

# Gingival Calcifying Epithelial Tumor - A Rare Case Report

Tejasvi MLA,<sup>1</sup> Bangi BB,<sup>1</sup> Geetha P,<sup>1</sup> Pavani D,<sup>2</sup> Harsha B<sup>1</sup>

<sup>1</sup>Department of Oral Medicine and Radiology

Kamineni Institute Dental Sciences

Sreepuram Narket Pally,

Nalgonda Dist, T.S- 508254, India

<sup>2</sup>Shri. Yashwantrao Chavan Memorial Medical and Rural Development

Foundation's Dental College and Hospital

Ahmednagar-414003, India

## Corresponding Author

Avinash Tejasvi ML

Department of Oral Medicine and Radiology

Kamineni Institute Dental Sciences

Narketpally, India

E-mail: avinash\_tejaswi@yahoo.co.in

## Citation

Tejasvi MLA, Bangi BB, Geetha P, Pavani D, Harsha B. Gingival Calcifying Epithelial Tumor - A Rare Case Report. *Kathmandu Univ Med J* 2014;47(3):215-8.

## INTRODUCTION

Calcifying epithelial odontogenic tumor (CEOT) was first described by Pindborg in 1955, and is also referred to as Pindborg tumor.<sup>1</sup> 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.<sup>2,3</sup> This lesion is an odontogenic benign tumor, occasionally invasive, which produces a homogeneous, eosinophilic, pale staining amyloid-like material which may become calcified.<sup>4</sup> 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 intermedium of the enamel organ.<sup>5</sup> Peripheral Calcifying epithelial odontogenic tumors generally occur as single, painless, non bleeding, gingival masses that commonly resemble oral hyperplastic or reactive lesions.<sup>5</sup>

## 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 23<sup>rd</sup> case.

## KEY WORDS

*Computed tomography, gingival calcifying epithelial tumor, odontogenic tumor, peripheral calcifying epithelial tumor, pindborg tumor*

The peripheral location suggests histogenesis from the dental lamina or basal cells of the oral epithelium.<sup>6</sup>

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.<sup>7</sup> 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 our search in pubmed this case will be the 23<sup>rd</sup> 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



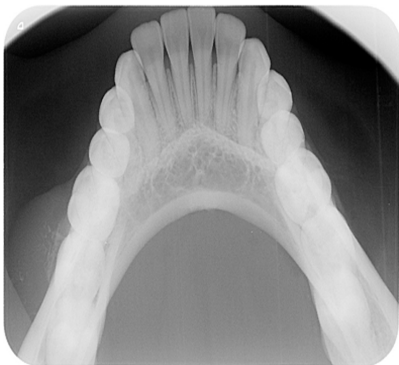
**Figure 1.** Well defined exophytic growth in relation to lower right molar region along the buccal side, measuring approximately 3x3 cm in diameter.



**Figure 2.** An IOPA radiograph showing no evidence of any pathological findings.



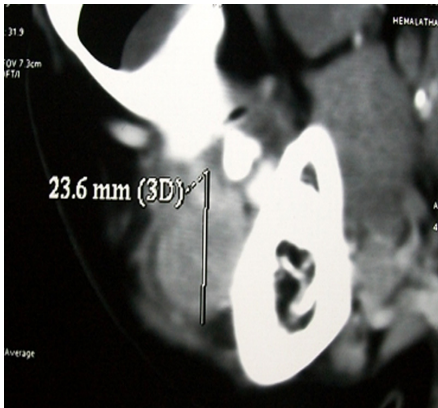
**Figure 3.** An OPG showing no evidence of any pathological findings in the radiograph.



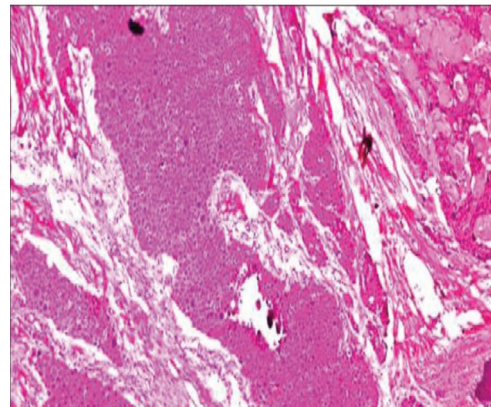
**Figure 4.** Occlusal radiograph showed a soft tissue shadow of lesion along the buccal aspect with multiple radiopaque mass seen within the lesion.



**Figure 5 and 6.** Computed tomography axial section (bone window) and Computed tomography axial section (soft tissue window) shows multiple hyperdense areas measuring 1-2 mm interspersed within the soft tissue shadow on the right buccal aspect with intact cortex.



