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COMPUTER GUIDED RESECTION AND RECONSTRUCTION OF A HYBRID ODONTOGENIC TUMOR OF THE ANTERIOR MANDIBLE: A CASE REPORT

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ABSTRACT

Purpose: Hybrid odontogenic tumors are considered to be rare differentiation forms of certain tumors. This study dealt with a case of rare hybrid odontogenic tumor involving a combination of the aggressive clear cell odontogenic carcinoma (CCOC) and the benign granular ameloblastoma (GA) and its computer guided resection and reconstruction.

Patients and methods: A 79 years old male, was referred to the oral and maxillofacial surgery department at the Faculty of Dentistry, Cairo University with a progressively facial swelling at the anterior mandible with palpable and tender submandibular lymph nodes (LN). He had a difficulty in breathing and inability to wear his removable denture. An intra-oral ulceration over the tumor was evident. Radiographic findings revealed a well-defined, multilocular lesion of 60 x 56 mm of size. Surgical resection of the lesion with safety free margins using computer guided 3D printed seating device used as an osteotomy guide with a stabilizing arm to prevent disarticulation of the condyles and collapse of the ramus of both sides. Immediate reconstruction plate and fibula vascularized flap were used.

Results: The excised lesion was sent to the pathology lab for microscopic examination. The specimen revealed a dominating clear cell carcinoma with minor granular ameloblastic appearance. The final diagnosis reached was a Hybrid odontogenic tumor.

Conclusion: Our study uncovered the presence of a rare hybrid tumor of the CCOC and granular ameloblastoma. Wide resection with safe margin is the only documented line of treatment due to its highly aggressive malignant nature with subsequent composite fibula free flap reconstruction.

The use of computer guided 3D printed devices is always beneficial. In this case, it was used as an osteotomy guide and to hold the mandible intra-operatively to prevent its collapse during tumor removal and before mandibular fixation.

KEYWORDS: Rare Hybrid tumor - Clear cell tumor - Granular ameloblastoma

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INTRODUCTION

Hybrid odontogenic tumors are rare conditions that may occur in the oral and maxillofacial region. Most of them do not fall under the standard World Health Organization (WHO) classification of odontogenic tumors. They usually occur sporadically and rarely.

Some authors regarded these so called hybrid tumors as simple anomalous histo- or morpho-differentitation process of certain tumors [(1)].

In this study, a rare hybrid odontogenic tumor was dealt with. It involved a combination of the aggressive clear cell odontogenic carcinoma (CCOC) and the benign granular ameloblastoma (GA).

Granular ameloblastoma is an infrequent benign odontogenic tumor which exhibits the presence of granular cells with an epithelial origin. Recent ultrastructural studies implied that the cytoplasmic granules most likely represent lysosomal aggregates. These granular cells in an ameloblastoma appeared to be of some prognostic significance in terms of the suggested treatment [(2)].

Clear cell odontogenic carcinoma (CCOC) is a rare intraosseous carcinoma of the jaw first discovered in 1985 by ***Hansen et al*** [(3)]. It was originally regarded as clear cell odontogenic tumor or clear cell ameloblastoma.

CCOC was considered to be a malignant tumor of odontogenic origin in the WHO classification of 2005 due to its aggressive tendency with local recurrence, regional lymph node metastasis, and distant metastasis [(4-6)].

CCOC occurs predominantly in the 5th to 7th decades usually in the mandibles of females. Painless swelling is the initial common symptom, while, subsequent pain, teeth loosening, and paresthesia might follow [(7)].

Only 8 true cases have been reported in the English literatures to date without our present report[(7-14)].

In this study, we reported a case of hybrid tumor of a dominating clear cell carcinoma with granular ameloblastoma. We provided an additional case to the rare existing literature.

In comparison with the previous studies, our case was a rare large case, extending from the right canine area to the left angle across the midline. Simultaneous reconstruction with a micro-vascular fibula free flap was also rare among previous cases. The present study aimed to report a rare CCOC case of a large lesion with free flap reconstruction compared to the previous literature.

