Peripheral Calcifying Epithelial Odontogenic Tumor: A Case Report

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1 Introduction

The calcifying epithelial odontogenic tumor (CEOT) was first described by Pindborg (1955) as a distinct entity in 1955. The eponym "Pindborg Tumor" was first introduced into the literature in 1967 to further describe this interesting and unique odontogenic tumor. The CEOT typically presents clinically as an intraosseous, expansile, painless mass that exhibits slow growth. It is most often encountered between the ages of 30 and 50 years, with no significant difference in occurrence based on sex. This tumor is typically associated with an unerupted or impacted tooth, usually a mandibular third molar (Neville et al., 1995).

The CEOT is composed microscopically of polyhedral epithelial cells that exhibit a granular eosinophilic cytoplasm and are believed to originate from the stratum intermedium. Other characteristic microscopic features include the presence of an amorphous, homogeneous, eosinophilic, amyloid-like material and foci of calcification, sometimes in large amounts and in the form of lamellar, concentric structures (Liesegang's rings). Occasionally the lesional cells may exhibit a clear, vacuolated cytoplasm (clear cell variant) (Shafer et al., 1983). Although most of these tumors are primarily intraosseous, an extraxo- sseous tumor is also known to occur; first observed by Pindborg in 1966. Additional extraosseous lesions have been reported; however, only four of the nine reported cases have exhibited the unique microscopic features of a prominent clear cell component. The purpose of this article is to describe a case of extraosseous form of CEOT.

Keywords Banana; Water deficit; Total chlorophyll; Soluble protein and yield
tional cases of extraosseous CEOT.

2 Case reports
A 35 year old female patient reported to the outpatient department, with the chief complaint of pain in the lower left back region from past 3 months. The history of present illness dates back to past 3 months when the patient felt pain w.r.t distal surface of 37 on the alveolar crest region. On examination, the patient was moderately built and nourished with all the vital signs within normal limits. Extra-oral examination presented with no facial asymmetry. On intra-oral examination a small swelling was seen w.r.t the alveolar crest region of 37. It was oval in shape, 1 cm × 0.08 cm in size. The color was similar to the normal color of mucosa, with smooth texture and margins, soft consistency. No induration was present. The swelling was tender and submandibular lymph nodes were palpable. On radiographic examination, a bony spicule was seen w.r.t the distal aspect of 37, and no other changes were observed. Under LA, the soft tissue was excised and the provisional diagnosis of hyperplastic gingival tissue was given and the tissue was submitted to the department of oral pathology on histopathological examination, the submitted H/E section revealed a parakeratotic epithelium with finger like rete ridges. The underlying connective tissue has dense bundles of collagen. Deep down, the connective tissue is slightly loose and 3 epithelial islands are present associated with multiple areas of calcification. The epithelial cells show prominent desmosomal junctions and are hyperchromatic and pleomorphic (Figure 1; Figure 2).

3 Discussion
Just as there is an uncommon peripheral variant of ameloblastoma, so there is also a distinctly uncommon peripheral variant of Pindborg tumor—limited to soft tissue only—that presents clinically as a nodular mass on gingiva mucosa (Krolls and Pindborg, 1974; Wertheimer et al., 1977), frequently in an anterior tooth location. The extraosseous CEOT is exceedingly rare with only nine cases previously reported (Pindborg, 1966; Takeda et al., 1983). It is a rare benign epithelial odontogenic neoplasm; its prevalence ranges from 0.6% to 1.7% of all odontogenic tumors (Pindborg, 1955; Shafer et al., 1983; Pindborg, 1966). In one investigation that reviewed the literature and combined the data reported on 181 patients who had Pindborg tumors, approximately 1 in 20 tumors (5%) were extraosseous in location (Ai-Ru et al., 1982). The tumour is basically one of adulthood. The mean age of occurrence is around 40 years. Chaudhary and his colleagues (Pindborg, 1966) reported the average age of patients with intraosseous lesions was 45 years.
whereas that with extraosseous tumour was 38 years. In reported cases that mentioned race, the great majority of patients were Caucasians. There have only been few reported cases in blacks (Pindborg, 1966; Abrams and Howell, 1967; Decker and Laffitte, 1967). There is a predilection for occurrence in the premolar-molar region of the mandible. Approximately two-thirds were located there (Abrams and Howell, 1967; Patterson et al., 1969). The intraosseous variant of CEOT are more common accounting for 87.8% as compared extraosseous tumors 6.1% with the former presenting most often in the mandible (premolar-molar area) whilst the latter in the anterior part of the jaws (Ai-Ru et al., 1982).

