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Granular Ameloblastoma a Case Report

Sushanth S. Bhat[1*], Laxmikanth Chatra[1], Prashanth Shenoy[1], K. M. Veena[1], Rachana Prabhu[1] and Sajad Buch[1]

[1]Department of Oral Medicine and Radiology Yenepoya Dental College Yenepoya (Deemed to be University) Karnataka, India.

Authors' contributions

This work was carried out in collaboration among all authors. Author SSB conducted the clinical procedures of the study and prepared the manuscript. Authors RP and SB assisted in clinical procedures and prepared the primary draft of the manuscript. Authors PS and VKM conducted the radiological investigation and gathered evidence for the treatment plan. Author LKC supervised the protocol. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Ameloblastoma usually presents as a painless, slow growing, locally aggressive swelling with expansion of the cortical bone, perforation of the cortical plates and infiltration of the soft tissues. It is a classic case of a true neoplasm of enamel organ tissue that lacks the potential to undergo differentiation. Granular cell ameloblastoma is a rare type of ameloblastoma being aggressive in nature with a marked propensity for recurrence and which can progress to metastasis.
This report discusses a case report of a 46 years old female patient diagnosed with granular cell ameloblastoma on the right side of mandible.

Keywords: Ameloblastoma; jaw neoplasm; mandibular disease; odontogenic tumor.

1. INTRODUCTION

Ameloblastoma usually presents as a painless, slow growing, and locally aggressive swelling of the face. It causes expansion of the cortical bone, perforation of the lingual or the buccal cortical plate and infiltration of the soft tissues. Robinson defined it as a "unicentric, non-

**Corresponding author: Email: bhatsushanth007@gmail.com;*

functional, intermittent in growth, anatomically benign and clinically persistent". It is a classic example of a true neoplasm of enamel organ type tissue that lacks the potential to undergo differentiation. Ameloblastoma is diagnosed between the ages of 30 and 50, and is very rare among children and elderly. About 80% of cases are seen in the mandible; of these, 70% develop in the posterior molar region, and often involve the ramus [1].

The current World Health Organization (2017) classification of odontogenic tumours classifies ameloblastoma into four types: solid/multisystem ameloblastoma, unicystic Ameloblastoma, the peripheral (or extra osseous) ameloblastoma, the desmoplastic ameloblastoma and malignant ameloblastoma. The six main histopathological subtypes of ameloblastoma are: follicular, plexiform, acanthomatous, granular cells, basal cell, and desmoplastic form. Granular cell ameloblastoma is a rare type of ameloblastoma, accounting for 3.5% of all ameloblastoma cases that shows marked transformation in the cytoplasm of tumor cells, which are usually stellate reticulum like cells.[2] The transformed cells possess very coarse, granular, eosinophilic cytoplasm. The "granular change" is thought to be due to a dysfunctional status of neoplastic cells, and the pathogenesis of this tumor seems to be age-related [3].

We present a case of a multicystic granular ameloblastoma in a 46 year old female.

2. CASE REPORT

A 46year old female reported to the Department of Oral Medicine and Radiology with chief complaint of swelling in the right side of the face since one month. It started as a small growth inside the mouth on buccal side of right jaw and attained the present size .She had dull aching and non radiating pain since two weeks.

Diffuse extra oral swelling was noted on right lower third of the face extending anterioposteriorly from the corner of the mouth to 2cm anterior to the angle of the mandible and superioinferiorly from the corner of the mouth to the lower border of the mandible. On palpation all inspectory findings were confirmed. The swelling was soft in consistency; tender, non compressible and non reducible .She had no parasthesia over the skin over the swelling. Intra orally: Diffuse swelling was noted on the buccal side extending from the mesial aspect of 43 to distal aspect of 46. Overlying mucosa was intact with no ulceration. The swelling was soft in consistency in the anterior region and bony hard in the posterior region. It was tender on palpation. Buccolingual expansion was noted and buccal sulcus was obliterated. Tooth number 17 and 48 were missing. Teeth were not displaced.

Investigations carried out were panoramic radiography, cone beam computed tomography, blood investigations, and excisonal biopsy. As the patient reported during the COVID-19 pandemic, nasopharyngeal and oropharyngeal swabs were taken for RT- PCR assay.

