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**Research Article**

# AGGRESSIVE BASAL CELL AMELOBLASTOMA: A RARE DIAGNOSTIC ENIGMA

**Dr. Mandeep Kaur[1], Dr. Nitish Bhat[2], Dr. Ashanka Bhardwaj[3] and Dr. Roopav Nargotra[4]**

[1]Associate Prof Department of Oral Pathology & Microbiology, Indira Gandhi Govt. Dental College, Jammu
[2]Registrar, Department of Oral Pathology & Microbiology, Indira Gandhi Govt. Dental College, Jammu
[3]Registrar, Department of Oral Pathology & Microbiology, Indira Gandhi Govt. Dental College, Jammu
[4]Associate Prof, Department of Oral Surgery, Indira Gandhi Govt. Dental College, Jammu

**A R T I C L E I N F O**

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**A B S T R A C T**

Ameloblastoma is a slow growing benign locally invasive polymorphic neoplasm which is derived from remnants of odontogenic epithelium. It is primarily located centrally within the jaw bones and is the second most common odontogenic tumor after odontomes. Distant metastatis of an ameloblastoma to lungs or regional lymph node involvement do exists in rare cases. Radiographically areas of well defined bone destruction with smooth scalloped hyperostotic borders in early stages and perforated cortical plates in advanced stages are observed. Basal cell ameloblastoma is believed to be the rare histopathologic subtype variant of ameloblastoma with fewer features of typical peripheral palisading. The main aim of presentating the case report of basal cell ameloblastoma in 56 years old male patient in anterior maxillary region is because of rarity of occurrence and unique features.



## INTRODUCTION

Ameloblastomas are benign, aggressive tumors which mainly arise in the jaw region and are known to have an odontogenic origin. These can be locally invasive and are often known to invade and destruct the adjoining structures. They usually show a diverse histological pattern including follicular, acanthomatous, granular, clear cell, basal cell, plexiform, desmosplastic and a cystic variant (unicystic ameloblastoma).[1] Out of these variants is the basal cell ameloblastoma which presents as a tumor with a follicular pattern constituting of hyperchromatic basal cells within these follicles invading the connective tissue stroma. This histological variant of ameloblastoma is very rare and known to constitute 2% of the total found cases of ameloblastoma with a prevalence toward the mandibular region.[2,3] Occurrence in the mandible is found to be five times higher than that in maxilla with an average age of 38.9 years.[4]

Due to limited number of cases reported in literature it sometimes becomes difficult to differentiate basal cell variant of ameloblastoma from basal cell carcinoma. So a proper history of the patient and proper knowledge about the case is necessary to make the diagnosis. Here we report a rare case of basal cell variant of ameloblastoma that was found in the anterior region of the maxilla and involving the nasal floor.

**Corresponding author:* **Dr. Mandeep Kaur**
Associate Prof Department of Oral Pathology & Microbiology, Indira Gandhi Govt. Dental College, Jammu

### Case Report

A 56 years old male patient reported to the department of oral medicine and radiology with chief complaint of pain in the upper front teeth region since 2 1/2 months. Patient had a history of extraction of upper right and left central incisors since two months. Root canal treatment was done in upper left lateral incisor since 6 months. Intra oral examination revealed extracted upper right and left central incisor.(Fig I) There was a small soft swelling in the region with mild pain and tenderness. There were no significant palpable and tender lymph nodes. There was no associated discharge from the region. CBCT showed complete destruction of bone from upper right canine to upper left canine region. Cortical plate resorption and discontinuity of nasal floor was also observed.(Fig II) Radiograph showed a radiolucent lesion involving upper right canine to upper left canine. (Fig III) A provisional diagnosis of osteomyelitis of maxillary anterior region was made. Incisional biopsy of the lesion showed features of basal cell ameloblastoma. Resection of the lesion was planned under general anaesthesia. Partial maxillectomy was done and sample was sent for histopathological examination which showed basaloid follicles of odontogenic epithelium with basilar hyperplasia.(Fig IV)Typical peripheral palisading and reversal polarity was not prominent and central cell were polygonal in shape. (Fig V) The features were consistent with the diagnosis of Basal Cell Ameloblastoma. Patient was put under observation for a period of six months and no recurrence was found.

