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Case Report

GRANULAR CELL VARIANT OF AMELOBLASTOMA- A UNIQUE AGGRESSIVE TUMOR

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A B S T R A C T

Ameloblastoma is the most significant epithelial odontogenic tumor of concern for the oral pathologist because of its high incidence among all odontogenic tumors and true neoplastic (infiltrative and recurrent) potential, combined with its diversity of histologic patterns. Six histopathologic subtypes of ameloblastoma are recognized, amongst which granular cell ameloblastomas are the most uncommon lesions accounting for about 3–5%. It appears to be aggressive in nature, with a marked propensity for recurrence and can show metastasis. It exhibits diverse microscopic patterns, which occur either independently or in combination with other patterns.

Hereby we are presenting a rare aggressive case of granular cell ameloblastoma in the left retromolar region of mandible affecting a 38-year old male.

INTRODUCTION

Odontogenic tumors comprises of a group of heterogeneous lesions that range from hamartomatous or non-neoplastic tissue proliferations to benign or malignant neoplasms with varying degrees of aggressiveness and metastatic potential.[1] These lesions are derived from epithelial, retromolar mesenchymal remnants of the tooth-forming apparatus.[2] Ameloblastoma is the second most common odontogenic tumor, being clinically and histologically diverse. The origin of tumor derived from: cell rests of enamel organ, epithelium of odontogenic cysts, disturbances of developing enamel organ, basal cells of surface epithelium, or heterotrophic epithelium in other parts of body.[3]

Under the current World Health Organization (WHO) classification of odontogenic tumors, ameloblastomacan be divided into: solid/multicystic, extra osseous/peripheral, desmoplastic, and unicystic types.[4] Conventional solid type histogically categorized into follicular, plexiform, acanthomatous, granular cell, desmoplastic and basal cell ameloblastoma.[5] Granular cell ameloblastoma accounts for about 3-5% of all histologic subtypes marking the rare occurrence. A unique microscopically picture is characterized by marked transformation of the cytoplasm of the tumor cells to coarse granular eosinophilic appearance.

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These granular cell changes seen usually in stellate reticulum like cells may extend to include peripheral columnar or cuboidal cells as well. This phenomenon can lead to degeneration or formation of cystic areas [6] or a dysfunctional status of neoplastic cells or an indicator of a more aggressive course.[5]

This report describes granular cell ameloblastoma that is a relatively rare histologic subtype and aggressive lesion with marked proclivity for recurrence.

Case Report

A 38years old male patient presented with an ulcer in the left lower back tooth region, which persisted since 2months. He also gave a history of swelling associated with an impacted tooth, 10-12yrs back, which was treated for the same. No much details about the past procedure. On examination, facial asymmetry was noticed on the left side of the face extra orally.Swelling was soft to firm in consistency. (Fig.1) There was no associated pain, no facial paralysis, no paresthesia or anesthesia and no palpable regional lymphadenopathy. Intraoral examination revealed an ulcer in the left retro molar region measuring 2x2cm.

Routine biochemical investigations were within normal limits. OPG revealed a multilocular radiolucency seen on the lower left mandibular region, which was extending from molar region anteriorly until the coronoid process posteriorly, measuring approximately 2x2cm in diameter. The margins showed irregularity with sclerotic borders. (Fig. 2)

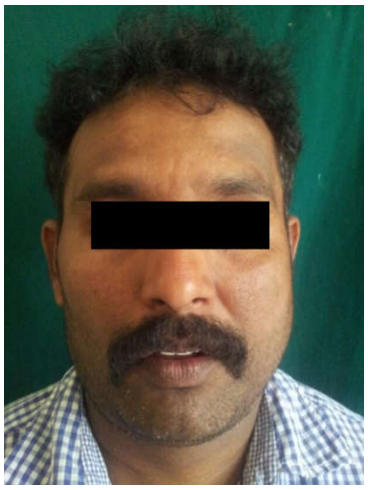

Figure 1 Facial asymmetry was noticed on the left side

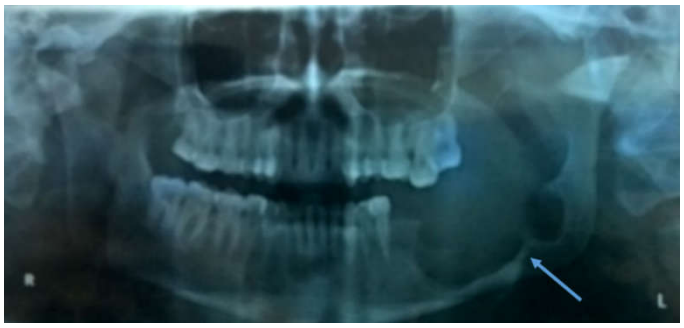

Figure 2 OPG revealing a multilocular radiolucency

3D Reconstruction of CT Images revealed bony destruction in the superior border of the body of the mandible extending up to anterior border of the ramus. The lower border of the body of the mandible is intact. (Fig.3)

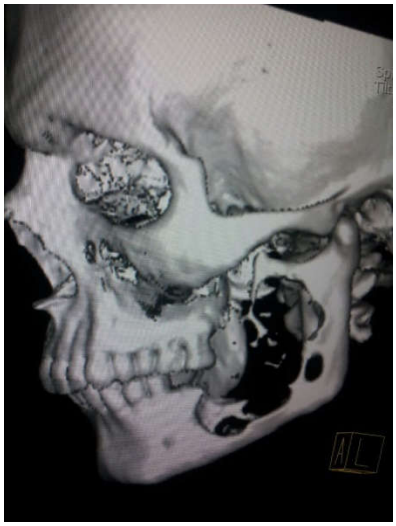

Figure 3:3 D Reconstruction of CT Images revealed bony destruction in the superior border of the body of the mandible.

