

Cytology diagnosis of Multilocular Ameloblastoma of the Mandible: A Case Report

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ABSTRACT

Ameloblastoma is most common benign epithelial odontogenic tumor with malignant potential usually located in jawbone. This tumor is reported to constitute 1-3% of tumors and cyst of jaws. It is common between 3rd to 5th decades of life with equal sex distribution. It often presents as a slow growing painless swelling causing perforation of buccal bone along with infiltration of surrounding soft tissue. It has a very high recurrence rate of over 50% even after wide excision.

We present a case of Ameloblastoma in a 71 year female to highlight its cytological features.

Keywords: Mandible, Multilocular Ameloblastoma, FNAC of Mandibular Tumour.

Introduction

There are many benign odontogenic and non-odontogenic lesions which may present with swelling in mandibular region. Ameloblastoma is a tumor of odontogenic epithelium that commonly occurs in the jaws. It constitutes about 1% of all tumors and cyst of the jaws. [1,2] The tumor is usually reported in age group from 30 to 60 years with equal sex distribution. [3]

Utilization of Fine needle aspiration cytology (FNAC) in the diagnosis of odontogenic tumors in general and ameloblastoma in specific are rare. FNAC diagnosis of ameloblastoma is a simple, inexpensive, convenient and comfortable method for evaluation. The sensitivity and specificity of FNAC in the diagnosis of ameloblastoma is reported to be 86.6% and 100% respectively. [1]

Case Presentation

A 71 year female patient presented with complaints of swelling over the left side of mandible since 5 years. There was history in difficulty in swallowing since two years. There no history of pain or pressure symptoms. There was no history of similar swelling in the body. Past history revealed tooth extraction two years back after which the swelling increased in size exponentially. Personal history and family history were insignificant.

On examination, a single huge swelling measuring 15x15x10 cm was seen on left side of the mandible distorting the face (Figure 1). Skin over the swelling was tense with engorged veins. External surface was nodular. On palpation, swelling was found to be firm, non-tender and non reducible. Intraorally, there was buccal and lingual

extension of the tumour. Mouthy opening was restricted to 1-2 finger breadth.

Computed tomography showed a large, expansile, multiloculated cystic mass with heterogeneously enhancing soft tissue component arising from hemi-mandible and a possible diagnosis of Ameloblastoma was given (Figure 2).

Fine needle aspiration cytology (FNAC) showed round to oval to spindle cells with bland nucleus, moderate amount of cytoplasm without definite cell margins. These cells were arranged in cohesive clusters, monolayered sheets and in singles. Focal areas showed palisading arrangement of cells with nucleus showing inverse polarization with fibro collagenous tissue (Figure 3). A cytological diagnosis of ameloblastoma was given.

Surgical excision of tumor was done and subjected to histopathological examination. Grossly, specimen consisted of single, nodular, soft tissue mass measuring 11x7x8.5cm with two teeth and part of mandible. Cut surface showed solid and cystic areas exuding about 150ml of brownish mucoid material. Histopathological examination revealed odontogenic epithelium consisting of elongated /columnar cells with nucleus having reversed polarity, arranged in palisading pattern. Majority of cells were arranged in follicular pattern, some in sheets, plexiform pattern, cords and trabecular pattern (Figure 4). A few cells showed abundant amount of eosinophilic granular cytoplasm (Granular variant). Focal areas showed squamous metaplasia with desmoplasia and involvement of bone. A diagnosis of Ameloblastoma- Follicular variant was given. Patient was followed up for 6 months, which was uneventful.



Fig. 1: Photograph showing huge tumour with intraoral extension.

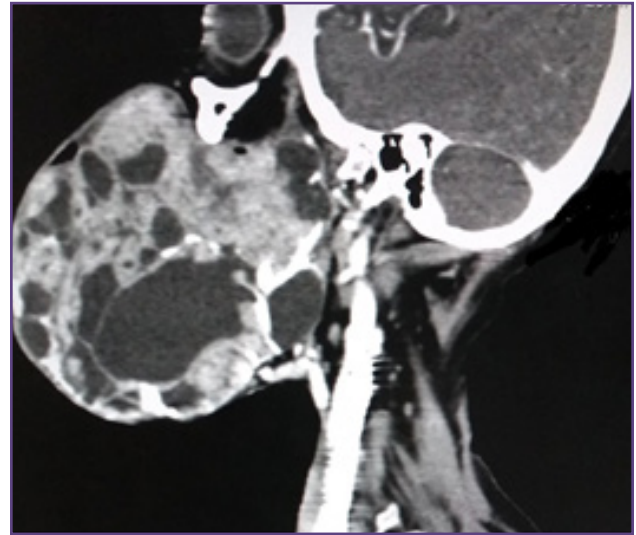


Fig. 2: CT Photograph showing multiloculated cystic lesion with bone destruction.

