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(CASE REPORT)

Ghost cell odontogenic carcinoma of the mandible: Case report and review of literature

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Abstract

Ghost cell odontogenic carcinoma (GCOC) is an extremely rare odontogenic carcinoma that is typically characterized by an admixture of atypical epithelial cells and ghost cells. Up to date, GCOC remains rare with around 58 cases have been reported. Herein,we report a case of GCOC of the mandible, the patient is particularly young, successfully treated with surgical resection, reconstruction, and radiation, followed for 2 years without any sign of recurrence. A comprehensive literature review was performed.

Keywords: Ghost cell odontogenic carcinoma; GCOC; Ghost cells; Mandible

1. Introduction

Ghost cell odontogenic carcinoma (GCOC) is an extremely rare malignant odontogenic neoplasm with a significant potential for aggressive growth. Although the literature on this tumor is limited, its high recurrence rates suggest that early and multimodal intervention may be beneficial. Herein, we report a case of GCOC of the mandible that was successfully treated with surgical resection, reconstruction, and radiation. A comprehensive literature review was performed.

2. Case report

A 19 year-old woman, without notable personal and family history, who consulted in August 2020 for the appearance for 3 years of a left jugal swelling progressively increasing in volume and becoming painful, treated with paracetamol, amox clavulanic acid without improvement, all evolving in a context of apyrexia and conservation of the general condition.

Panoramic dental x-ray done on 2020/25/08 revealed an important multi-compartmental osteolytic lesion of the left mandible, reaching the coronoid process, and the temporal condyle, coming in close contact with teeth 35, 36, 37 including tooth 38 (figure 1).

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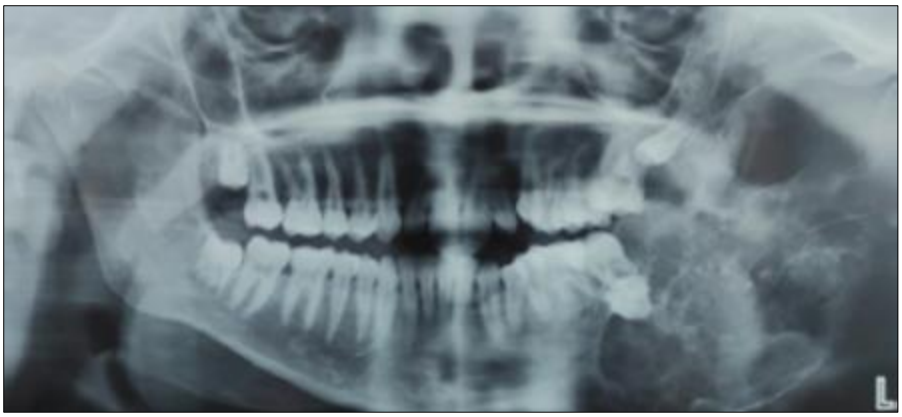

Figure 1 Panoramic dental x-ray with an important multi-compartmental osteolytic lesion of the left mandible

CT of the Facial Massif done on 2020/25/08 objectified a voluminous osteolytic mass involving the entire left mandible measuring approximately 80 mm in length with fleshy and liquid areas and multiple partitions, blowing the bony cortices and coming into intimate contact with teeth 35, 36, 37 and 38, lysing their alveolar margins, it pushed back the adjacent muscular structures and soft parts without invading them in favor of an odontogenic tumor.(figure 2)

