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Author(s)	Matsuzaka, K; Shibahara, T; Murakami, S; Lee, DH; Shimono, M; Inoue, T
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Case Report

ACINIC CELL CARCINOMA FOUND BY RECURRENCE OF A MUCOUS CYST IN THE SUBLINGUAL GLAND

KENICHI MATSUZAKA, TAKAHIKO SHIBAHARA*, SATOSHI MURAKAMI,
DONG-HO LEE, MASAKI SHIMONO** and TAKASHI INOUE

*Oral Health Science Center,
Department of Clinical Pathophysiology, Tokyo Dental College,
1-2-2 Masago, Mihama-ku, Chiba 261-8502, Japan*

** The 1st Department of Oral and Maxillofacial Surgery, Tokyo Dental College,
1-2-2 Masago, Mihama-ku, Chiba 261-8502, Japan*

*** Department of Pathology, Tokyo Dental College,
1-2-2 Masago, Mihama-ku, Chiba 261-8502, Japan*

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Abstract

This case report describes an acinic cell carcinoma found by a recurrence of a ranula in the sublingual gland. A 42-year-old male was admitted to the hospital of the Tokyo Dental College with a swelling in his right oral floor but without pain. The lesion was treated by windowing the same day under the diagnosis of a ranula, but the swelling appeared again at the same area eight months after the first operation. A resection was performed, and the specimen was sent to the clinical laboratory for pathological diagnosis. Proliferating serous cells were seen in part of the wall of an exudative mucous cyst. PAS staining was partially positive, and immunohistochemical staining for S-100 protein, lactoferrin, and amylase were also positive in cytoplasmic granules. This report concludes that the pathological diagnosis is beneficial in clarifying the reasons for the recurrence of a benign lesion.

Key words: Acinic cell carcinoma—Sublingual gland—Ranula—Mucous cyst—
Neoplasm recurrence—Local

INTRODUCTION

Acinic cell carcinomas are low-grade malignant epithelial neoplasms of the salivary gland that demonstrate some cytological differentiation toward acinar cells^{4,9,10}. This type of tumor is essentially limited to the parotid gland and constitutes about 2% of all salivary gland tumors, 3% to 5% of all parotid gland

tumors, and about 12% of all malignant salivary gland tumors⁵. Some authors have reported that acinic cell carcinomas originating from the sublingual gland are rare^{1-3,6,8,11,12}. This present case is an extremely rare case of an acinic cell carcinoma which was unexpectedly found in the excision specimen of a ranula arising in the sublingual gland.

CASE REPORT

A 42-year-old male had noticed a swelling in his right oral floor without pain in February of 2000 and was admitted to the hospital of the Tokyo Dental College on March 28th, 2001. The mass of approximately 1.5×1.0 cm was covered by normal mucosa, and he had experienced no history of trauma. The lesion was treated by windowing on the same day. Eight months after the first operation, the swelling reappeared in the same area. Under the diagnosis of a ranula, it was treated again by windowing and was sent to the clinical laboratory for pathological diagnosis.

The specimen was fixed with 10% formalin, rinsed with water, and dehydrated with graded ethanol before being embedded in paraffin. Paraffin sections about $5 \mu\text{m}$ in thickness were cut and stained with hematoxylin-eosin (H-E). Periodic acid-Schiff (PAS) staining and immunohistochemical staining were also performed. For immunohistochemical staining, primary antibodies to S-100 protein, lactoferrin, amylase, and vimentin were used. The sections were deparaffinized with xylene and washed with 100% alcohol and distilled water. Endogenous peroxidase activity was blocked by incubating the sections with 3% H_2O_2 in methanol for 30 minutes. To prevent non-specific reactions, sections were incubated with 10% serum for 30 min in a 100% humidity chamber. After washing in PBS, sections were incubated with the peroxidase-conjugated secondary antibody for 30 min in the humidity chamber. After washing in PBS three times for 5 minutes each, sections were stained with 3,3'-diaminobenzidine for 5 minutes and were finally counterstained in hematoxylin.

The pathological findings showed a mucous pool which was covered by fibrous connective tissue under the normal oral epithelium. This structure revealed an exudative mucous cyst without an epithelial lining (Fig. 1). Epithelial tumor cells with clear cytoplasm and dark, chromatin-dense nuclei were observed as sheet-like structures that contained proliferations of cells within lumen-like structures of

