

Clear Cell Odontogenic Carcinoma of the Mandible: An Unusual Case Diagnostic in Ho, Ghana

Magalys López Cuba^{1,2,3}, Richard Ampoh⁴, Fernando Acevedo Almaguer^{1,4}, Israel Bedzina^{3*}, Felix Kwame Kpor³, Eric Gyan¹, Cecelia Smith-Togobo⁵ and Emmanuel Akomanin Asiamah⁵

¹Department of Pathology, University of Health and Allied Sciences, Ho, Ghana

²Department of Pathology, Ho Teaching Hospital, Ho, Ghana

³Reinbee Health and Wellness Centre, Ho, Ghana

⁴Department of Surgery, Ho Teaching Hospital, Ho, Ghana

⁵Department of Medical Laboratory Science, University of Health and Allied Sciences, Ho, Ghana

Corresponding author: Israel Bedzina, Reinbee Health and Wellness Centre, Ho, Ghana, E-mail: 2017ibedzina@uhas.edu.gh

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Abstract

Clear Cell Odontogenic Carcinoma (CCOC) is a rare intraosseous tumor of the jaw. Currently, it is a low-grade malignant tumor according to the recent reclassification by the World Health Organization, capable of frequent recurrences and loco-regional and distant metastases. Histologically the tumors consist primarily of large sheets and islands of uniform vacuolated and clear cells without evidence of amyloid deposition, calcification, or glandular differentiation. Till date, a little over 100 cases have been reported in the literature. The case a 47-year-old male patient presented, with a tumor mass extending from the canine to the molar region on the right inferior maxillary without regional lymph node involvement or apparent metastasis, with a biopsy diagnosis of clear cell odontogenic carcinoma, the patient was treated by partial resection of the anterior mandible, believed to be the first case ever from a rural community in Ghana.

Keywords: Clear cell odontogenic carcinoma; Clear cell ameloblastoma; Clear cell tumors; Odontogenic tumors

Introduction

Clear Cell Odontogenic Carcinoma (CCOC) is an extremely rare lesion that usually appears in the anterior part of the lower jaw. The WHO classed CCOC as a low-grade malignant tumor in 2005, describing it as having a locally aggressive and destructive nature with local recurrence, regional lymph node metastases and seldom distant metastasis [1]. It was first described in 1985 and since then, several but few case reports have been published [2-4].

Its incidence is higher in females between the fourth and seventh decade of life. The most typical symptom is painless

swelling, followed by discomfort, teeth loss and paresthesia. Local symptoms are frequently limited to gingival/periodontal problems and edema, with or without pain [5-7].

Due to its unique location in the mandible, the sporadic presence of odontogenic epithelium mixed with the clear cell component, the potential for tall columnar cells resembling ameloblasts and the infrequent presence of dentinoid structures, several authors who study histogenesis postulate that it has an odontogenic origin [8].

After more than 30 years after the first description in 1985 by Hansen et al., [3] just a little over 100 cases have reported in literature and much is now known about the presentation, radiological findings, histological and immunohistochemistry characteristics of CCOC.

Case Presentation

History

A 47-year-old male, no significant past medical, surgical history, admitted in Maxillofacial Surgery Service at the Ho Teaching Hospital in August of 2021 with a history of 3 years progressive lower right jaw swelling. This followed an occasion when the patient accidentally hit his lower right jaw on a metal. There is moderate pain 3/12 prior to presentation. Pain becomes worse whenever patients lie on right side of the face. At the time of his presentation of him in the consultation, an increase in volume was observed towards the right side of the chin (**Figure 1**); Intraorally: Tooth irregularities and deformity, with increase in volume and expansion of the internal and external cortices on the right side of the body of the mandible, in the area corresponding to the teeth: 31 to 47. The intraoral examination revealed an alteration of the attached gingiva of the region of the lower right mandibular area with nodular swelling, hard in consistency. The underlying alveolar mucosa

was normal. The involved teeth were sound, positive at cold sensitivity test and without mobility. No swollen cervical lymph nodes were found, and the remainder of the physical examination was within normal limits, (**Figure 1A**).

The radiographs revealed an osteolytic lesion in the region of 47-31, multilocular radiolucency extending from mesial roots of 47-31 with some fleck of opacity; truncation of roots of 46,45 and 44, (**Figure 1B**).

Based on the clinical and radiographic findings the doctor proceeds to excisional biopsy, and finding buccolingual expansion of jaw and loose teeth in involved segment. The excised tumor was sent for histopathological examination.

On macroscopic examination, the tumor mass was of 9 cm in maximum diameter, with infiltration the maxillary bone. The bone measuring 12 cm × 4 cm × 1.5 cm, the tumor measuring 9 cm × 6 cm × 5 cm located at the 1 cm bone margin, encapsulated and complete excision seen. The tumor is located at 1 cm of the mucosal margin and the muscular tissue measuring 1 cm × 4 cm × 2 cm. Cut surface of the tumor white in color, solid in consistence, hard, with calcification seen. No cystic area seen **Figure 1C and 1D**.

