Case Report

Pindborg's tumor: Report of a case with 6 years of follow-up

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ABSTRACT

Calcifying epithelial odontogenic tumor (Pindborg's Tumor) is an odontogenic tumor derived from dental lamina. It is relatively an uncommon odontogenic tumor, which shares clinical features with ameloblastoma. Here, we are reporting a case of Pindborg's tumor with 6 years of follow-up we are presenting this case with all the available clinical data. There was no recurrence within the 6 years of follow-up.

Key words: Liesegang rings, odontogenic tumor, Pindborg's tumor

INTRODUCTION

The calcifying epithelial odontogenic tumor (CEOT) was first described as an entity by Danish Pathologist Pindborg in 1955.[1] Furthermore known as Pindborg's tumor, it is an uncommon, locally invasive, benign odontogenic tumor occurring in individuals over a wide age range, but peaking in incidence in the 40s. It usually presents as a hard painless mass, generally affecting the mandible. The characteristic histopathologic description consists of sheets and islands of polygonal cells that often have distinct intercellular bridges. The nuclei may be pleomorphic and hyperchromatic and bizarre in appearance. Mitoses are very uncommon. Pale eosinophilic masses (amyloid-like) may be found within the sheets of tumor cells and can undergo calcification, often concentrically in the form of Liesegang rings.[2]

CASE REPORT

This was a case report of a 35-year-old male patient who reported to Government Dental College, Trivandrum, South India in April 2007 with a chief complaint of swelling in the lower front jaw region since last 2 years. Patient also had difficulty in chewing. On examination, extra-orally there was hard, non-tender swelling in the lower anterior region of size 3 cm × 2 cm. Extension of swelling was not well-defined. Intra orally swelling was obliterating the mandibular labial vestibule and with lingual cortical expansion. Clinically swelling was extending from 37 to 47 region with a size of 7 cm × 4 cm in its greatest dimension. Lingual swelling was irregular and creating the problem in tongue movement. Lower anterior teeth were having grade II mobility and were misplaced. There was paraesthesia of the lower lip. Oral hygiene status was poor. Systemic and family history was non-contributory. By above mentioned clinical features, provisional diagnosis of benign odontogenic tumor was made and central giant cell granuloma was the differential diagnosis. As investigation, orthopantomogram (OPG) and computed tomogram (CT) scan were advised. OPG showed mixed radiolucent, radio-opaque lesion extending from 36 to 44 regions anterio-posteriorly with root resorption of 31, 32, 41, 42. Interesting thing noted in OPG was typical driven snow appearance that was diagnostic for Pindborg's tumor [Figure 1]. CT scan was taken to assess the exact extension of the lesion. CT scan showed mixed radiolucent, radio-opaque lesion with discontinuity of lingual and buccal cortex with lesion extending into the soft-tissues lingually and it was extending from 36 to 45 region [Figure 2]. To confirm the diagnosis, incision biopsy was taken. Hematoxylene and eosin stained section showed strands and nests of polygonal epithelial cells with eosinophilic cytoplasm and prominent nuclei. The intercellular bridges were distinct and were irregular in shape with pleomorphic nuclei. Numerous concentric ring of calcified material was also noticed [Figure 3]. The definitive diagnosis of Pindborg's tumor was made.

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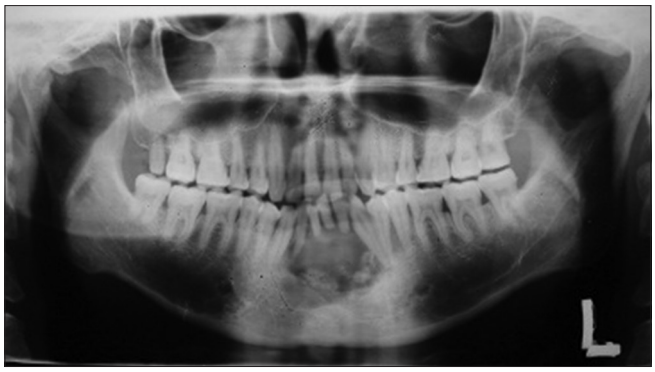

Figure 1: Orthopantomogram showing typical "Driven Snow" appearance

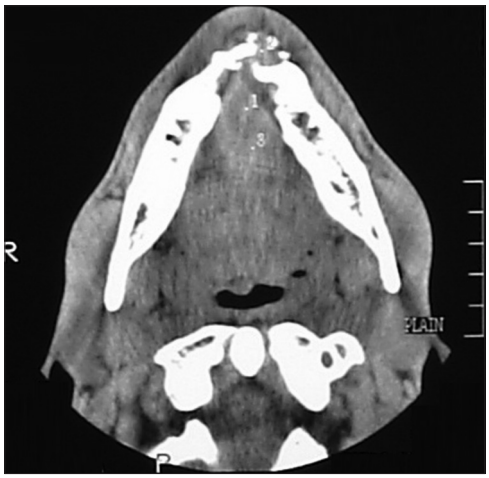

Figure 2: Computed tomogram scan showing breach in buccal and lingual cortical plate with soft-tissue enhancement

