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CASE REPORT

CALCIFYING EPITHELIAL ODONTOGENIC TUMOR MIMICKING LIKE AN CALCIFYING ODONTOGENIC CYST: A CASE REPORT

Suryadeep Kaushik[1], V. Ramesh[2], M. Jonathan Daniel[3], B. Premlatha[4], P. Durgadevi[5]

[1]Post graduate resident, [2]Dean, Professor & Head, [4]Professor, Department of Oral Pathology and Microbiology, [3]Associate Dean, Professor & Head, [5]Post graduate resident, Department of Oral Medicine and Radiology, Mahatma Gandhi Post graduate Institute of Dental Sciences, Puducherry

ABSTRACT:
Calcifying epithelial odontogenic tumor (CEOT) is a benign odontogenic neoplasm with characteristic radiological and histopathological picture. CEOT is also known as Pindborg tumor. We are presenting a case of CEOT which despite its typical radiological picture was mimicking like an odontogenic cyst on histopathology and a correct diagnosis and treatment will definitely prevent any unwanted recurrences.
Key words: Calcifications, odontogenic tumour, pindorg tumour.

Corresponding author: Dr Suryadeep Kaushik, Post graduate resident, Department of Oral Pathology and Microbiology, Mahatma Gandhi Post graduate Institute of Dental Sciences, Puducherry

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INTRODUCTION:

Calcifying epithelial odontogenic tumor (CEOT) is an rare benign odontogenic neoplasm most of the times associated with impacted tooth and consisting of prominent epithelial cells with intercellular bridges and calcifications. Tumor usually occurs as a painless slow growing mass.

The entity was first described by Pindborg in 1955 as benign but locally aggressive and was termed as calcifying epithelial odontogenic tumor [1]. The term was accepted by world health organisation (W.H.O) in 1971 [2]. The frequency varies from 0.4% to 3% among all odontogenic tumors [3-5]. Approximately, 200 cases have been reported till date [6].

CASE REPORT:

A 24 year old female patient reported to our institute with complain of swelling in relation left lower back teeth with associated since past two months. There is no history of numbness, swelling or paresthesia in that region and also no underlying systemic illness. On extraoral examination the swelling was bony hard in consistency with single ipsilateral node palpable and was soft in consistency, mobile and tender. On intraoral examination 37 and 38 were missing and a swelling of size 4*4 cm^2 was present extending anteroposteriorly from distal of 36 to retromolar region and buccolingually with obliteration of buccal vestibule upto the floor of mouth (Figure 1).

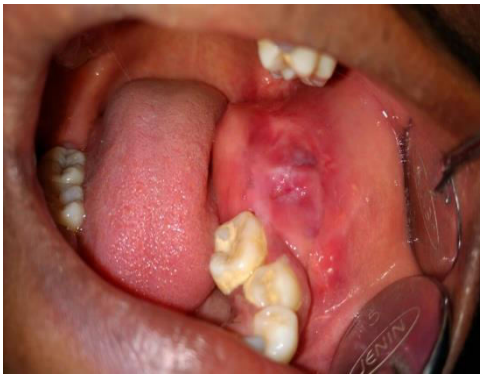


FIGURE 1: Intraoral swelling in retromolar region showing bony expansion and clinically missing 37 and 38.

On palpation the swelling was soft to firm in consistency, non fluctuant, non compressible with evidence of lingual cortical expansion and tenderness.

Intraoral periapical radiograph irt 36 and edentulous 37, 38 revealed a mixed radiolucent radiopaque lesion extending from distal aspect of 36 and the full extent is not covered. Internal aspect consists of foci of calcification and there was bone loss, root resorption with

respect to distal root of 36, widening of periodontal ligament space and loss of lamina dura irt 36 was noted. Digital Orthopantomogram revealed a well defined corticated mixed pericoronal radiolucency radiopacity extending from the mesial aspect of 36 till the sigmoid notch posteriorly. There was evidence of impacted tooth in close proximity to the inferior border with adjacent multiple scattered radiopaque foci. Inferior alveolar nerve canal could not be traced on the affected side (FIGURE 2).

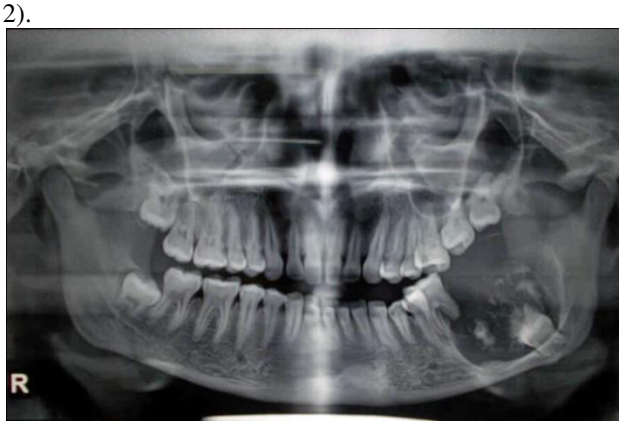
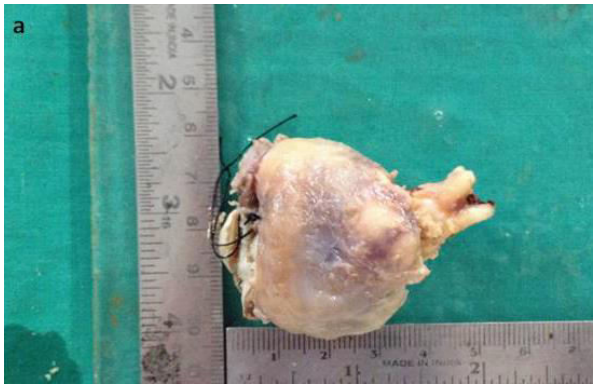


