(e) ISSN Online: 2321-9599
(p) ISSN Print: 2348-6805

CASE REPORT

INTRICACIES IN DIAGNOSING AMELOBLASTOMA- A CASE REPORT

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ABSTRACT:
Ameloblastoma is a true but rare neoplasm of odontogenic origin in the oral cavity (0.78%). It has a wide range of age for occurrence with mean age of 36 years and with equal frequency for both the sexes. It occurs most commonly in the mandible than in maxilla (5:1), involving the mandibular molar and ramus areas. It is usually asymptomatic therefore patients report late after tumor has reached a considerable size. Since it has high recurrence rate therefore complete surgical excision is the main aim of treatment. We hereby present a case of follicular ameloblastoma in a 30 year old female who presented with a swelling involving the lower left posterior region of jaw.

Keywords: Ameloblastoma, Odontogenic tumor, Surgical excision

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This article may be cited as: Hundal JK, Kaur H, Sandhu SK, Kaur I. Intricacies in diagnosing Ameloblastoma- A Case Report. J Adv Med Dent Scie Res 2016;4(3):103-107.

INTRODUCTION

Odontogenic tumors are lesions derived from epithelial, ectomesenchymal, and/or mesenchymal elements that are, or have been, part of the tooth-forming apparatus. These tumors, therefore, are found exclusively within the jawbones (intrabony or centrally located) or in the soft mucosal tissue overlying tooth-bearing areas (peripherally located).[1] Ameloblastoma falls under the category of 'benign neoplasm arising from odontogenic apparatus showing odontogenic epithelium with mature, fibrous stroma, odontogenic ectomesenchyme not present.[2] The tumor may be derived from disturbances of the developing enamel organ, residual epithelium from tooth germ, epithelium of the odontogenic cysts and cell rests of enamel organ, but it does not undergo differentiation to the point of hard tissue formation.[3]
Ameloblastoma is a true neoplasm which is described as a tumor which is usually unicentric, non-functional, intermittent in growth, anatomically benign and clinically persistent.
Ameloblastoma is the second most common odontogenic tumor after odontome and it presents as a slow growing, locally invasive tumor. Ameloblastoma has a wide range of occurrence, with a mean age of 36 years and most of the cases occur between 1st to 9th decades of life. No significant sex predilection has been reported. Ameloblastoma occurs in all areas of the jaws, but the most commonly affected area is the posterior region and the ascending ramus of mandible.[4]
Clinically it is generally asymptomatic, but sometimes it may present symptoms such as slow growing painless swelling, expansion of the cortical bone, perforation of buccal or lingual cortical plates and infiltration of soft tissue.[5] Others symptoms may include dental malocclusion, paresthesia and pain of the affected area.[3]
Radiographically this tumor generally presents as a unilocular or multilocular radiolucent area with a honeycomb appearance or soap bubble appearance. Depending upon the clinic-radiographic type of tumor there is variation in the radiographic appearance of ameloblastoma. CT is generally helpful in determining the contours of the lesion, its content and extension in the soft tissue.[6] For visualizing the papillary projections into cystic cavity MRI is superior to CT, as it helps in establishing the exact extent of ameloblastoma and thus determining the prognosis of the surgery.[7]
According to Reichart four variants of ameloblastoma categorized clinic-radiographically are solid multicystic ameloblastoma (SMA), unicystic ameloblastoma (UA), peripheral ameloblastoma (PA) and desmoplastic ameloblastoma (DA). SMA is further classified histopathologically into: follicular, plexiform, acanthomatous, granular cell, basal cell and desmoplastic.[8] Follicular ameloblastoma is the most

common with the incidence rate of 27.7% followed by plexiform, acanthomatous and desmoplastic type's respectively.[9]

Treatment of ameloblastoma depends upon clinic-radiographic variant, anatomic location and clinical behaviour of the tumor. Treatment includes radical and conservative surgical excision, curettage, chemical and electrocautery, radiation therapy or a combination of surgery and radiation. Curettage is least preferred because of high recurrence rate associated with it. Recurrence rate is 10-25% after radical treatment and 75-90% after conservative treatment.[2,10] The goal of treatment of ameloblastoma is to achieve complete removal of lesion and appropriate reconstruction of surgical defect. We, hereby illustrate a case of 30 year female who presented with a swelling on the left side of face and was diagnosed with ameloblastoma of the follicular variant.

CASE REPORT

A 30 year old female reported with a swelling in the lower left posterior region of jaw since 5 months. History revealed that, initially the swelling was small in size and gradually increased to reach the present size of 3 x 3 cm. Extraorally the extent of the swelling was seen involving the lower left and middle third of face extending supero-inferiorly from oro tragal line to the inferior border of the mandible and antero-posteriorly from commisure of the lip to the line joining the outer canthus to the gonion. Swelling was hard and firm, associated with pain on palpation but without any history of discharge. Patient had no complaint of dysphagia, trismus, dysphonia, fever, chills, loss of weight and parasthesia. Intraoral examination revealed obliteration of the left labial vestibule in relation to mandibular posterior teeth extending from #35 to distal aspect of #38 region. **(Figure 1)**

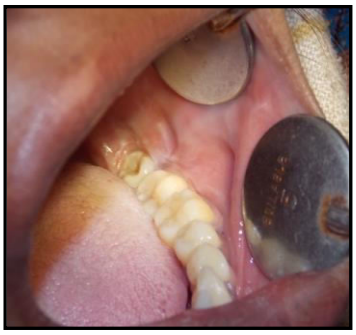

Figure 1: Intraoral view showing the vestibular obliteration with respect to left posterior mandibular region.

