Open Access Library Journal
2026, Volume 13, e15533
ISSN Online: 2333-9721
ISSN Print: 2333-9705

Pindborg Tumor, A Rare Entity: Clinical Case Report

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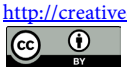
How to cite this paper: Zaher, B., Ouammou, W. and Bouzoubaa, S.M. (2026) Pindborg Tumor, A Rare Entity: Clinical Case Report. *Open Access Library Journal*, **13**: e15533.
https://doi.org/10.4236/oalib.1115533

Received: May 23, 2026
Accepted: June 21, 2026
Published: June 24, 2026



Abstract

Calcifying epithelial odontogenic tumor (CEOT), also called Pindborg tumor, presents a rare and extensive diversity in clinical, histopathological features, and biological behavior. Through this work, we expose a case of a 14-year-old male with a complaint of gradually increasing swelling on the left lower facial region for 18 months. On oral examination, a hard and fixed swelling, involving the left mandible, was noted extending from the mandibular second premolar to the second molar region. Radiographic examination showed a well-demarcated radiolucent lesion causing an expansion of the vestibular cortex. Surgical enucleation, along with curettage, was carried out under local anesthesia, and the histopathological examination confirmed the diagnosis of CEOT. The accurate final diagnosis needs to be established through a combination of clinical, radiographic, and histopathologic analysis. Diagnostic accuracy is essential to provide adequate treatment. However, regular follow-up over time allows us to monitor the stability of the case and prevent recurrence.

Subject Areas

Dentistry, Pathology

Keywords

Pindborg, Tumor, Rare, Case

1. Introduction

Calcifying epithelial odontogenic tumor (CEOT) is a rare odontogenic epithelial tumor. It was first described by the oral pathologist Pindborg, so CEOT is also called the Pindborg tumor. It accounts for <1% of all odontogenic tumors [1]. The eponym Pindborg tumor was first introduced to the literature in 1963 by Shafer to describe this remarkable and unique odontogenic tumor which typically con-

tains calcifying masses or homogeneous acellular material within the tumor epithelium and stroma [2]. The patient we are reporting is an exception and a rare case since it is a pediatric case, as the tumor occurs most commonly in middle age, and the mean age was 40 years. There is no sex predilection [3]. The origin of this tumor is controversial, and it is thought to be derived from the oral epithelium, reduced enamel epithelium, stratum intermedium, or dental lamina remnants. CEOT presents an extensive diversity in clinical, histopathological features, and biological behavior [4].

2. Cas Report

A 14-year-old male reported with a complaint of gradually increasing swelling with mild intermittent pain in the left lower facial region for 18 months. The medical history was noncontributory. On extraoral examination, a single, smooth, slightly tender, hard, and fixed swelling, involving the left mandible, was noted (**Figure 1(a)**, **Figure 1(b)**). Intraoral examination revealed the presence of a firm to hard oval-shaped tender swelling, extending from the mandibular left second premolar to second molar region, causing extensive buccal cortical plate expansion but no mobility of teeth. The overlying mucosa of the swelling was intact (**Figure 1(c)**).