Panoramic radiography revealed [Fig1] a well defined multilocular radiolucency extending anterioposteriorly from the mesial aspect of 43 to the distal aspect of 46, interdental septae were noted superior-inferiorly from the roots of 46 up to the inferior alveolar canal. Root resorption was seen in 46, but lower border of the mandible was intact.

Cone beam computed tomography revealed an extensive well defined heterogeneous multilocular hypo dense area, on the right body of the mandible. Multiple spider web type of internal septa was noted within the lesion and buccolingual expansion with loss of buccal and lingual cortical plate on axial view [Fig2a]. Anterioposteriorly the lesion was extending from the mesial aspect of 43 till the distal aspect of 46 and superioinferiorly from the crest of the alveolar bone to 6mm above the inferior border of the mandible along with scalloping of borders. Root resorption was seen in relation to 44, 45 and 46. Inferior alveolar canal could not be appreciated [Fig 2b].

46 showed well defined hypo dense area in the coronal aspect involving the pulp chamber and the radicular portion was combined with the lesion. Inferior border of mandible was intact.

Nasopharyngeal swab revealed the presence of SARS CoV 2 RNA. Patient was treated for COVID and after the recovery period, excisional biopsy was performed.

The specimen was sent for histopathological examination. H and E sections revealed large odontgenic islands showing peripheral ameloblast like cells and central stellate reticulum-like cells and with extensive granular cells transformation surrounded by stroma [Fig 3a,3b]. Hence a final diagnosis of ameloblastoma, granular cell variant was made based on the histopathological report.

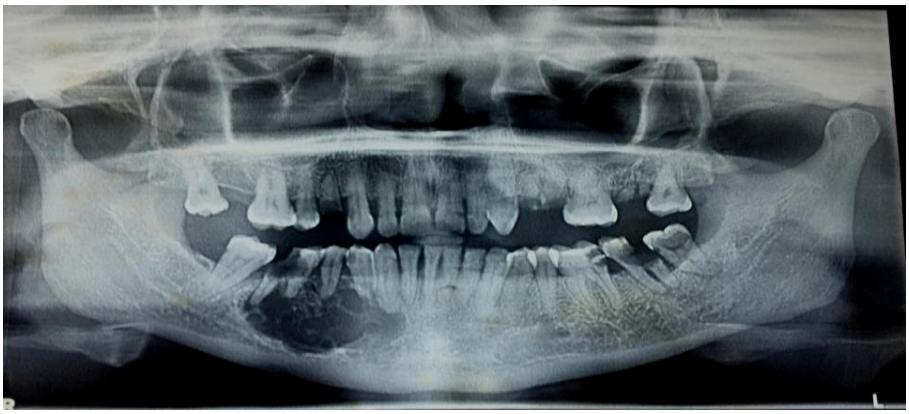

Fig. 1. showing the panoramic radiograph which shows a well defined multilocular radiolucency in the right mandibular arch

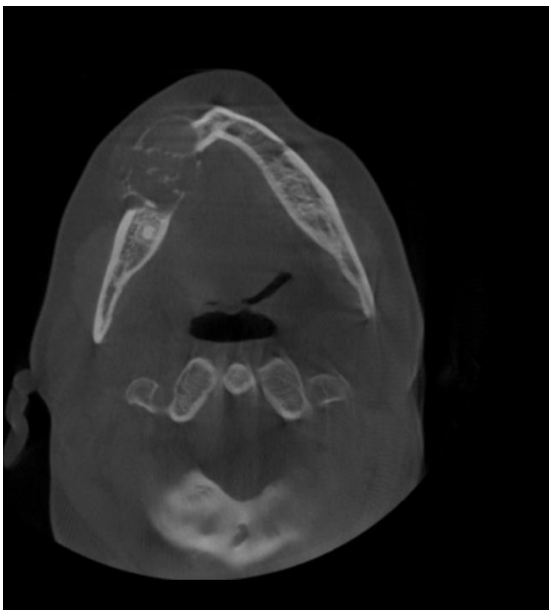

Fig. 2a. Showing the cone beam computed tomograhic in axial section extensive well defined heterogeneous multilocular hypo dense area, on the right body of the mandible. Multiple spider web type of internal septa was noted within the lesion and buccolingual expansion with loss of buccal and lingual cortical plate

