CASE REPORT

Basal cell adenoma of maxillary sinus mimicking ameloblastoma

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ABSTRACT

Basal cell adenoma (BCA) is a rare basaloid tumor, with only 20% of cases occurring in minor salivary glands. Histologically, BCA is characterized by the presence of basaloid cells and may frequently be mistaken with canalicular adenoma, basal cell adenocarcinoma, adenoid cystic carcinoma and basaloid squamous cell carcinoma. Immunohistochemistry may aid in arriving at a final diagnosis as in the present case. Reported here is a case of locally aggressive BCA. Histologically, the lesion mimicked ameloblastoma and other entities which posed a diagnostic challenge. There are no reports of BCA presenting as an aggressive lesion available in English literature so far; moreover, merely a single case of BCA of maxillary sinus has been previously reported to the best of our cognition. This case report highlights the rarity of this tumor with regards to its site of origin, clinical behavior and histopathological mimics.

Key words: Ameloblastoma, basal cell adenocarcinoma, basaloid, maxillary sinus

INTRODUCTION

Basal cell adenoma (BCA) is a rare entity belonging to the group of basaloid tumors and was named so by Kleinssaser and Klein (1967). It was initially included in the group of nonpleomorphic adenomas of the salivary gland, more specifically "monomorphic adenoma" along with canalicular adenoma. Subsequently, the 1991 WHO classification of salivary gland tumors included it in the category of benign epithelial neoplasms as a discrete entity.

It comprises approximately 1–2% of salivary gland tumors. Over 80% of cases appear in major salivary glands, primarily in the parotid. Less frequently, it may involve minor salivary glands, with upper lip being the most common site, followed by buccal mucosa. It is a slowly progressive benign neoplasm which on computed tomography scan indicates a well-circumscribed lesion. Clinically, it usually appears as painless, firm and mobile slow-growing mass, more common in the 7th decade with a female predilection (2:1), except for the membranous type which bears an equal male:female distribution. It gives a uniform histologic appearance dominated by basaloid cells arranged in solid, trabecular, tubular or membranous patterns. Solid BCA is formed by small cells organized in a compact manner. In the trabecular and tubular subtypes, groups of cells exist in narrow bands and ductal structures or a combination of both. A membranous subtype is constituted by external cells in a stockade pattern and by an intense hyalinized basal membrane.[1]

CASE REPORT

A 70-year-old male patient reported with the complaint of the unhealed wound in the mouth for 6 months after he pulled out a loose upper right molar himself. He also reported a gradually increasing swelling and numbness over the right maxillary sinus region, blocked nostril and decreased hearing from the same side of the lesion. Extraorally, the patient had diffuse swelling over the right maxillary sinus region extending over the ala of the nose [Figure 1]. Intraorally, there was an

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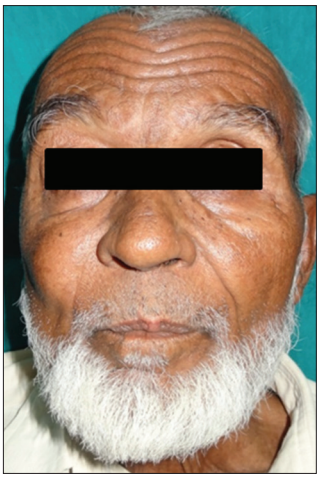

Figure 1: Extra orally diffuse swelling over the right maxillary sinus extending onto the ala of nose

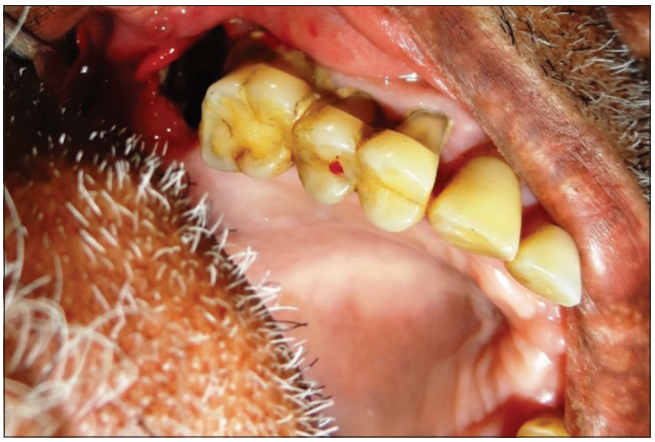

Figure 2: Intraoral examination revealed unhealed socket of 17 surrounded by everted margins and necrosed bone within

unhealed socket of 17 with necrosed bone inside, surrounded by everted margins [Figure 2].

