Clear Cell Variant Of Pindborg's Tumor In Anterior Mandible: Case Study And Overview Of Other Histological Variants.

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Citation

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Abstract

Pindborg tumour or calcifying epithelial odontogenic tumor (CEOT) is a rare and benign odontogenic neoplasm that was first described by a Dutch pathologist Jens Jorgen Pindborg in 1955. The most common clinical manifestation of CEOT is that of a localized, slow growing hard tissue swelling of the posterior jaws. Radiographically, it presents as a mixed radiolucent-radiopaque lesion. Though the histopathological presentation is quite distinct, cases have been reported to show slight variations from the usual picture. One such rare variant is the presence of abundant clear cells intermixed with the tumor cells. The significance of this variant is that it has been shown to have a more aggressive behaviour with greater tendency to recur if inadequately removed. We report a case of clear cell variant of CEOT with an uncommon site of presentation in the anterior mandible.

INTRODUCTION

Calcifying epithelial odontogenic tumor (CEOT) is a benign, locally invasive, slow growing neoplasm that has been identified under different denominations, such as ameloblastoma of unusual type with calcification, calcifying ameloblastoma, malignant odontoma and cystic complex odontoma, and has also been considered as a variant of simple ameloblastoma.[1] The eponym Pindborg tumor was first introduced to the scientific literature in deference to the renowned oral pathologist who first reported the case in 1955.[2]

The peak age of incidence is between fourth and sixth decades with the preferred site of occurrence being the posterior mandible (69%).[3] Microscopically, CEOT's are composed of sheets of strands of odontogenic epithelium, amorphous amyloid-like material and calcifications (Liesegang rings).[4]

The presence of clear cells in CEOT has been described and was identified as a distinct variant of CEOT. Only 18 cases of clear cell variant of CEOT (CCEOT) have been reported in the dental literature between 1958-2001.[5] CCEOT's are locally aggressive neoplasms requiring surgical resection with tumor free margins. Partial excision or enucleation results in recurrence with a reported case showing progression to a malignant form (odontogenic carcinoma).[6]

We report a case of CCEOT in the anterior mandible of a 43 year old male patient for its unusual site of presentation and a rare histological variant.

CASE REPORT

A 43 year old man reported to the out patient department of Indira Gandhi Institute of Dental Sciences with the chief complaint of swelling in the lower jaw since two years. Clinical examination revealed solitary, mildly tender, lobulated swelling of size 9 X 6 cms. in the anterior mandible (Figure. 1). Radiographic evaluation (CT and OPG) showed an ill defined mixed lesion with both intraosseous and extraosseous component extending from lower right molar region to canine region on the left side (Figure. 2). Numerous radio dense areas suggestive of calcifications were present with erosion of outer cortex of bone (Figure. 3).

Incisional biopsy was done under local anesthesia. Histopathological examination showed several islands and strands of normal appearing squamous epithelial cells in the connective tissue stroma with few inflammatory cells (Figure. 4). Some of these islands were surrounded by a homogenous eosinophilic material within which specks of calcifications were present (Figure. 5). Interestingly, several

areas of the tumour showed sheets of pale appearing squamous cells with clear foamy cytoplasm (Figure. 6). In some fields, the clear cells were the only predominant cells that were present.

Based on the clinical, radiological and histopathological observations, the lesion was diagnosed as calcifying epithelial odontogenic tumor – clear cell variant

The tumor was excised and the patient was recurrence-free after one year follow up.

