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Granular cell ameloblastoma in a case of recurrent follicular ameloblastoma

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Abstract: *Ameloblastoma is an epithelial odontogenic tumor which is locally aggressive and has tendency to recur. Granular cell ameloblastoma is a histological variant of ameloblastoma which occurs along with other histological variants and is characterised by presence of granular cells. It is usually seen in long-standing or recurrent cases of ameloblastoma. We report a case of granular cell ameloblastoma in a 34-year-old female patient who had been previously treated surgically for follicular ameloblastoma.*
Key words: *Granular cell ameloblastoma, recurrent ameloblastom*a

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I. Introduction:

Ameloblastoma is an epithelial odontogenic tumor which is defined as an unicentric, non functional, intermittent in growth and anatomically benign and clinically persistent tumor. Clinically it can manifest as solid and cystic variants [1]. It also has many histological variants of which, Granular cell ameloblastoma (GCA) is a relatively rare and is often found to occur along with other histological subtypes. GCA exhibits either follicular or plexiform pattern where the central stellate reticulum-like cells are replaced by granular cells [2].

Usually, granular cell change is usually found to occur in long standing or recurrent cases of ameloblastoma and the time of diagnosis is 8 years more than the mean age of ameloblastoma (36 years) [3,4]. We report a case of granular cell ameloblastoma in a 34-year-old female patient recurring from follicular ameloblastoma which was initially diagnosed 15 years back.

II. Case report:

A 34-year-old female patient presented with chief complaint of pain and swelling in the right side of face for the past one-and-half years. Her past surgical history revealed that she was surgically treated for follicular ameloblastoma fifteen years back which recurred after eight years following which segmental resection of the mandible was done for the same. Extraoral examination revealed a diffuse swelling of 4 x 5 cms in size on the right side of body of mandible which was tender on palpation and firm in consistency. Intraoral examination revealed a solitary, tender and firm swelling extending from 42 region to the retromolar area, obliterating the buccal vestibule (Fig 1 a & b). OPG revealed a soft tissue shadow in the lower jaw in the excised part of the mandible. The computerised tomographic images revealed a well circumscribed isointense mass in the mandibular body and ramus region and mild erosion of adjacent bone anteriorly.(Fig 2 a & b)

Correlating the previous history and the clinical findings, a provisional diagnosis of recurrent ameloblastoma was made. The lesion was excised in toto and the specimen was sent for histopathological examination.

Microscopic examination of H&E stained sections showed follicles of odontogenic epithelium with peripheral tall columnar ameloblast-like-cells showing pallisading of basal cells with hyperchromatic nuclei and central stellate reticulum like cells. In most of the follicles the central stellate reticulum-like cells are replaced by eosinophilic granular cells. The granular cells possessed small pyknotic nucleus which was peripherally displaced and prominent eosinophilic granular cytoplasm. The cell boundaries were distinct. The surrounding

stroma was moderately fibrocellular (Fig 3 a,b,c & d). Based on the above histopathological features, the final diagnosis of **granular cell ameloblastoma** was made.

III. Discussion:

As mentioned earlier, Granular cell ameloblastoma is a relatively rare histological variant. The clinical features in terms of location, signs and symptoms are similar to other types of ameloblastoma [2]. But the age of occurence is usually 8 years greater than that of conventional ameloblastoma. However, this is highly subjective to the time of diagnosis, as they are common in long standing and recurring cases [5]. Granular cell ameloblastoma commonly occurs in solid variants of ameloblastoma but occasional reports in cystic variants have also been encountered [6].

GCA is considered to be locally aggressive like other variants of ameloblastoma with comparitively higher incidence of recurrence [4]. However, the opinion varies as it is highly subjective to the type of surgical treatment provided [7]. However, there are incidences of metastasis recorded in literature [8].

Microscopically, granular cells are ovoid to polyhedral in outline with nucleus displaced to the periphery. The granules are coarse and distend the cytoplasm. The cell borders are usually distinctly demarcated. The granular cells can be demarcated from the deeply basophilic peripheral columnar cells by their weak eosinophilic staining, which is less pronounced than that seen in acanthomatous ameloblastoma [2]. The histopathological diagnosis of GCA can be made only when granular cell change is a prominent feature involving most of the follicles and not as a focal change as in some cases of follicular ameloblastoma [9].

The granular appearance has been ascribed to numerous lysosomes based on histochemical and electron microscopic findings. Ultrastructurally, the osmiophilic internal structure of the lysosomes varies considerably. Many of these granules are about 1 μm in size, but giant granules of 30 μm in diameter are rarely seen. The internal structure is composed of finger-print-like membranous structures, myelin figures, small particles, granules, vesicles, lattice structures, crystalloids and sometimes empty depending on the stage of digestion of lysosomal content [5].