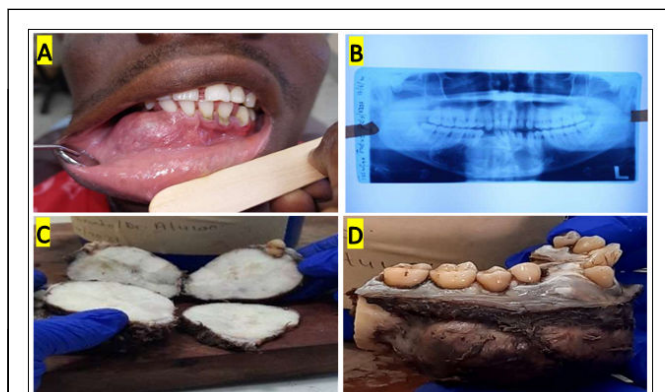


Figure 1: A: Physical appearance of patient with mass on the right lower mandibular; B: The radiograph reveals an increase multilocular radiolucency image in right mandibular; C and D: Completely excised nodular mass measuring 9 cm × 6 cm × 5 cm in dimensions, hard in consistency, good excisional margins. Tumor growth was inside and outside of the mandibula. Cut surface of the tumor showing a white in color, with focal calcification area, no cystic area seen, no necrosis and no hemorrhage seen.

Histological Findings: Shows malignant transformation composed by a neoplastic proliferation of cells of odontogenic origin, with a biphasic pattern of growth, epidermoid and large cells with clear cytoplasm arranged in nests or cords lack focal palisading of basal cells, ameloblastic-like cells seen. Low cellular pleomorphism, few atypical mitosis, stromal invasion seen, calcification and metaplastic bone seen in area. The margins look free of tumor. No vascular invasion seen. Squamous epithelium free of tumor. Focal area of chronic inflammation on the peripheral area. Bone margins free of tumor seen. The photomicrograph of the diagnosis of clear cell odontogenic carcinoma is shown in **Figure 2** below.

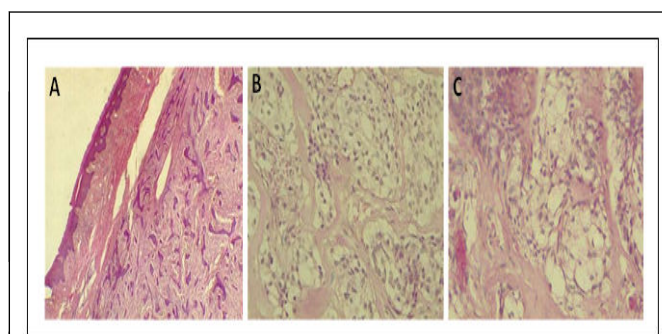


Figure 2: A: Shows subepithelial neoplastic proliferation of odontogenic cells, composed predominantly by epidermoid cells Haematoxylin & Eosin (H&E, 10 ×); B and C: Large cells with clear cytoplasm arranged in nests or cords with low mitotic activity, low cellular pleomorphism, no vascular invasion (H&E, 40 ×).

Results and Discussion

Odontogenic tumors are uncommon jaw tumors that mostly contain clear cell components. The first report of very malignant and aggressive clear cell odontogenic tumors was made by Hansen et al., [3]. The majority of patients present with tooth irregularities, painless swellings, or swellings that grew slowly over months and years. These tumors appear radiographically as radiolucent lesions with wavy edges and, in most instances, root resorption. Squamous cell carcinoma, small salivary gland carcinomas, and metastatic malignancies to the jaws are among the differential diagnoses based on the radiography picture [4].

CCOC is identified by the proliferation of neoplastic epithelial cells with clear cytoplasm arranged in islands and strands. Three kinds of cells can be found in CCOC: Basaloid to polygonal clear cells, basaloid to polygonal pale eosinophilic cells and columnar cells with ameloblast-like differentiation.

Depending on the proportion of these cells in the tumor, three different subtypes can be distinguished: (1) Monophasic: Formed almost entirely of clear cells with well-defined borders and centrally located nuclei; (2) Biphasic: Is characterized by oval and linear nests of large cells intermixed with smaller islands of smaller polygonal cells with eosinophilic cytoplasm (3) Ameloblastic: Characterized by columnar cells with ameloblastic differentiation at the periphery of islands [9,10].

Biphasic, monophasic and ameloblastomatous patterns are the three histopathological types of CCOCs. The biphasic pattern, which includes nests of cells with transparent cytoplasm interspersed with cells with eosinophilic cytoplasm, is the most prevalent [4,6]. Our case shows a biphasic pattern of growth, epidermoid and clear cells arranged in nests or cords lack focal palisading of basal cells, ameloblastic-like cells.

Due to their high glycogen content, the transparent cells exhibit Periodic Acid-Schiff (PAS) and diastase labile material staining. Immunohistochemistry reveals that clear cells are positive for the cytokeratin's CK8, CK13, CK14, CK18, and CK19 as well as for Epithelial Membrane Antigen (EMA) [6]. However, immunohistochemical analysis can be non-decisive.

In the majority of cases, surgery was required. A radical approach with excision in a wide margin is advised due to the high incidence of local and regional metastases and recurrences. The preferred course of treatment for CCOC is a wide-margin surgical resection [6,7].

Patient was administered Amoxil+Clavulanic Acid 1.2 g (G), Clindamycin 300 mg, Dexamethasone 4 mg, Paracetamol 1000 mg/100 ml post-surgery and was discharged satisfactorily to go home 3 days post-surgical procedure and improved recovery.

After resection, the CCOC reports a 34% recurrence rate. This recurrence rate plus the potential for metastasis to local lymph nodes and places like the lung and distant bone suggest that the current patient will need to be monitored for a considerable amount of time [11].

Conclusion

Initially, it was thought that clear cell odontogenic tumor was a benign, though locally invasive, tumor. Later, in few cases, it showed localized aggressive growth, frequent recurrences and sporadic metastasis, finally considered as a low-grade malignant and very rare tumor.

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