Pindborg tumor

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Abstract

Calcifying epithelial odontogenic tumor (CEOT), also known as Pindborg tumor, is a rare odontogenic epithelial neoplasm. So far, nearly 200 cases have been reported in the literature. We are reporting a case of CEOT in a 42-year-old male patient with painless bony swelling in the mandible. The clinical, radiographic, and histopathologic features are discussed with relevant references.

Keywords: Calcifying epithelial odontogenic tumor, odontogenic epithelial tumor, pindborg tumor

Introduction

Calcifying epithelial odontogenic tumor (CEOT) is an odontogenic tumor arising from the odontogenic epithelium.[1] 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] Here, we report a case of CEOT from our institution.

Case Report

A 42-year-old male patient reported to our institution with a chief complaint of a swelling in the right side of the mandible in the region of 43, 44, and 45. The mobile 43 and 44 were extracted in a private clinic 2 years back.

Extraorally, there was a diffuse swelling on the right side of the face with associated facial asymmetry.

The swelling was firm in consistency and nontender. Lymph nodes were not palpable, and the temporomandibular joint was normal.

Intraorally, there was an exophytic soft tissue overgrowth in the region of 43 and 44. Both these teeth were missing. On palpation, the intraoral swelling was smooth, firm, and nontender with bicortical expansion of both buccal and lingual cortical plates. The color was normal, but the shape was irregular. There was a displacement of 42 and 44 with grade III mobility [Figure 1].

Radiographic examination revealed missing 43 and 44 with a multilocular radiolucency, sclerotic borders, and displacement of 42 and 45. The lower border of the mandible was intact. Few radiopaque spots were seen within the radiolucency [Figure 2]. Mandibular occlusal view radiographs revealed bicortical expansion of both buccal and lingual cortical plates. Computed tomography scan with three-dimensional reconstruction showed an expansile soft tissue mass with areas of calcification [Figure 3].

Incisional biopsy was done, and a small bit of lesional tissue was given for histopathological examination. Histopathology of the incisional biopsy specimen showed features of CEOT.

The lesion was surgically excised, and the surgical site was reconstructed. Histopathology of the excised specimen showed islands and strands of polyhedral epithelial cells with prominent intercellular bridges. The connective tissue showed collagen fibers and eosinophilic homogenous, acellular areas resembling amyloid. Leisegang ring type of calcifications were also seen [Figures 4 and 5]. From the histopathology, a diagnosis of CEOT/Pindborg tumor was made.

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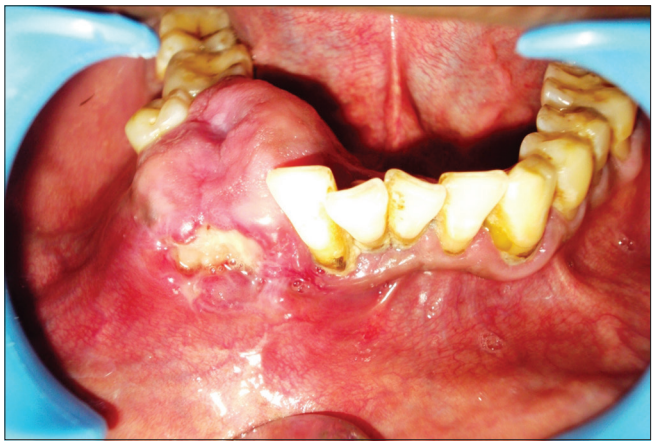

Figure 1: Exophytic growth in relation to 42, 43, 44, and 45

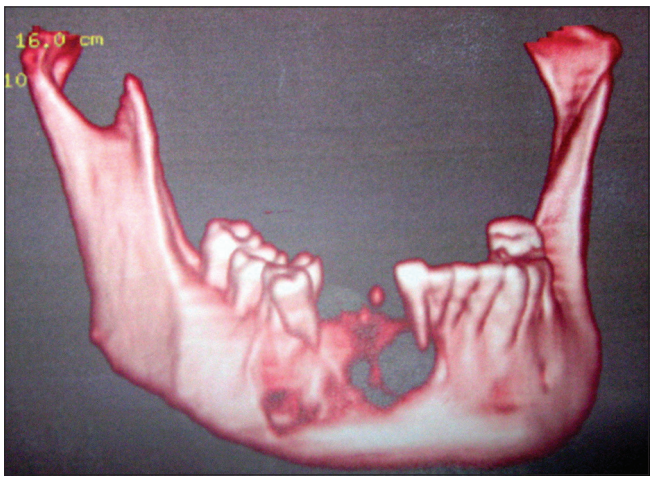

Figure 3: Computed tomography reconstructed image of the lesion

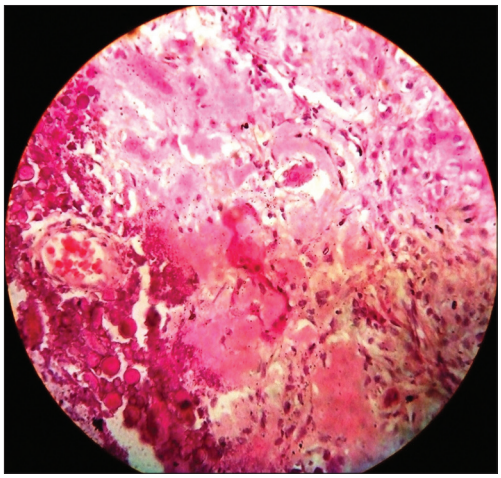

Figure 5: Leisegang ring calcifications and amyloid-like material in connective tissue stroma

