

Calcifying epithelial odontogenic tumor without calcification: A rare entity

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Abstract

Pindborg in 1955, for the first time described four cases of a rare benign odontogenic tumor. Shafer *et al* called this entity Pindborg tumor. This was later referred to as the calcifying epithelial odontogenic tumor. It accounts for less than 1% of all odontogenic neoplasm. This tumor is characterized mainly by the presence calcification and eosinophilic deposits resembling amyloid including presence of polygonal epithelial cells. It is very rare to find non calcifying epithelial odontogenic tumor with only 5 cases documented till .Here we present another case of non-calcifying Pindborg tumor, which first came to the department as a follicular tissue and was histologically diagnosed as a calcifying epithelial odontogenictumor. It is considered that noncalcifying Pindborgtumor is an aggressive variant, hence a long term follow-up after surgery is recommended.

Keywords: Calcifying Epithelial Odontogenic Tumor, Non-calcifying Odontogenic Tumor, Pleomorphic Cells, Pindborgs Tumor, Non-encapsulated.

Background

In 1955, Danish pathologist Jens Jorgen Pindborg first recognised an unusual entity which later came to be known as the Pindborg tumor, now known as calcifying epithelial odontogenic tumor (CEOT), it has been previously named as an adenoid ameloblastoma. Cystic odontoma, unusual ameloblastoma were few other names used for it.⁽¹⁾ Pindborgtumor is thought to be exclusively of epithelial origin.⁽¹⁾

The CEOT is a slowly growing, benign, but non encapsulated and locally invasive, epithelial, odontogenic neoplasm with a singular histomorphological pattern characterized by irregular sheets and islands of eosinophilic, polyhedral, and often pleomorphic cells, which eventually disintegrate into an eosinophilic, amorphous substance, which stains with amyloid markers and tend to calcify.⁽²⁾

Calcifying epithelial odontogenic tumor (CEOT) most commonly presents in the fourth to fifth decade of life as a slow growing intraosseous mass in the mandible. Male to female ratio of its presentation is nearly equal. The etiology is unknown, and no predisposing factors have been identified. This tumor is characterized mainly by the presence calcification and amyloid like eosinophilic deposits including presence of polygonal epithelial cells.⁽³⁾ Non-calcifying epithelial odontogenic tumor is rarely seen, with only five cases reported till date.⁽⁴⁻⁸⁾

In this article, we present an additional case of a non-calcifying Pindborg tumor. This is a unique case as it was diagnosed provisionally as a follicular tissue and histologically was later diagnosed as a non-calcifying epithelial odontogenic tumor.

Case Presentation

A 18- year old female patient was referred from department of Orthodontics to the Oral Surgery department for extraction of impacted para-premolar located between 34 and 35. There was no pain and swelling present in relation to para-premolar. No relevant family history present.

Investigations

On panoramic radiographic examination a parapremolar tooth was noticed situated periapically between 34 and 35. A radiolucency around the impacted tooth was noticed which was found to be within normal range thus denoting a follicle (Fig. 1). After extraction the follicular tissue was sent to our department for histopathological investigation (Fig. 2). H and E stained section showed that the tumor was well defined with a definite fibrous capsule. Microscopic examination revealed polyhedral epithelial cells arranged in sheets or strands and amorphous eosinophilic globules of amyloid-like materials in a loose fibrous connective tissue stroma (Fig. 3). The outline of the cell and the intercellular bridges between them were distinct. Cell cytoplasm was abundant and eosinophilic (Fig. 4). There were no calcified bodies (Liesegang rings) present in the tumor. A histopathological diagnosis of non-calcifying epithelial odontogenic tumour was made. The patient was kept under follow-up for any recurrence.



Fig. 1: Orthopantomograph showed a parapremolar tooth which was situated periapically between 34 and 35. A radiolucency around the impacted tooth was found to be within normal range thus denoting a follicle



Fig. 2: Grayish white follicular tissue for histopathological examination

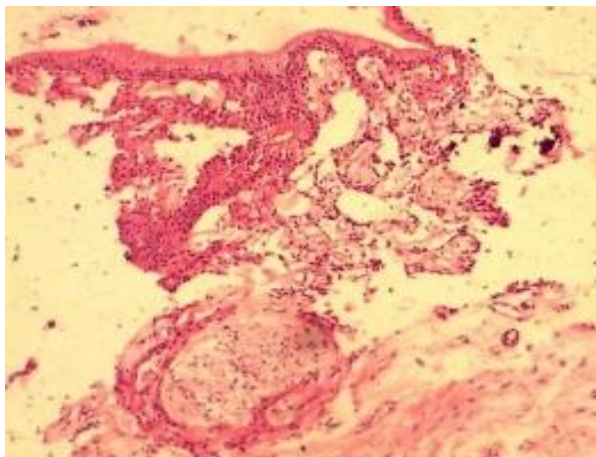


Fig. 3: 10xH and E stained section showed polyhedral epithelial cells arranged in sheets or strands and amorphous eosinophilic globules of amyloid-like materials in a loose fibrous connective tissue stroma

