Hybrid clear cell odontogenic carcinoma and ameloblastic carcinoma-report of a case

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Abstract

Ameloblastic carcinoma (AC) produces extensive local destruction, perforation of the cortical plate, extension into surrounding soft tissues, numerous recurrent lesions, and metastasis, usually to cervical lymph nodes. Clear cell odontogenic carcinoma (CCOC) which was previously designated clear cell odontogenic tumor also exhibits an aggressive biologic behavior and a tendency to metastasize to distant locations. Both lesions are rare. We report an odontogenic carcinoma with a dual histomorphologic feature of CCOC and AC coexisting in a single lesion. To the best of our knowledge, this is the first report of its kind in the literature.

Key words: Clear cell; Ameloblastic carcinoma; Odontogenic carcinoma; Histopathology

Introduction

Odontogenic carcinomas are malignancies arising from an odontogenic epithelium. These tumors are rare. According to the World Health Organization (WHO) classification 1992, odontogenic carcinomas include malignant ameloblastomas, primary intraosseous carcinomas, malignant transformation of odontogenic cysts and malignant variants of other odontogenic tumours[1].

Clear cell odontogenic tumor[1] CCOT is a benign but locally invasive neoplasm of odontogenic epithelium characterized by sheets and islands of uniform, vacuolated and clear cells. Subsequent reports in the literature[2,3] indicated that the tumor has the potential to metastasize and current opinion is that it should be designated as clear cell odontogenic carcinoma (CCOC). Clear cell ameloblastoma (CCAM) is histologically characterized by an ameloblastomatous component intermixed with an extensive clear cell component[4]. Braunshtein et al[5], reviewing 27 cases of CCOC and 8 cases of clear cell ameloblastoma from the literature reported that both lesions should be considered low-grade malignancies and could well represent a clinicopathologic continuum of a single disease entity rather than 2 separate lesions.
Ameloblastic carcinoma (AC)[6] on the other hand refers to any ameloblastoma with histologic features of malignancy either in the primary or recurrent tumour regardless of whether it has metastasized or not. It may appear de novo or originate from a pre-existing ameloblastoma or odontogenic cyst[7].

These tumors are very aggressive with extensive local destruction, perforation of the cortical plate, extension into surrounding soft tissues, numerous recurrent lesions, and metastasis, usually to cervical lymph nodes.
Earlier reports in the literature are essentially isolated cases of either clear cell odontogenic carcinoma or ameloblastic carcinoma. We report an odontogenic carcinoma with a dual histomorphologic feature of CCOC and AC coexisting in a single lesion. To the best of our knowledge, this is the first report of its kind in the literature.

Case Report

A 34-year-old male Nigerian presented with a recurrent mandibular swelling of 2 months duration (Figure 1). He had had four previous surgical operations at another centre based on a histopathological diagnosis of ameloblastoma. The swelling extended from the region of the lower right second premolar to the lower left second molar. Plain radiographs and orthopantomogram (Figure 2) revealed a multilocular radioluscent lesion extending from the lower second right molar to the lower third left molar. Hematological and biochemical parameters were within normal limits. An incisional biopsy was taken under Local Anaesthesia (LA) and sent for histopathological examination.

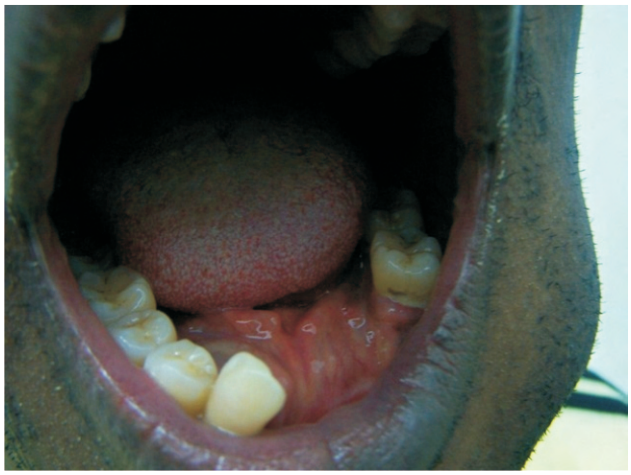

Figure 1- Clinical picture of the hybrid clear cell odontogenic carcinoma and ameloblastic carcinoma showing bucco-lingual expansion of the mandible.