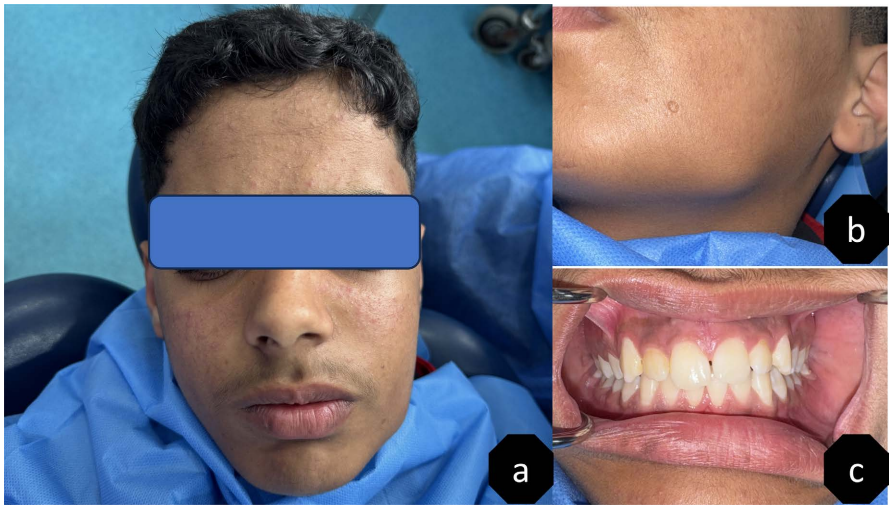


Figure 1. (a) (b) Extraoral photographs of the patient, showing swelling on the left side of the face. (c) An intraoral photograph showing lower left buccal vestibule obliteration.

The panoramic radiograph revealed a large, well-demarcated radiolucent lesion with a sclerotic border, involving the left mandible, extending from the second premolar to the second molar (**Figure 2**).

Axial, perpendicular-oblique, and frontal sections of the CBCT showed a radiolucent lesion measuring approximately 3.5 cm wide and 3 cm high. The lesion caused expansion of the vestibular cortex and was located vestibularly, distant from the teeth and the inferior alveolar nerve (**Figures 3(a)-(c)**).

As a diagnostic hypothesis, we considered an aneurysmal cyst, a dentigerous

cyst, or a keratocyst.

Surgical enucleation, along with curettage, was carried out under local anesthesia (Figures 4(a)-(c)).

The histopathological examination revealed an odontogenic epithelial proliferation arranged in sheets with trabeculae and islands of polygonal cells with abundant eosinophilic cytoplasm and well-defined cell boundaries. The stroma contained patches of amyloid-like eosinophilic extracellular deposits associated with

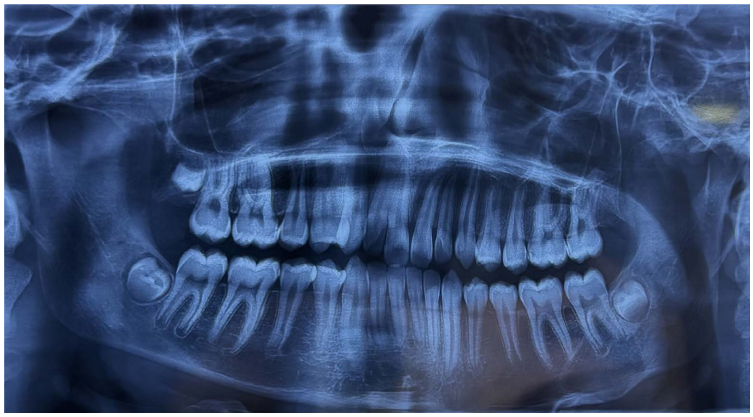


Figure 2. Panoramic radiograph showing a large, well-demarcated radiolucent lesion, involving the left mandible, extending from the second premolar to the second molar.

