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Case Report

A Case of Basal Cell Adenocarcinoma of the Parotid Gland

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Departments of Otorhinolaryngology and [1]Pathology, Tohoku University School of Medicine, Sendai 980-8574 and [2]Department of Pathology, Iwate University School of Medicine, Morioka 020-0023

IKEDA, K., WATANABE, M., OSHIMA, T., NAKABAYASHI, S., KUDO, T., SAWAI, T. and TAKASAKA, T. *A Case of Basal Cell Adenocarcinoma of the Parotid Gland.* Tohoku J. Exp. Med., 1998, **186** (1), 51-59 —— We present a 73-year-old female with an enlarged mass in the right parotid gland. Fine-needle aspiration cytology suggested pleomorphic adenoma. Diagnostic imaging revealed that the tumour had a well-defined margin arising from the deep lobe of the parotid gland. A total parotidectomy with preservation of the facial nerve was performed. The final histopathological diagnosis including immunohistochemical studies was basal cell adenocarcinoma, which is a recently defined entity and a rare epithelial neoplasm. No sign of local recurrence or metastasis 24 months postoperatively has been observed. ———— basal cell adenocarcinoma; parotid gland; immunohistochemistry

Basal cell adenocarcinoma (BCAC) is a recently defined entity in the World Health Organization classification of the salivary gland tumors (Seifert and Sobin 1991). BCAC is a rare epithelial neoplasm that is cytologically and histomorphologically similar to basal cell adenoma, but is infiltrative and has a slight potential for metastasis. There are a few reports of this tumor occurring at the major and minor salivary glands (Ellis and Gnepp 1988; Luna et al. 1989; Ellis and Wiscovitch 1990; Batsakis and Luna 1991; Ellis and Auclair 1991; Williams et al. 1993; Fonseca and Soares 1996). We report a case of BCAC of the salivary gland.

CASE REPORT

A 73-year-old female noted a painless, progressively enlarging mass of the

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right parotid region for two years before a medical evaluation was made. The physical examination revealed a nonfixed, slightly tender 50-mm mass. Facial nerve function was normal and there were no palpable cervical lymph nodes. Computed tomographic scan showed a lesion arising from the deep lobe of the parotid gland, resulting in a dumbbell shape. The mass was clearly separated from the normal parotid gland tissue covering the mass. The contrast study on computed tomography indicated a few of areas of low density without enhancement (Fig. 1). Magnetic resonance imaging revealed that the tumor had a well-defined margin in the parotid gland with moderate-to-low signal intensity on T1-weighted image (Fig. 2a). The T2-weighted image demonstrated high signal areas in the deep lobe comparable with cystic formation, which were separated from superficial areas of moderate-to-low intensity (Fig. 2b). Fine needle aspiration cytomorphologic features showed moderate cellular aspirates composed of small monomorphic cohesive sheets of epithelial cells with enlarged hypochromatic nuclei (Fig. 3), interpreted as the features of a pleomorphic tumor.

A total parotidectomy with preservation of the facial nerve was performed. At surgery the tumor was confirmed to arise from the deep lobe of the parotid gland. Monomorphic adenoma was suspected by frozen sections. The final histological diagnosis derived from paraffin-embedded tissues was BCAC. The tumor consisted of basaloid epithelial cells with scanty cytoplasm, variable atypia and a few counts of mitotic figures. The most conspicuous cells were relatively uniform with pale eosinophilic to amphophilic cytoplasm, indistinct cell borders, and round to oval, pale basophilic nuclei. An infiltrative growth into the surrounding tissue with perineural and perivascular invasion (Fig. 4) was observed. Basaloid epithelial cells were aggregated in variably sized and shaped nests that were separated from one another by thin septa or thick bands of collagenous

