Late recurrence of acinic cell carcinoma of the parotid gland

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Abstract : Acinic cell carcinoma of the salivary glands is a rare cancer representing a low grade malignancy. The recurrence of a tumor is sometimes encountered, usually within 5 years of initial operation. We describe an unusual recurrence after a long interval following primary surgery. In 1987, a 60-year-old woman underwent excision of a mass in the superficial lobe of the right parotid gland under the preoperative diagnosis of a benign tumor. A histologic diagnosis of acinic cell carcinoma was made by examining sections from the resected mass. The patient noted several small nodules in the right parotid region in 1995, but she did not visit our clinic until 1998 when tenderness developed. A locally recurrent tumor and cervical lymph nodes containing metastases were resected and postoperative radiotherapy was given 11 years after the first operation. At least 10 years of follow-up may be necessary for patients with acinic cell carcinoma because of slow-tumor growth. J. Med. Invest. 46 : 213-216 1999

Key words **:** *acinic cell carcinoma, parotid gland, recurrence*

INTRODUCTION

Acinic cell carcinoma (ACC) is a rare, slow-growing, low-grade salivary gland cancer. Most ACC occur in the parotid gland, representing between 2 and 3% of all epithelial parotid neoplasms (1-3). The reported 10-year survival rate is approximately 70% (4-5). However, ACC has a tendency to develop local recurrence in 40 to 50% of the patients, usually within 5 years of surgery (3, 6).

We recently encountered an ACC patient in which a local recurrence was resected more than 10 years after initial surgery. Thus, prolonged, careful follow-up for 10 years or more seems essential, because the relatively indolent course of ACC may delay recurrence in some patients rather than precluding it.

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CASE REPORT

A 60-year-old woman who had noted enlargement of a small, painless mass in the right lateral region of the face for 10 years was referred in January 1987 to our hospital. An elastic hard mass with a smooth surface and a diameter of 3.5 cm was palpated in the right parotid gland at the time of initial presentation. No facial paralysis or enlargement of cervical lymph nodes was present. Computed tomography (CT) and ultrasonography both revealed a spherical solid tumor with a sharp border, leading to a preoperative diagnosis of benign parotid neoplasia. In January 1987, the patient underwent excision of the tumor from the superficial lobe of the right parotid gland, including two nearby lymph nodes. The histologic diagnosis was not established until permanent sections were examined, showing a microcystic pattern of acinic cell carcinoma associated with abundant lymphocytic infiltrates (Fig.1). Macroscopically, the tumor seemed completely resected. However, infiltration surrounding parotid tissue was found microscopically. Malignant cells were not detected in the two lymph nodes. No facial nerve paralysis was evident after tumor resection.

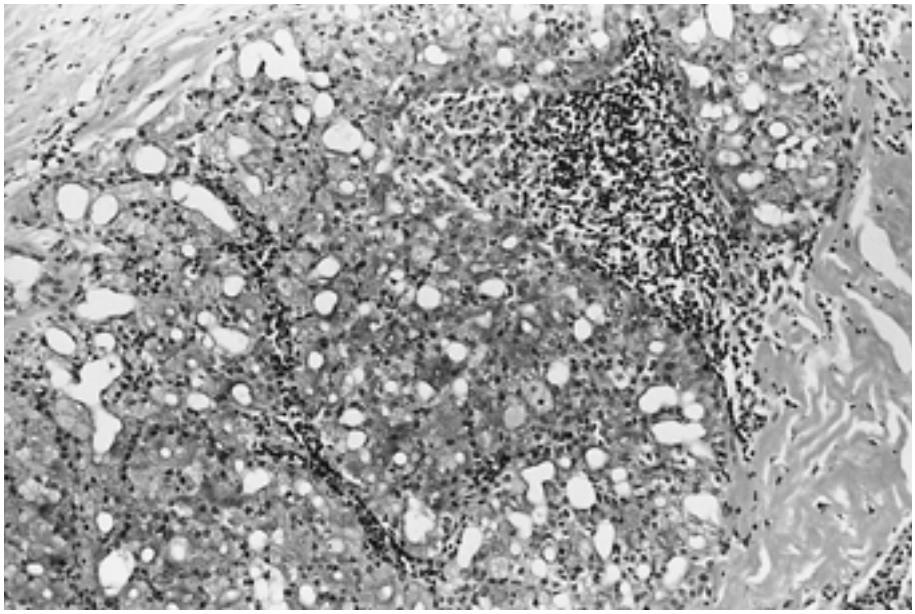

Fig.1. Microscopic appearance of primary acinic cell carcinoma. It showed a microcystic pattern associated with abundant lymphocytic infiltrates (hematoxylin and eosin).

No postoperative radiotherapy or chemotherapy was administered.

