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A Rare Case of Granular Cell Type of Ameloblastoma: A Case Report

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ABSTRACT

Granular cell ameloblastoma is a rare histopathological variant of ameloblastoma accounting for about 5% of all subtypes. It is characterized by a nest of large eosinophilic granular cells. The present case report describes the surgical management of a 25-year-old male with granular cell ameloblastoma involving the body of the mandible. The lesion was confirmed by clinical, radiological, and histological findings and was managed by enucleation. A six-month follow-up reveal satisfactory outcomes concerning the healing and absence of recurrence.

Keywords: Granular cells, odontogenic tumors, Ameloblastoma, enucleation.

INTRODUCTION

Ameloblastoma is a benign yet locally aggressive odontogenic neoplasm that can cause extensive destruction of the jaws. Recent reports have described its incidence rate among all oral lesions to be between 1 to 2.5% in India.[1,2] Thus, while the tumor is the commonest odontogenic neoplasm following odontoma in its prevalence, its actual prevalence in the general population is quite low. Furthermore, numerous histopathological variants of AM have been described based on the pattern of odontogenic epithelium such as follicular, plexiform, papilliferous, adenoid, and admixed, or its surrounding connective tissue such as desmoplastic, hemangiomatous, keratinizing and dentinoid. While the tumor generally exhibits cells having ameloblast-like features, additional types such as granular cell AM, clear cell AM, acanthomatous AM, or basaloid AM are based on the differentiation or appearance of the constituent odontogenic cells of the tumor.[1,3]

Of these, the granular cell type is extremely rare, representing less than 5% of AM.[4] Consequently, the entity is quite rare with relatively fewer cases reported in the literature and therefore, not much is known about this type of AM. From a histopathological perspective, it is distinguished by an abundance of big eosinophilic granular cells.Various gold standard techniques CT and MRI are used to establish the extent of the lesion.[5]

This case report highlights the unusual case of granular cell type and its incidence, histological features, diagnosis, and management of a rare variant of ameloblastoma.

Case Report

A 25-year-old male complained of swelling in the right mandibular region for eight months. A history of extraction of the mandibular right first molar eight months ago was elicited following which the swelling developed. The swelling was initially small and increased to the present size. It was diffuse when observed extraorally and of size 3 x 2 cm. Intraorally, a well-defined swelling was observed extending from right mandibular canine to the second molar region with obliteration of buccal vestibule (Figure 1). Orthopantomogram revealed a multilocular radiolucency extending from the distal surface of the mandibular right lateral incisor to the mesial root of the second molar (Figure 1). Sharp knife-edge resorption of the roots of premolars was evident. A well-defined hypodense area surrounded by corticated borders was observed on a three-dimensional computed tomography scan. Expansion of the buccal and lingual cortical plates was evident in the involved area.

Given the clinical and radiographic features, a provisional diagnosis of conventional ameloblastoma was imparted. Differential diagnoses included odontogenic keratocyst and other benign and malignant odontogenic tumors. An incisional biopsy was obtained under local anesthesia and fixed in 10% neutral buffered formalin for 24 hours. The tissue was subjected to routine histopathological processing followed by H and E staining of 4 um sections. Microscopic examination showed odontogenic islands arranged in nests and islands in a background of fibrocellular connective tissue stroma (Figure 2). The neoplastic cells mostly included large eosinophilic cells rich with cytoplasm

rich in granules. Peripherally, ameloblast-like tall columnar cells with hyperchromatic nuclei exhibiting reversal of polarity resembling the lining of a typical AM follicle were present. The nucleus was directed away from the basement membrane of the islands and nests with evident subnuclear vacuolization. The histopathological features were suggestive of granular cell AM.

