Unusual Variant of Ameloblastoma with Calcifying Epithelial Odontogenic Tumor-like Areas in a Three-Year-Old Child

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Case Report

Unusual Variant of Ameloblastoma with Calcifying Epithelial Odontogenic Tumor-like Areas in a Three-Year-Old Child

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Abstract

The Calcifying Epithelial Odontogenic Tumor (CEOT) is a benign odontogenic tumor, comprising approximately 1% of all odontogenic tumors. It presents as a slow-growing tumor in posterior mandibular region. A three-year-old female child from Multan, reported a swelling in right lower jaw progressively increasing in size eventually causing facial asymmetry. On intraoral examination, a protuberance was seen arising from posterior mandibular region and causing displacement of overlying teeth. The swelling was cystic on palpation and mildly tender. The patient underwent complete excision of lesion at a tertiary care center in her locality. The excision biopsy specimen was received at our lab which was intact and well preserved. A diagnosis of ameloblastoma with Calcifying epithelial odontogenic tumor-like areas was made. The patient was followed up after six months however no complaint of any recurrent swelling in the area of was recorded.

Keywords: Ameloblastoma, Calcifying Epithelial Odontogenic Tumor (CEOT), Pindborg Tumor

Introduction

The term "ameloblastoma" originates from English word "amel" meaning enamel and Greek word "blastos" meaning germ.[1] The neoplasm is thought to arise from odontogenic epithelium, particularly from dental lamina or enamel organ.[2] Ameloblastoma is one of the most common odontogenic tumors (approximately 10-50% of all odontogenic tumors). It predominantly occurs in the third to fourth decades of life, with a 1:1 male-to-female ratio, and often presents in the mandibular molar-ramus region. Radiologically, ameloblastoma appears as unilocular or multilocular radiolucency. Although it is a benign tumor, it demonstrates local aggressiveness and a high propensity to recur. Histologically, it consists of islands of proliferating odontogenic epithelium within fibrous stroma. Ameloblastma is separated into three clinical categories because of its presentation, treatment, and prognosis i.e Conventional, Unicystic, and Peripheral. Plexiform and follicular are most common histologic variants. When specific changes like granular transformation and squamous metaplasia occur, they are classified as granularcell and a anthomatousvariants, respectively. Granlarcell variantismost aggressive, characterized by a higher incidence of malignant transformation and a tendency to metastasize followed by desmoplastic variant having high recurrence rates owing to infiltrative margins.[3]

The Calcifying Epithelial Odontogenic Tumor (CEOT) is a benign odontogenic tumor, comprising approximately 1% of all odontogenic tumors. It was initially described by Pindborg, hence known as "Pindborg tumor". It presents as a slow-growing tumor in posterior mandibular region. Peak occurrence is in fourth and fifth decades of life CEOT is characterized by aunilocular or multilocular lesion, displaying mixed radiolucent-radiopaque pattern. Microscopically, it consists of three principal histological components: amyloid-like deposits, sheets of polyhedral epithelial cells, and calcifications.[4]

Both entities differ significantly based on their clinical course, etiology, histogenesis, treatment, and prognosis. Although Ameloblastomas often show a wide spectrum of histomorphological features however incidence of CEOT like areas in conventional solid multicystic ameloblastoma is exceedingly rare. This article presents a unique case of Ameloblastoma with areas resembling CEOT accompanied by a review of literature.

Case Presentation

A three-year-old female child, resident of Multan, a city in the province of Punjab reported a swelling in right lower jaw progressively increasing in size eventually causing facial asymmetry in the subject. On intraoral examination, a protuberance was seen arising from posterior mandibular region and causing displacement of overlying teeth. The swelling was tense (cystic) on palpation and mildly tender. The patient underwent complete excision of lesion at a tertiary care center in her locality. The excision biopsy specimen was received at our lab which was intact and well preserved.