Contrast-enhanced computed tomography revealed an ill-defined heterogeneously enhancing neoplastic mass of size 6.1 cm × 3.8 cm × 3.3 cm involving the right maxillary sinus and extending to ethmoid sinus, masticator space and infratemporal region causing destruction of the walls of the maxillary sinus. Medially, it involved pterygopalatine fissure and pharyngeal mucosal space. Posteriorly, it was abutting retropharyngeal space and laterally the ramus of mandible. No cervical lymphadenopathy was evident [Figure 3].

Incisional biopsy showed an encapsulated mass consisting of isomorphic basaloid cells forming trabecular and tubular structures in a scanty stroma. These structures were seen infiltrating into the overlying epithelium [Figure 4] and showed two cell populations: peripheral tall columnar and central round to ovoid cells. The central cells displayed discohesiveness giving stellate reticulum-like appearance in a considerable portion of the lesion. Some of the central areas showed the presence of mucous cells and mucin filled spaces [Figure 5]. At places, the tumor cells formed canalicular structures enclosing hemorrhagic spaces [Figure 6]. A provisional diagnosis of ameloblastoma and differential diagnosis of basal cell adenocarcinoma (BCAC), BCA and canalicular adenoma were made. Negative immunostaining for calretinin ruled out ameloblastoma [Figure 7]. Periodic acid–Schiff stain revealed patchy positivity at the periphery of islands and in the cytoplasm of the central cells indicating a salivary gland origin [Figure 8]. On immunostaining, strong positivity for calponin in the peripheral basaloid cells was suggestive of myoepithelial cells [Figure 9]. Cytokeratin (CK 19) positivity in the central cells and cells in cords was suggestive of luminal cells[2] [Figure 10]. Ki-67 index was <5% [Figure 11]. Final diagnosis of locally aggressive BCA was given based on the histopathologic and immunohistochemical findings. Because of the extensive lesion, the patient was referred to the higher center for treatment. However, patient refused treatment because of general illness.

DISCUSSION

BCA is a rare benign neoplasm characterized by the basaloid appearance of the tumor cells and absence of the myxochondroid stromal component as present in pleomorphic adenoma (WHO 2005). It was reported as a distinct entity for the 1st time by Kleinsasser and Klein in 1967. Batsakis is credited for reporting the first case in the American literature in 1972 and suggesting the intercalated duct reserve cell as the histogenetic source of BCA. Later studies endorsed the origin of BCA to myoepithelial and/or intercalated duct lineage.[3,4]

In the Armed Forces Institute of Pathology, series up to 75% of BCAs are reported in parotid gland, 5% in submandibular gland and 6% in intraoral location with upper lip being the most common site, followed by the buccal mucosa. An incidence of approximately 1% in the oral minor salivary gland has been reported by Takahashi *et al*.[5] It occurs most commonly in the 6th or 7th decade of life[1] In the present case, age of the patient was in accordance with the literature, whereas the site of neoplasm was rare and unique with only single case reported previously in English literature (2012).[6] The ill-defined and extensive nature of the lesion along with a patient's complaint of decreased hearing from the same side ear, blocked nostril and numbness over the involved region exhibited an aggressive nature of the lesion in contrast to the usual presentation of the BCAs.

