Gingival Calcifying Epithelial Tumor - A Rare Case Report

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ABSTRACT
Calcifying epithelial odontogenic tumors are rare odontogenic neoplasms that account for approximately one percent of all odontogenic tumors. Extra osseous variant is very rare with very few cases being reported in literature. Peripheral Calcifying epithelial odontogenic tumor (CEOT) commonly resemble oral hyperplastic or reactive lesions and are histologically similar to their intraosseous counterparts. Here we report a rare case of gingival calcifying epithelial odontogenic tumor in the mandibular posterior region in a 31 year old female patient. Interestingly on literature review in on PubMed search only 22 case reports were available in English literature, added to that this report will be the 23rd case.

KEY WORDS
Computed tomography, gingival calcifying epithelial tumor, odontogenic tumor, peripheral calcifying epithelial tumor, pindborg tumor

INTRODUCTION
Calcifying epithelial odontogenic tumor (CEOT) was first described by Pindborg in 1955, and is also referred to as Pindborg tumor.1 In 1966, Pindborg described a peripheral counterpart that was histologically similar to the intraosseous tumor, and it represents approximately six percent of the total cases of Calcifying epithelial odontogenic tumor.2,3 This lesion is an odontogenic benign tumor, occasionally invasive, which produces a homogeneous, eosinophilic, pale staining amyloid-like material which may become calcified.4 This tumor accounts for approximately one percent of all odontogenic tumors. Most Calcifying epithelial odontogenic tumors are intraosseous lesions of the mandible. It is believed that Calcifying epithelial odontogenic tumors arise from dental lamina remnants or the stratum intermediate of the enamel organ.5 Peripheral Calcifying epithelial odontogenic tumors generally occur as single, painless, non bleeding, gingival masses that commonly resemble oral hyperplastic or reactive lesions.5 The peripheral location suggests histogenesis from the dental lamina or basal cells of the oral epithelium.6

Studart Soares EC et al have reviewed literature since 44 years form 1966-2010 and stated, only 22 cases of peripheral Calcifying epithelial odontogenic tumor have been reported in English literature.7 Here we report a case of peripheral Calcifying epithelial odontogenic tumor which is very rare in occurrence, which appeared in the premolar molar area of the mandibular gingiva, according to out search in pubmed this case will be the 23rd case in literature till date.

CASE-REPORT
A 31 year old female patient attended the out patient Department of oral medicine and radiology with a complaint of a mass in right lower posterior teeth region
of her jaws since one year in her gingiva. Lesion started as a small painless nodule from the attached gingiva of right lower first molar region and gradually increased in size with no history of bleeding, paraesthesia and pain. The medical and dental history was not contributory. Intra-oral clinical examination revealed a well defined exophytic growth in relation to lower right molar region along the buccal side, measuring approximately 3x3 cm in diameter, extending mesiodistally from middle third of right lower second premolar to middle third of lower second molar and from marginal gingiva above and approximately 2 cm below the occlusal level of the involved teeth supero-inferiorly (Fig 1). The overlying mucosa was normal in color; none ulcerated and showed no vascular markings. On palpation the mass was firm in consistency, pedunculated, nontender and no bruit or pulse was felt. Considering the history and clinical examination, following differential diagnosis was considered, peripheral ossifying fibroma, peripheral odontogenic fibroma, Peripheral CEOT and pyogenic granuloma. Pulp vitality test was conducted, involved teeth...
responded within normal limits. Routine hematological investigation values were also found to be within normal limits. Later intraoral periapical radiograph (Fig-2) and Orthopantomogram (Fig-3) were taken which did not reveal any pathological findings. Further mandibular cross sectional occlusal radiograph (Fig-4) was taken which revealed a soft tissue shadow of lesion along the buccal aspect of the involved teeth and multiple radiopaque mass seen within the lesion which was appearing to be a calcified structure within soft tissue shadow. Computed tomography was done, in the axial section hard tissue window (Fig-5) and axial section soft tissue window (Fig-6) shows multiple hyperdense areas measuring 1-2 mm interspersed within the soft tissue shadow on the right buccal aspect with intact cortex. Paraxial view (Fig-7) showed intact buccal cortex with soft tissue shadow interspersed with small hyperdense foci. An excisional biopsy was performed under local anesthesia and subjected to histopathologic examination, (Fig-8 ) which revealed sheets of polyhedral epithelial cells exhibiting perichromatic nuclei and prominent intercellular bridges. Eosinophilic amyloid like material and calcifications are also seen. These are features are consistent with final diagnosis of gingival calcifying epithelial tumor.

**DISCUSSION**

Soft tissue CEOTs are extremely rare, accounts for six percent of all CEOTs. Only 16 peripheral cases have been previously reported in the English literature. Most of these occurred in the canine-premolar gingival regions with a single growth. Our present case the lesion was present in the right lower first molar region which is the characteristic site for intraosssous CEOT which was solitary. There are reports in literature stating multifocal distribution of PCEOT in the jaws. According to Sedghizadeh et, odontogenic lesions involving multiple sites may be associated with chromosomal or genetic abnormalities, as supported by reports of multifocal odontogenic lesions in patients with known genetic disorders.

Although it is an odontogenic tumor, its histogenesis is still uncertain. It has been proposed that CEOTs originate from the stratum intermedium of the dental organ or from dental lamina. CEOT can occur at any age; however, it is most often seen between 30 and 50 years of age, with no sex predilection. The vast majority of cases occur in the posterior mandible. Rare extraosseous examples have also been reported with the anterior gingiva being the most frequent location.

Clinically, gingival CEOT is a slow-growing solitary painless mass, slight female gender preference, with most cases occurring in middle aged women. Clinically, presented case appeared in a 31 years old female patient as a painless, firm, sessile, well-circumscribed nodule covered by smooth, regular, and erythematous mucosal surface. All the features in the present case was not unique to PCEOT, except for the location of the lesion where gingival CEOTs are mostly anterior region of the jaws but a differentiation was very much necessary.

Peripheral CEOT have resemblance to the reactive lesions such as (pyogenic granuloma, traumatic fibroma, peripheral giant cell granuloma, peripheral ossifying fibroma). However, the etiology of these conditions is usually known and careful examination is necessary when any local irritant is defined or the lesion recurs after surgical treatment. Diagnosis of peripheral ossifying fibroma was made for the present case based on the clinical and radiographic
appearance with the presence of radiopaque clusters. Possible clinical and radiologic differential diagnosis is considered in Table 1.

Peripheral CEOT is histologically similar to its intraosseous counterpart in that both contain islands of polyhedral eosinophilic epithelial cells, a connective tissue stroma, and variable amounts of amyloid and Liesegang rings. Most peripheral CEOTs appear to be nonaggressive lesions with low recurrence and excellent prognosis. Intraosseous CEOT recurrence rates vary from 10% to 15% and surgical excision with clear margins and close follow-up are mandatory.

CONCLUSION
Peripheral CEOTs are extremely rare, but oral physician should include in the differential diagnosis of gingival lesions. Present case demonstrates the importance not to ignore any peripheral gingival lesion without appropriate histopathological evaluation, which in turn helps in the correct initial management of gingival overgrowth. Hence, complete surgical excision generally leads to cure, the lack of complaints and recurrence of GCEOT. A recommendation for long-term monitoring especially required for such rare tumors because there are limited literature about long term prognosis.

REFERENCES