CASE REPORT

Basal cell adenocarcinoma of the minor salivary gland: Case report and cell adhesion molecules immunocytochemical profile

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Summary Basal cell adenocarcinoma (BCAC) is a rare salivary gland tumour, especially in minor glands, and is considered to be the malignant counterpart of basal cell adenoma. The clinical, histological and immunohistochemical features of a case involving the upper labial mucosa are described. In conclusion any tumor that shows the characteristics of basal adenoma, on initial examination but has limited encapsulation, even a small number of mitotic figures, or even a suggestion of infiltration, and altered expression of cell adhesion molecules, should be carefully examined for the possibility of BCAC.

KEYWORDS Basal cell adenocarcinoma; Salivary gland neoplasms; Cell adhesion molecules

Introduction Basal cell adenocarcinoma (BCAC) is a rare salivary gland tumour, which is considered to be the malignant counterpart of basal cell adenoma. BCAC is accounting for only the 2.9% of malignant salivary neoplasms.¹ It affects predominantly the major glands and there are very few reports of lesions in minor glands.

Diagnosis of BCAC depends on finding features similar to adenoma but with an infiltrative growth pattern and exlusion of adenoid cystic carcinoma, sialoblastoma and basaloid carcinoma.² BCAC is considered a low-grade malignancy but the clinicopathologic behaviour of this tumour, is still unclear.

A case of BCAC of a minor salivary gland in the upper labial mucosa is reported and its cell adhesion molecules immunocytochemical profile is described.
Case report

A 61 year-old male presented with a complaint of swelling involving the upper labial mucosa that had enlarged slowly for 3 years. Intraoral examination revealed a 1.5 cm diameter, painless, lobulated mass with well defined borders and normal overlying mucosa (Fig. 1). No neck masses or palpable lymph nodes were noticed. The patient was otherwise in good health. No clinical signs of malignancy were present. The patient complained of spontaneous pain caused by irritation from the adjacent tooth. The clinical impression was that this was a salivary gland neoplasm.

Following local excision, histological examination with H&E staining revealed an unencapsulated salivary gland neoplasm consisting of basaloid cells with basophilic cytoplasm and hyperchromatic nuclei, and cuboidal ductal cells. The neoplasm showed solid and tubulo-trabecular growth patterns, which were intermixed with areas of necrosis and haemorrhage. The histologic diagnosis was BCAC (Fig. 2a).

Neoplastic cell population was composed of two cellular forms: basaloid cells positive stained for S100, vimentin and partially for a cocktail of cytokeratins (CK5, 6, 8, 17) and cuboidal, ductal cells, strongly expressed cytokeratins and only partially S100 protein. Neoplastic cells were almost totally negative for SMA.

Furthermore by immunohistochemical methods was detected the expression of cell adhesion molecules E-cadherin, desmoglein-2, CD-44s, ICAM-1 and β4-integrin. Almost all neoplastic cells were membrane positive for β4-integrin (Fig. 2b), especially peripherally, E-cadherin was expressed mainly on the membrane of cuboidal, ductal cell (Fig. 2c), desmoglein-2 was detected in almost all neoplastic cells, intracellularly and furthermore on the membrane of luminal cells (Fig. 3a), CD44s expressed in the majority of both basaloid and ductal cell especially in the centre of solid areas (Fig. 3b) and finally ICAM-1 was positive in a small number of neoplastic cells (Fig. 3c).

The lesion was considered completely excised histologically, the postoperative period was uneventfull, and there was no evidence of recurrence after 3 years of follow up.

Discussion

BCAC is uncommon in the minor salivary glands. Ninety percent of cases reported are in the major glands, usually the parotid,1 four cases reported in the submandibular gland and one in the sublingual gland.3 Very few cases4–10 have been reported in minor salivary glands of the palate, buccal mucosa, and labial mucosa as in our case. The average age of reported cases was in the sixth decade11 and lesions presented as asymptomatic swellings of long duration with pain and sudden increase in size reported in only a few cases in the major

Figure 1  Clinical appearance of a broad based mass on the upper labial mucosa which was diagnosed as the BCAC.

Figure 2  (a) Histological appearance of BCAC (H&E, original magnification ×40), (b) β4 integrin positive cellular membrane expression for most of the neoplastic cells (original magnification ×40) and (c) E-cadherin positive expression mainly on the membrane of cuboidal, ductal cell (original magnification ×40).
glands. Presentation in minor glands appears identical.

The diagnosis of BCAC may be difficult because clinical examination, computed tomography, ultrasonography and magnetic resonance imaging occasionally do not exclude other benign and malignant salivary gland tumours, and microscopic examination of biopsy specimens sometimes is unable to distinguish it from basal cell adenoma. If a tumour has a marked similarity to basal cell adenoma, further examination is necessary to determine the final diagnosis. Semi-serial sections of the excised tumour should be examined to determine whether the tumour is infiltrating into the surrounding normal tissues. Although no specific histologic parameters are known to be predictors of biologic behaviour, the mean mitotic index is slightly higher in recurrent and metastatic tumors. Furthermore it has been reported that the examination of cell proliferation, apoptosis, and expressions of p53, bcl-2 and epidermal growth factor receptor may be useful in distinguishing malignant basal cell tumours arising in the salivary glands from their benign counterparts.

Reciprocal mesenchymal-epithelial interactions are important for the development of salivary glands and for the maintenance of the structure of the adults glands. Our findings indicate that cell adhesion expression may be a characteristic constitutive phenotypic feature of salivary epithelial and myoepithelial cells, which is preserved in neoplastic proliferation. Whereas a possible downregulation of CD 44 expression, concomitant with loss of other cell-cell and cell — matrix adhesion molecules, such as integrins and E — cadherin, may be involved in tumor dedifferentiation and may promote stromal invasion, eventually contributing to the development of a more aggressive phenotype.

BCAC is a low-grade malignancy and local recurrence has been previously reported. Metastasis is rare with few lesions spreading to lymph nodes and one to the lung. The solid type appears to have the highest risk of metastasis. There was no evidence of metastasis in our case. As primary treatment especially for the major glands, it has been proposed surgical excision with wide margin to ensure complete removal of the tumour. Regional lymph node dissection is recommended only if there is evidence of metastatic disease. In minor glands it has been proposed wide excision with follow-up radiotherapy on the basis of a higher likelihood of neural and vascular invasion. Further studies concerning the clinical behaviour of BCAC of minor salivary glands after primary treatment including radiation therapy is necessary. If BCAC is radiosensitive, conservative surgery for BCAC in minor salivary glands may be a viable option.

In conclusion any tumor that shows the characteristics of basal adenoma, on initial examination but has limited encapsulation, even a small number of mitotic figures, or even a suggestion of infiltration, and altered expression of cell adhesion molecules, should be carefully examined for the possibility of BCAC.

References


