BASAL CELL AMELOBLASTOMA: A RARE CASE REPORT AND REVIEW OF LITERATURE

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Abstract:

Ameloblastomas are an enigmatic group of oral tumors. The name implies a resemblance to cells of the enamel-forming organ. The general agreement that ameloblastomas are odontogenic in origin occurs largely on the basis of the histologic similarities of the tumor and the developing enamel organ. Basal cell ameloblastoma is believed to be the rarest histologic subtype. It is reported to occur primarily in peripheral location but has been seen intraosseously, albeit rarely. Till date only 6 cases of Basal cell Ameloblastoma has been reported in literature out of which five were in 3rd – 4th decade and only one case was reported in 2nd decade. Numerous cases of ameloblastomas have been reported in adults in the literature. However, only a few articles discuss ameloblastomas in children and adolescents. Considering the rarity of the lesion, we report here an interesting and unique case of Basal cell ameloblastoma of the mandible occurring in a very young patient.

Key Words: odontogenic tumors, Basal cell ameloblastoma, acanthomatous ameloblastoma, granular cell ameloblastoma

Introduction

Odontogenic tumors comprise of a complex group of lesions of diverse histopathologic types and clinical behaviour.[1]. Of all swellings of the oral cavity, 9% are odontogenic tumors, and within this group, ameloblastoma accounts for 1% of lesions . WHO defines it as a locally-invasive polymorphic neoplasia that often has a follicular or plexiform pattern in a fibrous stroma. Its behavior has been described as being benign but locally aggressive[2]

Ameloblastomas are benign locally aggressive, polymorphic neoplasms of proliferating odontogenic epithelial origin. The incidence, clinical & radiological features, behavior and histopathology of ameloblastomas have been extensively reviewed in numerous publications. Several histopathologic patterns of ameloblastomas are commonly described and include the follicular, plexiform, acanthomatous, granular cell and basal cell patterns. There appears to be rather general agreement that these variations in histopathology patterns do not have any significant bearing on prognosis except unicystic ameloblastoma because of less aggressive behavior and favorable prognosis.[3]

Ameloblastomas in young people (ie, those 19 years old and younger) are thought to be rare. They account for approximately 10% to 15% of all reported cases of ameloblastoma.[4]Ameloblastomas in children differ from adults, with a higher percentage of unicystic tumors[5]

Basal cell variant of ameloblastoma is the least common type. These lesions are composed of nests of uniform baseloid cells and histopathologically very similar to basal cell carcinoma of skin. No stellate reticulum is present in the centre portion of the nest. The peripheral cells around the nest tends to be cuboidal rather than columnar.[1]

Though very little information appears in literature either due to insufficient number of cases reported or due to variations in the clinical or radiological criteria, the pathologist may sometimes fail to differentiate it from intraoral basal cell carcinoma[3]

The purpose of this article is to present a case of rarest variant of ameloblastoma (Basal cell ameloblastoma) that has occurred in an unusual age and to provide a brief review of literature.

Case Report

A 12 year old male was referred to the government dental college and hospital, Raipur in with the chief complaint of painless swelling in relation to right lower mandibular 2 nd and 3rd molar region. Past medical, dental & family history of the patient was non contributory. There was no history of trauma, sinus opening or pus discharge. Extraoral examination revealed facial asymmetry due to swelling on right side of the face extending from 2cm anterior to ear till corner of mouth anteroposteriorly. Superioinferiorly, the swelling extended from infraorbital margin till inferior border of

mandible.(Fig 1) Clinical examination revealed firm to bony hard swelling in the right mandibular region with normal overlying skin. No lymphadenopathy on palpation was noted. Intra-oral examination revealed obliteration of buccal sulcus in the region of 43,44,45,47. History of extraction of 46 was given during incisional biopsy, the details of which are not available. Poor oral hygiene was noted. There was no intra oral sinus or ulceration.(Fig 2)

Investigations:

Radiographic report revealed multilocular radiolucency with root resorption in relation to 43,44,45,47. The inferior border of mandible was not traceable from 43,44,45,47. region. Third molar was seen in the right ramus area Left side of mandible appears normal. .(Fig.3) Hematological investigations [CBC, ESR] were all normal. From these clinical and radiographic findings a possible diagnosis of ameloblastoma or odontogenic keratocyst was made. To obtain a specific diagnosis an excisional biopsy was done as segmental resection of right mandible and the lesion was diagnosed as basal cell ameloblastoma.

