International Journal Dental and Medical Sciences Research
Volume 5, Issue 4, July-Aug 2023 pp 296-299 www.ijdmsrjournal.com ISSN: 2582-6018

Granular Cell Ameloblastoma–A Case Report

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Date of Submission: 20-07-2023 Date of Acceptance: 31-07-2023

ABSTRACT: Ameloblastoma is a slow-growing, locally aggressive neoplasm that originates within the mandible or maxilla from the epithelium involved in the formation of teeth. Ameloblastoma is classified into many variants histologically. Among them, Granular Cell Ameloblastoma is a rare entity, characterized by large eosinophilic granular cells. This article describes a case of Granular Cell Ameloblastoma in a 21-year-old male patient with clinical, radiological, and histological features along with a systemic review of the literature.

KEYWORDS: Ameloblastoma, Granular Cell Ameloblastoma

I. INTRODUCTION

Odontogenic tumors (OT) are a heterogeneous group of lesions of diverse clinical behavior and histopathologic types, ranging from hamartomatous lesions to malignancy. OT is derived from ectomesenchyme and/or epithelial tissues that constitute the tooth-forming apparatus. Like normal Odontogenisis, the Odontogenic tumors represent inductive interactions between Odontogenic mesenchyme and epithelium.[12]Ameloblastoma is a benign but locally aggressive epithelial neoplasm that is one of the most common Odontogenic tumors. Current genetic studies show mutations in genes that belong to the MAPK pathway in many Ameloblastomas. BRAFV600E is the most common mutation.[3]

The 5th edition of the World Health Organization (WHO) Classification of Head and Neck Tumors (2022) classified Ameloblastoma into Conventional, Unicystic, Peripheral, Metastasizing, and Adeno Ameloblastoma[4]. There are two basic histopathologic patterns in Solid/Multi-cystic Ameloblastoma:(1) follicular and(2) plexiform. Other microscopic patterns of Ameloblastoma include acanthomatous, basal cell-like,and granular cell. These patterns can be uniform or mixed. In different parts of the world, the distribution of Ameloblastoma varies to a certain extent.[5]Granular Cell Ameloblastoma is a term applied to a tumor, most often of the follicular type, shows an extensive granular transformation of the central stellate reticulum- like cells. In some lesions, all tumor islands or nest cells are composed of granular cells[6].

II. CASE REPORT

A 21-year-old male patient was referred to our Department with the complaint of swelling of the face of four months duration. The patient noticed a swelling on the right side of the mouth for four months which gradually increased in size and reached the present size. It was not associated with pain or paresthesia.

The patient's medical history and review of systems were unremarkable. On extraoral examination, no visible swelling was noted on the right side of the face. The over lying skin appears to be normal with no secondary changes noted. There was no tenderness noted on palpation. There was no cervical mass or lymphadenopathy found.

On intraoral examination, a swelling of size 2X3cm was noted over the right mandibular body region in relation to 44 to 47. The swelling was hard in consistency. It is non-tender on palpation, expansion of the lingual and buccal cortical plate of the right mandible was noted. No peripheral eggshell cracking was noticed. Grade I mobility noted on 44 to 46 and grade II mobility on 47.

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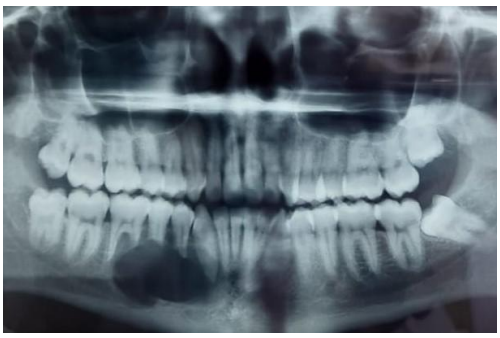

Figure.1: OPG showing a well-defined unilocular radiolucency extending from 46 to distal aspect of 43 with a well-corticated border. Resorption of roots noticed from 43 to46.

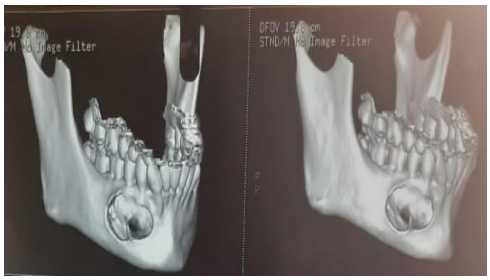

Figure 2:CBCT showing a unilocular expansile, lytic lesion in the mandible of size 34.6mm X 2.0mm X21.9mm extending from 43 to 46. Cortical thickening around the lesion can be seen.

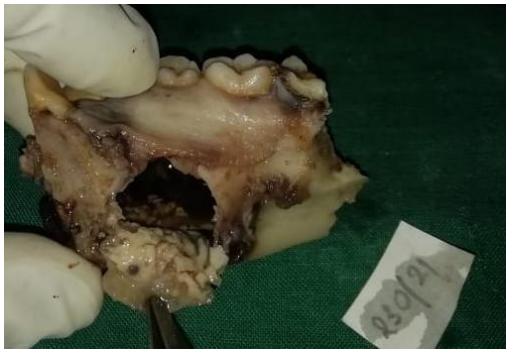

Figure.3:Gross specimen showing resected mandible from 42 to 47 with cystic degeneration

Histopathological examination revealed interlacing cystic spaces lined by columnar odontogenic epithelium in a moderately collagenous connective tissue stroma. Most of the cells in the stellate reticulum beneath have abundant deeply granular eosinophilic cytoplasm with eccentrically placed nuclei. A focal collection of inflammation is also noted within the connective tissue (Figure 4).