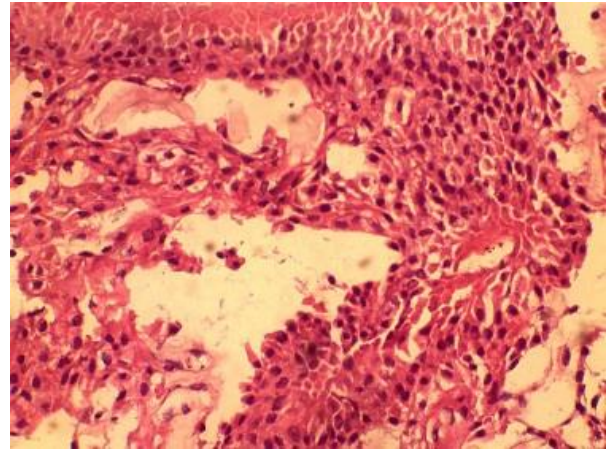


Fig. 4: 40x H & E stained section showed cell outlines and intercellular bridges were distinct. Cell cytoplasm was abundant and eosinophilic

Differential diagnosis: On the basis of radiograph and, clinical examination, a diagnosis of Dental follicle was made.

Treatment: Complete surgical excision of the lesion was performed.

Outcome and follow-up: After surgical excision of the lesion, regular follow up of the patient with repeated scans and radiographs has not revealed any signs.

Discussion

The histopathology and clinical features of CEOT have been well documented since its original description by Pindborg in 1955.^(9,10)

Classically, CEOT presents in the 4th to 6th decade of life with an equal incidence amongst the men and women. It mostly presents in the mandible as a painless slow growing mass. CEOT may present intraosseously that is the central type or it may present extraosseously, that is the peripheral type. 85% of the cases of CEOT of the mandible are of the intraosseous type. The incidence of extraosseous CEOT is reported to be about 6%.^(4,11)

Both intraosseous and extraosseous types present in a similar manner histologically, however there are differences radiographically. The intraosseous CEOT shows radiolucent areas with occasional calcification, while the extraosseous type shows bone erosion near the tumor.⁽¹²⁾

In our case the radiolucency around the tooth was within normal limits so radiographically it was diagnosed as a follicle. All the cases of CEOT which have been reported so far in literature have shown extensive radiolucency. Our case is the first case of CEOT showing an inconspicuous radiolucency around the impacted tooth and no such case has been reported till date.

The intraosseous tumor is thought to originate from the stratum inter medium of the enamel organ, while the extraosseous type is thought to be derived from the dental lamina, or the basal cells of the gingival

epithelium.⁽³⁾ The intraosseous CEOT is thought to be more aggressive than the extraosseous type. It reportedly has a recurrence rate of 14%.⁽¹²⁾

Histologically, CEOT characteristically has presence of nests and sheets of polygonal epithelial cells interspersed with amyloid like eosinophilic droplets along with presence of calcification.

The epithelial cells may have a clear to eosinophilic cytoplasm, with a vesicular nuclei. The epithelial cells may also be present in a cribriform or a pseudoglandular pattern. Necrosis is uncommon. Atypical mitosis is also not seen usually. However, moderate amount of pleomorphism may be present. The presence of eosinophilic material resembling amyloid is quite characteristic of CEOT. Although the exact origin of this amyloid is not known it is believed to be derived from filamentous degradation of keratin filaments secreted by tumor epithelial cells.⁽³⁾

After reviewing the literature, we found that our case revealed striking histological resemblance to the five cases reported as non-calcifying variant of CEOT.⁽⁴⁻⁸⁾

These reported cases and our case all revealed odontogenic epithelial cells arranged in thin strands or small nests and globules of amyloid materials. Noticeably, no calcifications were presented in contrast to the stereotypic CEOT.

According to Kroll & Pindborg, absence of calcification denotes less tumor differentiation, which could mean that there will be more chances of recurrences.⁽¹³⁾ It is interesting that although there are few case reports of intraosseous CEOTs with minimal or no calcification, it is the general view that lack of calcification is more common in the peripheral or extraosseous variant.

The present case of CEOT was situated in the mandible that is intraosseously. Interspersed with the epithelial cells there was presence of amyloid like eosinophilic deposits. However, Langerhans cells were not seen.

Learning points/take home messages

- An unusual histological presentation of CEOT was reported.
- Difficulties were faced in diagnosing the tumor due to the absence of calcifications.
- Knowledge about this unusual presentation of CEOT can alert the pathologists to guide the surgeons for an extended follow up.

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