Basal Cell Ameloblastoma: A Rare Histological Variant of an Uncommon Tumor

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Abstract

Ameloblastomas are an inscrutable group of oral tumors. Basal cell ameloblastoma is a rare variant of ameloblastoma with very few cases reported until date. The tumor is composed of more primitive cells and has less conspicuous peripheral palisading. It shows remarkable similarity to basal cell carcinoma, basal cell adenoma and intra-osseous adenoid cystic carcinoma. This report describes the case of a 27-year-old male with an ameloblastoma in the right posterior mandible. Orthopantomography computed tomography and finally histopathological examination directed us toward the confirmatory diagnosis of basal cell variant of ameloblastoma. Considering the rarity of the lesion and histological paradox regarding its diagnosis, we report here an interesting and rare case of basal cell ameloblastoma of the mandible with emphasis on differential diagnosis from other entities with basaloid differentiation having varying prognosis. After surgery, long-term follow-up at regular intervals is recommended as no sufficient statistical information regarding the behavior of this tumor is available.

Keywords: Ameloblastoma, basal cells, desmoplastic, odontogenic

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Access this article online

Quick Response Code:

Website: www.nigerianjsurg.com

DOI:
10.4103/1117-6806.152730

Introduction

Odontogenic tumors comprise of a complex group of lesions of diverse histopathologic types and clinical behavior. Ameloblastoma represents 1% of all tumors and cysts that involve the maxillomandibular area and about 10% of odontogenic tumors. WHO defines it as a benign, but locally invasive polymorphic neoplasm consisting of proliferating odontogenic epithelium, which usually has a follicular or plexiform pattern, lying in a fibrous stroma.[1] Adebiyi *et al.* analyzed histological variants of ameloblastoma and found that follicular ameloblastoma was the most common histological type (64.9%), followed by plexiform meloblastoma (13.0%), desmoplastic (5.2%) and acanthomatous (3.9%) while the basal cell variant accounted for (2.6%) cases.[2] Basal cell ameloblastoma is a rare variant with only a few cases described till date with insufficient information about his biological behaviour. It is reported to occur primarily in peripheral locations, but has been seen intraosseously, albeit rarely.[3] Histologically, basal cell ameloblastoma consists of darkly stained cells distributed predominantly in a trabecular pattern with little evidence of palisading at the periphery.[2] Although the small number of cases makes a precise assessment difficult, the tendency for recurrence or malignant transformation in this variant is considered to be the same as other variants of ameloblastoma.[4] The microscopic features of basal cell ameloblastoma, however, are similar to those of several malignant tumors, including basaloid squamous cell carcinoma (BSCC),[5,6] cutaneous basal cell carcinoma (BCC) and solid-type adenoid cystic carcinoma (ACC).[1] The pathologist may sometimes fail to differentiate it from intraoral BCC leading to erroneous diagnosis. Hence, careful analysis of all the clinical and pathological data should be carried out before arriving at final diagnosis.

Case Report

A 27-year-old male patient complained of painless swelling in relation to left lower mandibular posterior region. Past medical,

dental and family history of the patient was unremarkable. There was no history of trauma or pus discharge. Extra-oral examination revealed facial asymmetry due to swelling on the left side of the face extending anteroposteriorly from 3 cm anterior to the ear till the corner of mouth and superior-inferiorly, extended from infraorbital margin to inferior border of mandible. Clinical examination revealed firm to bony hard swelling in the left mandibular region with normal overlying skin. Intra-oral examination revealed obliteration of buccal sulcus in the region of 34, 35, 37 as seen in Figure 1. Orthopantomograph as observed in Figure 2 revealed multiple multilocular radiolucencies in the left side of mandibular body and ramus area involving coronoid and condylar process. There was thinning of inferior border of mandible. Posterior border of ramus and right side of mandible appeared normal. Coronal slice of computed tomography (CT) scan showed expansion of medial and lateral border of the left side of ramus with thick and curved bony septa and homogenous density [Figure 3a]. Axial slice CT at level of mandible showed soft tissue mass in left side of mandible with complete destruction of buccal and lingual plates and remnant of bone within mass extending into adjacent soft tissue with loss of flat plane [Figure 3b]. From these clinical and radiographic findings, differential diagnoses of ameloblastoma or odontogenic keratocyst were considered. Fine needle aspiration was carried out, but it did not yield any fluid.

