CASE REPORT

A rare case of hybrid odontogenic tumor: Calcifying epithelial odontogenic tumor combined with ameloblastoma

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ABSTRACT

A hybrid odontogenic tumor comprising two distinct lesions is extremely rare. Nevertheless, such tumors have been reported in the literature for academic and research interest. However, it is still obscure whether they behave as a new entity or they solely present separate histopathologic patterns. Here, we present a true hybrid neoplasm of combined ameloblastoma and calcifying epithelial odontogenic tumor showing intermixed histopathologic patterns of both the tumors.

Key words: Ameloblastoma, calcifying epithelial odontogenic tumor, hybrid odontogenic tumors

INTRODUCTION

The process of odontogenesis is prolonged and complex and the tumors derived from epithelial, ectomesenchymal and/or mesenchymal elements that are, or have been, part of the odontogenic apparatus are called as odontogenic tumors.[1] Despite the World Health Organization (WHO) classification of odontogenic tumors, unique odontogenic lesions with combined histologic features have been encountered occasionally.[2] Hybrid tumors are very rare tumor entities, which are composed of two different tumor entities, each of which confirms with an exactly defined tumor category whereas a hybrid odontogenic tumor is defined as follows: "A lesion showing the combined histopathological characteristics of two or more previously recognized tumors and/or cysts of different categories."[3] Ameloblastoma is a benign odontogenic tumor arising from the odontogenic apparatus showing odontogenic epithelium with mature fibrous stroma, without ectomesenchyme. It is the best known and the most frequently seen odontogenic tumor. Calcifying epithelial odontogenic tumor (CEOT) is also benign neoplasm reported by Pindborg in 1955,[4] with a frequency varying between 0.4% and 3%. It is a relatively rare occurring tumor with one of the lowest frequency rankings amongst the list of odontogenic tumors.[1] A hybrid odontogenic tumor comprising two distinct lesions is extremely rare. Earlier, hybrid tumors of CEOT in association with adenomatoid odontogenic tumors (AOTs) have been reported.[1,5-7] But, nothing indicates a true combination of two distinct and separate odontogenic tumor entities. Also, there are very few reported cases of ameloblastoma in which CEOT-like areas predominate. Only two such cases have been reported till date.[8,9] Here, we describe an extremely rare and interesting case of an intraosseous ameloblastoma associated with CEOT that appeared to be a histologic hybrid tumor.

CASE REPORT

A 65-year-old edentulous female patient reported to the Department of Orthopedics with a complaint of fractured leg. The Oral and Maxillofacial Surgery Department was informed with an incidental finding of huge swelling of the right maxilla causing asymmetry of the face [Figure 1].

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The massive swelling was painless and asymptomatic. The swelling was bony hard, noncompressible and nonfluctuant. The mass extended superio-inferiorly from the infraorbital region till the alveolus of the maxilla, laterally till the malar prominence, posteriorly till the anterior border of the masseter muscle and anteriorly extended till the angle of the mouth obliterating the nasolabial fold. Buccal and palatal cortical plate expansion was observed with adequate mouth opening. Intraorally, an irregular, nodular soft tissue mass was present in the vestibule of the posterior region and anteriorly over maxillary central incisor region [Figure 2]. Patient was a hookah smoker for last 4 years. Paranasal sinus view radiograph showed a large radiopaque mass in the right maxillary region [Figure 3]. Incisional biopsy of the lesion was sent to the Department of Oral and Maxillofacial Pathology which revealed intermingled areas of the plexiform type of ameloblastoma and CEOT [Figure 4]. Interlacing cords and strands of odontogenic epithelium were observed within a mature fibrous connective tissue stroma. Scanty stellate reticulum-like areas were appreciated in between the interlacing cords, suggesting the plexiform type of ameloblastoma [Figures 5 and 6]. However, many areas showed sheets and nests of polyhedral epithelial, eosinophilic cells which showed prominent nuclei and also cellular and nuclear pleomorphism in some areas [Figure 7]. Some cells were binucleated and prominent intercellular bridges were observed in focal areas. Among the sheets and cords of cells, numerous homogeneous eosinophilic hyaline masses were evident in most part of the section, suggestive of amyloid-like material [Figure 8]. Calcifications were not observed in the received specimen. The case was signed out as combined CEOT with ameloblastoma. The patient was not willing to undergo treatment for the lesion and hence was discharged from the hospital against medical advice.