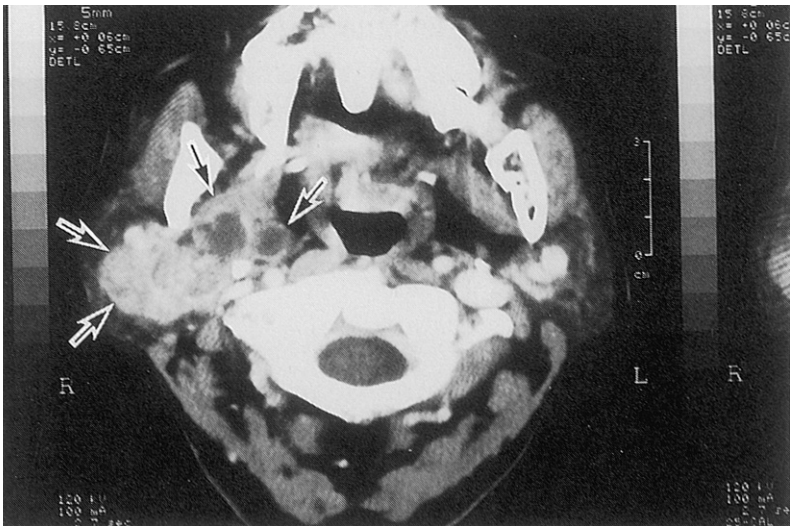

Fig. 1. An axial computed tomographic view showing a tumour mass arising from the deep lobe, a dumbbell shape (arrows), which is clearly separated from the normal parotid gland tissue covering the mass.

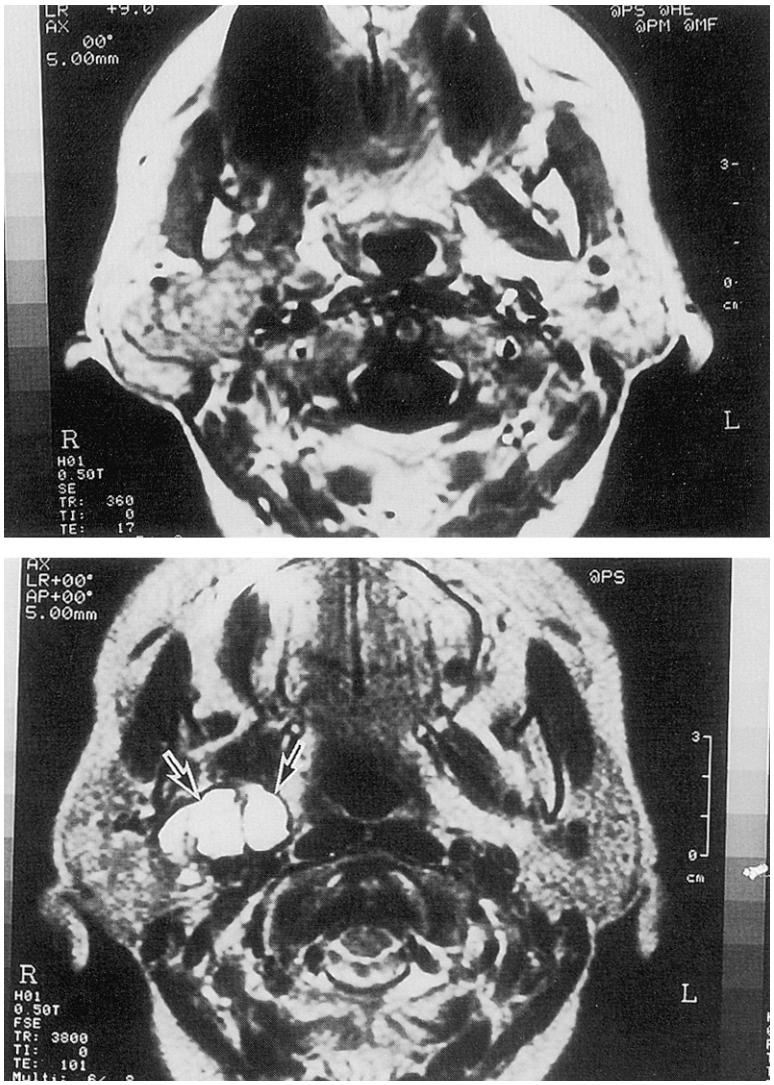

Fig. 2. Axial T1-weighted (a) and T2-weighted (b) magnetic resonance images. On the T2-weighted image, high signal intensity is present in the deep lobe of the parotid gland (arrows), indicating cystic formation.

