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CASE REPORT

Cystic Variant of Calcifying Epithelial Odontogenic Tumor

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ABSTRACT

Calcifying epithelial odontogenic tumor (CEOT) is an uncommon benign, locally invasive odontogenic epithelial neoplasm, which accounts for 1% of all odontogenic tumors. Two important features distinguish CEOT from other odontogenic tumors. Firstly, nuclear pleomorphism of lesional cells, although suggestive of malignancy, is considered benign. Secondly, the tumor is locally invasive with indolent biological behavior. Microscopically, CEOT exhibits distinct diagnostic features. Seldom histological diversities are reported in the literature. We report a rare case of cystic variant of CEOT along with focus on other variants with a review of literature.

Keywords: Amyloid, Calcifying epithelial odontogenic tumor, Pindborg tumor.

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INTRODUCTION

Calcifying epithelial odontogenic tumor was first described in 1955 by a Dutch pathologist Pindborg[1] as a distinct entity derived from the stratum intermedium of the bell-staged enamel organ showing characteristic histopathological features. The CEOT can be present both intraosseous and rarely in extraosseous locations. It occurs over a wide range with a peak incidence in the 40s. It is twice common in the mandible compared with the maxilla with a predilection for the posterior region.[2] Wide radiological appearance is noted from an early complete radiolucent lesion to a mixed lesion. It may be present as a unilocular radiolucency to a multilocular radiolucency mimicking soap bubble appearance. The World Health Organization in 1992 classified it as a benign odontogenic tumor, which is exclusively epithelial in its tissue of origin. The differential diagnosis for CEOT should include adenomatoid odontogenic tumor (AOT), calcifying odontogenic cyst, ameloblastic fibro-odontoma, and odontoma.[3] Few CEOT cases have been associated with impacted teeth resembling dentigerous cyst radiographically; true cystic variants are less reported. We report a case of a cystic variant of a CEOT in a young male patient.

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CASE REPORT

A 26-year-old male patient visited our center with history of pain and swelling in the right posterior region of the mandible since 1 year. The patient started noticing an asymptomatic swelling about a year back, which was of the size of a walnut, and which eventually grew to the present-day size and was associated with pain for a week. The swelling, about 3 × 3 cm in the right body of mandibular region, extended from 1st premolar region to the 2nd molar region and was just behind the corner of the mouth until just below the lower border of mandible. The skin over the swelling appeared normal, intraorally extending from 1st premolar to the 2nd molar and causing obliteration of the sulcus, with the mucosa over it appearing inflamed. The consistency of the swelling was hard and tender on palpation. Right submandibular lymph node is palpable and tender. Tooth 46 was missing. Radiographically, a well-circumscribed radiolucent lesion in the right body of mandible associated with an impacted molar in the lower border was seen. Aspiration of the lesion revealed pus. Surgical excision of the same considering it to be an infected dentigerous cyst was planned. Under local anesthesia, a vestibular incision was placed, and complete enucleation of the lesion and the removal of the tooth were carried out. The provisional clinical diagnosis of ossifying fibroma, CEOT, ameloblastoma, and odontogenic myxoma was made. The specimen was sent for histopathological evaluation. Histopathology findings revealed a neoplasm composed of cells arranged as sheets and anastomosing small and large islands. These cells were interspersed by prominent homogeneous hyaline acellular material. Areas of concentric lamellated calcifications were seen. The neoplastic cells have well-defined cell borders, abundant eosinophilic cytoplasm, and hyperchromatic mildly pleomorphic nuclei. A few bizarre nuclei were seen; however, no abnormal mitosis was seen. Normal mature lamellar bony trabeculae were seen between tumor islands interspersed with large areas of hemorrhages, as

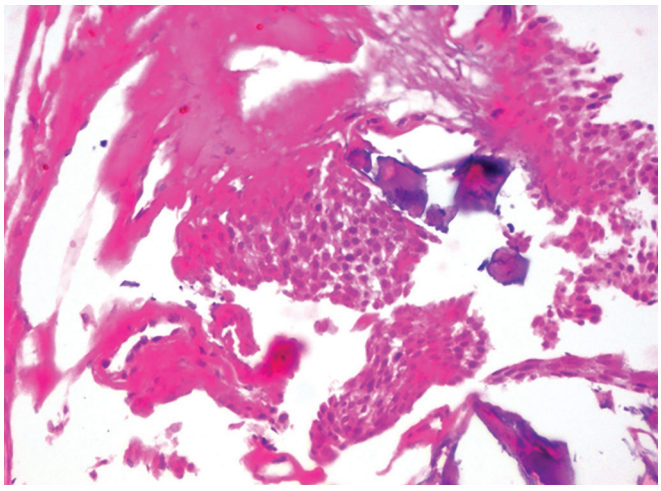

Fig. 1: Histopathological finding of CEOT

seen in Figure 1. The eosinophilic material was confirmed as amyloid upon special staining, diagnosing the lesion to be a CEOT.

