CASE REPORT

Role of Polarizing Microscopy in Unveiling a Unique Cystic Variant of CEOT.

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Abstract

Context: Calcifying epithelial odontogenic tumor (CEOT) is a rare benign odontogenic neoplasm characterized by the presence of amyloid-like material and calcifications. The cystic variant of CEOT is exceedingly uncommon and may closely mimic other odontogenic cysts histopathologically, posing a diagnostic challenge. Recognition of its distinct microscopic and histochemical features is essential for accurate diagnosis and appropriate management.

Case Presentation: We report a unique case of a 76-year-old partially edentulous male presenting with a cystic lesion in the posterior mandible. Histopathological examination revealed a cystic epithelial lining containing amyloid-like eosinophilic material associated with dystrophic calcifications, suggestive of cystic CEOT. Polarizing microscopy demonstrated characteristic birefringence of the amyloid deposits, thereby confirming the diagnosis and helping distinguish the lesion from histopathological mimics such as odontogenic keratocyst (OKC) and residual cyst.

Management and Prognosis: The lesion was managed surgically, and the excised specimen was subjected to detailed histopathological evaluation. Identification of amyloid deposition and calcifications, along with confirmation by polarizing microscopy, played a crucial role in establishing the diagnosis. The patient was placed under regular postoperative follow-up to monitor for recurrence, considering the rare nature of the lesion.

Conclusion: This case highlights the importance of comprehensive assessment of clinicopathologic features and adjunctive polarizing microscopic examination in diagnosing rare cystic variants of CEOT. Awareness of its overlapping features with common odontogenic cysts is essential to avoid diagnostic pitfalls. Accurate recognition of this entity contributes to appropriate treatment planning and improved understanding of the biological spectrum of CEOT.

Keywords: Amyloid, Calcifying Epithelial Odontogenic Tumor, Odontogenic Cysts, Polarization Microscopy, Pindborg Tumor.

Introduction

Odontogenic tumors are a heterogeneous group of lesions arising from odontogenic epithelium, ectomesenchyme, or both. Among these, the calcifying epithelial odontogenic tumor (CEOT) is a rare benign neoplasm first described by Pindborg in 1958, hence also called the Pindborg tumor.[1] CEOT accounts for less than 1% of all odontogenic tumors and is most frequently reported in the posterior mandible.[2] It usually affects patients between the ages of 30 and 50 years, with no strong sex predilection. Histologically, CEOT is composed of sheets and nests of polyhedral epithelial cells with prominent intercellular bridges, nuclear pleomorphism, and deposition of an amyloid-like substance. The latter often undergoes calcification, producing concentric liesegang ring calcifications that are pathognomonic.[3] The cystic variant of CEOT was first described by Franklin and Pindborg in 1976.[4] This variant is extremely rare, with only a handful of reported cases in the literature. Unlike the conventional

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solid form, the cystic variant presents as a cystic cavity lined by CEOT-like epithelium containing amyloid material and calcifications. Most CEOTs are associated with impacted teeth or arise in dentate regions of the jaws.[5] The occurrence of CEOT in partially edentulous jaws is exceptional, making the present case noteworthy. Additionally, the sequential occurrence of a residual cyst followed by CEOT in the same patient raises diagnostic and biological interest. This report describes a rare case of cystic CEOT in a 76-year-old male and highlights its clinical, radiographic, and histopathological features, with emphasis on diagnostic challenges and pathogenesis.

Case History

A 76-year-old partially edentulous male initially presented with a swelling in the upper left posterior alveolar region. Clinical examination revealed a soft-tissue swelling involving the edentulous maxillary alveolar ridge, and radiographic evaluation demonstrated a radiolucent lesion. Surgical enucleation was performed, and histopathological examination confirmed the diagnosis of a residual cyst. During subsequent follow-up evaluation, the patient reported a swelling in the lower right posterior jaw region (45–47), which had been present for approximately two weeks. Intraoral examination revealed a brownish-red, ovoid swelling of soft consistency with well-circumscribed margins and associated yellowish discharge. Based on the clinical and radiographic findings, odontogenic keratocyst (OKC) and residual cyst were considered as provisional differential diagnoses. Radiographic examination demonstrated a well-defined oval radiolucency extending from the 45 to 47 region. (Figure 1)

The lesion was surgically excised and submitted for histopathological analysis. Microscopic examination revealed a cystic lining composed of 5–10 layers of polyhedral epithelial cells with prominent intercellular bridges (Figures 2 and 3). Areas of dystrophic calcification (Figure 4) were identified within the epithelial lining, along with deposits of amyloid-like material. Congo red staining demonstrated minimal apple green to yellow to orange red birefringence under polarized light (Figure 5), confirming the presence of amyloid. Collectively, these features were diagnostic of the cystic variant of calcifying epithelial odontogenic tumor (CEOT), a rare presentation, particularly in an edentulous jaw.

