Basal Cell Ameloblastoma: Report of a Rare Case with Review of Literature
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ABSTRACT
Ameloblastoma is a benign tumour of odontogenic epithelial origin with several histological variants comprising of follicular, plexiform, acanthomatous, desmoplastic, granular cell and basal cell types. Basal cell ameloblastoma (BCA) is the rarest histologic subtype in which the tumour is composed of numerous odontogenic epithelial islands supported by fibrous connective tissue stroma. It exhibits variations in the clinical course and histological features compared with the classic type of ameloblastoma. Till date, only 11 cases of BCA have been reported in the literature. Considering the rarity of the lesion, we report here a unique case of BCA occurring in the 8th decade with review of the literature of this rare variant.

KEYWORDS: Basal cell ameloblastoma (BCA), Histological features, Mandible, Odontogenic tumours.

INTRODUCTION
Odontogenic tumours comprise of a complex group of lesions of diverse histopathologic types and clinical behaviour.[1] Ameloblastoma, though rare, is the second most common odontogenic tumour, accounting for 1% of all tumours of the head and neck region and approximately 11% of all odontogenic tumours.[2-4]

Ameloblastoma is a benign but locally aggressive odontogenic tumour of epithelial origin, arising from one of the following sources: (a) cell rests of the enamel organ, (b) epithelial rest of Malassez, (c) disturbances of the developing enamel organ, (d) basal cells of the surface epithelium, or (e) heterotropic epithelium in other parts of the body, especially the pituitary gland and epithelium of the odontogenic cysts.[5]

The clinical, radiological and histological features of ameloblastomas have been extensively studied and well reported in various scientific literatures. The tumour has been classified based on the clinical and radiographic features as multilocular, unicystic and peripheral types. Histologic variants of the tumour include follicular, plexiform, acanthomatous, granular cell, desmoplastic and basal cell types. Basal cell ameloblastoma (BCA) is the least common type and accounts for only 2% of the histologic types.[6]

Till today, only 11 cases of BCA have been reported in the literature. Due to its rare occurrence and their heterogeneous nature in terms of clinical and radiographical appearance, sometimes, the involved clinician fails to make an early diagnosis.

Therefore, this article aims to create awareness not only by reporting a case of the rarest variant of ameloblastoma (BCA) that has occurred in the 8th decade
of the age but also through its comprehensive review.

Case Report
A 72-year-old male patient was reported to the Care Multispecialty Dental Hospital with a chief complaint of swelling in the left lower jaw for the past 5 months. The swelling was associated with pain for the past 1 month. The patient gave a history of extraction of his lower left last tooth 1 year ago. Extraoral examination revealed a diffuse swelling on the left side of the face in the angle of the mandible region (Figure 1). Superioinferiorly, the swelling was extended from the line joining the tragus of the ear to the angle of the mouth till the inferior border of the mandible. Anteroposteriorly, the swelling was extending from 2 cm lateral to the angle of the mouth till the posterior border of the mandible. On palpation, swelling was bony hard in consistency. Intraoral examination revealed obliteration of the buccal vestibule in the molar region with ulceration of the mucosa in the third molar region (Figure 2). Radiographic examination of Orthopantomogram revealed a multilocular radiolucent lesion in the left angle of the mandible, extending from the second molar till the mandibular notch, involving the entire ramus of the mandible (Figure 3). Based on the clinical and radiographic findings, a provisional diagnosis of ameloblastoma was made. An incisional biopsy was performed under local anaesthesia, to obtain a final diagnosis. The histopathological examination demonstrated features of BCA. The resection of the lesion was done from the distal of the left first molar to the neck of the condyle and reconstruction was done with a 2.5-mm titanium reconstruction plate.
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Figure 4: Basaloid cells with no stellate reticulum seen in the central portion of the follicles

Figure 5: Peripheral cells are cuboidal to columnar with basaloid appearance

Histopathological examination of the lesion showed numerous odontogenic islands supported by fibrous connective tissue stroma. In some areas, the peripheral cells were columnar with reverse polarity and showed basaloid appearance and basophilic cytoplasm. In a few islands, cuboidal cells were also seen at the periphery. Stellate reticulum was not present in the central portion of the follicles. The connective tissue stroma was fibrovascular(Figures 4 & 5).

