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Case Report

Granular Cell Type of Ameloblastoma: A Case Report

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Abstract

Ameloblastoma is a locally invasive tumor derived from odontogenic epithelium. An uncommon variant of ameloblstoma is granular cell type, which cannot distinguish from other ameloblastoma subtypes by clinical and radiographic findings alone. Only review of its microscopic features allow, distinction from other subtype. The purpose of this article is to present a case of granular cell ameloblastoma. This subtype should be distinguishing from the other histopathologic subtypes because of its higher recurrence rate and more aggressive biological behavior. Radiographic and histologic findings as well as treatment are also discussed.

Keywords: Ameloblastoma; Granular Cell Change; Jaw Neoplasm; Lysosome; Mandibular Disease; Odontogenic Tumor

Introduction

Ameloblastoma is a locally invasive tumor derived from odontogenic epithelium [1]. Majority of patients present in the fourth decade [2]. Men are involved more than female [3] and More than 80% of Ameloblastomas are in the mandible (mostly angle and ramus) [4]. Clinically, jaw swelling and pain are the most frequent presenting symptoms [5]. Radiographically, Ameloblastoma is included solid (multicyctic) and unicystic [4,6,7]. Microscopically The follicular and plexiform patterns are the most frequent and less common histopathology subtypes include the acanthomathous, granular cell, desmoplastic and basal cell [5]. Granular cell Ameloblastoma is a rare subtype (less than 3/5%) [8]. It cannot be distinguished from other Ameloblastoma subtypes by clinical and radiographic findings alone [9] histopathology features of Granular cell type of Ameloblastoma is characterized by the groups of granular cells, which have abundant cytoplasm filled with Eosinophilic granules [5]. The granular cells usually form the central mass of the epithelial tumor islands and cords. The periphery of the islands consists of non-granular columnar cells [10]. Sometimes granular cells phenotype has been attributed to an aging or degenerative change in long-standing lesions [6]. But this tumor usually shows higher recurrence rate and more aggressive behavior which demand a close post operation follow up [10]. The purpose of this article is to present a case of granular cell Ameloblastoma and review its microscopic features that allow its distinction from other Ameloblastoma subtype.

Case Report

A 47-year-old male presented with a chief complaint of a painless swelling in his right mandible and mobility of lateral and canine teeth in same side. Swelling was begun from 3 years ago until 8 months before was reached to present size; mobility of teeth revealed from 3 months ago. There were no lymphadenopathy and tenderness (Figure 1).

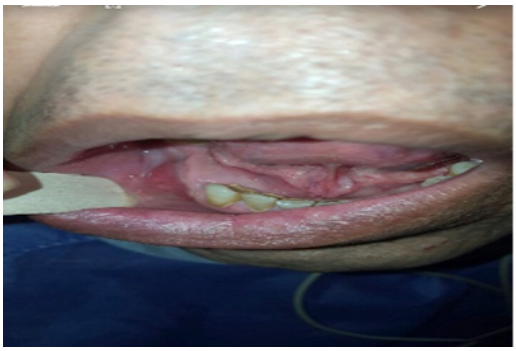

Figure 1: Patient with a swelling on the right mandiblular vestibule.

Panoramic radiograph showed a large, multilobular radiolucency with ill-defined borders, located in the body of partial endentulous right mandible and extending from the lateral to the first molar area (Figure 2).

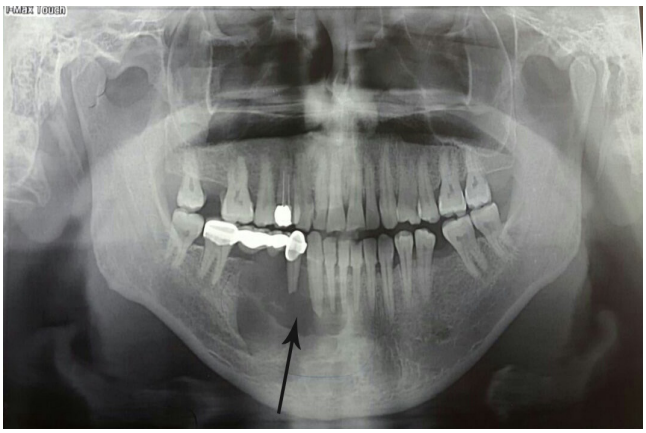

Figure 2: Panoramic radiograph showed a large, multilobular radiocency,located in the right mandible,with resorption of the canine and premolar teeth (arrow).

