



## A RARE TUMOUR JUXTAPOSES HIGHLY INFECTIOUS SYSTEMIC DISEASE: CYSTIC VARIANT OF PINDBORG'S TUMOUR IN AN HBSAG-POSITIVE ADOLESCENT- A CASE REPORT

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**KEYWORDS :** Calcifying Epithelial Odontogenic Tumor, Pindborg Tumor, Cystic Variant, Mandible, Odontogenic Tumor, Liesegang Rings, HbsAg Positive

### INTRODUCTION

The cystic variant of CEOT is extremely rare, with only a limited number of cases reported in the literature. We present a rare case of cystic CEOT occurring in a 16-year-old male involving the posterior mandible.

### Case Report

A 16-year-old male presented to the Department of Oral and Maxillofacial Surgery with complaints of pain and swelling involving the right lower posterior jaw for one month. The swelling had gradually increased in size. The patient reported a past history of jaundice during infancy and chickenpox at approximately one and a half years of age. He was not receiving any regular medications.



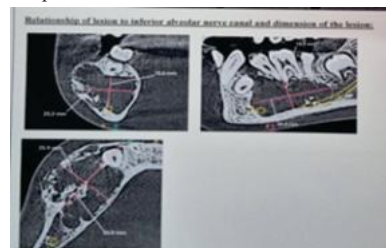
Extraoral examination revealed facial asymmetry approximately  $2.5 \times 2.5$  cm over the right mandibular body from the commissure of the lip to approximately 3 cm anterior to the angle of the mandible. The swelling was hard, fixed, and non-tender. Ipsilateral level IB lymph nodes measuring approximately  $1.5 \times 1.5$  cm and  $1 \times 1$  cm were palpable, firm, mobile, and tender.



Intraoral examination revealed vestibular obliteration extending from teeth 42 to 47. A hard swelling measuring approximately  $4 \times 2$  cm occupied the buccal vestibule in relation to teeth 42–46. The overlying mucosa appeared erythematous, and a superficial ulcer measuring approximately  $0.5 \times 0.5$  cm was present over the alveolar region adjacent to tooth 44.



Panoramic radiography demonstrated a mixed radiolucent-radiopaque lesion measuring approximately  $3 \times 3$  cm extending from the mesial aspect of tooth 43 to tooth 47. Tooth 44 was impacted and displaced with loss of the apical one-third of its root.



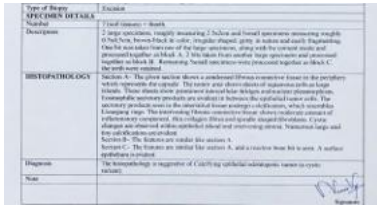
Cone-beam computed tomography revealed an ovoid lesion measuring approximately 36 mm involving the right posterior mandible. Inferior displacement of the inferior alveolar canal was noted. Resorption of the apices of teeth 45, 46, and 47 was observed. Tooth 44 showed an oblique orientation with crown directed toward the buccal cortex and root toward the lingual cortex, with root dilaceration.

Based on the clinical and radiographic findings, a provisional diagnosis of central giant cell lesion was considered, with other mixed odontogenic lesions included in the differential diagnosis. Routine hematological investigations were within normal limits. HIV serology was non-reactive, while hepatitis B surface antigen (HBsAg) tested positive.

The lesion was surgically excised under appropriate precautions



Histopathological examination demonstrated a fibrous connective tissue capsule enclosing sheets and islands of polygonal epithelial cells exhibiting prominent intercellular bridges and mild nuclear pleomorphism. Abundant eosinophilic extracellular amyloid-like material was present within the epithelial islands and stroma. Multiple concentric calcifications resembling Liesegang rings were identified throughout the lesion. The cystic lining exhibited features similar to the tumor islands, confirming a cystic architecture.



The histopathological diagnosis was calcifying epithelial odontogenic tumor (cystic variant).

## DISCUSSION

CEOT is an uncommon benign odontogenic epithelial neoplasm first described by Pindborg<sup>[1]</sup>. Approximately two-thirds of cases occur in the mandible, particularly in the posterior region, and nearly half are associated with impacted teeth<sup>[2]</sup>. The present case shares these characteristic features but differs significantly in patient age, as CEOT is rare during adolescence<sup>[2][5]</sup>.