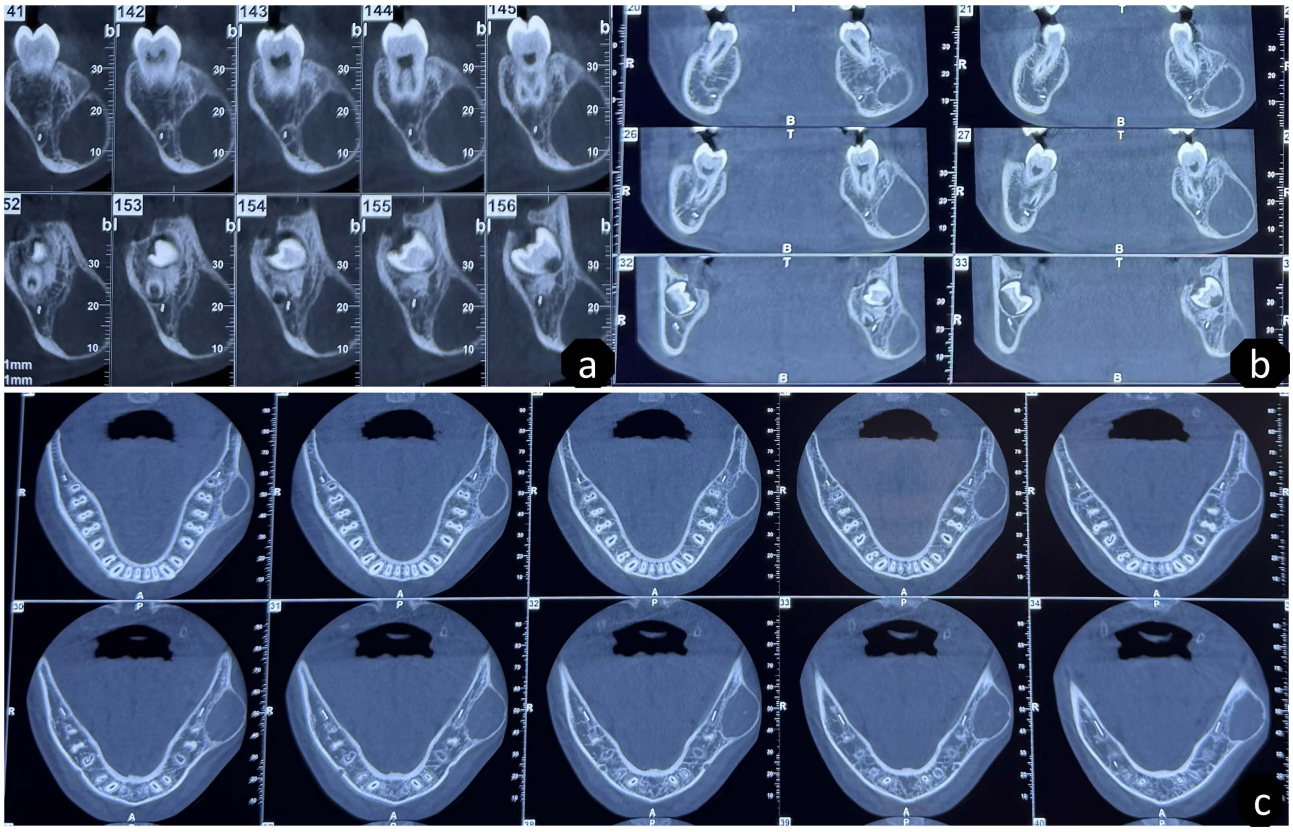


Figure 3. (a) Perpendicular-oblique view; (b) Frontal view; (c) Axial view of computed tomography shows a radiolucent lesion causing an expansion of the vestibular cortex.

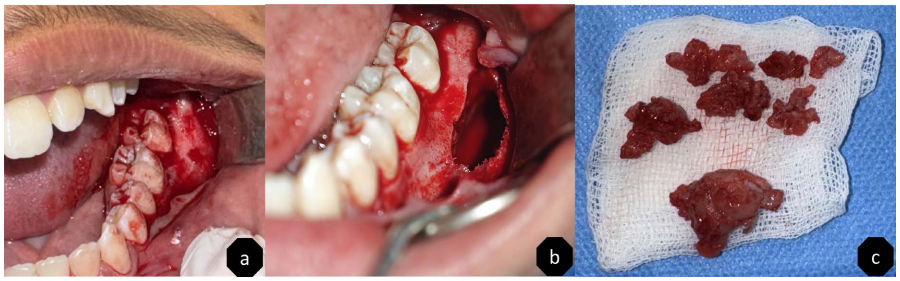


Figure 4. (a) (b) Surgical procedure; (c) Excised specimen.

concentric foci of calcification. Focal reactive multinucleated giant cells were also present, particularly in contact with the calcification foci (**Figure 5(a)**, **Figure 5(b)**).

