Basal Cell Adenocarcinoma of the Salivary Gland
- A Case Report -

Basal cell adenocarcinoma of the salivary gland is a very rare disease entity as the malignant counterpart of basal cell adenoma. On the basis of morphologic pattern, basal cell adenoma can be divided into four subtypes; trabecular, solid, tubular, and dermal analogue (membranous). We report a case of basal cell adenocarcinoma in the left parotid gland of a 33-year-old woman. Light microscopically, the tumor cells were composed of relatively uniform, monotonous basaloid cells. The tumor cell nests commonly had peripheral palisadings. The main growth pattern of the tumor cells was tubulotrabecular. However, other portions were similar to dermal analogue monomorphic adenoma and showed cribriform patterns reminiscent of adenoid cystic carcinoma. The tumor had frequent perineural invasions and an infiltrative margin. On immunohistochemistry, only scattered numbers of tumor cells showed irregular positive reaction for S-100 protein, suggesting a few myoepithelial cell components of the tumor compared with most tumor cells derived from epithelial cells.
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Key Words : *Adenocarcinoma; Salivary gland neoplasms; Immunohistochemistry*

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INTRODUCTION

Kleinsasser and Klein (1) proposed monomorphic adenoma of the salivary gland as a distinguishing term from pleomorphic adenoma. Monomorphic adenoma was classified as basaloid type and nonbasaloid type. Basal cell adenocarcinoma is a very rare disease entity as the malignant counterpart of basal cell adenoma (basal cell monomorphic adenoma). Basal cell adenocarcinoma can be divided into four subtypes: tubulotrabecular, solid, canalicular, and dermal analogue (membranous) (2, 3, 4). Most cases have arosen exclusively in the major salivary glands (parotid) except canalicular type which occurs almost always in intraoral minor salivary glnads in site of origin (3, 5). We report a case of basal cell adenocarcinoma in the left parotid gland and the results of the immunohistochemistry.

CASE REPORT

A 33-year-old woman had complained of an infraauricular painless mass of the left side for 5 years. The size of the mass was not greatly changed but seemed to be enlarged. She had no previous history of malignancy. Physical examination revealed a 2 cm sized, round, hard, fixed mass with tenderness at the left infraauricular area. Routine laboratory studies showed no abnormal findings. A sialogram demonstrated no abnormal ductal dilatation or stenosis. On operative field, there was a hard, fixed mass between the cervical and marginal mandibular branch of the deep lobe of the left parotid gland. Grossly, the mass, measuring $2.5 \times 1.6 \times 0.5$ cm in dimension, was moderately demarcated, round, whitish gray, multinodular, and solid. The cut surface showed an infiltrative margin into the surrounding normal parenchyme, with two small hemorrhagic foci (Fig. 1).

Light microscopically, the tumor cells were composed of relatively uniform, monotonous basaloid cells. Individual tumor cells were small, round with a dark basophilic nucleus and scanty cytoplasm. The stroma intervening the nests was focally myxoid. Nuclear pleomorphism or mitotic activity could not be found. The main growth pattern of the tumor cells was tubulotrabecular type (Fig. 2) forming definite central lumina. However, some portions were similar to dermal analogue monomorphic adenoma with solid, small nests of tumor cells having hyperchromatic nuclei and peripheral palisadings (Fig. 3). Other foci showed cribriform patterns reminiscent of adenoid cystic carcinoma (Fig.

