Cytology of Granular Cell Ameloblastoma of Jaw: A Rare Case Entity

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Abstract

Ameloblastoma is a benign neoplasm of odontogenic epithelium occurring more commonly in the mandible than the maxilla. The granular cell variant of ameloblastoma is a rare condition, accounting for 3.5% of all ameloblastoma cases. It is known to be locally more aggressive, with a higher chance of recurrence compared with other subtypes. Though fine-needle aspiration cytology of the jaw lesions are rarely done, it provides a means of rapid initial evaluation and offers reliable pre-operative cytological diagnosis. This case report illustrates the diagnosis of granular cell ameloblastoma based on correlation of clinicoradiologic and cytologic features.

Key words: Ameloblastoma, Basaloid cells, Cytology, Granular cell

INTRODUCTION

The odontogenic tumors constitute a heterogeneous group of lesions that arise from the epithelial and/or mesenchymal elements of tooth germ.¹ Ameloblastoma is a benign but locally aggressive tumor affecting the jaw bones. It occurs most often in the posterior mandible in the region of the third molar. There is no definite sex predilection and is seen most commonly in 4th and 5th decades.²

The most common clinical presentations are painless slow-growing swelling, which is often accompanied by facial deformity, malocclusion, tooth loss, and pain and paresthesia of the affected region.¹

Radiologically, ameloblastoma presents as an expansile uni or multiloculated lytic lesion, with thin internal septations giving the classic "soap bubble" appearance. They may be associated with resorption of roots of adjacent teeth. The internal septations represent differential cortical resorption, rather than true compartmentalization of tumor tissue.³

Access this article online



Month of Submission: 02-2015
Month of Peer Review: 03-2015
Month of Acceptance: 03-2015
Month of Publishing: 04-2015

Histologically the tumor exhibits diverse patterns. The morphologic spectra include follicular, plexiform, acanthomatous, granular cell, desmoplastic and basal cell patterns, the most common being follicular form. The granular cell ameloblastoma (GCA) is one of the rarest entities and accounts for only 5% of all ameloblastomas. Only around 30 cases of GCA's have been reported in English literature.²

On histopathology, GCA is characterized by a granular change of stellate-like cells located in the central portion of the epithelial islands. The periphery of these islands shows palisading tall columnar cells. This granular change is attributed to accumulation of lysosomal granules within the cytoplasm of stellate reticulum like cells. With increasing age, the lysosomes lose their ability to digest unwanted cellular material and their aggregates impart a granular appearance to the cytoplasm. 5

Whereas histopathological and radiological findings for ameloblastomas have been extensively studied, fine-needle aspiration cytology (FNAC) reports are rare with poor documentation in the literature.⁶ However, studies show that FNAC is an excellent diagnostic aid for the preoperative diagnosis of jaw lesions. We discuss here a case of GCA of the mandible, which was diagnosed based on characteristic clinical, radiological and cytomorphological features.

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CASE REPORT

A 42-year-old male patient presented to us with a painless swelling over the right side of the lower jaw. He had first noticed a small swelling 2 years back. Since then the swelling had grown slowly to its present size. There was no associated difficulty in opening the mouth, chewing or articulating.

On physical examination, there was a firm to hard, non-compressible, non-tender mass measuring 6 cm × 5 cm located at the angle of the right side of the mandible. The overlying skin was stretched, normal in color and texture and not adherent to the mass. No sinus or discharge was observed. Intraoral examination revealed a diffuse swelling in the posterior mandibular region on the right side, irregular in shape and firm to hard in consistency.

Computed tomography scan of the face revealed a uniloculated, expansile, lytic lesion of size 5.7 cm × 5.3 cm in the ramus of right mandible extending up to the angle of mandible, showing multiple areas of cortical dehiscence with associated heterogeneous soft tissue (Figure 1a-d). The radiological differentials were ameloblastoma and odontogenic keratocyst.

Aspiration of the swelling yielded thick reddish brown material. Air-dried smears were stained with May-Grunwald Giemsa. The smears showed moderate cellularity. There were sheets and clusters of uniform appearing basaloid cells with peripheral palisading. The basaloid cells showed scant, poorly defined cytoplasm, elongated nuclei, finely distributed chromatin and inconspicuous nucleoli. Closely admixed with these cellular fragments were large cells seen

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Figure 1: Computed tomography scan face and mandible.

