Case Report

An unusual case report of basal cell ameloblastoma and a detailed review of literature

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Abstract

Ameloblastoma is an odontogenic tumour derived from the remnants of odontogenic epithelium. Ameloblastoma shows versatile clinical and histological variants. Basal cell ameloblastoma is a rare variant among all histopathological variants of ameloblastoma. We present a case of a 25-year-old male patient with painless swelling on the lower left side of the face for 1 year, which had gradually increased in size and histologically showed features of basal cell ameloblastoma. The aim of the present paper is to understand this rare variant of ameloblastoma and review the literature.

Keywords: Ameloblastoma, basal cell variant, odontogenic tumor

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INTRODUCTION

Ameloblastoma is stated to be the second most common locally aggressive tumour among all odontogenic tumors.[1] They may arise from odontogenic epithelium like cell rest of Malassez or cell rests of Serre, heterotopic epithelium in the pituitary gland, odontogenic cysts such as dentigerous cyst, odontomas and basal cells of the surface epithelium of the jaws. Ameloblastoma comprises 2%–3% of all pathologies of the jaw.[2] Ameloblastoma most commonly occurs in the mandible than the maxilla, the ratio being 5:1. The average age of occurrence reported is about 38.9 years.[3]

In the recent classification of the World Health Organization (WHO) 2017, ameloblastoma has been classified clinically as conventional, unicystic and peripheral variant, whereas histopathologically under conventional, it has been further classified as follicular and plexiform (being most common) and others (granular, basal cell variant, acanthomatous, clear cell variant etc.) To avoid any misperception, 'solid or multicystic amelobalstoma' term has been omitted as it was confused with unicystic. The desmoplastic variant is considered only a histopathologic variant than a clinical entity.[4] Granular cell ameloblastoma, clear cell variant of ameloblastoma and basal cell variant of ameloblastoma are considered rare variants.[5] Moreover, as the basal cell variant of ameloblstoma is an odontogenic variant (benign), it may share a similar pathological picture as basal cell carcinoma (BCC) or basaloid squamous cell carcinoma (BSCC) (malignant). As published data on basal cell variant of ameloblastoma is scarce, it is difficult to understand certain molecular pathology and diagnosis.[6] In the present paper, we present the basal cell variant – a

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rare histopathological variant of ameloblastoma along with a detailed review of the literature.

CASE REPORT

A 25-year-old male patient with a complaint of swelling in the left posterior part of the mandible for the last 1 year was referred by a surgeon. There was no relevant medical, dental, family or habit history of the patient. Extraoral findings were remarkable, and facial asymmetry was evident. The overlying skin was normal, and no signs of trauma or pus discharge were seen. Clinical examination suggested non-mobile, non-fluctuant diffuse and hard swelling extending from the inner canthus of the eye to the lower border of the mandible. [Figures 1 and 2] Intraorally, the buccal sulcus was obliterated. Radiographical findings seen in an orthopantomography were suggestive of diffuse radiolucency involving the body of the mandible and angle of the mandible, thinning of the inferior border of the mandible was seen, and the right-side mandible was normal. [Figure 3] Computed tomography showed expansion of cortical plates with well-demarcated borders. The medial and lateral borders showed homogenous density throughout. [Figure 4]. The provisional diagnosis was an odontogenic tumor or odontogenic keratocyst. An incisional biopsy was taken, and hematoxylin and eosin–stained section was reported as ameloblatoma by a general pathologist.

Under general anesthesia, the patient underwent a partial mandibulectomy extending from the premolar region to the neck of the mandible. The excisional tissue was referred for histopathological reporting. The excisional biopsy report was the same as the provisional diagnosis. The slide was referred to a private laboratory for a second opinion where we received hematoxylin and eosin–stained slide. The slide showed numerous follicles and islands containing basaloid cells in mature fibrous connective tissue stroma. At the periphery of the follicles, cuboidal to tall columnar cells were seen with hyperchromatic nuclei, and no reverse polarity of nuclei was seen. Cells in the center were basaloid. There was a lack of stellate reticulum-like cells in the center. Few cystic spaces were evident in the follicles. Overall, the picture was suggestive of the basal cell ameloblastoma. [Figures 5, 6 and 7] The patient was followed up for 1 and 1/2 years, and there was no recurrence.