In 1995, 8 years after the operation, the patient noted several small nodules in the right parotid region, but did not consult our outpatient clinic even though the size and number of these nodules gradually increased. She finally returned when tenderness developed in the right parotid region in January 1998. CT revealed a mass 2.0 cm in diameter with an irregular shape showing soft-tissue density in the right parotid gland, and several small nodules consistent with enlarged cervical lymph nodes were seen near the lateral portion of the right sternocleidomastoid muscle (Fig. 2). MRI and ultrasonography revealed similar findings. A fine-needle aspiration biopsy specimen was characterized by loosely cohesive, granular cells with mildly atypical nuclei (Fig. 3). Local recurrence of acinic cell carcinoma and metastasis to cervical lymph nodes were diagnosed, and resection of the locally recurrent tumor in the parotid gland combined with right cervical lymphadenectomy was performed 11 years after initial surgery, in February 1998. The recurrent tumor in the superficial lobe measured 2.5× 1.8 cm in area, and was adherent to the facial nerve. A hard, nodular mass measuring 7.0×3.1×1.5 cm, located laterally to the parotid tumor, represented metastatic lymph nodes adherent to one another. Histologically, the recurrent and metastatic tumors showed the microcystic pattern of well-differentiated acinic cell carcinoma, appearing very similar to the initially resected tumor (Fig. 4). Abundant lympho-

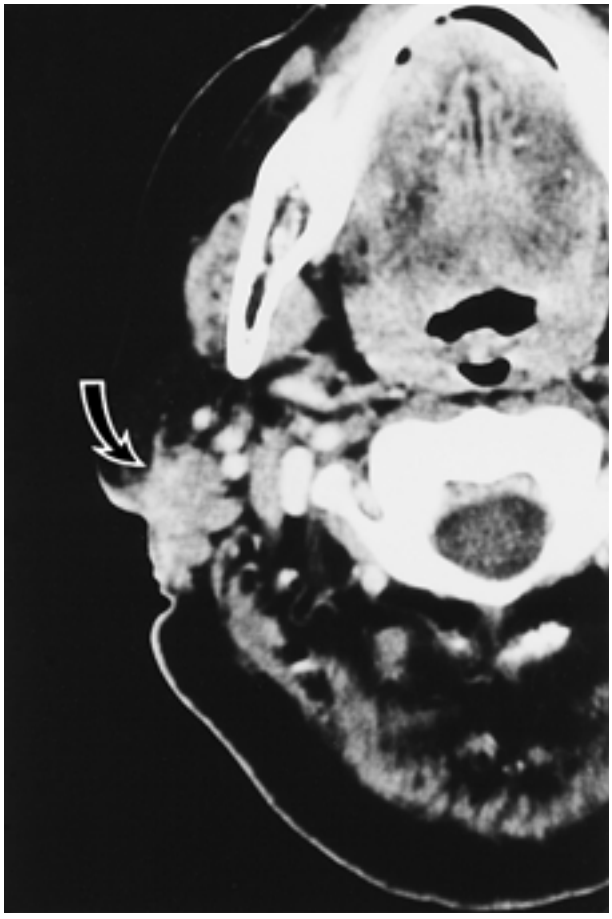

Fig. 2. Computed tomography of the right parotid region. The tumor (arrow), an acinic cell carcinoma 2.0 cm in diameter, has recurred at the initial operative site.

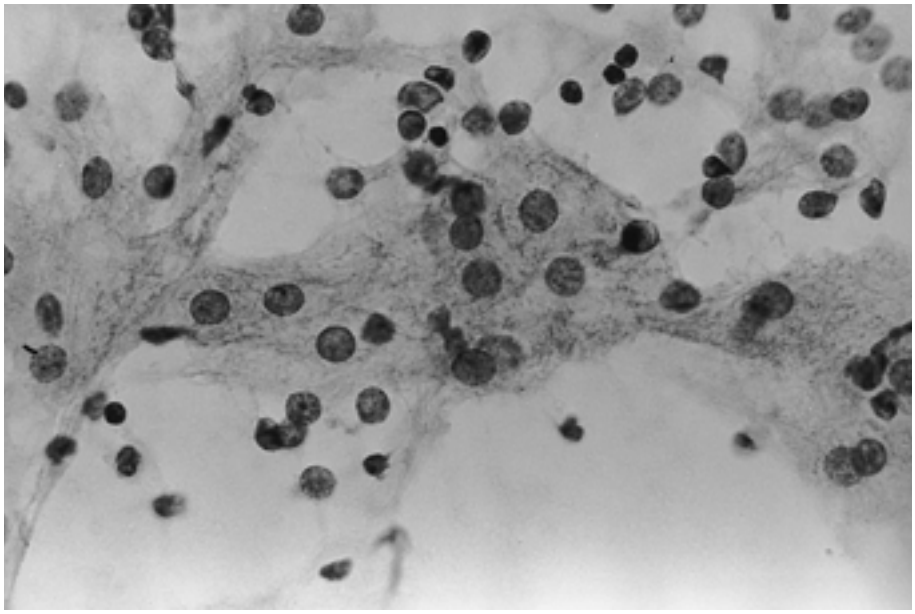

Fig. 3. Fine-needle aspiration biopsy specimen of the recurrent right parotid mass. Loosely cohesive, granular cells with mildly atypical nuclei are seen (Papanicolaou's, ×400)

