Review Article

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Calcifying Epithelial Odontogenic Tumor of Jaws: An Overview

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Abstract

The Calcifying Epithelial Odontogenic Tumor (CEOT) was first described by Pindborg as a distinct entity in 1955. Odontogenic tumors are derived from epithelial, ectomesenchymal and mesenchymal elements that have been a part of the tooth-forming apparatus. Of all the odontogenic tumors, calcifying epithelial odontogenic tumor accounts for 1% of the cases. There is no sex predilection with a 2:1 predilection for the mandible. CEOT is mostly found in the premolar or molar region. Clinically calcifying epithelial odontogenic tumor typically presents as a slow growing intra-osseous, expansile, painless mass. It is often locally invasive. Most often it is associated with an impacted tooth and remains asymptomatic. Although most of CEOTs are primarily intra-osseous however an extra-osseous counterpart of CEOT is also known to occur and first reported by Pindborg in 1966. This article presents a review of this unique and interesting tumor of jaws.

Keywords: Calcifying Epithelial Odontogenic Tumor; Intra-osseous; Extra-osseous; Maxilla; Odontogenic Tumors; Supernumerary Tooth.

Introduction

Odontogenic tumors comprise a diverse group of exceptional lesions derived from epithelial elements of the tooth forming apparatus that account for about 1% of all jaw tumours [1]**.** According to Mosqueda and Taylor, some of these are hamartromas that present variable degree of differentiation whereas the remaining ones are benign or malignant neoplasms of variable aggressiveness characterized by a metastatic potential [2]. The calcifying epithelial odontogenic tumor (CEOT) was first described by Pindborg as a distinct entity in 1955 [3].The eponym Pindborg Tumor was first introduced into the literature in 1967 to further describe this interesting and unique tumor [4]. Calcifying epithelial odontogenic tumor is an asymptomatic, benign, slow-expanding and a locally invasive tumor of jaws that account for approximately 1% of all odontogenic tumours [5,6]. Different terminologies have been used to describe this tumor such as ameloblastoma of unusual type with calcification, calcifying ameloblastoma, malignant odontoma and cystic complex odontoma [7].

It is most often encountered between the ages of 8 and 92 years with the peak occurrence in 40 years of age without significant difference in occurrence based on sex [8]. It may be classified as intra-osseous or extra-osseous. The extra-osseous variant has a predilection for anterior gingiva where it appears as a sessile mass capable of destroying the underlying bone. Intra-osseous type is more commonly found in the mandibular posterior region. More than half of these are associated with an impacted tooth [9]. On radiographic evaluation this lesion usually presents as unilocular or multilocular radiolucent area. In some cases this neoplasm may exhibit calcified structures of variable density and size [10]. 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 includes the presence of an amorphous, homogeneous, eosinophilic, amyloid like material and foci of calcification and in the form of lamellar, concentric structures (Liesegang's rings). Occasionally the lesional cells may exhibit a clear, vacuolated cytoplasm (clear cell variant) [11]. The differential diagnosis includes Adenomatoid odontogenic tumor, calcifying odontogenic cyst,

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dentigerous cyst, Ameloblastic fibro-odontoma and odontoma [7,12].

Discussion

Calcifying epithelial odontogenic tumor was first described by Jens J. Pindborg in 1955 as a separate entity among epithelial tumors and the eponym Pindborg tumor has also been used for this pathologic condition [3,13]. Most cases (94%) are intra-osseous and only 6% are extraosseous[7].The first report of extra-osseous CEOT is dated back to 1966 when Pindborg published two cases of gingival growth in the anterior jaw region of young patients [14]. Some other studies says the prevalence of extraosseous CEOT ranges from 0.6% to 1.7% of all odontogenic tumors [3,11,14]. Al-Ru et al. calculated 1 in 20 cases of CEOT were extraosseous in location in their case study of 181 patients [15]. In jaws there is predilection for occurrence anatomically. The intraosseous variant of CEOT has ratio of 1:2 (maxilla : mandible). The intraosseous variant is more common accounting for 87.8% of all CEOTs as compared to extra-osseous variants accounting for 6.1% of all CEOT. The intra-osseous variant is most often reported in the premolar-molar area while the extra-osseous variants is more commonly found in the anterior part of the jaws [7,13,15,16]**.** Kaplan et al. reported 41 cases of one or more impacted teeth (60%) associated with a total of 67 cases of calcifying epithelial odontogenic tumor. Out of these, the most prevalent site involved by CEOTs were the molars (62%) followed by premolars, canines, incisors and the least were the supernumerary or unidentified teeth (4%) [19].The initial consensus regarding the pathogenesis of CEOT was attributed to Pindborg in 1955. He stated that the CEOT was indeed of Odontogenic origin i.e. reduced enamel organ-related as the previous cases have been associated to unerupted teeth [14]. However according to Philipsen et al with the reports of central cases not presenting with unerupted tooth and the gingival variants other sources of origin were debated [7]. The soft tissue location of this tumor strongly suggests that these tumors may arise from rests of dental lamina or from basal cells of the oral epithelium. After disintegration of the dental lamina complex numerous epithelial remnants (rests of serres) persist in the jaw bones and supraperiosteally in the gingiva when odontogenesis is completed. Furthermore the focal proliferation of the basal layer of the gingiva epithelium has also been proposed as a possible origin [7,17]. Wertheimer et al stated that the intra-osseous CEOT is derived from the stratum intermedium of the enamel organ. In contrast the extra-osseous CEOT arises from the dental lamina, epithelial rests in gingiva and / or basal cells of the gingival surface epithelium [8,18].

