] An early tumor may appear completely radiolucent, As the tumor matures and enlarge, they will have a mixed radiolucent-radiopaque appearance, although some larger tumors will remain radiolucent.[5] Further they become completely radiopaque. In one study of 67 Pindborg tumors by Kaplan et al, the mixed radiolucent and radiopaque pattern occurred most often (65%), followed by the completely radiolucent pattern (32%) and, least often, the totally radiopaque pattern (3%). According to Franklin and Pindborg the most characteristic presentation of CEOT is a radiolucency associated with an impacted or unerupted tooth, mostly first or second mandibular molar that may be displaced inferiorly causing bulging of the inferior cortex. The calcified material may be seen within the radiolucency around the crown of unerupted tooth. This radiopaque flecks of calcification may sometimes coalesce or form linear streaks that crisscross resembling “driven-snow” in appearance, though rarely reported this characteristic feature is unique to CEOT.[7-8]

The present case showed a completely radiolucent pattern with impacted second molar displaced towards inferior border of the mandible. Although there was absence of calcification radiographically, other findings of the present case were in confinement with the characteristic features of CEOT as reported by Franklin & Pindborg. Similar radiographic findings were noted in a case of CEOT reported by Deboni et al.[9], which showed a lytic lesion in right body of mandible with impacted second molar dislocated to the inferior border.

The histological features of CEOT are identical. It shows sheets or nests of polyhedral epithelial cells with prominent intercellular bridges, pleomorphic nuclei and varying amounts of amyloid like homogenous eosinophilic material interspersed between the polyhedral cells. These characteristic microscopic features of CEOT were evident in our case. Occasionally

amyloid like substance undergo mineralization in the form of concentric calcifications known as liesegang rings, pathognomic of this tumor.[10]

The management of CEOT ranges from simple enucleation or curettage to hemimandibulectomy or hemimaxillectomy. In mandible mostly enucleation with curettage is recommended for smaller lesions but aggressive lesions may require surgical resection of the tumor with a safety margin of 1cm of normal bone.[5-11] Maxillary lesions appear to grow faster and may be associated with vital structures and hence a more aggressive treatment is needed. In a study by Pindborg it showed a recurrence rate of about 14%[11]. Since the lesion was of greater extension and to reduce the chances of recurrence, the choice of treatment in our case was segmental resection of left mandible followed by surgical reconstruction. Patient was under follow-up for two years and no recurrence is reported till date. The treatment rendered in present case is in confinement with other case reports by Deboni et al.[9] and Zanakis et al.[11]However treatment varies in each case depending on site, size, extent of the lesion and amount of bone destruction.[11]

Salient features of our case:

- Radiographically CEOT shows varied appearance with calcifications around unerupted/impacted tooth, In our case CEOT presented with multilocular radiolucency associated with impacted second molar but there were no calcifications evident.
- Histopathological examination demonstrated the characteristic features of CEOT with presence of calcifications.

CONCLUSION:

All the characteristic or pathognomonic features of any lesion or disease may not be evident in every case, the presentation varies in each patient, and therefore it is important to consider the variations in appearance of each entity that may provide a clue to the correct diagnosis.

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FIGURES:

