CASE REPORT

Adenocarcinoma of the Sublingual Gland

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(Received for publication on June 23, 1990)

Abstract. Primary adenocarcinoma arising in the sublingual gland is very rare. In this report, we have described the details of a case of adenocarcinoma of the sublingual gland. A 27-year-old Japanese male was referred to our department with a swelling of the floor of the mouth on the right side. The patient underwent a wide resection of the lesion and dissection of the right upper neck. Twelve months after his primary surgery, he was readmitted to hospital because of a metastasis in the lower lobe of the right lung and a right lower lobectomy was performed. He has undergone periodical controls for 3 years. No sign of recurrence or metastasis has been observed. (Keio J Med 40 (1): 20-24, March 1991)

Key words: salivary gland tumor

Introduction

Among tumors of the head and neck, the salivary glands are relatively common. Most such tumors arise in the parotid, submandibular and minor salivary glands; tumors of the sublingual gland are extremely rare. Among the malignant tumors of the salivary glands, adenoid cystic carcinoma, mucoepidermoid tumor, carcinoma of pleomorphic adenoma and adenocarcinoma are frequent.

In this paper, a case of an adenocarcinoma of the sublingual gland with subsequent pulmonary metastasis is reported. Special reference to its incidence, pathological features and treatment is made.

Case Report

A 27-year-old Japanese male came to our clinic with a small, painless swelling under his chin for the first time in April, 1985; no other symptoms had been noted during the preceding 3 months. Upon examination a mass on the right side under his tongue was found. The mass, which was non-tender and 2.5×3.0 cm in size, was located in the region of the salivary gland. The skin in the area was of normal color, and there was no cervical lymphadenopathy. The mass, examined from the oral cavity, was firm, well-demarcated, non-ulcerated and 2.0×1.5 cm in size (Fig. 1). Sonography demonstrated a heterogeneous mass located in the area corresponding to the sublingual gland which was 3.0×3.0 cm in size and contained a cystic region. A scintigram of the salivary gland revealed that the major salivary glands (parotid and submandibular) were intact and that the sublingual gland was not distinct (Fig. 2). The histopathological findings in the biopsy specimen were consistent with adenocarcinoma. Chest X-rays did not reveal any abnormality and laboratory data were within normal limits. Based on the above findings a clinical diagnosis of malignant tumor of the sublingual gland was made.

Treatment

The patient underwent a resection of the lesion and lymph nodes dissection of the right upper neck in October, 1985. All the tissue enclosing the mass was removed en bloc. The tumor, measuring 3.5×3.0×1.5 cm, was partially attached to the soft tissue of the floor of the mouth. The sublingual gland had for the most part been invaded by the tumor, which was separable from the surrounding tissue (Fig. 3). Twelve months after surgery, a coin lesion, possible metastatic, 1.0×1.0 cm in size was observed on a chest X-ray and was confirmed by CT scan (Fig. 4a, b). Right lung lower lobectomy was performed; pathologically the specimen was consistent with undifferentiated carcinoma. Two courses of postoperative chemotherapy, combined CDDP + adriamycin, were instituted. The patient has survived for more than 4 years.
Gross pathology

The mass was surgically removed from the right sublingual fossa. A grayish-white solid tumor was identified on the cut surface.

Histopathology

The primary tumor was diagnosed as adenocarcinoma (Fig. 5a, b). The tumor cells had large, irregularly arranged nuclei, rich in chromatin, and inconspicuous nucleoli. The moderately profuse cytoplasm stained clear.
and eosinophilic with H-E. The tumor cells grew invasively in solid focal and fascicular patterns. Mitoses were scattered. Tubular patterns, containing an amorphous, homogeneous and eosinophilic substance and, in some parts, a small amount of mucoid secretory product, were observed. Some areas with relatively large microcystic compartments were also present.

The metastatic tumor was diagnosed pathologically as undifferentiated carcinoma (Fig. 6a, b). The tumor cells had pathological features similar to those of the sublingual tumor. The atypical tumor cells had hyperchromatic, large, oval and irregularly shaped nuclei with scattered mitoses and their cytoplasm was profuse and stained clear and slightly eosinophilic. Necrotic areas were observed and the cells grew in a solid pattern. Areas with tubular patterns and microregions of keratinization were observed rarely. The above findings were consistent with the diagnosis of undifferentiated carcinoma showing characteristic differentiation in both directions.

**Discussion**

Recently salivary gland tumors have been clearly classified into malignant and benign tumors, and a WHO histological classification was proposed in 1972. It defines adenoma as a benign and mucoepidermoid, acinic cell tumor, and carcinoma as malignant. Adenocarcinoma in particular is defined as an epithelial malignant tumor with high malignant potential.