Orthopantamogram (OPG) revealed a multilocular radiolucency involving the lower left posterior region of the jaw **(Figure 2)**. Radiolucency was seen involving the left body of rami of mandible, extending from #34 region to the condylar process measuring about 6 X 3 cm in size with bony septa in between the radiolucent area giving it a soap bubble appearance. Superiorly radiolucency was seen causing knife edge resorption of left lower posterior teeth(#35, #36, #37, #38). Inferiorly the radiolucency was extending 1 cm above the inferior border of the mandible.

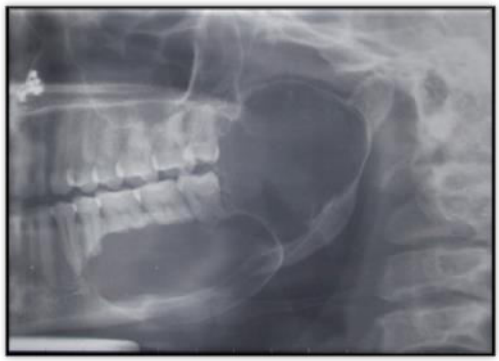

Figure 2: Orthopantamogram showing multilocular radiolucency involving the lower left posterior region of jaw.

Based on clinical and radiological features a provisional diagnosis of ameloblastoma was arrived keeping in view other multilocular radiolucencies like central giant cell granuloma, odontogenic keratocyst, aneurysmal bone cyst and hyperparathyroidism.

Incisional biopsy was performed and histologically the section revealed numerous small discrete islands of tumor mass in mature connective tissue stroma **(Figure 3)**. The islands were composed of peripheral layer of tall columnar ameloblast like cells which enclosed a central mass of polyhedral stellate reticulum like cells. Some of the tumor islands show cystic degeneration while in some there was evidence of squamous metaplasia of stellate reticulum like cells **(Figure 4)**. Few central cells gave a granular appearance. The intervening stroma sowed proliferating fibroblasts, focal aggregates of chronic inflammatory cells and blood vessels.

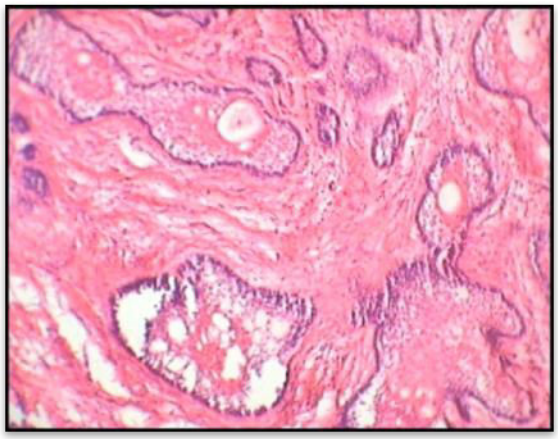

Figure 3: Ameloblastic follicles in mature connective tissue stroma (H&E, X10).

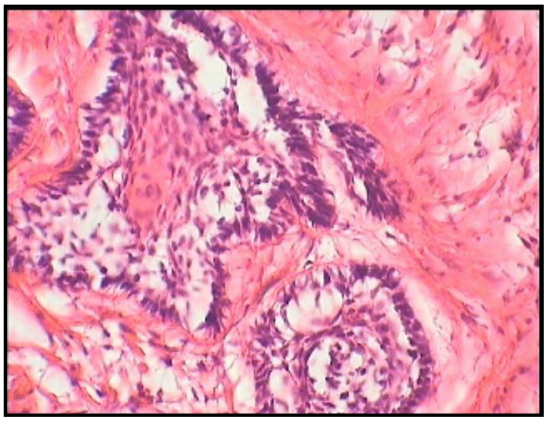

Figure 4: Ameloblastic follicles showing pallisading arrangement of peripheral cells and central stellate reticulum like cells along with squamous metaplasia (H&E, X40)

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Final diagnosis of ameloblastoma with follicular variant was given based upon histological and radiographic findings.

Treatment plan implemented was surgical resection followed by placement of reconstruction plate. Left submandibular incision was given 2 cm below the inferior border of mandible after dissection of platysma and superficial layer of deep cervical fascia. Lateral surface of mandible was exposed, #38 was extracted and resection was accomplished through the socket of the extracted tooth. After severing all muscular attachments specimen was removed **(Figure 5)**. Thorough irrigation and IMF was done followed by suturing.