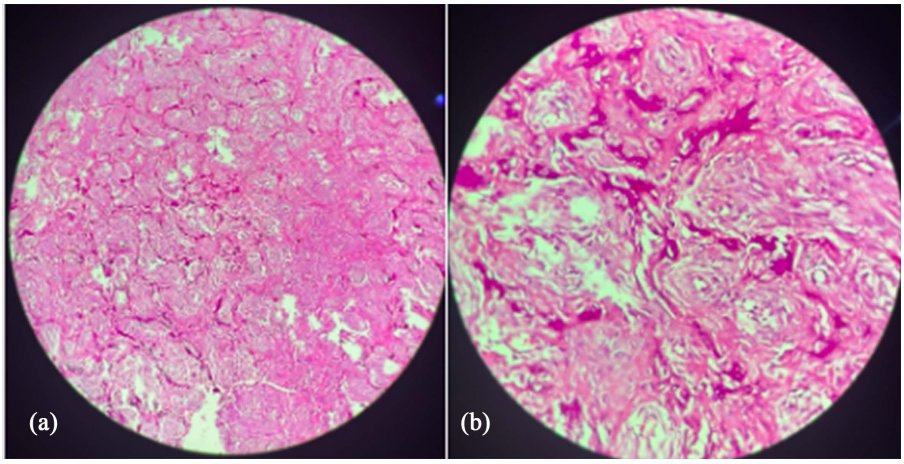


Figure 5. (a) (b) Histological sections show an odontogenic epithelial proliferation arranged in sheets, trabeculae, and islands of polygonal cells with abundant eosinophilic cytoplasm and well-defined cell boundaries. The nuclei are of variable size, sometimes large, without marked cytonuclear atypia or significant mitotic activity. The stroma contains extracellular eosinophilic deposits resembling amyloid, associated with concentric foci of calcification. Focal reactive multinucleated giant cells are noted, particularly in contact with the foci of calcification, without predominance or aggressiveness.

Based on all the above findings, the final diagnosis of intraosseous CEOT was made. The patient has been on regular follow-up for the last 3 months, and no recurrence has been reported.

3. Discussion

3.1. Etiology and Biological Mechanism

CEOT is a "locally invasive epithelial neoplasm characterized by the development of intra-epithelial structures, probably of an amyloid-like nature, which may be calcified and which may be liberated as the cell's breakdown [5].

CEOT usually appears between the second and sixth decades of life, but mainly occurs in the fourth decade, and has no gender predilection. It is most often located in the premolar-molar region of the mandible and associated mostly with

one or more impacted teeth [6].

CEOT may present as an intraosseous (central) or extraosseous (peripheral) tumor. The intraosseous tumor is the most common type, usually seen in the posterior mandible, and is more aggressive, accounting for more than 95% of the cases. Whereas the extraosseous tumor accounts for <6% and most commonly occurs on the anterior gingiva as a sessile mass capable of destroying the underlying bone [7].

Different clinical presentations of the tumor have distinguished histogenetic origins. The central tumors are said to be derived from the stratum intermedium of the enamel organ, whereas the extraosseous form arises from dental lamina epithelial rests in the gingival and/or basal cells of the gingival surface epithelium [8].

The initial consensus regarding the pathogenesis of CEOT was attributed to Pindborg in 1955. He stated that the CEOT was indeed of odontogenic origin, that is, reduced enamel organ-related, as the previous cases have been associated with unerupted teeth. However, according to Philipsen and Reichart, with the reports of central cases not presenting with an unerupted tooth and the gingival variants, other sources of origin were debated. The soft-tissue location of this tumor strongly suggests that these tumors may arise from rests of dental lamina or basal cells of the oral epithelium. After the disintegration of the dental lamina complex, numerous epithelial remnants (rests of Serres) persist in the jaw bones and supra-periosteally in the gingiva when odontogenesis is completed [9].