DISCUSSION

Ameloblastoma was known as 'Admantinoma', the term 'Ameloblastoma' was coined by Churchill in 1933. It is

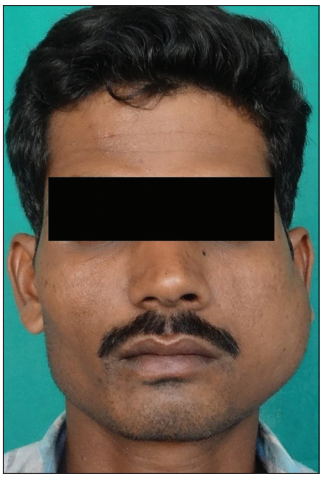

Figure 1: Clinical photograph showing diffuse swelling at the left posterior mandible from the inner canthus of the eye involving the inferior border of the mandible. (Anterior View)

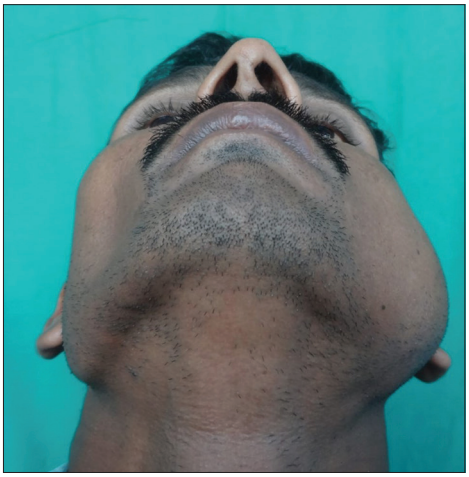

Figure 2: Clinical photograph showing diffuse swelling at the left posterior mandible from the inner canthus of the eye involving the inferior border of the mandible. (Inferior view)

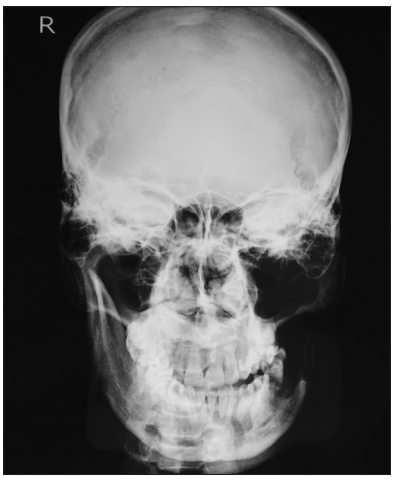

Figure 3: Orthopantomography showing diffuse radiolucency involving the body of the mandible and angle of the mandible, thinning of the inferior border of the mandible.

a benign odontogenic tumor arising from odontogenic epithelium which is aggressive in nature.[5] The first detailed report published was by Falkson 1879.[6] Literature suggests that about 80% of ameloblastoma cases are seen in the mandible and 20% in the maxilla. Basal cell variant of ameloblastoma is also more commonly seen in the mandible. Most cases reported are in the 4th decade.[7] Various hybrid variants of ameloblastoma are reported in the literature.[8] Basal cell ameloblastoma (BCA) is the rarest, it comprises only 2% of all histopathological variants of conventional ameloblastoma with only 28 cases reported till date. Hence, it is difficult to understand nature and clinicopathological relation.[9] We did a PubMed and google scholar database search for basal cell variant of ameloblastoma [Table 1].[2,7,10-23] We found that most of the reported cases of BCA were seen in the 3rd to 5th decade, and the maxilla to mandible ratio reported was 1:1.2 with no gender predilection.

Histopathologically, the tumour essentially consists of follicles and strands of basaloid cells. Cells at the periphery show hyperchromatic nuclei. The central cells of the islands or follicles are devoid of stellate reticulum-like cells and instead show basaloid cells only. These features were evident in our case too. [Figure 7] There is no columnar cell layer and palisading nuclei like other variants of ameloblastoma. Some cases might show cystic degeneration. Alterations in the peripheral cell nuclei can be seen.[9,20] Histopathologically, differential diagnosis can be intraoral basal cell carcinoma and basal cell adenoma.[2,25] Immunohistological (IHC) studies have been conducted on variants of ameloblastoma including six cases of BCA. Cytokines were consistent with all variants of ameloblastoma, but an elevation of p53 and ki-67 was evident in the basal cell variant. Two cases showed BRAF gene mutation.[22] Further studies on histopathological and immunohistochemical features of basal cell ameloblastoma are needed to understand its resemblance with basal cell carcinoma (BCC), as one is benign, and another is malignant with a concern of metastasis. Jawad and Abdullah conducted a study on basal cell variant of ameloblastoma and basal cell carcinoma using a panel of markers, and they found MMP-2 is higher in basal cell ameloblastoma whereas p53 and MMP-9 were increased in basal cell carcinoma.[25] Due to the scarcity of data, the exact nature of BCA cannott be predicted.[9]