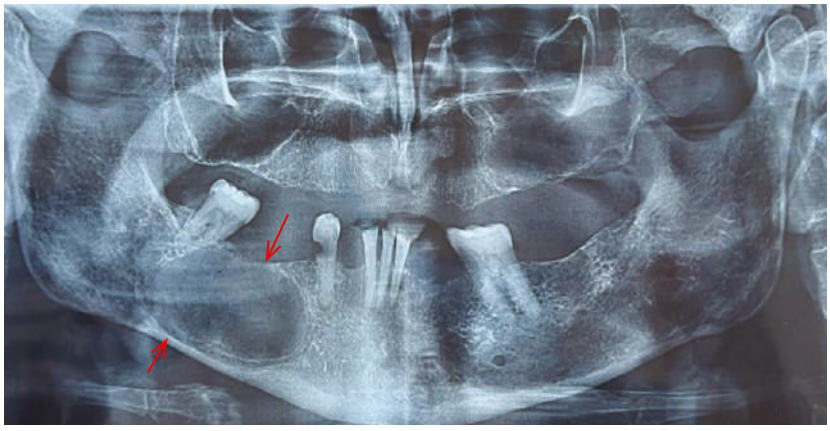

Figure 1: Orthopantomogram (OPG) showing a well-defined unilocular radiolucent lesion involving the right posterior mandibular region extending from 45 to 47

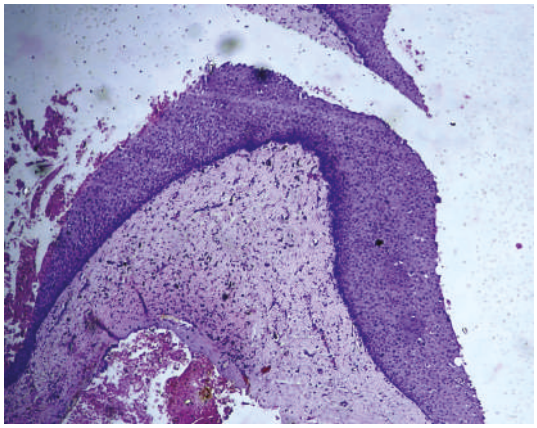

Figure 2: Cystic lining having 5-10 layers of polyhedral epithelial cells.

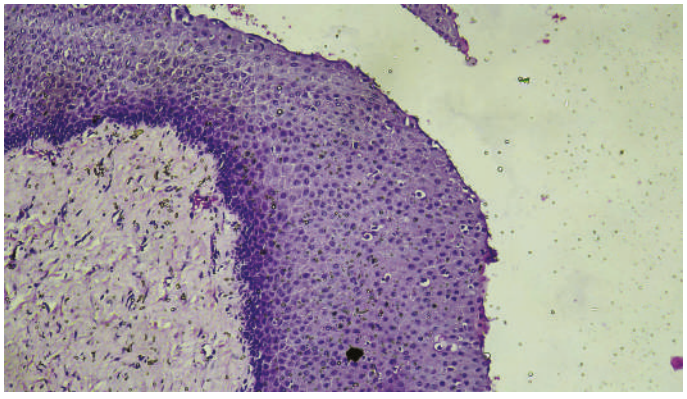

Figure 3: Polyhedral epithelial cells in the cystic lining displaying prominent intercellular bridges