The granular cell change is usually related to age-related or degenerative changes due to increased lysosomal activity. It is theorised that as the age of the tumor cells increases, unnecessary or aged components in the cytoplasm of some tumour cells become increasingly more numerous, but the ability of lysosomes to dispose these materials decreases with age. Hence, the cytoplasm of these cells contain numerous lysosomal granules [5]. Also the immunohistochemical and ultrastructural studies conducted by Kumamato et al demonstrate apoptotic cell death initiated by lysosomal granules followed by phagocytosis of apoptotic fragments by adjacent neoplastic granular cells in GCA [3].

The Granular cells in GCA are similar to the granular cells in other oral granular cell tumors like congenital epulis of new born, granular cell myoblastoma, granular cell leiyomyoma, oncoyctoma,etc. But most of these tumors are of mesenchymal origin (exception oncocytoma) whereas granular cells of GCA originates from odontogenic epithelium [9]. Another odontogenic tumor which has granular cell similar to GCA is Granular cell odontogenic tumor. But the granular cells are found within the fibrous stroma unlike GCA, with few strands of odontogenic epithelium interspersed between them [7].

GCA can be differentiated from other oral granular cell lesions by special stains and immunohistochemistry. All the granular cells of GCA shows positivity for PAS and stain intensively for acid phosphatase, a lysosomal enzyme, thus indicating aging or degenerative process. These lesions show histopathological resemblance to oncocytoma, when the entire island, including the peripheral cells are composed of granular cells. But can be differentiated from GCA by PTAH staining, as the granules in oncocytoma represent mitochondria, whereas the granules of GCA are lysosomal [1,5].

Immunohistochemically, granular cells showed positive immunoreactivity for anticytokeratin antibodies and negativity to vimentin, desmin and s-100, indicating its epithelial origin and thus can be differentiated from other granular cell lesions of oral cavity which are mesenchymal in origin [9]. The granular cells also show CD68, α- antichymotrypsin positivity, indicating lysosomal aggregations and positive reaction to anti-ssDNA antibody, thus indicating the presence of apoptotic bodies [3].

IV. Conclusion

Granular cell ameloblastoma has unique histopathological, electron microscopical and immunohistochemical findings. Although the behavior is similar to other types, it has higher incidence of recurrence and hence should be distingushed from other variants of ameloblastoma, In our case, the lesion was well circumscribed and was completely excised and bone margins were trimmed. The patient showed no signs of recurrence after one year and hence no further treatment was required.

References:

[1]. Rajendran R, ed (2009). Shafer's Textbook of Oral Pathology, ed 6, pp. 254–262, Elsevier, Noida, India.
[2]. Kessler HP (2004). Intraosseous ameloblastoma. Oral Maxillofacial Surg Clin N Am 16:309-322.
[3]. Kumamoto H, Ooya K (2001). Immunohistochemical and ultrastructural investigation of cell death in granular cell ameloblastoma. J Oral Pathol Med 30:245-250.
[4]. Reichart PA, Philipsen HP, Sonner S (1995). Ameloblastoma: biological profile of 3677 cases. Eur J Cancer B Oral Oncol 31B(2):86-99.
[5]. Nasu M, Takagi M, Yamamato H (1984). Ultrastructural and histochemical studies of granular cell ameloblastoma. J Oral Pathol 1984;13: 448-456.
[6]. Thillaikarasi R, Balaji J, Gupta B, Ilayarja V, Vani NV, Vidula B, Saravanan B, Ponniah I (2010). Cystic granular cell ameloblastoma. J Maxillofac Oral Surg 9(3):310-313.
[7]. Nikitakis NG, Tzerbos F, Triantafyllou K, Papadimas C, Sklavounou A (2010). Granular Cell Ameloblastoma: an unusual histological subtype report and review of Literature. J Oral Maxillofac Res 1:e3
[8]. Takahashi K, Kitajima T, Lee M, Iwasaki N, Inoue S, Matsui N, Ohki K, Nagao K, Akikusa B, Matsuzaki O (1985) Granular cell ameloblastoma of the mandible with metastasis to the third thoracic vertebra. A case report. Clin Orthop Relat Res 197:171-80.
[9]. Lo Muzio L, Mignogna MD, Staibano S, De Rosa G (1996) Granular cell ameloblastoma: A case report with histochemical findings. Oral Oncol, Eur J Cancer 3:210-212.

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Legends:

[1]. Fig 1 : a. Extraoral clinical photograph showing a diffuse swelling on right side of body of the mandible. b. Intraoral clinical photograph showing swelling extending from 42 region to the retromolar area, obliterating the buccal vestibule.
[2]. Fig 2 : a. Contrast enhanced Axial CT image showing a well circumscribed isointense mass. b. Contrast enhanced coronal CT image showing well circumscribed isointense mass
[3]. Fig 3 : a. Histopathological photomicrograph showing follicles of odontogenic epithelium with peripheral tall columnar cells and central stellate cells replaced by granular cells (x 40X). b. Histopathological image showing large follicles of ameloblastoma with peripheral ameloblast-like cells showing pallisading and granular cells occupying the whole follicle. (x 100X) c. A small follicle of ameloblastoma where stellate cells are replaced by granular cells in the center (x 400X) d. High power magnification of granular cells showing peripherally displaced nucleus, granular cytoplasm and distinct cell borders (x 400X).