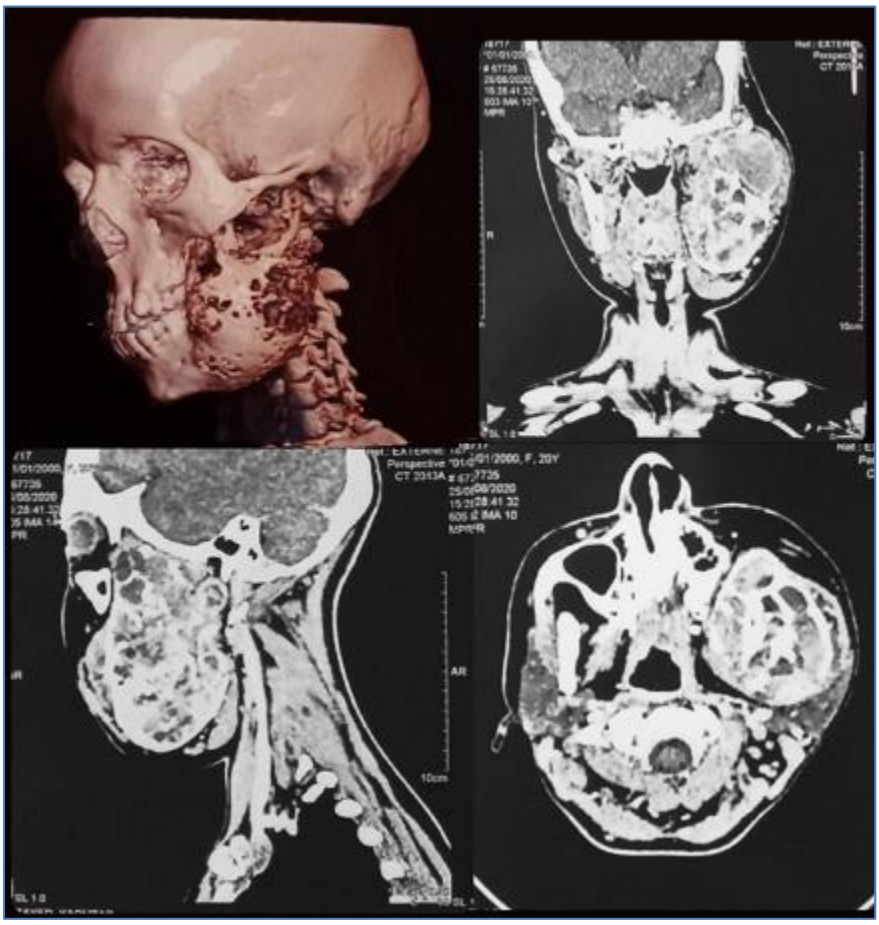

Figure 2 Cervicofacial CT with axial, coronal, sagital sections and 3D reconstruction showing a voluminous osteolytic mass involving the entire left mandible measuring approximately 80 mm.

On 2020/20/10, the patient underwent a left parasymphyseal interrupting left hemimandibulectomy removing the tumor in one piece with the placement of a mandibular reconstruction plate (figure 3)

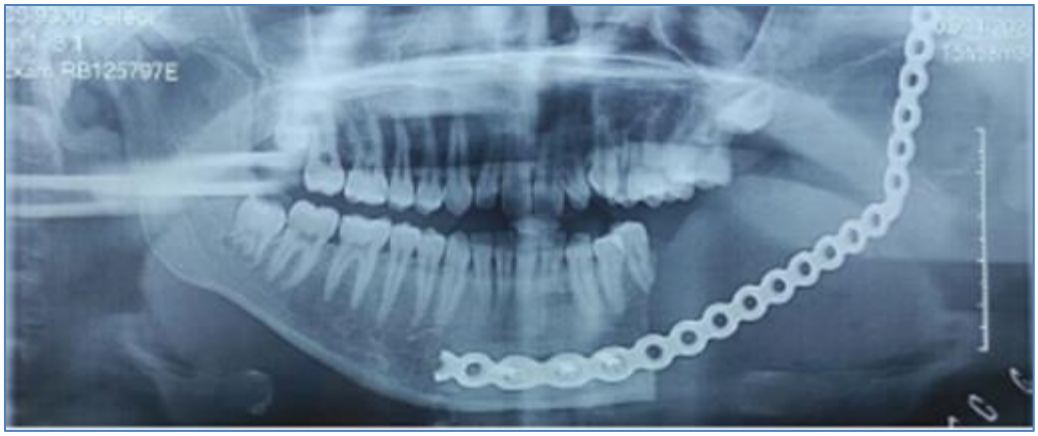

Figure 3 Postoperative dental panoramic after left hemimandibulectomy and plate reconstruction

Histopathological examination of the surgical specimen showed an odontogenic carcinoma whose histological appearance is compatible with ghost cell odontogenic carcinoma and the bone boundaries were tumorous

Histologically, it is a tumor proliferation infiltrating preexisting bone tissue arranged in clumps of varying size and in spans. In places, tumor cells are basaloid in appearance, rounded, with an enlarged nucleus, heterogeneous chromatin, and low basophilic cytoplasm. Certain massifs are bordered at the periphery by cylindrical cells of palisade arrangement provided with a nucleus projected towards the apical pole. At the center of these massifs, cellularity is looser. Several clusters of eosinophilic ghost cells are often seen in the center of clumps.

On 2020/07/12 , a complementary excision of the tumorous bone boundaries was performed with a margin of 1 cm taking away the two premolars and the anatomopathological examination found a buccal mucosa with bony tissue, histologically healthy, internal cuts healthy.

The indication for adjuvant radiotherapy was established in a multidisciplinary consultation meeting on 14/01/21 and the patient underwent radiation treatment on the operating bed at a dose of 50 Gy in 25 fractions, which was completed in March 2021. The immediate tolerance to the treatment was marked by a radiodermatitis and a radiomucitis grade 1 and an edema of the left cheek which did not interfere with the continuation of the treatment.

The patient underwent a cervicofacial scan on 19/09/2022 which did not show any recurrence.

Clinically she complains of a xerostomia grade 1

3. Discussion

GCOC is a malignant odontogenic tumor that is aggressive and devastating. Since Ikemura et al[1],first well documented one case in 1985, about 58 cases have been described thus far.

