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Acinic Cell Carcinoma of the Sublingual Gland Accompanied by Bone Formation

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A rare case of acinic cell carcinoma of the sublingual gland accompanied by bone formation is reported. The patient is a 79-year-old male who was referred to Yokohama Minami Kyosai Hospital with sublingual swelling. A tumor mass, 20 × 10 mm in diameter, was detected on the right side of the floor of the mouth. Computed tomography (CT) revealed a mass lesion with calcification in the sublingual gland. The patient underwent total sialadenectomy of the sublingual gland with conservation of the lingual nerve. Histologically, the lesion showed amylase-positive atypical cells with thyroid gland-like arrangement, and mature bone tissue in the stroma. Based on these findings, the tumor was diagnosed as acinic cell carcinoma accompanied by bone formation. Postoperative recovery was uneventful, and two years after surgery, there are no signs of distant metastases or recurrence.

Key words : Acinic cell carcinoma, Bone formation, Sublingual gland

INTRODUCTION

Sublingual gland tumors account for less than 1% of all salivary gland tumors, and about 80% of sublingual gland tumors are malignant [1]. This report describes a case of acinic cell carcinoma of the sublingual gland accompanied by bone formation.

CASE REPORT

A 79-year-old male was referred to Yokohama Minami Kyosai Hospital on October 15, 1999 with a ten-month history of swelling of the floor of the mouth. A tumor mass, 20 × 10 mm in diameter, was detected on the right side of the floor of the mouth (Fig. 1). Mobility of the tongue was not restricted, and sensory paralysis was not evident. Cervical lymph nodes were not palpable. CT revealed a mass lesion with calcification in the sublingual gland which was clinically diagnosed as a sublingual gland tumor. Total sialadenectomy of the sublingual gland with conservation of the lingual nerve was performed under general anesthesia on November 30, 1999. Macroscopic examination of the resected specimen revealed a solid tumor covered by a thin fibrous capsule in the sublingual gland. The cut surface of the tumor was dark yellowish in color (Fig. 2). Tumor sections showed proliferation of mildly atypical cells in a follicular thyroid-like pattern arrangement and colloid-like pools in small and large follicles. Mature bone trabeculae were seen in the stroma, and cells similar to osteoblasts were observed in some areas surrounding the bone tissue. Atypical cells resembling serous acinar cells, a characteristic finding of acinic cell carcinoma, were not observed (Fig. 3a, b). Immunohistochemical staining was performed using the LSAB method. Tumor cells were positive for salivary amylase (Fig. 3c), lactoferrin and EMA, but negative for S-100 protein, smooth muscle actin and thyroglobulin. The tumor was therefore diagnosed as acinic cell carcinoma; a follicular variant with bone formation in the stroma.

Two years after surgery, the patient continues to be followed on an outpatient basis and his postoperative course has been uneventful to date. Systemic examinations have revealed neither distant metastases nor recurrence.

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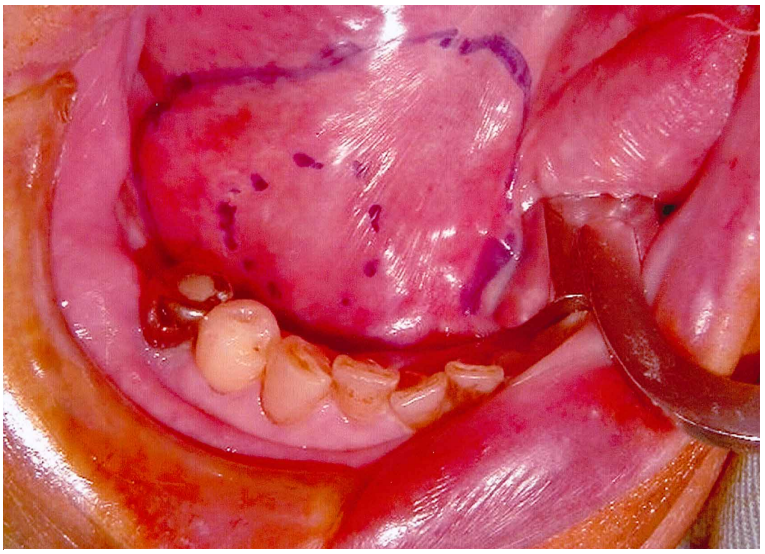

Fig. 1 Intraoral findings at surgery. Swelling of floor of the mouth was found. The dotted line indicates the tumor and the solid line indicates the incision line.