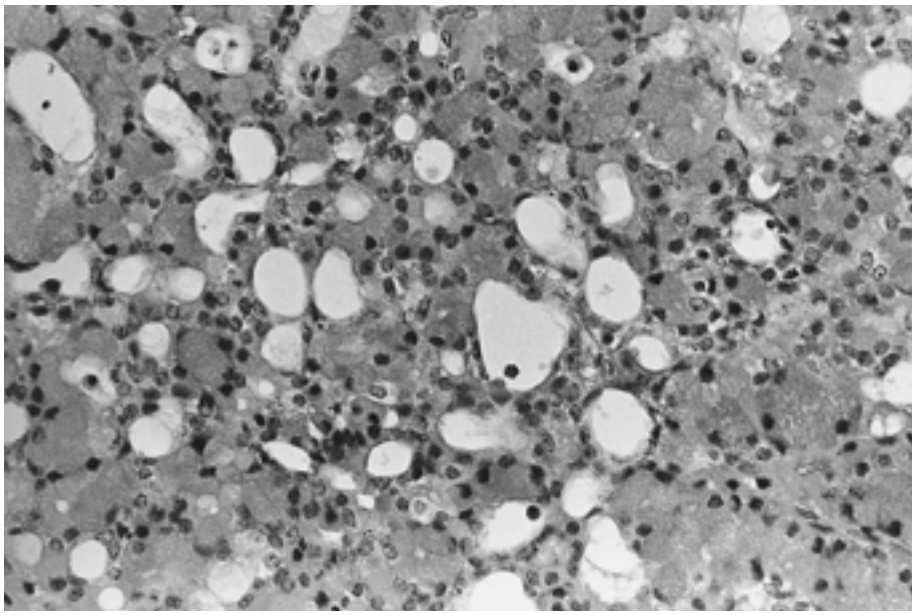

Fig. 4. Microscopic appearance of the acinic cell carcinoma recurrence. The recurrent tumor showing cells with darkly stained cytoplasm in a microcystic pattern appeared similar to the initial resection specimen (hematoxylin and eosin).

cytic infiltrates were also found in the recurrent tumor.

A right facial nerve palsy persisted for 4 months after the second operation, although the nerve was preserved during surgery. Postoperative radiotherapy was given at a dose of 50 Gy to eradicate microscopic residual disease. No major complications related to radiotherapy were observed. No new recurrences of a tumor are evident to date.

DISCUSSION

ACC is an uncommon cancer accounting for 2 to 3% of all salivary gland neoplasms and 10 to 16% of malignant salivary gland tumors (1-3, 7). Women are affected more often than men, and peak occurrence is seen in the fifth decade (1, 2, 8). More than 90% of reported ACC has arisen in the parotid gland(2). Pain or tenderness occurs in about 20% of the patients (9, 10), but most of these tumors have no symptoms except for the development of a solitary firm mobile mass in the parotid region, that is clinically indistinguishable from benign tumors. The presence of a mass was the only symptom in our patient at initial presentation. Preoperative facial paralysis is rare (<3%) (2).

Noteworthy characteristics of ACC are low-grade

malignant behavior and slow growth. ACC has a relatively good prognosis with 5- and 10-year survival rates between 76 and 100% and between 63 and 87%, respectively (1, 4-5). However, ACC tends to recur locally with a frequency between 40 and 50% (2-3). The evolution of ACC seems to be largely influenced by the type of surgery employed. Chong, *et al*. (11) have found that 67% of patients eventually develop recurrences at sites of initial local excision. Recurrent disease is often multifocal, invasive, and difficult to treat (2). Our patient initially underwent tumor extirpation and later developed a local recurrence. More aggressive surgery, such as superficial parotidectomy or total parotidectomy with nerve preservation, has been recommended as a first operation by Spafford, *et al*. (2) and Oliveira, *et al*. (1). The location in the deep lobe of the parotid gland and large size are also thought to indicate a tendency towards recurrence. However, no relationship between either morphological pattern or cytological type and prognosis was found. Recurrence usually develops within 5 years of the initial operation (3). Chilla, *et al*. have reported a mean interval for recurrence of 4.5 years (6). However, Oliveira, *et al*. (1) and Napier, *et al*. (8) have observed recurrences 17 and 20 years after initial surgery. Our patient had a second operation for the local recurrence 11 years after the initial treatment. Although with our patient, the reason for recurrence after such a long interval is unclear. At least 10 years of careful follow-up may be necessary with ACC patients.

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