CASE REPORT

Basal Cell Adenocarcinoma - 2 Case Series of a Rare Entity

Arunachalam Meyyappan, Sathish Kumar, Mary Tresa Jeyapriya, Pradeep Sankar

Abstract

Introduction: Basal cell adenocarcinoma is a rare well recognized low grade malignant salivary gland tumor often resembles basal cell adenoma. the infiltrating growth pattern and likelihood of vascular and perineural involvement distinguishes basal cell adenocarcinoma with basal cell adenoma.

Case presentation: Here we present histopathological and immune histochemical analysis of two such rare cases of basal cell adenocarcinoma and review of literature is discussed.

Conclusion: Basal cell adeno carcinoma is a low grade malignancy and doesn't metastasize. With only few hundreds of cases that has been reported in literature, basal cell adeno carcinoma should be included as differential diagnosis in salivary gland tumors in order not to miss the diagnosis.

Key words: basal cell adenocarcinoma, basal cell adenoma, salivary gland neoplasm

Oral and Maxillofacial Pathology Journal (2023): ***https://www.ompj.org/archives***

Introduction

Basal cell adenocarcinoma (BCAC), is a rare salivary gland tumor that has been recently added to the subtypes of salivary gland carcinoma in the year 1991 by World Health Organization classification. Although there have been occasional references in the literature over the past 30 years to malignant basaloid tumors, malignant transformation of basal cell adenoma and salivary gland carcinomas associated with basal cell adenoma, BCAC has only recently been characterized.[1] It is classified as a malignant epithelial tumor in the latest WHO histological classification of salivary gland tumors (2017).[2]

This tumor was reported under a variety of names, including malignant basal cell tumor, malignant basaloid tumor, hybrid basal cell adenoma/ adenoid cystic carcinoma, basaloid salivary gland carcinoma and atypical monomorphic adenoma, Before the term was universally accepted.[3]

Ellis and Wiscovitch have published the largest series of cases (29), and have defined the clin- icopathological features.[4,5]

BCAC of the salivary gland, occurs mostly in the major salivary glands, particularly in the parotid gland, it is a rare neoplasm comprises 1.6% of all salivary gland neoplasms and 2.9% of malignant salivary gland neoplasms. BCAC is a tumor similar to basal cell adenoma (BCA) except that it grows in an invasive destructive fashion, often with perineural and/or vascular invasion but without histologic evidence of preexisting basal cell adenoma.[1] Based on growth pattern they are divided into the following four subtypes: solid, trabecular, tubular and membranous; of these, the solid subtype is the most common.[4]

Department of Oral Pathology and Microbiology, Karpagavinayaga Institute of Dental Sciences, Chinnakolambakam, Maduranthakam taluk, Chengalpet District, Tamil Nadu, India.

Corresponding author: Arunachalam Meyyappan, Department of Oral Pathology and Microbiology, Karpagavinayaga Institute of Dental Sciences, Chinnakolambakam, Maduranthakam taluk, Chengalpet District, Tamil Nadu, India. Email:drarunsoralpath@gmail.com

How to cite this article: Arunachalam Meyyappan, Sathish Kumar. Mary Tresa Jeyapriya, Pradeep Sankar. Basal Cell Adenocarcinoma -2 Case Series of a Rare Entity. Oral Maxillofac Pathol J 2023;14(1): page no. 127-130

Source of Support: Nil

Conflict of Interest: None

It is important to differentiate BCAC from other basaloid cell tumors of salivary gland tumors because of the differences in prognosis and potential differences in treatment.

In this article we present histopathological and immunohistochemical analysis of two cases of basal cell adenocarcinoma, reported to the Department of Oral Pathology and Microbiology, Karpaga Vinayaga Institute of Dental Sciences, Chengalpet.