Sandra *et al*.[26] conducted a study using immunohistochemical (IHC) markers, monoclonal anti-proliferating cell nuclear antigen and Ki-67 in all variants of ameloblastoma, they stated that in BCA these two labeling indices were highest than that of other variants of Ameloblastoma. Sridhar *et al*.[17] reported a case of BCA with a detailed review, they mentioned that it is needed to differentiate BCA, intraosseous adenoid cystic carcinoma, BCC and basal squamous cell carcinoma (BSCC). Hence, the

Table 1: Reported cases of basal cell variant of ameloblastoma with age, gender and location details

Author	Year	No. of cases reported	Age/Gender	Site
Kameyama *et al*.[24]	1987	2	3rd Decade, 7th decade	Maxilla, Mandible
Matsuo *et al*.[10]	1988	1	7th Decade (64)/Male	Maxilla
Iordanidis *et al*.[11]	1999	1	6th Decade (63)/female	Maxilla
Hirota M *et al*.[12]	2005	1	2nd Decade (17)/Male	Maxilla
Kehinde E Adebiyi *et al*.[13]	2006	2	4th Decade, 4Th Decade	Mandible
Fatema Saify *et al*.[7]	2010	1	2nd Decade (12)/Male	Mandible
Giraddi *et al*.[14]		3	6th Decade (55)/Male, 2nd Decade (17)/Female, 4th Decade (38)/Female	Mandible Mandible Maxilla
Shakya *et al*.[2]	2013	1	5th Decade (50)/Female	Mandible
Pendyala *et al*.[15]	2014	1	8th Decade (72)/Male	Mandible
Kumar *et al*.[16]	2014	1	8th Decade (72)/Male	Mandible
Sridhar *et al*.[17]	2015	1	3rd Decade (27)/Male	Mandible
Virmani *et al*.[18]	2015	1	4th Decade (37)/Male	Maxilla
Ghattamaneni *et al*.[19]	2015	1	3rd Decade (30)/Male	Maxilla
Kosanwat *et al*.[20]	2016	1	5th Decade (46)/Female	Maxilla
Lee *et al*.[21]	2018	1	3rd Decade (30)/Female	Mandible
You *et al*.[22]	2019	6	3rd Decade (22)/Female 5th Decade (42)/Male 5th Decade (42)/Female 4th Decade (37)/Female 9th Decade (82)/Female 5th Decade (48)/Male	Mandible Mandible Maxilla Maxilla Maxilla Maxilla
Kazakydasan *et al*.[9]	2019	1	1st Decade (08)/Female	Mandible
Mandeep Kaur *et al*.[8]	2020	1	6th Decade (56)/Male	Maxilla
Abrishami *et al*.[23]	2021	1	4th Decade (34)/Female	Mandible

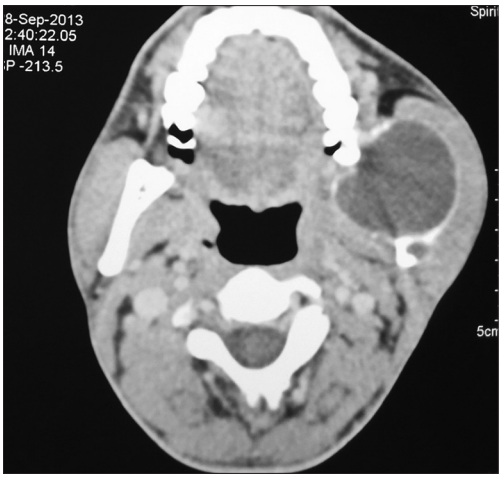

Figure 4: Computed tomography showing an expansion of cortical plates with well-demarcated borders. The medial and lateral borders show throughout homogenous density.

