A Case Report of Salivary Duct Carcinoma

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IKEDA, K., YOSHIDA, F., SUZUKI, H., OSHIMA, T., HIRANO, K. and TAKASAKA, T. *A Case Report of Salivary Duct Carcinoma*. Tohoku J. Exp. Med., 1997, **183** (3), 233–238 — We present a 65-year-old man with an enlarged mass in the right parotid gland. A fine-needle aspiration cytology suspected Warthin's tumor. The ill-defined margin of the tumor in diagnostic imaging and unexpected clinical course of the occurrence of facial nerve paralysis suggested malignant neoplasm. The patient was treated with surgical resection of the primary site and neck following radiation therapy. Pathologic diagnosis was a salivary duct carcinoma. Difficulty in early diagnosis of this tumor may result in local extension, early metastasis to regional lymph nodes and distant sites, and death. — salivary duct carcinoma; fine-needle aspiration; parotid gland; electron microscope
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Salivary duct carcinoma (SDC) is a rare but high grade adenocarcinoma arising from the ductal epithelium of salivary glands. SDC was first described by Kleinsasser et al. (1968). The tumors are characterized by their resemblance histologically to breast carcinoma of the distal type and by their highly aggressive behavior. Confusion in early diagnosis including the potential pitfalls of fine-needle aspiration (FNA) cytology and nonspecific patterns of diagnostic images may result in local extension, early metastasis, and significant mortality. This paper presents a case of SDC, showing some of the characteristic clinical and pathological features.

CASE REPORT

A 65-year-old man was referred to our clinic in April 1995 with a half year history of a swelling arising in his right neck. Physical examination revealed a single painless, firm, and mobile 3-cm mass in the parotid area. No cervical lymph nodes were palpable. Facial nerve function was intact. Computed tomographic scan showed a lesion almost entirely replacing the parotid gland.

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Fig. 1. An axial computed tomographic view showing nearly total replacement of parotid gland with the tumor (an arrow).

Fig. 2. A coronal magnetic resonance imaging showing a low signal intensity mass with ill-defined infiltrative margins (an arrow).

Magnetic resonance imaging revealed that the tumor had an ill-defined infiltrative margin in the parotid gland with low signal intensity in both T1- and T2-weighted images (Figs. 1 and 2). Metastatic cervical nodes were recognized by computed tomography. FNA cytomorphic features showed moderate cellular aspirates composed of large, flat and monomorphic sheets of epithelial cells with abundant dense eosinophilic cytoplasm and enlarged nuclei with prominent nucleoli

together with lymphocyte infiltration, interpreted as the features of Warthin's tumor.

The patient showed an abnormality in electrocardiogram and was referred to cardiologists. During treatment of arrhythmia, partial facial paralysis in the right side was noticed. Re-examination of FNA biopsy, however, showed no evidence of malignancy. A total parotidectomy and right radical neck dissection were performed in May 1995, since frozen sections of the parotid tumor during

Fig. 3. Salivary duct carcinoma with characteristic comedonecrosis (an arrow). Hematoxylin and eosin $\times 200$.

Fig. 4. Electron micrograph of salivary duct carcinoma showing polygonal to columnar cells containing mitochondria and whorl-like rough endoplasmic reticulum (an arrow). $\times 4000$.

operation were suspected of being oncocytic adenocarcinoma. Light microscopic examination of surgical specimens (Fig. 3) showed solid, cribriforming, and papillary nests of tumor cells with central comedonecrosis. The tumor cells are polygonal with eosinophil cytoplasm, prominent nucleoli, and minimal pleomorphism. Metastatic carcinoma was identified in 7 out of 9 cervical lymph nodes by a histopathologic examination. Furthermore, the tumor were examined ultrastructurally (Fig. 4). The polygonal to columnar tumor cells contained a large oval nucleus with dense chromatin. The cytoplasm contained several mitochondria and rough endoplasmic reticulum forming circles. However, there was no cluster of enriched mitochondria as shown in oncocytic adenocarcinoma. These morphological features suggested the diagnosis of salivary duct carcinoma (Garland et al. 1984; Brandwein et al. 1990; Delgado et al. 1993; Yoshihara et al. 1994). Postoperatively the patient received 60 Gy of radiation to the parotid and neck. The patient is well 25 months later.

DISCUSSION

SDC is histologically characterized by solid cribriforming and papillary nests of tumor cells with central comedonecrosis. The tumor cells are large polygonal with abundant eosinophilic cytoplasm prominent nucleoli, and minimal pleomorphism. The differential diagnosis of SDC includes high grade mucoepidermoid carcinoma, oncocytic adenocarcinoma, acinic cell carcinoma, and metastatic adenocarcinoma. Mucoepidermoid carcinoma is distinguished by its more variable cell population, including the presence of squamous cell and goblet cells and the absence of a cribriform pattern. The cytoplasm of oncocytic adenocarcinoma is finely granular because of the presence of a large number of mitochondria, which can be detected by phosphotungstic acid hematoxylin stain or electron microscopy. Oncocytoma and epithelial-rich Warthin's tumor may resemble SDC, showing large flat and monomorphic sheets of epithelial cells with abundant dense eosinophilic cytoplasm and enlarged nuclei with prominent nucleoli. On the other hand, SDC usually shows a higher nuclear cytoplasmic ratio and less granular cytoplasm. FNA biopsy is an efficient and cost-effective diagnostic tool for significant masses of the salivary glands (Frable and Frable 1991). It is still difficult to confirm the cytologic differential diagnosis of SDC by FNA biopsy (Layfield and Glasgow 1991; Elsheikh et al. 1994). Although this case was first misdiagnosed as Warthin's tumor or oncocytic adenocarcinoma, light and electron microscopic findings in the surgically resected materials showed the characteristic features of SDC.

SDC is uncommon, but clinically is characterized by its aggressive behavior with early metastasis, local recurrence, and significant mortality. The patients having SDC typically present with a rapidly enlarging firm mass in the form of extrasalivary extension that is accompanied by facial paralysis or pain. Cervical adenopathy is commonly detected. The clinical course is rapid and dismal. Hui

et al. (1986) reported poor prognostic features including the presence of infiltrative borders, lymphatic invasion, invasion of adjacent structures, lymph node metastasis, recurrence and distant metastasis. However, statistical analysis in recent studies (Colmenero Ruiz et al. 1993; Grenko et al. 1995; Lewis et al. 1996) demonstrated no correlation with outcome for lymph node metastasis, vascular invasion or perineural invasion. These findings suggest the aggressive clinical behavior of SDC, with frequent and early metastasis and death due to disease.

The treatment most frequently given to the patients was a combination of operations on the neck and primary site, and radiotherapy. Delgado et al. (1993) have suggested that smaller tumors may be amenable to total parotidectomy with preservation of the facial nerve. Although radiotherapy has not yet proven to be a significant factor in overall survival, many of the reported long-term survivors had received postoperative radiotherapy (Afzelius et al. 1987; Luna et al. 1987; Colmenero Ruiz et al. 1993). The present patient also received radiation following radical operation for both primary site and neck, which may have contributed to the 25 month survival after diagnosis.

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