FIGURE 2: Orthopantomogram shows well defined corticated mixed pericoronal radiolucency radiopacity and impacted 37 with multiple scattered radiopaque foci.

Based on these clinical and radiographic findings a provisional diagnosis of calcifying epithelial odontogenic tumor was made.

Incisional biopsy was taken to plan for the appropriate treatment. The biopsy revealed thickened fibrous capsule with collagen fibres arranged in parallel wavy bundles and no cystic epithelial lining or any eosiniophilic structures even after serial sectioning and a diagnosis of capsule of a cyst was made. Based on the report surgery was done and the specimen attached to the neck of impacted tooth alongwith extracted 35, 36 and embedded 37 was sent to our department for histopathological confirmation.

On macroscopic, the excised specimen with embedded 37 was whitish yellow in colour interspersed brownish areas on cutting the specimen into 2/3rd and 1/3rd proportion the central lesional area showed papillary growth surrounded by thick fibrous capsule (FIGURE 3).



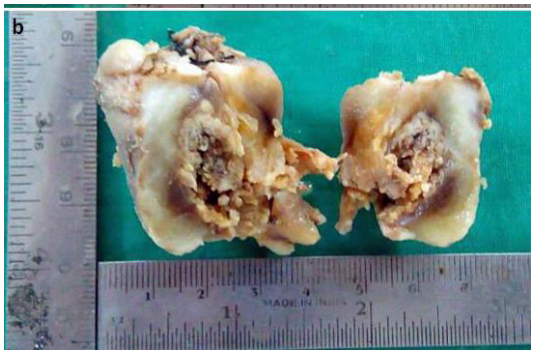


FIGURE 3: a) Macroscopy shows 37 embedded into the specimen and b) on cutting revealed thick fibrous capsule with central lesional area.

Noting all these features tissue was taken comprising both of lesional area and the capsular part. The histopathology revealed 1) Sheets and islands of epithelial cells enclosed within the fibrous capsule 2) large epithelial cells surrounded by both intracellular and extracellular eosinophilic material 3) Areas showing globular calcified masses 4) No pleomorphism or mitoses were noted. Due to thickened fibrous capsule again serial sectioning was done and also a new tissue was cut from the specimen so as to rule out calcifying odontogenic cyst whose epithelium might have been stripped off during sectioning (FIGURE 4).

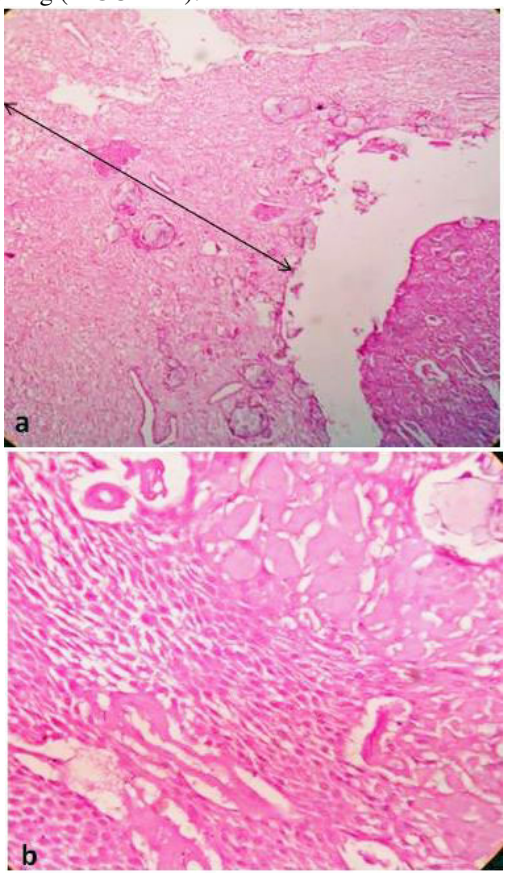


FIGURE 4: Histopathology shows a) thick fibrous capsule on left marked by an arrow (100 x magnification) and b) epithelial cells with prominent intercellular junctions and eosinophilic areas (400 x magnification).

Based on these histopathological features a diagnosis of calcifying epithelial odontogenic tumor was made.
The section was also stained for special stain Congo red which didn't reveal much apple green birefringence under polarized light apart from few eosinophilic areas lying adjacent to collagen fibers (FIGURE 5).