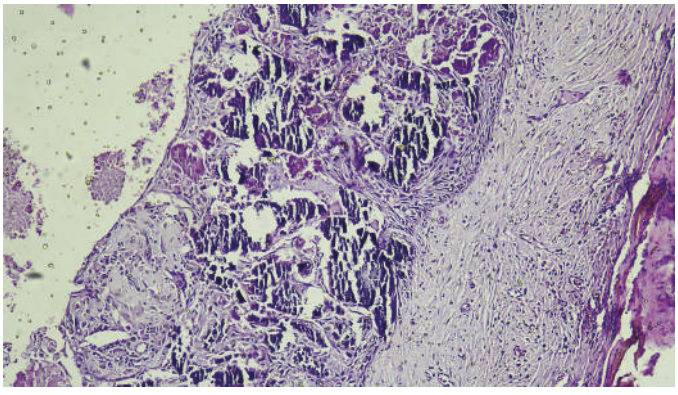

Figure 4: Dystrophic calcifications within the cystic epithelial lining

Discussion

Rarity of CEOT and Its Variants

Calcifying epithelial odontogenic tumor (CEOT) is an uncommon benign odontogenic neoplasm, accounting for less than 1% of all odontogenic tumors.[2] The cystic variant is exceedingly rare, with only a limited number of cases reported in the literature.[4] Most documented cases occur in dentate jaws and are frequently associated with unerupted or impacted teeth. Therefore, the occurrence of cystic CEOT in a partially edentulous jaw, as seen in the present case, represents a highly unusual presentation.

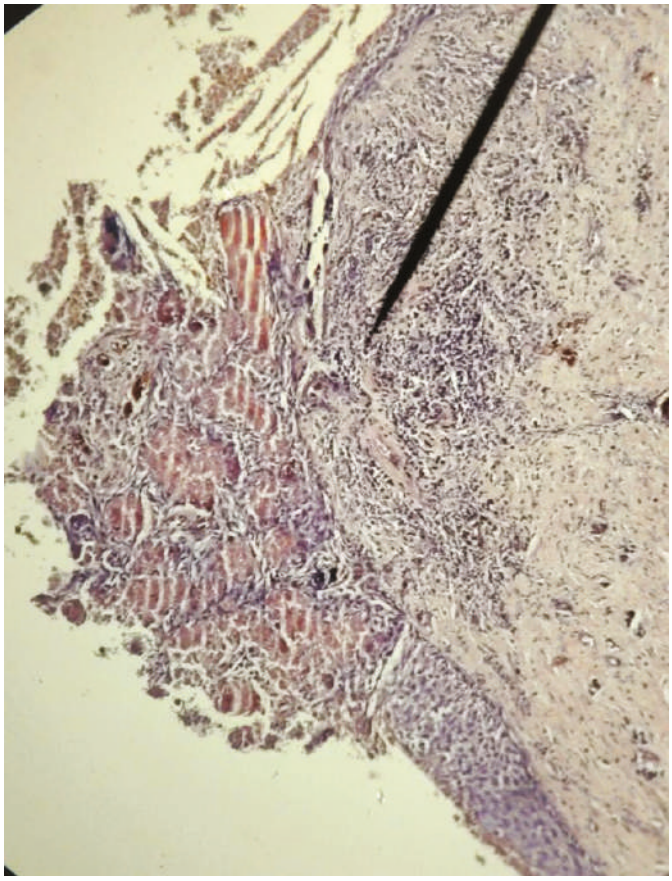

Figure 5: Amyloid deposits in the epithelium showing minimal apple birefringence under polarising microscopy.

Clinical and Radiological Features:

Clinically, CEOT usually presents as a slow-growing and painless swelling, although secondary infection may occasionally result in pain or discharge, as observed in the present patient. Radiographically, CEOT commonly appears as a unilocular or multilocular radiolucency with varying degrees of internal radiopacity depending on the extent of calcification[5]. In this case, the lesion appeared as a well-circumscribed unilocular radiolucency, closely simulating common odontogenic cysts and tumors.