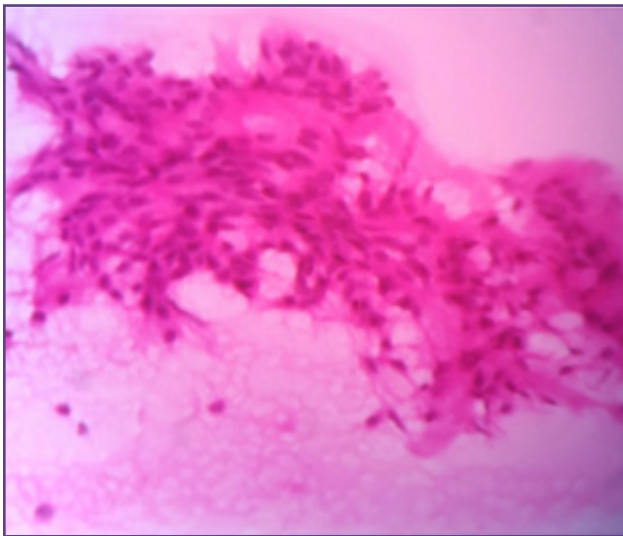


Fig. 3: Microphotograph of FNAC smear showing oval to spindle cells in cohesive clusters with palisading pattern at the margins. (H&E X400).

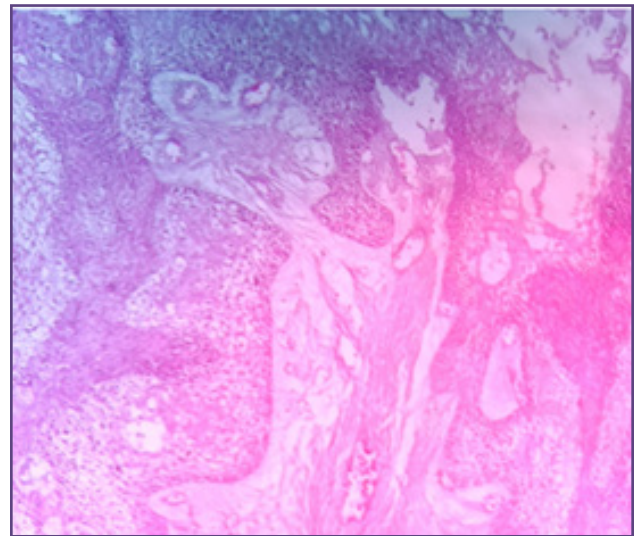


Fig. 4: Microphotograph of the tumour showing oval to spindle cells in palisading pattern and reverse polarization. (H&E X400).

Discussion

Ameloblastoma is most common benign odontogenic tumour arising from odontogenic epithelium. Though benign, they are locally aggressive epithelial odontogenic neoplasm and can rarely transform to malignancy with metastatic deposits (less than 2%).^[1,2] They constitute about 1% of all head and neck region tumours and 11% of odontogenic tumours.^[3,4]

It is commonly seen in the 3rd to 5th decades of life with average age of 36 years. Its peak incidence is in 4th decade of life. In developing countries it is reported in younger

patients.^[1,2,3,4] Odontogenic tumors in early teenage years presents as a well circumscribed radiolucency lesion associated with mal positioned and unerupted teeth mostly in posterior mandible.^[2] Cystic ameloblastomas occur in any age group. Unicystic ameloblastomas are reported occurring in younger patients in approximately 6% of cases.^[1] Ameloblastoma is considered fairly rare in elderly.^[3,4] In present case the patient is an elderly female.

Men and women are affected equally and there is no sex predilection. Usually women are four years younger than men and ameloblastoma appear to be larger in females than

in males. ^[1,3,4] In present case the patient is female with huge swelling.

Mandible is the most common site of occurrence. ^[1,2,3,4] The ratio of ameloblastoma of the mandible to the maxilla is 5:1 and that of mandible occur 12 years earlier than the maxilla. It commonly occurs in the molar region of the mandible. Two percent of ameloblastomas are peripheral tumors. In Blacks races, it commonly occurs in the anterior region of the jaws. ^[1,2] In present case the tumour was mandibular in origin.