DISCUSSION

Ameloblastomas was first described by Falkson et al. in 1879 and the term was coined by Churchill in 1933[7]. It is a benign odontogenic tumour that tends to arise from odontogenic epithelium. But being locally aggressive, this tumour has the potential to cause deformity and debilitation if left untreated. The ameloblastoma can be classified based on the predominant histologic features as follicular, plexiform, granular cell, acanthomatous, desmoplastic and basal cell variants[8].

BCA is a rare variant of ameloblastoma and scarce information available in the literature either due to insufficient number of cases reported or because of variations in the clinical and radiological features. It shows a remarkable resemblance to the basal cell carcinoma and the pathologist may sometimes fail to differentiate it from intraoral basal cell carcinoma[9].

The review about this least reported variant, BCA was done through PubMed and Google search. Only articles in English were included for review. Through our review, we found only seven published articles comprising of 11 cases of BCA. Parameters, such as age, sex and involved site, were evaluated and compiled in the form given in Table 1.

In the literature, BCAs have been reported most commonly at 4–6th decade, in contrast with our case which has occurred in the 8th decade. But with respect to the site of occurrence, our case was in accordance with other reports, as it occurred in the posterior region of the mandible. As stated by Girish et al. [10] our case exhibited ulceration of oral mucosa, which remains the most common clinical feature for this tumour.

Histologically, BCA reveals multiple follicles and strands of odontogenic epithelium in connective tissue stroma. The peripheral cells of follicles exhibit hyperplasia and are basaloid in appearance similar to our case. Colour gradation of this type of variant becomes difficult as basaloid-appearing cell (in place of stellate reticulum-appearing cells) occupies the central portion of the tumour. The central portion in some of the follicles shows cystic degeneration[10]. Deep basophilic stains can be observed in both basaloid and peripheral layers of cells.[11] Though hyperchromatism and palisading of nuclei are retained, alterations can be noted in nuclear orientation of peripheral cell.[12]
Diagnosis of such rare variants should not be only on the basis of clinical and radiological appearances but has to be correlated with relevant histopathological interpretations. Surgical resection remains to be the principal modality of treatment for this tumour, taking into consideration with regards to its aggressive nature. As this tumour remains the least reported variant, information regarding recurrence rates is limited. After 2 years of postoperative follow-up at regular intervals, there are no signs of recurrence in our patient. However, considering the aggressive nature of this lesion, long-term follow-up has been recommended.

CONCLUSION

Because of variations in the clinical and radiological features, and histological similarity of BCA with basal cell carcinoma, it requires to be diagnosed carefully and accurately. Long-term follow-up at regular intervals after the appropriate surgical management is recommended for this aggressive variant of ameloblastoma.

REFERENCES


Table 1: BCA cases reported in the literature till date.

<table>
<thead>
<tr>
<th>Year</th>
<th>Author</th>
<th>Number of cases reported</th>
<th>Age/Sex</th>
<th>Site</th>
</tr>
</thead>
<tbody>
<tr>
<td>2013</td>
<td>Hemant Shakya et al.</td>
<td>1</td>
<td>50/F</td>
<td>Mandible – Right side, Posterior region</td>
</tr>
<tr>
<td>2012</td>
<td>Girish B Giraddi et al.</td>
<td>3</td>
<td>55/M, 17/F, 38/F</td>
<td>Mandible – Right side – from left lateral incisor (32) to right second molar (47) region; Mandible – Right side – from second molar to retromolar region; Maxilla – Right side – from second molar region to pterygoid plates</td>
</tr>
<tr>
<td>2010</td>
<td>Fatema Saify et al.</td>
<td>1</td>
<td>12/M</td>
<td>Mandible – Right side, Posterior region – between 2nd and 3rd molar region</td>
</tr>
<tr>
<td>2006</td>
<td>Kehinde E Adebiyi et al.</td>
<td>2</td>
<td>4th Decade/M, 4th Decade/M</td>
<td>Mandible, Mandible</td>
</tr>
<tr>
<td>2005</td>
<td>Hirota M et al.</td>
<td>1</td>
<td>17/F</td>
<td>Maxilla – Right side, Anterior region</td>
</tr>
<tr>
<td>1999</td>
<td>Iordanidis et al.</td>
<td>1</td>
<td>63/F</td>
<td>Maxilla – Right side, Posterior region – between 2nd premolar and 3rd molar region</td>
</tr>
<tr>
<td>1987</td>
<td>Kameyama Y et al.</td>
<td>2</td>
<td>3rd Decade, 7th Decade</td>
<td>Maxilla, Mandible</td>
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