Case report 1

A 57 year old female reported with diffuse swelling in left side of the face since five months and pain since two months

On examination, swelling was present on left side of the

face in the pre auricular region measuring approximately 2x2 cms in size; radiograph revealed no abnormality and on the basis of clinical features, lesion was thought to be a salivary gland lesion with possibility of pleomorphic adenoma. Surgical excision was done and biopsy sent for histopathological and immune histochemical analysis. macroscopically specimen was greyish brown in color and firm in consistency. Microscopically, the section showed salivary parenchyma with an adjacent poorly circumscribed malignant neoplasm arranged predominantly in solid sheets, nests with jig saw pattern, and cribriform pattern. Many of tumor cell lobules exhibit predominant peripheral palisading of nuclei. the individual tumor cells are round to polyhedral in shape with scant cytoplasm and pleomorphic, hyper chromatic nuclei with moderate increase in mitotic activity. the intervening fibrous stroma show adundant myxohyaline basement membrane material. Occasional areas exhibit squamous metaplasia of tumor cell lobules. The tumor exhibits capsular infiltration and two lymph nodes were seen in adjacent connective tissue with features of reactive follicular hyperplasia. histopathologic diagnosis of basal cell adenocarcinoma was given. (Figure A and B)

Immunohistochemistry analysis showed focal positivity for tumor protein (p63), smooth muscle actin (SMA), smooth muscle myosin heavy chain (SMMHC) (Figure C, D and E) and strong positivity for cytokeratin (CK) (Figure F)

Case report 2

A 85 year old male patient reported with localized swelling present in right maxillary palatal region since two months. On examination the swelling was present in the right maxillary palatal region in relation to 13, 14 and 15 measuring approximately 1.5*1.5 cms in size, soft in consistency and nontender, occlusal radiograph revealed a mild radiolucency in 13,14,15 region. lesion was thought to be a minor salivary gland tumor. Surgical excision was done and biopsy sent for histopathological and immunohistochemical analysis. Macroscopically the specimen was greyish white in color and firm in consistency.

Microscopically the section showed mucous salivary gland acini with an adjacent infiltrating neoplasm arranged in trabeculae and solid sheets. The tumor cells are basaloid with scant cytoplasm and hyperchromatic nuclei. Some of the epithelial nests exhibit squamoid metaplasia. the intervening fibrous stroma shows myxoid basement membrane. Tumor is seen infiltrating into adjacent tissue. (Figure G and H)

Immune histochemistry analysis showed strong positivity for P63, SMA, SMMHC and CK (Figure I, J, K and L)

Discussion

Basal cell adenocarcinoma composes only 1.4% of all major salivary gland malignant tumors, and it primarily arises from the parotid gland (88%) followed by the submandibular

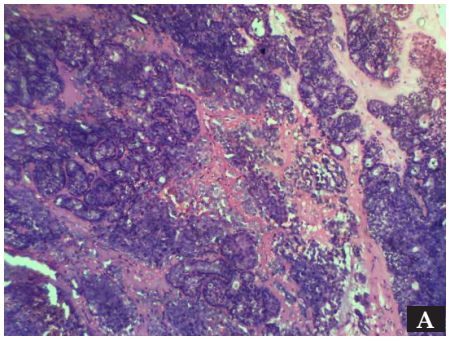
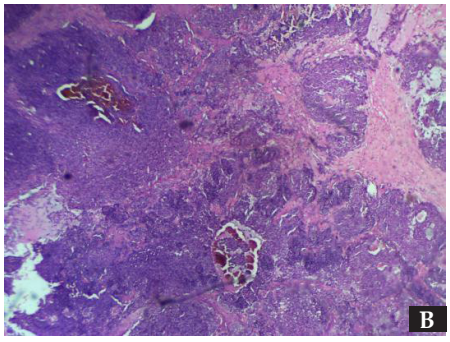
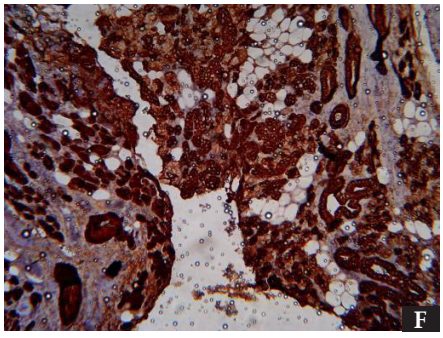


