

## Case Report

### Granular cell type of ameloblastoma

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#### ABSTRACT

Ameloblastoma is a locally invasive tumor derived from odontogenic epithelium. An uncommon variant of ameloblastoma is granular cell type, which cannot distinguish from other ameloblastoma subtypes by clinical and radiographic findings alone. Only review of its microscopic features allows distinction from other subtypes. The purpose of this article is to present a case of granular cell ameloblastoma. This subtype should be distinguished from the other histopathologic subtypes because of its higher recurrence rate and more aggressive biological behavior. Radiographic and histologic findings as well as treatment are also discussed.

**Key Words:** Ameloblastoma, jaw neoplasms, lysosome, mandibular disease, odontogenic tumor

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## INTRODUCTION

Ameloblastoma is a locally invasive tumor derived from odontogenic epithelium.<sup>[1]</sup> Majority of patients present in the fourth decade.<sup>[2]</sup> Men are involved more than female<sup>[3]</sup> and more than 80% of ameloblastomas are in the mandible (mostly angle and ramus).<sup>[4]</sup> Clinically, jaw swelling and pain are the most frequent presenting symptoms.<sup>[5]</sup> Radiographically, ameloblastoma is included solid (multicystic) and unicystic.<sup>[4,6,7]</sup> Microscopically, the follicular and plexiform patterns are the most frequent and less common histopathology subtypes include the acanthomatous, granular cell, desmoplastic, and basal cell.<sup>[5]</sup> Granular cell ameloblastoma (GCA) is a rare subtype (<3/5%).<sup>[8]</sup> It cannot be distinguished

from other ameloblastoma subtypes by clinical and radiographic findings alone<sup>[9]</sup> histopathology features of GCA are characterized by the groups of granular cells, which have abundant cytoplasm filled with eosinophilic granules.<sup>[5]</sup>

The granular cells usually form the central mass of the epithelial tumor islands and cords.

The periphery of the islands consists of nongranular columnar cells.<sup>[10]</sup>

Sometimes, granular cells' phenotype has been attributed to an aging or degenerative change in long-standing lesions.<sup>[6]</sup> However, this tumor usually shows higher recurrence rate and more aggressive

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behavior which demand a close postoperation follow-up.<sup>[10]</sup> The purpose of this article is to present a case of GCA and review its microscopic features that allow it's distinction from other ameloblastoma subtypes. Radiographic and histologic findings as well as treatment are also discussed.

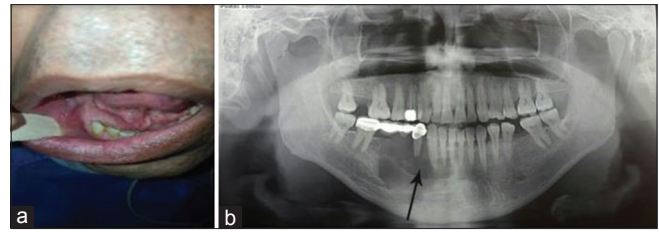
## CASE REPORT

A 47-year-old male presented with a chief complaint of a painless swelling in his right mandible and mobility of lateral and canine teeth in the same side. Swelling was begun from 3 years ago until 8 months before was reached to present size; mobility of teeth revealed from 3 months ago. There were no lymphadenopathy and tenderness [Figure 1a].

Panoramic radiograph showed a large, multilobular radiolucency with ill-defined borders, located in the body of partial edentulous right mandible and extending from the lateral incisor to the first molar area [Figure 1b]. According to preoperative management of patient, routine biochemical and hematological investigations were done, and all were within normal limits. With a differential diagnosis of central giant cell granuloma or odontogenic tumors or any other centrally located mesenchymal tumors, the patient posted for further evaluation.

Incisional biopsy was done, but the resected tissue was found to be insufficient to arrive at a histopathological diagnosis. The patient refused further incisional biopsy; based on the suggestion of the surgeon, the patient posted for surgery. Under general anesthesia, removing part of the jawbone including tumor with right lateral and canine teeth was performed. In gross, tumor appeared as a combination of cystic and solid areas [Figure 2].

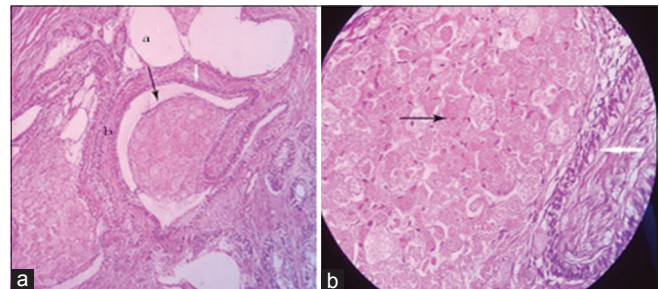
Histopathology survey of surgical specimen revealed a combination of cystic and solid areas. The peripheral layer of cystic areas consisted of a parallel arrangement of tall cylindrical cells with reverse polarity [Figure 3a, white arrow] of their hyperchromatic nuclei and vacuolization of the cytoplasm, and in solid area, the accumulations of cell rich in eosinophilic granular cytoplasm were found [Figures 3b, black arrows]. Furthermore, in the periphery of solid part of islands and cords of epithelium, a row of cell similar to ameloblast was found. According to above findings, diagnosis of GCA was given. After 2 months, the radiography which was taken showed acceptable healing improvement



**Figure 1:** (a) Patient with a swelling on the right mandibular vestibule. (b) Panoramic radiograph showed a large, multilobular radiolucency, located in the right mandible, with resorption of the canine and premolar teeth (arrow).