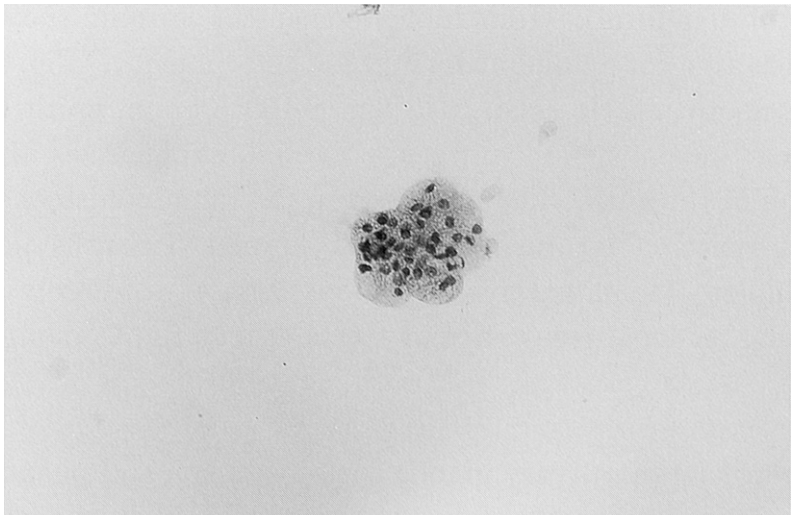

Fig. 3. A cytological feature obtained by fine needle aspiration biopsy form BCAC. May Grunwald Giesma staining ($\times 236$).

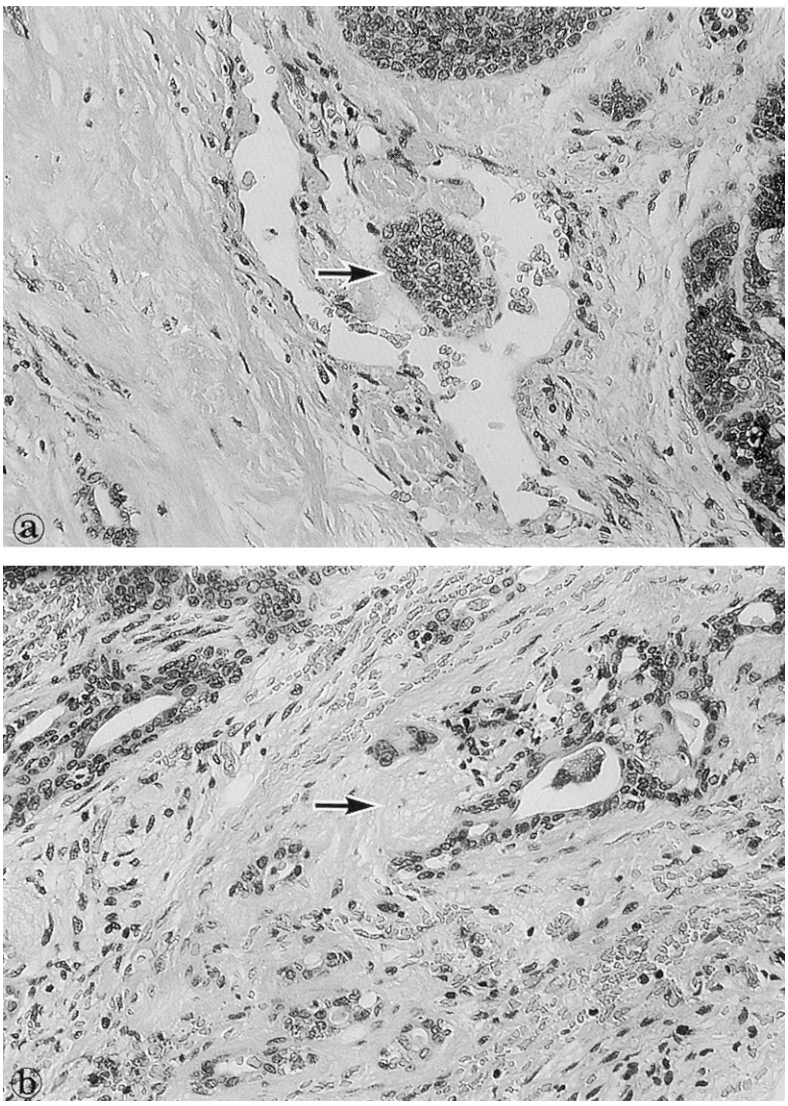

Fig. 4. Perineural (a) and perivascular (b) invasion (arrows) of BCAC. Hematoxylin and eosin (×236).

stroma (Fig. 5a, solid form). In some areas, lumens or pseudolumens were prominent (Fig. 5b, tubular form). Immunohistochemical staining showed that all of the cells were reactive for cytokeratin (Fig. 6a) and vimentin (Fig. 6b), with focal reactivity for smooth muscle actin (Fig. 6c), S-100 protein, epithelial membrane antigen, and carcinoembryonic antigen. Table 1 summarizes the source and optimal dilution of the antibodies employed in the immunohistochemical studies.