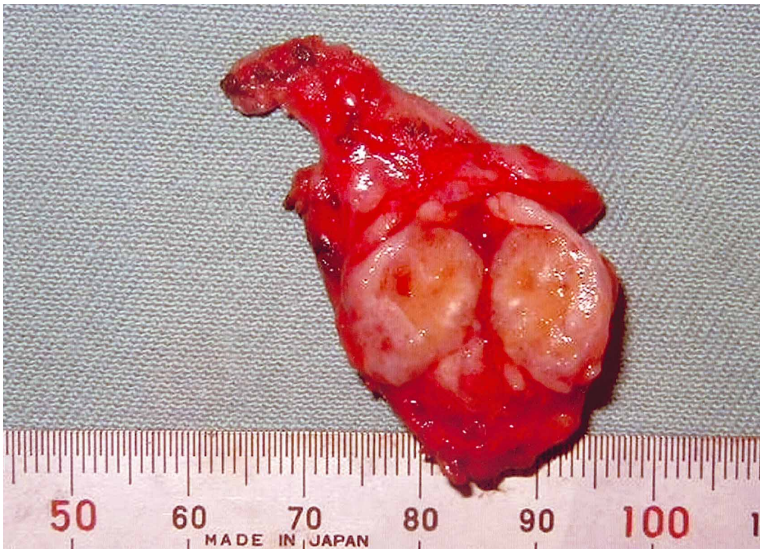

Fig. 2 Resected specimen. The tumor in the sublingual gland is covered with a thin fibrous capsule.

DISCUSSION

Acinic cell carcinoma is a rare tumor consisting of cells resembling serous acini of the salivary gland. The tumor accounts for 1–3% of all salivary gland tumors, with about 90% of acinic cell carcinomas occurring in the parotid gland [1, 2]. Tanamoto *et al.* reported an incidence of acinic cell carcinoma in sublingual tumors of 4.5% [3]. Accurate preoperative diagnosis of this tumor remains difficult because the tumor shows slow growth and is thus difficult to differentiate from benign tumors. Confirmation of the diagnosis relies on histopathological examination. In this case, although there were no clinical signs of malignancy, histopathological and immunohistochemical

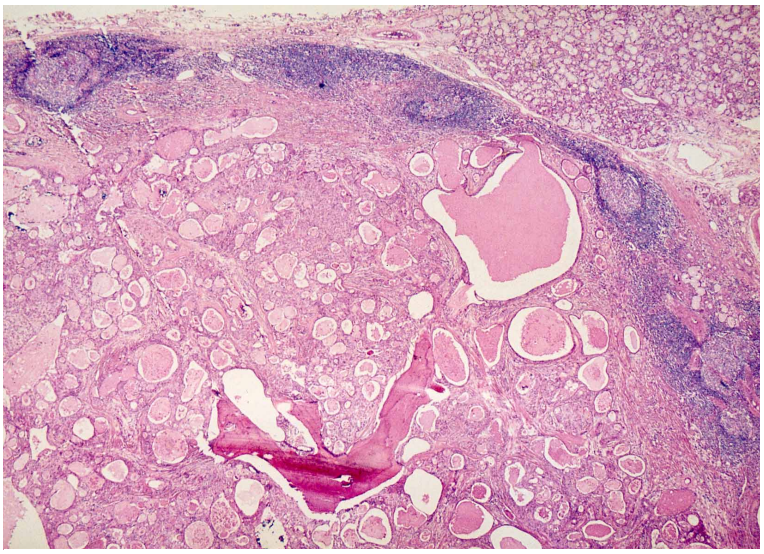

Fig. 3a Histological findings.
Microphotograph showing proliferation of relatively mild atypical cells with follicular arrangement and bone formation in the stroma. (× 5, H-E stain)