Radiographically, the intraosseous lesion presents as radiolucency. Later as the lesion ages, calcium salts are deposited and it becomes increasingly radio-opaque. It also simultaneously erodes bone and thus the lesion is often mixed radiolucent and radio-opaque giving a characteristic 'driven snow' appearance on the radiograph. Further the lesion may be unilocular or more commonly multilocular in appearance. Thoma reported 65% lesions out of his 67 cases of Pindborg tumor were radiographically mixed (radiolucent and radio-opaque) type followed by 32% complete radiolucent and 3% radio-opaque [21]. The peripheral variant of CEOT can display a range of radiographic features with regards to lesion size and bone pattern as compared to the intraosseous forms. It presents with no radiographic changes to a superficial erosive pattern [14,20] to a radiolucency with scattered radiopaque foci [7].

Histologically calcifying epithelial odontogenic tumor exhibit considerable variations. It is characterized by a fibrous tissue stroma with sheets or islands of polyhedral epithelial cells with intercellular bridges. Nuclei are often pleomorphic and giant nuclei may be visible. Eosinophilic amorphous hyaline-rich material which stains positive for amyloid, may be present. Calcifications in the form of concentric rings known as Liesegang rings may develop within the amyloid-like material. This material stains with Congo red and exhibits an apple-green birefringence under polarized light. It also fluoresces under ultraviolet light with thioflavin T. This amyloid-like material may contain either basement membrane components (type IV collagen) [22] or a mixture of cytokeratins [23]. The origin of amyloid is unclear. It could either be an active secretion product or a degeneration product of keratin filaments which originate from tumor epithelium due to developmental or aging processes [23].

Some of these tumors may be epithelium-predominant with minimal amyloid whereas others may be amyloid-predominant with small islands of epithelium. Still others may have abundant clear cells [24].A mixed lesion along with Adenomatoid odontogenic tumor has also been reported [25].

Al-Ru et al [15] subclassified this tumor into four distinct microscopic patterns, two or more types may be present in same tumor. Type -1 is characterized by sheets, nests and masses of polyhedral epithelial cells exhibiting prominent intercellular bridges, marked

nuclear size variation, regular nuclear pleomorphism, scarce mitotic figures and calcified corpuscles in the fibrous stroma however Type-2 is characterized by a cribriform arrangement of tumor cells, less nuclear cell pleomorphism, absence of prominent intercellular bridges and masses of calcified tissue showing Liesegang rings. Type-3 consists of scattered or densely populated tumor cells accompanied by marked cellular pleomorphism in a myxoid stroma and frequent multinucleated giant cells. Type-4 is characterised by small nests and cords of epithelial cells, some of them containing abundant cytoplasm separated by fibrous stromal tissue. In addition several cellular variants such as clear cells, pigmented, langerhans cells, myoepithelial cells, and noncalcifying subtypes have been reported [7,26].

Calcifying epithelial odontogenic tumor is less aggressive than ameloblastoma although cases of malignant transformation have been reported [27]. The aggressiveness is a prominent finding in posterior maxilla. In addition root resorption is reported as a rare finding in calcifying epithelial odontogenic tumor (4%) unlike solid ameloblastoma (81%) [19]. Kaplan et al reported 28 cases out of a total of 67 cases (41%) of calcifying epithelial odontogenic tumor which had caused displacement of teeth [19].

Treatment of calcifying epithelial odontogenic tumor involves enucleation of smaller lesions and resection of large tumors [9]. The resection should include a rim of the surrounding bone. A long follow-up is recommended as a recurrence rate of 14% has been reported, particularly for those which have been curetted [28]. However according to literature a minimal follow-up of five years is highly recommended [5,8].

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