Figure 1

Fig. 1

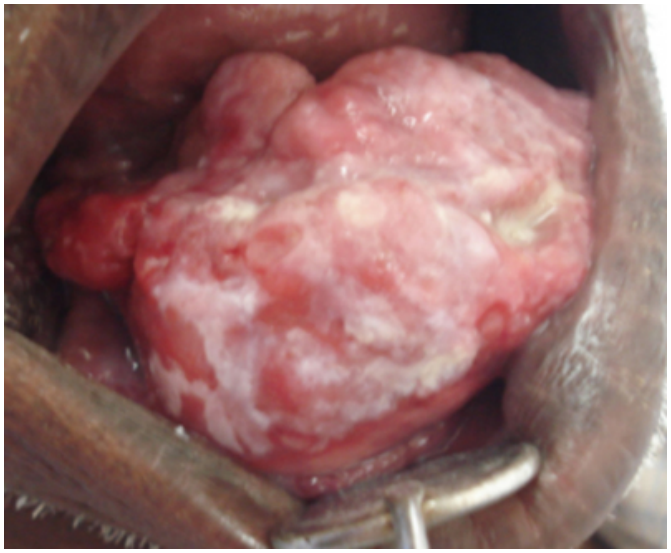


Figure 2

Fig. 2

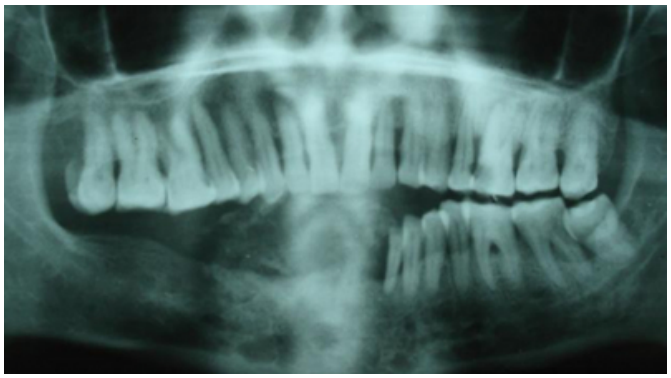


Figure 3

Fig. 3

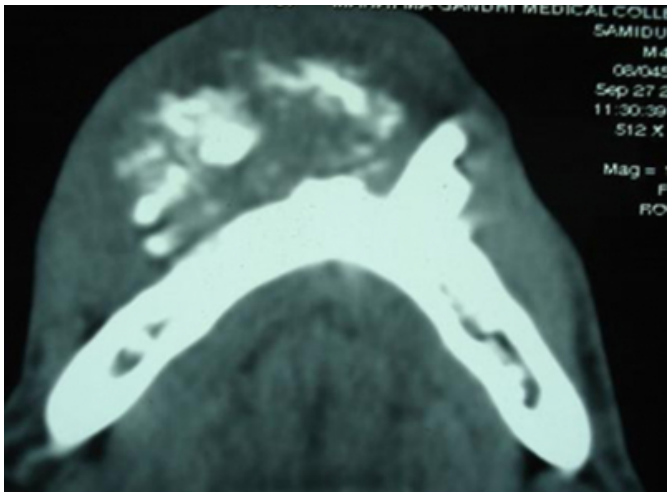


Figure 4

Fig.4

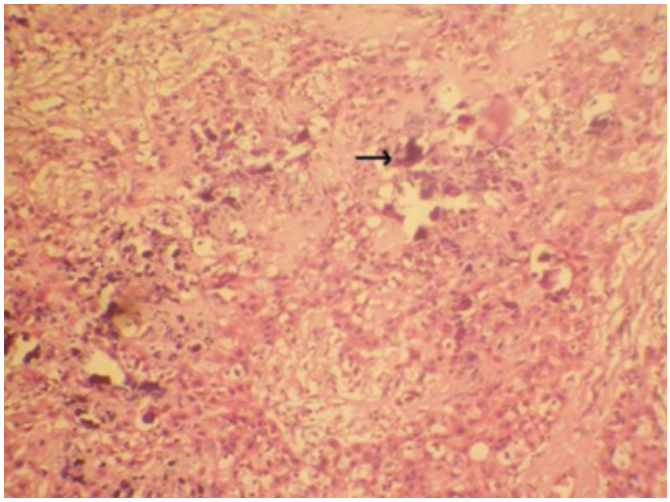


Figure 5

Fig. 5

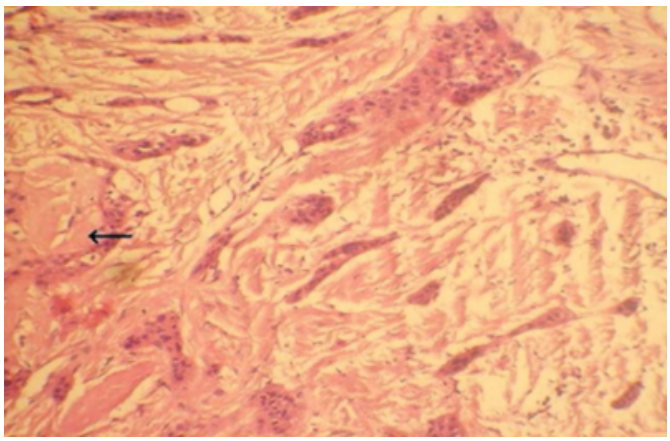
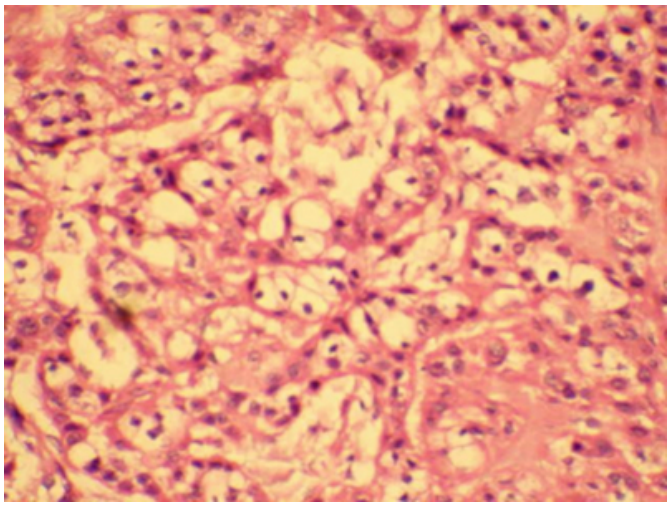


