Granular Cell Ameloblastoma-A case report

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ABSTRACT:
Granular cell ameloblastoma is a rare variant of ameloblastomas, considered as an aggressive and recurrent tumour. Microscopically the cells of granular cell ameloblastoma show cytoplasm filled with the granules. There is a marked transformation of the cytoplasm usually of the stellate reticulum like cells. So, it takes very coarse granular eosinophilic appearance. Ultrastructural studies show that cytoplasmic granules represent lysosomal aggregates with no recognizable cellular component. This variant is seen commonly in younger age group. A differential diagnosis with granular cell odontogenic tumour, granular cell tumour and congenital epulis was also made for proper treatment prognosis.

Keywords: Aggressive odontogenic tumour, Granular cell ameloblastoma, Granular Cell Tumor

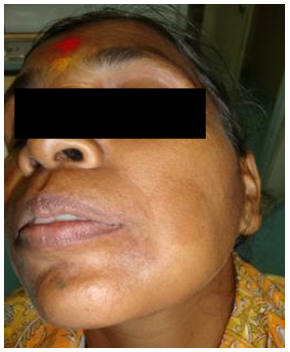
INTRODUCTION:

Granular Cell ameloblastoma is a rare variant of ameloblastomas. It is a very aggressive tumour as well as the recurrence rate is very high. Also this tumour is seen in younger age group which is the uniqueness of this tumour. Very few cases have been reported till date in India. In this article we will be discussing a case of a 50 year old female patient with a complaint of swelling in the lower left jaw since two years. Also a differential diagnosis has to be made so as to impart a proper treatment to the patient.

CASE HISTORY

A 50 year old patient reported to the outpatient department of Bharati Vidyapeeth Dental College, Pune for the complaint of swelling in the lower left side of the jaw since two years.

On extra oral examination there was facial asymmetry seen. The swelling extended from the midline till the angle of the mandible. On palpation the swelling seems to be firm but no pain. The overlying skin appeared normal. The swelling extended from the midline to angle of the mandible of the left side measuring 6cm x 3.5 cm. The intraoral examination reveals 33, 34, 35, 36, 37 teeth missing. The cortical plates of buccal and lingual sides seems to show expansion. The overlying mucosa was normal. The swelling was non ulcerated, non-tender.



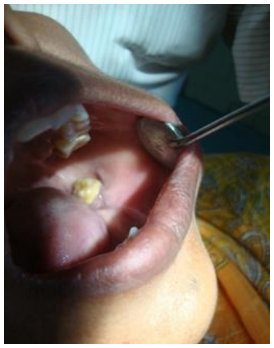


Figure 1: extraoral swelling on left side of the face and Figure 2: intraoral swelling in the retromolar region

The patient did not give history of any illness. The routine haematological examination was in normal limits. OPG revealed multilocular radiolucency with sclerotic border extending from 31 to 38 and bone destruction seen till the lower border of the left side of the mandible

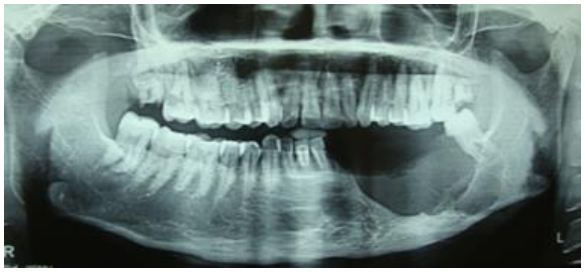


Figure 3: OPG showing radiolucency extending from mandibular left canine till the third molar anteroposteriorly and inferiorly extending till the border of the mandible.

Excisional biopsy of the lesion was done under general anaesthesia.

MACROSCOPY

The surgical specimen consists of hemimandible with the lesion measuring about 5.5 cm x 3.5 cm firm in consistency, with regular border.

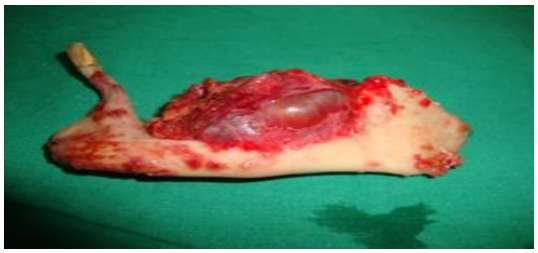


Figure 4: Gross specimen showing resected hemi-mandible

HISTOLOGICAL FINDINGS

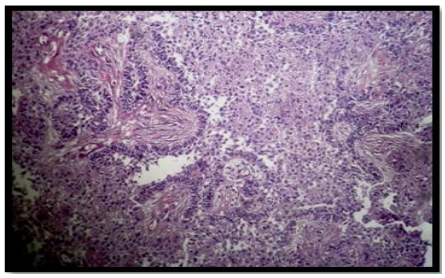


Figure 5: Odontogenic islands resembling follicular ameloblastoma in 4 x magnification with peripheral tall columnar cells

