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An Exceptional Case of Expedited Recurrence and Progression of Granular Cell Ameloblastoma: Extension to the Temporal Bone

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ABSTRACT

Odontogenic tumors are any abnormal growth in and around the mandible and teeth. Ameloblastoma is an aggressive odontogenic tumor of the mandible. Its extensive tumefaction accords to a high malignant transformation potential. Asymptomatic slow growth of the affected area is a consensus in the literature for the manifestation of ameloblastoma which is consistent. It comprises nearly 1% of all oral tumors, 13-58% of all odontogenic tumors, and is far more typical indeveloping countries with an elevated risk of recurrence. Granular cell ameloblastoma (GCA) is a less prevalent histological subtype of ameloblastoma with a pronounced transformation of the cytoplasm of stellate reticulum like cells into a rough, granular eosinophilic appearance. This article reports a case of GCA involving the posterior part of the right mandible region in a 37-year-old female patient which even after surgical resection recurrence and spread to the frontal bone.

Keywords: *Aggressive, Ameloblastoma, Granular cell ameloblastoma, Recurrence*

1. INTRODUCTION

Ameloblastoma is a steady growing odontogenic epithelial tumor of the mandible. It is well identified as a localized invasive benign neoplasm perceived to arise from the cellular components of the enamel organ [1]. Granular cell ameloblastomas (GCA) are unusual lesions, a rare infirmity, accounting for 3-5% of all ameloblastoma cases. Granular cells are an intermediate or matured stage in the life cycle of ameloblastomas, starting with normal stellate reticulum-like cells, accompanying a production of granules, eventually leading to degeneration and initiation of cystic areas [2].

Granular cells are of epithelial origin, and distinct ultrastructural and histochemical studies have cataloged them as lysosomes [3]. Here, we present a rare case of granular cell ameloblastoma in a 37-year-old female highlighting its unique microscopic features.

2. CASEREPORT

A 37 - year - old female patient reported to our department with a chief complaint of gradually increasing mass in the right mandibular posterior region since 1 month. Swelling was associated with pain and difficulty in chewing. The patient gave a past history of surgical removal of the ameloblastoma (marsupialization; cystic/pathological lining was removed) involving the same mandibular posterior region. In 2019, patient's medical, social, and family history was non-contributory. Extra-oral examination showed slight facial asymmetry with right mandibular deviation. No evidence of draining sinus, pus discharge, or cervical lymphadenopathy was observed. The swelling was tender on percussion, near the angle and body of mandible. Intra-orally, a 4 cm× 2 cm solitary, nodular growth in the right mandibular edentulous posterior region was observed extending from 46 to the ramus.The overlying mucosa was erythematous and fissured. On palpation, the growth was non-tender, sessile, and firm in consistency. Based on the clinical examination, differential diagnosis of reactive lesions like pyogenic granuloma, peripheral giant cell lesion was considered. Keeping previous history of surgical removal of the ameloblastoma in mind, consideration to recurrent ameloblastoma was also kept in mind. Radiographic examination revealed a multilocular radiolucency [Figure1]. Multiple punched out lesions seen in mandible extending to the initial portion of temporal bone and the whole of zygomatic bone [Figure 2].The previous occurrence of ameloblastoma in the patient involved just the mandible but recent OPG revealed much distant recurrence with spread to the zygomatic and temporal bone.

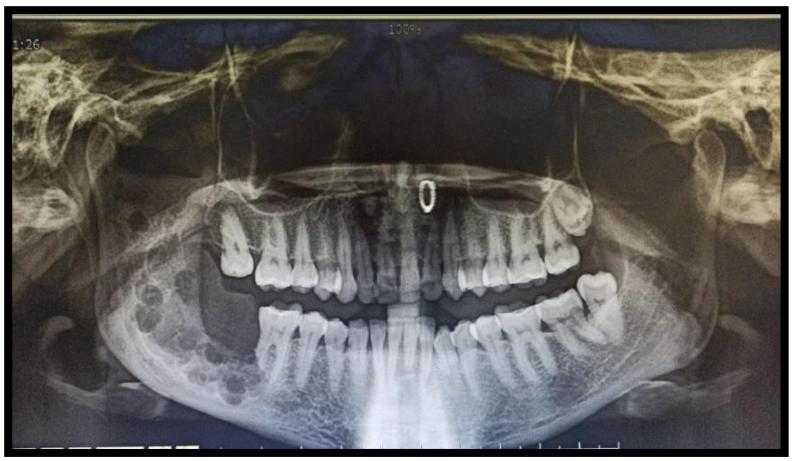

Figure 1- Multilocular radiolucency in the right mandibular posterior region

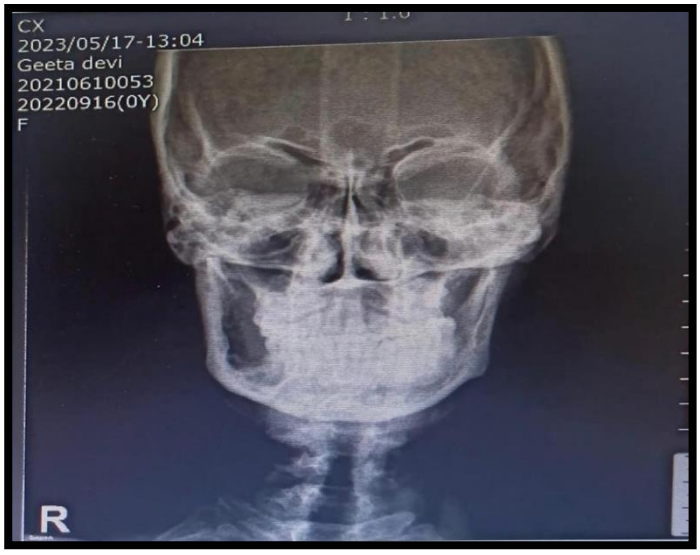

Figure 2: Multiple punched out lesions seen in mandible extending to the initialportionoftemporalboneandthewholeofzygomaticbone