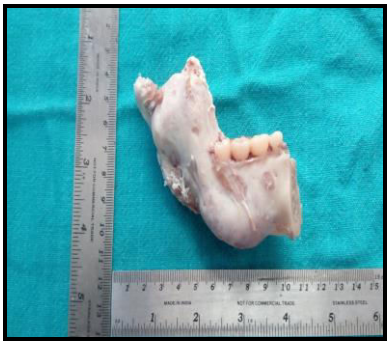

Figure 5: Resected hemimandibular section

DISCUSSION

Ameloblastoma is often an aggressive and destructive tumor with the capacity to attain great size, erode bone and invade adjacent structures.[11] In the mandible (80% of ameloblastoma), 70% occurs in the area of molars, 20% in the premolar region and 10% in the anterior region.[12] In the case presented, follicular ameloblastoma was found in the left body of mandible extending from #34 region uptill the condylar process, in a 30 year old female which is in concordance with the epidemiological findings and location of ameloblastoma. Majority of patients of ameloblastoma are asymptomatic and symptoms appear after the expansion of the tumor. Tumor involves both buccal and lingual cortical plate, cause expansion of the bone with the growth of the tumor and show resorption of the roots of the affected teeth.[12] In the case presented here, patient had asymptomatic swelling on the lower left posterior region of jaw which showed resorption of the roots of the involved teeth (#35 #36 #37 #38) radiographically. In the case reported, on OPG; a soap bubble appearance, multilocular radiolucency could be seen which is in concordance to the typical radiographic presentation of ameloblastoma.

Histologically ameloblastoma compises of a moderately to densely mature fibrous connective tissue stroma. The epithelial component of the tumor proliferates in the form of follicles, islands, strands and cords within the mature connective tissue stroma. A prominent budding growth pattern is seen, with small, rounded extensions of epithelium projecting from larger islands, recapitulating the various stages of enamel organ formation.[13] Tall columnar cells with hyperchromatic nuclei forms the peripheral boundary of the tumor follicles. Nuclei tend to be oval to round in shape and the nuclei of adjoining cells are roughly in same location within the cytoplasm giving it a characteristic palisading appearance. The palisaded nuclei are oriented away

from the basement membrane area of the cell, and a small clear vacuole can be seen between the nucleus and basement membrane. The characteristic features of ameloblastoma such as the peripheral layer of tall columnar cells with hyperchromasia, reverse polarity of nuclei and subnuclear vacuole formation mimic the normal embroyologic development of tooth bud at the stage of enamel matrix formation[14] emphasising on the concept of odontogenic tumors recapitulating phylogenesis.

According to Reichart PA et al[9], follicular ameloblastoma is the most commonly encountered variant, in which the peripheral cells resemble ameloblasts and preameloblasts and enclose a central mass of polyhedral loosely arranged cells resembling the stellate reticulum. Some tumor islands undergo cystic degeneration of stellate reticulum. Formation of intrafollicular cystic is considered due to deficiency in absorption and diffusion of nutritive elements(coming from the perifollicular blood capillaries) to the centre of the cellular islands, causing their degeneration by nutritive insufficiency, since the neoplastic growth causes extremely large follicles. Polarization of the nuclei at the cellular end facing the stellate reticulum. This causes the cells of the peripheral layer of the follicles to remove nutririve elements from the interior of the cellular elements and not from connective tissue facing the other cellular extremity. Nutritive competition can cause metabolic deficiencies for the cells of the stellate reticulum, which explains the degeneration of the central cells of the islands and consequent formation of cystic cavities. [15]

In the case presented histologically the section revealed numerous small discrete islands of tumor mass in mature connective tissue stroma. Majority of the islands showed a typical follicular pattern along with cystic degeneration in some, squamous metaplasia in others and central granular cell appearance in few. Though it resembled acanthomatous and granular cell type but majority pattern was follicular; therefore the final diagnosis of ameloblastoma of follicular variant was concluded upon. Most of our findings were in concordance with the data given in literature.

Histopathologically it could be differentiated from other multilocular radiolucencies like central giant cell granuloma, aneurysmal bone cyst, odontogenic keratocyst and hyperparathyroidism. Surgical excision is the most preferred treatment for ameloblastoma. According to various authors[16-18], surgery is the only treatment for ameloblastoma, due to its resistance to radiation therapy. Surgical plan should be influenced by whether the lesion is in maxilla or mandible, as the maxillary lesion behave differently from mandibular. Due to high content of cancellous bone in maxilla it facilitates the spread of ameloblastoma whereas the density of cortical bones in mandible limit the spread of the tumor. Regardless of the form of treatment plan patient should be put up on long term follow up because the tumor has high recurrence rate and once the tumor recur the treatment become more challenging.[2] In the present case left mandibular resection was done followed by placement of reconstruction plate. Six months follow up till date has been uneventful

Ameloblastoma is an aggressive odontogenic tumor. Based upon the condition of the individual patient, treatment plan is selected. Cases of ameloblastoma should be carefully studied to correlate the histologic pattern with biologic behaviour so that slight changes in histology can be detected which could predict the aggressive nature of the tumour. Early and prompt diagnosis of the lesion and timely surgical intervention can improve the prognosis.

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Source of support: Nil **Conflict of interest:** None declared