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CASE REPORT

Plexiform Ameloblastoma : A Case Report and a Brief Review

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Abstract

Ameloblastoma is a true neoplasm of enamel organ type tissue which does not undergo differentiation to the point of enamel formation. It appears most commonly in the third to fifth decades with equal sex predilection. Multicystic ameloblastoma is the most frequent subtype while unicystic ameloblastoma can be considered as a variant of the solid or mul- ticystic. Ameloblastoma is usually benign in growth pattern but frequently invade locally and occasionally metastasize. Recurrence frequently appears after inadequate treatment. Here we present a case of plexiform ameloblastoma in a 30 year old female patient who presented with a swelling on the left side of mandible.(2018, Vol. 02;Issue 02: Page 9 - 14)

Key words: Ameloblastoma, En bloc surgical excision, Mandible, Plexiform, Recurrence

Introduction

According to the WHO in 1992, ameloblas- toma is a benign but locally invasive poly- morphic neoplasm consisting of proliferat- ing odontogenic epithelium, which usually has a follicular or plexiform pattern, lying in a fibrous stroma (1). It represents 1% of all tumors and cysts that involve the max- illomandibular area (2). Ameloblastoma was first described by Cusack in 1827. Churchill coined the term ameloblastoma in 1930 (3). Ameloblastoma occurs in all areas of jaws, but the mandible is the most commonly affected area. Within the man- dible, the molar-angle-ramus area is com- monly involved and they are occasionally associated with unerupted third molar teeth (4). Radiographically, an ameloblas- toma can be unilocular or multilocular ra- diolucent lesion with a honeycomb or soap bubble appearances. Based on histo- pathology, ameloblastoma is classified into: follicular, acanthomatous, granular cell, basal cell, and plexiform (5). Follicu- lar and plexiform ameloblastomas are the most common, with incidence rates of 27.7% and 21.1% respectively, followed by acanthomatous and the granular types
(6). Treatment of ameloblastoma should involve surgical removal of complete lesion followed by appropriate reconstruction.

We report a case of plexiform ameloblas- toma in a 30 year old female patient, elu- cidating its clinical features, differential diagnosis and management.

Case report

A 30 years old female patient reported to the Department of Oral Medicine and Ra- diology with a chief complaint of swelling in left lower back tooth region since 6 months. History of present illness revealed that patient was asymptomatic 6 months back when she experienced swelling in mandibular left posterior region. Initially swelling was smaller in size which gradu- ally increased to the present size and was constant past one month. The swelling was not associated with pain, trauma and any kind of discharge in that region. Med- ical and family history was non contribu- tory. General examination revealed that all vital signs were within normal range. On extra oral examination, a diffuse swelling was noted on left lower third of face ex- tending superoinferiorly 1 cm below the ear lobule and 0.5 cm into the subman- dibular region and anteroposteriorly 1 cm from commissural area to angle of mandi- ble (Fig 1).

Swelling was non tender, firm and skin over swelling appeared to be normal. No rise in temperature was appreciated over the swelling. Left submandibular lymph node was palpable and non tender. On in- tra oral examination, the swelling was noted in mandibular posterior region ex- tending from mesial surface of 35 till distal surface of 37 (Fig 2).

Fig 2: Intraoral view

It was oval in shape, approximately 1 cm x 1.5 cm in size and of normal mucosal colour. No discharge was evident. Vestib- ular obliteration could be seen along with buccal cortical plate expansion. The swell- ing was non tender, firm and non fluctu- ant on palpation. No mobility of the adja- cent teeth or decortications was evident. Based on the history and clinical exami- nation a provisional diagnosis of ame- lobalstoma on mandibular left region was made. Differential diagnosis of odonto- genic myxoma, central giant cell granu- loma, odontogenic keratocyst was given. The radiographic investigations (OPG) showed a unilateral, well defined, multi- locular radiolucency in mandibular left re- gion measuring about 2 cm x 3 cm in di- ameter, extending from distal surface of

33 to mesial surface of 38 anteroposteri- orly, superoinferiorly it extended till the inferior border of mandible (Fig 3).

Figure 3: OPG of the lesion

Lesion had corticated borders. There was displacement of 33 mesially and 37 dis- tally. Resorption of distal root of 36 was evident. There was displacement of infe- rior alveolar canal inferiorly towards infe- rior border of mandible. Surrounding bones appeared to be normal. Blood inves- tigation reports were within normal range. En-bloc resection of tumor was done and the specimen sent for histopathologic ex- amination. Histopathologic examination revealed epithelium arranged as a tangled network of anastomosing strands (Fig 4).