Fig 1 Extraoral swelling on the right side of the face.

Microscopy:

H & E stained sections showed lesional tissue composed of uniform baseloid cells. The peripheral cells were columnar with reverse polarity. In few islands, cuboidal cells were also seen at the periphery. No stellate reticulum was seen in the central portion of the follicles. At one place these nests, tends to form a net like pattern with interconnecting strands. The lesional tissue was covered by fibrous capsule with fibrous septa intervening into it giving it a lobular pattern. (Fig 4,5,6,7)

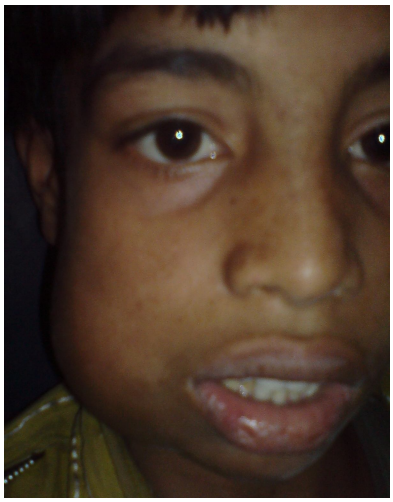


Fig 2 Intraorally obliteration of buccal sulcus in the region of 43,44,45,47 is seen

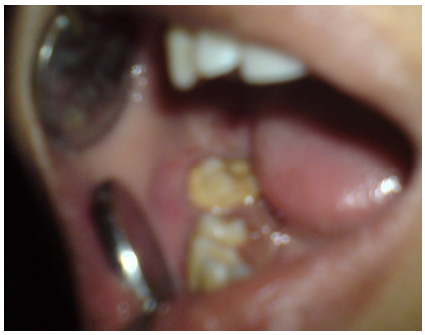


Fig 3 OPG reveals multilocular radiolucency with root resorption in relation to 43,44,45,47

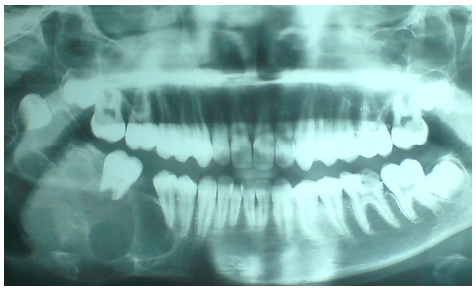


Fig 4 The lesional tissue covered by fibrous capsule with fibrous septa intervening into it giving it a lobular pattern (4x)

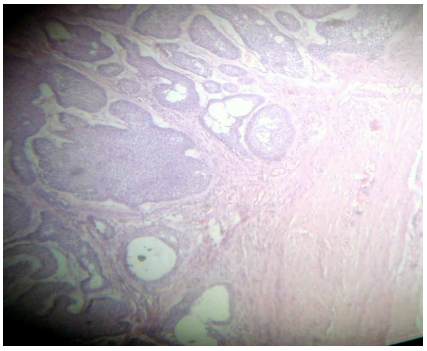


Fig 5 Baseloid cell island (10x)