DISCUSSION

The CEOT/Pindborg tumor is a rare benign, but locally aggressive tumor. Neville et al,[2] in their textbook of oral and maxillofacial pathology, assert that the lesion is a distinct entity and probably represents less than 1% of all odontogenic neoplasms. Clinically, it may present as a slow-growing painless mass with predilection for mandible. Maxillary lesions are more aggressive when it invades the maxillary sinus, orbit, and ethmoid sinus causing pain, nasal obstruction, epistaxis, and headache.[4] Histogenesis of the tumor is controversial. Most investigators are of the opinion that it arises from stratum intermedium as tumor cells are morphologically similar and possess a high alkaline phosphatase and adenosine triphosphate activity.[5] Other possible considerations for the cell of origin are from oral epithelium, dental lamina or its remnants, and reduced enamel epithelium.[2] The CEOT shows three important histological features. It is composed of sheets and strands of polyhedral eosinophilic cells showing prominent intercellular bridges. Some cells may be spindle shaped. Nuclear variations like nuclear pleomorphism, binucleation, prominent nucleoli, hyperchromatism, and giant nuclei are seen. Despite nuclear atypia, there is no detectable mitotic activity.[6] The connective tissue contains a finely fibrillar or hyaline eosinophilic matrix component. An extracellular eosinophilic homogeneous material staining like amyloid is characteristic of this tumor. Nature of amyloid material is uncertain and the following hypotheses have been suggested[6]:

- It represents degradation of lamina densa.
- It is thought to be extracellular deposits (collagen type IV and laminin).
- Amelogenins and enamelin, precursors of enamel matrix protein, have been identified and support the secretory rather than degenerative hypothesis.

Amyloid material can be enhanced and observed using special stains like Periodic acid–Schiff. It stains positively with congo red (apple green birefringence) under polarized light and fluorescence with thioflavin T. Mineralization exhibits Liesegang-ring phenomenon showing multiple, concentric, hematoxyphilic deposits.[6]

Histological variants of CEOT:

- Calcifying epithelial odontogenic tumor with cementum-like substance[7]
- Noncalcifying variant[7]
- Clear cell variant[8]
- *Calcifying epithelial odontogenic tumor with Langerhans cells*: due to the secondary inflammatory changes[7]
- *Calcifying epithelial odontogenic tumor with myoepithelial differentiation*: spindle cells with features of smooth muscle cell with no desmosomes[7]
- Malignant CEOT[9]
- Hybrid tumors (CEOT and AOT, CEOT and ameloblastoma)[10]
- Cystic variant

Cystic degeneration is an expected phenomenon in a solid tumor. The other possibility is that tumor can exhibit a cystic growth pattern as it occurs in unicystic ameloblastoma. Histological variants of CEOT may not influence the biological activity of the tumor. But, it is considered that clear cell variant is more aggressive than conventional types.[8]

TREATMENT

Numerous surgical treatment modalities have been suggested, and the treatment plan is dependent on multiple factors, such as size and location of neoplasm, general condition of patient, and operator skill. Small, intrabony mandibular lesions with well-defined borders are treated by simple enucleation or curettage followed by judicious removal of a thin layer of bone adjacent to the tumor.[5] Large tumors require aggressive approach by segmental resection, hemimandibulectomy and hemimaxillectomy, which cause bone discontinuity requiring reconstruction procedures, such as grafting or distraction osteogenesis.[7] Recurrence rate of 10 to 20% following conservative treatment is reported. Malignant transformation and metastasis are rare.[5,9]

CONCLUSION

The CEOT does not show intramedullary spread and, thereby, behaves less aggressively with a recurrence rate of 14%. Around 200 cases of CEOT have been reported until date with very few reports on cystic variant of CEOT.

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