According to preoperative management of patient, routine biochemical and hematological investigations were done and all were within normal limits. With differential diagnosis of central giant cell granuloma or odontogenic tumors or any other centrally located mesenchymal tumors, the patient posted for surgery. Incisional biopsy was done but the resected tissue was found to be insufficient to arrive at a histopathological diagnosis. Based on the suggestion of the surgeon, incisional biopsy was not repeated. The patient posted for excisional biopsy. Under general anesthesia removing part of the jawbone including tumor with right lateral and canine teeth performed. In gross, tumor appeared as a combination of cystic and solid areas (Figure 3).

Figure 3: Photograph of surgical specimen appearing as a combination of cystic (black arrow) and solid areas (white arrow).

Histopathology survey of surgical specimen revealed combination of cystic and solid areas. The peripheral layer of cystic areas consisted of a parallel arrangement of tall cylindrical cells with reverse polarity (Figure 4a, white arrow) of their hyper chromatic nuclei and vacuolization of the cytoplasm and in solid area the accumulations of cell rich in Eosinophilic granular cytoplasm were found (Figures 4b,4c, black arrows).

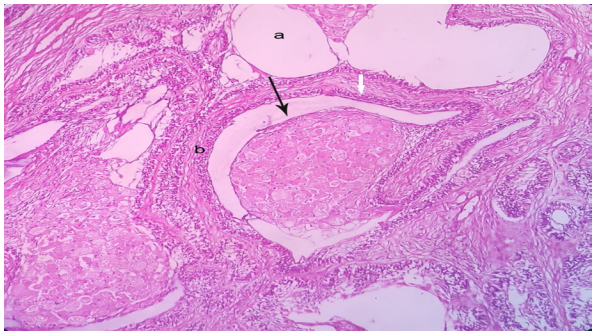

Figure 4a: Hematoxylin and eosin stain, original magnification ×40.

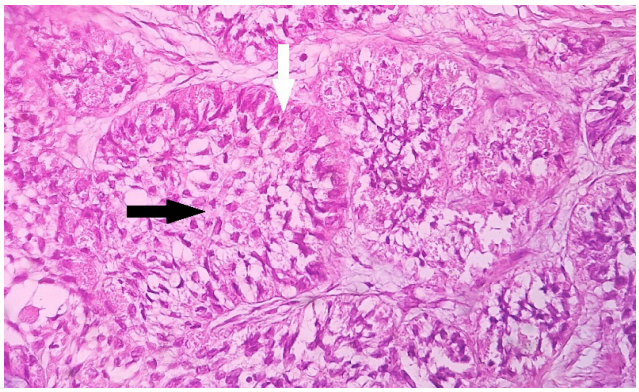

Figure 4b: Hematoxylin and eosin stain, original magnification ×400.

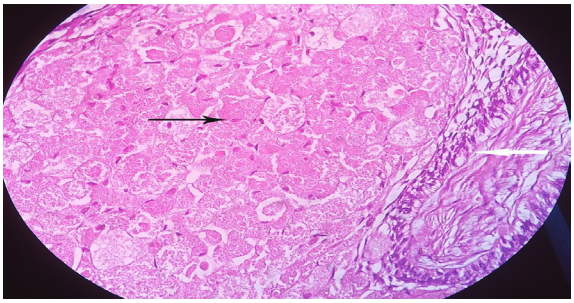

Figure 4c: H and eosin stain, original magnification ×400.

(Figures 4a, b, c) Photomicrographs showing tumor appears in cystic(a) and islands(b) with a peripheral layer of ameloblastsle (white arrow) and extensive central areas of granular cells with large, eosinophilic cytoplasmic granules (black arrow).

Also in the periphery of solid part of islands and cords of epithelium a row of cell similar to Ameloblast were found. According to above findings diagnosis of Granular cell Ameloblastoma was given. The segmental resection removing part of the jawbone including tumor with right lateral and canine teeth performed. After2 month the radiography which was taken showed acceptable healing improvement and patient schedule for follow up in six months' interval (Figures 5a,5b).