The facial function returned to almost normal 3 months postoperatively, although incomplete facial paralysis was observed immediately after surgery. There was no sign of local recurrence or metastasis at the 24 month follow-up.

Discussion

Neoplasms arising in salivary glands with histologic and phenotypical resemblances to BCAC include basaloid salivary carcinoma, carcinoma ex monomorphic adenoma, malignant basal cell adenoma, and basal cell carcinoma. As early as 1970, Evans and Cruickshank (1970) mentioned the existence of malignant basal

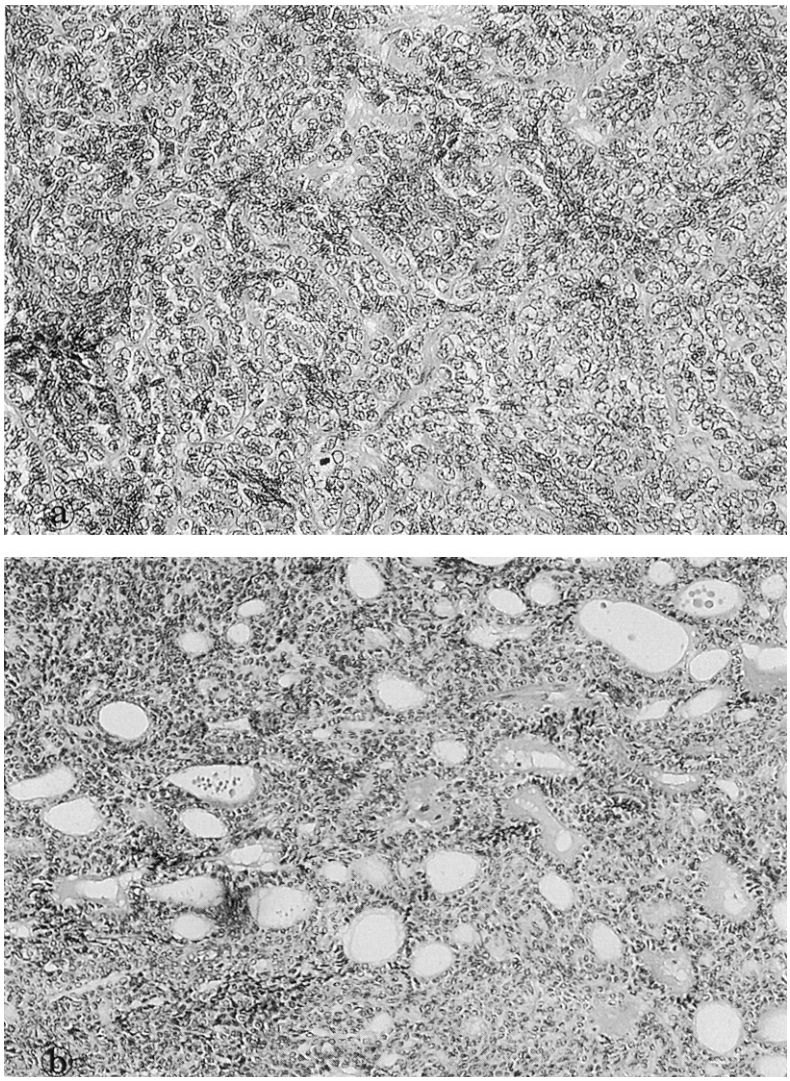

Fig. 5. Solid pattern (a, ×236) and tubular pattern (b, ×118) of BCAC. Hematoxylin and eosin.

cell tumors. A few investigators have described adenoid cystic carcinoma arising from or developing in association with basal cell adenomas and speculated that basal cell adenoma is the benign counterpart of adenoid cystic carcinoma (Evans and Cruickshank 1970; Bernacki et al. 1974). Chomette et al. (1991) reported basaloid carcinoma of salivary glands, a variety of undifferentiated adenocarcinoma, however, they clearly indicated that the tumors were solid variants of adenoid cystic carcinoma. Likewise, Seifert et al. (1986) considered malignant basal cell adenoma to be synonymous with the basaloid type of adenoid cystic carcinoma. Recently, the histomorphologic features and biologic behavior demonstrate that the tumors classified as BCACs are not solid variants of adenoid cystic carcinoma.