Most of the imaging studies done on BCA concluded it of having well-defined and smooth marginal morphologies. A few cases have shown nonhomogenous enhancements with either cystic areas, linear bands or stellate-shaped areas of nonenhancement.[7,8] In the present case, ill-defined, nonhomogeneous enhancing mass causing local destruction a feature not previously reported in BCA so far raised the suspicion of malignancy.

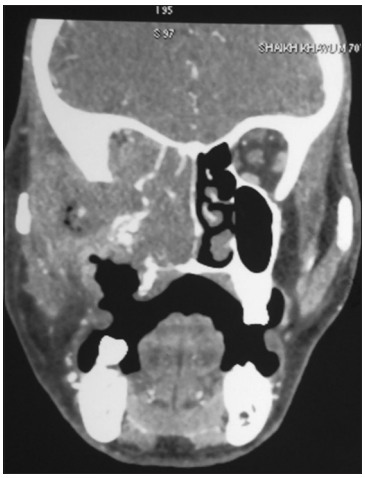

Figure 3: Contrast-enhanced computed tomography exhibiting nonhomogeneous enhancing lesion destroying walls of maxillary and ethmoid sinus, upper right alveolus and extending into masticator space and infratemporal region

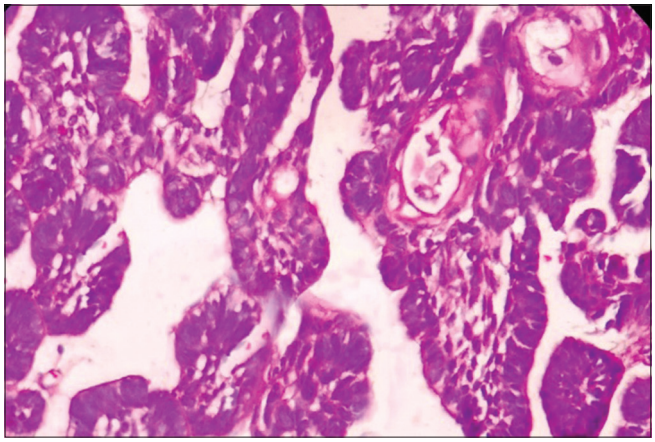

Figure 5: Peripheral palisaded arrangement of columnar cells and central loose cells showing stellate reticulum-like appearance with interspersed mucous cells (H&E stain, ×400)

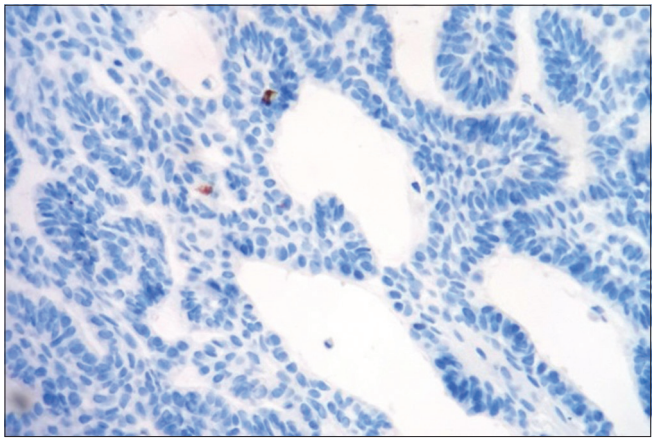

Figure 7: Calretinin negativity ruled out ameloblastoma (IHC stain, ×400)

Histologically, BCA is characterized by the presence of uniform and regular basaloid cells. These cells have two different morphologies and are blended. The first cell type is a basaloid

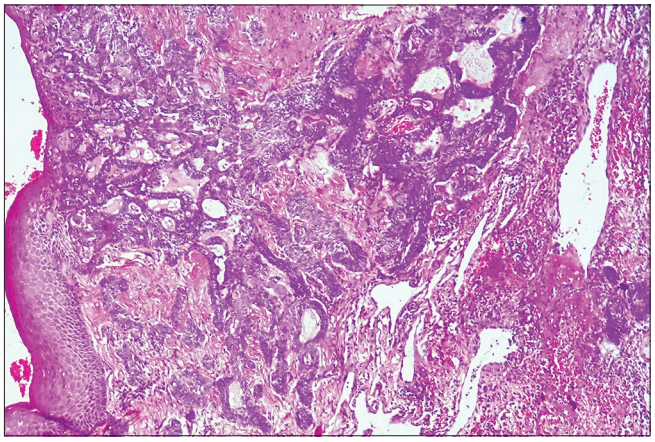

Figure 4: Basaloid islands and cord-like structures infiltrating the overlying epithelium, areas of mucous cells and mucin filled spaces are also seen (H&E stain, ×40)