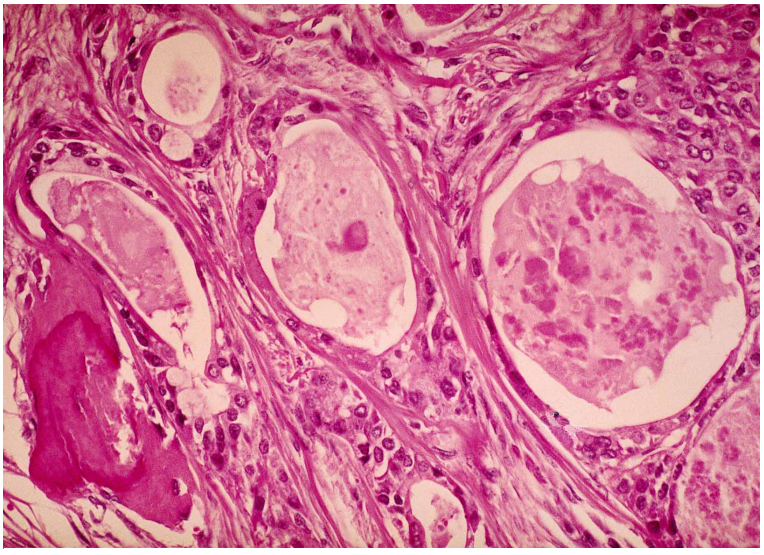

Fig. 3b Histological findings.
Microphotograph showing proliferation of relatively mild atypical cells with thyroid-like appearance and mature bone formation in the stroma. (× 160, H-E stain)

findings confirmed the diagnosis.

Histopathologically, acinic cell carcinomas consist of cells resembling serous acini that show a positive immunohistochemical reaction to anti-amylase antibody. Of the four growth patterns of acinic cell carcinoma that have been identified - solid, microcystic, papillary-cystic and follicular - the follicular pattern is the rarest [4]. In this case, the tumor predominantly demonstrated thyroid-like follicular proliferation. As follicular acinic cell carcinoma is not characterized by typical serous acinar cells histologically, there is a risk of misdiagnosis [5]. In our case, typical

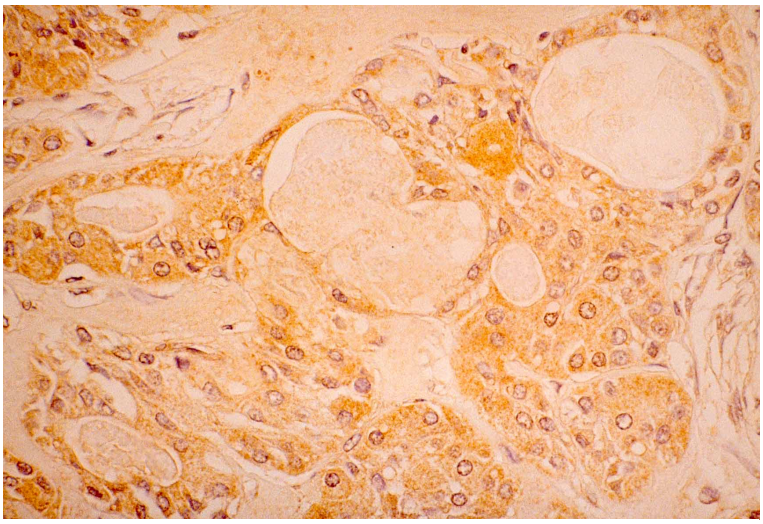

Fig. 3c Histological findings.
Microphotograph showing tumor cells with a positive amylase reaction. (× 66, SAB method)

serous acini were largely absent. However, since the tumor cells showed a positive immunohistochemical reaction to anti-amylase antibody and a negative reaction for thyroglobulin, the tumor was diagnosed as acinic cell carcinoma. Ultrasonography to rule out metastasis from a thyroid cancer (follicular pattern) found no evidence of a tumor in the thyroid gland.

Bone formation inside the tumor was confirmed in the present case. Stromal metaplasia tends to be the cause of bone formation in salivary gland tumors [6], and the literature contains only a few reports of bone formation from parenchyma in cases of mixed tumors [7, 8]. In our case, because there were no findings of parenchymal bone formation, bone formation was attributed to stromal metaplasia. To our knowledge, no previous study has reported acinic cell carcinoma accompanied by bone formation. Our case therefore appears to be an extremely rare subtype of acinic cell carcinoma.

Because the 5-year survival rate of patients with acinic cell carcinoma is statistically favorable at 76–89% [5], acinic cell carcinoma was once considered benign. However, it is now recognized to be predominantly malignant. No characteristic histopathological findings have been identified, and prognosis is largely dependent on the extent of local invasion and on the completeness of surgical resection [5]. In the present case, the good mid-term outcome is likely due to the fact that the tumor did not infiltrate the sublingual space and could be resected completely. However, due to the malignant nature of this disease, careful long-term follow up is advised.

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