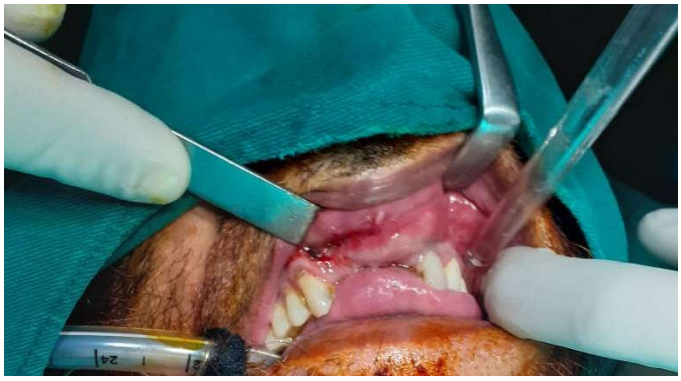

**Fig I** Swelling in maxillary anterior region

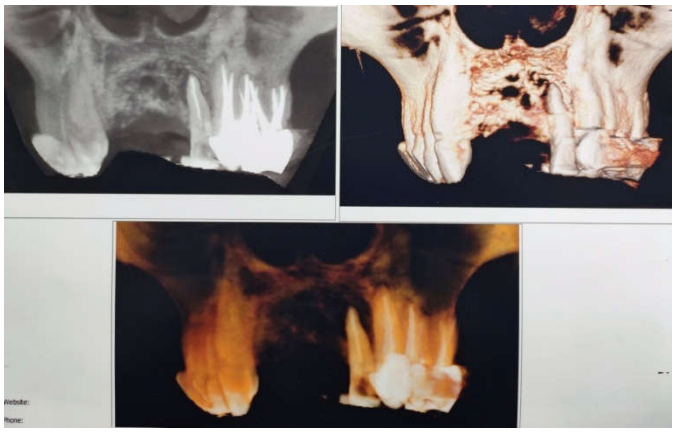

**Fig II** CBCT showing bone destruction in anterior region

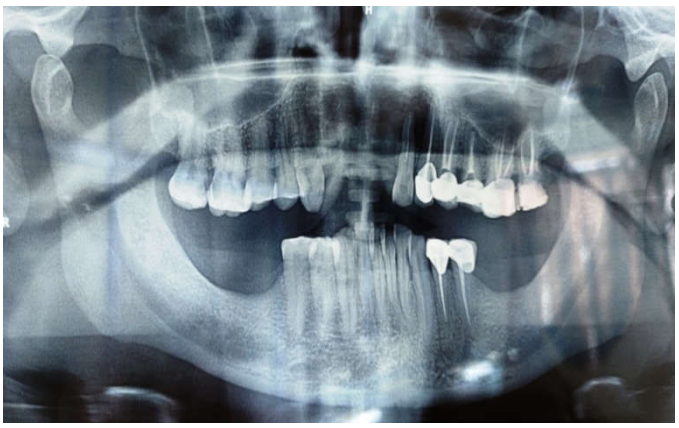

**Fig III** Radiolucent lesion

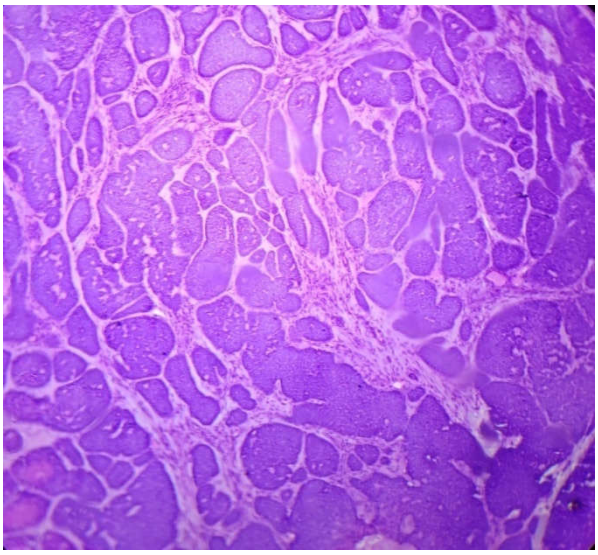

**Fig IV** Follicles showing basal hyperplasia (10X)

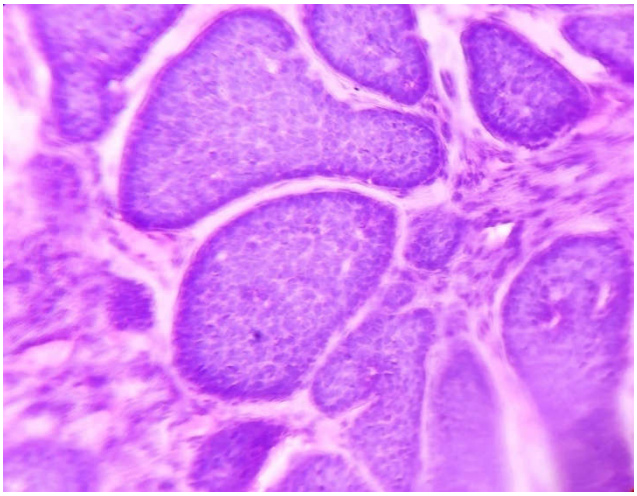

**Fig V** Basaloid cells at the periphery and polygonal cells in centre(40X)

## DISCUSSION

Ameloblastoma, previously known as admantinoma, is a benign but locally aggressive tumor of the jaw that tends to arise from the odontogenic epithelium. The first detailed description of this tumor was published by Falkson in 1879, but the term 'ameloblastoma' was coined by Churchill in 1933.[5]

