

Multinucleated Giant Cells in Unicystic Ameloblastoma: A Very Rare Case Report and Review of the Literature

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ABSTRACT

Background: Occurrence of giant cells has been reported in malignancies of pancreas, breast and thyroid but rarely in benign tumors. However, giant cells were observed in some benign odontogenic neoplasms like solid multicystic ameloblastoma. Studies were carried out in the past to establish the origin and nature of these giant cells and the results were quite variable. Here, a very rare case of unicystic ameloblastoma (UA) is presented which revealed focal multinucleated giant cells in close proximity to the calcifications seen in the connective tissue. An attempt was made to identify the nature and origin of these giant cells by immunohistochemical staining, the cells were negative for cytokeratin and positive for CD68. This suggests that the giant cells were non-epithelial in origin and probably were of foreign body type and stromal in origin.

Keywords: Ameloblastoma, Giant cells, Oral cavity.

INTRODUCTION

Unicystic ameloblastoma (UA) was first described as a distinct entity in 1977 by Robinson and Martinez¹. Ackermann et al recommended the use of term UA to describe all ameloblastomas which macroscopically consist of a single unilocular epithelial lined cystic cavity². UA constitutes approximately 10 to 15 percent of all intraosseous ameloblastomas³. It is usually less aggressive with distinctly lower recurrence rate than conventional ameloblastomas⁴. Giant cells have been reported in a variety of malignancies, their occurrence in odontogenic neoplasms is a relatively rare phenomenon. This report describes a very rare case of UA with stromal giant cells.



CASE REPORT

A 25 year old female presented with a chief complaint of numbness of lower lip since

eight months. History revealed that her lower right premolars were extracted about six months back and subsequently she developed numbness of the lower lip. The patient was referred to our institute with an incisional biopsy report as odontogenic fibromyxoma. Extra-oral examination did not showed any gross asymmetry. Intra-oral examination showed mild expansion of buccal cortical plate at lower right first molar region and absence of lower right premolars (Figure 1). Numbness of lower lip and mental region on right side was also noticed. Orthopantomograph (before extraction of premolars) revealed a well-defined unilocular radiolucency from lower left canine to lower right first molar (Figure 2). Computerized tomography scan showed a bilocular defect on the right side crossing the midline. Surgical enucleation of the tumor was done and the specimen was sent for histopathologic examination. Haematoxylin and eosin sections revealed a well-defined cystic lumen bordered by odontogenic epithelial lining overlying

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Fig 1: Intra-Oral view.



Fig 2: OPG.

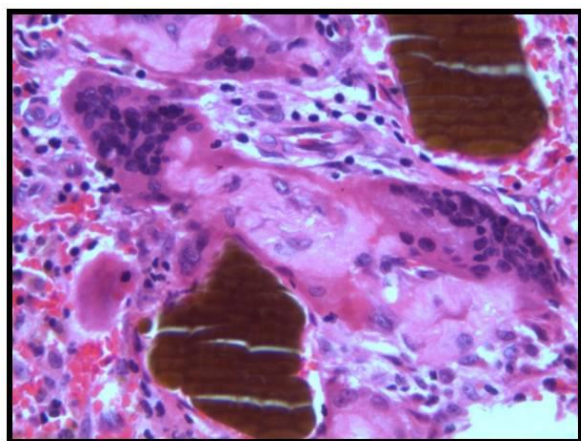


Fig 3: H & E Photomicrograph showing multinucleated giant cells adjacent to calcifications.

a delicate to dense connective tissue stroma. Cystic epithelium revealed basal columnar to cuboidal cells with hyperchromatic nuclei and superficial loosely arranged stellate reticulum-like cells. Underlying connective tissue stroma showed mild inflammatory component and few islands of odontogenic epithelium.