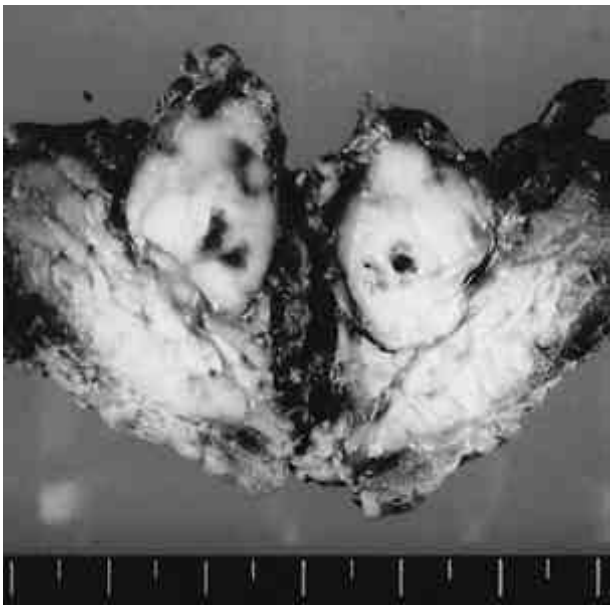

Fig. 1. The cut surface of the tumor showed a moderately demarcated, unencapsulated, gray-tan, solid, mutinodular appearance.

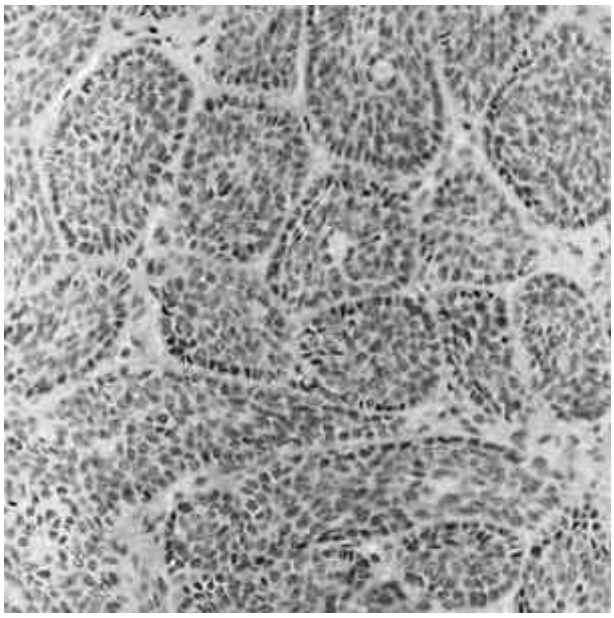

Fig. 3. Membranous growth pattern showing strong similarity to basal cell adenoma, dermal analogue type (H&E, ×200).

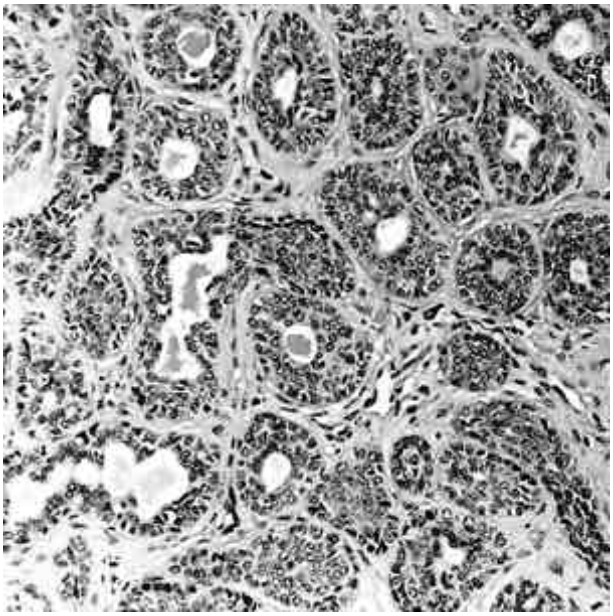

Fig. 2. Neoplastic cells arranged in solid nests forming early lumen corresponding to tubulotrabecular type (H&E, ×200).