Case Presentation

A 79 years old male, was referred to the oral and maxillofacial surgery department at the Faculty of Dentistry, Cairo University with chief complaint of progressively facial swelling at the anterior mandible. He stated that there was a recent difficulty in breathing and inability to wear his removable denture. He stated that the lesion started to expand his face one year ago. The past medical history revealed that the patient was asthmatic, a controlled diabetic and controlled hypertensive patient. An ethical clearance with a consent taken from the patient.

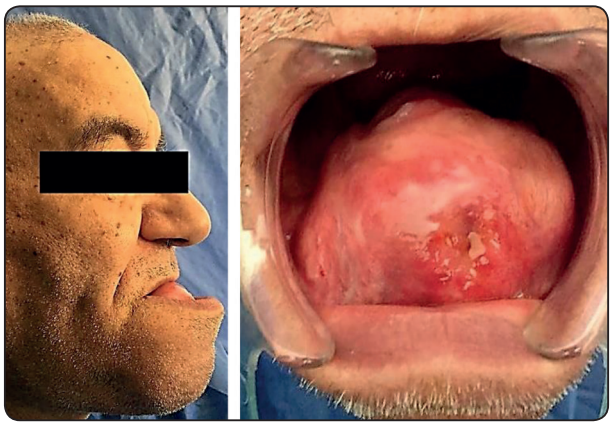

Fig. (1) Photographs showing the preoperative clinical picture of the lesion.

Clinically, the patient was completely edentulous with an intra-oral swelling at the ridge of the anterior mandible. Upon examination, the lesion was hard, fixed, painful swelling with palpable and tender submandibular lymph nodes (LN). An intra-oral ulceration over the tumor was evident with inverted edges.

