

RADIOGRAPHIC ENIGMA: A CASE REPORT OF CLEAR CELL ODONTOGENIC NEOPLASM IN THE ANTERIOR MANDIBLE DIAGNOSED BY CONE BEAM COMPUTED TOMOGRAPHY

Enigma radiográfico: Informe de un caso de carcinoma odontogénico de células claras en la mandíbula anterior diagnosticada mediante Tomografía Computarizada de Haz Cónico

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ABSTRACT

Introduction: Clear cell odontogenic carcinoma (CCOC) is a rare and aggressive odontogenic tumour, seldom involving the anterior mandible.

Case Report: This report presents an atypical case in a 38-year-old female, emphasizing the diagnostic challenges, the role of CBCT in detecting aggressive features, and the importance of considering CCOC in the differential diagnosis of anterior mandibular lesions. The patient presented with gingival swelling, and cone-beam computed tomography revealed a diffuse, irregular, expansile lesion with labial cortical perforation, thinning of the lingual cortex, and hyperdense foci. Periosteal bone formation was also noted anteriorly.

Results: These atypical features initially suggested alternative diagnoses, but histopathological evaluation confirmed CCOC.

Conclusions: This case reinforces the diagnostic challenges of CCOC and demonstrates the utility of radiographic evaluation for identifying aggressive features of rare mandibular tumours.

Keywords: Case report; Mandibular neoplasms; Odontogenic tumors; Cone-beam computed tomography; Diagnosis, differential; Mandible.

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RESUMEN

Introducción: El carcinoma odontogénico de células claras (CCOC) es un tumor odontogénico raro y agresivo, que rara vez afecta la mandíbula anterior.

Reporte de Caso: Este informe presenta un caso atípico en una mujer de 38 años, destacando los desafíos diagnósticos, el papel de la tomografía computarizada de haz cónico (CBCT) en la detección de características agresivas y la importancia de considerar el CCOC en el diagnóstico diferencial de las lesiones mandibulares anteriores. La paciente presentó inflamación gingival, y la CBCT reveló una lesión difusa, irregular y expansiva con perforación de la cortical labial, adelgazamiento de la cortical lingual y focos hiperdensos. También se observó formación ósea perióstica en la zona anterior.

Resultados: Estas características atípicas sugirieron inicialmente diagnósticos alternativos, pero la evaluación histopatológica confirmó el CCOC.

Conclusión: Este caso refuerza los desafíos diagnósticos del CCOC y demuestra la utilidad de la evaluación radiográfica para identificar características agresivas de tumores mandibulares raros.

Palabras clave: *Informe de caso; Neoplasias mandibulares; Tumores odontogénicos; Tomografía computarizada de haz cónico; Diagnóstico diferencial; Mandíbula.*

INTRODUCTION

Clear cell odontogenic carcinoma (CCOC) is a rare malignant odontogenic epithelial tumour, recognized in the World Health Organization (WHO) classification for its aggressive behaviour, high recurrence rate, and metastatic potential.¹ Since its first description in 1985, approximately 120 cases have been reported worldwide, mostly as isolated case reports, highlighting its rarity.²⁻⁴ CCOC occurs predominantly in the mandible, often in the posterior mandible followed by the anterior region, with a female predilection and peak incidence in the fifth to sixth decades of life.^{2,5} Radiographically, it typically presents as a unilocular or multilocular radiolucency with well- or ill-defined margins.⁶ Histopathology demonstrates a spec-

trum from low- to high-grade morphology, with recurrence rates reported at 41% and metastasis at 31%.⁷ Here, we present an unusual radiographic manifestation of CCOC in the anterior mandible of a 38-year-old female, a pattern not previously documented, to expand the spectrum of known imaging findings. Given its rarity, aggressive biological behaviour, and potential for recurrence and metastasis, each additional case contributes valuable information to the literature. The present report is important because it documents an atypical radiographic presentation of CCOC in a younger patient, thereby contributing to the understanding of its imaging features and reminding clinicians to consider this entity in the differential diagnosis of anterior mandibular lesions

