CASE REPORT

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Basal Cell Adenocarcinoma in the Tongue: An Unusual Presentation

Geetanjali Gupta, Reecha Singh, Kathirvelu Shanmugasamy, Dhananjay S. Kotasthane and Vaishali D. Kotasthane

Department of Pathology, Mahatma Gandhi Medical College and Research Institute, Puducherry-607402, India.
Corresponding author email: gng_82@yahoo.co.in

Abstract: We present a case of basal cell adenocarcinoma (BCAC) in the tongue in a 65-year old male. This is an extremely rare presentation. BCAC generally occurs in the parotid gland and rarely involves the minor salivary glands. Few cases have been reported in literature with a variable presentation. The biopsy was formalin-fixed and paraffin-embedded. The sections were stained with routine Hematoxylin and Eosin. Immunohistochemistry was performed. Hematoxylin and eosin staining showed tumour composed of variable sized and shaped, nests and sheets of basaloid epithelial cells having hyperchromatic to vesicular nuclei. Immunohistochemistry was positive for Pancytokeratin, Epithelial membrane antigen and p53. The clinicopathological features and the cellular immunophenotype addressed the diagnosis towards BCAC of the tongue. The goal of this report is to increase awareness of this rare disease and to review and discuss the differential diagnosis and important considerations in treatment.

Keywords: basal cell adenocarcinoma, minor salivary gland, tongue

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Introduction

Basal cell adenocarcinomas (BCAC) are slow-growing tumours that most commonly involve the parotid gland and very rarely involve the minor salivary glands of the oral cavity.¹ The average age of patients is 60 years, with no sex predilection.² We present a case of basal cell adenocarcinoma in the tongue in a 65 year old male. Such a presentation is extremely rare. BCAC is believed to arise from pluripotent ductal reserve cells. Histopathology of BCAC is characterized by two cell types: small basaloid epithelial cells at the periphery of tumour clusters, and larger epithelial cells situated centrally in tumour clusters. Different histological growth patterns are seen, ie, solid, trabecular, tubular and membranous types. Criteria for the diagnosis of BCAC include infiltrative growth with possible perineural or vascular invasion. Differential diagnosis with high grade malignancies like adenoid cystic carcinoma is important because of the low malignant behaviour and good prognosis of BCAC. Immunohistochemistry reveals reactivity for cytokeratins, p53 and focally reactive for epithelial membrane antigen and S100 protein. It metastasizes seldom, but may recur locally. Although BCAC is a malignant counterpart of Basal Cell Adenoma, it often grows *denovo*.

Case Presentation

A 65-year old male reported with swelling in the throat for two months. Swelling was also accompanied with pain in the involved region. On examination, an ulceroproliferative growth was noticed in the posterior one-third of the tongue extending into the vallecula and crossing the midline. No lymph nodes in the neck were identified. Radiography revealed no abnormality. Imaging studies revealed no evidence of tumour elsewhere in the body. Wide surgical excision with complete removal of the tumour was performed.

The surgical specimens were formalin-fixed and paraffin embedded. The sections were stained with routine Hematoxylin and Eosin stain. Special stains were performed including PAS-diastase and Mucicarmine. Immunohistochemistry was performed using avidin biotin complex technique and diaminobenzidine as chromogen. The antibodies used included Pan-cytokeratin, Epithelial Membrane Antigen, p53, Smooth Muscle Actin, and S-100, at suggested

dilution. We also performed appropriate routinely positive and negative controls.

Results

Macroscopically, the biopsy specimen consisted of a single irregular soft tissue bit measuring $1 \times 0.8 \times 0.4$ cm, gray-tan in colour and firm in consistency. Microscopically, the section showed ulceration through the mucosa. Variable sized and shaped, nests and sheets of basaloid epithelial cells having hyperchromatic to vesicular nuclei (Fig. 1) were seen. Two types of basaloid cells were observed: dark basophilic cells towards the periphery and pale basophilic cells towards the centre of the proliferation. Nuclear palisading was observed along the interface with the collagenous stroma. Intervening thick bands of collagenous septa were observed. Tumour showed an infiltrative growth pattern and focal areas of squamous cell differentiation. Mitotic activity was seen. No salivary gland tissue was identified in the sections studied. PAS-diastase and mucicarmine were negative.

Immunohistochemical evaluation revealed Pan-cytokeratin (Fig. 2), Epithelial Membrane Antigen, p53 (Fig. 3) and S100 positivity. In contrast, Smooth Muscle Actin (SMA) was negative.

The clinical presentation and the macroscopic aspect, together with the histological pattern, the cytological characteristics and the cellular immunophenotype addressed the diagnosis towards Basal Cell Adenocarcinoma (solid pattern mixed with trabecular pattern), in the tongue.

Discussion

Basal cell adenocarcinoma (BCAC) was first recognised in 1978.³ Ellis and Gnepp first defined the histological features of BCAC in 1988. The clinic-pathologic features of this tumour were defined in 1990 by Ellis and Wiscivitch in their study of 29 cases.⁴ In 1991, BCAC was included in the World Health Organization classification of salivary gland neoplasms.⁵ The 2005 World Health Organization classification categorizes BCAC as a low-grade tumour with a favourable prognosis.¹

BCAC's comprise 1.6% of all salivary gland neoplasm and 2.9% of malignant salivary gland neoplasms.⁵ This tumour most commonly involves the parotid gland (90%) and very rarely the minor salivary glands of the oral cavity.¹ Jones et al (2008)⁶

Figure 1. Histologic appearance of solid/trabecular nests of cells with scant cytoplasm, vesicular nuclei and peripheral palisading. Overlying squamous epithelium is seen. Arrow indicates vascular invasion. Hematoxylin and eosin stain, magnification $\times 100$.

states that minor salivary gland tumours typically occur on hard palate. Peel et al (2007)⁷ reported a case of minor salivary gland BCAC in buccal mucosa. Parashar et al (2007)⁸ has also described this tumour in the lip and tongue.