The treatment plan comprised enucleation and peripheral osteotomy under local anesthesia. An investigation of routine blood parameters was done, which were found within normal limits. Universal aseptic precautions were maintained. An incision was given from the mesial side of the mandibular right central incisor to the distal side of right second mandibular molar. A full-thickness mucoperiosteal flap was raised and the tumour was exposed. tumor margins were explored and enucleation and peripheral osteotomy were done (Figure 3). Thorough betadine irrigation was performed and the tissues were sutured with 3-0 vicryl sutures. Histopathological examination of the enucleated tumor mass confirmed the diagnosis of granular cell AM. A one-year follow-up. Patient was kept under follow-up for 1 year, and no recurrence was found to date (Figure 4).

DISCUSSION

Despite AM being one of the most commonly observed odontogenic tumors, the granular subtype is exceedingly rare. In a clinicopathological study, In an extensive study of the literature on AM in the earlier years, Reichart et al. found only 56 cases of granular cell AM among a total of 1593.[6] Kameyama et al. reported that only one out of 77 cases of AM could be classified as granular cell AM.[7] Because of its increased prevalence of recurrence, malignancy, and metastasis, it is treated as a different entity even though it is a variation of ameloblastoma. The present case report, thus, sought to add to the existing scant literature related to the lesion.

The aggressive nature of the lesion was evident in its destruction of a significant portion of the mandibular body and resorption of the roots of the premolars in the present case. Interestingly, the patient had a history of molar extraction eight months before the diagnosis of granular cell AM. Whether the extraction caused the lesion to occur or whether it was present inherently before the extraction or arose on its own is still unclear. Nevertheless, the likelihood of the former is more given the circumstances of the present case.

Generally, there are no distinguishing clinical and radiological findings reported when compared from granular cell ameloblastoma and other histopathological variants of ameloblastoma.[8] The unique blend of central large eosinophilic granular cells with peripheral ameloblast-like cells is a pathognomonic feature of granular cell AM. The presence of these histopathological features was the biggest clue in the diagnosis of the present case. Large eosinophilic granular cells are also observed in normal and neoplastic human tissues of the parathyroid glands (oxyphils), salivary glands (oncocytes), thyroid (Hürthle cells), granular cell ameloblastic fibroma, granular cell myoblastoma, and congenital epulis.[4,8] The main histogenetic difference is that the granular cells in AM are of epithelial tissue origin, while those in the latter three entities are derived from mesenchyme.

Numerous theories have been proposed on the origin and nature of granular cells in ameloblastomas. These granular cells are epithelial in origin, and several ultrastructural and histochemical studies have described them and stated that granular cells of ameloblastoma are due to lysosomal overload, metabolic change in response to aging, degenerative mechanism, or an indicatory of a more aggressive course, of which the most accepted concepts are aging phenomenon and degenerative changes.[9] The most common perception is that the granularity of cells is a product of aging or cell senescence representing degenerative changes in chronic long-standing cases of AM. However, the occurrence of this change in only a handful of individuals including those of younger age groups contradicts the statement to some extent. Immunohistochemistry-based studies have demonstrated the positivity of various biomarkers such as cytokeratins, CD68, lysozymes, and α-1-antichymotrypsin in the granular cell component of AM. Expression of the latter two markers is indicative of the fact that the cells have acquired a secretory phenotype rather than senescence. An upregulation in the synthesis of signaling molecules such as β-catenin and Wnt-5a has been demonstrated in the granular cells, but their transportation or secretion is impaired, resulting in their accumulation within granular cells, as autophagosomes. [10] Naturally, the cells exhibit negative staining for mesenchymal markers such as vimentin, desmin, and S-100 protein.

Surgical options include segmental resection, en bloc resection, simple curettage, and excision with peripheral osteotomy.[11] While aggressive treatments are preferred, enucleation and peripheral osteotomy were performed in the present case which yielded acceptable outcomes in terms of healing and absence of recurrence for a six-month follow-up period. The histopathological features being sufficient for diagnosis, immunohistochemistry was not required for confirmation in the present case although performing them would have greatly benefited the scientific community in understanding the lesion more clearly. Another drawback of the present case report is a relatively shorter long-term period to report any conclusive findings about the success of the outcome which warrants further long-term follow-up in the future.

