Follicular Ameloblastoma: A Case Report

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ABSTRACT

An ameloblastoma is benign and locally aggressive an odontogenic tumor of epithelial origin. Histologically it resembles the epithelial odontogenic apparatus, such as enamel organ and dental lamina, in some respects; however, the detailed mechanism of oncogenesis, cytodifferentiation, and tumor progression remains unknown. This tumor comprises about 1% of tumors and cysts arising in the jaws. Because of its slow-growing nature, there is often a delay in the diagnosis. Treatment decisions for ameloblastoma are based on the individual patient situation and the best judgment of the surgeon. This case report describes a case of an ameloblastoma in the mandible, which radiographically appeared as a multilocular radiolucency and histologically was predominantly follicular with a few plexiform areas.

KEYWORDS: Ameloblastoma, Follicular Ameloblastoma, Plexiform ameloblastoma

INTRODUCTION

Odontogenic cysts and tumors represent a surprisingly diverse group of pathologic lesions of the jaws and overlying soft tissues. The origin of ameloblastoma is not known with certainty, but in concert with concepts of neoplasia in general, it is likely the result of alterations or mutations in the genetic material of cells that embryologically are preprogrammed for tooth development. It is very aptly described by Robinson as being a tumor that is "usually unicentric, non-functional, intermittent in nature, anatomically benign and clinically persistent".[1] According to World Health Organization, three or four subtypes of ameloblastomas can presently be distinguished: Cassic Solid/ Multicystic Ameloblastoma (SMA), Unicystic Ameloblastoma (UA), Peripheral Ameloblastoma (PA), Desmoplastic Ameloblastoma (DA), including so-called hybrid lesions of ameloblastoma (HLA). Its incidence, combined with its clinical behavior, makes ameloblastoma the most significant odontogenic neoplasm to oral and maxillofacial surgeons.[2] The incidence of ameloblastoma is equal to the incidence of all the other odontogenic neoplasms combined excluding odontoma. The most common appearance of ameloblastoma on radiograph is that of a multilocular cystlike radiolucency, surrounded by a radiopaque border. However, unilocular radiographic appearances are also observed.[3] As seen with nearly every odontogenic neoplasm, the ameloblastoma may occur centrally within the bone or peripherally, without an intraosseous component, in the soft tissues overlying the alveolar ridge. Intraosseous lesions outnumber peripheral lesions by at least a 9:1 margin.[2]

In the present paper, we describe a case of solid multicystic ameloblastoma - predominantly follicular pattern mixed with few areas of plexiform pattern in a 42-year male patient. A comprehensive review of literature is also added.

CASE REPORT

A 42-year old male patient reported with a complaint of swelling in lower left a back region of the jaw since 3-4 months. The patient gave no relevant medical history. About 4 months back a small pea sized swelling was noticed by the patient on the left side of the jaw and it gradually increased to the present lemon size. (Figure no.1)

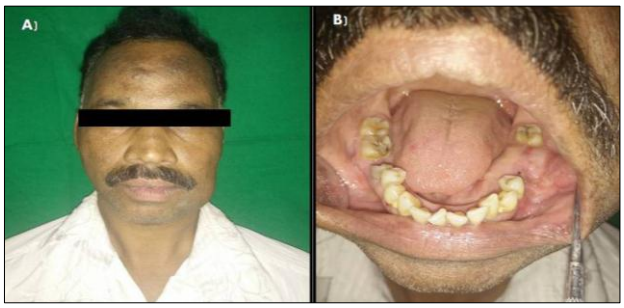

Figure 1: A) Extraoral photograph, B)Intraoral photograph

Orthopantomograph revealed a multilocular expansile radiolucency extending anterior posteriorly from mesial of 33 to distal of 38 and superoinferiorly from alveolar ridge descending down expanding the lower border of the mandible with a displacement of mandibular nerve canal inferiorly. The internal structure consisted of septae dividing the radiolucency into multiple locules with larger locules giving a soap bubble appearance inferiorly, smaller locules giving honey comb appearance superiorly and a few septae were seen emanating from center to

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periphery giving a spider web like appearance too. An intraoral periapical radiograph revealed a knife edge external resorption of the roots of 34, 35 and 37.The occlusal radiograph showed bicortical plate expansion with marked scalloping at the peripheral border. Cone beam computerized tomographic view further showed the expansile nature and the displacement of mandibular nerve inferiorly. It was surprising that the patient had no complaints of any altered sensation in that area.(Figure no.2, 3)