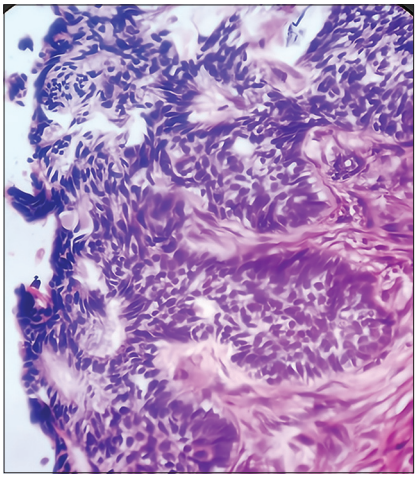

Figure 6: 10 × view showing basaloid cells in a follicle lacking reverse polarity of nuclei. The centre of the follicle is filled with hyperchromatic basaloid cells lacking stellate reticulum-like cells in the center.

expression of IHC markers in BCA are lack of CK7, CK8, CK10, CK18, CK20, EMA and positivity for MNF116, AE1/AE3, KL1,34, EL12.Whereas Ghattamaneni *et al*.[19] stated that CK19, amelogenin or enamelin can be used for differentiating BCA, BCC, and BSCC. They used CK19 IHC marker and found that it shows a diffuse positivity in BCA. We searched for papers where IHC markers in BCA were studied. [Table 2][9,17,19,22,25,26]

Treatment modalities of BCA are controversial due to the nature of ameloblastoma, which is benign but still aggressive. Enucleation, curettage, cautery, laser, chemotherapy, radiotherapy, and marginal, composite sectional resection are considered conservative treatment modalities.[17] Curettage and enucleation show the highest recurrence rates that is 55%–90%.[27] Radical resections having 1 cm resected margins show 0%–15% recurrence

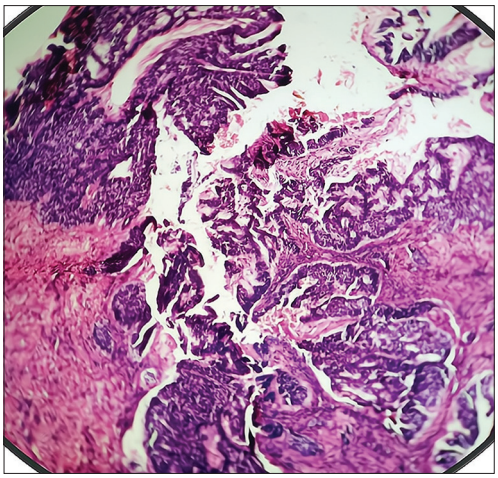

Figure 5: Scanner view showing islands of basaloid cells.

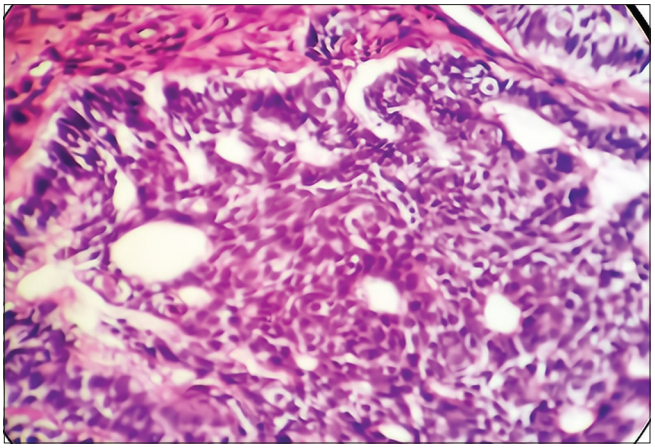

Figure 7: 40 × view of basaloid cells with few cystic spaces.

Table 2: Immunohistochemical markers positive in Basal cell ameloblastoma

Author	Year	Positive IHC Markers in Basal Cell Ameloblastoma cases
Sandra *et al*.[26]	2001	Highest labelling indices for proliferating cell nuclear antigen (PCNA) and Ki67
Sridhar *et al*.[17]	2015	Positive for AE1/AE3, KL1,34, E12 and MNF116 CK
Ghattamaneni *et al*.[19]	2015	Cytokeratin 19
Bajpai *et al*.[25]	2017	Bcr-EP4
You *et al*.[22]	2019	CK5, CK14, CK-Pan, 34βE12, CK19, Ki67, p53
Kazakydasan *et al*.[9]	2019	AE1/AE3

rate and is considered the best modality till date.[28] Exclusive treatment modalities for the basal cell variant are no different than conventional; however, the prognosis may be controversial due to the lack of reported cases.

CONCLUSION

Basal cell ameloblastoma is a rare variant of ameloblastoma. Its histopathological picture resembles basal cell carcinoma.

Hence, the diagnosis must be given considering clinical, radiographical, histopathological and by using IHC markers. In the present paper, we have tried to explain this rare entity with its review of the literature. This is an endeavour to report a case for a better understanding of its histopathological and immunohistochemical nature and treatment modalities.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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