DISCUSSION

According to the 1992 WHO classification, a CEOT is a locally invasive epithelial neoplasm characterized by the development of intraepithelial structures, probably of an

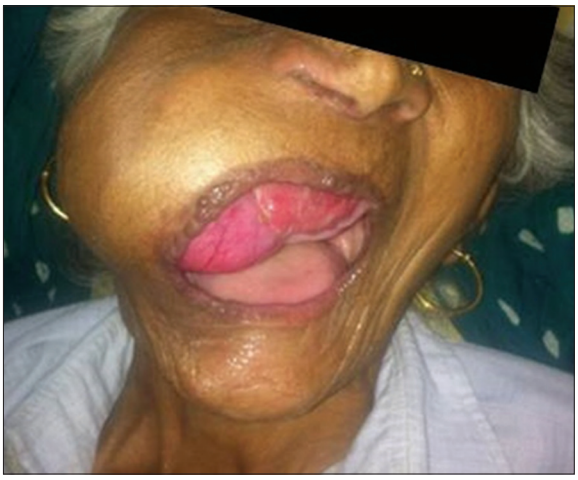

Figure 1: Extraoral view of the patient showing diffuse swelling of the right maxillary region causing facial asymmetry

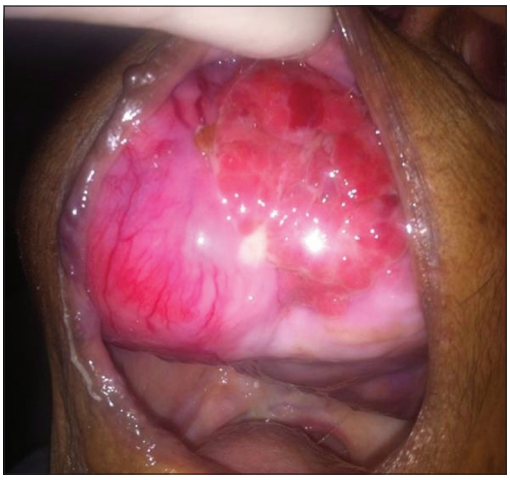

Figure 2: Intraoral view showing irregular and nodular swelling crossing the midline and obliterating the buccal and labial vestibule

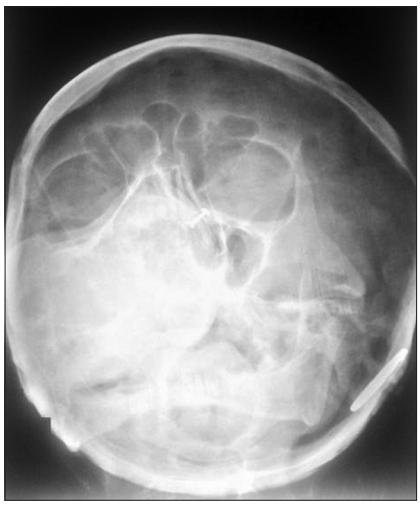

Figure 3: Posterior nasal spinal radiographic view showing large radioopaque mass in the right maxilla

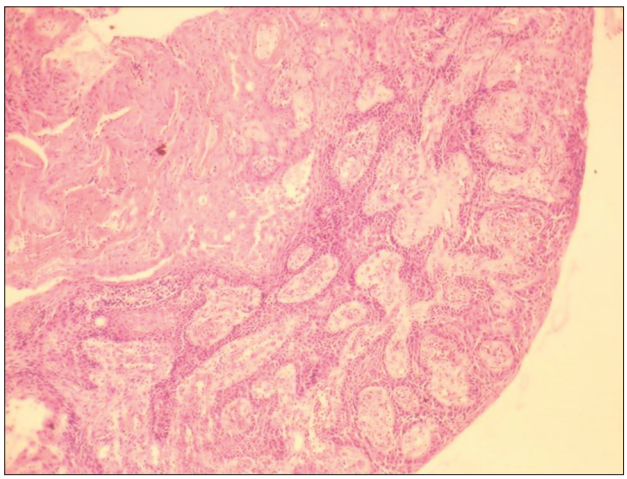

Figure 4: Photomicrograph showing intermingled areas of calcifying epithelial odontogenic tumor and plexiform ameloblastoma (H&E stain, x100)

