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Case Report

Mural ameloblastoma- A case report

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ABSTRACT

Ameloblastoma is a benign neoplasm of odontogenic epithelial origin which comprises of several clinical, radiological and histological varieties. Among these, unicystic variant is the least explored and its mural subtype shows a high aggressiveness and risk of recurrence and comparable with that of conventional ameloblastoma. Herein, we present a case of mural ameloblastoma of maxilla in a 32-year old female.

1. Introduction

Ameloblastomas, although locally invasive, are considered to be benign neoplasms derived from odontogenic epithelium.[1] WHO 2017 classified these lesions as ameloblastoma, unicystic ameloblastoma (UA), and peripheral type. Metastasizing ameloblastoma is another type now classified under ameloblastoma, although, this decision was not unanimous.[2] Robinson and Martinez in 1977 first described UA as a distinct variant with considerably better prognosis and a much reduced incidence of recurrence compared to conventional ameloblastoma.[3,4] These lesions histologically classified as luminal, intraluminal or mural type.[4] Mural types are those cystic lesions that behave clinically as cyst and show radiological characteristics of odontogenic cyst. But in histopathological examination these lesions show macroscopically a thickening or a nodule in some part of their cystic lining which microscopically is composed of ameloblastic tissue.[5] Here we report a distinctive case of mural variant of unicystic ameloblastoma.

2. Case Report

A 32-year-old female reported to our institution referred by a private practitioner for the management of pain and swelling in relation to her right upper back teeth region. History revealed that the pain and swelling was present since 2 years for which she had approached a private clinic and had undergone extraction of right lower back tooth before 1year even after which the swelling did not subside. Then she had been referred to an Otolaryngologist and had undergone cyst removal along with right middle meatal antrostomy. Follow-up after 1 year revealed no regression of the swelling. Thus she had been referred to our institute for further opinion and management.

Extra oral examination revealed a mild diffuse swelling present in right side middle third of face of size 4 x 3 cm^2, extending superiorly from infra orbital margin, inferiorly in line with right corner of mouth, anteriorly in line with right ala of nose and posteriorly in line with right outer canthus of eye. Skin over the swelling was normal with no secondary changes (Figure 1). On palpation swelling was firm, non-tender, non-reducible, non-compressible and non-fluctuant. Single right submandibular lymph node was palpable of size approximately 0.5×0.5 cm^2, tender and mobile. Intra

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oral examination revealed a well-defined swelling on right buccal vestibule of size 1 x 2cm^2 extending from distal of 13 to distal of 16. Surface was smooth with no secondary changes. On palpation it was firm and non-tender. A palatal extension of the swelling was also noticed of size 2 x 1cm^2 extending from distal of 13 to distal of 16 (Figure 2).

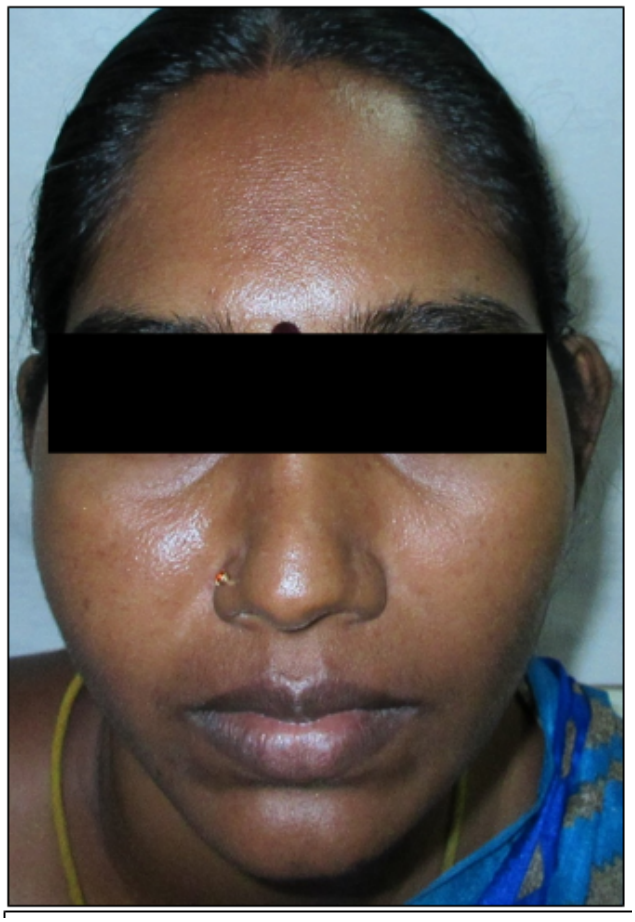

Fig. 1: Clinical photograph showing extra oral swelling in the right middle third of face

Intra oral periapical radiograph revealed a well-defined radiolucency extending from distal of 13 to distal of 16 with root resorption in relation to 14, 15 and 16. Orthopantomogram revealed similar findings along with missing teeth of all the third molars (Figure 3).