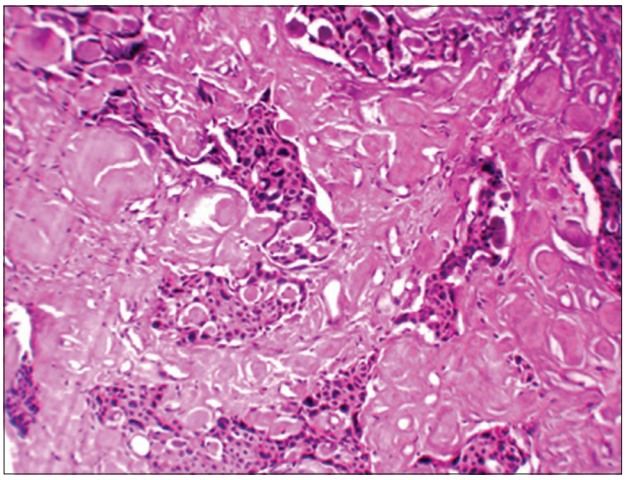

Figure 3: H and E section showing polygonal cell with distinct intercellular bridges, hyperchromatic nuclei and concentric calcified rings (Liesegang rings)

Segmental resection of mandibule from 37 to 46 region done under general anesthesia. As in CT there was infiltration into soft-tissue and with root resorption as shown in OPG. This case was put under regular follow-up and after 6 year also there was no evidence of recurrence.

DISCUSSION

In general, the CEOT is considered an uncommon to rare odontogenic neoplasm. Although Pindborg's tumor shares clinical features with ameloblastoma as to the site and age predilection, it is significantly less frequent in incidence. The Pindborg's tumor probably represents less than 1% of all odontogenic neoplasms.[3] Pindborg's tumor is most commonly reported in patients during the fourth, fifth and sixth decades of life (ages 30-50 years). The mean patient age for this lesion is variably reported in different studies between the late 30s to early 40s and is probably within the range of 37-43 years of age. Age of patient in this reported case was 35 years. No gender predilection had been reported for Pindborg's tumor.[4] An excellent description of clinical features of Pindborg's tumor was given by Robert in 2004. Like ameloblastoma, the Pindborg's tumor presents most often (75% of the time) as an intrabony, mandibular lesion. In the mandible, usually (60% of the time) is found in the posterior body to ascending ramus region. Though observed less frequently in the maxilla than the mandible (ratio: 3:1), when present in the maxilla, the calcifying epithelial tumor is again preferentially located in the area of the posterior teeth. Pindborg's tumor may present in a patient who lacks symptoms and consequently may be discovered only through routine radiographic examinations by the dentist, or it may present symptomatically as a slow-growing, painless, expansile, hard, bony swelling causing cortical bone to become egg-shell thin before perforation and subsequent soft-tissue infiltration. Pindborg's tumor may potentially cause associated tooth tipping, rotation, migration and/or mobility secondary to root resorption.[4] All the above mentioned clinical feature were observed in our case. Our patient reported to the clinician because of the difficulty in chewing. Radiographically Pindborg's tumor is a mixed radiolucent radio-opaque lesion with honeycomb or soap bubble appearance. Flakes of calcified mass will give "driven snow" appearance, which was noted in the presented case in OPG. On CT examination, Pindborg's tumor has been reported in the mandible as demonstrating expansion and thinning of buccal and lingual cortical bony plates by a well-defined mass containing scattered radiopaque areas of varying size and signal intensity.[4] In our case, there was the destruction of lingual and buccal cortical plates with soft-tissue involvement. Treatment is dependent on the size and location of the neoplasm, the patient's overall medical condition or tolerance to withstand the surgical procedure and the skill or experience level of the operator. Recurrent or persistent disease and tumors diagnosed late in the duration of their clinical course, which over an extended time have become larger and more extensive (greater than 4 cm), may not be curable by conservative surgical measures such as en-bloc resection only.[4] In the presented case, en-bloc

resection was done with reconstruction by reconstruction plate. CT scan showed soft-tissue involvement; and root resorption was evident in OPG. We have categorized this case as aggressive in nature and planned for long-term follow-up. After 6 years of follow–up, there was no recurrence.

CONCLUSION

Pindborg's tumor is an uncommon tumor of odontogenic origin. Recurrence rate is extremely low after en-bloc resection, yet it takes 5-10 years to find clinically diagnostic recurrence. So whether the lesion is clinically aggressive or not' it should be regularly followed-up by routine radiographic evaluation.

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