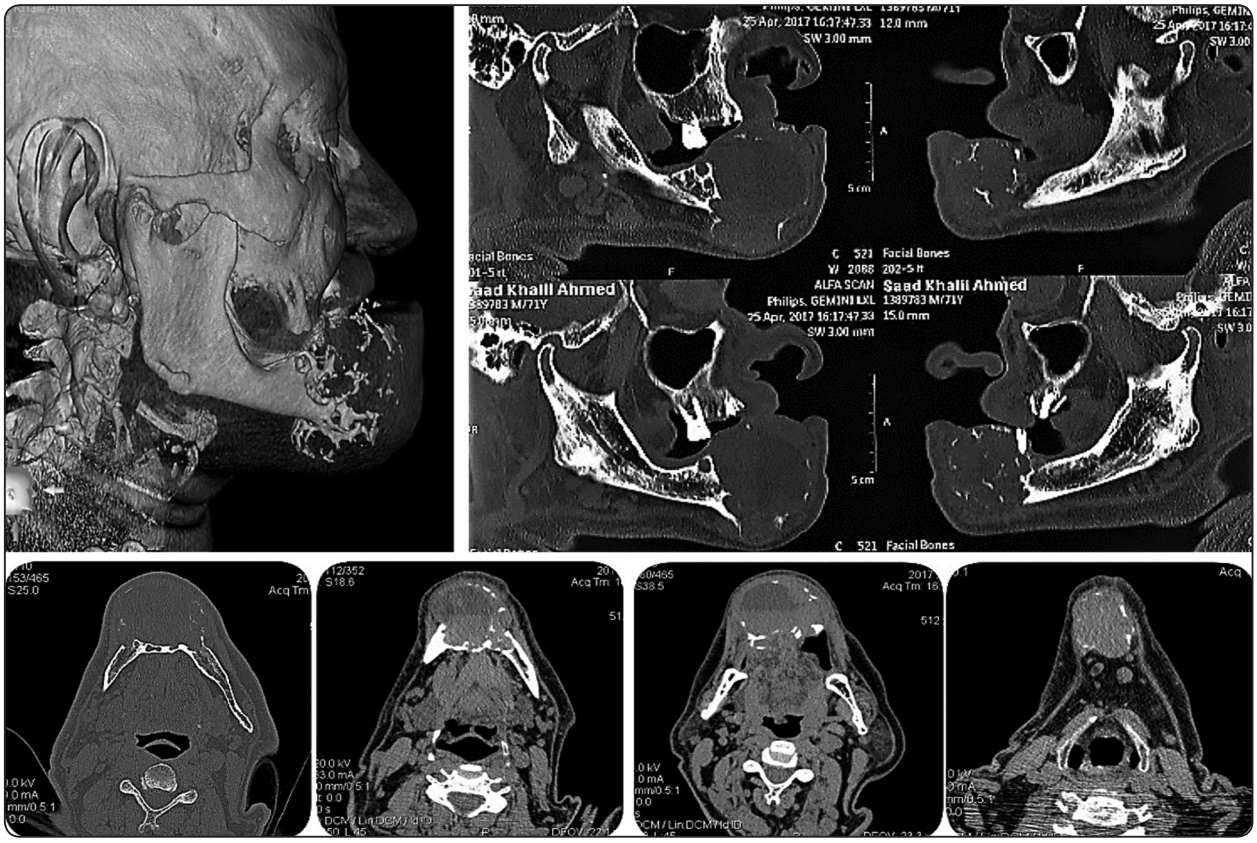

Fig. (2) Radiographic views showing the extension of the lesion.

Radiographic findings revealed a picture of well-defined, round, multilocular lesion, extending bucco-lingually, mesio-distally and cervico-facially (60 x 56 mm of size).

Adjunctive aids in diagnosis included the aspiration biopsy result which was negative. An incisional biopsy was collected from multiple areas through the lesion. The Microscopic Picture of the specimen formed from sheets, nests of columnar odontogenic cells with scattered eosinophilic rounded large cells and eosinophilic non-calcified globules were seen surrounded by odontogenic epithelium suspecting a follicular-type of Adenomatoid Odontogenic Tumor (AOT).

MATERIALS AND METHODS

The treatment plan was surgical resection of the lesion with safety free margins using computer guided 3D printed seating device to be used as an osteotomy guide with a stabilizing arm to prevent dislocation, disarticulation of the condyles and collapse of the ramus of both sides. This device aimed to preserve the contour of the anterior mandible during insertion of the reconstruction plate and the fibular vascularized flap.

Surgery:

Subcutaneous administration of 1:100000 adrenaline was performed. An extra-oral approach was done though apron incision to expose the lesion and the computer guided device was seated in the planned position.

The anterior slope of the device on both sides was planned to be used as guides for the osteotomy lines of resection. After the resection of the lesion, the seating device remained in place until the placement of the pre-bended reconstruction plate and its fixation to the mandible with the used immediate fibular vascularized flap for grafting.