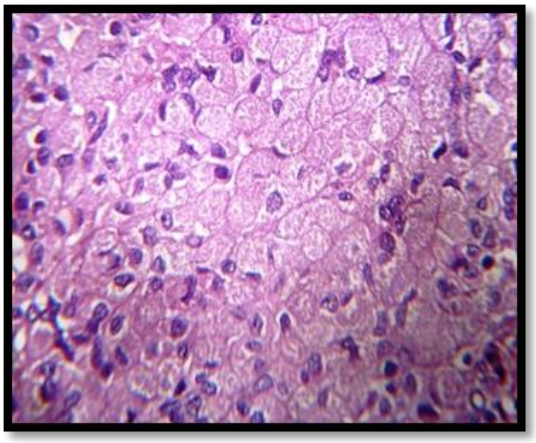


Figure 6: Showing large eosinophilic cells with granular cytoplasm in 40X magnification

The H & E section shows odontogenic epithelium in the connective tissue stroma in the form of islands and follicles. Peripherally the cells are tall columnar with nucleus showing reversal of polarity. The inner cells are polygonal in shape with nucleus placed at the center. Their cytoplasm is eosionophilc. The polygonal cell also shows granular cytoplasm throughout the section.

A diagnosis of granular ameloblastoma was made based on histological features.

DISCUSSION

Granular cell ameloblastoma is a rare variant of ameloblastoma accounted for 3.5% of ameloblastomas.[1, 2, 3] They resemble the more common follicular type.

Its age distribution is considered to be quite similar to the other types of ameloblastoma, for which an average median age of 35 years old, ranging from 4 to 92 years, is reported.[4] All tumours occurred in the mandible, the vast majority of which affected the posterior regions. Jaw swelling and pain were the most frequent presenting symptoms.[5,6]

This variant shows granular transformation of odontogenic epithelial cells. Microscopically these cells show cytoplasm filled with the granules.[6] Ultrastructural, histochemical and immunohistochemical studies have revealed that cytoplasmic granularity is caused by the

lysosomal overload. Other studies postulate that this cytoplasmic granularity might be caused due to increase apoptotic cell death of odontogenic epithelial cell and associated phagocytosis of the neighbouring cells. Some author considers it to be aggressive and recurrent.[7,8] Granular cell formation was thought to be an aging or degenerative change, but can be seen in ameloblastoma in young person

Immunohistochemical investigation proved that the granular cells are positive for cytokeratin, CD68, lysozyme and alpha-1-antichymotrypsin, but negative for vimentin, desmin, S-100 protein, neuron-specific enolase CD15, indicating epithelial origin and lysosomal aggregation.[8,9] Proliferative activity of granular cell ameloblastoma have been found to be greatest among the variants of ameloblastoma, accounting for it aggressive behaviour.[10]

The differential diagnosis of granular cell ameloblastomas includes other oral lesions with a similar morphology of granular cell accumulation, including granular cell odontogenic tumour, granular cell tumour and congenital epulis. These lesions have different biologic behaviour and should be separated from granular cell ameloblastomas.

In contrast to granular cell ameloblastoma, the granular cell odontogenic tumour are not located within epithelial islands but constitute part of the tumour stroma. Small islands or cords of the odontogenic epithelium may be seen interspersed among the granular cells, while cementum-like deposits and dystrophic calcifications are often found within the lesion.[10,11]

The granular cell tumour is an uncommon benign soft tissue neoplasm which generally occurs in the tongue followed by the buccal mucosa.[11] Differential diagnosis from a granular cell ameloblastoma is necessary. Granular cell tumour usually occurs in the fourth to sixth decades of life and shows a female predilection. Clinically, it appears as an asymptomatic sessile nodule of small size. Microscope examination reveals large polygonal cells with abundant, pale, eosinophilic granular cytoplasm, small vesicular nucleus, arranged into sheets or nests. The occasional presence of pseudoepitheliomatous hyperplasia of the overlying epithelium can result in misdiagnosis of squamous cell carcinoma.[10,11]

Congenital epulis is an uncommon soft tissue tumour which occurs almost exclusively on the alveolar ridges of newborns or rarely on the tongue. Although this lesion is also composed of granular cells, the necessity of distinction from a granular cell ameloblastoma appears unlikely, considering occurrence of the latter in patients of older age.[10]

TREATMENT PLAN

The types of treatment that have been include both radical and conservative surgical excision, curettage, chemical and electrocautery, radiation therapy or combination of surgery and radiation. Curettage is least desirable since it is associated with the highest incidence of recurrence. Treatment decision of ameloblastoma is based on individual patient situation and best judgement of the surgeon.

Since granular cell ameloblastoma is an aggressive tumour with high recurrence rate, radical surgery (hemimandiblectomy) was performed. Regular follow up the patient is done.

CONCLUSIONS

The granular cell ameloblastoma is a rare condition with unique histopathologic and immunohistochemical findings; its treatment and prognosis do not significantly differ from those of other subtypes of solid/multicystic ameloblastomas.

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