

Follicular Ameloblastoma : A Case Report

Kale UB*, Munde AD**, Biradar S***, Shaikh SS****

Abstract

Ameloblastoma, a true neoplasm of odontogenic epithelium is a persistent, locally invasive and an aggressive tumor. The ameloblastoma represents about 1% of all odontogenic epithelial tumors. Clinicopathologically ameloblastoma is divided into solid/multicystic type (86%), the unicystic type (13%) and peripheral or extraosseous (1%) variant. A painless swelling and expansion of the jaws is the most common clinical presentation; neurosensory changes are uncommon, even with large tumors. It has a high recurrence rate due to its capacity to infiltrate trabecular bone and potential to undergo malignant transformation and to metastasize (1- 2%). In this paper we report a case of follicular ameloblastoma in a 22 year old female.

Key words: Ameloblastoma, follicular, mandible, solid multicystic

Introduction

Robinson defined ameloblastoma as a tumor which is “usually unicentric, nonfunctional, intermittent in growth, anatomically benign, and clinically persistent.” The term ameloblastoma was coined by Churchill in 1934 to replace the term adamantinoma coined by Malassez (1885).[1] It is the second most common odontogenic neoplasm and accounts for approximately 11% to 18% of all odontogenic tumors. The tumor is thought to originate from, cell rests of the enamel organ, epithelium of odontogenic cysts, disturbances of the developing enamel organ, basal cells of the surface epithelium or heterotropic epithelium in other parts of the body especially pituitary gland.[1, 2,3] There are 4 distinct clinicopathologic subtypes: unicystic ameloblastoma (UA), solid / multicystic ameloblastoma (SMA), peripheral and malignant forms. Unicystic ameloblastoma usually appears as a cystic lesion with either intraluminal or an intramural proliferation of cystic lining. SMA has great infiltrative potential and a higher recurrence rate.[4,5] It has 6 histologic subtypes:

follicular, plexiform, acanthomatous, granular cell, basal cell and desmoplastic.[1,2,3] Treatment may vary from curettage to broad bone resections, with or without reconstruction. Curettage should be avoided due to its high recurrence rate. Larger, aggressive lesions require a more radical surgical approach. Prognosis of ameloblastoma is more dependent on the method of surgical treatment rather than histologic type of tumor.[6,7]

Case report

A 22 year old female patient reported with the complaint of swelling over the lower left face since 15 months. Initially the swelling was small, painless, of peanut size and gradually grew to attain the present size. There was no history of paraesthesia or anaesthesia. Personal and medical history of patient was not significant.

Extraoral examination revealed a solitary swelling on lower left side of the face, approximately 6X5cm in size, extending anteroposteriorly from angle of mouth to angle of mandible and superoinferiorly from line joining tragus of ear and angle of mouth to 1cm below inferior border of mandible. The skin over the swelling was normal in color and intact. (Figure 1,& 2) On palpation the swelling was non tender and hard in consistency. Intraorally, there was expansion of buccal and lingual cortical plates extending from mandibular left canine to mandibular left third molar causing obliteration of the buccal and lingual vestibule. Overlying mucosa was of normal color and intact. Grade I mobility was present with mandibular second premolar, first molar

*Post-Graduate Student, **Professor, ***Sr. Lecturer, ****Professor and Head,

Corresponding author:

Dr. Ujwala B. Kale
Post-Graduate Student, Dept. of Oral Medicine and Radiology,
Rural Dental College, PIMS, Loni-413736,
Tal: Rahata, Dist: Ahmednagar, Maharashtra, India
E-mail- drujwalak@gmail.com

and second molar and second molar was displaced distally. (Figure 3) Based on clinical examination provisional diagnosis of benign tumor or cyst was made. Differential diagnosis included ameloblastoma, dentigerous cyst, ameloblastic fibroma and central giant cell granuloma.



Figure 1: Extraoral photograph (front view)



Figure 2 : Extraoral photograph (lateral view)



Figure 3 : Intraoral photograph

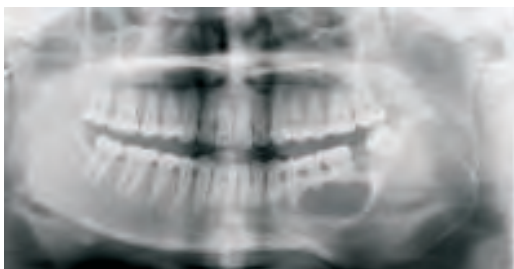


Figure 4 : Panoramic radiograph showing extensive multilocular radiolucency

Imaging/radiographic examination

Patient was subjected to radiographic and CT examinations. Orthopantomogram (OPG) revealed a large multilocular radiolucent lesion extending from mandibular left canine involving mandibular body and ramus causing thinning and expansion of its inferior border. The numbers of locules were few and large with curved septae. External root resorption was seen with mandibular left first and second premolars and molars (Figure 4). Occlusal radiograph and CT scan revealed significant thinning and expansion of buccal and lingual cortical plates and loss of cortex on the lingual aspect (Figure 5,6,7)