**Figure 2:** Photograph of surgical specimen appearing as a combination of cystic (black arrow) and solid areas (white arrow).



**Figure 3:** (a and b) Photomicrographs showing tumor appears in cystic (a) and islands (b) with a peripheral layer of ameloblasts (white arrow) and extensive central areas of granular cells with large, eosinophilic cytoplasmic granules (black arrow). a: H and E,  $\times 40$ ; b: H and E,  $\times 400$ .

and patient schedule for follow-up in 6-month interval [Figures 4a and b].

## DISCUSSION

The age distribution of granular cell variant is similar to the other types of ameloblastomas which shows an approximately equal prevalence in the third to seventh decades of life. About 85% of tumors occurred in the mandible, the vast majority of which affected the molar–ramus region.<sup>[5]</sup>

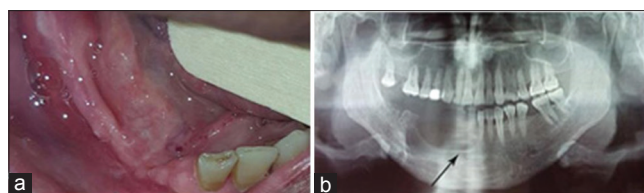
Jaw swelling and pain were the most frequent presenting symptoms. Compared to the other ameloblastoma subtypes, no distinguishing radiographic findings have been reported, the patient in this study was completely matched to above finding. In review of literature and case report which was done by Arora *et al.*, similar clinical and histopathological features with our case also could be found.<sup>[8]</sup>

Histopathologically, GCA has numerous large eosinophilic granular cells. These cells usually form the central mass of the epithelial tumor islands and cords. The periphery of the islands consists of nongranular tall columnar cells. GCA is diagnosed by the presence of granular cells, which usually occur within the central area of tumor and progressively replace the stellate reticulum.<sup>[11]</sup> Our case also showed similar features. Ultrastructurally, it has been revealed that the lysosome accumulation in these cells provides the characteristic granularity.<sup>[5]</sup>

It is evident from the literature; there are two main lines of interpretation about nature of granular cells, some consider it as a metabolic, while others of the view that it represent a degenerative process. More recent observation supports the later view to be more tenable based on the increased expression of death signaling molecules. Taneeru *et al.* suggested that the synthesis of signaling molecules such as  $\beta$ -catenin and Wnt-5a is upregulated in the granular cells, but their transportation or secretion is impaired, resulting their accumulation within granular cells, as autophagosomes.<sup>[12]</sup>

The GCA has a more aggressive behavior compared with the other subtypes; it may be locally aggressive and has relatively higher recurrence rate.<sup>[9]</sup> Unlike the case reported in this article, despite curettage with peripheral osteotomy which was done, after 2 months, radiography showed acceptable healing improvement. However, we need an extended period of follow-up in this patient for better judgment.

The differential diagnosis of GCAs includes other oral lesion with a similar morphology of granular cell accumulation such as granular cell tumor, granular cell odontogenic tumor, and congenital epulis, but these lesions usually could differentiate easily.<sup>[5]</sup> Treatment of ameloblastomas should be based on patient's history, clinical, radiographic examination, and finally histopathology findings.<sup>[13,14]</sup> However, similar to the other types of solid ameloblastoma, the prognosis



**Figure 4:** (a) Intraoral view which shows normal healing after 2 months. (b) Follow-up after 2 months which show sclerotic bone formation at the base of the surgical area (arrow).

is more dependent on the surgical procedures, i.e., GCAs treated by enucleation or curettage exhibit a high recurrence rate.<sup>[5]</sup>

Surgical options include segmental resection, *en bloc* resection, simple curettage, and excision with peripheral osteotomy.<sup>[13,14]</sup> The last one which was done for our patient and after 2 months clinically [Figure 4a] and radiography which was taken [Figure 4b] showed acceptable healing improvement and patient schedule for follow-up in 6 months interval.

## CONCLUSION

GCA is a rare condition with unique histopathology findings; this subtype should be distinguished from the other histologic subtypes because of its higher recurrence rate and more aggressive behavior and necessity of a long period of follow-up.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

The authors of this manuscript declare that they have no conflicts of interest, real or perceived, and financial or nonfinancial in this article.

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