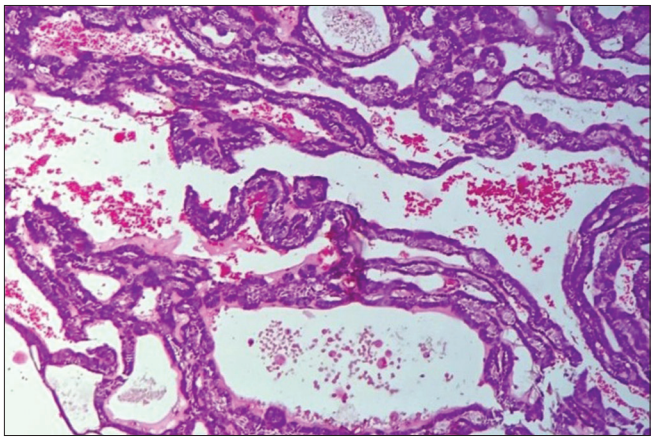

Figure 6: Tumour cells forming canalicular and tubular structures enclosing haemorrhagic spaces (H&E stain, ×100)

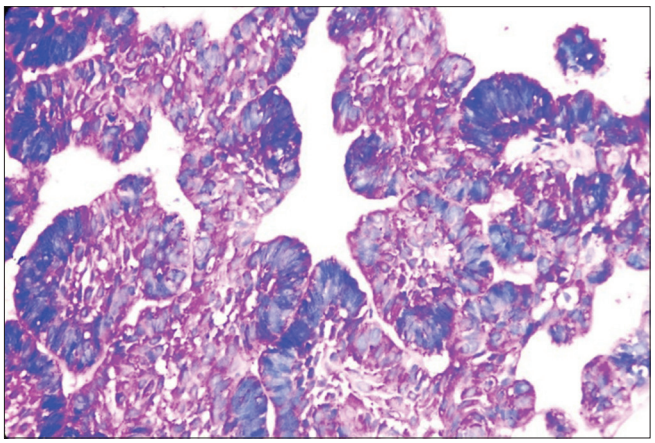

Figure 8: Periodic acid–Schiff (PAS) stain positivity at periphery of islands and cytoplasm of central cells (PAS stain, x400)

cell frequently found peripherally, in the cell nests and cords arranged in a palisaded manner. They are cuboidal or columnar cells and are usually single-layered, but multiple layers may be seen peripherally. The second cell type is larger with modest cytoplasm, indistinct cell borders and a pale-staining oval nucleus located at the center of the tumor nests.[1] The

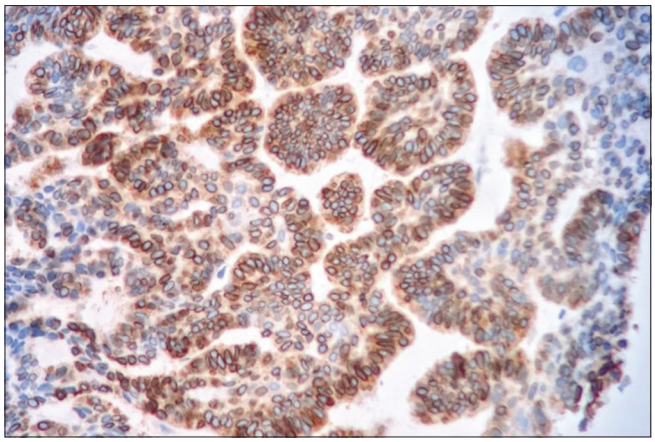

Figure 9: Calponin immunostaining shows strongly positive peripheral cells of the lesion suggestive of myoepithelial cells (IHC stain, ×400)