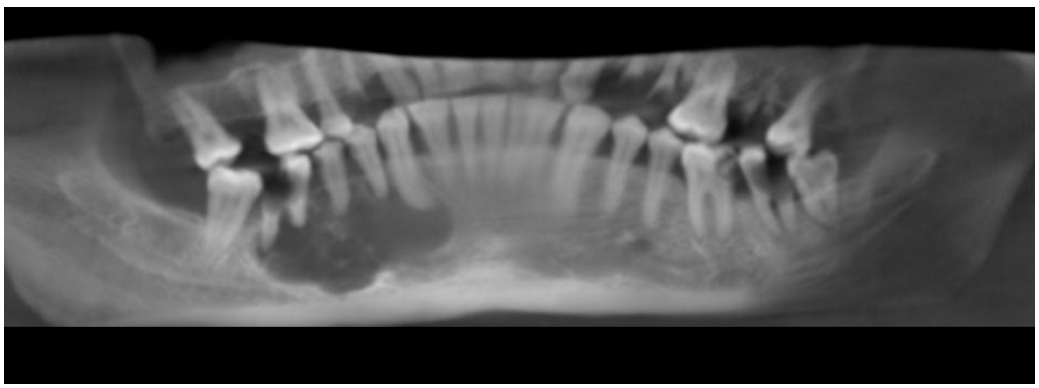

Fig. 2b. Shows the cone beam computed tomographic image in the panoramic view

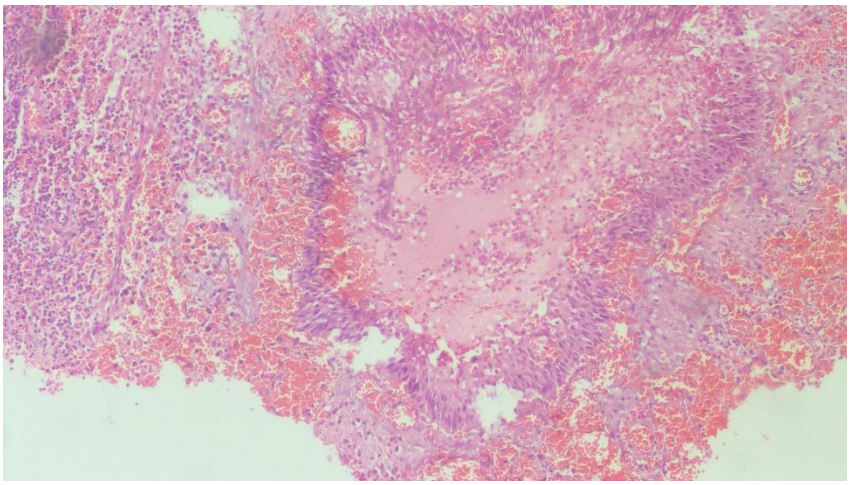

Fig. 3a. shows the photomicrograph of10x ameleoblastomas follicle which is lined by tall columnar ameloblast like cell which is hyper chromatic nuclei and reversal of polarity and central stellate reticulate like cell and the central reticulate is replaced by the granular cells