Coronal (a), axial (b) and three-dimensional reconstruction images (c,d) show a large expansile lytic lesion (arrow) involving the right mandibular ramus extending up to the angle of mandible, with areas of cortical dehiscence

in loosely arranged groups as well as scattered singly. The cells showed abundant granular pale eosinophilic cytoplasm with ovoid nuclei and vesicular chromatin (Figure 2 a-d).

DISCUSSION

GCA is an uncommon variant of the many types of ameloblastomas. Though it possesses distinctive cytomorphological features, there are very few documented cases in the literature which have been diagnosed based on FNAC. Deshpande *et al.*, had described two cases of GCA diagnosed based on cytology and had stated that "recognition of large cells with abundant cytoplasm exhibiting eosinophilic granules dispersed singly or in loosely cohesive groups along with individual tightly cohesive basaloid epithelial cell clusters might facilitate its diagnosis in cytology." These cases were subsequently confirmed on histopathology lending credibility to their observation.

On cytology, the characteristic triad of ameloblastoma consists of cohesive sheets of basaloid cells with peripheral palisading, squamous metaplastic cells and cells resembling stellate reticulum.⁸ Fragments of basaloid cells with peripheral palisading intermixed with numerous large granular cells were seen in the present case, representing the classic features of GCA.

In regards to the tumor location, the various differentials considered in our case were salivary gland tumors such as mucoepidermoid carcinoma, warthin's tumor, oxyphilic adenoma/carcinoma, acinic cell carcinoma and granular cell tumor.

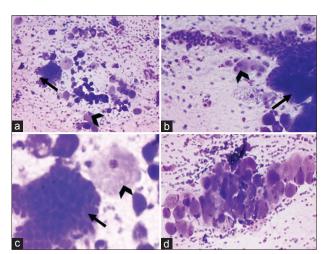


Figure 2: (a-c) Epithelial islands of basaloid cells with peripheral palisading (arrow) along with large granular cells (arrow head), (a) Giemsa, ×40, (b) Giemsa, ×100, (c) Giemsa, ×400, (d) Cluster of granular cells, Giemsa, ×200

In mucoepidermoid carcinoma, there is an admixture of mucin-secreting columnar cells, intermediate cells and squamoid cells in a dirty background of mucus and debris. Smears of warthin tumor aspirate show sheets of oncocytes with sharp cytoplasmic borders and abundant granular cytoplasm in a lymphocyte rich dirty granular background. Oxyphilic adenoma/carcinoma arising from salivary glands exclusively show clusters of oncocytes. Acinic cell carcinoma exhibits cells with abundant basophilic granular cytoplasm with eccentric nuclei often arranged in acinar-glandular pattern with frayed borders and a centrally placed vessel. The granular cell tumors of the jaw predominantly show granular cells in tight clusters and scattered singly. 10 However, all the above lesions can be reliably distinguished from GCA based on the presence of basaloid cell clusters showing peripheral palisading, admixed with large granular cells.

The pre-operative diagnosis of GCA is important as it enables the surgeon to plan a more radical surgery instead of a conservative approach. This is important as GCAs are known to be more aggressive and are associated with a high recurrence rate. A radical surgery is warranted in cases of GCA as their extension within the cancellous bone tends to exceed their macroscopic and radiologic boundaries. Simple enucleation or curettage may lead to recurrence of the tumour.

CONCLUSION

To conclude, in the presence of appropriate clinicoradiologic findings, FNAC provides a simple, low-cost, rapid and reliable pre-operative diagnosis. A meticulous search for basaloid clusters and large granular cells can aid in making a cytological diagnosis of GCA with reasonable accuracy thus avoiding unnecessary surgical biopsy and ensuring an adequate surgical excision in a planned manner.

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How to cite this article: Sharma S, Hasan R, Kumar S, Shivamurthy A, Shetty T. Cytology of Granular Cell Ameloblastoma of Jaw: A Rare Case Entity. Int J Sci Stud 2015;3(1):224-226.

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