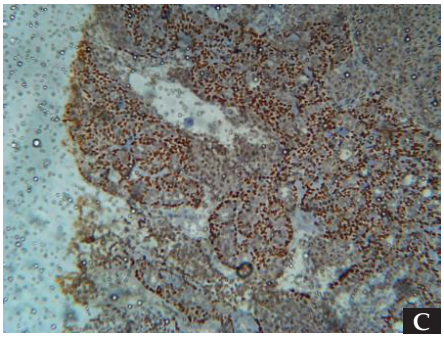
Fig. 1: (A) Malignant cells arranged in nests and solid sheets, and also in cribriform pattern.(H and E, 10x)

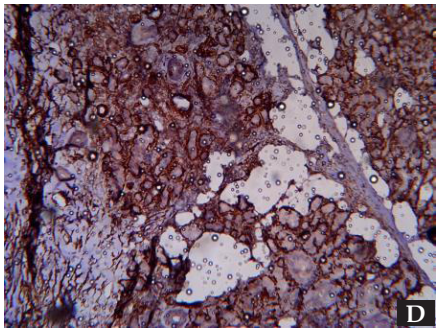


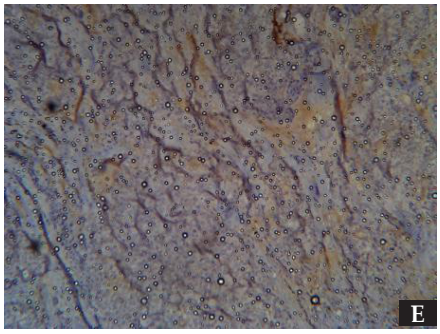
(B) Malignant neoplasm arranged predominantly in solid sheets, nests with jig saw pattern. Many of tumor cell lobules also shows predominant peripheral palisading and hyper chromatic nuclei (H and E, 40x)



CK
(F) Strong positivity for CK (IHC, 10X)







(C), (D) and **(E)** Focal positivity of p63,SMA and SMMHC respectively (IHC, 10x)

(11.2%) and sublingual (0.8%) glands. this tumor is also referred to as basaloid salivary gland carcinoma, carcinoma ex – monomorphic adenoma, and malignant basal cell.[6]

Eighty percent of the patients are over 50 years, with average age of 60 years with no sexual predilection. Swelling is the principal symptom, but pain or tenderness occasionally may be an associated complaint. Occasionally, the swelling has been described as rapid in onset, but this has no prognostic significance.[7]

The average symptom duration prior to diagnosis has been reported to 1.8 years by Ellis and wiskovitc, additional cutaneous adnexal lesions are also observed in about 10% of BCAC cases.[4] According to the retrospective database review of the national cancer database in the US between 1998 and 2012 by zhan et al, the average age at diagnosis is 64 years (range 18 years to 92 years) and 47.3% of the tumors were 2cm to 4cm in diameter, and 35.8% of them were less than 2cm in diameter.[6]

Histologically, BCAC can be divided into four subtypes: solid, trabecular, tubular and membranous. The solid pattern is characterized by contiguous tumor cells arranged in islands and masses within the fibrous connective tissue stroma. These tumor islands can be round to oval or large irregular masses.

The membranous type is distinguished by thick, eosinophilic, periodic acid schiff positive hyaline laminae that surround and separate one tumor nest from another and may create a jigsaw puzzle image in portions of tumor.[7]

Trabecular type is characterized by anastomosing cords and bands of basaloid epithelial cells which may be likened to the configurations shaped like Chinese characters that are formed by bony trabeculae in fibrous dysplasia of bone. Conspicuous small lumina or pseudolumina characterize the tubular type of BCAC.[7]

First case has shown malignant neoplasm arranged predominantly in solid sheets, nests with jig saw pattern, and cribriform pattern similarity with solid type intermingled with membranous type showing capsular infiltration. second case showing infiltrating neoplasm arranged in solid sheets like proliferation intermingled with trabeculae due to which diagnosis of solid pattern intermixed with trabecular pattern of BCAC was justified.