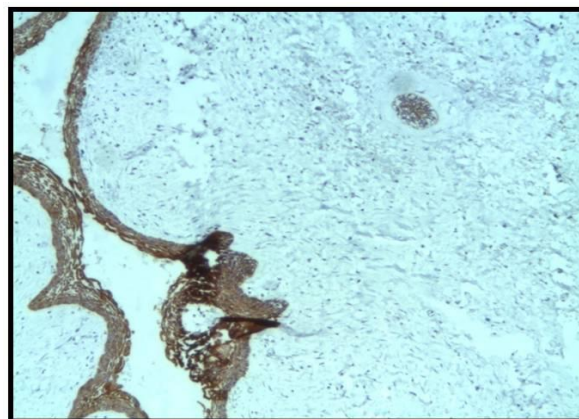


Fig 4: IHC showing negative for cytokeratin.

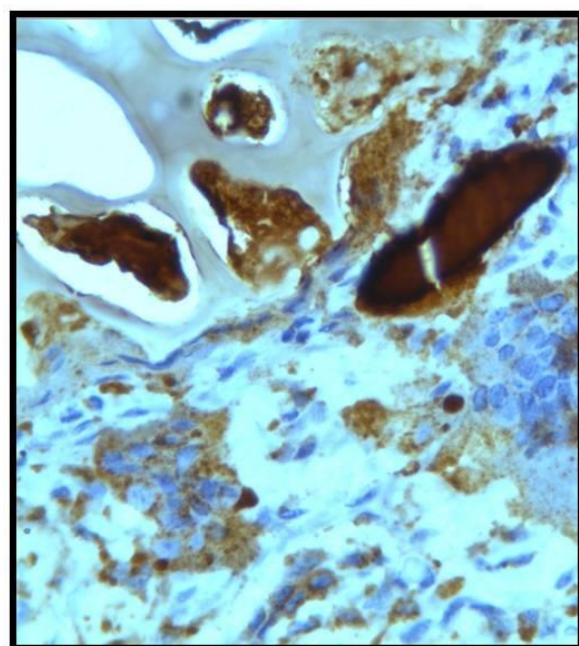


Fig 5: IHC showing CD68 positivity.

Areas of hemorrhage with calcifications were seen at few places. At focal areas, multinucleated giant cells with hyalinization of connective tissue stroma were evident (Figure 3). Based on the above findings a diagnosis of UA was given. Further immunohistochemical (IHC) studies were carried out with cytokeratin and CD68 to know about the origin and nature of the giant cells. The giant cells expressed CD68 but not cytokeratin (Figures 4 and 5).

DISCUSSION

The presence of giant cells has been documented in malignancies of breast, thyroid and pancreas but rarely in odontogenic neoplasms.

Though many attempts were made to know about the origin and nature of giant cells, the knowledge is still obscure.

Donath et al analyzed giant cells in 11 cases of salivary gland neoplasms. They suggested that giant cells may be of a reactive nature or may be a true tumour component in giant cell tumours. According to them reactive giant cells are derived from mononuclear monocytic or histiocytic stromal cells and they are either part of a local foreign body reaction or of a sarcomatoid stromal reaction. They classified giant cells as foreign-body giant cells, fibroblast-like giant cells, osteoclast-like giant cells, neoplastic giant cells and giant cells in granulomas. In their 11 cases, four cases showed foreign-body giant cells and seven cases showed osteoclast-like giant cells. All the giant cells were negative for cytokeratin and positive for CD68⁵.