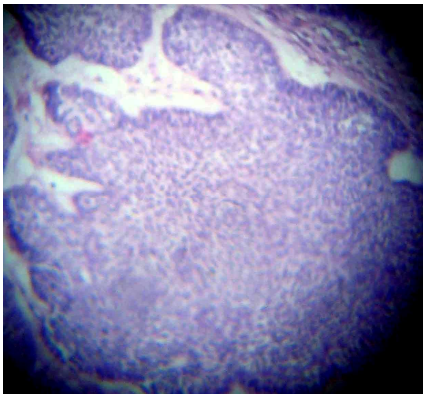
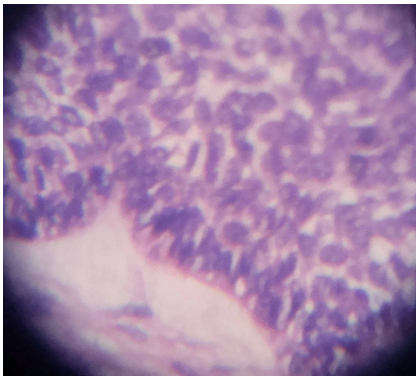


Fig 6 . The peripheral cells were columnar with reverse polarity(40x)



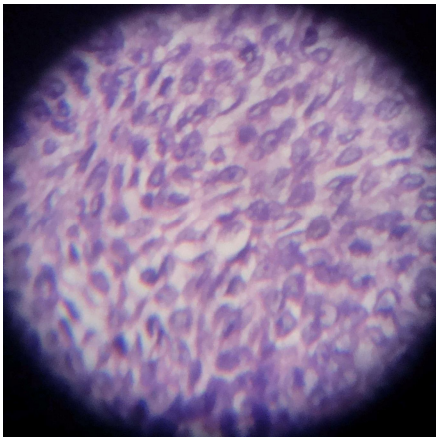


Fig 7 uniform baseloid cells with no stellate reticulum seen in the central portion of the follicles (40x)

Discussion

Ameloblastoma is chiefly a lesion of adults. It occurs predominantly in 4th – 5th decade of life and the age range is very broad[6]. The average age of patients with intraosseous ameloblastoma has been reported to be 39 years[2] The rare lesions occurring in children are usually cystic and appear clinically as odontogenic cysts. In this study, we document the occurrence of ameloblastoma in a significantly younger age group.

The basal cell ameloblastoma is a rare variant of ameloblastoma, which shows a remarkable resemblance to basal cell carcinoma and published cases of intraoral basal cell carcinoma most likely are basal cell ameloblastomas. Too few cases of basal cell subtype were available for valid statistical analysis but recurrence was reported.[7]

According to published articles on this variant, 6 cases have been reported in literature out of which five were in 3rd – 4th decade and only one case was reported in 2nd decade.[8,9,10,11] The average incidence of basal cell ameloblastomas was found to be less than 3%.

Basal cell ameloblastoma tends to grow in an island like pattern. The characteristic color gradation seen in other ameloblastomas is often difficult to appreciate in the basal cell subtype, because baseloid appearing cells rather then stellate reticulum like appearing cells occupy the central portion of the tumor

islands. The baseloid cells tends to stain deeply basophilic and are nearly equivalent in staining intensity with the peripheral layer of cells. The cells in the central portion may be polyhedral to spindle shape but stellate reticulum like areas are notably absent. The typical cellular morphology and nuclear orientation of the peripheral cells often are altered. They tend to be low columnar to cuboidal and often do not demonstrate reverse nuclear polarity with subnuclear vacuole formation. However, hyperchromatism and palisading of the nuclei normally are retained. [7]

Historically ameloblastoma has been recognized for over a century and a half. Its frequency, persistent local growth and ability to produce marked deformity before leading to serious debilitation probably account for its early recognition .Thus we conclude on the available information that basal cell ameloblastoma needs a perfect diagnosis based not only on clinical and radiographical appearance but also on histopathological findings. The lesion in the present case deviates from the usual ameloblastoma in terms of age and histological appearance. Therefore when the diagnosis in young people remains in doubt after clinical and radiologic examination, a biopsy is necessary. Long term follow-up at regular intervals after surgery is also recommended.

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