GCOC has different synonyms due to its diverse histopathological characteristics, including calcifying GCOC, malignant epithelial odontogenic ghost cell tumor, carcinoma arising from the calcifying odontogenic cysts (COCs), aggressive epithelial ghost cell odontogenic tumor, malignant COC, and malignant calcifying ghost cell odontogenic tumor [2,3]. This carcinoma belongs to the odontogenic ghost cell group together with calcifying cystic odontogenic tumor (CCOT) and dentinogenic ghost cell tumor (DGCT) and it may arise de novo or as a result of other odontogenic lesions; mainly CCOT or DGCT [4, 5].

Up to date, GCOC is an extremely rare malignancy and its diagnosis remains challenging with about 58 cases have been reported.

Of the reviewed 58 cases, The Asian population appears to be more susceptible to such tumor and men are at a higher risk than women with a ratio of 3.2:1. It can occur at any age, ranging from 13 to 80 years with a peak incidence in the fourth to the sixth decade of life [3, 6].

The maxilla is the most commonly involved site, with a ratio of 2.1:1 . In addition, the molar area and ramus are the most affected parts of the mandible.

GCOC can develop newly or in a known COC /DGCT due to malignant transformation after a long evolution, and sometimes from other odontogenic tumors.[6, 7].

Few cases have reported the simultaneous existence of a COC and GCOC[1, 7].

The diagnosis of GCOC should be based on clinical symptoms, radiological examination, and histopathology.

The clinical manifestations are not specific, patients are often referred to the hospital with a chief complaint of painful or painless swelling of the bone, and local paresthesia, teeth mobility, and mucosal ulcer can be seen in some patients(3,6)

Because of the different compositions of GCOC tissues, the imaging of GCOC showed that mixed unicameral or multilocular radiolucent-radiopaque lesions with or without dfined borders are more common than radioactive lesions[8]. Displacement of tooth roots and various degrees of bony destructions or infiltration of adjacent structures are common; tooth impaction and root resorption can also occur(9-10). No typical or special presentation is found on CT of such lesions. Although bone destruction is frequently observed, the manifestation of tooth resorption was unusual.

Diagnosis of GCOC requires adequate biopsy samples with meticulous histopathological examination of multiple sections along with immunohistochemistry.

The presence of ghost cells is not a specific feature, as they can also be seen in pilomatricoma, craniopharyngioma, odontoma, and ameloblastic fibro-odontoma[2,11].

Histologically, ‘‘ghost cells,” are enlarged polygonal eosinophilic epithelial cells without a nucleus stained with hematoxylin and eosin. The formation of ghost cells is still unclear, and ischemia induced degeneration or metaplastic of epithelial cells may play an important role[12].

The stroma is fibrous. Cellular pleomorphism is prominent and mitoses are frequently observed. The malignant contingent can be separated or mixed with the benign contingent. Ghost cells vary in number and can be isolated or grouped into clusters [3, 13]

Foci of calcification, necrosis and dysplastic dentin can also be seen [3, 6].

Immunohistochemical staining with a panel of antibodies demonstrated that neoplastic cells were strongly positive for cytokeratin and EMA. Positivity for NSE and the p53 protein has been reported. and negative for vimentin, desmin, SMA,S100protein and CD34[3,14]. The expression of Ki-67 and MMP-9 may be associated with the proliferation, invasion, and prognosis of GCOC [15]

Rappaport et al[16] reported that mutation of the β-catenin gene was noted at codon 33 in GCOC. Compared to CCOT and DGCT, GCOC demonstrates extensive bone destruction, and tartrate-resistant acid phosphatase and vitronectin receptor were strongly expressed in the ghost cells of GCOC[17].

Further research is required to determine if these biomarkers can be used to determine malignant transformation or high recurrence rate.

The recommended treatment for GCOC is wide surgical excision with clean margins (2,18) .No research to date has been able to draw definite conclusions on whether adjunctive radiotherapy with or without chemotherapy has efficient outcome(7,19)

Adjuvant cetuximab may be an option; however, it has been used in only one case.

4. Conclusion

GCOC has an unpredictable biologic behavior as it may range from slowly progressing to rapidly destructive growth, with high local aggressive features, recurrence, and occasional distant metastases.

To date only a few cases of GCOC have been reported in the scientific literature.

It is commonly recognized that Wide surgical excision with clear pathological margins is a comparatively radical cure for GCOC. However, adjunctive chemotherapy or radiotherapy is controversial and of questionable value in preventing recurrence.

To develop a full picture of GCOC treatment, additional studies will be needed to obtain overall information about its pathological and biological features. Taken together, a multidisciplinary team is essential to determine proper therapy and optimal outcome.

Compliance with ethical standards

Acknowledgments

We thank all participants in this study.

Disclosure of conflict of interest

The authors declare that they have no conflict of interest, and all authors have read and approved the final draft.

Statement of ethical approval

The present research work does not contain any studies performed on animals/humans subjects by any of the authors

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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