Although, it is the most common odontogenic tumor, it represents only around one per cent of oral tumors. Around 80 per cent of ameloblastomas are reported to occur in the mandible.[4] The incidence of maxillary ameloblastomas has been variably reported in the literature. Although, it is generally accepted that only 20 per cent of ameloblastomas occur in the maxilla, some reports indicate an incidence as low as one per cent, and of those, 47 per cent occur in the molar region, 15 per cent in the antrum and the floor of the nose, 9 per cent in the premolar areas, 9 per cent in the canine regions and 2 per cent in the palate.[5,6] Iordanidis reported an incidence of 5 percent in his clinical practice.

Ameloblastoma is a developmental odontogenic tumor and possibilities for its development are from enamel organ, remnants of dental lamina, remnants of hertwig's sheath, epithelium of odontogenic cyst i.e, from Dentigerous cyst and odontomes and basal cell layer of oral mucosa. Other etiologic factors include trauma, oral sepsis, dietary deficiency, inflammation, viruses mainly polyoma virus and HPV type 16 or 18 and type 6, chronic irritation and infection with history of traumatic injury or extraction.[7]

Ameloblastomas begin as unilocular lesions and evolve into multilocular lesions, according to the fact that the mean age of patient with unilocular lesion is 26 years, whereas it is 38 years for multilocular ameloblastomas. Radiographically it may appear unilocular or multilocular with soap bubble or honeycomb appearance; buccal and lingual expansion of the cortex invariably accompanies ameloblastoma. Thinned and intact cortex shows egg shell appearance.[8]

The most common clinical symptom of the maxillary ameloblastoma is a painless swelling of the involved part of the jaw. Other symptoms that may occur are nasal obstruction, epistaxis, sinusitis or symptoms due to extension of disease into orbit. The maxillary bone has a richer blood supply compared to the mandible which could also contribute to the accelerated spread of this tumor in this location.[9]

Ameloblastoma shows different clinical features and radiographic appearances. Among histologic types of Ameloblastoma, follicular and plexiform patterns are the most common; less frequent variants are acanthomatous and granular cell types. Less common cellular variants are the desmoplastic Ameloblastoma, basal cell Ameloblastoma, keratoameloblastoma, papilliferous keratoameloblastoma, clear cell Ameloblastoma and unicystic Ameloblastoma. Except for the unicystic type, which has low recurrence rate, no significant differences in the behaviour of these variants have been observed. Few hybrid forms having combinations of histologic variants is also reported.[10]

The basal cell ameloblastoma is a rare variant of ameloblastoma, which shows a remarkable resemblance to the basal cell carcinoma and published cases of intraoral basal cell carcinoma most likely are basal cell ameloblastoma; only few cases of basal cell subtypes were available for valid statistical analysis. Basal cell ameloblastoma tends to grow in an island-like pattern. The characteristic color gradation in other ameloblastoma is often difficult to appreciate in basal cell type, because baseloid appearing cell rather then Stellate reticulum-like appearing cell occupies the center portion of the tumor island. The baseloid cells stain deeply basophilic and equivalent in staining intensity with peripheral layer of cells. These histological features are similar to our case.[11]

Maxillary ameloblastoma is more cellular than the mandibular counterpart and is further characterized by less distinct peripheral palisading, fewer columnar cells, frequent focal acanthomatous metaplasia and more cellular stroma.[9]

Among various histological variants, basal Cell Ameloblastoma is our case of discussion which shows equal gender predilection and mean age of occurrence is 40. In our case the age of the patient was 56 years. As basal cell Ameloblastoma is a rare entity its statistical data is also less, according to this data most common site of occurrence is mandible but in the case reported it occurred in maxilla which also makes it a rare entity. Basal cell Ameloblastoma is sometimes misdiagnosed as basal cell carcinoma which is an extraossous lesion as former is an intraosseous lesion. Intraoral basal cell carcinoma is the most common adnexal tumor and very rare occur in oral cavity. Basal cell Ameloblastoma showing a close resemblance to basal cell carcinoma histologically, however a demarcation can be made on immunohistochemical grounds by using Ber-EP4 marker. The prognosis & biological behaviour of basal cell ameloblastoma is not clear due to its rarity and few reported cases.[12]

## CONCLUSION

Basal cell ameloblastoma is a rare histopathologic subtype of ameloblastoma. The frequency of ameloblastoma and its persistent growth causes facial asymmetry so accurate diagnosis followed by adequate treatment is mandatory. The recurrence rate of basal cell ameloblastoma has not been reported due to limited cases in the literature. Basal cell ameloblastoma presented needs appropriate diagnosis based on the clinical, radiographical and histopathologic analysis as it bears close resemblance to BCC. Long term followup at regular intervals of this aggressive variant is required to establish the recurrence.

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