Boss JH, Kawakami et al, Richard et al and Takeda et al reported giant cells in ameloblastomas⁶⁻⁸. Boss JH suggested that giant cells in their three cases were stromal reactive giant cells and divided giant cells into three types, foreign-body giant cells, giant cells as an integral component of a reparative granulomatous transformation of the connective tissue and osteoclasts resorbing newly formed bone spicules in the connective tissue stroma. However, Kawakami et al argued that determination of nature of giant cells is impossible by haematoxylin and eosin stained sections alone and without histochemical, immunohistochemical and ultrastructural studies being carried out. According to them giant cells associated with neoplasms are of two types neoplastic and non-neoplastic and that the later type mainly arises from stromal elements reactive to malignant epithelial elements which act as a foreign body as in their case⁶. Richard et al showed that osteoclast-like giant cells in their case was in response to the woven bone seen in connective tissue⁷. Takeda et al suggested that giant cells in their case were associated with prominent stromal ossification and these were reactive in nature^{8,9}. In addition, few reports have also suggested that the giant cells were epithelial in origin particularly in carcinosarcomas⁵.

Histopathologic findings in this case were suggestive of UA but connective tissue stroma showed unusual calcifications and irregular giant

cells with about 20-25 nuclei. Presence of giant cells has been reported in Solid Multicystic Ameloblastoma (SMA) but not in UA. After searching the literature, it was noticed that the present case was the first case of UA which showed stromal giant cells. The giant cells are supposed to help in the tumor progression by resorbing the bone, hence usually seen in more aggressive lesions. Analysis was done to find the origin of giant cells in this case and the results were similar to that of Donath et al⁵ and Richard et al⁷. The expression of cytokeratin by giant cells was negative suggesting their non- epithelial origin. Moreover giant cells were positive for CD68 suggesting their macrophage or histiocytic origin. The presence of giant cells in close proximity to the unusual calcifications may suggest that these giant cells were formed due to fusion of macrophages and are in the process of removal of calcifications, probably indicating foreign-body giant cell. Probably carrying out studies with different markers and assessing some more cases of UA with giant cells in near future may enlighten us in identifying the nature and origin of these giant cells.

CONCLUSION

Evidence of giant cells in benign neoplasms is rare and even more so in benign odontogenic neoplasms. So, a case of UA with stromal giant cells is reported here. Further, there is a need to thoroughly evaluate more number of such cases in future in order to understand about the origin and nature of giant cells.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES

1. Philipsen HP, Reichart PA. Unicystic ameloblastoma. A review of 193 cases from the literature. *Oral Oncol.* 1998; 34(5): 317-25.
2. Manas Bajpai, Deshant Agarwal, Anindya Bhalla, Malay Kumar, Rakesh Garg, and Manish Kumar, "Multilocular Unicystic Ameloblastoma of Mandible," *Case Reports in Dentistry*, vol. 2013, Article ID 835892, 4 pages, 2013. doi:10.1155/2013/835892.

3. Dandriyal R, Gupta A, Pant S, Baweja HH. Surgical management of ameloblastoma: Conservative or radical approach. *Natl J Maxillofac Surg.* 2011;2(1):22-7.
4. Yunus M, Baig N, Haque AU, Aslam A, Atique S, Bostan S, et al. Unicystic ameloblastoma: A distinct clinicopathological entity. *Pakistan Oral and Dent J.* 2009;29:9-12..
5. Donath K, Seifert G, Röser K. The spectrum of giant cells in tumours of the salivary glands: an analysis of 11 cases. *J Oral Pathol Med.* 1997;26(9):431-6.
6. Kawakami T, Antoh M, Minemura T. Giant cell reaction to ameloblastoma: an immunohistochemical and ultrastructural study of a case. *J Oral Maxillofac Surg.* 1989;47(7):737-41.
7. Richard BM, Thyveetil M, Sharif H, Athanasou NA. Ameloblastoma with stromal multinucleated giant cells. *Histopathology.* 1994; 25(5):497-9.
8. Takeda Y, Kuroda M, Suzuki A. Ameloblastoma with prominent stromal ossification. *Acta Pathol Jpn.* 1990;40(10):780-4.
9. Benjamin Fomete, Ezekiel Taiwo Adebayo, Joseph Osamudiamen Ogbeifun. Ameloblastoma: Our clinical experience with 68 cases. *J Orofac Sci.* 2014;6(1):17-24.

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