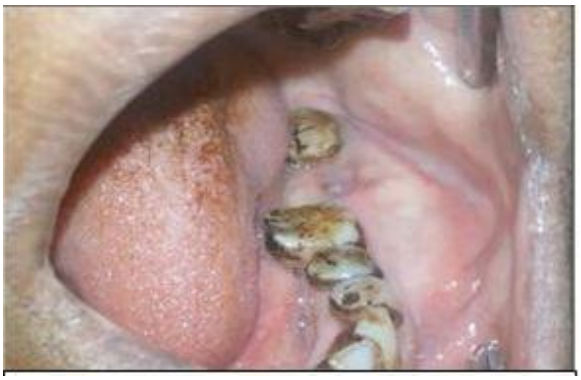

Figure 1: Swelling in the left mandible with missing second molar

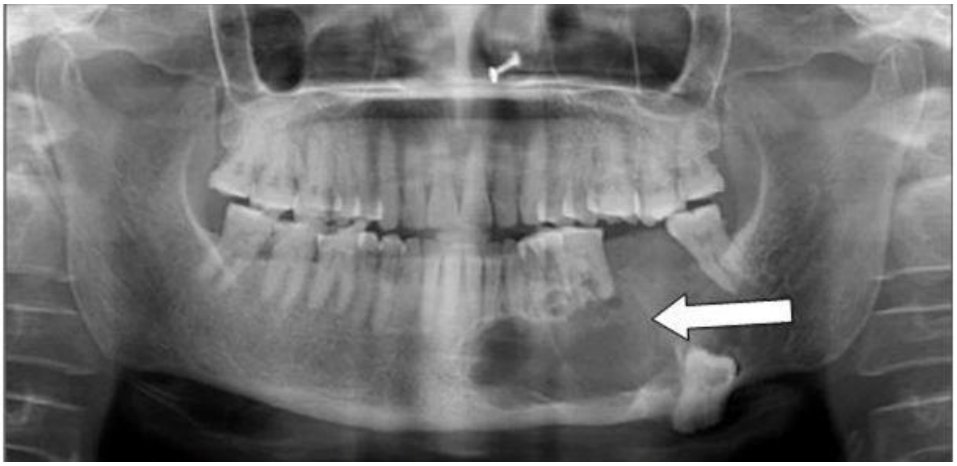

Figure 2: Orthopantomograph (OPG) showing a multilocular radiolucency with displaced unerupted second molar (white arrow)

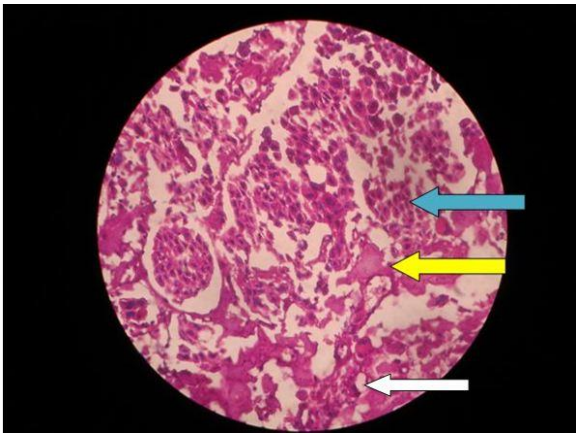

Figure 3: Microphotograph demonstrates closely packed sheets of polyhedral epithelial cells (blue arrow) with eosinophilic material (yellow arrow) and few calcifications (white arrow) {H & E stained, 40X}

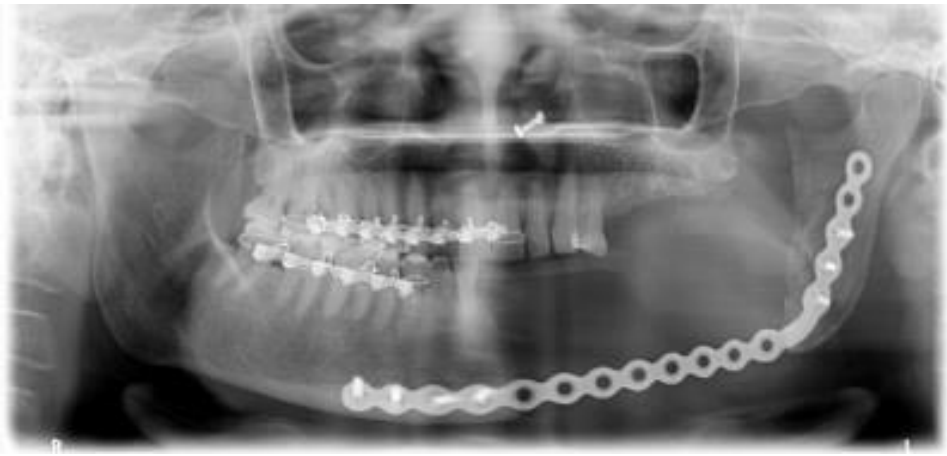

Figure 4: Post-operative orthopantomograph depicting reconstruction of bone with titanium plates and arch bar fixation