**Figure 7.** Paraxial view showed intact buccal cortex with soft tissue shadow interspersed with small hyperdense foci.



**Figure 8.** H & E stain, 10X showing sheets of polyhedral epithelial cells, amyloid like material and calcifications.

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, parasthesia 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

**Table 1. Clinical and radiographic differential diagnosis for gingival CEOT.**

Lesion	Site	Age	Clinical Feature	Radiographic Feature	Prognosis
1. Peripheral Adenomatoid odontogenic tumor	Maxillary canine	9-16 years	- Painless gingival sessile swelling	Shallow erosion of the underlying bone. - Bony depression below the lesion. - Characteristic mineralization within the lesion which is radiopaque in nature	-Good
2. Peripheral ossifying fibroma	-Anterior to molar region	Any age	Well demarcated focal mass of tissue on gingiva with a sessile or pedunculated base with normal colored mucosa.	- No bone involvement. - Radiopaque ossification seen within the lesions.	-Good without any recurrence
3. Peripheral Odontogenic Fibroma	Maxilla and mandible		Slow growing Solid firmly attached gingival mass between the teeth. Lesion displaces the teeth.	Mineralization seen with the lesion. Radiopaque flecks are seen with the lesion.	Good without any recurrence
4. Peripheral Ameloblastoma	Maxillary and mandibular canine – premolar region.	5 <sup>th</sup> – 6 <sup>th</sup> decade.	Painless, Firm exophytic mass with normal color.	No evidence of bone involvement. Superficial saucerization of alveolar bone.	Good
5. Peripheral Ameloblastic fibroma	Mandibular posterior region is most common site.	1 <sup>st</sup> -2 <sup>nd</sup> decade Mean age - 14 years	Presents as a painless localized gingival enlargement or epulis. Associated with delayed eruption of the teeth.	No definable radiographic features present	Good

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 perchromatic nuclei and prominent intercellular bridges. Eosinophilic amyloid like material and calcifications are also seen. These are features consistent with final diagnosis of gingival calcifying epithelial tumor.

## DISCUSSION

Soft tissue CEOTs are extremely rare, accounts for six percent of all CEOTs.<sup>6</sup> 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 intraosseous CEOT which was solitary. There are

reports in literature stating multifocal distribution of PCEOT in the jaws. According to Sedghizadeh et al, 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.<sup>8</sup>

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.<sup>9</sup> 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.<sup>10</sup>

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 were 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.<sup>11</sup> 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.<sup>12</sup>

## 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

- Franklin CD, Pindborg JJ. The calcifying epithelial odontogenic tumor. A review and analysis of 113 cases. *Oral Surg Oral Med Oral Pathol.* 1976; 42:753-65.
- Da Silveira EJ, Gordon-Nunez MA, Seabra FR, Bitu Filho RS, Lima EG, de Medeiros AM, et al. Peripheral calcifying epithelial odontogenic tumor associated with generalized drug-induced gingival growth: a case report. *J Oral Maxillofac Surg.* 2007; 65(2):341-345.
- Mesquita RA, Lotufo MA, Sugaya NN, De Araujo NS, De Araujo VC. Peripheral clear cell variant of calcifying epithelial odontogenic tumor: report of a case and immunohistochemical investigation. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2003;95(2):198-204.
- Shafer WG, Hine MK, Levy BM. A textbook of Oral Pathology. 2<sup>nd</sup> ed. Philadelphia: W.B. Saunders; 1963. p. 217.
- Mesquita RA, Lotufo MA, Sugaya NN, De Araujo NS, De Araujo VC. Peripheral clear cell variant of calcifying epithelial odontogenic tumor: report of a case and immunohistochemical investigation. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2003;95(2):198-204.
- Takeda Y, Suzuki A, Sekiyama S. Peripheral calcifying epithelial odontogenic tumor. *Oral Surg Oral Med Oral Pathol.* 1983;56(1):71-5.
- Stuart Soares EC, Gurgel Costa FW, Silveira Esses DF, Alves Filho EP, Nunes Alves APN. Gingival calcifying epithelial odontogenic tumor: case report and review of the literature comprising a period of 44 years. *J Clin Exp Dent.* 2011;3(5):491-6.
- Sedghizadeh PP, Wong D, Shuler CF, Linz V, Kalmar JR, Allen CM. Multifocal calcifying epithelial odontogenic tumor. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007;104(2): 30-4.
- Takeda Y, Suzuki A, Sekiyama S. Peripheral calcifying epithelial odontogenic tumor. *Oral Surg Oral Med Oral Pathol.* 1983;56:71-5.
- Neville BW, Damm DD, Allen CA, Bouquot JE. Oral and maxillofacial pathology. Philadelphia: W. B. Saunders; 2002.p. 623-5
- Philipsen HP, Reichart PA. Calcifying epithelial odontogenic tumour: biological profile based on 181 cases from the literature. *Oral Oncol.* 2000;36(1):17-26.
- Anavi Y, Kaplan I, Citir M, Calderon S. Clear-cell variant of calcifying epithelial odontogenic tumor: clinical and radiographic characteristics. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2003; 95(3):332-9.