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CLEAR CELL VARIANT OF CALCIFYING EPITHELIAL ODONTOGENIC TUMOR OF MANDIBULAR ANTERIOR REGION: REPORT OF A RARE CASE

Dr. Biju Babu	Junior Resident Department Of Oral Pathology And Microbiology, Government Dental College, Trivandrum
Dr. Padmakumar S K*	Professor And Head, Department Of Oral Pathology And Microbiology. Government Dental College Thiruvananthapuram*Corresponding Author
Dr.Manju M Stephen	Assistant Professor, Department Of Oral Pathology And Microbiology. Government Dental College, Thiruvananthapuram

ABSTRACT Calcifying epithelial odontogenic tumor (CEOT) is a uncommon benign odontogenic neoplasm of the jaws, accounting for less than 1% of all odontogenic tumors. CEOT was included in the 1971 WHO classification of odontogenic tumors and a since then number of variants have been described, which have added confusion to the diagnostic criteria. CEOT is also known as Pindborg tumor. The classic pattern of CEOT comprises sheets of polyhedral epithelial cells with well-defined cell borders and distinct intercellular bridges with pleomorphic nuclei, which might show clear appearance due to intra cytoplasmic accumulation of glycogen but only rarely shows typical mitosis. Clear cell variant of calcifying epithelial odontogenic tumor of mandibular anterior region is one of the rarest presentations of this tumor and one such case associated with left mandibular canine of 44 year old female have been reported in this literature. CEOT are less aggressive in nature, so the conservative local resection to be the treatment of choice. The recurrence rate of these tumors are 15 %. The overall prognosis of these tumors is good. Clear cell variant of CEOT is the rarest form. In this literature we discuss one of such rarest variant of CEOT.

KEYWORDS : CEOT (calcifying epithelial odontogenic tumor), Odontogenic tumor, Pindborg tumor, Pleomorphic nuclei.

INTRODUCTION

Calcifying epithelial odontogenic tumor (CEOT) is a unusual benign odontogenic neoplasm of the jaws, accounting for less than 1% of all odontogenic tumors was first identified by Thoma and Goldman. It was not until 1955, however, that CEOT was recognized as a separate entity by Pindborg. So CEOT is also known as Pindborg tumor(Badrashetty et al., 2013). Commonly occur in the posterior mandible and found in patients between 30 and 50 years of age, with no sex predilection(Badrashetty et al., 2013). The origin of the neoplasm is controversial, though it is believed to be derived from the oral epithelium, stratum intermedium, or remnants of dental lamina(M. M. R. Bouckaert et al., 2000). In general, two varieties of CEOT were recognized – extraosseous and intraosseous with an incidence of 6% and 94%, respectively. Intraosseous tumors commonly involve mandible than maxilla with a ratio of 2:1 in the molar and premolar region(Sahni et al., 2012). Clinically, CEOT is usually presented as asymptomatic, slowly enlarging swelling associated with expansion of cortical plates(Chatterjee et al., 2017). On radiographic examination, CEOTs vary from small, unilocular radiolucent lesions to extensive multilocular, mixed radio-dense lesions often associated with an impacted tooth(Chrcanovic & Gomez, 2017). Presence of radio-opaque flecks is the characteristic feature of this lesion. 50% of the central lesions exhibit evidence of cortical bone perforation while 40% of peripheral CEOTs have subjacent bone erosion(Chaudhry et al., 1962). CEOT is microscopically characterized by cords and nests of round to polygonal eosinophilic cells with nuclear pleomorphism and conspicuous intercellular bridges in a fibrous stroma that typically contains variable amounts of the Congo red-positive amyloid-like material and calcified structures(Siriwardena et al., 2021). Tumor may show variations in the classic histological appearance such as noncalcifying CEOT with Langerhans cells, CEOT displaying cementum and bone-like material, and clear cell CEOT, of which clear cell CEOT shows more aggressiveness with a high recurrence rate. In this paper, a rare case of CCEOT of anterior mandible is reported and clinical, radiographic histological features of the same are discussed.

CASE REPORT

A 44-year-old female patient reported to the Department of Oral and Maxillofacial Pathology of Government dental college, Thiruvananthapuram, Kerala, India, with a chief complaint of swelling involving the lower front side of the face since one and half months. Initially swelling was very small in size which had gradually increased in size accompanied by localized intermittent pain and associated tooth mobility. The patient medical history and review of system were unremarkable.

On intraoral examination revealed the presence of a diffuse, moderately tender, noncompressible, firm to hard swelling involving buccal vestibular region in relation to mandibular left lower canine and premolar region which obliterating the buccal vestibule. Surface over the swelling appear normal. On palpation the swelling is bony hard and mildly tender which is obliterating the buccal vestibule.

Orthopantomogram [Figure 1] view revealed the presence of well-defined multilocular radiolucency at the anterior body of mandible. CT mandible (plane & contrast) view revealed a well-defined expansile heterogeneous soft tissue density lesion noted involving anterior half of body of mandible on left side.

Routine hematological investigations were within the normal limits. Under local anesthesia, excision biopsy was done.

The Histopathological examination of excised specimen shows sheets of polyhedral, neoplastic, odontogenic epithelial cells with prominent cellular outlines and intercellular bridges along with the presence of homogeneous, eosinophilic, amorphous materials. Calcified areas were also noted within the sheets of epithelial cells [Figure 2]. One of the most interesting features was the presence of clear cells having vacuolated cytoplasm within the odontogenic epithelial islands which stains positively with periodic acid–Schiff (PAS) [Figure 3].