To obtain a specific diagnosis, an incisional biopsy was done. H and E stained sections showed lesional tissue composed of nests of uniform basaloid cells. No stellate reticulum was seen in the central portion of the nests. The peripheral cells were cuboidal to short columnar with reversal of polarity. Fibrous septa divided the lesional tissue giving it a lobular pattern [Figure 4]. Based on the available supporting evidence, final diagnosis of basal cell Ameloblastoma was given.

Under general anesthesia, tumor mass was exposed buccally and lingually and osteotomy cut was placed and completed buccally and lingually and tumor mass was excised with bone margin of 1.5 cm and also by encompassing surrounding healthy soft tissue. Free fibula osteocutaneous graft of 16 × 4.5 × 3 cm in dimension was harvested from right leg along with peroneal artery and vessels. Antibiotics, analgesics and antiinflammatory

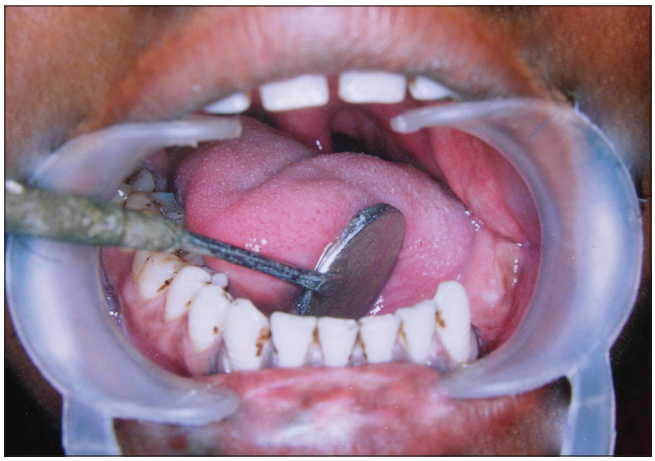

Figure 1: Intra-oral photograph showing swelling in the left mandibular region with obliteration of buccal sulcus

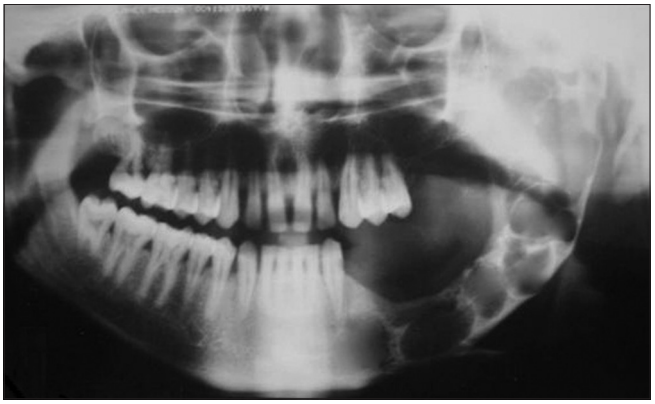

Figure 2: Orthopantomograph showing multiple multilocular radiolucencies in left side of body of mandible and ramus area involving coronoid and condylar process

