

# Granular cell ameloblastoma as a solitary peripheral growth after twenty years of segmental resection of the mandible: A rare case of recurrence

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## Abstract

The ameloblastoma is a slowly growing, locally invasive, benign epithelial odontogenic neoplasm of the jaws with a high rate of recurrence if not removed adequately. We report an interesting case of granular cell ameloblastoma, which presented as a solitary, peripheral, soft tissue growth 20 years after initial segmental resection of the left mandible. The basal layer of oral mucosa could be the possible source of peripheral ameloblastoma in our case. In order to reduce the chances of recurrence, we suggest to incorporate mucosal stripping along with the conventional treatment as a mandatory rather than an elective procedure while treating ameloblastoma.

**Keywords:** Ameloblastoma, granular cell ameloblastoma, recurrence

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## INTRODUCTION

The ameloblastoma is the most common benign intraosseous epithelial odontogenic neoplasm predominantly affecting the posterior mandible. It is a slow growing, locally invasive tumour characterized by expansion and a tendency for recurrence if not removed adequately.<sup>[1]</sup> Although histological variants have no prognostic implication, the six main histological types include (i) follicular, (ii) plexiform, (iii) acanthomatous, (iv) granular, (v) basaloid, and (vi) desmoplastic forms.<sup>[2]</sup>

We report an unusual case of granular cell ameloblastoma (GCA) which presented as a peripheral, soft tissue growth in the left posterior mandible of a

40-year-old female patient, 20 years after the primary segmental resection of the left mandible. Our case also highlights the importance of life-time follow-up for a patient with ameloblastoma.

## CASE REPORT

A 40-year-old female patient reported to our department with a chief complaint of gradually increasing painless mass in the left mandibular posterior region since 1 month. The patient gave the past history of surgical removal of the ameloblastoma involving the left mandibular posterior region in 2001. Patient's medical, social, and family history was non-contributory.

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Extra-oral examination showed slight facial asymmetry with left mandibular deviation. No evidence of draining sinus, pus discharge, or cervical lymphadenopathy was observed [Figure 1a]. Intra-orally, a 2 cm × 1 cm solitary, nodular growth in the left mandibular edentulous posterior region was observed. The overlying mucosa was erythematous and fissured [Figure 1b]. On palpation, the growth was non-tender, sessile, and firm in consistency. Based on the clinical examination, differential diagnosis of reactive lesions like pyogenic granuloma, peripheral giant cell lesion, and fibrous hyperplasia was considered. Keeping previous history of surgical removal of the ameloblastoma in mind, due consideration to recurrent ameloblastoma was also kept in mind.

A panoramic radiograph showed the presence of radio-opaque bone plate extending from the left symphysis to the left condylar region suggestive of a previous segmental resection of the left mandible followed by reconstruction of the left mandible with metallic bone plate [Figure 2]. No evidence of lytic lesion was observed in the area corresponding to the clinical lesion.

Surgical excision of the mass was performed under general anaesthesia. Grossly the tumour was white in colour, ovoid in shape, measuring about 2.5 cm × 1.5 cm in size. The cut surface was smooth, white in colour, and firm in consistency [Figure 3].

Microscopic examination of the haematoxylin and eosin stained sections showed fibrous connective tissue exhibiting numerous odontogenic epithelial islands with the peripheral tall columnar cell revealing nuclear reversal of polarity, resembling ameloblast-like cells [Figure 4]. Centre of the islands showed stellate-reticulum-like cells with numerous large eosinophilic rounded or polyhedral granular cells. The final diagnosis of peripheral GCA was rendered [Figure 5]. The post-operative healing was uneventful. However, the patient was lost to follow-up.

## DISCUSSION

The GCA is a rare variant accounting for only 5% of all ameloblastoma and found as an admixture with other histologic patterns, mostly the follicular variant.<sup>[3,4]</sup> Microscopically, GCA is characterised by the groups of granular cells, which have abundant cytoplasm filled with eosinophilic granules that resemble lysosomes, both ultrastructurally and histochemically. The acquisition of granular cell phenotype has been attributed to an ageing or degenerative change in long-standing lesions like ours.<sup>[5,6]</sup>



**Figure 1:** (a) Extra-oral view showing left deviation of the chin. (b) Intraoral growth in the edentulous left mandibular posterior region