The differentiated diagnosis of BCAC includes basal cell adenoma, adenoid cystic carcinoma, basaloid squamous carcinoma, small cell (neuroendocrine) carcinoma, and cutaneous basal cell carcinoma. Cytologic features alone are frequently insufficient for making a distinction. Cellular atypia and the number of

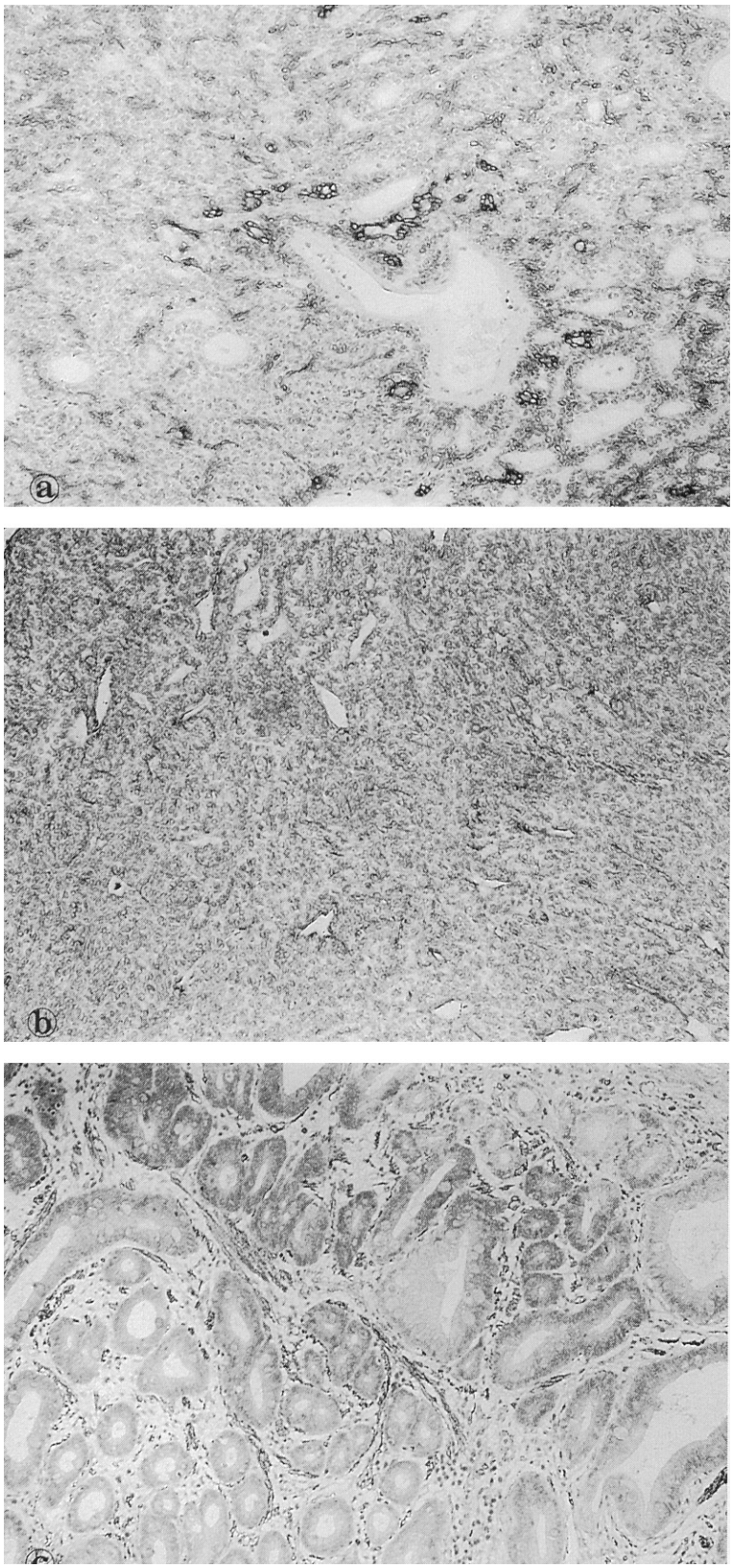

Fig. 6. Immunohistochemical staining of BCAC (a, cytokeratin; b, vimentin; c, smooth muscle actin). ×118.