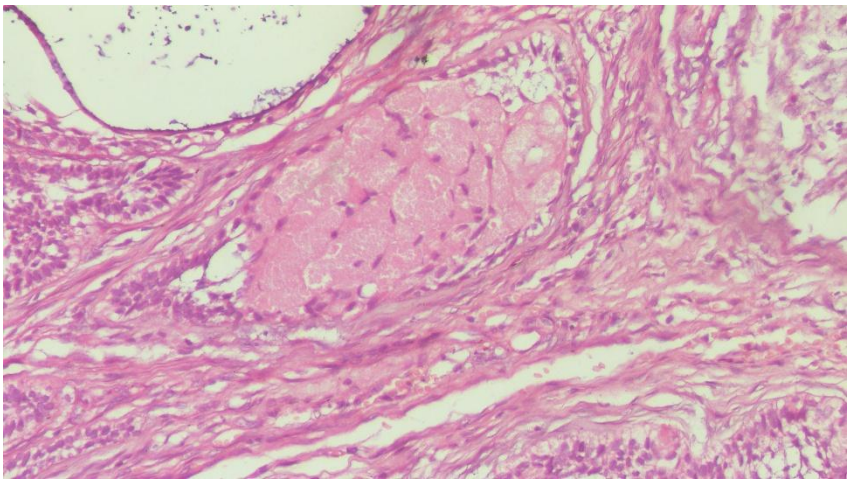

Fig. 3b. Shows the photomicrograph of 20x of ameleoblastomas follicle

3. DISCUSSION

Churchill defined ameloblastoma as unicentric non functional, intermittently growing anatomically benign but clinically persistent tumor.[4] It is the second most common odontogenic tumor, arising from rests of the dental lamina or from a developing enamel organ or from the epithelial lining of an odontogenic cyst, or from the basal cells of the oral mucosa. Rosenstein et al. describes a series of cystic ameloblastoma that showed more recurrence and aggressiveness as manifested by cortical perforation, large size of the lesion, multilocularity and extensive bone destruction.[5] The rarity of the granular cell ameloblastoma subtype and the possibility of confusion with other odontogenic and non-odontogenic lesions with a granular cell component require an understanding of the salient features of this locally aggressive neoplasm.[6]

In a recent Meta analysis of ameloblastoma, the mean age was 34 years and the peak age incidence in the third decade of life. A slight male preference (53%) was found, and the mandible appeared to be the preferred site. The most common type of ameloblastoma was multicystic. The histopathologic patterns were mostly follicular and plexiform. The incidence rate was determined to be 0.92 per million person-years [7].

80% of ameloblastomas occur in the mandible mainly the third molar region and the remaining 20% in the upper jaw [8]. The mandibular molar

and the ramus region are the most commonly affected sites and it is usually asymptomatic. It has seen in the both the genders. Radiographically, granular cell ameloblastoma resemble all the classical features of ameloblastoma. The most typical radiographic feature is multilocular radiolucent lesion [9].

The lesion is described as having a classical “soap bubble”. Buccal and lingual cortical expansion is present. Resorption of the roots of teeth adjacent to the tumor is common. All the classical radiographic features were noted in the case.

Granular cell change in ameloblastoma histopathology was first reported by Krompecher [10]. in 1918 and called it pseudoxanthomatous cells. Granularity is due to the marked transformation of the cytoplasm of the stellate reticulum cells into granular eosinophilic appearance. It is also known to be an aggressive histological variant of ameloblastoma [11]. There is not much change in clinical and radiological findings reported when compared from granular cell ameloblastoma and other histopathological variants of ameloblastoma.

Granular cell ameloblastoma is characterized by the presence of eosinophilic granular cells, measuring approximately about 1mm in size. The granular cells are seen in the central area with marked transformation of stellate reticulum cells into granular eosinophilic cells, surrounded by tall columnar cells [12]

The nuclei of granular cells are pyknotic, hyper chromatic showing reverse polarization which is seen in our case. This subtype should be distinguished from the other histopathologic subtypes because of its higher recurrence rate and more aggressive biological behaviour. [13] Very rarely, it was found to metastasize to lymph nodes and lung [14].

The surgical management of ameloblastoma is challenging due to the high recurrence rates and significant morbidity associated with radical treatment. A higher recurrence rate is associated with conservative treatment for ameloblastoma, while radical treatment leads to an increased number of postoperative complications that affect the patient's quality of life.[15].

In our case, the COVID state of the patient further negated the complexity of the case and surgical management had to be deferred.

4. CONCLUSION

Granular cell ameloblastoma is rarest variant of ameloblastoma with higher recurrence rate. Hence, it should be differentiated from the other granular cell lesions primarily because of its aggressiveness and higher recurrence rate. Patient has to be periodically followed because of its high recurrence rate.

CONSENT

The authors certify that they have obtained appropriate patient consent and she has given consent for her images.

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

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COMPETING INTERESTS

Authors have declared that no competing interests exist.

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