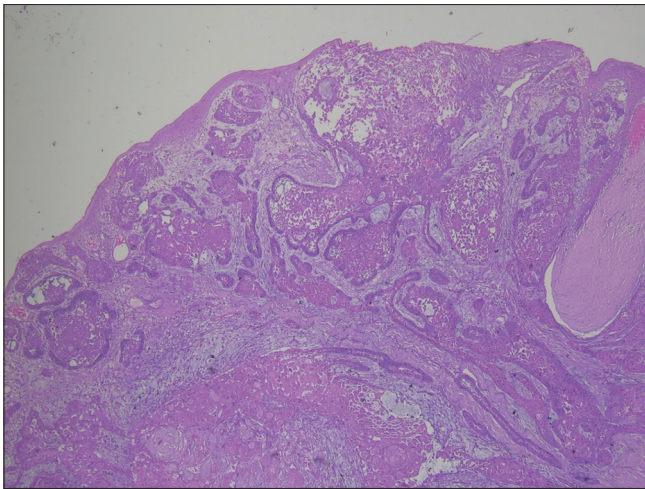
**Figure 2:** The panoramic radiograph showing a well-defined characteristic radio-opacity in the form of bone plate on the left side of mandible extending from the mandibular symphysis to left condylar region



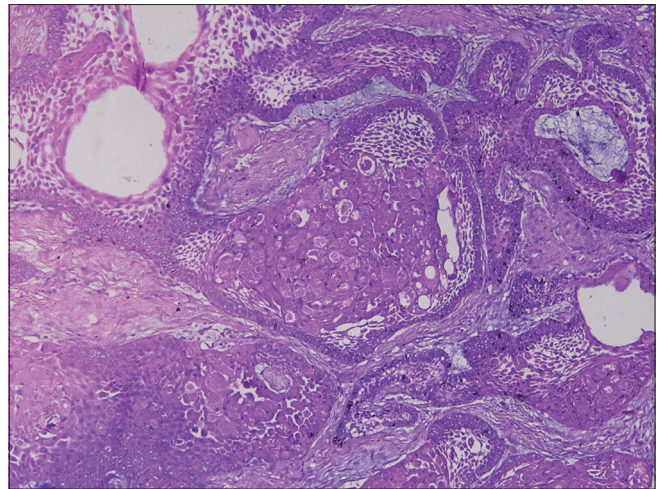
**Figure 3:** Gross specimen appearing ovoid soft tissue mass measuring 2 cm × 1.5 cm. Consistency of the specimen was firm. Inset: solid, smooth, and whitish cut surface

GCA may rarely behave in a malignant fashion giving rise to regional or distant metastasis.<sup>[7,8]</sup> Extra-oral soft tissue recurrent ameloblastoma as a granular cell variant 41 years after extirpation of a follicular type ameloblastoma has been reported in the literature.<sup>[9]</sup>





**Figure 4:** Histopathology showing numerous odontogenic epithelial islands composed of peripheral tall, columnar, ameloblast-like cells, and centrally placed stellate reticulum-like cells with coarse granular eosinophilic cells (Haematoxylin & eosin stain 4x)



**Figure 5:** Histopathology showing stellate-reticulum like cells along with numerous large eosinophilic rounded or polyhedral granular cells (Haematoxylin & eosin stain 10x)

The present case is different from the previous reports of ameloblastoma because the tumour occurred as a solitary intra-oral soft tissue growth 20 years after the first surgery and did not show intra-osseous component or malignant alteration after such a long period.<sup>[10,11]</sup>

The differential diagnosis of GCA includes granular cell odontogenic tumour, granular cell tumour and congenital epulis, all of which have different clinical presentation and biological behaviours.<sup>[12]</sup>

Immunohistochemically, the granular cells are found to be positive for cytokeratin, CD68, lysozymes, and alpha -1-antichymotrypsin, indicating epithelial origin and lysosomal aggregation.<sup>[8,13]</sup> Various researchers have demonstrated a link between the neuroectoderm, ameloblastoma, and normal oral ectoderm, suggesting the possible origin of ameloblastoma from the basal layer of the oral mucosa.<sup>[14,15]</sup> Occurrence of GCA as a peripheral growth 20 years after the first surgery favours the hypothesis that the basal cell layer of oral mucosa could be the sought out culprit for recurrence in our case. In view of the above, we recommended the stripping of the overlying mucosa along with the conventional treatment to reduce the chance of recurrence. The present case also emphasizes the life-time follow-up for the patient with ameloblastoma.

#### Author contributions

R.D. and N.A. have been involved in drafting the manuscript.

S.B. and C.F. revised it critically. All the authors have read and approved the final manuscript.

#### 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

There are no conflicts of interest.

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