[Downloaded free from http://www.jpbsonline.org on Thursday, May 05, 2016, IP: 223.236.174.99]

A case report of granular cell ameloblastoma associated with aneurysmal bone cyst like features

Sriram Kaliamoorthy, Pazhani Ambiga[1], Shivakumar Sivaraman[1], Kanmani Shanmugasundaram[2], Mahendirakumar Nagarajan[3]

Department of Oral and Maxillofacial Pathology, Chettinad Dental College and Research Institute, Chettinad Health City, Kelambakkam, Kancheepuram, [1]Department of Oral Medicine and Radiology, Vivekananda Womens Dental College and Hospital, Thiruchencode, Namakkal, [2]Department of Oral Medicine and Radiology, SRM Dental College and Hospital, Ramapuram, Chennai, [3]Department of Prosthodontics, Crown and Bridge and Implantology, Raja Muthiah Dental College and Hospital, Annamalai University, Chidambaram, Tamil Nadu, India

Address for correspondence:
Dr. Sriram Kaliamoorthy,
E-mail: ksrirammds@gmail.com

Received : 28-04-15
Review completed : 28-04-15
Accepted : 22-05-15

ABSTRACT

Granular cell ameloblastoma is a rare variant of ameloblastoma, which is histopathologically characterized by the presence of large eosinophilic granular cells within the ameloblastic follicle. Its accurate preoperative diagnosis is based upon clinical, radiological, and incisional biopsy findings. This article reports a case of granular cell ameloblastoma in a 65-year-old female, which on incisional biopsy showed the features suggestive of aneurysmal bone cyst. Furthermore, the influence of macroscopic presentation of the current lesion on its accurate preoperative incisional biopsy diagnosis is discussed in detail.

KEY WORDS: Ameloblastoma, diagnosis, incisional biopsy

Access this article online

Quick Response Code:

Website: www.jpbsonline.org

DOI: 10.4103/0975-7406.163569

Ameloblastoma is an epithelial odontogenic neoplasm that generally presents as solid/multicystic lesion. Its biological behavior is similar to the low grade malignant tumor because of its local invasive potential.[1] Surgical excision of the lesion with an adequate margin is considered to be an ultimate treatment modality to prevent the recurrence, which is primarily based upon the accurate preoperative incisional biopsy report.[1]

How to cite this article: Kaliamoorthy S, Ambiga P, Sivaraman S, Shanmugasundaram K, Nagarajan M. A case report of granular cell ameloblastoma associated with aneurysmal bone cyst like features. J Pharm Bioall Sci 2015;7:S797-9.

[Downloaded free from http://www.jpbsonline.org on Thursday, May 05, 2016, IP: 223.236.174.99]

Case Report

A 62-year-old female presented with gradually increasing swelling of the right side face for 5-years duration. Extra orally she had a diffuse, bony hard, non-tender swelling involving right side mandible (expansion of ramus, angle, and posterior body region) [Figure 1a]. Color and texture of the overlying skin was normal. Intraorally mild buccal vestibule obliteration was evident in relation to the edentulous molar region extending posteriorly to the ascending ramus area.

Panoramic radiograph showed multilocular radiolucency involving the ascending ramus, angle and posterior body of the right side mandible.

Computed tomography scan showed an ill-defined expansile lytic lesion seen within the ramus and angle of right side mandible measuring approximately of size 4.2 cm × 3.2 cm × 4.5 cm, with the lesional cavity partially filled with fluid and irregular soft tissue density in the residual portion with thinned out cortex. No calcification/fat attenuation was seen [Figure 1b].

Aspiration yielded a fluid mixed with dark venous blood, and the mild oozing was stopped on applying gentle pressure for few minutes.

Incisional biopsy was done, and the sample on histopathologic evaluation did not reveal any specific features except for loosely arranged fibro cellular areas with focal hemorrhage [Figure 2a].

Based on the incisional biopsy findings, a working diagnosis of aneurysmal bone cyst like change secondary to the unknown primary intraosseous lesion was made. Considering the clinical and radiographic features and the age of the patient it was decided to perform surgical resection and the patient underwent resection of the right side mandible (ramus to the molar area).

Macroscopically the resected specimen showed buccal cortical expansion with the cavitation of ramus and angle of the mandible with soft tissue mass confined to the peripheral parts of the lesion cavity [Figure 2b].

Microscopic examination of the Hematoxylin and Eosin sections of the soft tissue of the resected specimen showed ameloblastic follicular islands showing extensive granular transformation of the central stellate reticulum – like cells in a fibrous stroma [Figure 3a and b].

Immunohistochemical study using anti-cytokeratin antibody showed a uniform and intense positive staining with peripheral ameloblastic cell and central granular cells [Figure 4a] and anti CD-68 antibody showed intense positivity with central granular cells and negative reaction for peripheral ameloblastic cells [Figure 4b].

With the support of histopathology and immunohistochemical findings, a final diagnosis of granular cell ameloblastoma was made.