TABLE 1. *Specifications of antibodies*

Antibody	Origin	Dilution
Smooth muscle antigen	DAKO (M851)	1 : 1000
S-100 protein	DAKO (Z311)	1 : 16 000
Vimentin	Boehringer (V9)	1 : 80
Cytokeratin	Immunotech (KL1)	1 : 300
Epithelial membrane antigen	DAKO	1 : 400
Carcinoembryonic antigen	Mochida (CEM010)	1 : 400

mitotic figures, in general, are greater in BCAC than in basal cell adenoma. Infiltrative, destructive growth, and neural and vascular invasion are characteristic features of BCAC (Table 2). Recently, Nagao et al. (1998) reported that cell proliferation, apoptosis, and expression of *p53*, *bcl*-2, and epidermal growth factor receptor were found to be useful in distinguishing BCAC from basal cell adenoma. Adenoid cystic carcinoma is also considered in the differentiated diagnosis. BCAC lacks the cribriform pattern and pseudocysts of amorphous, basophilic glycosaminoglycans characteristic of adenoid cystic carcinoma. Cytologic features are also helpful for differentiated diagnosis. The pale to clear cells with irregular, angular nuclei characteristic of adenoid cystic carcinoma contrast with the oval to round, eosinophilic cells and round nuclei of BCAC. Basaloid squamous carcinoma is a specific clinicopathologic entity that must be differentiated from BCAC since its clinically aggressive behavior justifies a multimodal therapeutical approach. In contrast to BCAC, basaloid squamous carcinoma has a malignant squamous component that involves the mucosal epithelium in the form of epithelial dysplasia, carcinoma in situ, or invasive squamous cell carcinoma. Immunohistochemically, basaloid squamous cell carcinoma is reported to be unreactive for muscle actin (Banks et al. 1992). In contrast to BCAC, small cell carcinoma often show endocrine differentiation and may therefore be positive for synaptophysin and neuron specific enolase, but negative for smooth muscle antigen and carcinoembryonic antigen (Muller and Barnes 1996). Cutaneous basal cell carcinoma that deeply invades the parotid

TABLE 2. *Diagnostic histological hallmarks between basal cell adenoma and adenocarcinoma*

Hallmarks	Adenoma	Adenocarcinoma
Cellular atypia	−	+
Mitosis	−	+
Invasive and multifocal growth	−	+
Perineural invasion	−	+
Vascular invasion	−	+

TABLE 3. *Immunohistochemical studies for differentiated diagnosis*

Antibody	BCAC	Basaloid squamous cell carcinoma	Small cell carcinoma	Cutaneous basal cell carcinoma
Smooth muscle antigen	+	−	−	−
S-100 protein	+	+	−	−
Vimentin	+	+	+	+
Cytokeratin	+	+	+	+
Epithelial membrane antigen	+	+	+	+
Carcinoembryonic antigen	+	−	+	−

gland from BCAC may be impossible by histology alone. The immunohistochemical identification of cells with myoepithelial differentiation of cell are features more compatible with BCAC (Muller and Barnes 1996). Differentiated diagnosis using immunohistochemical studies is summarized in Table 3.

BCACs are low-grade carcinomas. They are infiltrative, locally destructive, and tend to recur, but they only occasionally metastasize. The recurrence rate varies between 28% and 76% (Luna et al. 1989; Ellis and Wiscovitch 1990; Atula et al. 1993; Gallimore et al. 1994) with a mean rate of 35% (Fonseca and Soares 1996). Mortality is low. This clinical evolution is similar to that of the other subentities of the same family of adenocarcinomas of low-grade malignancy (adenoid cystic carcinoma, epithelial-myoepithelial carcinoma, and polymorphous low-grade adenocarcinoma). However, BCAC arising in previous, dermal analogue types of monomorphic adenomas are known to be especially aggressive neoplasms locally (Luna et al. 1989). Therefore, careful follow-up is needed. Considering the low-grade biologic potential of BCAC, surgical excision is appropriate treatment. Enucleation or curettage are to be avoided.

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