3.2. Clinical Aspect

Intraosseous CEOT may manifest as asymptomatic swelling and grow by infiltration, causing cortical plate expansion, tooth movement, and root resorption. Rarely, the tumor may be associated with paresthesia. CEOT, when located in the maxilla, may present with nasal stuffiness, epistaxis, and headache. Although CEOT is a benign neoplasm, its biologic behavior is variable, ranging from very mild to moderately invasive behavior.

3.3. Radiological Representation

The radiographic appearance of CEOT is variable and depends on the stage of development; either as a well-defined radiolucency, a mixed radiolucent-radiopaque mass, or a completely radio-opaque mass [7].

The intraosseous lesion presents as radiolucency. Later, as the lesion ages, calcium salts are deposited, and it becomes increasingly radio-opaque. It also simultaneously erodes bone and thus, the lesion is often mixed radiolucent/radiopaque, giving a characteristic "driven-snow" appearance on the radiograph. Further, the lesion may be unilocular or, more commonly, multilocular in appearance [9].

The peripheral variant of CEOT can display a range of radiographic features with regard to lesion size and bone pattern as compared to the intraosseous forms. It presents with no radiographic changes to a superficial erosive pattern, and then

to a radiolucency with scattered radiopaque foci [9].

3.4. Differential Diagnosis

The differential diagnosis of CEOT will depend on radiographic appearance. In case of radiolucent lesion-dentigerous cyst, odontogenic keratocyst, ameloblastoma, odontogenic myxoma, whereas in mixed radiolucent radiopaque lesion-Calcifying odontogenic cyst, adenomatoid odontogenic tumor, complex odontoma, ameloblastic fibro-odontoma, fibro-osseous lesions, osteoblastoma should be considered [10] [11].

3.5. Histology

The histologic pattern of CEOT is typical and well defined. The tumor consists of polyhedral cells arranged in masses, sheets, islands, cords, rows, or strands in a scanty connective tissue stroma. The cells are pleomorphic with well-defined borders, prominent nucleoli, and abundant finely granular cytoplasm filled with an eosinophilic "amyloid-like" material, which gradually becomes concentric calcified deposits, resembling psammoma bodies called the "Liesegang rings," which is considered pathognomonic for this tumor. The round-shaped eosinophilic amyloid material will stain positive for Congo red and will appear as an apple-green birefringence under a polarized microscope [12] [13].

3.6. Treatment

The treatment methods can range from simple enucleation or curettage to hemimandibulectomy or hemi-maxillectomy.

Small intrabony lesions may be treated with conservative tumor enucleation and curettage, followed by removal of a thin layer of normal bone adjacent to the tumor. Larger lesions may require resection of the tumor into the disease-free bony margin, as these tumors are more invasive and destructive [13].

Enucleation with a margin of normal tissue is usually recommended for mandibular lesions. CEOT of the maxilla should be treated more aggressively, as maxillary tumors grow more rapidly and are usually not well confined [7].

Treatment, however, should be individualized for each case. Our patient underwent enucleation, and no recurrence is reported in 3 months of follow-up.

Long-term follow-up, at least 5 years, is a must as there is a high risk of recurrence if the tumor was incompletely resected, especially with the clear cell variant, which is locally more aggressive [7].

The lesions treated with enucleation and curettage procedures show a high recurrence rate ranging from 15% to 30% after just 2 - 4 years. Therefore, CEOT is best treated with a resection using 1.0 - 1.5 cm margins in bone [14].

For our patient, we have planned follow-up sessions every 4 months to monitor healing through clinical examination and panoramic radiographs. Recurrence control can be established after one year of clinical and radiological monitoring.

4. Conclusion

CEOT is a rare odontogenic tumor and does not have a pathognomonic presentation. Thereby, the classic histopathologic pattern will always confirm the diagnosis. Treatment of CEOT involves the enucleation of smaller lesions and the resection of large ones. Recurrence prevention needs the patient's cooperation for frequent recall visits.

Conflicts of Interest

The authors declare no conflicts of interest.

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