Basal cell ameloblastoma—review of literature with report of three cases

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ABSTRACT

The ameloblastoma is the most common epithelial odontogenic tumor of the jaw with several histologic variants viz. follicular, plexiform, acanthomatous, desmoplastic, and granular cell and basal cell types. The basal cell ameloblastoma is a rare histological variant which tends to demonstrate microscopic features similar to cutaneous basal cell carcinoma and basaloid squamous cell carcinoma. In the current article we report three cases and review the literature of this rare tumor.

Keywords: Basal cell ameloblastoma.

INTRODUCTION

The odontogenic tumors are a complex group of lesions which exhibit diverse clinical behavior and histopathologic variations. Ameloblastoma, an odontogenic tumor, is a benign, locally aggressive tumor arising from the odontogenic ectoderm. Several histologic patterns of ameloblastoma are commonly described which include follicular being the most common, plexiform, acanthomatous, granular cell, basal cell, and desmoplastic patterns, although the histopathologic variations do not have significant bearing on the prognosis. The basal cell variant of ameloblastoma accounts for only 2% of the histologic types¹ and majority of the cases have been shown to occur in the mandible.² Apart from the primary manifestation as basal cell ameloblastoma, a desmoplastic ameloblastoma can also convert into a more aggressive basal cell ameloblastoma as reported by Makoto Hirota.³ One of the cases in our series started as dentigerous cyst and later progressed to basal cell ameloblastoma. The microscopic appearance of basal cell ameloblastoma is similar to that of basaloid squamous cell carcinoma and cutaneous basal cell carcinoma.⁴ In this article we report two cases in the mandible and one in the maxilla of basal cell type of ameloblastoma and discuss relevant literature related to this rare tumor.

CASE REPORT 1

A 55-year-old male patient complained of swelling in the right lower jaw for 1 year. The swelling was associated with pain

for the past 1 month. It was not associated with any discharge. The swelling was hard in consistency which extended clinically from the right second molar to the left lateral incisor of the mandible. The swelling showed ulceration intra-orally (Figure 1) in the buccal vestibule. Radiographic examination revealed multilocular radiolucency extending from the right first molar to the left lateral incisor. Root resorption was seen of the first molar and the two premolars. Under local anesthesia, an incisional biopsy was done. The histopathology reported a basal cell ameloblastoma. The resection of the lesion was done from the distal of the right third molar to the left canine, followed by reconstruction with a 2.5-mm titanium reconstruction plate. The resected specimen was sent for histopathologic examination. The section revealed each follicle lined by hyperchromatic odontogenic epithelial cells showing basaloid pattern. Few follicles also showed basal cell hyperplasia. After 3 years of follow-up, there was no recurrence.

CASE REPORT 2

A 17-year-old female patient reported with a swelling in the lower right jaw for 3 months. The swelling was not associated with pain or any discharge. The swelling was hard in consistency which extended from the lower right second molar to the retromolar region. Radiograph showed unilocular (Figure 2) radiolucency below the lower right second molar to the retromolar region. An incisional biopsy revealed dentigerous cyst. Enucleation of the lesion was done under local anesthesia and the patient was kept under observation. Three years later, the

Figure 1 (A) Intra-oral clinical photograph. (B) Resected specimen. (C) Panoramic image showing multilocular radiolucency from right lower canine to right lower first molar, root resorption seen in right lower first and second premolar and first molar. (D) Histological examination shows each follicle lined by hyperchromatic odontogenic epithelial cells showing basaloid pattern.

patient reported back with mild swelling and pain in relation to the previously operated site. Radiograph revealed multilocular radiolucency in relation to the previous lesional site. An incisional biopsy was done which revealed ameloblastoma. The patient was taken for resection of the lesion under general anesthesia. The lesion was resected from the distal of the second premolar to 2 cm from the mandibular notch. Reconstruction was done with iliac crest bone graft and a 2.5-mm titanium reconstruction plate. The resected specimen on histopathologic examination revealed aggressive basal cell ameloblastoma. The patient has been kept under observation. Eight months postoperatively, there are no signs of recurrence.