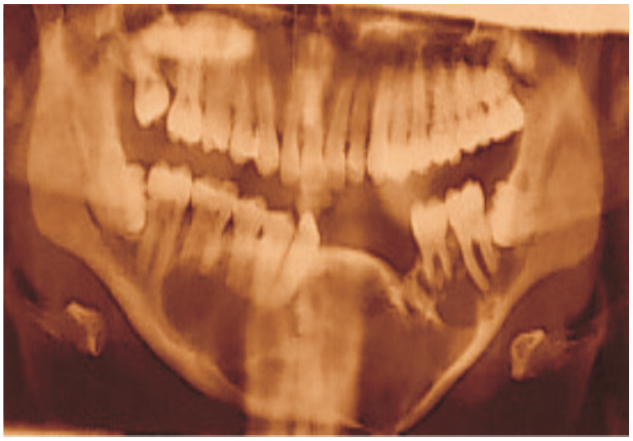

Figure 2- Orthopantomogram of the same lesion showing a multilocular radioluscency extending from the lower second right molar to the lower third left molar.

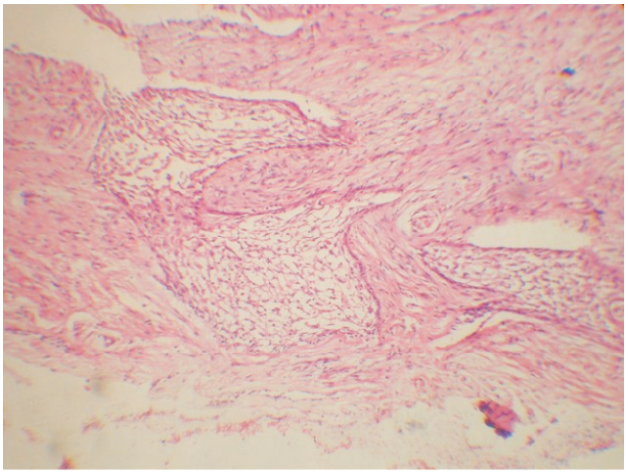

Figure 3 - Photomicrogragh of the lesion showing islands of neoplastic odontogenic epithelium made up of cuboidal or polyhedral cells exhibiting clear cytoplasm [H&E X100].

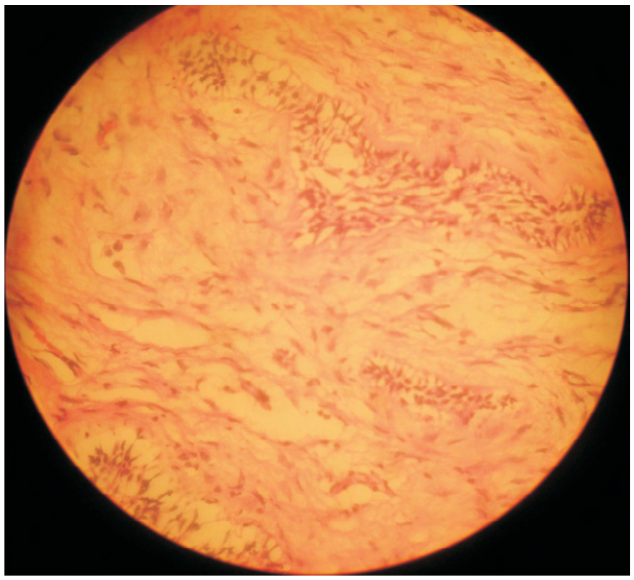

Figure 4 - Photomicrograph showing the isomorphic nature and well-defined outlines of the clear cells (H&E X400).

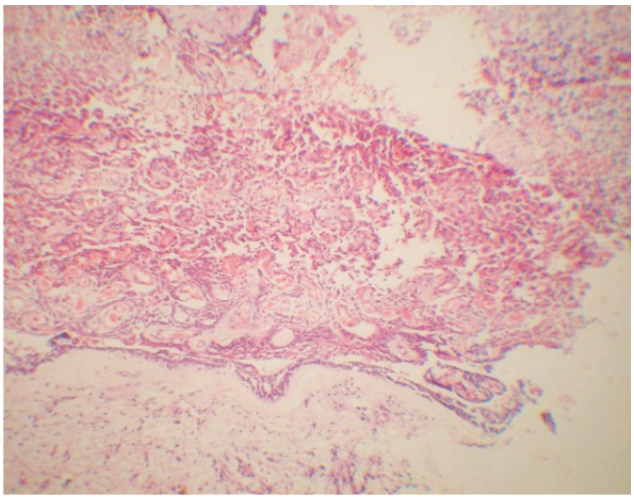

Figure 5 - Photomicrograph of the lesion showing peripheral, palisading ameloblastoma-like columner cells and central basaloid cells showing cellular and nuclear pleomorphism and hyperchromatism [H&E X40].