Based on the site of the swelling, history of tooth extraction, well-defined radiolucency, a provisional diagnosis of odontogenic cyst was given.

Histopathological examination of incisional biopsy tissue revealed flattened lining epithelium with few satellite cysts like areas in subepithelial zone. These satellite cysts have peripheral columnar cells and central stellate reticulum type of cells, resembling ameloblastic follicle. However, deeper areas appear to be normal (Figure 4). Based on these findings, histopathological diagnosis of odontogenic cyst

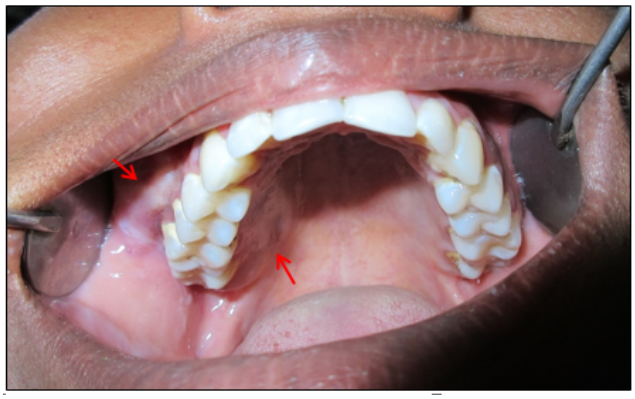

Fig. 2: Clinical Photograph showing intra oral swelling from 13 to 16 region

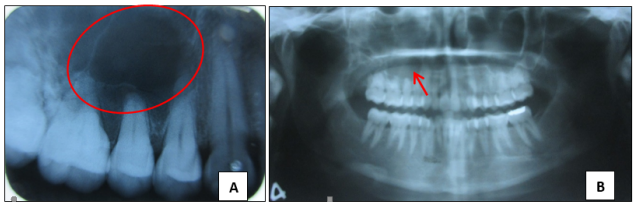

Fig. 3: Radiographic presentation. (**A**) Periapical radiograph revealing well defined radiolucency from distal of 13 to distal of 16 with root resorption in relation to 14, 15 and 16. (**B**) OPG revealing similar findings along with missing teeth of all the third molars

with early ameloblastic changes was given.

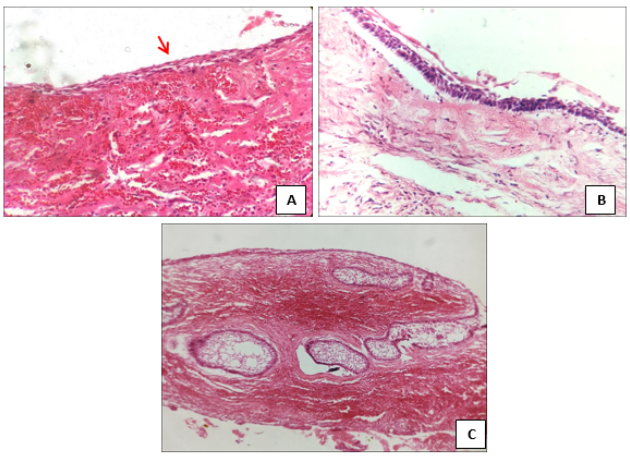

Fig. 4: Histopathology picture showing (**A**): Flattened lining epithelium (**B**): lining epithelium showing basal tall columnar cells with reversal of polarity and a superficial thin layer of degenerating stellate reticulum like cells (**C**): In growth of ameloblastic epithelium lining in to the fibrous capsule and few satellite cysts characterized by peripheral columnar cells and central stellate reticulum type of cells.

Cystic lining was then removed by Enucleation under general anesthesia and specimens send for histopathological examination (Figure 5). Specimen revealed thin odontogenic epithelial lining, fibrous capsule and deeper bone whereas some other areas exhibited multiple cystic lesions in separate follicles, peripheral cells as columnar showing reversal of polarity. A part of the lining epithelium also exhibited arcading arrangement. The central areas of the follicles showed degenerative changes (Figure 6). Considering these findings and clinical history of extraction of right maxillary posterior tooth, a final diagnosis of follicular ameloblastoma probably derived from dentigerous cyst lining epithelium was given.