The average age of patients for BCAC is 60 years. No predilection for the occurrence of BCAC in either men or women is apparent. In our case, BCAC has been reported in a 65 year old male. Swelling is the predominant symptom in BCAC, which may occasionally show a rapid onset. Pain and tenderness may be present as an associated complaint.¹ In our case, the patient reported a rapidly growing swelling associated with pain. Patients with BCAC may also develop multiple skin adnexal tumours and parotid basal cell adenocarcinomas.¹

Figure 2. Immunohistochemical staining showing diffuse positivity for cytoplasmic antigen Pancytokeratin ($\times 200$).

Figure 3. Increased proliferation detected by p53 immunostain. Magnification ($\times 100$).

BCAC most frequently occurs in the superficial lobe of the parotid gland. The cut-surface is homogenous, grey, tan-white or brownish with cystic changes. The tumour cells are basaloid epithelial cells which vary from small, dark cells to larger, paler stained cells. BCAC has four major histological patterns: solid, membranous, trabecular and tubular.¹ The solid pattern, as seen in our case, is the most common and is characterized by variable sized and shaped, islands and nests of tumour cells separated by thin septa or thick bands of collagenous stroma. The membranous type is distinguished by excessive amounts of eosinophilic, Periodic Acid Schiff positive hyalinised basal lamina material that forms intercellular droplets and peripheral membranes. Trabecular pattern is identified by the presence of interconnecting bands of basaloid cells. In the tubular type, there are luminal spaces among the basaloid cells. There are foci of squamous differentiation in some tumors.¹

Most cases of BCAC are believed to develop de novo but as many as 25% of cases may arise from a pre-existing basal cell adenoma.^{9,10} The major pathologic differential diagnostic considerations for BCAC are basal cell adenoma, adenoid cystic carcinoma and Basaloid squamous cell carcinoma (BSCC).¹¹ The distinguishing features of basal cell neoplasms have been illustrated in Table 1. Infiltration of tumour cells into parotid parenchyma, dermis, skeletal muscle, or periglandular fat distinguishes BCAC from basal cell adenoma. Vessel or peripheral nerve invasion is evident in about a fourth of the tumors.¹ Distinguishing BCAC

Table 1. Distinguishing features of basal cell neoplasms.¹

Tumour type	Pattern	Cellular features	Perineural invasion
BCA	Syncytial/non-invasive	Uniform basaloid	No
BCAC	Syncytial/invasive	Mild pleomorphism/invasive	Yes
BSCC	Syncytial	Marked pleomorphism, focal keratinization	Rare
ACC solid	Syncytial	Mild pleomorphism	Yes
ACC tubular/cribriform	Ductal/cylindromatous	Uniform biphasic, mild pleomorphism	Yes

Abbreviations: BCA, basal cell adenoma; BCAC, basal cell adenocarcinoma; BSCC, basaloid squamous cell carcinoma; ACC, adenoid cystic carcinoma.

from adenoid cystic carcinoma (ACC) is important due to poor prognosis and higher prevalence of the latter disease. Major distinguishing features of ACC are the presence of dark hyperchromatic angulated nuclei which are in contrast to the vesicular nuclei in BCAC. The cribriform pattern which is commonly seen in ACC, is distinctive and not found in BCAC.¹² Due to the above differences, ACC was ruled out during diagnosis. Another basaloid tumour showing similarity with BCAC is BSCC, a distinct variant of squamous cell carcinoma. The basaloid component of BSCC arranged in solid or trabecular pattern may resemble that of BCAC. The presence of squamous differentiation and invasive squamous cell carcinoma, form an integral component in BSCC but are not features of BCAC.¹² The diagnosis of BSCC was also negated in the present case due to the absence of invasive squamous cell carcinoma component.

Immunohistochemical staining is variable. Tumour cells are reactive for cytokeratins and focally reactive for S100 protein, epithelial membrane antigen (EMA), and carcinoembryonic antigen (CEA).¹ The present case showed positivity for Pan-cytokeratin, EMA, S100 and p53. SMA was negative.

BCAC's have a favourable prognosis as they seldom metastasize and death of patients is rare.¹ They are locally destructive with a propensity to recur. Surgical excision with a wide margin to ensure complete removal has been suggested as the primary treatment for BCAC.⁸ Enucleation or curettage is to be avoided. Radiotherapy has been proposed for lesions in the minor salivary glands because of the higher likelihood of vascular and neural invasion.¹³ Radiotherapy has also been used for tumours with a diffuse infiltrating pattern to adjacent tissue. Scholtz A et al¹⁴ suggest that the recurrence rate is about 25% to 30% and approximately 10% metastasize to regional lymph

nodes or distant organs. It is necessary to differentiate BCAC from other basaloid cell tumours because of the differences in prognosis and potential differences in treatment.

Very little is known about the genetic makeup of BCAC. The role of specific chromosomal aberrations remains unclear, and additional studies are needed to determine important chromosomal deviations in the pathogenesis of these tumours.¹⁴

Conclusions

BCAC of the tongue is an extremely rare presentation. Few cases of BCAC have been reported in literature involving the minor salivary glands and the tongue. Basal cell adenocarcinoma should be one of the diagnostic considerations in a patient with a rapidly growing swelling in the tongue associated with pain and tenderness.

Disclosures

This manuscript has been read and approved by all authors. This paper is unique and not under consideration by any other publication and has not been published elsewhere. The authors and peer reviewers report no conflicts of interest. The authors confirm that they have permission to reproduce any copyrighted material. Written consent was obtained from the patient or relative for publication of this study.

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