The clinical differential diagnosis included odontogenic keratocyst (OKC), residual cyst, calcifying odontogenic cyst (COC), and adenomatoid odontogenic tumor (AOT). OKC was considered because of the well-defined radiolucency and cystic presentation.[6] Residual cyst was included due to the patient's partially edentulous status and the occurrence of the lesion within an extraction site region.[7] COC was considered because it may present as a cystic radiolucent lesion with calcifications.[8] AOT was also considered because it can appear as a well-circumscribed radiolucent lesion, often mimicking other odontogenic lesions radiographically.[9]

Histopathological Distinction:

Histopathological examination played a critical role in distinguishing cystic CEOT from its mimics. Odontogenic keratocyst (OKC) is typically lined by parakeratinized stratified squamous epithelium with a corrugated surface and palisaded basal cell layer,[6] features that were absent in the present case. Residual cysts are generally lined by non-keratinized stratified squamous epithelium and do not demonstrate amyloid deposition or calcifications.[7] Calcifying odontogenic cyst (COC) characteristically exhibits ghost cells and calcifications but lacks amyloid-like material.[8] Similarly, adenomatoid odontogenic tumor (AOT) demonstrates pathognomonic duct-like epithelial structures.[9]

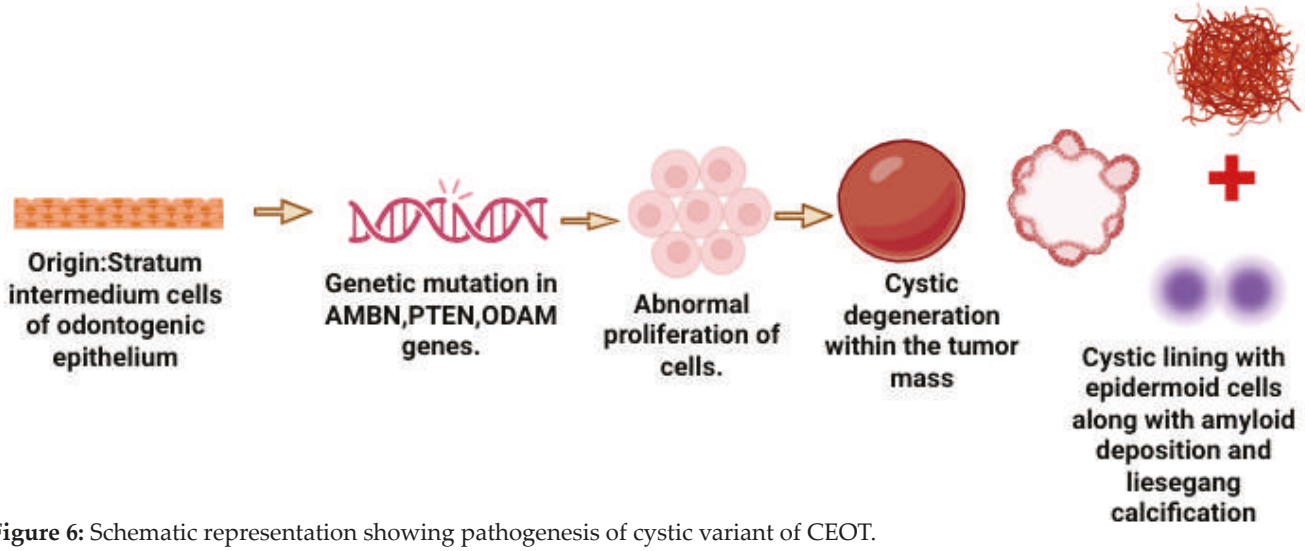

Figure 6: Schematic representation showing pathogenesis of cystic variant of CEOT.

Role of Polarizing Microscopy:

Polarizing microscopy serves as a valuable adjunctive diagnostic tool in the evaluation of calcifying epithelial odontogenic tumor (CEOT), particularly in cystic variants that may closely mimic other odontogenic cysts and tumors histologically. Although the identification of amyloid-like material on routine hematoxylin and eosin sections may raise suspicion of CEOT, definitive confirmation can be achieved by Congo red staining followed by examination under polarized light. The demonstration of characteristic apple-green birefringence confirms the amyloid nature of the extracellular deposits and provides strong diagnostic evidence in favor of CEOT. In the present case, polarizing microscopy played a pivotal role in establishing the diagnosis by confirming amyloid deposition and excluding histopathological mimics such as odontogenic keratocyst, residual cyst, calcifying odontogenic cyst, and adenomatoid odontogenic tumor, all of which lack Congo red–positive amyloid exhibiting apple-green birefringence. Thus, polarizing microscopy was instrumental in achieving an accurate diagnosis of this rare cystic variant of CEOT.[10]

In the present case, microscopic examination revealed a cystic lining composed of polyhedral epithelial cells with prominent intercellular bridges, associated with dystrophic calcifications and amyloid-like deposits. Congo red staining demonstrated minimal apple green birefringence and ranged from green to, yellow and orange to red under polarized light, confirming the presence of amyloid.[10] These collective features established the diagnosis of the cystic variant of CEOT.