A provisional diagnosis of ameloblastoma was proposed. An incisional biopsy involving the ulcer was taken. Macroscopically, the specimen was creamy white with cheese like material on the surface having irregular borders measuring 1.5x1.2cm. (Fig.4)

Microscopically the H &E stained section displays epithelial islands in the connective tissue stroma. The epithelial nests consisted of centrally arranged angular cells resembling stellate reticulum. At the periphery, a single layer of tall columnar ameloblast like cells with reversal of polarity was seen. In some regions, the ameloblast like columnar cells were arranged in anastomosing cords. (Fig.5) Regions showing transformation of groups of lesional cells to granular cells were also observed with the cells having abundant cytoplasm filled with eosinophilic granules. (Fig.6)

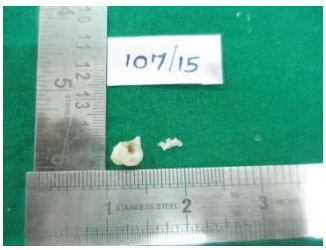

Figure 4 Incisional Biopsy Specimen

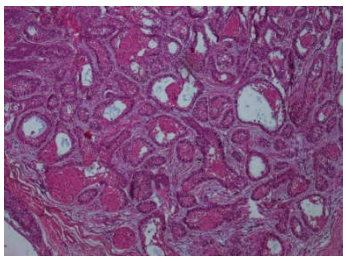

Figure 5 Histological section in 10X

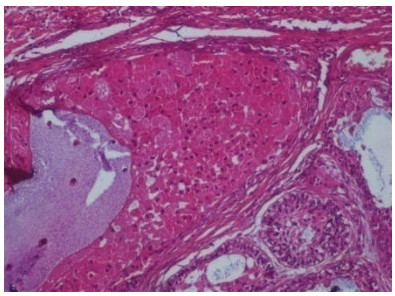

Figure 6 Histological section in 40X

On correlating the clinical and radiographic findings with the histopathological appearance, the final picture was suggestive of 'Granular Cell Ameloblastoma'.

The case was managed by resecting the tumor mass, sparing the condyle until the angle of the mandible and reconstruction with free fibula graft under general anesthesia. (Fig.7) Post operatively the healing was uneventful.The patient has been followed up regularly, and free of disease until date. (Fig.8 and 9)

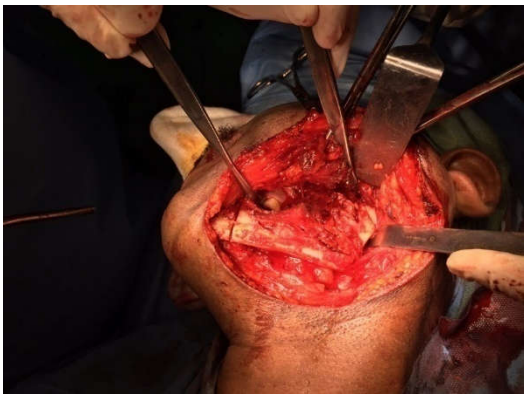

Figure 7 Intraoperative image depicting resection and reconstruction of mandible.

Figure 8 Post operative intraoperative image

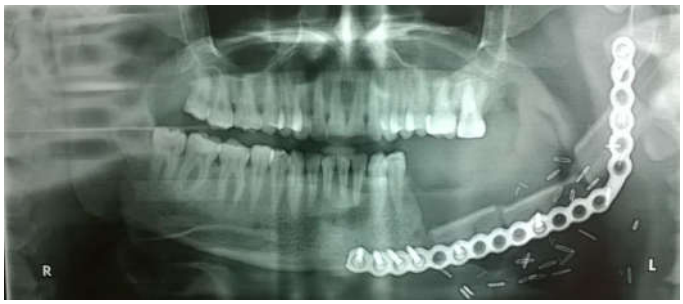

Figure 9 Post operative OPG.

DISCUSSION

Ameloblastoma, solid/multicystic variant (SMA) has been known for about 180 years.[7] Ameloblastoma can be histopathologically characterized by the presence of peripheral columnar cells with hyperchromatic, reversely polarized nuclei, arranged in a palisaded pattern[8]. Approximately 30 cases have been reported in the English literature search involving this rare subtype of ameloblastoma. Hartman conducted a study that included, only 1-4 cases of granular cell ameloblastoma, indicating the rare histological entiity[9]. In a review on available literature on ameloblastoma of the jaws from 1960 to 1993, Reichart *et al*. reported only 56 (3.5%) cases of the granular cell variant among 1593 cases[10]. Granular cell ameloblastomas accounted only for 5% out of 408 cases of ameloblastomas retrieved from the files of the Armed Forces Institute of Pathology[9]. In a study of 20 cases, Hartman reported that the average age of the patients was 40.7 years and that there was no gender predilection[4]. The age distribution of granular cell variant shows an approximately equal prevalence in the third to seventh decade of life that is similar for all subtypes of ameloblastomas[3]. About 85% of tumors, the site of occurrence is mandible, with vast majority being the molar–ramus region[2]. The granular transformation of tumor cells was thought to be a degenerative change. The ultrastructural and histochemical studies indicate that, accumulation of lysosomes gives granular appearance of cytoplasm[9]. The biological behavior of granular cell ameloblastoma involves locally aggressiveness and a relatively high chance of recurrence[11]. Reichart *et al*. reported a higher recurrence rate (33.3%) for granular cell ameloblastoma, when compared to the more common subtypes of ameloblastoma[12]. However, granular cell ameloblastomas exhibit a high recurrence rate when treated by enucleation or curettage because the border of the tumor within cancellous bone lies beyond the apparent macroscopic surface and the radiographic boundaries of the lesion. Therefore, radical surgical methods are mandatory[11].

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