CASE REPORT

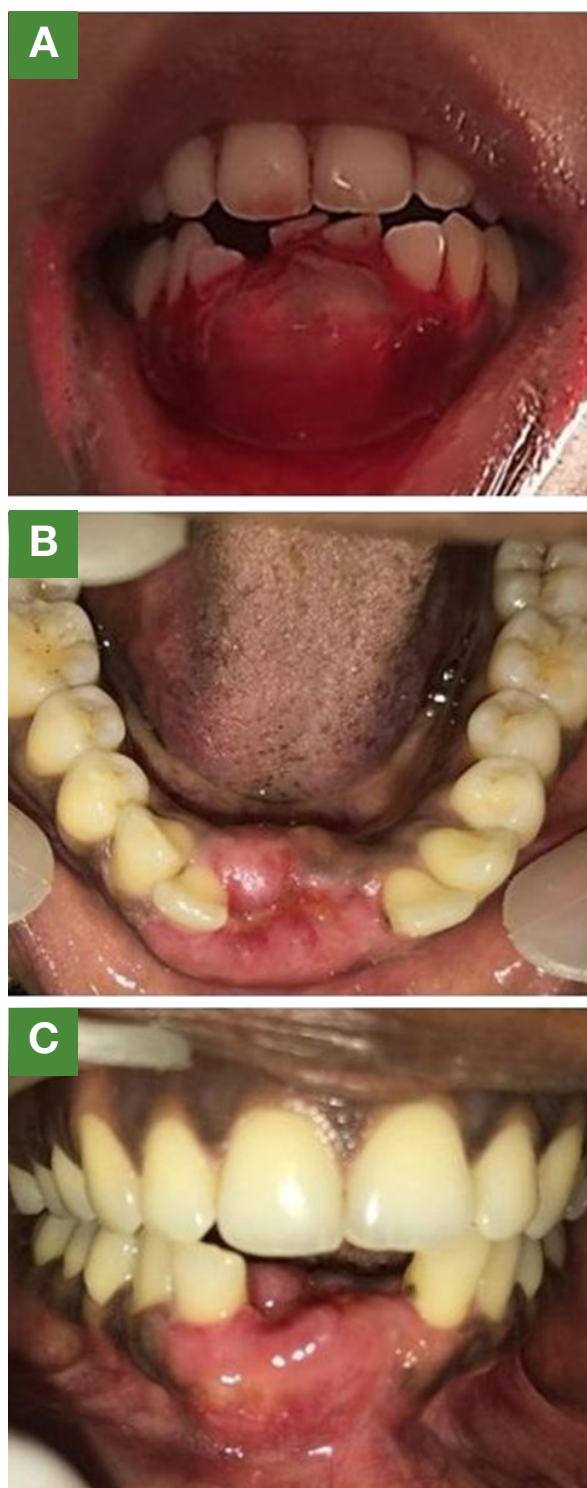
A 38-year-old female presented with swelling and bleeding in the lower anterior gingiva for one week, accompanied by pain during brushing. Her dental history revealed a root canal treatment in the same region 20 years earlier, after which she remained symptom-free until the current episode. Initial dental management involved extraction of the mobile teeth and excision of the gingival swelling under local anaesthesia, which was uneventful. She was otherwise healthy, with no significant medical, family, social, or occupational history, and reported no systemic illness or long-term medication use.

On intraoral examination, a diffuse lobulated swelling extended from tooth 32 to 42. The overlying mucosa appeared normal except for an ulcerated area with greyish-black pigmentation on the alveolar ridge. The lesion was firm, sessile, mildly tender, and irregular, without pus discharge or active bleeding. No regional lymphadenopathy was detected (Figure 1).

Cone-beam computed tomography (CBCT) of the mandible revealed an expansile lesion involving the anterior mandible, extending from tooth 32 to 42. The periphery is irregular with ill-defined borders with labial cortical perforation and thinning of the lingual cortex. A periosteal reaction was evident along the anterior aspect. The internal structure is predominantly radiolucent, interspersed with hyperdense foci. The lesion caused localized periodontal bone loss in relation to adjacent teeth, with