Microscopic examination of the Haematoxylin and Eosin stained sections showed fibro-cellular connective tissue stroma infiltratedwith islands and chords of tumor follicles showing peripheral cellswhich are tall columnar with reversal of polarity

of hyperchromaticnuclei and vacuolization of the cytoplasm [Figure 3].The central cells showed a marked transformation of the cytoplasm into course granular eosinophilic appearance [Figure 4].The intervening connective tissue is infiltrated with inflammatory cells and blood vessels of varying shape and size. The final diagnosis of GCA was rendered.

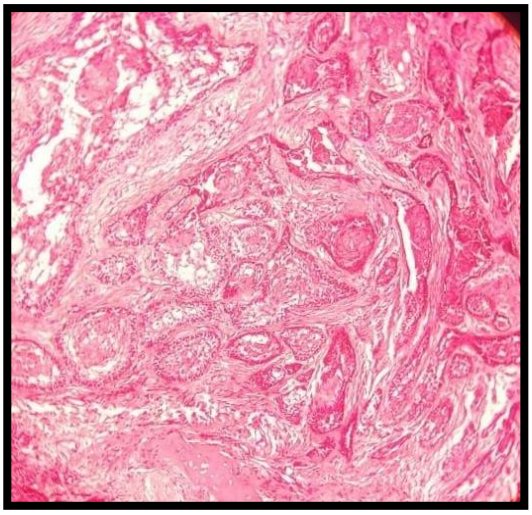

Figure 3: Histopathology (10X) shows fibro-cellular connective tissue stroma infiltrated with islands and chords of tumor follicles

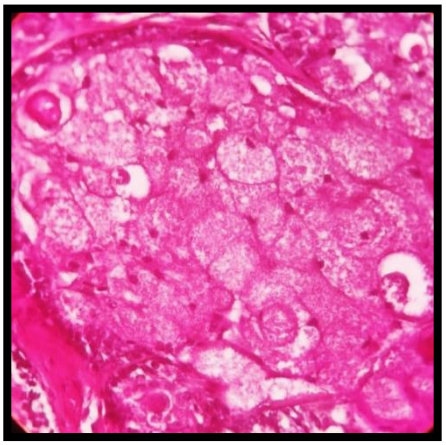

Figure 4: Histopathology (40X) shows the central cells with a marked transformation of the cytoplasm into course granular eosinophilic appearance

3. DISCUSSION

Ameloblastoma is a locally invasive neoplasm stemming from odontogenic epithelium. The tumor is composed of proliferating odontogenic epithelium, notably of enamel organ-like tissue that has not sustained differentiation to the point of hard tissue formation. Peculiarly, the tumor is devoid of enamel and dentin. The GCA is an odd version, accounting for just 5% of all ameloblastomas and found as a combination of other histologic patterns, primarily the follicular variant [4, 5]. It was originally appreciated by Krompecher in 1918 and was called pseudo-xanthomatous cells. Whether granular cell change in ameloblastoma is a degenerative progression or a prelude to a more aggressive direction is a matter of debate [6].

It seems to be aggressive in its attributes, with a marked inclination for recurrence and metastasis. Considerable interest has been shown with respect to the nature of granular cells in ameloblastoma since it was initially described. Studies report that these granular cells are derived from the odontogenic epithelium, rather than the connective tissue, as they majorly exhibit cytokeratins [7]. Microscopically, GCA is distinguished by the clusters of granular cells, which have profuse cytoplasm occupied with eosinophilic granules that resemble lysosomes, both ultrastructurally and histochemically. Granular cells, which prominently occur inside the central area of the tumor, increasingly replace the stellate reticulum [8].

Initially, they were said to symbolize an aging or degenerative process, but recent immunohistochemical studies show that this occurrence is due to increased apoptotic cell death of the cells of the lesion and the phagocytosis by adjacent neoplastic

cells. Immunohistochemical investigation is positive for S-100 protein, which may indicate neural origin. The incidental existence of pseudo-epitheliomatous hyperplasia in the overlying epithelium, which can induce a misdiagnosis of squamous cell carcinoma, is influential. The basement membrane corresponds to the barrier between the adjacent tissues, which renders mechanical support and symbolizes an important role in the progression of the disease by regulating cell migration, adhesion, and proliferation. Proteins from the basement membrane have been shown to be expressed in a variety of odontogenic tumors and may define contrasts between their biological nature [9].

The phenomenon of a GCA as a peripheral growth 2 years after the first surgery backs the hypothesis that the basal cell layer of oral mucosa could be the sought-out transgressor for recurrence in our case. The differential diagnosis of GCA covers granular cell odontogenic tumor, granular cell tumor, and congenital epulis, all of which have different clinical appearances and biological behaviors [10].

4. CONCLUSION

Although the morphology of all granular cells is comparable, the tissue of origin is unique to each condition. However, it should be distinguished from the other granular cell lesions especially due to its higher recurrence rate. An advanced comprehension of the molecular etiopathogenesis of ameloblastoma and its variants is in progress and may provide diagnostic and treatment advantages.

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