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Calcifying Epithelial Odontogenic Tumor (Pindborg Tumor): A case Report

DR.MANJEET SINGH *(SENIOR RESIDENT)*
DEPARTMENT OF DENTISTRY,
SLBSGMCH,NERCHOWK,MANDI,(HP)

Abstract: *Calcifying Epithelial odontogenic tumor(CEOT) is a rare benign odontogenic neoplasm described in 1955 by Danish pathologist Jens J.Pindborg and is now known as Pindborg tumour. It representing less than 1% of all odontogenic tumors. I am reporting a case of CEOT in a 33-years-old male patient with painful bony swelling in the mandible on left side with embedded permanent canine. The clinical, radiographic, and histopathologic features are discussed with relevant references.*

Keywords: *Calcifying epithelial odontogenic tumor, odontogenic epithelial tumor, pindborg tumor,CEOT.*

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I. Introduction

Calcifying epithelial odontogenic tumor (CEOT) is an odontogenic tumor arising from the odontogenic epithelium.It was first described in 1956 by the Late Dr. Jens J Pindborg. Hence, CEOT is also called Pindborg tumor.[1,2] It is very uncommon and accounts for <1% of all odontogenic tumors.[1,3] Approximately, 200 cases have been reported till date.[3] Histogenesis is uncertain and is believed to arise from remnants of dental lamina and stratum intermedium.[2,4] The most commonly involved part is posterior aspect of mandible in premolar and molar region and less frequently in maxilla. The lesion is locally aggressive, slow growing benign odontogenic neoplasm tends to invade local structures and has a potential for recurrence. It occurs most commonly in adult in 3rd to 6thdecade of life with no gender predilection.

II. Case Report

A 33-years-old male reported to the dental OPD,SLBSGMCH NERCHOWK,MANDI, HP. with pain and swelling in the left side of of lower jaw since 6 months. The swelling was tender and had progressively increased over a period of time leading to facial asymmetry. The dentition was normal with normal occlusion except 33 was absent in oral cavity. Orthopantomograph [Figure 1] revealed a mixed radiolucent-radiopaque lesion, which was uilocular with coarse trabeculae and scattered foci of calcification extending from left lower lateral incisor to first molar region with a radio opaque mass representing embedded 33. There is resorption of the roots of lateral incisor and both the pre-molors. A presurgical computed tomography (CT) scan [Figure 2] was obtained to ascertain the boundaries of the neoplasm. It [Figure 3] revealed an osteolytic lesion with foci of calcifications. The inferior border of the mandible was intact and about 3-4 mm bone was present in the pathological area. The provisional clinical diagnosis of ossifying fibroma, calcifying epithelial odontogenic tumor, ameloblastoma and odontogenic myxoma was made. Patient was systemically healthy and all other blood investigations were with in normal limits. An enucleation and curettage of the lesion along with extraction of embedded canine and surrounding teeth from the pathological region was planned under local anesthesia. The removed tissue sent for HPE.

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, 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 haemorrhage. The eosinophilic material was confirmed as amyloid upon special staining, diagnosing the lesion to be CEOT.

III. Discussion

Pindborg tumor is a rare benign but locally aggressive tumor. Neville and colleagues assert that the lesion is a distinct entity and probably represents less than 1% of all odontogenic neoplasms. There are differences of opinion within the oral and maxillofacial pathology community regarding degree of