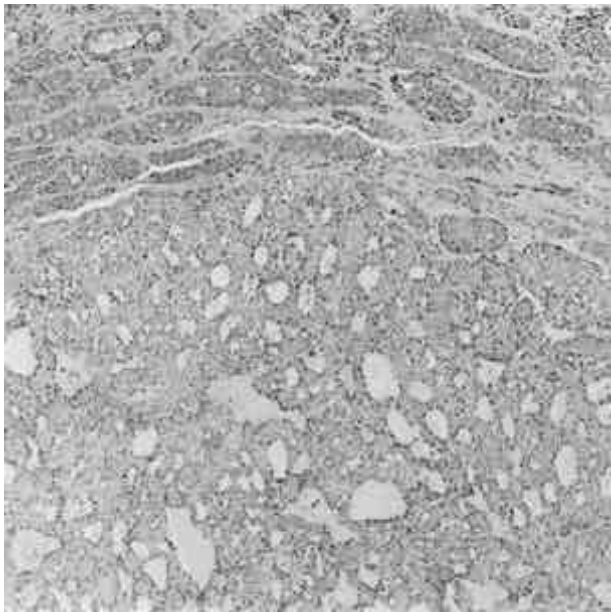

Fig. 4. Cribriform pattern reminiscent of adenoid cystic carcinoma. Nodular growth was characteristic in this portion (H&E, × 100).

4). The latter form was located in the center of the main tumor, which characteristically showed a multinodular growth pattern pushing the adjacent tumor cells. This feature is unique point in contrast to the previously reported cases. The tumor frequently showed perineural invasion (Fig. 5) and an infiltrative margin into the periglandular tissue (Fig. 6).

Immunohistochemical staining was done using mouse monoclonal anti-human antibody to cytokeratin (Zymed, Sanfrancisco, CA, USA) and rabbit polyclonal anti-human antibody to S-100 protein (Zymed, Sanfrancisco, CA, USA). All the tumor cells were strongly reactive for cytokeratin, and several tumor cells in the nests showed positive reaction for S-100 protein.

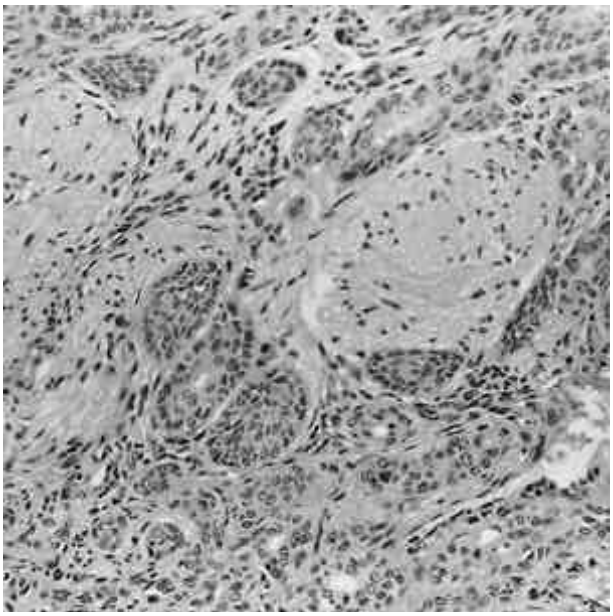

Fig. 5. Perineural invasion of the tumor cells was evident in many fields (H&E, ×200).

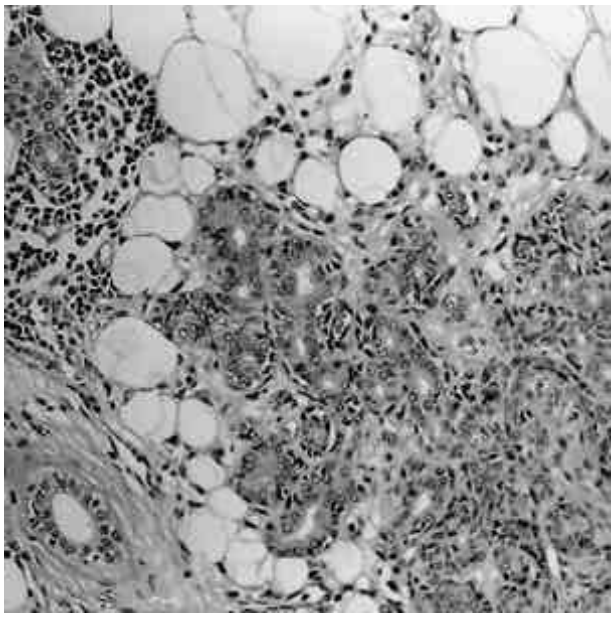

Fig. 6. Tumor cells infiltrating into periglandular adipose tissue which is part of the evidence of malignancy (H&E, ×200).