CASE REPORT 3

A 38-year-old female patient complained of growth in the upper right back tooth region for 6 months. The patient gave a history of extraction of the upper right second molar 6 months before, after which she noticed the growth. On

clinical examination, an erythematous overgrowth in the region of the right maxillary tuberosity region was seen (Figure 3). The lesion involved both buccal and palatal mucosa. There was pain and tenderness on palpation of the lesion. The right submandibular lymph node was palpable and tender. There was no associated discharge from the lesion. Radiograph showed a radiolucent lesion distal to the upper right second molar, and computed tomography (CT) scan revealed well-defined lytic soft tissue lesion extending from the right upper second and third molar measuring about $3.3\text{ cm} \times 2.2\text{ cm} \times 2.2\text{ cm}$ with extension into the right posterior maxillary antrum. Posteriorly, the lesion extended to the pterygoid plates. An incisional biopsy of the lesion showed basal cell ameloblastoma. The resection of the lesion was planned under general anesthesia. Partial maxillectomy was done and the specimen was sent for histopathologic examination which showed ameloblastoma with follicles exhibiting basilar hyperplasia and basaloid appearance of the odontogenic epithelial cells consistent with basal cell ameloblastoma. Seven months postoperatively, there are no signs of recurrence.

Figure 2 (A) Panoramic image showing unilocular radiolucency suggesting dentigerous cyst. (B) Completely healed lesion 1 year after enucleation. (C) Multilocular radiolucency seen 2 years after enucleation. (D) Histopathology showing features of basal cell ameloblastoma.

Figure 3 (A) Intra-oral clinical photograph. (B) Computed tomography scan showing radiolucent lesion in the right maxilla. (C) Panoramic radiograph showing radiolucency in the right posterior maxilla. (D) Histopathology showing features of basal cell ameloblastoma.

DISCUSSION

Ameloblastoma is a benign but locally aggressive odontogenic jaw tumor arising from the odontogenic epithelium. The maxillary ameloblastoma tends to behave in a more aggressive fashion with rapid direct extension beyond the site of origin, compared with that in the mandible.³ The ameloblastoma can be classified according to predominant histologic pattern as follicular variant which is the predominant variant, plexiform, granular cell, basal cell, desmoplastic, and acanthomatous type.¹

The basal cell ameloblastoma occurs predominantly in the fourth decade, mean age being 57.3 years.¹ In this article, this variant is seen in a 17-year-old patient. Most of the published cases report that basal cell variant presented predominantly in males but we present two cases in females. The posterior mandible has a definite predilection for this variant although cases have been reported in the maxilla.^{3,5,6}

The most common presenting symptom in basal cell variant was swelling followed by ulceration of the oral mucosa as seen in two of the three cases reported. The radiologic appearance of this variant is most commonly multilocular–mulicystic pattern as seen in these cases.

Histologic tumor reveals multiple follicles, strands, and cords of odontogenic epithelium in connective tissue stroma that is minimal. The peripheral cells of follicles exhibit hyperplasia and are basaloid in appearance. The central portion in most of the follicles shows cystic degeneration.

Ameloblastomas are known to arise from a variety of cells: (1) cell rests of the enamel organ, (2) epithelium of odontogenic cysts, (3) disturbances of the developing enamel organ, (4) basal cells of the surface epithelium, or (5) heterotrophic epithelium in other parts of the body of epithelial origin.⁷ However, for the first time, we have come across a basal cell ameloblastoma arising from a dentigerous cyst. In the literature, one case has been reported arising from a desmoplastic ameloblastoma.³

The similarity in the histopathology of this tumor with basal cell carcinoma might point toward the aggressiveness of this lesion. Further studies are required to establish this.

The surgical resection of the tumor remains the mainstay treatment modality. The maxillary ameloblastoma needs to be treated more aggressively due to the potential for spread of

the tumor due to increase in the cancellous spaces in the maxillary bone and the proximity to vital structures.⁵ Segmental resection was done in two cases of the mandible; and partial maxillectomy was done for the lesion in the maxilla.

The recurrence rate for histologic variants has been reported the highest for follicular type. The recurrence rates for basal cell ameloblastoma have not been reported due to the limitation in the number of cases. There is no recurrence seen in the three cases reported in this article also.

The frequency of ameloblastoma and its persistent growth causing facial asymmetry requires it to be diagnosed accurately followed by adequate management. Basal cell variant like reported in the article needs appropriate diagnosis based not only on clinical and radiologic principles but also on sound histopathologic analysis. Long-term follow-up at regular intervals of this aggressive variant is necessary to establish the recurrence rate.

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