differentiation of the odontogenic epithelium which forms the basis for tumor pathogenesis. Some authors suggest that the epithelial cells of the Pindborg tumor are reminiscent of the sequestered cells in the stratum intermedium layer of the enamel organ.[3-5] This idea is based on the morphological similarity of the tumor cells to the normal cells of stratum intermedium and a high activity of alkaline phosphatase and adenosine triphosphate.[6] It has been suggested that amyloid deposition within Pindborg tumor is an immunologic response to these stratum intermedium cells. Others insist that it arises from remnants of the primitive dental lamina found in the initial stage of odontogenesis, and these epithelial rests are the more likely true progenitor cell.[7] However, the exact etiology is unknown. CEOT occurs most commonly between 20 to 60 years of age with mean around 40 years. In 113 cases reviewed by Franklin and Pindborg,[8] patients ranged from 8 to 92 years of age with mean at 40 years. In 2004, Cicconetti and colleagues reported that tumor more frequently affects adults in the age range of 40 to 60 y ears with peak incidence in the 5th decade with an equal sex distribution.[9] Ninety-four percent of the lesions are central and intraosseous and 6% are extraosseous. Intraosseous CEOT shows a maxilla-to-mandible site ratio of 1:2 and is mainly located in the premolar/molar region. Half of the cases are associated with an impacted tooth.[7] Fifty-two percent cases have been associated with unerupted or embedded tooth. Similar finding was present in the case reported as canine was embedded. CEOT may lead to tooth tipping, rotation, migration, and/or mobility secondary to root resorption. This lesion is often symptomless and discovered on routine radiography. Alternatively it may present symptomatically as a slow-growing, painful, expansile, bony swelling with cortical bone resorption and finally perforation, as was seen in the case reported. In the initial stage, it is totally radiolucent, simulating a dentigerous cyst because of its relation with impacted tooth. Small intratumoral calcification starts appearing in the second phase, which is characteristic but not diagnostic. The final stages are associated with osseous destruction and the tumoral calcification giving it a honeycomb appearance. Quite similarly the radiographic finding in our case showed a loculated/ trabeculated honeycomb mixed radiolucent radiopaque finding. The diagnosis of CEOT is also based on histopathological 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 anastomosing sheet like masses. An extracellular eosinophilic homogenous material staining like amyloid is characteristic of this tumor with concentric calcific deposits called Liesigany Ring. [5] The case described also depicted calcific foci in abundance with fused amorphous calcified aggregates.

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.[6] Large tumors require aggressive approach by segmental resection, hemimandibulectomy and hemimaxillectomy, which causes bone discontinuity requiring reconstruction procedures such as grafting or distraction osteogenesis.[3,4,6] Recurrence rate of 10−20% following conservative treatment is reported.[5,9] Malignant transformation and metastasis is rare.[9] Patient reported here underwent simple enucleation and curettage, extraction of embedded and adjacent teeth with resorbed roots and there has been no recurrence on 6 months follow-up.[Figure 4]

Figures

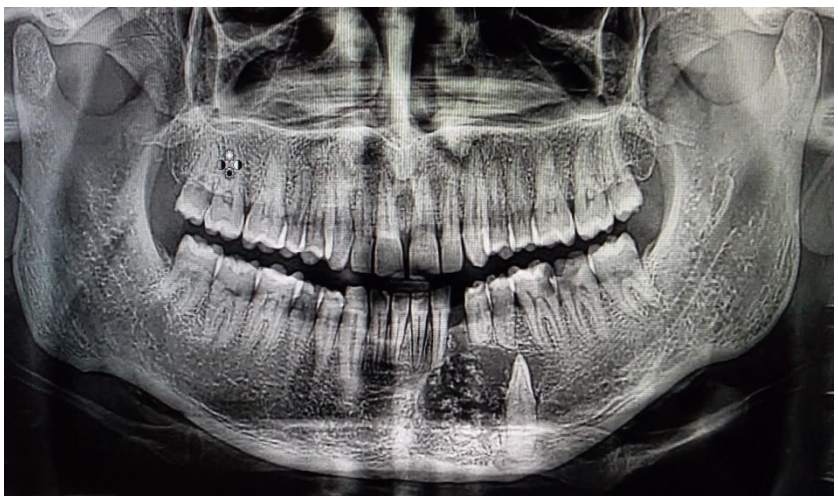

Figure 1. Orthopentomograph showing lesion in mandibular left side with embedded canine.