Foote and Frazell reported proportions of 63% and 37% for benign and malignant tumors, respectively, in a series of 877 salivary tumors; Chaudhry et al reported 61% and 39%, respectively, in a series of 1414 minor salivary tumors; Eneroth reported 79% (1831) and 21% (480), respectively, in a series of 2311 salivary tumors. The percentages reported in all of these papers were comparable.

The sites involved, as reported by Eneroth, were the parotid gland (80%), submandibular gland (5–10%), and sublingual gland (less than 10%), Conley reported
that sublingual tumors comprised 1% of 1280 major salivary tumors, 90% of which were malignant. Rankow also reported that a majority of sublingual tumors were malignant. Since Brunshwig reported the first case of malignant mixed tumor in 1930, a number of cases of similar malignancies arising in the sublingual gland have followed. Reviewing the literature, we found 73 such cases, including our own, which have been reported in the last 58 years from 1930 to 1988. The results indicate that malignant tumors of the sublingual gland comprise 0.2-5.9% of tumors of the major salivary glands and 0.5-5.9% of malignant tumors of the major salivary glands. Histologically, these included adenoid cystic carcinoma in 28 patients (38.4%), mucoepidermoid tumor in 15 (20.5%), carcinoma in pleomorphic adenoma in 11 (15.8%), and adenocarcinoma in 8 (11.0%), the first case of which was reported by Shimada in 1955. Thus, adenocarcinoma arising in the sublingual gland appears to be a rare entity. In the report by Foote and Frazell, adenocarcinoma accounted for 4% of tumors of the major salivary glands, 82% arising in the parotid gland, 13% in the submandibular gland, and 5% in the sublingual gland. Seifert and Schulz reported that adenocarcinoma accounted for 1.2% of salivary gland tumors and 10% of malignant epidermoid tumors of the salivary glands, 58.5% arising in the parotid gland, 28.5% in the minor salivary glands, 11.5% in the submandibular gland and 1.5% in the sublingual gland.

Occurrence is predominantly after middle age. One fourth of all of the tumors, reported by Seifert and Schulz, occurred in the patients' seventies and 66% of them occurred between 50 and 80 years of age. Forty-five males (58.8%) and 32 females (41.5%) were affected, and the male/female ratio has been reported elsewhere to be 2.8:1, suggesting male preponderance.

The tumors, exhibit rapid local invasive growth and give rise to clinical symptoms, such as pain, numbness, and swelling, depending on nerve and subcutaneous involvement. In our patient, swelling extended rapidly inside and outside the oral cavity within three months since the initial swelling was noted in the sublingual region. The tumors tend to recur locally and often...
metastasize to lymph nodes, lung, and bones. The tumors spread by direct extension, via the lymphatic or the blood stream. The prognosis is poor when a distant metastasis is present. Among cases of postoperative recurrence distant metastases to the lung are observed in about 50% of the cases. Due to facts exposed above, a total scincigraphy is specially needed.

As an imaging technique, scincigraphy was carried out in first place; however, there were no signs of metastasis in it. Regarding the operation, although it is necessary in most cases, a computed tomography was not done in this case because: a) clinically, a solid mass was detected in the area corresponding to the sublingual gland; b) the mucosa of the floor of the mouth was normal; c) there was no relation between minor salivary glands and the solid mass; and, d) the patient wanted to be operated on as soon as possible. In our own case, metastasis, presenting as a solid mass in the lower lobe of the right lung, was discovered 18 months after the patient's initial symptoms and 12 months after the surgery and might have developed due to surgical intervention.

The tumor, originating in multipotential epithelial duct cells has diverse histopathological features. Seifert and Schulz classified it into 4 types, i.e., solid, tubular, papillary, and tubular-papillary. The solid type, comprising 13%, is poorly differentiated histologically and tends to be invasive with frequent metastases. The tubular type, accounting for 52%, is highly differentiated with mucus production and few microcysts. The papillary type comprises 28.5%, 50% of which are usually accompanied by mucus production and the presence of microcysts. The tubular-papillary type is infrequent (6.5%). The tumor in our own case can be classified as solid type.

The tumor is unresponsive to radiation therapy. Chemotherapy with 5-FU was tried and proved effective in the prevention of micrometastases and tumor growth. The treatment of first choice is surgery, which thus far seems to be the most beneficial. Our patient, whose primary tumor was localized in the sublingual region, underwent a radical tumor excision. The lower lobe of the lung was then excised to remove the subsequent metastatic lesion, apparently localized in the lower lobe. The courses of chemotherapy, combined CDDP + adriamycin, were administered. At the present, 5 years after surgery of the primary lesion and 4 years after removal of the metastatic lesion, our patient has followed an uneventful course with no further recurrence or metastasis.

References