A solid pattern is predominant in about two-thirds of the tumors. The membranous type is the second most frequent and comprises about 20% of these tumors. The trabecular and tubular types are occasionally the dominant patterns.[7]

Basaloid salivary gland neoplasms presents either as adenoma or carcinoma, their relationship with surrounding tissue plays an important role to distinguish between the two. and BCAC shows perineural invasion and the histopathologic appearance of the cells with its invasiveness which distinguish it from basaloid adenoma.[7]

CASE 2
HISTOPATHOLOGY

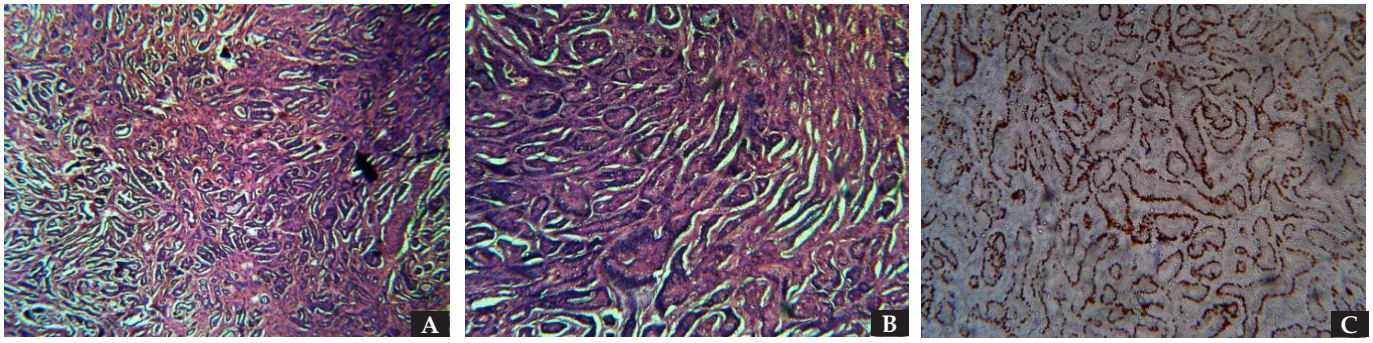
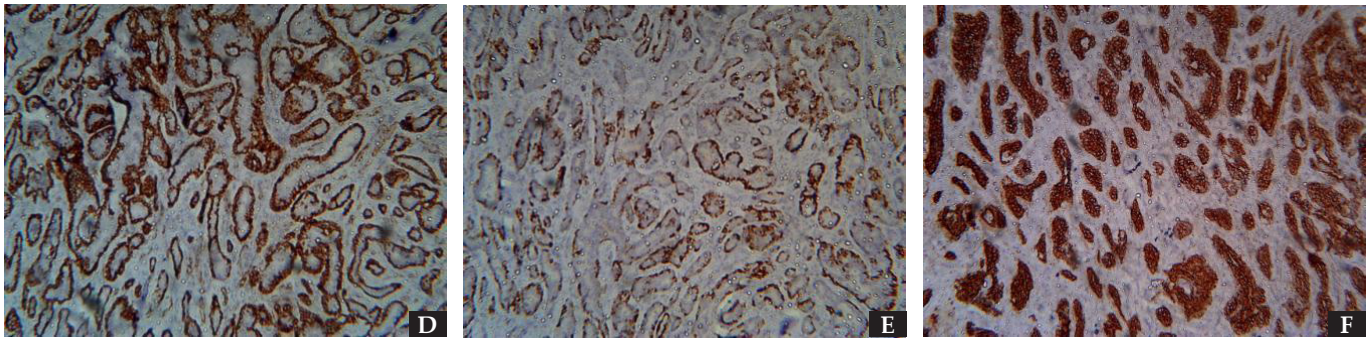


Fig. 2: (A) Mucous salivary gland acini with an adjacent infiltrating neoplasm arranged in trabeculae and solid sheets. (H and E, 4x view)

(B) Tumor cells that are basaloid with scant cytoplasm and hyperchromatic nuclei.(H and E 10X)



CK

(C), (D), (E) and **(F)** Strong positivity for p63,SMA,SMMHC and CK (IHC, 10x view)

Epithelial cells of basal cell adenocarcinoma present in two forms; one is small round cell with scanty cytoplasm and a dark basophilic nucleus while the other one is a large, polygonal to elongated cell with eosinophilic cytoplasm and a large, pale basophilic nucleus. Frequently, the small dark cells are located peripherally to the larger pale cells and produce palisading of the nuclei of cells along epithelial stromal interface. Surrounding this there will be varying degree of collagenous stroma. First case showed cells that are round to polyhedral in shape with scant cytoplasm and pleomorphic, hyper chromatic nuclei and second case showed basaloid cells with scant cytoplasm and hyperchromatic nuclei.