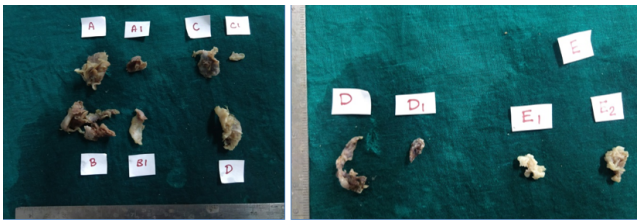

Fig. 5: Tissue specimens removed by enucleation

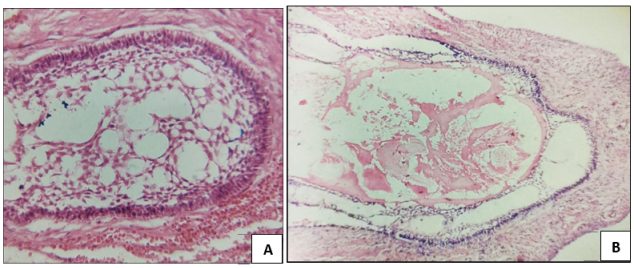

Fig. 6: Histopathology picture showing separate follicles (**A**) arranged as peripheral tall columnar cells with reversal of polarity; inner stellate reticulum like cells (**B**) large cystic spaces within the follicle

3. Discussion

The unicystic ameloblastoma (UA) has for several decades been given separate consideration based on its clinical, radiographic, and histopathologic features.[6] It is a rare type of ameloblastoma, accounting for about 6% of ameloblastomas, may be associated with impacted tooth, which is most often seen in young patients with 50% of such tumors diagnosed during the second decade of life.[7]

The pathogenesis of UA remains obscure. Whether it originates de novo as a neoplasm or a result of neoplastic transformation of non-neoplastic cyst epithelium has long been debated.[8] The proposed three pathogenic mechanisms for the evolution of UA are: (1) the reduced enamel epithelium associated with a developing tooth undergoes ameloblastic transformation with subsequent cystic development; (2) ameloblastomas may arise in a dentigerous or other type of dental cyst in which the neoplastic ameloblastic epithelium is preceded temporarily by a non-neoplastic stratified squamous epithelial lining; (3) a solid ameloblastoma undergoes cystic degeneration of ameloblastic islands with subsequent fusion of multiple microcysts to develop a unicystic lesion.[9,10]

Ackermann et al classified UA into three histological subgroups: luminal, intraluminal and mural.[11] Among the three subtypes, mural type has the highest recurrence rate since the epithelium penetrates and invades the fibrous wall with a high potential to infiltrate the adjacent cancellous bone.[12]

Histopathologically mural ameloblastoma is characterized by ingrowth of ameloblastic epithelium lining in to the fibrous capsule. Epithelial cells are of tall columnar in basal layer with subnuclear vacuoles, reverse polarity of hyperchromatic nucleus and a thin layer of edematous degenerating stellate reticulum like cells on the surface.[4] Our case also showed these typical features of mural ingrowth in some areas along with typical ameloblastic follicles.

Thus a definitive diagnosis of mural growth in ameloblastoma can only be done by histological examination of entire lesion and cannot be predicted preoperatively on clinical or radiographic grounds. As preoperative incisional biopsy is not representative of entire lesion it may result in an incorrect classification. True nature of the lesion becomes evident only after enucleation when entire specimen is available for microscopy.[8]

Thus with any presumed unicystic ameloblastoma, multiple sections particularly in nodular thickenings of specimen are necessary to rule out the possibility of mural invasion of tumor cell.[8] When mural involvement is identified the tumour may behave biologically as conventional ameloblastoma and requires either additional surgery or more careful follow-up.[13]

4. Conclusion

Ameloblastoma may appear as a simple cyst, points out the necessity for histopathological examination of all cystic specimens prior to rendering a specific diagnosis. It is particularly necessary to make a thorough histological examination of the cyst wall with special attention to all mural nodules or thickenings in order to avoid missing tumor outgrowths that may be result in multiple recurrences and reveal the existence of the already transformed neoplasm.

5. Source of Funding

None.

6. Conflict of Interest

The authors declare no conflict of interest.

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