DISCUSSION

To understand the disease entity, basal cell adenocarcinoma, it is important to know the pathogenesis of basal cell adenoma. For the first time, Kleinsasser and Klein (1) proposed monomorphic adenoma of the salivary gland as a distinguishing term from pleomorphic adenoma. In 1972, the classification of the World Health Organization subdivided the monomorphic adenomas into three groups: adenolymphoma, oxyphilic adenoma, and other types (6). Since then, Bastakis et al. (4) reclassified monomorphic adenoma as basaloid (basal cell adenoma or basaloid adenoma) and nonbasaloid type. Basal cell adenocarcinoma of the salivary gland is the malignant counterpart of basal cell adenoma. Basal cell adenoma and basal cell adenocarcinoma can be divided into four subtypes: trabecular, solid, tubular, and dermal analogue (membranous) (4). Basal cell adenocarcinoma has been called basaloid salivary carcinoma, atypical variants of monomorphic adenomas, malignant basaloid tumors, hybrid monomorphic adenomas, or carcinomas ex monomorphic adenoma (7). Luna et al. (8) reported 8 cases with a mean size of 3.7 cm (3.0 to 7.0 cm) and 6 of these had recurrence. Among 29 cases of basal cell adenocarcinomas reported by Ellis and Wiscovitch (3), 26 cases arose from the parotid, and 3 cases occurred in the submandibular glands. Mean size was 2.1 cm (0.7 to 4.0 cm). Seven cases among 25 traceable patients recurred. One of them died after 10 years.

Cho and Kim (9) studied 12 cases of monomorphic adenomas of the salivary glands with immunohistochemistry. They supported the hypothesis that basal cell adenoma and myoepithelioma may be located at the two extremes of one spectrum, with pleomorphic adenoma interposed between the two with a variable phenotypic expression of myoepithelial nature. The nature of a basal cell adenoma seems to be a tumor with more differentiated epithelial cells than that of pleomorphic adenoma in which a certain role for myoepithelial cells is considered to be important in morphogenesis (8). Batsakis et al. (4) also agreed with this proposal. However, basal cell adenoma and carcinoma have not yet been fully defined.

In this case, the majority of tumor cells were composed of cell nests with a tubulotrabecular growth pattern. Despite the abscence of mitotic feagures, pleomorphism, and vascular invasion, there was definite perineural invasion and infiltration into the periglandular adipose tissue. Other portions showed membranous and cribriform growth patterns. The morphology of the latter is similar to adenoid cystic carcinoma, so it is important to differentiate this type of basal cell adenocarcinoma from adenoid cystic carcinoma. Cytologic features are helpful to distinguish the two carcinomas : 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 basal cell adenocarcinoma. The mixture of large pale and small dark cells common to basal cell adenocarcinoma is not a feature of adenoid cystic carcinoma. Also, basal cell adenocarcinoma usually has a prominent myoepithelial component around

the ductal elements which is not commonly seen in adenoid cystic carcinoma. The cribriform portions in this case characteristically had a multiple nodular growth pattern. This is a unique point in this case in contrast to the previously reported cases. On immunohistochemistry, all the tumor cells were strongly reactive for cytokeratin, but they showed an irregular positive reaction for S-100 protein in several portions of tumor cell nest. So, it might be deduced that the components of the epithelial cells differentiate more than do those of the myoepithelial cells.

Because of the rarity of this tumor, the prognosis has not been statistically established. However, basal cell adenocarcinomas are low grade with recurrence rates which approximate 28% (4, 8).

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