

CASE REPORT

Pindborg Tumor in an Adolescent- An Atypical Presentation

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Abstract

Calcifying epithelial odontogenic tumor (Pindborg tumor), is a rare benign odontogenic neoplasm representing about 0.4-3% of all odontogenic tumors. This tumor more frequently affects adults in an age range of 20-60 years, with a peak incidence in the 5th decade of life. Calcifying epithelial odontogenic tumour has a much lower recurrence rate than ameloblastoma and malignant transformation with metastasis is rare.

Key Words

Calcifying Epithelial Odontogenic Tumour, Pindborg Tumour, Mandible

Introduction

The calcifying epithelial odontogenic tumour (CEOT) is a rare tumour. It was first described as a separate pathologic entity by Dutch pathologist Jens Jorgen Pindborg (1,2) in 1955 . The term “Pindborg’s tumour” was first used by Shafer and colleagues in 1963 (3).

Case Report

A 16 year old male, presented with a hard nodule, 6.5 x 4.5 cm in size, on the buccal aspect of right molar region of the mandible above the angle of jaw. The swelling was tender and increased progressively over a period of 1 year. Roentgenogram on admission revealed a loculated /trabeculated radioluscent cyst above the angle of right mandible measuring 6 x 4 cm. The provisional clinical diagnosis of Aneurysmal bone cyst/ Ameloblastoma/ odontogenic keratocyst was made. Resection of the right mandible from the first bicuspid through the condyle including the whole growth was performed and the specimen was submitted for histopathology.

Gross examination revealed a globular bony tissue with attached soft tissue piece measuring 6 x 3 cms. On cut section, a cystic growth was seen with bony loculations, along with cartilaginous to haemorrhagic areas (*Fig. 1*). Microscopic examination of the tissue section revealed sheets of polyhedral epithelial cells in definite lobules. The closely packed cells were small with mild nuclear pleomorphism and moderate eosinophilic cytoplasm. Numerous spherical calcified masses were seen in a

background of cellular degeneration with scant fibrous stroma (*Fig. 2a & b*). The diagnosis of calcifying epithelial odontogenic tumour (Pindborg tumour) was made.



Fig. 1 Gross Specimen of CEOT Showing Rounded & Predominantly Cystic & Haemorrhagic Growth on Cut Section

Discussion

CEOT is a rare, benign, but locally aggressive tumour, which accounts for less than 1% of all odontogenic tumours and most often located in the posterior mandible (4,5,6). Local recurrence rates of 10-15% have been reported, and malignant transformation is rare (5). Etiology of this lesion is not clear. Majority of the investigators are of the

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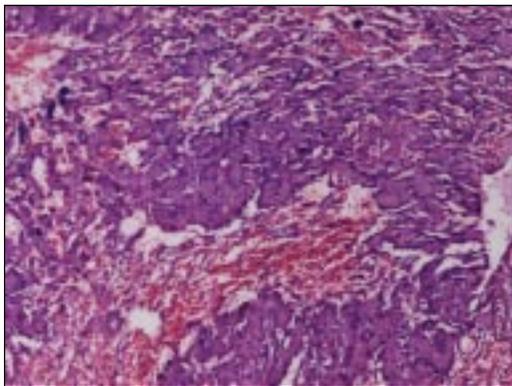


Fig. 2 a. CEOT: Histologic Examination Reveals Sheets of Polyhedral Epithelial cells with Calcific Foci in a Background of Cellular Degeneration and Haemorrhage. H&E X 200

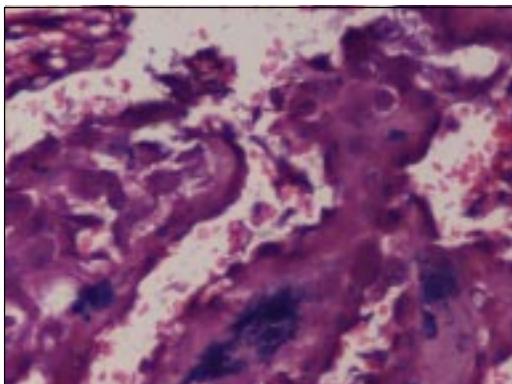


Fig 2 b. CEOT: Tissue Section Shows Sheets of Polyhedral Epithelial Cells with mild Nuclear Pleomorphism and Moderate Eosinophilic Cytoplasm with Spherical Calcific Masses H&E X 400

opinion that, the tumour cells originate from the striatum intermedium of the normal dental lamina (7), an idea based on the morphologic similarity of the tumour cell to the normal cells of stratum intermedium and a finding of high activity of alkaline phosphatase and adenosine triphosphate at both sites (8).

In the 113 cases reviewed by Franklin and Pindborg (9), patients age ranged from 8 to 92 years with a mean age of 40 years. According to Cicconetti and colleagues (10), the tumor more frequently affects adults in an age range of 20-60 years, with a peak incidence in the 5th decade of life with equal sex predisposition. There is a marked predilection for the molar – premolar area of mandible with about 50% cases associated with unerupted or embedded teeth. Radiologically, this tumour is often mistaken for dentigerous cyst or ameloblastoma (6). Quite similarly; the radiographic findings in our case showed a loculated /trabeculated radioluscent cyst. The diagnosis of calcifying epithelial

odontogenic tumour is based on histological examination, revealing polyhedral neoplastic cells which have abundant eosinophilic, finely granular cytoplasm with nuclear pleomorphism and prominent nucleoli. Most of the cells are arranged in broad ramifying and anastomosing sheet-like masses with little intervening stroma; similar morphologic features were visualized in our case. An extracellular eosinophilic homogenous material staining like amyloid is characteristic of this tumour with concentric calcified deposits, resembling psammoma bodies called “Liesegang rings. This case also depicted calcific foci in abundance and also fused amorphous calcareous aggregates. The CEOT is considered to have a lower recurrence rate than that of ameloblastoma and malignant transformation and metastasis is rare (11). Our patient underwent hemimandibulectomy and no recurrence was reported in 6 months of follow up.

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