CONCLUSION

Granules in GCA are due to the marked transformation of stellate reticulum cells into granular cells, which are considered lysosomal aggregates due to dysfunction of lysosomal enzymes or other related proteins. GCA should be differentiated from the other variants of ameloblastoma and also from other granular cell lesions due to its high recurrence rate. Patients should be kept under periodic observation due to their high recurrence rate even up to years after initial treatment. An immunohistochemistry finding helps to identify the molecular pathogenesis behind the granular cell formation in granular cell ameloblastoma.

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Figure Legends:

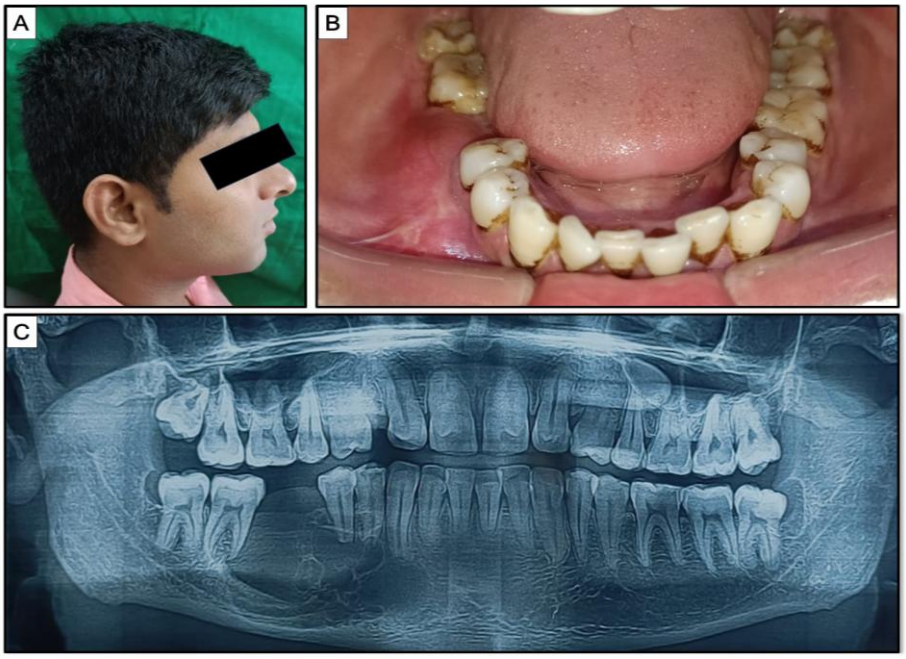

Figure 1: A) Lateral profile view showing extraoral swelling in the lower third of the face; B) Intraoral swelling leading to buccal vestibule obliteration; C) Orthopantomogram showing multilocular radiolucency involving the right mandibular body

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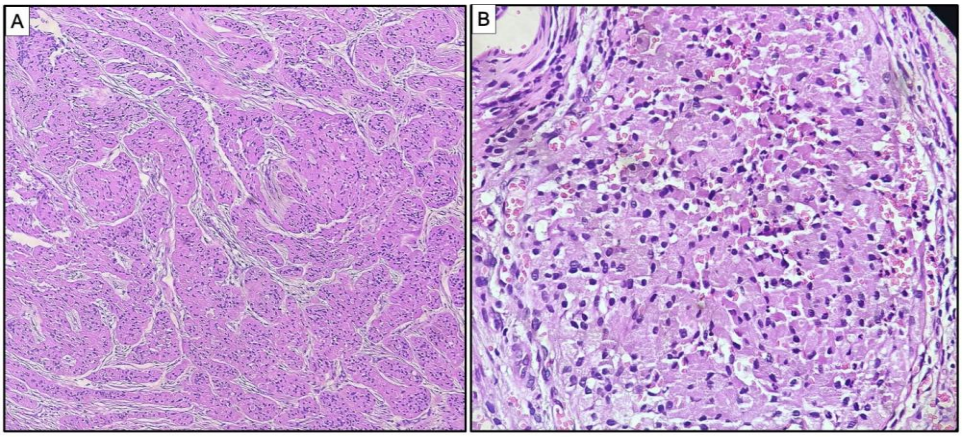

