### **CASE REPORT**

# Basal cell adenocarcinoma of the upper lip: a case report

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#### **Abstract**

Basal cell adenocarcinoma (BCAC) is an extremely rare low grade, malignant salivary gland neoplasm, affecting minor salivary glands, comprising about 3% of all malignant salivary neoplasms. Very few cases of BCAC affecting the upper lip have been reported in the literature till date. We describe an usual case of BCAC affecting the upper lip in a 58-year-old woman. The present case reiterates the limitations of microscopic examination of an Incisional biopsy to distinguish BCAC from basal cell adenoma (BCA).

#### **Key words**

Basal cell adenocarcinoma, Lip

#### 1 Introduction

Minor salivary gland neoplasms account for a significant proportion (9%-23%) of all salivary gland tumors <sup>[1]</sup>. Basal cell adenocarcinoma (BCAC) is an extremely uncommon salivary gland malignancy that very rarely affects the minor glands <sup>[1]</sup>. BCAC may arise either de novo or from a pre-existing basal cell adenoma (BCA) with which it shares demographic and histologic features <sup>[2, 3]</sup>. It is a low-grade malignant tumor which has microscopic features similar to BCA, together with infiltrative growth pattern and its ability to metastasize <sup>[4]</sup>.

We report an interestingly rare case of BCAC affecting the upper lip in a 58-year-old female.

# 2 Report of case

A 58-year-old female represented with a painless swelling of the upper lip for two years duration with a gradual increasing in size over the last six months. Patient's medical, personal and family history was non-contributory, and she denied any history of trauma or having a habit of upper lip biting.

Extra-oral examination revealed a solitary, diffuse swelling of the upper lip without ulceration. Intra-orally, there was a well-defined mass, about  $2 \text{ cm} \times 2 \text{ cm}$  in size with a raised surface and an area of depression in the centre (see Figure 1).

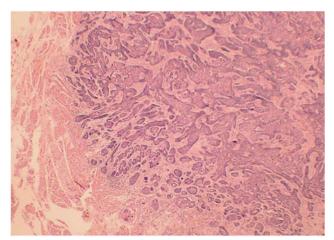
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The mass was firm, not tender and non-fluctuant on palpation. The overlying mucosa was intact and normal in appearance. No extra-oral sinus or cervical lymphadenopathy was detected. Cranial nerve examination revealed no sensory or motor deficit. Based on the size of the lesion and apparent induration, a presumptive diagnosis of salivary gland neoplasm was made.

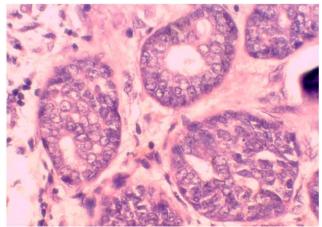


Figure 1. Intra-oral mass at time of initial biopsy

Under local anaesthesia, excisional biopsy was performed, and the histopathologic examination of hematoxylin and eosin stained tissue sections showed a solid, basaloid tumor islands infiltrating the adjacent fibrous tissue with prominent intramuscular invasion. The tumor islands were composed of two distinct cell types, namely peripherally placed compact cells with hyperchromatic nuclei arranged in a palisading manner, surrounding centrally located polygonal cells with eosinophilic cytoplasm. No abnormal mitotic activity was detected (see Figure 2-3). Because of the infiltrative pattern of growth, the final diagnosis of solid variant of low-grade basal cell adenocarcinoma of the upper lip was made. Thence, the residual lesion was completely re-excised which was subsequently examined histologically to confirm the diagnosis of BCAC. Neither radiotherapy nor neck dissection was performed. After a 12-month period, follow-up examination revealed complete healing with no clinical evidence of recurrence or metastasis. Written consent for publication was obtained from the patient.



**Figure 2.** Low-power view of the tumor demonstrating an obvious infiltrative nature at the periphery (Hematoxylin and eosin stain,  $\times 250$ ).



**Figure 3**. High-power view of the tumor islands showing a distinctive peripheral palisading basaloid cells (Hematoxylin and eosin stains, ×400).

# 3 Discussion

Basal cell adenocarcinoma is a relatively recently described uncommon entity that occurs almost exclusively in the major salivary glands. Most cases arise de novo, but some examples develop from malignant transformation of a pre-existing basal cell adenoma. This cancer accounts for about 2.9% of all salivary gland malignancies and 1.6% of all salivary gland tumors. About 90% of the tumors are situated in the parotid gland, where they account for 0.6% of parotid tumors [5, 6]. Basal cell adenocarcinoma of a minor salivary gland is extremely rare. The tumor affecting the minor salivary glands occur most frequently in the palate, tongue, buccal mucosa and upper respiratory tract. This tumor predominates at the seventh decade without sex predilection [7], and is believed to arise from pluripotent ductal reserve cells [8]. Basal cell adenocarcinoma is a low-grade malignant neoplasm with a cytologic resemblance to BCA, but demonstrating invasive growth pattern and sometimes demonstrates perineural or vascular invasion [4]. This tumor has four major histologic growth patterns, solid, tubulotrabecular, cribriform and membranous. Histologically, the small basaloid cells possess round, uniform basophilic nuclei and scanty cytoplasm. Nuclear pleomorphism and mitosis are not seen [8]. It is important to differentiate BCAC from other basaloid cell tumors of the minor salivary glands because of the prognostic and potential differences in treatment, particularly basal cell adenoma, solid variant of adenoid cystic carcinoma, basaloid squamous cell carcinoma, sialoblastoma, and undifferentiated carcinoma [3,7].

Immunohistochemically, BCAC expresses immunoreactivity for S -100 proteins, cytokeratins (CK), epithelial membrane antigen (EMA), and smooth muscle actin (SMA) [1, 9].

Basal cell adenocarcinoma is optimally treatable by wide surgical excision with negative margins. Radiotherapy has been proposed for lesions with close or positive resection margins <sup>[10]</sup>. The outcome is favourable after adequate surgical treatment although tumors arising in minor salivary glands having a higher local recurrence and mortality rates are low <sup>[8]</sup>.

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