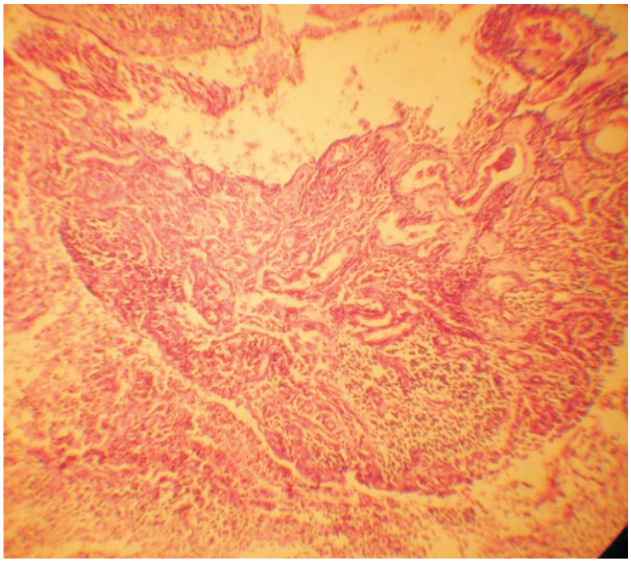

Figure 6 - Photomicrograph of the same lesion showing ameloblastic carcinoma exhibiting absence of the classic histologic features of ameloblastoma[H&E X80].

The surgical specimens were fixed in 10% neutral buffered formalin for 12 hours and paraffin embedded. Five-micrometer-thick sections were cut and stained with H&E, Mucicarmine and Periodic acid-Schiff (PAS). The histological sections showed pieces of soft tissue composed of moderately cellular and collagenized fibrous connective tissue within which are scattered sheets and islands of neoplastic odontogenic epithelium made up of cuboidal or polyhedral cells with centrally placed round nuclei and clear cytoplasm. Majority of the cells were isomorphic with well-defined outlines (Figure 3 & 4). In other areas the neoplastic islands are seen exhibiting peripheral, palisading ameloblastoma-like columner cells and central basaloid cells showing cellular and nuclear pleomorphism and hyperchromatism (Figure 5). Other areas show loss of this classic histologic architecture of ameloblastoma (Figure 6). Mitotic figures are also seen.

The tumor cells were negative for mucicarmine but PAS stain for glycogen was positive. A diagnosis of Odontogenic carcinoma exhibiting histologic patterns of clear cell odontogenic carcinoma and ameloblastic carcinoma was made.

Discussion

In the 1992 WHO classification of odontogenic tumors[1], clear cell tumor was classified as benign but a locally invasive tumor. However, high rate of recurrence, local and distant metasasis and tumor related deaths have lead to its reclassification as clear cell odontogenic carcinoma[(8)].

CCOC is a rare odontogenic tumor with female predilection and peak age incidence in the 5th to 7th decades of life[(5, 9)]. These are inconsistent with the demographic characteristics of our patient but the location of the tumor in the anterior mandible is consistent with reports in the literature[(2,10)].

Three different histomorphologic patterns of CCOC have been described[(11)]. The commonest is a biphasic tumor characterized by oval and linear nests of clear cells intermixed with smaller islands of polygonal cells with eosinophylic cytoplasm. Occationally these two cell-types co-exist in a tumor nest yielding a "glomeruloid" appearance. The second variant is represented by islands that show only the clear cell phenotype whereas the third and least common variant is comprised of clear cell nests with a tendency for ameloblastoid palisading around the periphery.

The present report is typical of this second variety. Special staining for mucin is recommended to rule out mucoepidermoid carcinoma. Furthermore, CCOC can be distinguished from the clear cell variant of calcifying epithelial odontogenic tumor because it lacks the characteristic calcification and amyloid deposition[(9)].

Contrary to CCOC, ameloblastic carcinoma tends to occur in the posterior segments of the jaws. There is no gender predilection but it occurs more in the mandible than the maxilla. Histologically, it presents as clusters of neoplastic epithelium within a collagenous stroma surrounded by polarized cells enclosing basaloid cells. In addition, it exhibits nuclear hyperchromatism, pleomorphism and focal areas of necrosis[(2)].

Of most head and neck tumors, clear cell tumors present a singular challenge to the pathologist since the classic morphological features of malignant neoplasia exemplified by cytological atypia are frequently absent in malignant clear cell variants, thereby excluding reliance on this histopathological hallmark for the establishment of a diagnosis[(12)].

The rare coexistence of dual histomorphologic patterns of clear cell odontogenic carcinoma and ameloblastic carcinoma in the present case may be an indication of these separate malignant entities occurring together in the same lesion. It may also be a singular lesion differentiating to exhibit another histomorphologic pattern.

The aggressive and destructive behavior of both clear cell odontogenic carcinoma and ameloblastic carcinoma has important clinical significance. It is therefore recommended that a lesion with this dual histologic presentation be treated with wide surgical resection leaving at least 1 cm of tumor free margin.

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