Our provisional diagnosis was clear cell variant of odontogenic tumor, and to confirm this diagnosis, we had gone for immunohistochemical (IHC) evaluation. IHC markers such as cytokeratin 8 confirmed the presence of odontogenic epithelium within the neoplasm[Figure 4]. After considering the microscopic features, the diagnosis of clear cell variant of CEOT was made, and the patient was referred for surgical treatment and management.

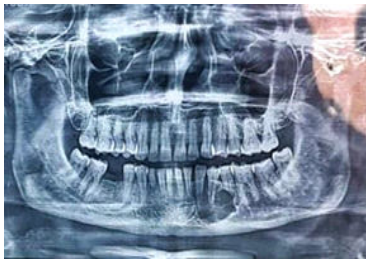


Figure 1: Panoramic radiographs showing a multilocular radiolucent area at the canine – premolar region.

Figure2: Microphotograph shows polyhedral, epithelial cells with homogeneous, polyhedral epithelial cells with clear polyhedral epithelial cells with clear

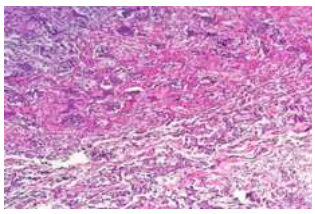


Figure 3. Microphotograph shows– Nests of polyhedral epithelial cells with clear Cytoplasm.

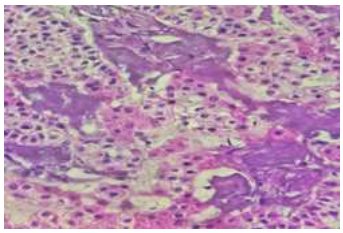
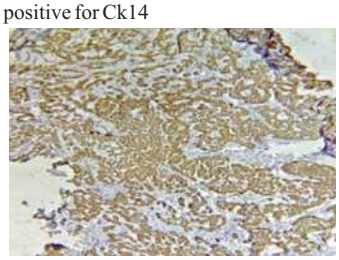


Figure 4. Immunohistochemical finding shows Sheets of polyhedral epithelial cells positive for Ck14



DISCUSSION

Clear cell variant of CEOT is rare, benign, but locally aggressive neoplasm of epithelial origin that accounts for <1% of all odontogenic tumors(Chatterjee et al., 2017).Histogenesis of this neoplasm is uncertain, mostly believed to arise from stratum intermedium of dental lamina because of the morphological resemblance of tumor cells to that stratum intermedium and a high activity of alkaline phosphatase and adenosine triphosphate in both these cells(M. M. Bouckaert et al., 2000).The classic pattern of CEOT comprises sheets of polyhedral epithelial cells with well-defined cell borders and distinct intercellular bridges with pleomorphic nuclei, which might show clear appearance due to intra cytoplasmic accumulation of glycogen but only rarely shows typical mitoses. Additionally, the other most important characteristic findings are the presence of amyloid-like substances and calcified concentric Liesegang rings. Amyloid-like substances showed positive, apple- green birefringence under polarized light. five histopathologic patterns of CEOT have been documented: (i) strands/ sheets/islands of polyhedral cells with intracellular bridges; (ii) a cribriform arrangement with many spaces containing an eosinophilic (amyloid-like) substance; (iii)

densely populated neoplastic cells with interspersed multinucleated giant cells; (iv) nests of epithelial cells similar to neoplasm of the salivary gland; and (v) prominent clear-cell arranged in a pseudo glandular manner(Krolls & Pindborg, 1974). In these five histopathologic types, the last pattern is referred to as the clear-cell variant of CEOT, and the histopathological findings of the case discussed in this literature were consistent with this pattern, showing numerous clear cells associated with amyloid-like material. It should be noted that clear cells are also seen on other epithelial odontogenic lesions such as ameloblastoma and calcifying odontogenic cyst(Aguiar et al., 1996).The clear cells of the ameloblastoma was clearly of odontogenic epithelial origin and clear cells of calcifying odontogenic cyst are odontogenic epithelial cells, which have undergone aberrant degeneration. In Clear cell variant of CEOT, it has been claimed that the clear cells are arise as a degenerative process, whereas another suggestion indicated that the clear tumor cells represent a feature of cytodifferentiation rather than the degenerative phenomenon(Chen et al., 2013).Radiographically CEOT exhibits a unilocular or multilocular radiolucency's with radiopaque flakes of calcifications within the radiolucent area producing a typical "driven snow" type of appearance. In our case there is no radiopaque flakes are noticed and not associated with an impacted tooth. The nature of arrangements of tumor tissue and staining characteristics of cells in our case was strongly mimicking the features of clear cell variant of CEOT. Histopathologically, clear cell variant of CEOT contains amyloid-like material which showed apple-green birefringence after staining with Congo red under polarized light. These features help differentiate it from clear cell odontogenic carcinoma from other lesions. The method of treatment will depend on multiple factors such as size, location of the tumor, general condition of the patient, histopathological findings, and operator skills. Small intrabony lesions are treated by simple enucleation and curettage whereas large tumors require aggressive approach, either by hemi mandibulectomy or hemi maxillectomy. The recurrence rate was reported to be 10%–20%; hence, periodic follow-up is essential to prevent further recurrence.

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