Figure 1

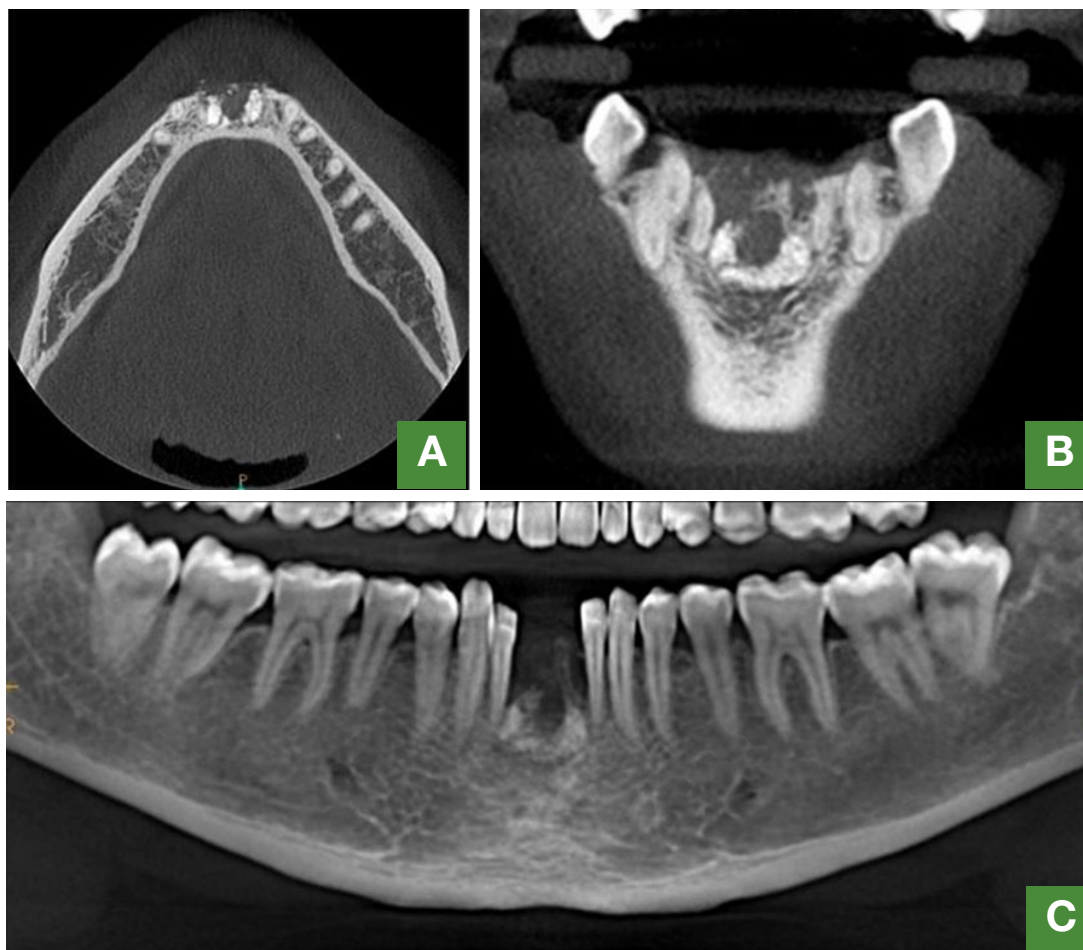
Clinical presentation of the lesion before surgical excision



A: View of bleeding and swelling from the patient's first visit prior to surgical excision. **B:** Occlusal view revealing an ill-defined swelling extending from teeth 32-42 with ulceration and greyish-black pigmentation of the alveolar ridge. **C:** Frontal view demonstrating the lesion's clinical extent and associated mucosal changes.

Figure 2

CBCT images of the anterior mandible demonstrate a diffuse expansile lesion (teeth 32-42) with ill-defined borders, labial cortical perforation, lingual cortical thinning, hyperdense foci greater than enamel density, and periosteal reaction along the anterior surface



A: Axial slice. **B:** Coronal slice. **C:** Panoramic reconstruction.

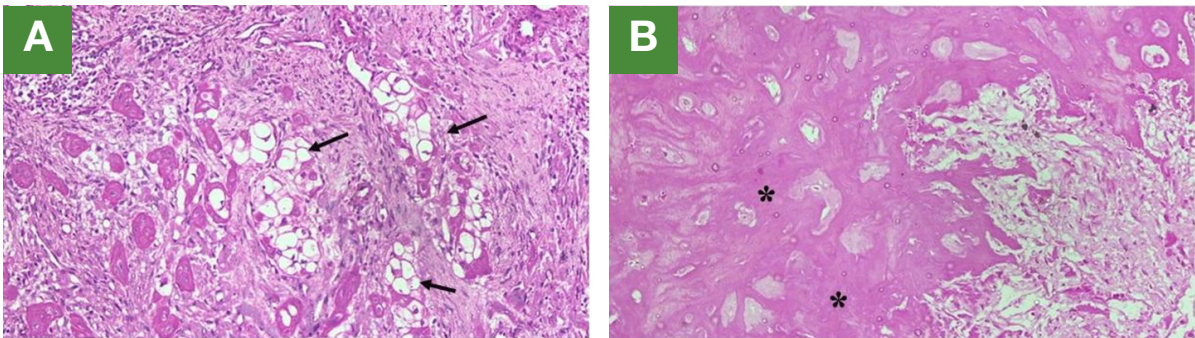
widening of the periodontal ligament space and partial loss of lamina dura. No displacement or root resorption of teeth was observed, (Figure 2). Based on these imaging features, the differential diagnoses included peripheral ossifying fibroma, foreign body reaction, chronic osteomyelitis, and malignancy.