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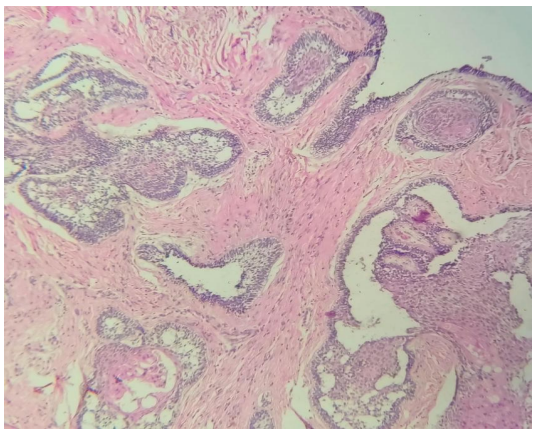

Figure.4:Microscopy showing interlacing cystic spaces lined by columnar ameloblastic epithelium in a moderately collagenous connective tissue stroma.

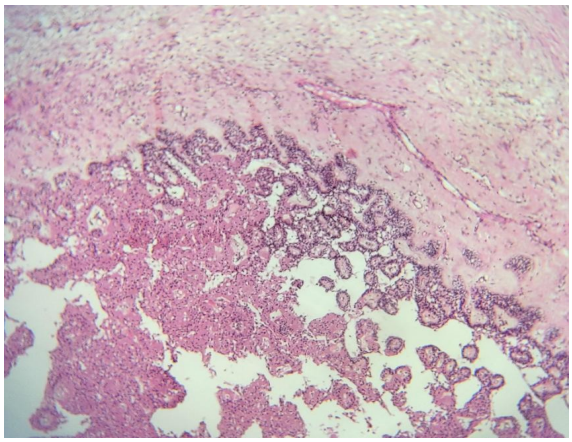

Figure.5: Most of the cells in the stellate reticulum beneath have abundant deeply granular eosinophilic cytoplasm

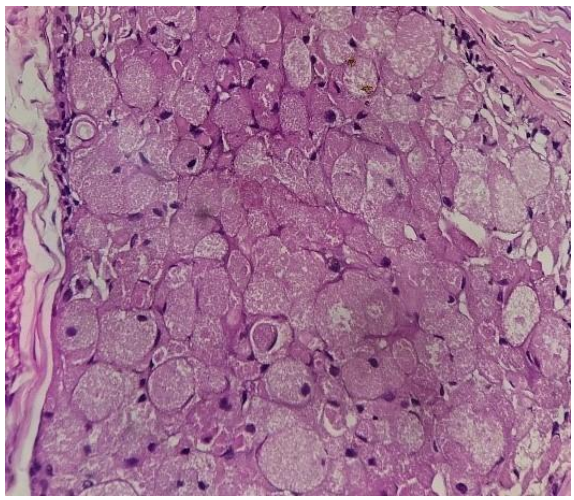

Figure6: Large ovoid cells with eosinophilic granular cytoplasm and eccentrically placed nuclei.

III. DISCUSSION

Granular Cell Ameloblastoma is a rare variant of Ameloblastoma. It accounts for about 5% of all Ameloblastomas.[7] Histopathologically, it is characterized by having numerous large eosinophilic granular cells. These cells usually form the central mass of the epithelial tumor islands and cords. Sometimes, even peripheral cells also exhibit similar features which are usually non-granular cells.[8] It was first seen by Krompecher in 1918 and was called pseudoxanthomatous cells.[9]

Granular cells can appear in various Odontogenic and Non Odontogenic tumors. Granular cells of Granular Cell Ameloblastoma are clearly of epithelial origin staining exclusively for cytokeratin. Granular cell tumors stain positive for vimentin and S-100. Only vimentin is positive in granular cells of Granular Ameloblastic fibroma. The term "Granular Cell Ameloblastic Fibroma" is a misnomer, as a number of these cases are probably central Odontogenic Fibromas exhibiting granular cell change. Ultrastructurally, it has been revealed that the lysosomal overload in these cells imparts characteristic granularity.

The granular appearance of the tumor cells is seen in various or altumors, such as Granular Cell Myoblastoma, Congenital Epulis, and Granular Cell Ameloblastic Fibromas. The morphology of granular cells is similar, but their origins are different. According to histogenesis, Granular Cell Ameloblastoma is epithelial, while others are of mesenchymal origin.[10]

Granular Cell Ameloblastoma is locally aggressive and has a relatively high rate of recurrence which was reported to be 33.3% and is higher as compared to that of follicular, plexiform, and acanthomatous subtypes.

IV. CONCLUSION

Granular Cell Ameloblastoma is a rare condition with unique histopathologic and immunohistochemical findings; its treatment and prognosis do not significantly differ from those of the other sub types of Solid/Multi-cystic Ameloblastoma. However, it should be differentiated from the other granular cell lesions primarily because of its higher recurrence rate. A better understanding of the molecular pathogenesis of Ameloblastoma will provide benefits in diagnosis and treatment.

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