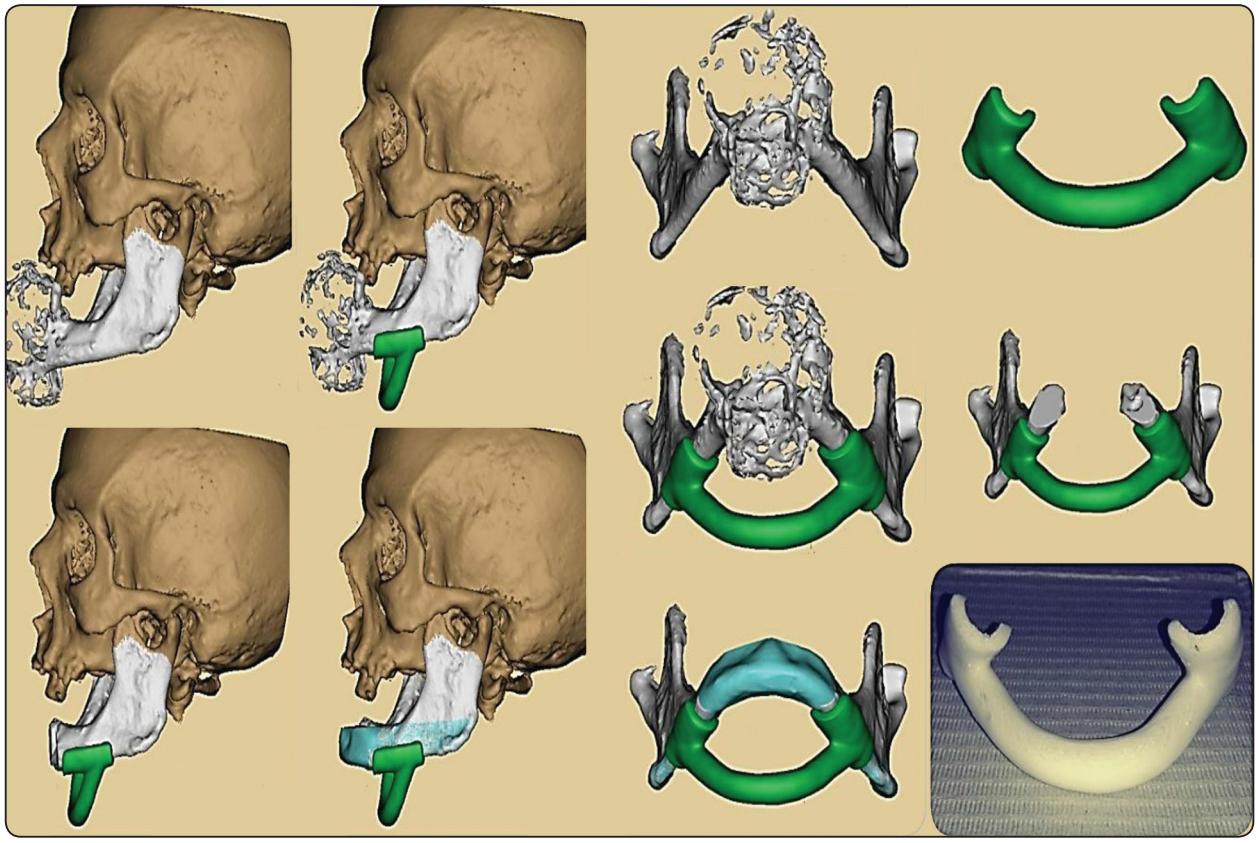

Fig. (3) Photographs showing computer guided 3D printed seating device.

Closure was done in 3 layers; platysma muscle and subcutaneous using 3/0 vicryle and skin using 5/0 polyprolyne

Follow-Up

Postoperative, 100mg of intravenous corticosteroid was administered every 8 hours for 3 days, Penicillin 1500 mg every 12 hours and NSAIDs for the post-operative pain and inflammation and was admitted into ICU for close observation. The patient was unstable after the admission, and chest infection was detected. After one week, the patient died from the acquired chest infection in the ICU hospitalization period, which made the post-operative CT not available as an option.

The excised lesion was sent to the pathology lab for microscopic examination. The specimen revealed a dominating clear cell carcinoma with minor granular ameloblastic appearance.

The microscopic features contained a diffusely infiltrative invasive tumor. Histopathological examination of H&E stained sections demonstrated islands of odontogenic epithelium. The outer cells were ameloblast-like showing reverse polarity, nuclear palisading and hyper-chromatism. The inner cells were stellate reticulum like cells and granular in other areas. Other areas demonstrated hypercellular masses formed of sheets of vacuolated clear cells that were oval or polyhedral in shape with small dark-staining eccentric nuclei with intervening basaloid cells and focal areas of palisaded cells.

The connective tissue was formed of fibroblasts, collagen and blood vessels. Inflammatory cells were around the tumor. Areas of skeletal bone invasion were also seen. There were no metastatic cervical lymph nodes in the dissected mass. Perineural