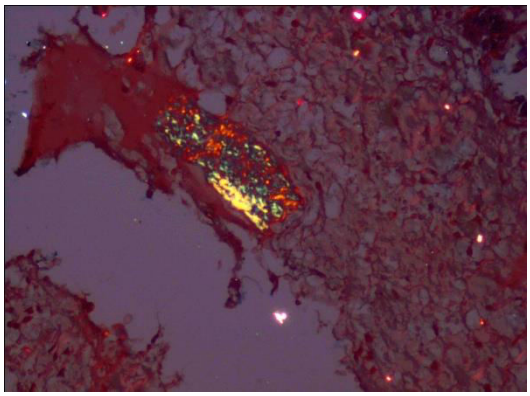


FIGURE 5: Special stain Congo red showing apple green birefringence.

DISCUSSION:-

Calcifying epithelial odontogenic tumor, as famously know as Pindborg tumor is a rare epithelial odontogenic tumor and derives attention because of the presence and nature of amorphous eosinophilic material. This material is reported to be derived from immune amyloid or amyloid of unknown origin by Franklin et al [7] while Mori et al reported the material to be positive for almost all protein reactions, which resembled those in enamel matrix [8]. A recent report also says this protein to be unique to tumor and both protein structure and DNA sequence of the responsible tumor has been described [6]. We used polarized microscopy to show the amyloid like material exhibiting apple green birefringence using the Congo red special stain. The birefringence was not shown in globular calcified regions but only in amorphous extracellular eosinophilic areas.
CEOT most commonly occurs as intraosseous variant but peripheral variants have also been reported. The intraosseous variant most commonly occurs in posterior mandibular region with a wide age range but most commonly encountered in 30-50 years of age. The tumor exhibits a unilocular or a multilocular radiolucent defect with margins of lytic defect often scalloped and well defined [6]. Scattered flecks of calcification have given arise to the term of driven snow appearance [9]. Reichart and phillipsen reported that 53% of CEOT's have an association with an unerupted molar and most frequently associated tooth is mandibular molar [10]. In our case all findings are confirmed the location is posterior mandible and the associated unerupted tooth was found out to be mandibular second molar based on its macroscopic examination which leads us to obvious assumption that third molar must be missing but the reason for missing molar creates confusion as it can be due to the effects of this slowly developing tumor or congenitally missing. But, comparison with other quadrants leads us to a hypothesis that the missing third molar might be due to the resorptive effects of the locally aggressive tumor and also the amount calcified specks seen on radiograph probably suggests that the lesion must be long standing and might have caused the damage to third molar in its early stages of calcification.
Due to less calcification in its early stages a unilocular radiolucency associated with impacted tooth can be mistaken for dentigerous cyst. In our case it was a unilocular mixed radiolucent – radiopaque lesion.
Histopathologically, CEOT usually shows typical picture of discrete islands, strands or sheets of polyhedral cells in a fibrous stroma. Intercellular bridges can be prominent among the epithelial cells. Some tumors may show pleomorphism but this should not lead to misdiagnosis of malignancy. Calcifications are seen within the amyloid material and form characteristic concentric liesegang rings [6]. These rings appear basophilic and were noted in our case. Another feature which can be noted in CEOT is cementum like material in stroma, clear tumor cells with foamy cytoplasm and if these cells are in majority then it know as clear cell CEOT. This clear cell variant can be distinguished from the CEOT as it lacks characteristic calcifications and amyloid like depositions. Langerhan cells and myoepithelial cells also have been observed in rare occasions. Combined CEOT and adenomatoid odontogenic tumor have also been reported [10-13]. Examples of CEOT showing aggressive growth and malignant behaviour have also been reported [14, 15].
In our case the diagnostic dilemma was due to too thickened fibrous capsule seen histologically surrounding the lesion in incisional biopsy, which brought the diagnosis of capsule of cyst. So, the reason which can lead to a non specific diagnosis as in our case was due to the deep lesion encapsulated by a capsule. If the lesion would not have been deep then it would have been easy to come to a conclusion in incisional biopsy and probably in the first few sections of the excisional biopsy itself reducing the chances of non specific diagnosis.
Though, the sections from excisional biopsy showed sheets of epithelial cells but a possibility of epithelial lining of a calcifying odontogenic cyst getting damaged or separated from the main section while sectioning due to calcifications present in the section was always there which led us to go for serial sectioning and new tissue to be taken from different area. Also another clue for going deep into the lesion was from the typical radiographic picture of the lesion.
Finally the diagnosis of tumor was made ruling out any possibility of it being a cyst as the treatment of the tumor requires a narrow rim of surrounding normal bone to be removed to prevent recurrences whereas cyst has just to be enucleated.

CONCLUSION:-

Based on all the findings in our case we have reached a conclusion that despite of its typical features its diagnosis should be made ruling out its cystic companion and its locally aggressive behaviour should not be underestimated as evident from missing third molar.

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