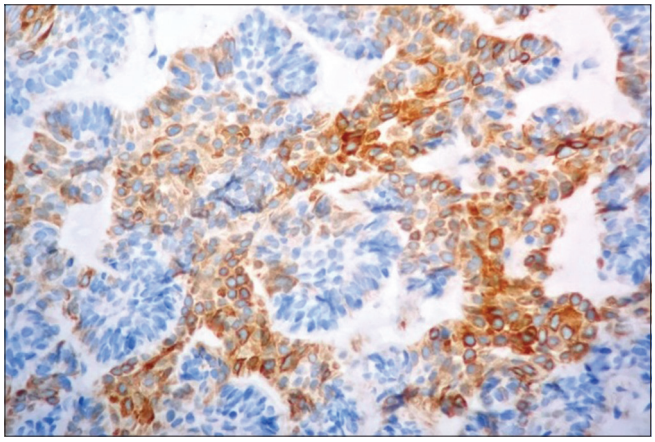

Figure 10: CK 19 showed patchy positivity in the center of islands at places and the tubular structures (IHC stain, ×400)

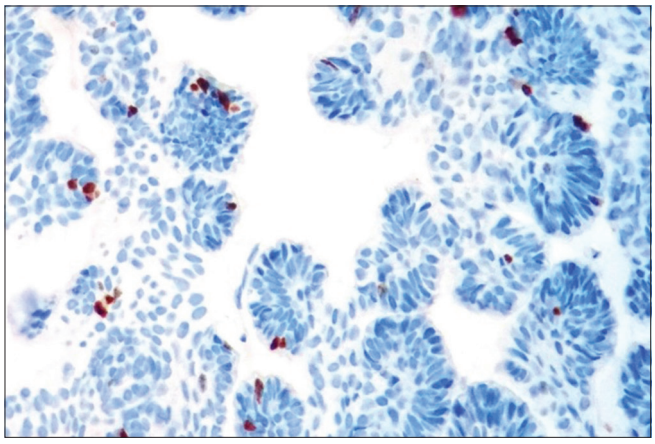

Figure 11: Ki-67 index <5% (IHC stain, ×400)

Electron microscopic and immunohistochemical evaluation of the BCA have shown luminal (ductal) and nonluminal cells (myoepithelial and basal cell) of variable differentiation. The luminal cells may show basosquamous or rarely true squamous cell differentiation, even forming keratin pearls. Nonluminal cells may be devoid of specific differentiations or show ultrastructure features indicative of myoepithelial cell development.[2,9] Cytokeratin was demonstrable in nearly all BCAs with variable expression. Similarly, immunoreactivity to S100 and vimentin was seen in most BCAs but was typically localized to peripheral tumor cells. In a study of 14 cases of BCA, Zarbo established that staining with antibodies to α-SMA and calponin was of equivalent measure in all the BCAs. In an another study by de Arujo *et al*. on 114 cases of minor salivary gland tumors, it was observed that luminal cells from BCA, pleomorphic adenoma, adenoid cystic carcinoma and epithelial myoepithelial carcinoma expressed CK 7, 8, 14 and 19.[2] P63 a nuclear marker for basal/stem cells of stratified epithelium and myoepithelial cells is strongly positive in BCA. This marker, however, is of limited use in the differential diagnosis because most other diagnostic considerations, aside from canalicular adenoma, are positive as well.[1] Basal/myoepithelial cell component is a key and essential diagnostic criterion.[9] In the present case, immunopositivity for calponin and CK 19 confirmed participation of both myoepithelial and luminal cells aiding the final diagnosis of BCA.