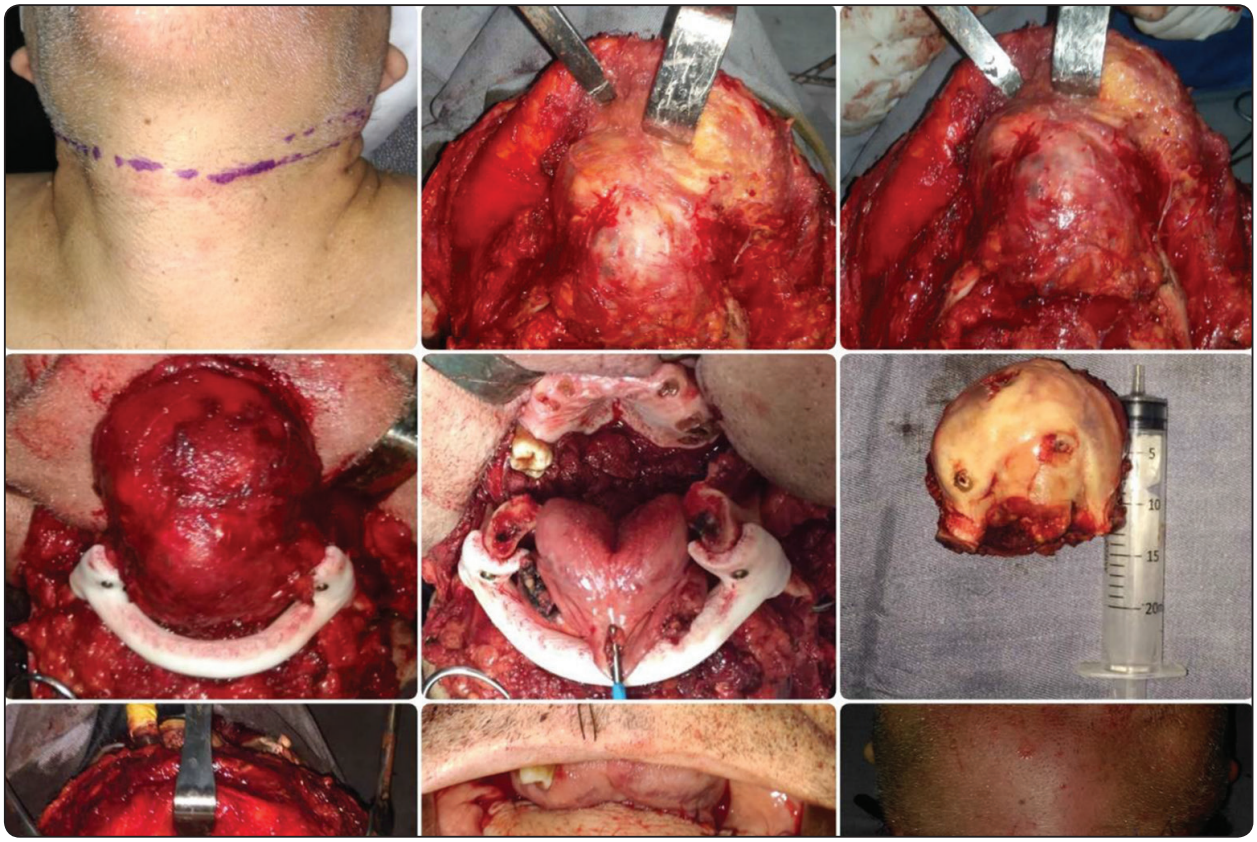

Fig. (4) Photographs showing the surgical procedure, tumor size and closure.

infiltration and vascular invasion were not seen. Bony margins were tumor free. The final pathologic staging was T4aN0M0 stage IVA.

On immune-histochemical staining, PAS showed glycogen positive, indicating clear cells. Mucicarmine was negative, eliminating mucoepidermoid carcinoma. Expression of CK-7, which is seen in the majority of cases of carcinoma, was positive focally. In addition, S-100 was negative, ruling out melanoma. SMA, a marker of proliferation of peri-endothelial smooth muscle cells and myofibroblasts was also negative.

Because of these results and the consistency with clear cells, the final readings of the rare CCOC were established. When added with the outer features of granular ameloblastoma, the final diagnosis reached up to be the Hybrid odontogenic tumor. (Fig. 7)

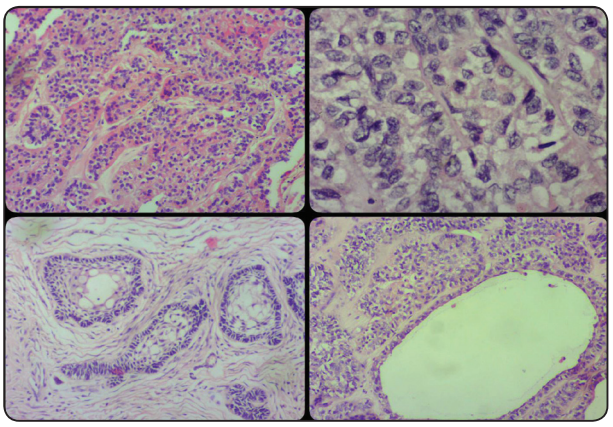

Fig. (5) Photographs showing the histological picture of the tumor.