On gross examination, it was a round mass 4.5cm, with solid to cystic cut surface. The cystic areas were filled with chocolate brown material. Histologically the neoplasm was composed of islands of stellate reticulum having loosely arranged oval to spindled cells with peripheral palisading. Overlying odontogenic epithelium showed classic reverse polarity.

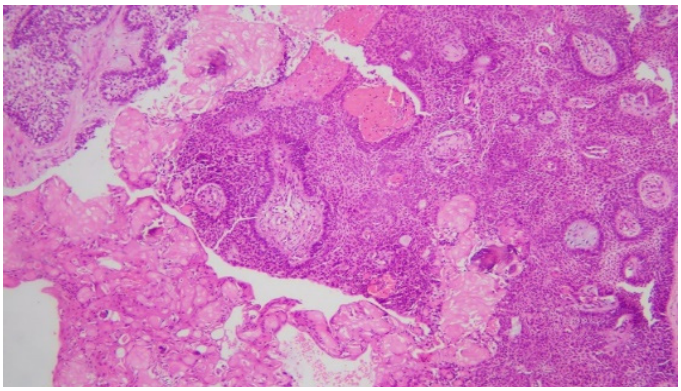

Figure 1: Low power view showing ameloblastoma with CEOT-like areas.

The follicular pattern was predominant, however, areas of plexiform pattern with anastomosing cords and sheets were also appreciated Figure 4)

Some of the foci showed polygonal epithelial cells with clear cytoplasm (ghost cells) and calcifications against a background of amyloid-like material (Figure 1,2,3). A diagnosis of Ameloblastoma with Calcifying epithelial odontogenic tumor-like areas was rendered to the patient. The patient was followed up after six months however no complaint of any recurrent swelling in the area of interest was recorded.

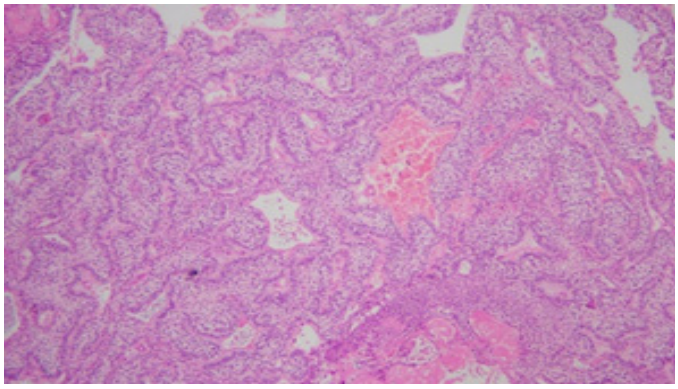

Figure 2: Low power view showing polygonal epithelial cells with ghost cells

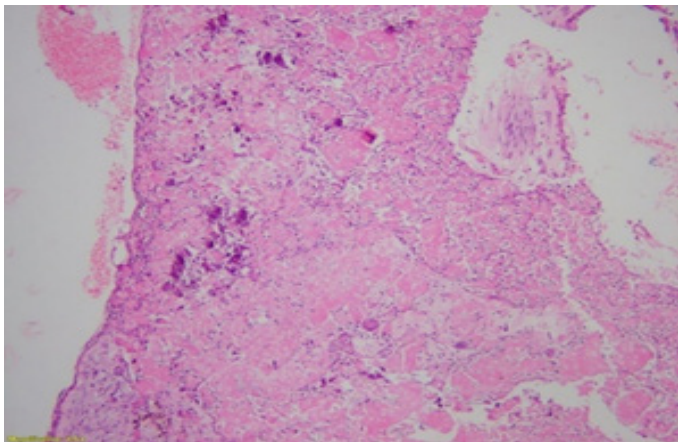

Figure 3: Low power view showing calcifications, ghost cells and amyloid like areas