The CEOT has also been reported as hybrid tumors (Wertheimer et al., 1977; Ai-Ru et al., 1982; Takeda et al., 1983) in combination with adenomatoid odontogenic tumour (AOT) which has been reported to be frequent in women and present at a younger age. Association with impacted teeth was difficult to ascertain, since this information was not uniformly recorded. It would appear that less than half of the reported tumors have developed in close proximity to an un-erupted tooth. The typical clinical presentation is a slowly enlarging intraosseous mass which causes expansion of the affected mandible and is asymptomatic. For extraosseous CEOT, bone adjacent to the tumour shows a superficial erosive pattern. The histomorphologic pattern of the CEOT consists of scanty connective tissue stroma that supports clusters of polyhedral epithelial cells with eosinophilic cytoplasm. The nuclei vary in size and staining quality, producing a pleomorphic appearance. Intercellular bridging is present. Small, usually round calcifications with Leisegang rings are present among the epithelial cells and also in the connective tissue, as were seen in the present case. Besides, these characteristic features the presence of homogeneous substance that has been variously described as amyloid (Hicks et al., 1994; Franklin and Pindborg, 1976), comparable glycoprotein (Franklin and Pindborg, 1976), or keratin or enamel matrix (Pindborg, 1966). A clear cell variant of CEOT has also been described (Abrams and Howell, 1967; Patterson et al., 1969; Thoma and Goldman, 1946). The clinical differences among the intraosseous and the extraosseous variants CEOT types have been thought to be attributed to their origin (Wertheimer et al., 1977; Ivy, 1948; Chaudhry et al., 1972). It has been shown that the intraosseous CEOT is derived from the stratum intermedium of the enamel organ. In contrast the extraosseous form arises from the dental lamina epithelial rests in gingiva and/or basal cells of the gingival surface epithelium. With the hybrid tumour between CEOT/AOT, the AOT portion arises from all three components of the enamel organ (preameloblasts, stellate reticulum, stratum intermedium (Wertheimer et al., 1977).

The peripheral variant of CEOT can displays a range of radiographic features with regard to lesion size and bone pattern as compared to the intraosseous forms. The intraosseous CEOT can show completely radiolucent character or, in the case of more mature lesions, exhibit a mixed radiolucent and radiopaque appearance. In one study of 67 Pindborg tumors (Thoma and Goldman, 1946), 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%). For extraosseous CEOT, bone adjacent to the tumour shows a superficial erosive pattern (Ivy, 1948). The peripheral CEOT can present with no symptoms and can be an incidental finding. As in the present case the provisional diagnosis was given as the hyperplastic tissue as there were no radiographic changes seen. The histopathological features showed the features similar to that of CEOT and based on its location a diagnosis of peripheral CEOT was given. As noted by Buchner and Sciubba, follow-up information for the extraosseous CEOT is limited and long-term follow-up is rare; therefore observation of these patients over a long period of time is necessary in order to further our understanding of the biologic behavior of this rare group of odontogenic tumors. No recurrence has been
reported in the 11 cases known to have been treated with simple local excision (Chaudhry et al., 1972; Abrams and Howell, 1967).

4 Conclusion
The case presented here showed a well-defined radiographic lesion and was completely enucleated. However, the histological appearance of the lesion was typical of a CEOI. A large amount of amyloid tissue and calcification was present in the lesion. A conservative approach was adopted. Although various approaches have been suggested by some authors we believe that the treatment should be individualized for each lesion because the radiographic and histologic features may differ from one lesion to another. The management of this patient, thus required close monitoring.

References
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