The radiographic appearance of CEOT varies according to the amount of calcification. Early lesions are predominantly radiolucent, whereas mature lesions demonstrate mixed radiolucent-radiopaque patterns due to progressive calcification. The present lesion exhibited mixed radiopacity with cortical expansion, internal septations, impacted tooth association, root resorption, and inferior alveolar canal displacement, findings consistent with previously reported cases<sup>[5][6]</sup>.

Histopathological examination remains the gold standard for diagnosis<sup>[4][5]</sup>. The characteristic microscopic features include polygonal epithelial cells with distinct cell borders, intercellular bridges, extracellular amyloid-like deposits, and concentric calcifications (Liesegang rings)<sup>[4][7]</sup>. Congo red staining with apple-green birefringence under polarized light may demonstrate the amyloid material, although routine hematoxylin and eosin staining is usually sufficient when classical features are present<sup>[5][6]</sup>.

The differential diagnosis includes calcifying odontogenic cyst, adenomatoid odontogenic tumor, ameloblastoma, ameloblastic fibro-odontoma, odontoma, central giant cell granuloma, ossifying fibroma, and cemento-osseous dysplasia<sup>[5][6]</sup>. Careful correlation of clinical, radiographic, and histopathological findings is necessary to establish the diagnosis.

Conservative curettage has been associated with recurrence because CEOT lacks a true capsule and may infiltrate adjacent cancellous bone<sup>[2][5][8]</sup>. Therefore, complete surgical excision with adequate margins is generally recommended, especially for larger lesions<sup>[5][6]</sup>. Reported recurrence rates are approximately 10–15%, emphasizing the importance of long-term postoperative surveillance<sup>[2][8]</sup>. The cystic variant is exceedingly uncommon, and only isolated cases have been documented. Recognition of this unusual presentation expands the histopathological spectrum of CEOT and highlights the importance of considering this diagnosis even in young patients presenting with mixed radiolucent-radiopaque mandibular lesions.

HbsAg positivity makes this case much more of a rare occurrence. Even though HbsAg positivity does not cause the tumour, it complicates the surgical management of the patient. The patient requires strict medical precautions regarding Liver function status & viral load before planning and executing the procedure. Post surgically on a long term basis, special care to be given to the patient's liver function along with long term follow up of surgical site for possible recurrence in future.

## CONCLUSION

Calcifying epithelial odontogenic tumor is a rare odontogenic neoplasm, particularly in adolescents<sup>[2][5]</sup>. The cystic variant is even more uncommon and may present diagnostic challenges because of its overlapping clinical and radiographic features with other odontogenic lesions. Histopathological examination remains the definitive diagnostic modality<sup>[4][5]</sup>. Early diagnosis, complete surgical excision,

and long-term follow-up are essential for preventing recurrence and ensuring favorable clinical outcomes<sup>[5][6]</sup>. Special care to be given for systemic HbsAg positivity, taking into consideration long term care and follow up of liver function of the patient.

## REFERENCES

1. Pindborg JJ. A calcifying epithelial odontogenic tumor. *Cancer*. 1958;11(4):838–43.
2. Philipsen HP, Reichart PA. Calcifying epithelial odontogenic tumour: biological profile based on 181 cases from the literature. *Oral Oncol*. 2000;36(1):17–26.
3. Barnes L, Eveson JW, Reichart P, Sidransky D, editors. *WHO Classification of Tumours: Pathology and Genetics of Head and Neck Tumours*. Lyon: IARC Press; 2005.
4. Wright JM, Vered M. Update from the 4th Edition of the World Health Organization Classification of Head and Neck Tumours: Odontogenic and maxillofacial bone tumors. *Head Neck Pathol*. 2017;11(1):68–77.
5. Neville BW, Damm DD, Allen CM, Chi AC. *Oral and Maxillofacial Pathology*. 4th ed. St Louis: Elsevier; 2016.
6. Regezi JA, Sciubba JJ, Jordan RCK. *Oral Pathology: Clinical Pathologic Correlations*. 7th ed. St Louis: Elsevier; 2017.
7. Speight PM, Takata T. New tumour entities in the 4th edition of the World Health Organization classification of head and neck tumours: odontogenic and maxillofacial bone tumours. *Virchows Arch*. 2018;472(3):331–9.
8. Franklin CD, Pindborg JJ. The calcifying epithelial odontogenic tumour. A review and analysis of 113 cases. *Oral Surg Oral Med Oral Pathol*. 1976;42(6):753–65.