Figure 2: A) Nests and locules of eosinophilic cells (4X magnification) B) Cells exhibiting granularity in the cytoplasm. (10X magnification)

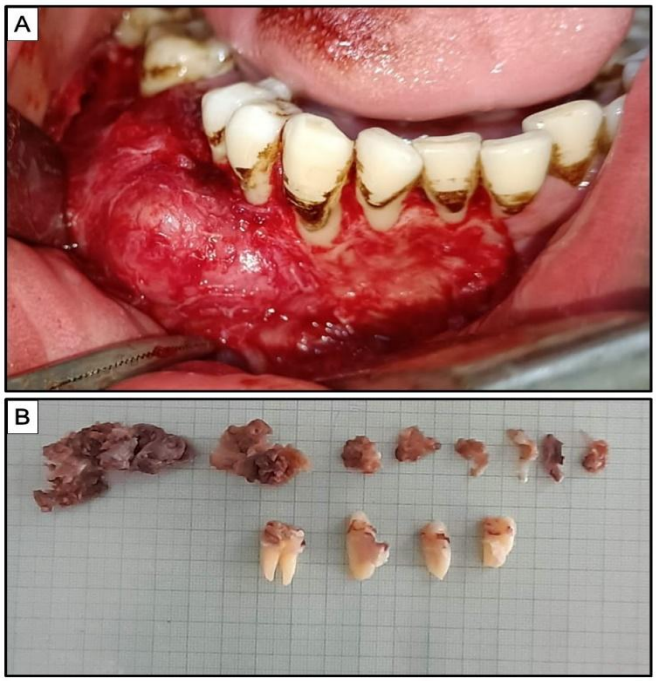

Figure 3: A) Surgical enucleation and B) Enucleated bits of tumor with extracted teeth

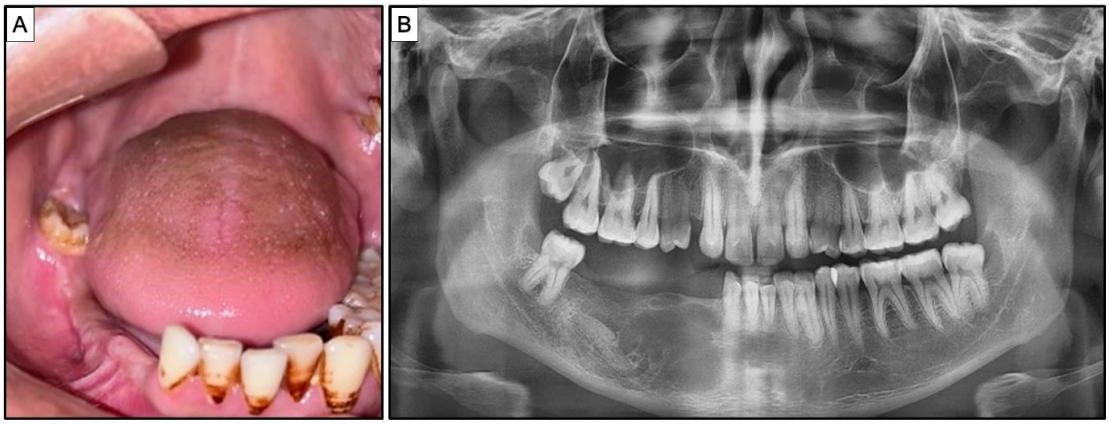

Figure 4: Adequate healing observed at 6-month follow-up on A) Intraoral examination and B) Orthopantomogram