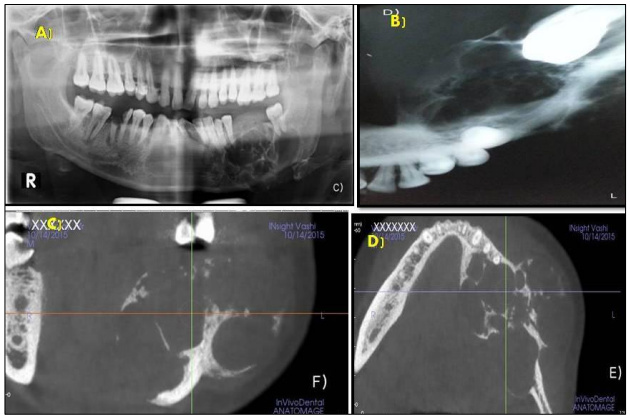

Figure 2: Photograph A)Orthopantomagraph, B) Occlusal view, C) CBCT coronal view D) CBCT Axial view

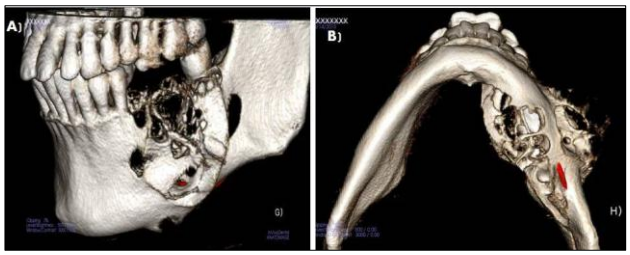

Figure 3: 3D Reconstruction view

Based on clinical and radiographic features, a provisional diagnosis of ameloblastoma was made, and an incisional biopsy was performed under local anesthesia after which the tissue was sent for histopathological examination. Histopathological examination revealed the presence of ameloblastic follicles in a fibrous connective tissue stroma. Following this, surgical excision was performed with complete enblock resection of the lesion under general anesthesia followed by reconstruction. There were no post operative complications and the wound healed uneventfully.

Macroscopically, the resected mass was grayish black in color showing multicystic appearance, grayish black in color. The specimen was solid at its periphery but was cystic in the center suggesting degeneration of the tumor mass.

Microscopically, the section showed ameloblastic follicles in a fibrocellular connective tissue stroma. The peripheral cells of the follicles were tall columnar ameloblast like cells, demonstrating reversed nuclear polarity and central stellate reticulum like area. Some of these follicles also showed cystic degeneration in the center.(Figure no.4-A) Also, one area showed a plexiform pattern with long anatomizing cords of ameloblastomatous epithelium bounded by columnar to cuboidal ameloblast like cells.(Figure no.4-C) The marginal area of the lesion revealed bony infiltration. (Figure no.4-D)As histopathology predominantly showed a follicular pattern, final diagnosis of solid multicystic ameloblastoma of follicular type was made.

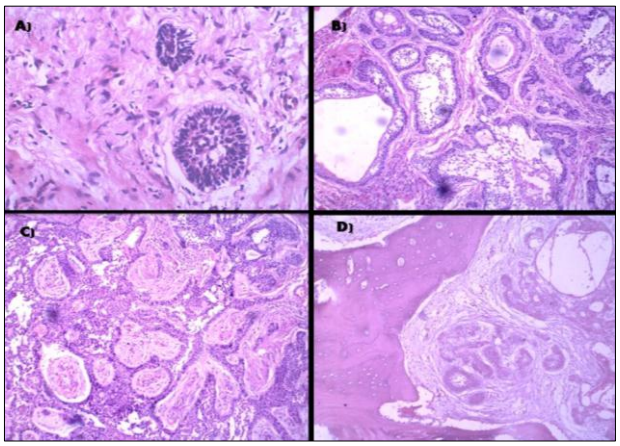

Figure 4: Photograph A) H & E (10x) showing histopathology of incision biopsy, B)H& E (10x) showing follicular pattern with areas of cystic degeneration, C)H & E (10x) plexiform pattern, D) H & E (10x) infiltration of bone by ameloblastic follicles.