Pathogenesis

The exact pathogenesis of calcifying epithelial odontogenic tumor (CEOT) remains incompletely understood, and two principal pathogenetic theories have been proposed for the cystic variant. The first theory suggests neoplastic transformation arising within the lining epithelium of a pre-existing odontogenic cyst. This hypothesis is supported by the cystic architecture of the lesion and the presence of neoplastic epithelial lining exhibiting characteristic CEOT features, including polyhedral epithelial cells, amyloid deposition, and calcifications. Such transformation may occur in pre-existing odontogenic cysts through proliferation and neoplastic alteration of odontogenic epithelial remnants.[4]

The second theory proposes that the cystic variant represents secondary cystic degeneration occurring within a conventional solid CEOT. According to this concept, degenerative changes within epithelial islands of a pre-existing solid tumor may lead to central necrosis and cystic cavitation, ultimately producing a predominantly cystic lesion.[4] The absence of a significant solid tumoral component in some reported cases, however, has made the exact mechanism controversial. In the present case, the lesion predominantly demonstrated a cystic lining with characteristic CEOT histopathological features, favoring a true cystic variant rather than extensive degeneration of a solid tumor.

CEOT is believed to originate from stratum intermedium cells of the enamel organ or remnants of the dental lamina.[9] The stratum intermedium is metabolically active during enamel formation and expresses alkaline phosphatase and adenosine triphosphatase, which may explain the metabolic activity observed in CEOT cells.[11]

A defining feature of CEOT is the presence of amyloid-like material. Immunohistochemical studies have demonstrated that this material is related to enamel matrix proteins, particularly amelogenin and odontogenic ameloblast-associated protein (ODAM).[12] Congo red positivity with apple-green birefringence under polarized light confirms the amyloid nature of these deposits. The amyloid material may subsequently act as a nidus for Liesegang ring formation and dystrophic calcification, which are characteristic microscopic findings in CEOT.[3]

Recent molecular studies suggest that odontogenic tumors, including CEOT, may involve alterations in ameloblastin, amelogenin, and keratin expression; however, the precise molecular pathways responsible for tumor initiation and cystic transformation remain under investigation.[13]

The cystic variant of CEOT is thought to arise from neoplastic transformation of odontogenic epithelial remnants entrapped within a pre-existing odontogenic cyst, where amyloid-like protein deposition and subsequent calcification occur within the cystic lining which is explained in a diagrammatic representation below. (Figure 6)

Thus, the unique histopathology of CEOT — epithelial nests, amyloid, and calcifications — can be explained by its odontogenic epithelial origin and aberrant protein deposition, which also help distinguish it from odontogenic cysts.

Biological Behavior and Management

CEOT is generally regarded as a benign but locally aggressive tumor with recurrence rates ranging from 10–15% after conservative excision.[14] The cystic variant is less well studied but is believed to have a relatively indolent course. Complete surgical excision with regular follow-up remains the recommended treatment approach.

Significance of This Case

Unique Presentation: Unlike typical CEOTs associated with impacted teeth, this case arose in a partiailly edentulous mandible.

Sequential Pathology: The occurrence of a residual cyst in the maxilla followed by CEOT in the mandible within a short interval suggests either a predisposition to odontogenic cystic lesions or coincidental sequential pathology.

Diagnostic Challenge: Clinically and radiographically, the lesion strongly resembled common odontogenic cysts, highlighting the importance of clinicopathologic factors in establishing the correct diagnosis.

Conclusion

This case emphasizes the importance of considering rare odontogenic tumors such as cystic CEOT in the differential diagnosis of jaw cysts, even in partially edentulous patients. Sequential cystic pathology in the same patient further underscores the need for careful follow-up. Histopathology with special stains such as Congo red remains indispensable for accurate diagnosis, and polarizing microscopy serves as a crucial adjunct by demonstrating the birefringence of amyloid

deposits, thereby unmasking this rare entity and differentiating it from histological mimics.

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