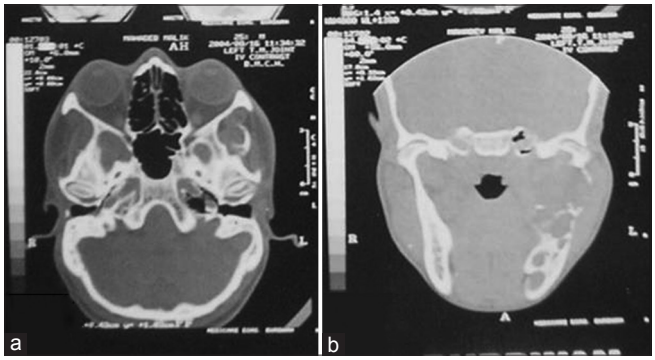

Figure 3: (a) Coronal slice of computed tomography (CT) scan showing expansion of medial and lateral border of left side of ramus with thick and curved bony septa and homogenous density; (b) Axial slice CT at level of mandible showing soft tissue mass in left side of mandible with complete destruction of buccal and lingual plate and remnant of bone within mass extending into adjacent soft tissue with loss of flat plane

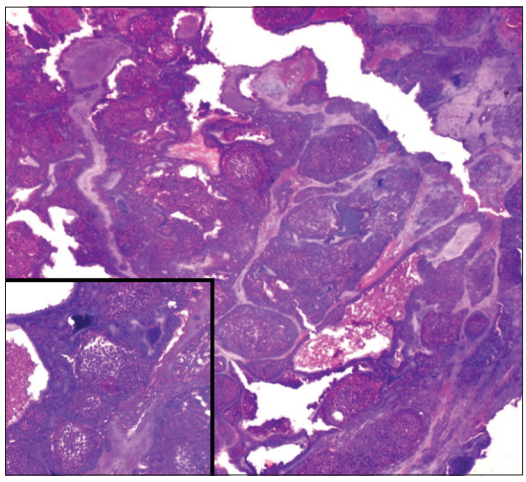

Figure 4: Photomicrograph showing nests of uniform basaloid cells (H and E, scanner view) and inset showing basaloid cells with fibrous septa divided the lesional tissue giving it a lobular pattern (H and E, ×10)

drugs were given postoperatively. Histopathological examination of excised specimen reconfirmed the initial diagnosis. Surgical wound healed uneventfully, and sutures were removed on 10th postoperative day. The postoperative recovery of the patient was uneventful and followed-up of 2 years revealed no evidence of recurrence.

Discussion

Published data showed that the ameloblastoma is a rare benign odontogenic tumor, representing only 1% of all tumors and cysts of maxilla and mandible. It attacks either sex and can attack at any age, 50% occurs between age of 20 and 30 years. Adebiyi *et al*. clinicopathologically analyzed the histological variants of ameloblastoma and observed that basal cell variant of ameloblastoma occurs most commonly in mandible of males with 31-40 year age as common age of occurrence.[2,3] The age, sex and site in the present case of basal cell ameloblastoma described here agree with the data given in the literature.

The basal cell ameloblastoma is a rare variant of ameloblastoma.[3] Histological feature is apparently benign, constitutes a puzzling paradox. Basal cell ameloblastoma tends to grow in an island like pattern. The basaloid appearing cells in basal cell ameloblastoma tend 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 as seen in other ameloblastomas are often altered. They tend to be low columnar to cuboidal and usually do not demonstrate reverse nuclear polarity with sub-nuclear vacuole formation. However, hyperchromatism and palisading of the nuclei normally are retained.[7] Our histological findings were in unison with the described features. Basal cell ameloblastoma shows a remarkable resemblance to BCC. Ide *et al*.[6] disagreed with the diagnosis of oral BCC, the case earlier reported by Wedenberg *et al*.[5] believing it to be a basal cell ameloblastoma on the basis of available clinical and pathological data. They believed that it may often be difficult to establish an accurate diagnosis of basal cell ameloblastoma. They claimed that at least two alleged cases of oral BSCC reported by Wedenberg *et al*.[5] and de Araújo *et al*.[8] published in literature do not satisfy most of the strict criteria for the diagnosis.