Figure 5 : Occlusal radiograph showing expansion of buccal and lingual cortical plates

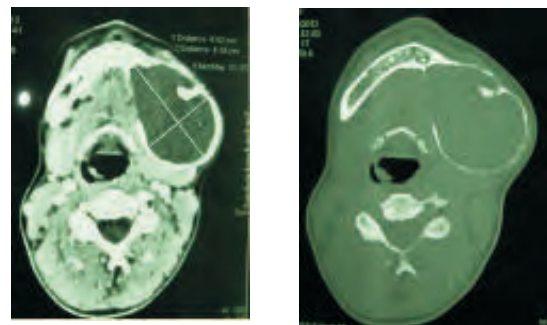


Figure 6 : CT Scan (axial view)

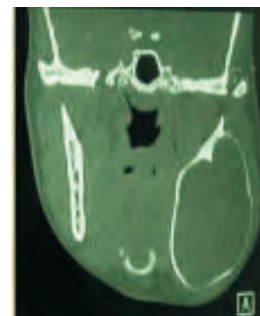


Figure 7 : CT Scan (coronal view)

Treatment

Surgical resection of the lesion along with hemimandibulectomy and reconstruction was done with titanium plates. The resected specimen was sent for histopathological examination. Patient is on regular follow up and there is no evidence of recurrence (Figure 8)

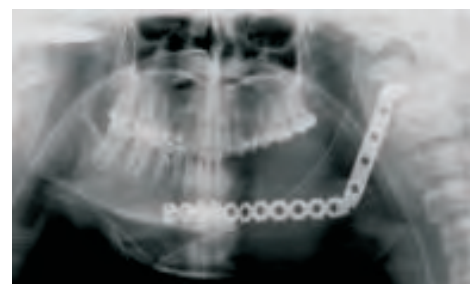


Figure 8 : Post operative OPG

Histopathological examination

H & E stained section showed follicles of ameloblastoma. Few areas showed lining of stratified squamous epithelium. Fibro-vascular connective tissues consisting of numerous endothelial lined blood vessels, chronic inflammatory giant cells were also seen. Overall features were suggestive of "follicular ameloblastoma".

Discussion

Ameloblastoma is a neoplasm of odontogenic epithelium, especially of enamel organ-type tissue that has not undergone differentiation to the point of hard tissue formation. It is a benign but locally aggressive tumor.[2,3] A wide range of occurrence of the tumor from 10 years to 90 years has been reported. Most patients are between ages of 20 and 60 years,[1] with no significant sex predilection. About 80% of all cases occur in mandible of which 70% are seen in molar ramus area, 20% in premolar region, and 10% in anterior region.[8] In our case a 22 year old female reported with swelling in molar ramus area of mandible which is consistent to the data given in the literature.

Patients of ameloblastoma generally present with slow growing painless swelling causing facial asymmetry and loose teeth and rarely paresthesia. Lesions generally progresses slowly and extensive. Untreated lesions may cause cortical expansion and perforation and infiltrate into the adjacent soft tissue.[6]

A multilocular expansile radiolucency usually in the molar ramus area is the classic radiographic appearance of the ameloblastoma. Depending on the size and number of locules it can give either honeycomb or soap bubble appearance.[9] Our case showed multilocular radiolucency involving mandibular molar angle ramus area with thinning and expansion of inferior border, and occlusal radiodiograph and CT showed expansion of buccal and lingual cortical plates. CT also revealed loss of lingual cortex.

Among the histologic types, follicular and plexiform ameloblastoma are most common and acanthomatous, granular cell, desmoplastic and basal cell types are less common.[1,2,3] The case reported is of follicular type. Follicular ameloblastoma is characterized by higher recurrence rate (29.5%) compared to plexiform ameloblastoma (16.7%) and acanthomatous ameloblastoma (4.5%).[1,2].

Malignant transformation of ameloblastoma is very rare (1%-2%). Malignant ameloblastoma is defined as a tumor that shows histopathological features of ameloblastoma

both in primary tumor and metastatic deposits. Ameloblastic carcinoma is an ameloblastoma exhibiting the histologic criteria of a malignant neoplasm.[3,9]

Recommendation

Ameloblastoma, especially follicular type has a high rate of local recurrence if it is not adequately removed. In cases of large, expansive tumors a radical surgical protocol is a very good option to prevent relapse of the tumor on a long-term basis.[3] In general, annual follow up for at least 10 years is recommended. Others recommend annual follow-up until 5 years and every 2 years thereafter for at least 25 years.[7]

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