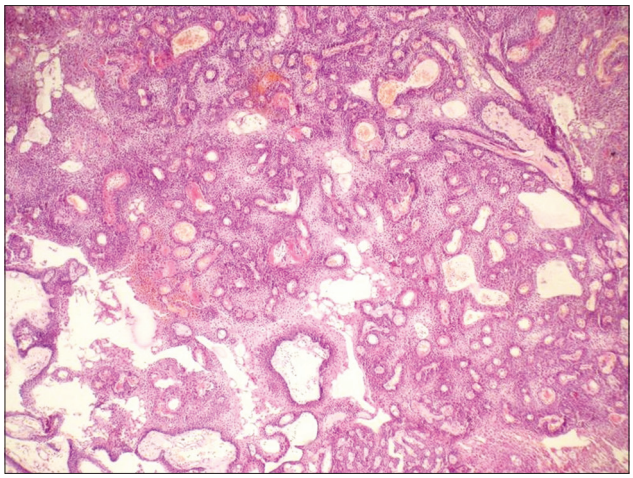

Figure 5: Photomicrograph shows odontogenic epithelial cells arranged in interlacing cords suggestive of plexiform ameloblastoma (H&E stain, x100)

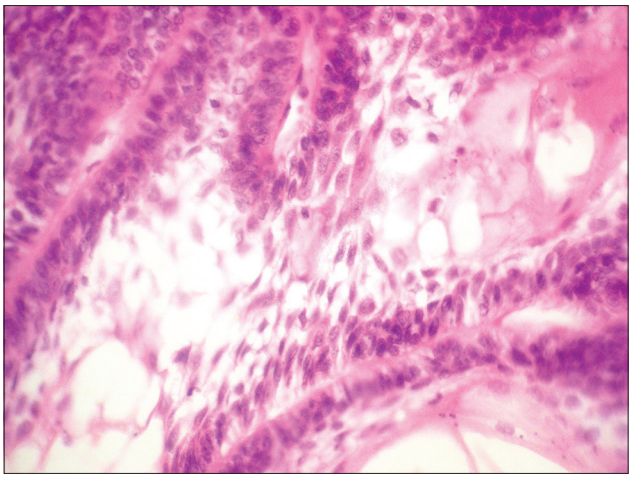

Figure 6: Photomicrograph shows plexiform ameloblastoma like areas (H&E stain, x400)

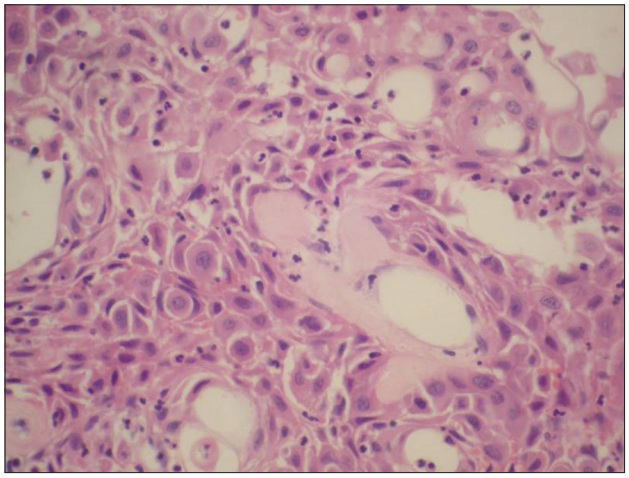

Figure 7: Photomicrograph shows slight pleomorphism, intercellular bridges and hyperchromatic nuclei in calcifying epithelial odontogenic tumor-like areas (H&E stain, x400)

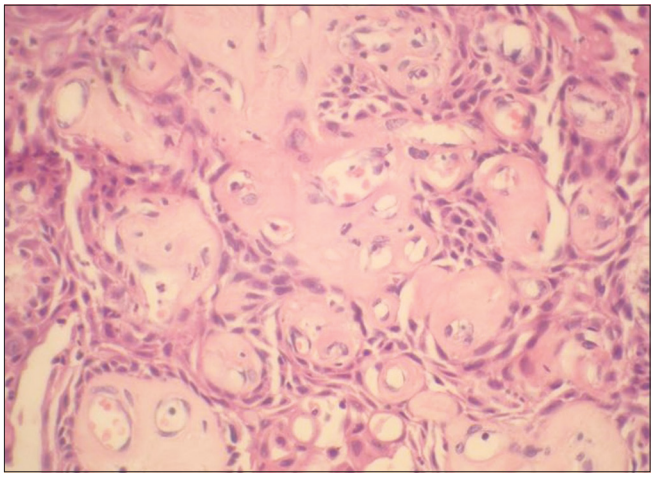

Figure 8: Photomicrograph shows acellular eosinophilic areas suggestive of amyloid-like areas (H&E stain, x400)