If basal cell type of growth occurs in the jaws, special care is necessary to distinguish between this type of ameloblastoma and an intra-osseous (ACC.[1] BSCC and ACC are a well-defined, highly malignant tumor both clinically and histologically. Hence, distinction between basal cell ameloblastoma and these differentially considered lesions is of paramount importance. Immunohistochemical analysis of lesion with histological perplexity can direct pathologists toward confirmatory diagnosis. Table 1 shows expression of different markers in basal cell ameloblastoma, BSCC, ACC and BCC.[2,5,6,9,10]

Recently Sandra *et al*. immunohistochemically compared proliferative activity of different variants of amelobalstoma using monoclononal antiproliferating cellular nuclear antigen (anti-PCNA) antibody and monoclonal anti-Ki-67 antibody which are considered as reliable markers for cell proliferation, the authors found that the basal cell solid multicystic ameloblastoma (SMA) had the highest labeling indices for both PCNA and Ki-67, indicating that the basal cell type is the most actively proliferating type and therefore the most immature cells in an (SMA).[11]

It is very difficult to predict the prognosis of Basal cell type of ameloblastoma as very few cases of basal cell subtype has been reported for valid statistical analysis, but recurrence of this lesion has been reported. There has been some debate regarding the most appropriate method for management of ameloblastomas. Conservative modalities include curettage, enucleation, cryotherapy, cautery, laser usage, radiotherapy, chemotherapy and radical approach encompasses marginal, segmental and composite resections.[12] Some authors favor the conservative approach as they believe that ameloblastomas though, locally invasive, are essentially benign in nature. Therefore, they should be treated as such because of serious cosmetic, functional and reconstructive problems associated with it. Proponents of the radical approach to the treatment of ameloblastomas argue that, albeit these tumors are histologically benign, but are locally aggressive, and their clinical behavior may be regarded as lying somewhere between benign and malignant lesions.[13] Satisfactory results have been achieved either with radical treatment or more conservative approaches whereas enucleation and curettage have

Table 1: Immunohistochemical expression of different markers in basal cell ameloblastoma, BSCC, adenoid cystic carcinoma, basal cell carcinoma

Lesion	Immunohistochemical marker			
	Ber-EP4 (epithelial specific antigen)	C-kit (CD 117)	CK	Others
Basal cell ameloblastoma	Negative	Negative	Positive for AE1/AE3, KL1, 34, E12, and MNF116 CK; devoid of CK7, CK8, CK10, CK18, CK20, and EMA	-
BSCC	Negative	Negative	Positive for high molecular weight CK	Weak p63 positivity
Adenoid cystic carcinoma	Negative	Positive	Positive for CK 7, 14	Strong p63 positivity
Basal cell carcinoma	Positive	Negative	Predominant expression of K17 and the frequent expression of K8 and K19 little K6/K16 and K1/K10	Negative for EMA

EMA: Epithelial membrane antigen, BSCC: Basaloid squamous cell carcinoma, CK: Cytokeratin

been reported with the highest recurrence rate of 55-90% of the cases. Metastases following conservative management have also been reported.[12,13] In addition to being a radio-resistant tumor, the intra-osseous location of the ameloblastoma prevents the use of radiotherapy as an effective therapeutic option because radiation enhances the potential development of secondary tumors but may be performed in cases when surgery is not considered to be a method of choice.[12,13] There is a need to run more evidence-based clinical studies of clinical practice guidelines as there is a lack of complete consensus regarding appropriate treatment modality for ameloblastomas. It is very difficult to predict the prognosis of it as very few cases of basal cell subtype were reported for valid statistical analysis, but recurrence of it is reported.[10]

Conclusion

Rarity of basal cell variant of ameloblastoma in conjunction with atypical histological feature constitutes a puzzling paradox. Hence, diagnosis should be based not only on clinical and radiographic appearance, but also on histopathological findings. Long-term follow-up at regular intervals after surgery is recommended.

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How to cite this article: Sridhar M, Bhaskar Reddy LR, Kharat S, Mahesh BS, Gandi L, Mahendra A, *et al*. Basal cell ameloblastoma: A rare histological variant of an uncommon tumor. Niger J Surg 2015;21:66-9.

Source of Support: Nil, **Conflicts of Interest:** None declared.