Discussion

Ameloblastoma is one of the most common odontogenic tumors that do not always grow as uniform solid mass due its inherent potential to undergo cystic degeneration.[1]

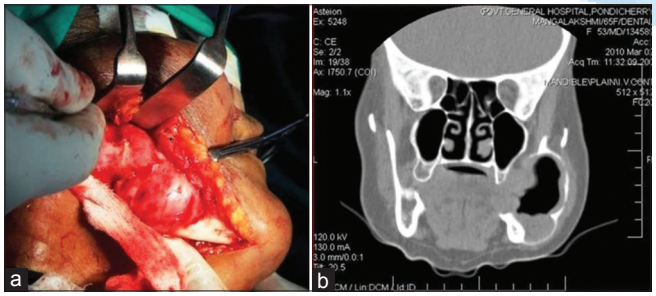


Figure 1: Cortical expansion of right ramus (a) and computed tomography scan axial view showing an expansile osteolytic lesion involving ramus, angle, and body of right mandible (b)

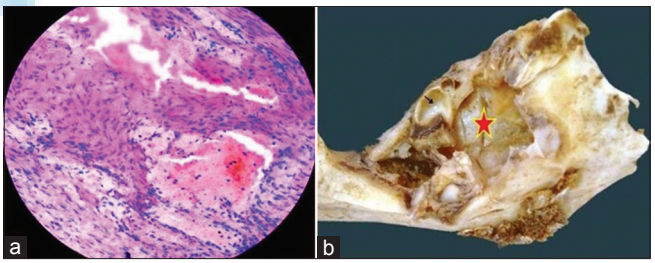


Figure 2: Blood filled spaces without endothelial lining in the fibro cellular area (a) and Gross specimen showing central cavitation (star), cystic spaces (arrow), and cortical expansion (b)

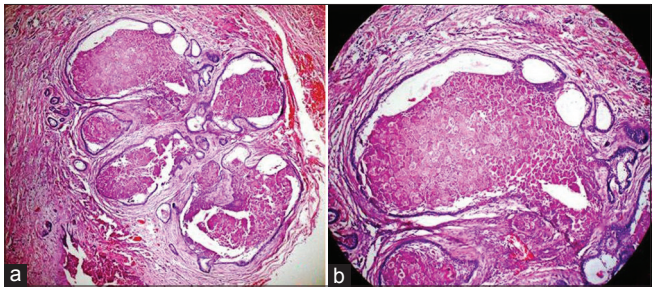


Figure 3: Ameloblastic follicles are exhibiting cuboidal to columnar cells at the periphery and extensive granular cell changes at the center. (a) H and E, ×4 and (b) H and E, ×40

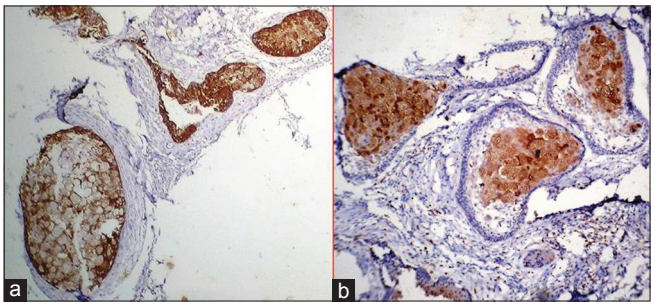


Figure 4: Peripheral ameloblastic cell and central granular cell showing cytokeratin positivity (a) and CD-68 positivity in central granular cells alone (b) (×40)

[Downloaded free from http://www.jpbsonline.org on Thursday, May 05, 2016, IP: 223.236.174.99]

Microscopically cystic degeneration usually starts as a small area within the ameloblastic follicle and as they grow gradually, lack of nutrition leads to complete degeneration of central region of the follicle leading to clear space centrally within the follicle. Often the periphery of the space shows inconspicuous flattened ameloblasts lining.

The extent of cystic degeneration logically seems to be more in long standing cases, and it may not be rare to encounter a lesion with gross feature showing complete cavitations of the involved jaw bone.

This macroscopic picture can potentially compromise the adequate representative sample procurement during the routine incisional biopsy.

Although any histopathological variant of ameloblastoma is prone for cystic degeneration, it can be speculated that the process is more aggressive and faster in granular cell variant of ameloblastoma. This is based on the fact that, phagocytosis of apoptotic remnants and adjacent neoplastic cells (digestion of solid tumor components) by the granular cells within the follicle as evidenced by the macrophage-specific lysosomes within the granular cells.[2]

Macroscopic presentation of complete cavitations in the current case may be explained with the above said facts and the incisional biopsy sample did not show any evidence of ameloblastomous follicle, except for the presence of loosely fibrous stroma with multiple blood-filled spaces without any specific endothelial lining, simulating aneurysmal bone cyst histopathological feature.

These stromal features that is, areas of hemorrhage are generally considered to be a secondary phenomenon in long-standing lesion[3] as similar to the present case that facilitated us to give a more practical working diagnosis for the further management.

Conclusion

Ameloblastoma with its varied macroscopic presentation may offer difficulty in its representative sample procurement during the incisional biopsy and its accurate interpretation at the histopathological level. This may be more significant in granular cell ameloblastoma, hence more practical approach with proper clinical correlation will avoid unnecessary advanced imaging modalities such as magnetic resonance imaging and repeat of surgical, diagnostic procedure, thereby reduce the cost and stress of the patients in its management.

References

1. Reichart PA, Philipsen HP, editors. Solid/multicystic ameloblastomas. In: Odontogenic Tumours and Allied Lesions. 1st ed. London, UK: Quintessence Publishing Co. Ltd.; 2004. p. 41-55.
2. Kumamoto H, Ooya K. Immunohistochemical and ultrastructural investigation of apoptotic cell death in granular cell ameloblastoma. J Oral Pathol Med 2001;30:245-50.
3. Shear M, Speight PM, editors. Aneurysmal bone cyst. In: Cysts of the Oral and Maxillofacial Regions. 4th ed. Oxford, UK: Blackwell Munksgaard; 2007. p. 150-5.

Source of Support: Nil, **Conflict of Interest:** None declared.