amyloid-like nature, which may become calcified and may be liberated as the cells break down.[10] CEOT arises from the reduced enamel epithelium of the closely related unerupted tooth. The peripheral variant may arise from rests of the dental lamina or from the basal cells of the oral epithelium.[1] CEOT shares many clinical features with ameloblastoma. However, microscopically there is no resemblance to ameloblastoma and distinct differences will be noted radiographically.[11] Solid multicystic ameloblastomas may arise from rests of a dental lamina or from the enamel organ. The basic histologic pattern of CEOTs, is characteristic and unique, is an unusual and variable combination of odontogenic epithelium and calcified structures. We observed the sheet-like proliferation of polyhedral cells along with numerous amyloid-like areas but no calcifications were evident in the received specimen histologically. Earlier literature studies have reported CEOTs without calcifications.[8,12] Intermingled areas of polyhedral cells with ameloblast-like cells were evident throughout the section. In both CEOTs and ameloblastomas, the mandible is affected twice as often as the maxilla and there is a predilection for the molar-ramus region. But in the present case, swelling was observed in the maxillary region.

Occasionally, hybrid odontogenic tumors have been reported in the literature, such as keratoameloblastoma,[13] hybrid tumor of ameloblastoma and glandular odontogenic cyst,[14] ameloblastoma and calcifying odontogenic cyst,[15] ameloblastoma and ameloblastic fibroma.[16] Also, most of them do not appear in the standard WHO classification of odontogenic tumors. These ambiguous tumors should not generally be considered true hybrids; rather they should be identified simply as anomalous histodifferentiation and/or morphodifferentiation process.[3] To call them as hybrid tumors would be a misnomer. Melrose wrote that the designation hybrid tumor serves no real purpose. It is probably due to the expression of the histomorphodifferentiation

Table 1: Clinicopathological features of all hybrid odontogenic tumors (CEOT+ameloblastoma) reported in the English literature

Author (year)	Age/sex	Clinical features	Radiological features	Histological features
Seim *et al.* (2005)[9]	53 years/ male	Right posterior maxilla; asymptomatic; mild erythematous gingiva and missing maxillary third molar	IOPA showed unilocular radiolucency	Features of both ameloblastoma (both follicular and plexiform) and CEOT; mild nuclear atypia; no mitotic figures and acellular eosinophilic interstitial material suggestive of amyloid
Etit *et al.* (2010)[8]	62 years/ female	Right maxilloalveolar edentulous ridge; 25 mm size; eroding overlying mucosa and no lymphadenopathy	Coronal computed tomography scan showed a soft tissue mass involving the right maxilloalveolar junction, associated with destruction of the maxillary sinus	Ulcerated surface epithelium; islands of odontogenic epithelium in mature connective tissue stroma; palisading arrangement of basal cells; dispersed islands of epithelial cells and amyloid-like areas
Present case	65 years/ female	Maxilla; huge; irregular, nodular soft tissue mass; buccal and palatal cortical expansion; bony hard	PNS view-a large radioopaque mass in the right maxillary region	Plexiform ameloblastoma with CEOT

CEOT: Calcifying epithelial odontogenic tumor, IOPA: Intraoral periapical radiograph, PNS: Paranasal sinus

potential of the epithelium and the mesenchyme and their complex inductive interaction, from which these lesions are derived.[9] Ameloblastomas commonly exhibit more than one histologic type in a single lesion. Thus, in such cases, the term "hybrid" is actually a misnomer. The present reported case is a very rare hybrid neoplasm consisting histologically of plexiform ameloblastoma and CEOT, appears to be the third reported case in the literature. The clinicopathological features of the present case have been compared with the previously reported cases in Table 1. Because both these neoplasms develop from a common or similar odontogenic epithelial source, a single tumor could exhibit histological features of both neoplasms.

Even though, the lesion was diagnosed as a hybrid odontogenic tumor showing the features of both CEOT and ameloblastoma, few shortcomings were encountered since the type of biopsy received was incisional and the entire specimen was not available for further investigations as the patient had left against medical advice. At First, no calcifications could be noticed in the present tissue sections although the lesion appeared radiopaque; second, paraffin-embedded tissue was insufficient to demonstrate amyloid using special stains and immunohistochemistry and finally, the appropriate treatment regimen required for this type of rare tumor could not assessed by postoperative follow-up.

In general, the biologic behavior of hybrid odontogenic tumors does not differ from that of other solid ameloblastomas. However, since the long-term behavior of this particular hybrid neoplasm remains unknown, the most appropriate approach has yet to be determined. CEOT has invasive potential but not to the extent of ameloblastoma. It is slow growing and compromises the patient through direct extension. Although, enucleation and excision appear to cure the hybrid lesion, long-term follow-up data and additional cases are still needed to substantiate the clinical significance of these lesions.

The authors would like to suggest that such lesions should be included in the WHO classification of Odontogenic tumors as they show the propensity of exclusively occurring in the maxillary region.

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Conflicts of interest

There are no conflicts of interest.

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