The patient was advised to come for regular follow-up to monitor the healing and prognosis. The follow-up of 1 year revealed good healing of the site and no recurrence.

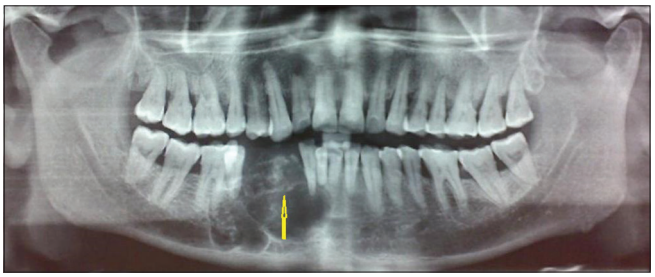

Figure 2: Multilocular radiolucency with many radiopaque spots of driven snow appearance

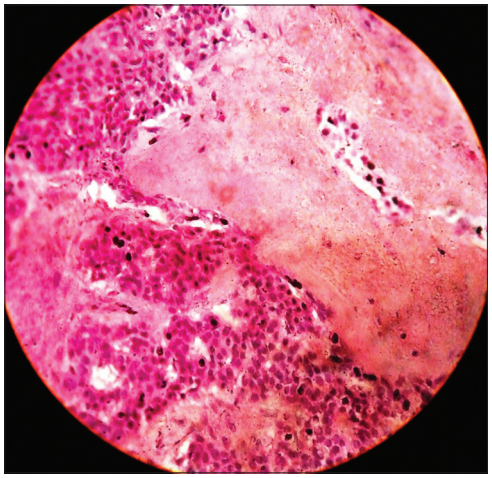

Figure 4: Epithelial cells with prominent intercellular bridge and amyloid-like material

Discussion

CEOT is a rare odontogenic epithelial tumor.[1,3] It was first described by the oral pathologist Pindborg, so CEOT is also called Pindborg tumor.[1-3] It accounts for <1% of all odontogenic tumors.[1-3]

Histogenesis is uncertain, mostly believed to arise from stratum intermedium of dental lamina.[1-3] This is based on the morphological resemblance of tumor cells to that stratum intermedium and a high activity of alkaline phosphatase and adenosine triphosphate in both these cells.[4]

The tumor occurs most commonly in the middle age, and the mean age was 40 years.[5] There is no sex predilection.[4] A painless, slow growing swelling is the most common presenting sign.[2-4] About two-thirds of the cases were reported in the mandibular posterior region.[2,3,4,6-8] The present case was reported in a 42-year-old patient with a painless swelling in the mandibular premolar region. This concurs with earlier reports of common clinical findings of CEOT.

The radiographic features of the tumor may be unilocular or multilocular radiolucency.[2,3,6,8] In our case, it was multilocular. Radiopaque flecks of calcifications are usually seen within

the radiolucency.[5-9] This is described as "driven snow" appearance.[1,2,4] Our case also revealed similar radiopaque mass and driven snow appearance.

Approximately, 50% of the cases are associated with an unerupted tooth or odontome,[2,3,6] but was not so with our case.

Histopathologic feature usually shows islands and strands of polyhedral epithelial cells with nuclear pleomorphism, prominent nucleoli, and intercellular bridges.[4,6,7,9,10] One of the characteristic microscopic features of this tumor is the presence of amorphous, eosinophilic hyalinized, and acellular areas resembling amyloid within or adjacent to epithelial islands.[2,3,6,9-11] These epithelial islands and amyloid-like materials were seen in our case also. The presence of amyloid can be confirmed with Congo Red Stains.[1-3,5,6,9]

Many histologic, histochemical, immunohistochemical, and electron microscopic investigations were done to identify the nature of amyloid-like material in Pindborg tumor.[8,10-14] Yamaguchi *et al*. and Page *et al*. studied the histologic, histochemical, fluorescent, and ultrastructural study of CEOT and found that eosinophilic substance was not amyloid.[13,14] It is now identified that this material has a unique protein that has similarity with enamel proteins and is produced by this tumor cells.[13,14]