Predominant clinical symptoms are slow growing painless swelling which is non-characteristic. It frequently becomes large, destructive and multilocular. It causes expansion of the cortical bone, perforation of the lingual and/or buccal plates and infiltration of soft tissue. The slow growing nature of the tumour leads to delay in diagnosis. ^[1,2,3,4] In the present case, the tumour was also slowing growing since five years.

Radiologically, ameloblastoma shows features of expansileuni or multiloculatedexpansilelytic lesion along with thin internal septations presenting as classic “soap bubble” appearance. Approximately 50% of the ameloblastomas appear as multilocular radiolucent lesions with a sharp delineation. They may be associated with resorption of roots of adjacent teeth with erosion of adjacent tooth/bone and eroding into soft tissue. The internal septations indicates differential cortical resorption than true compartmentalization of tumor. Ameloblastomas have a characteristic appearance on CT, but are not specific. ^[1,2,5] In this case report also the lesion was expansile, multiloculated cystic mass with involvement of soft tissue.

FNAC has been less commonly used as a diagnostic tool in diagnosis of odontogenic tumors and cysts. A few reports of ameloblastoma and ameloblastic carcinoma are reported to be diagnosed by FNAC. FNAC of jaw lesions is a simple, rapid and non-invasive procedure for the initial diagnosis of these lesions as benign / malignant and prevents unnecessary surgical biopsy. Ameloblastomas can be aspirated conveniently and the cytologic features are quite characteristics to offer diagnosis. Arrangement of cells resembling odontogenic islands or ameloblast like epithelial cells in palisading pattern along with digitated / stellate reticulum-like cells is clue for diagnosis. The other cytological features are tightly cohesive clusters or as pseudopapillary projections of basaloid epithelial cells, squamous differentiation as larger cells with a central nucleus, abundant eosinophilic cytoplasm with keratohyalin granules and whorls. Another study has reported two-cell population having small, hyperchromatic, basaloid-type cells and scattered larger cells with more open chromatin. Fragments of mesenchymal cells with

elongated nuclei with abundant, clear cytoplasm are reported. False negative FNAC are reported in inadequate specimen or sampling error and most are related to cystic tumors. Tissue sampling at FNAC can be done at multiple sites and deeper aspect of the tumor which can help in arriving at the accurate preoperative diagnosis. ^[1,2,4] In this case, most of the cytomorphological features were present in the FNAC smears.

The differential diagnosis of Ameloblatoma in FNAC is ameloblastic fibroma, a primary intraosseous tumor. Both show predominance of basaloid cells with peripheral palisading of tumor cell. However Ameloblastic fibroma has more stromal fragments than ameloblastoma. The other differential diagnosis are other basaloid cell tumors involving jaw. ^[2]

Histomorphologically the subtypes are follicular (1/3rd), plexiform (1/3rd), acanthomatous (occur in elderly), papilliferous, keratotic, granular cell, desmoplastic and vascular. In follicular type, the cells resemble dental organ epithelium. These cells are tall, columnar with polarization of nuclei away from the basement membrane (Inverse polarization). The central portion of epitheloidisland is composed of loose network of cells resembling stellate reticulum. Acanthomatous type is composed of squamous metaplasia within stellate reticulum. The plexiform type shows irregular mass and interdigitating cords of epithelial cells. ^[1,4,6] In this case report, the predominant pattern was follicular.

Ameloblastomas has a very high recurrence rates after surgery (up to 50%) and hence it is placed in Borderline category of tumours rather than benign category. Hence there is a need for long term followup of these patients. ^[4,5,6] The prognosis depends on recurrence of the tumour after surgical excision. Usually, ameloblastoma in elderly patients carry a bad prognosis. Recurrence may be due to the persistence of the original tumour which was not resected or due to new neoplastic cells. ^[4,6] We followed the case only for 6 months which was uneventful.

Hence presence of clinico-radiologic findings helps in evaluation FNAC diagnosis. FNAC provides a simple, low cost, rapid and reliable pre-operative diagnosis of ameloblastoma. It provides the initial evaluation of these lesions, avoids unnecessary surgical biopsy and ensures an adequate surgical excision in a planned manner. ^[1,2] In the present case also the clinical and radiological findings were correlated in issuing cytological diagnosis.

Conclusion

It is important to know the cytological features for the diagnosis of Ameloblastoma and plan for surgery. In addition, knowing cytological features also help in early

diagnosis of recurrence cases. Overall, early diagnosis of Ameloblastoma increases the patient survival rate. In our case the patient was an elderly female which is a rare feature.

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Financial or other Competing Interests: None.