Globally, it has been accepted that the BCA may adopt an ameloblastoma-like pattern because of the palisaded nature of the peripheral layer of basaloid cells. The distinction could be especially problematic for lesions that occur in the odontogenic regions, which fortunately are uncommon.[10] In the present case, the site, histopathology and radiographic presentation of the lesion channeled more toward ameloblastoma. However, ameloblastoma can be ruled out by the fact that, reverse polarization of nuclei typical of peripheral ameloblast-like cells is not observed in BCA.[1] Further calretinin negativity ruled out ameloblastoma in the present case.

WHO (2005) has defined BCAC as cytologically and histomorphologically similar to BCA but is an infiltrative epithelial neoplasm with potential for metastasis. Owing to the extensive local destruction, BCAC was also considered in the differential diagnosis, but the low Ki-67 index did not support the suspicion of a malignant neoplasm, i.e., BCAC or basaloid squamous cell carcinoma (BSCC). BCAC can be distinguished from BCA by its infiltrative and metastatic potential, but its extensive infiltrative growth has been rarely encountered and is essentially a low-grade malignancy. Regional recurrences or distant metastases have been described in BCAC since it was first reported, but later authors have noted that certain of these cases have unusual histologic features that are seen in some other similar tumors such as adenoid cystic carcinoma. To identify additional morphological and immunohistochemical characteristics that can assist in differentiating BCACs from BCAs, Jung *et al*. carried out a study on BCAC and BCAs with or without capsular invasion. They concluded that at cytomorphologic level, the lesions could not be distinguished. None of the two had clearly different immunohistochemical profile as well. Further, BCAs with capsular invasion shared several pathologic features with BCACs. Based on these findings, they suggested that it can be difficult to distinguish early stage BCACs from BCAs that exhibit minimal capsular

invasion because of lack of immunohistochemical or molecular evidence supporting the notion that BCAs with capsular invasion may be early BCACs. None of their patients diagnosed as BCAC manifested recurrences or metastases after surgery with or without radiotherapy. Therefore, they posited that the BCA can be viewed as an infiltrative neoplasm and questioned the category of its malignant counterpart "BCAC."[11] The present case supports this hypothesis.

In some of the histopathologic sections, basaloid islands were seen either infiltrating from or into the epithelium giving the impression of BSCC. However, the deeper tissue showing the presence of many canaliculi with double layered cells, enclosing hemorrhagic spaces with very scanty stroma a feature of canalicular adenoma[10] ruled out BSCC. Tubular trabecular variants of BCA at low power can be confused with canalicular adenoma, but closer examination can reveal participation of both luminal and basal cells and more collagenized stroma compared to canalicular adenoma. Lack of myoepithelial cells in canalicular adenoma further helps in the diagnosis. In the past, both the tumors were encompassed in the group of monomorphic adenomas. Later, in 1983, Gardner and Daley showed the distinction between BCA and canalicular adenoma. Both the tumors have different immunohistochemical and histogenetic profile. None of the smooth muscle cell antibodies bind cells of canalicular adenoma allowing specific immunohistochemical differentiation.[4,12]

BCA warrants adequate cure rate with only conservative resection. However, recurrence has been reported in about 25% patients with membranous BCA. Sometimes, it may undergo malignant transformation into BCAC and adenoid cystic carcinoma. Malignant transformation rate of 4.3% has been reported.[13] On the other hand, recurrence and malignant transformation in canalicular adenoma are rare.[10] In ameloblastoma, the rate of recurrence reported in various reviews is diverse and range from 20% to 90%. The recurrence rate after conservative therapy (34.7%) comes out to be about twice that associated with radical therapy (17.3%).[1] Therefore, due to prognostic implications, differential diagnosis of these lesions is mandatory.

CONCLUSION

This rare entity can thus be summarized as a great mimicker and may mislead a clinician causing diagnostic threat leading to severe morbidity and fatality. Thus, recognizing and aiding proper therapy is a must. Moreover, larger studies and literature review for BCA and BCAC is must to distinguish these two varieties or confirm the behavioral status of these lesions. Although BCA is a rare lesion in the minor salivary glands, it should be considered in the differential diagnosis of a palatal lesion.

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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