Histopathological examination of the incisional biopsy showed epithelial islands within a vascular stroma, with some cells exhibiting clear cytoplasm. Hyalinized connective tissue containing

dentinoid-like material was also observed (Figure 3). Immunohistochemistry demonstrated CK19 and CK7 positivity, membranous β -catenin positivity, MUC4 negativity, and a low Ki-67 index (1–2%) (Not shown). These findings confirmed the diagnosis of clear cell odontogenic carcinoma (CCOC).

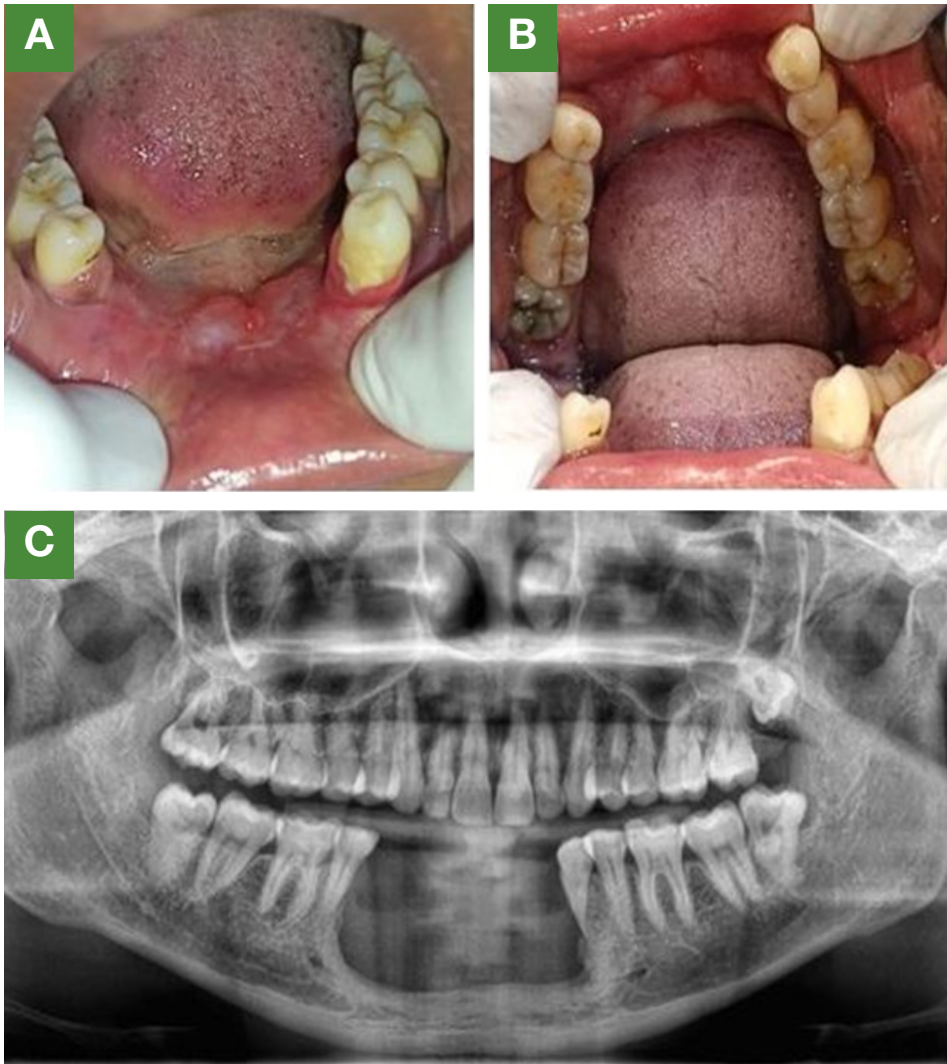
The patient underwent wide excision with marginal resection of the anterior mandible. At the six-month follow-up, an orthopantomogram revealed a stable bony defect at the symphysis with

Figure 3
Histopathological features of the tumor showing epithelial islands



A: Large eosinophilic and clear cells (arrows) within a connective tissue stroma. **B:** Hyalinized stroma and focal dentinoid-like deposits (asterisk).