Another characteristic feature of this tumor is the presence of concentric ring of basophilic calcifications called Leisegang rings within the amyloid-like masses.[2,5,6] These were also evident in our case. These Leisegang ring calcifications were thought to be a form of dystrophic calcification and cemental masses.[2,3,6,15,16] They usually fuse to form large masses of calcification.[2,3]

On rare occasions, clear cells, langerhan cells, and myoepithelial cell may be observed in CEOT.[2,5,17] Sometimes, CEOT can occur along with adenomatoid odontogenic tumor.[2,18] On extremely rare occasions, CEOT can show aggressive growth and features of malignancy.[19,20]

The lesion was surgically excised, and reconstruction was done. The recurrence rate reported was 10–20%.[1-3] The patient was called for regular follow-up to monitor the prognosis. The follow-up of 1 year showed good healing of the site and no recurrence.

Conclusion

We have reported a rare case of CEOT or Pindborg tumor which was diagnosed and confirmed as CEOT by incisional and excisional biopsy. It is an extremely rare odontogenic tumor and included in differential diagnosis of all the odontogenic tumors.

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Conflicts of interest
There are no conflicts of interest

References

1. Rajendiran R, Sivapathasundaram B. Shafer's Textbook of Oral Pathology. 6th ed. India: Elsevier; 2009. p. 279-81.
2. Reichart P, Philpsen HP. Odontogenic Tumors and Allied Lesions. 1st ed. USA: Quintessence Publishing Co. Ltd.; 2004. p. 93-103.
3. Neville BW, Damm DD, Allen CM, Bouquat JE. Oral & Maxillofacial Pathology. 3rd ed. India: Saunders; 2009. p. 716-8.
4. Franklin CD, Pindborg JJ. The calcifying epithelial odontogenic tumor. A review and analysis of 113 cases. Oral Surg Oral Med Oral Pathol 1976;42:753-65.
5. Regezi JA, Scuibba JJ, Richard JC. Oral Pathology: Clinical Pathologic Correlation. 5th ed. USA: Saunders; 2008. p. 268-9.
6. Philipsen HP, Reichart PR. Calcifying epithelial odontogenic tumour: Biological profile based on 181 cases from the literature. Oral Oncol 2000;36:17-26.
7. Ai-Ru L, Zhen L, Jian S. Calcifying epithelial odontogenic tumors: A clinico-pathologic study of nine cases. J Oral Patho1 1982;11:399-406.
8. Maranda G, Gourgi M. Calcifying epithelial odontogenic tumor (Pindborg tumor). Review of the literature and case report. J Can Dent Assoc 1986;52:1009-12.
9. Robert ME, Diane S. Oral and Maxillofacial Pathology: A Rationale for Diagnosis and Treatment. 1st ed. USA: Quintessence Publishing Co. Ltd.; 2003. p. 660-3.
10. Chomette G, Auriol M, Guilbert F. Histoenzymological and ultrastructural study of a bifocal calcifying epithelial odontogenic tumor. Characteristics of epithelial cells and histogenesis of amyloid-like material. Virchows Arch A Pathol Anat Histopathol 1984;403:67-76.
11. Mori M, Makino M. Calcifying epithelial odontogenic tumor: Histochemical properties of homogeneous acellular substances in the tumor. J Oral Surg 1977;35:631-9.
12. Franklin CD, Martin MV, Clark A, Smith CJ, Hindle MO. An investigation into the origin and nature of 'amyloid' in a calcifying epithelial odontogenic tumour. J Oral Pathol 1981;10:417-29.
13. Yamaguchi A, Kokubu JM, Takagi M, Ishikawa G. Calcifying epithelial odontogenic tumor: Histochemical and electron microscopic observations on a case. Bull Tokyo Med Dent Univ 1980;27:129-35.
14. Page DL, Weiss SW, Eggleston JC. Ultrastructural study of amyloid material in the calcifying epithelial odontogenic tumor. Cancer 1975;36:1426-35.
15. el-Labban NG. Cementum-like material in a case of Pindborg tumor. J Oral Pathol Med 1990;19:166-9.
16. Slootweg PJ. Bone and cementum as stromal features in Pindborg tumor. J Oral Pathol Med 1991;20:93-5.
17. El-Labban NG, Lee KW, Kramer IR. The duality of the cell population in a calcifying epithelial odontogenic tumour (CEOT). Histopathology 1984;8:679-91.
18. Philipsen HP, Reichart PA. Adenomatoid odontogenic tumour: Facts and figures. Oral Oncol 1999;35:125-31.
19. Bouckaert MM, Raubenheimer EJ, Jacobs FJ. Calcifying epithelial odontogenic tumor with intracranial extension: Report of a case and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2000;90:656-62.
20. Basu MK, Matthews JB, Sear AJ, Browne RM. Calcifying epithelial odontogenic tumour: A case showing features of malignancy. J Oral Pathol 1984;13:310-9.