DISCUSSION

According to literature, a total of 81 CCOC cases were recognized up to date. After ***Zhang et al*** [7] documented 6 cases and reviewed 67 cases, additional 8 cases were found in the literature [8-14].

CCOC was documented with a female tendency, and a Male/Female ratio of 1:2 and an average mean age of 55 (from 14 to 89). In addition, most of the cases were located in the mandible with a Mandible to maxilla ratio of 3:1. Our case was in a mandible of a male [3].

The typical clinical presentation of CCOC has been documented to be of a painless intra-bony swelling, followed by pain and paresthesia [15]. Contrarily, in our case, the clinical symptoms were quite different. Initially, the patient had a painful lesion, but there was no swelling and so it was misdiagnosed as a toothache in a private clinic before presenting to our hospital.

In general, CCOC is difficult to diagnose. ***Kim et al*** [14] presented a case of a well-defined unilocular radiolucent lesion that was similar to a cystic lesion and was misdiagnosed at first as an infected cyst. In our review of the previous cases, the most frequent radiologic appearance was radiolucent (only 4 cases were mixed type). Thus, the possibility of radiographic misdiagnosis is relatively high. A radiolucent lesion with jaw swelling and teeth loosening should be carefully assessed to allow for proper diagnosis and treatment as it may possibly be malignant CCOC.

Moreover, CCOC is also difficult to diagnose histo-pathologically. Other jaw tumors with prominent clearing cytoplasm include intraosseous salivary gland tumors (epithelial-myoepithelial carcinoma) and meta-static tumors (clear cell renal carcinoma). Additionally, some odontogenic tumors may also reveal clearing of their cells as calcifying epithelial odontogenic tumor, identified by the presence of amyloid deposits, and clear cell ameloblastoma which is difficult to distinguish from CCOC [15].

Accordingly, authors believed that clear cell ameloblastoma and CCOC might represent a single clinico-pathological neoplastic entity [16]. In addition, despite the different origin of cells, clear cell ameloblastoma and CCOC are difficult to separate morphologically and immune-histo- chemically. ***Bilodeau et al*** [17] suggested that location is the main criterion to distinguish between these tumors.

In CCOC, surgical resection with a wide safety margin is a must [15]. Therefore, proper jaw reconstruction is vital and should be performed simultaneously. In addition, this case was relatively rare in terms of its large size and simultaneous reconstruction with a micro-vascular free flap was needed.

In this study, we used the Fibular free flap reconstruction to compensate for the large defect of the mandible. Generally, it provides several benefits over other donor sites; adequate bone length, ease of graft dissection and good contouring. In addition, it offers the advantages of long pedicles with proper vessels and less donor site morbidity along with the use of a two-team approach. In this case, we obtained an adequate bone length (115 mm) and were able to reconstruct the mandible with satisfactory esthetics and no complications.

Recent studies have been trying to incorporate computer guided programs and devices, pre- and intra-operatively, in attempts to enhance the surgical accuracy. In this study, the unique use of the computer guided 3D printed seating device was utilized as an osteotomy guide. It was also beneficial in preventing the disarticulation of the condyles and collapse of the mandible bilaterally through its stabilizing arms.

Despite the patients' conditions, the only documented line of treatment was the complete resection of the lesion with safety margin due to the highly aggressive nature of the lesion with no documented alternative line of treatment [15]. Unfortunately, the patient here passed away after ICU admission. This was attributed to the patient preoperative medical condition together with the acquired postoperative ICU chest infection.

CONCLUSIONS

Our study uncovered the presence of a rare hybrid tumor of the CCOC and granular ameloblastoma. The survey of the literature clarified the characteristics of the rare CCOC which occurs in the 5th to 7th decades in the mandible of females appearing as a painless swelling.

The possible differential diagnosis is wide, recommending a careful assessment both clinically and immuno-histochemically.

In large CCOC cases of the mandible, wide resection with safe margin is the only documented line of treatment with subsequent composite fibula free flap reconstruction due to its highly aggressive malignant nature.

The use of computer guided 3D printed devices is always beneficial. In this case, it was used to hold the mandible intra-operatively to prevent its collapse during tumor removal and before mandibular fixation.

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