DISCUSSION

Odontogenic tumors are lesions derived from epithelial, mesenchymal and/or ectomesenchymal elements that still are, or have been, part of the tooth-forming apparatus.[4,5] Ameloblastoma is a rare odontogenic tumor accounting for around 1% of all the cysts and tumors in the jaw. The term ameloblastoma was coined by Churchill in 1933 and the first detailed description of this lesion was by Falkson in 1879. [3] Ameloblastoma appears in the age group 20-50 years with equal frequency between sexes, [6,7] although a higher frequency in females than in males has been described.[8] Clinically, it frequently manifests as a painless swelling, which can be accompanied by facial deformity, malocclusion, ulceration and paraesthesia of the affected area. The site predilection for ameloblastomas are about 5 times more common in the mandible than in the maxilla.[9] Radiographically, the tumor may be unilocular or multilocular, with a tendency for expansion.[10] Unicystic lesions, present more commonly as unilocular radiolucencies. The internal structure varies from totally radiolucent to a mixed radiolucent-radiopaque caused by presence of bony septae which creates internal compartments, that give rise to soap bubble, spider-like, honeycomb appearances. The appearance of septae on the radiograph usually represents differential resorption of the cortical plate by the tumor and not actual separation of tumor portions.[11] Ameloblastomas have a tendency to cause extensive root resorption and teeth may be displaced apically.

In our case, the patient was a male, in the fourth decade of life. Clinical examination revealed a large, expansive

mass in the body of the mandible which was hard, painless on palpation and covered by normal mucosa with no altered sensation. Radiographically multilocular expansile radiolucency with all three appearances coexisting in the same lesion makes this case unique. External root resorption with scalloped margins at periphery were also noted.

There are several histopathological subtypes-follicular, plexiform, acanthomatous, desmoplastic, granular cell, and basal cell pattern, that may exist singly or as a combination of two or more types.[12] Follicular and plexiform are the commonly encountered variants accounting for 32.5% and 28.2% respectively; followed by the acanthomatous subtype 12.1% while desmoplastic is extremely uncommon with incidence rates ranging from 4-13%. Follicular ameloblastoma consists of discrete follicles with similarity to the stellate reticulum of enamel organ and with the varying quantity of tissue stroma. Because the follicular subtype is the most common variant, some pathologists believe that the acanthomatous, granular cell, basal cell, and desmoplastic variants are subsets of the follicular ameloblastoma .[9]

In our case, we observed bony infiltration in marginal areas; this could be correlated with various factors secreted by the ameloblast cells. Abdel sayed et al., (2004) found an increased expression of the parathyroid hormone-related protein (PTHrP) in ameloblastoma and suggested that it has a role in local bone resorption and also provided an explanation for infiltrative growth and destructive behavior of ameloblastoma. Also, other authors found that the RANKL, tumor necrosis factor-α secreted by ameloblast cells could induce the osteoclastogenesis, which in turn provide space for it to expand.[13] The growth pattern of ameloblastoma can be classified into two broad categories: (1) conventional ameloblastoma and (2) unicystic ameloblastoma.[14] In the conventional ameloblastoma, the neoplasm grows in island, strand, and cord-like patterns. In areas where islands of ameloblastic epithelium attain large size, cystic degeneration in the center of the tumor an islands may be seen. This feature can be a prominent component of the neoplastic proliferation, to the point that cyst-like areas may be identified grossly at the time of surgery.[15] This was noticed in our case. Cystic degeneration in an otherwise solid ameloblastoma does not alter the prognosis or the incidence of recurrence, and it should not affect surgical treatment decisions. Islands of neoplasm at times can be found far from the main tumor mass and often are seen within trabecular spaces. These islands may hinder the surgeon's ability to completely remove the neoplasm when conservative forms of therapy are used. For this reason, block resection is often the treatment of choice, and surgical margins generally are established at a distance of at least 1 cm from the clinical or radiographic boundary of the neoplasm. The treatment of conventional ameloblastoma by curettage alone is associated with increased incidence of recurrence when compared with the recurrence rate after block resection. If a more conservative surgical option than block resection is chosen, mechanical or chemical fulguration of the margin is included in the surgical treatment plan to reduce the chance of recurrence.[16]

Hong et al. recently showed that the histopathological variant of an ameloblastoma is significantly associated with a recurrence. It was shown that the follicular, acanthomatous and granular cell types have a relatively high likelihood of recurrence. In contrast, the unicystic desmoplastic and plexiform types show a relatively low potential for recurrence.[17] So, Follicular Ameloblastoma is characterized by higher recurrence rate (29.5%) compared to plexiform ameloblastoma (16.7%) and acanthomatous ameloblastoma (4.5%).[18]

CONCLUSION

Ameloblastoma is considered to be a benign, but locally invasive odontogenic tumor with a high rate of recurrence. Treatment decisions for ameloblastoma are based on the individual patient situation and the judgment of the surgeon. Resection with some safe margin is the best primary method for treating solid/multicystic ameloblastomas to avoid recurrence.

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