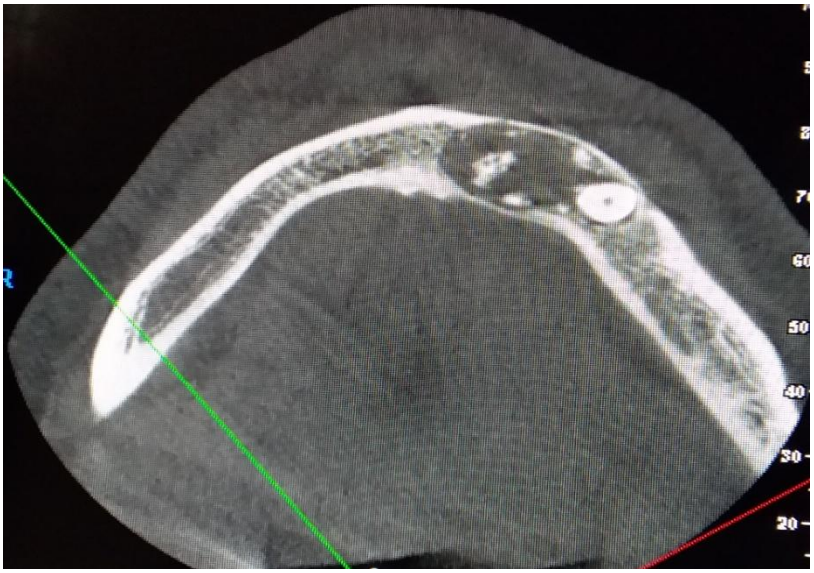

Figure 2

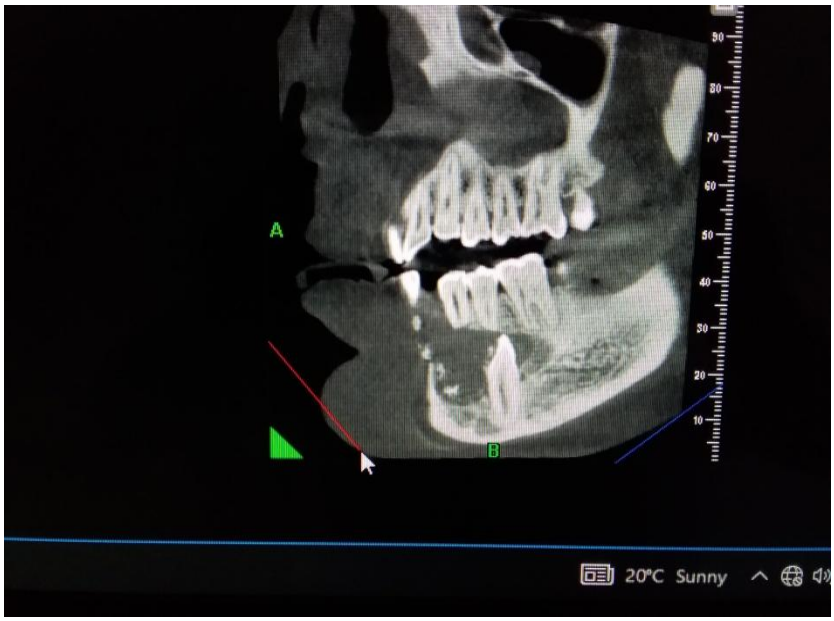

Figure 3

Figure 2&3 showing extension of bony lesion with calcified tissue with in radiolucent area and resorption of the roots of surrounding teeth.

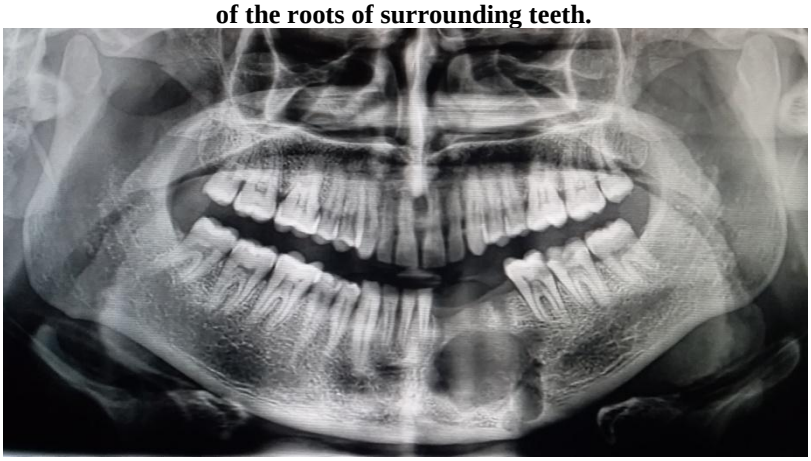

Figure 4. An enucleation and curettage of the lesion along with extraction of embedded canine.

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