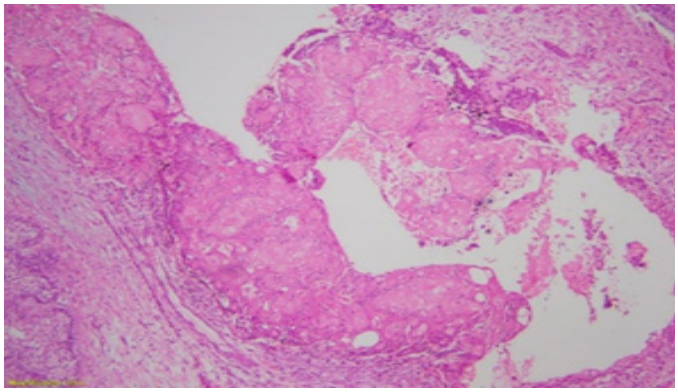

Figure 4: Low power view showing conventional ameloblastoma with follicular and plexiform pattern

Discussion

CEOT and ameloblastoma, might have overlapping clinical features. However, there are clear distinctions in radiographs and no similarities in terms of histology.

While reviewing the literature, we found quite a few reported cases of hybrid odontogenic tumors with dual morphology. Examples include Kerato-Ameloblastoma, Ameloblastoma and Ameloblastic fibroma, Ameloblastoma, and Calcifying Odontogenic Cyst, and Ameloblastoma and Glandular Odontogenic Cyst. These lesions should not be recognized as true hybrids; rather can be considered anoalous morphodifferentiation/ histodifferentiation process. Melrose writes that the term hybrid tumor is a misnomer.The odontogenic epithelium has eminent histomorphodifferentiation potential and undergoes complex inductive interactions with mesenchyme, giving rise to such lesions.[5]

Ameloblastoma are frequently aggressive with ability to destroy bone and grow upto varied sizes, leading to pathological fractures, facial asymmetry, tooth displacement, and malformities. The key prognostic factors for ameloblastoma are primary surgical approach and morphologic variant.[6,7] Recurrence is common with conservative therapy, ranging from 60% to 90% . The preferred method of treatment for biologically aggressive subtypes of primary and recurrent ameloblastoma is radical surgery i.e en bloc tumor excisiom with a safe bone margin of 1 to 2 cm.[8]

Contrary to ameloblastoma, CEOT is considered an expansile lesion that does not invade intertrabecular spaces. Various treatment modalities for CEOT have been proposed and recent data reinforces the idea that conservative surgery is preferred approach for mandibular CEOT.[9,10]

The available data suggests that when it comes to hybrid odontogenic tumors, they usually behave in a manner that is similar to other solid/multi-cystic ameloblastoma & none of these variants shows significant behavioral variability. Optimal treatment of hybrid lesions with an ameloblastoma component should follow the management guidelines for conventional

ameloblastoma.[10] This constitutes an important evidence to the proposition that such lesions are mere variants of Ameloblastoma and not composite tumors. However the long-term behavior of this hybrid tumor is uncertain, the best treatment strategy is yet to be defined. Even though enucleation and resection seemed to resolve the hybrid lesion in our case, additional data and long-term surveillance are required to establish the clinical importance of these lesions.

Conclusion

Ameloblastoma is well known to have multiple variants in a single lesion. The biological behavior of hybrid odon to genic tumors is typically the same as that of other solid or multicystic ameloblastomas. Therefore, we propose that ameloblastoma with CEOT-like areas may be a rare variant of ameloblastoma rather than a true hybrid neoplasm. To better understand the relevance of these lesions, more cases and follow-up information are required because the long-term behavior of this lesion is still unknown.

Authors' Contribution: The authors hereby validate their participation in the preparation of the manuscript in the subsequent manner: the inception and formulation of the study were done by MF, the collection of data was undertaken by AA and ZR, the analysis and interpretation of the obtained results were carried out by FWK and ZB, and the initial draft of the manuscript was prepared by MC.

Conflict of Interest: The authors have no conflicts of interest.

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