



REVIEW ARTICLE

Non-calcifying and Langerhans cell-rich variant of calcifying epithelial odontogenic tumor



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Abstract This study reported the clinicopathological features, treatment and prognosis of nine cases of noncalcifying and Langerhans cell (LC)-rich calcifying epithelial odontogenic tumor (CEOT) collected from the English literature. Of the nine cases, seven were intraosseous and two were extraosseous. All nine tumors were found in Asian patients. The age of the nine patients ranged from 20 years to 58 years with a mean age of 41 years. There were five female and four male patients. The seven intraosseous cases included six in the anterior and premolar region of the maxilla and one in the posterior region and ascending ramus of the mandible. The two extraosseous cases were located at the upper lateral incisor and premolar gingivae, respectively. Of the seven intraosseous cases, five showed unilocular and two multilocular radiolucency without foci of calcification. Six of the seven intraosseous cases showed resorption of the tooth roots in the tumor-involved region. Histologically, noncalcifying and LC-rich CEOTs were composed of small nests and thin strands of tumor epithelial cells with a relatively high number of LCs among them. This was the reason why we classed these nine cases as

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noncalcifying and LC-rich CEOTs. Two extraosseous cases received total excision of the gingival mass. For the seven intraosseous cases, four accepted partial maxillectomy or mandibulectomy, two received total excision or enucleation, and one underwent curettage. The six cases with the follow-up information available showed no tumor recurrence after a follow-up period of 6 months to 10 years.

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Introduction

Calcifying epithelial odontogenic tumor (CEOT) is a rare, benign, locally-invasive, and slow-growing odontogenic neoplasm which accounts for 1–2% of all odontogenic tumors.¹ It was firstly reported by Pindborg² in 1955 and thus it has also been known as Pindborg tumor for ~50 years. CEOT can be divided into either intraosseous (central, 94%) or extraosseous (peripheral, 6%) type.¹ The intraosseous type appears radiographically as a unilocular or multilocular radiolucent lesion containing calcified structures of varying size and density. Intraosseous CEOT occurs more frequently in the mandible (especially in the premolar/molar region of the mandible) than in the maxilla. Approximately 60% of intraosseous CEOT are associated with an unerupted tooth (or odontoma). The extraosseous type appears as a painless, firm, and sessile gingival mass and it may cause the depression or erosion of the underlying bone.¹

Histologically, the conventional CEOT is composed of sheets, islands, or strands of polyhedral and eosinophilic epithelial cells, large areas or globules of homogeneous and eosinophilic amyloid-like substance, and multiple concentric Liesegang ring calcifications in a fibrous stroma. The tumor epithelial cells may show cellular and nuclear pleomorphism and giant cell formation. However, no increased mitotic figures are found. Based on various histological features, the histological variants of CEOT include CEOT with cementum-like components, clear-cell CEOT, Langerhans cell (LC)-containing CEOT, CEOT combined with adenomatoid odontogenic tumor, and CEOT with myoepithelial cells.¹

The conventional CEOT has more or less foci of calcification. Another variant of CEOT that does not contain structures of calcification within the tumor is reported to be noncalcifying variant of CEOT with LCs.^{3–9} Although the tumor nests of conventional CEOT may occasionally contain LCs, the LC to tumor epithelial cell ratio is ~ 0.8–1.7:100. However, the tumor epithelial nests of noncalcifying variant of CEOT with LCs often contain abundant LCs with the LC to tumor epithelial cell ratio being 42–83:100.⁸ Therefore, we classed this specific type of noncalcifying variant of CEOT with LCs as noncalcifying and LC-rich variant of CEOT. In this study, nine cases of noncalcifying and LC-rich variant of CEOT were collected from the English literature.^{3–9} The clinical, radiographic, and histological features as well as treatment and prognosis of these nine cases of noncalcifying and LC-rich CEOT were analyzed and described in this study.

Materials and methods

Well-documented case reports of noncalcifying and LC-rich CEOT published between 1990 and 2015 were collected from English literature using Medline and from cross-references. The search was made using the keywords “calcifying epithelial odontogenic tumor”, “noncalcifying variant” and “Langerhans cell”. In total, nine accepted cases retrieved from seven articles were selected.^{3–9} The LC-containing conventional CEOT were excluded from the study samples. Data on age, gender, duration, location, symptoms and signs, radiographic features, resorption of tooth roots, histological findings, treatment modalities, and follow-up information were obtained from the original articles, analyzed, and reported.

Results

Clinical features

The demographic and clinical data of nine cases of noncalcifying and LC-rich variants of CEOT are shown in [Table 1](#). All nine noncalcifying and LC-rich CEOTs occurred in Asian patients. The ages of the nine patients at the time of diagnosis ranged from 20 years to 58 years with a mean of 41 ± 13 years. The seven patients with intraosseous noncalcifying and LC-rich CEOT had a higher mean age (45 ± 12 years) than that (30 ± 13 years) of the two patients with extraosseous noncalcifying and LC-rich CEOT. There were five female patients (including two with extraosseous type) and four male patients. The duration of the lesion (from the onset of the lesion to the time of diagnosis) was not stated in two cases. The duration of the resting seven tumors varied from 1 month to several years.

Of the nine cases of noncalcifying and LC-rich CEOT, seven were intraosseous and two were extraosseous. The seven intraosseous cases included six in the anterior and premolar region of the maxilla and one in the posterior region and ascending ramus of the mandible. The two extraosseous cases included one on the left upper premolar gingiva and the other on the labial gingiva of the right upper lateral incisor ([Table 1](#)). Thus, the anterior and premolar area of the maxilla was the most common location (8/9, 88.9%) for the noncalcifying and LC-rich CEOTs. For the symptoms and signs of the tumor, the two extraosseous cases had no symptoms and signs except a gingival swelling. Of the seven intraosseous cases, two had no symptoms, two had both pain and loose teeth, two had loose teeth only,

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