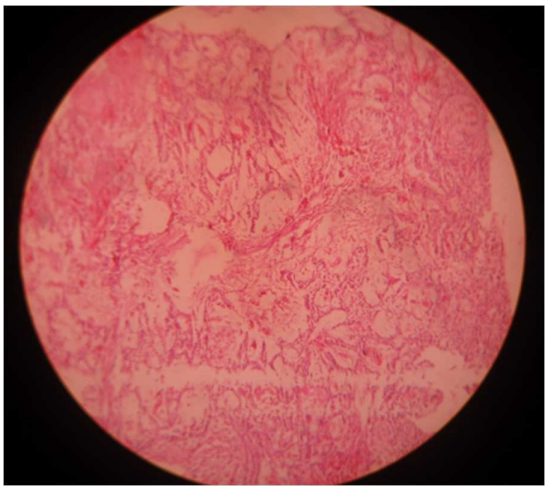

Figure 4: Histopathologic view of plexi- form ameloblastoma

The cords or sheets of epithelium were bounded by columnar or cuboidal amelo- blast like cells surrounding more loosely arranged epithelial cells. The supporting stroma was loosely arranged and vascular. Histopathological finding was suggestive of plexiform ameloblastoma. So the final diagnosis of plexiform ameloblastoma was given.

Discussion

The term “plexiform” refers to the appear- ance of anastomosing islands of odonto- genic epithelium in contrast to a follicular pattern. Initially some pathologists con- sidered it as a hyperplastic epithelial pro- liferation of the cystic lining instead of true ameloblastoma because it does not fulfills the histologic criteria as published by Vickers and Gorlin (7). To determine this Gardner studied histologic sections of 19 cystic lesions of the jaws exhibiting this plexiform pattern of epithelial proliferation in 1981, according to which he concluded that this pattern should be considered as ameloblastoma (8).

In the mandible, 70% are located in the area of the molars or the ascending ra- mus, 20% in the premolar region and 10% in the anterior region. About 10-15% of ameloblastomas are associated with a non-erupted tooth. In the present case, a large plexiform ameloblastoma found in the molar region of the mandible and it was not associated with a non-erupted tooth. Ameloblastoma appears with equal frequency between sexes, although higher frequencies in males have been reported occasionally (9). However, in our case, the patient was female. Clinically it manifests as a painless swelling with facial deform-

ity. In our case, clinical examination re- vealed painless swelling with facial de- formity on left side of patient. Radiograph- ically, ameloblastoma may be unilocular or multilocular with root resorption seen in some cases. Similar manifestation was encountered in our case. Differential diag- nosis of ameloblastoma involving mandi- ble includes odontogenic keratocyst, cen- tral giant cell granuloma, odontogenic myxoma, giant cell lesions of hyperpara- thyroidism, central haemangioma etc (Ta- ble 1) (9, 10).

Treatment of ameloblastoma varies from conservative approach to radical resec- tion. Different treatment options include enucleation followed by chemical cauteri- sation with carnoy`s solution, marsupiali- sation followed by enucleation, marginal resection or aggressive resection (10).

Table 1: Differential diagnosis of ameloblastoma

S.no	**Lesions**	**Characteristics features**
1.	Multilocular cysts	Male = Female Mean age is 16 years Mandible is predominantly affected Posterior mandible more commonly affected Soap bubble radiographic appearance
2.	Central giant cell granuloma	Female: Male = 2.4:1 Mean age group is 30 years Mandible is predominantly affected Anterior to second molar in mandible Soap bubble radiographic appearance Lesion usually crosses the midline
3.	Giant cell lesion of hyperpara- thyroidism (secondary)	Female: male= 2:1 Usual age group is 50-80 years. Mandible is predominantly affected Soap bubble radiographic appearance Serum calcium level is decreased Serum phosphorus level is increased Serum alkaline phosphatase level increased
4.	Cherubism	Male is more commonly affected Usual age group is 2-20 years Mandible, maxilla and zygoma is affected Ramus, molar region, sinus, orbital floor Soap bubble radiographic appearance

Continue…

S.no	Lesions	Characteristics features
5.	Odontogenic myxoma	Male = female Average age group is 25-35 years Ramus, premolar and molar area Tennis racket radiographic appearance
6.	Aneurysmal bone cyst	Male = Female 70% under age of 20 years Mandible is predominantly affected Ramus, premolar and molar area Soap bubble radiographic appearance
7.	Central haemangioma of bone	Female : Male =2:1 Usual age group is 10-20 years Mandible is more commonly affected Body and ramus Honeycomb radiographic appearance Pumping action of tooth
8.	Odontogenic keratocyst	Usual age group is 2nd to 3rd decade Mandible is more commonly affected No expansion is evident Have scalloped borders

Conclusion

Ameloblastoma is an aggressive tumor of odontogenic origin. Treatment decisions for ameloblastoma are based on the indi- vidual patient situation and the best judg- ment of the surgeon. Owing to the higher prevalence among odontogenic tumors cases of ameloblastoma should be studied carefully, correlating their histologic pat- tern with biologic behavior to detect subtle changes in histology that may predict ag- gressive behavior. Prognosis is good if an early diagnosis of the lesion is made with prompt surgical intervention.

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