Figure 6

Fig. 6



DISCUSSION

Pindborg tumor is an uncommon benign odontogenic tumor with a variable biological behaviour that according to Neville et al. account for less than 1% of all odontogenic neoplasms.[7]

Odontogenic epithelium is evidently the origin of this tumor, but the particular cell remains unknown. The original hypothesis of Pindborg who first described this tumor, as arising from the stratum intermedium of the enamel organ remains the most likely one.[8]

Other possible considerations for the cell of origin includes remnants of the dental lamina and basal stratum of the gingival epithelium.[9]

Microscopic section of the tumor will show nests of closely packed polyhedral cell that frequently demonstrate nuclear pleomorphism. Mitoses are rare. An extracellular eosinophilic homogenous material staining like amyloid is characteristic of this tumor. Concentric calcified deposits called ''Liesegang ring'' are formed in the amyloid material.

The nature of the amyloid-like material is unknown, but the material appears to be derived from epithelial cells.[10]

Histological variants of CEOT includes presence of clear cells, cementum-like components in CEOT, Langerhans cells in CEOT, presence of myoepithelial cells in CEOT , combined epithelial odontogenic tumor (CEOT associated with adenomatoid tumor –like areas) [11] and non-calcifying type of CEOT.[12]

Our case demonstrated a distinct clear cell component. The clear cells in CEOT represent a feature of cytodifferentiation rather than a degenerative phenomenon.[13]

Presence of cytoplasmic glycogen granules makes the odontogenic epithelial cells appear clear. These cells are positive for periodic acid Schiff's.

Positive immunoreactivity was demonstrated in these clear cells for cytokeratins 8, 13 and 19, filaggrin and anti ameloblastoma antibodies suggesting an odontogenic epithelial origin.[14]

OVERVIEW OF OTHER VARIANTS

LANGERHANS CELLS IN CEOT

Langerhans cells have been reported in association with CEOT. Compared to typical CEOT, the tumor islands in these cases were thin and composed of small number of polyhedral epithelial cells. Almost no calcification of homogenous eosinophilic material was observed. [15]

Few of these cells were + ve for S-100 protein and electron microscope examination revealed the presence of Birbeck granules in the cytoplasm, highly suggestive of Langerhans cells. [16]

MYOEPITHELIAL CELLS IN CEOT

El Labban [17] in his reported case has identified that tumor islands consist of two different cell populations. Along with the classic polyhedral epithelial cells, few cells were arranged peripherally with elongated profiles and juxtaposed to the tumor epithelial cells.

These cells exhibited large number of cytoplasmic filaments with occasional electron dense areas similar to smooth muscle type cell. No desmosomes between these cells and tumor epithelial cells were present. Ultrastructurally these cells were interpreted as myoepithelial cells.

COMBINED EPITHELIAL ODONTOGENIC TUMOR

First described in 1983, two cases of adenomatoid odontogenic tumor (AOT) showed CEOT-like areas within the tumour.[18] So far, 24 such cases have been reported. In all the cases reported, the histological picture showed predominantly AOT-like areas. The biologic behaviour of these combine tumor is identical to that of AOT.[19]

CONCLUSION

Clear cells in CEOT are an uncommon presentation that requires the pathologist to differentiate these tumors from other clear cell tumor of the jaws. These include clear cell salivary gland tumor, clear cell variant of squamous cell carcinoma, primary intraosseous mucoepidermoid carcinoma, metastatic clear cell adenocarcinoma and epithelial-myoepithelial carcinoma.

Tumors containing clear cells generally show a locally aggressive behaviour. Cortical perforation is common (67%) compared to CEOT without a clear cell component (6.7%). Recurrence is also more common for clear cell containing CEOT's (17%). Hence surgical resection of these neoplasms requires wider tumor-free margins and long term follow up.

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