In general, BCAC shows positivity for cytokeratin, anti-smooth muscle actin and S-100 protein. Staining pattern varies with its architectural type. both cases of us showed positivity for cytokeratin, P 63, SMA and SMMHC.

Similar case was reported in 2007 by Ruchi Sharma et al, a case of basal cell adenocarcinoma in sub mandibular salivary gland with cells arranged as sheets and strands of proliferating monotonous basaloid cells having hyperchromatic nuclei showing Two types of basaloid cells dark basophilic cells towards the periphery and pale basophilic cells towards the center of the proliferation with no encapsulation. IHC was not done unlike in our case.[8]

The differential diagnosis of BCAC is basal cell adenoma, adenoid cystic carcinoma (ACC), basaloid squamous cell carcinoma (BSCC) and acinic cell carcinoma. The basal cell adenocarcinoma shares histologic features with the basal cell adenoma (BCA). Both exhibit myoepithelial differentiation, reactivity patterns indicative of ductal epithelium, and closely similar immunohistochemical profiles. Basal cell adenocarcinoma is distinguished from BCA by the histologic features of invasion, mitotic activity, and neural or vascular involvement. ACC must be differentiated from BCAC for prognostic values. BCAC lacks cribriform pattern and pseudocysts of amorphous, basophilic material characteristic of adenoid cystic carcinoma. BSCC is usually located in the hypopharynx, base of tongue, and supraglottic larynx, regions in which basal cell adenocarcinoma is rarely found. Unlike basal cell adenocarcinoma, basaloid squamous carcinoma shows squamous differentiation that involves the mucosal epithelium, which may demonstrate dysplasia, carcinoma in situ, or invasive squamous cell carcinoma, whereas acinic cell carcinoma is histologically defined as a tumor with predominant differentiation toward serous acinar cells, admixed with ductal and myoepithelial elements with predominantly cells arranged in organoid pattern which BCAC lacks. Right now, basal cell adenocarcinoma is considered as a low-grade malignancy with less metastatic potential.[7]

Conclusion

Basal cell adeno carcinoma is a low-grade malignancy and doesn't metastasize, with only few hundreds of cases that has been reported in literature, basal cell adeno carcinoma should be included as differential diagnosis in salivary gland tumors in order not to miss the diagnosis as local recurrence was reported in 28% of cases. Immunohistochemistry has to be done as an adjunctive aid in diagnosis.

References

1. Quddus MR, Henley JD, Affify AM, Dardick I, Gnepp DR. Basal cell adenocarcinoma of the salivary gland: An ultrastructural and immunohistochemical study. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1999;87:485-92.
2. Paul M speight, A William barrett, Salivary gland tumours:diagnostic challenges and an update on the latest WHO classification. Diagnostic histopathology 26:4
3. Raslan WF, Leonetti JP, Sawyer DR. Basal cell adenocarcinoma of the parotid gland: A case report with immunohistochemical, ultrastructural findings and review of the literature. J Oral Maxillofac Surg 1995;53:1457-62.
4. Ellis GL, Wiscovitch JG. Basal cell adenocarcinomas of the major salivary glands. Oral Surg Oral Med Oral Pathol 1990;69:461–9.
5. Seifert G, Sobin LH, eds. Histological Typing of Salivary Gland Tumours. Berlin, Germany: Springer-Verlag; 1991. World Health Organization International Histological Classification of Tumours. 2nd ed.
6. Zhan KY, lentsch EJ. Basal cell adenocarcinoma of the major salivary glands: a population- level study of 509 cases. Laryngoscope 2016; 126:1086-90.
7. Ellis GL, Auclair PL. Basal cell adenocarcinoma. In: Major problems in pathology, Surgical pathology of the salivary glands. WB Saunders Company: 1991. v.25. p. 441-54.
8. Ruchi sharma, Susmita Saxena, Rani Bansal.Basal cell adenocarcinoma: Report of a case affecting the submandibular gland. JOMFP: Vol. 11 Issue 2 Jul-Dec 2007