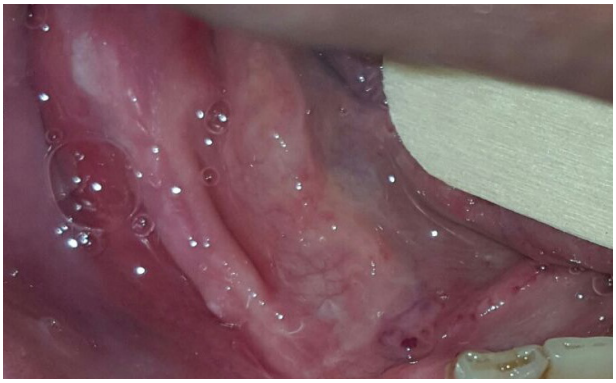

Figure 5a: Intra oral view which show normal healing after2 month.

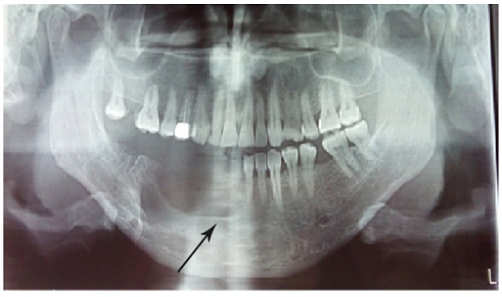

Figure 5b: Fallow up after2 month which show sclerotic bone formation at the base of the surgical area Arrow).

Discussion

The age distribution of granular cell variant is similar to the other types of Ameloblastomas which shows an approximately equal prevalence in the third to seventh decade of life [5], about 85% of tumors occurred in the mandible, the vast majority of which affected the molar–ramus region [5]. Jaw swelling and pain were the most frequent presenting symptoms. Compared to the other Ameloblastoma subtypes, no distinguishing radiographic findings have been reported [8]; the patient in this study was completely matched to above finding. In reveiw of litreature and case report which was done by Shelly Arora et.al., similar clinical and histopathological features with our case could be found [8]. Histopathologically GCA has numerous large eosinophilic granular cells. These cells usually form the central mass of the epithelial tumor islands and cords.

The periphery of the islands consists of non-granular tall columnar cells. Granular cell Ameloblastoma is diagnosed by the presence of granular cells, which usually occur within the central area of tumor and progressively replace the stellate reticulum [11]. Our case also showed similar features. It is evident from the literature, there exist two main lines of interpretation about nature of granular cells, some consider it as a metabolic, whilst others of the view that it represents a degenerative process. More recent observation supports the later view to be more tenable based on the increased expression of death signaling molecules. Ara et.al., suggested that the synthesis of signaling molecules, such as β-catenin and Wnt-5a is upregulated in the granular cells of GCA, but their transportation or secretion is impaired, resulting their accumulation within granular cells, as auto phagosomes [12]. The granular cell Ameloblastoma has a more aggressive behavior compare with the other histologic subtypes; it may be locally aggressive and has relatively higher recurrence rate, [9] Unlike the case reported in this article, despite of curettage with peripheral osteotomy which was done, after two months, radiography showed acceptable healing improvement. But we need an extended period of follow up in this patient for better judgement.

Ultra-structurally it has been revealed that the lysosome accumulation in these cells provide the characteristic granularity [5]. The differential diagnosis of granular cell ameloblastomas includes other oral lesion with a similar morphology of granular cell accumulation such as granular cell tumor, granular cell odontogenic tumor and congenital Epulis but these lesions usually could differentiate easily [5]. Treatment of Ameloblastomas should be based on patient's history, clinical, radiographic examination, and finally histopathology findings [13,14]. However, similar to the other types of solid Ameloblastoma, the prognosis is more dependent on the surgical procedures, i.e. granular cell Ameloblastomas treated by enucleation or curettage exhibit a high recurrence rate [15]. Surgical options include segmental resection, en-block resection, simple curettage and excision with peripheral osteotomy [13, 14]. The last one which was done for our patient and after

2 months clinically (figure 5a) and radiography which was taken (figure 5b) showed acceptable healing improvement and patient schedule for follow up in six months' interval.

Conclusion

Granular cell Ameloblastomas is a rare condition with unique histopathology findings; this subtype should be distinguished from the other histologic subtypes because of its higher recurrence rate and more aggressive behavior and necessity of long period of follow up.

Conflict of Interest

The authors disclose no potential conflicts of interest.

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