Figure 4
Post-operative views of the lesional site



A: Frontal view. **B:** Occlusal view. **C:** Follow-up orthopantomogram showing a bony defect in the anterior mandible with preservation of the inferior mandibular border.

preservation of the inferior mandibular border and no evidence of recurrence (Figure 4).

DISCUSSION

Clear cell odontogenic carcinoma was first described by Hansen in 1985 as a clear-cell odontogenic tumour. In 2005, 2017 and 2022, the WHO classified CCOC as a malignant odontogenic tumour due to its aggressive growth, local recurrence, and metastatic potential.¹ Historically, “clear cell ameloblastoma” and “clear cell odontogenic tumour” were used interchangeably.⁸

CCOC typically presents as a painless, slowly progressive swelling, often for about four years before diagnosis. This case involved mild pain and swelling initially mistaken for a pulpal issue. Age of occurrence ranges from 17–89 years, peaking in the sixth decade, with a female predominance (male-to-female ratio 1:1.8).⁹ While the anterior mandible is a relatively common site, this case is reported because of its unusual radiographic characteristics.²

Diagnosis can be challenging due to non-specific clinical and radiological signs. Recurrence after years may reflect lesion progression and delayed detection. Radiographically, CCOC is usually radiolucent with poorly defined margins; 66.3% of cases show poorly defined lesions, and 25% well-defined ones.^{2,10} Additional features include cortical perforations, root resorption, and occasional soft-tissue invasion.^{11,12} This case displayed a rare mixed internal structure, with hyperdense-hypodense areas.

Clear cells support but do not confirm CCOC diagnosis, as they appear in various odontogenic and non-odontogenic tumours. Due to atypical radiographic features and clinical history, CCOC was initially excluded from the differential diagnosis, highlighting

the role of histopathology and immunohistochemistry.

Clear cell odontogenic carcinoma (CCOC) typically shows clear cells within epithelial islands and may contain hyalinized or dentinoid-like stroma. Immunohistochemical features include positivity for CK8, CK13, CK14, CK18, CK19, and EMA, and negativity for vimentin, S-100, desmin, SMA, HMB-45, CD31, CD45, and GFAP, with generally low Ki-67 and p53 expression. Molecular studies often reveal EWSR1-ATF1 translocations, aiding diagnostic precision. In our case, the tumor demonstrated epithelial islands with clear cells and dentinoid-like material.

Immunohistochemistry showed CK19 and CK7 positivity, membranous β -catenin positivity, MUC4 negativity, and a low Ki-67 index (1–2%), consistent with the low proliferative activity expected in CCOC and confirming the diagnosis. These findings highlight the importance of correlating histology, immunohistochemistry (IHC), and molecular features to accurately distinguish CCOC from other clear cell-rich odontogenic or metastatic tumors.^{2,13}

Literature shows recurrence rates of 86.7% after curettage and 29.8% after resection.¹⁴ This patient showed no recurrence after 6 months post-surgery.

Limitations

A limitation of this report is the unavailability of IHC microphotographs, as the analysis was conducted externally. However, the detailed IHC results, when correlated with the histopathological features, were sufficient to establish the diagnosis of CCOC.

CONCLUSIONS

This case highlights an uncommon presentation of clear cell odontogenic carcinoma in a female in her late 30s, involving the anterior

mandible with an atypical mixed radiolucent-radiopaque pattern. Such variation can obscure early recognition, reinforcing the role of CBCT and immunohistochemistry in accurate diagnosis. Clinicians should maintain a high index of suspicion and ensure wide excision

with prolonged follow-up given the tumour's recurrence and metastatic potential. Future collaborative studies are needed to better characterize long-term outcomes in this rare malignancy.

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CONFLICT OF INTERESTS

The authors declare no conflict of interest.

ETHICS APPROVAL

Written informed consent was obtained from the patient for publication of this case report and accompanying clinical and radiographic images.

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AUTHORS' CONTRIBUTIONS

Prarthana Sudeep: Concept development, study design, definition of intellectual content, literature search, data acquisition, data analysis, manuscript preparation, manuscript editing, manuscript review, and served as guarantor of the work.

Aravind Meena Shanmugham: Concept development, study design, definition of intellectual content, literature search, data acquisition, data analysis, manuscript preparation, manuscript editing, manuscript review, and served as guarantor of the work.

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