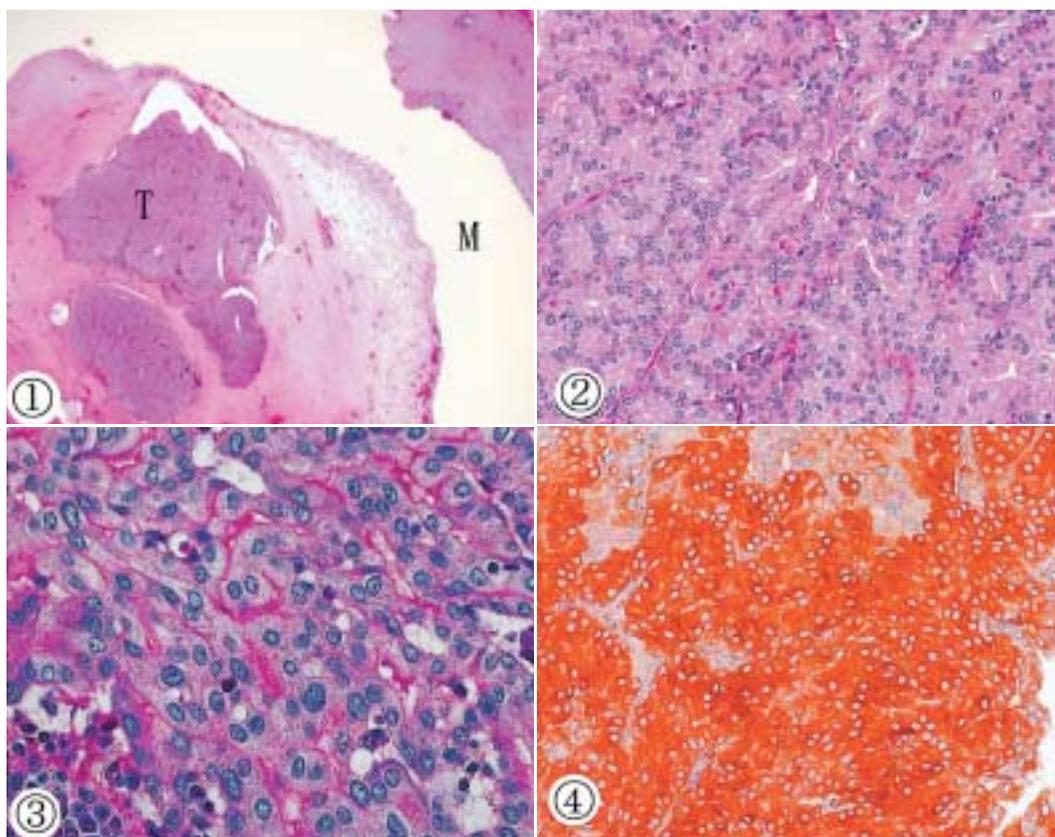
the duct in the paramucous cyst (Fig. 2). Some of the epithelial projections had thin fibrovascular cores. Intercalated duct-like and non-specific glandular cells usually predominated, although vacuolated cells were often numerous, and acinar cells could be seen. PAS-positive granules were seen in the cytoplasm (Fig. 3), and the cells were immunohistochemically reactive for lactoferrin (Fig. 4), S-100 protein, and amylase, but were negative for vimentin.

An additional resection was performed after the diagnosis of acinic cell carcinoma, and recurrence of the carcinoma has not been observed in the two years and five months of follow-up.

DISCUSSION

Acinic cell carcinoma is a low-grade malignant tumor that arises predominantly in the parotid gland⁹. This is the first case described of an acinic cell carcinoma recurring as a mucous cyst. Wahlberg *et al.*¹³ reported that acinic cell carcinomas have the best prognosis with a 10-year relative survival of 88% in over 2,000 patients with major salivary gland carcinomas. The WHO reveals that 81% of acinic cell carcinomas occurs in the parotid gland, only about 4% occur in the submandibular gland, and 13% develop in the minor salivary gland⁹. Levin *et al.*⁷ reported that the ratio of acinic cell carcinomas arising in the parotid gland was 92.5%, in submandibular glands was 1.5%, with only 0.6% originating in sublingual glands. The tumor cells are usually polygonal, resembling those of serous acini⁵. Suzuki and Henderson¹¹ reported a higher incidence of this tumor in the parotid gland, which is a pure serous gland, which seems related to the serous acinar cells of the gland, while the sublingual gland is a mixed gland, predominantly mucinous. This explains why acinic cell carcinomas arising in sublingual glands are extremely rare.

van den Akker *et al.*¹² reported that the differential diagnosis of a swelling in the floor of the mouth includes a variety of lesions;



- Fig. 1 H-E staining at loupe findings
The acinic cell carcinoma can be seen at the ranula wall; it increases in the lumen and infiltrates around the tissue. T: Tumor, M: the cavity of the mucous cyst
- Fig. 2 H-E staining at high magnification (original magnification: $\times 25$)
Epithelial tumor cells with chromatin-dense small nuclei proliferate at high density
- Fig. 3 PAS staining (original magnification: $\times 50$)
Tumor cells are partially PAS reactive in the cytoplasm.
- Fig. 4 Immunohistochemical staining for lactoferrin (original magnification: $\times 25$)

some of these lesions, such as ranula, are almost always easily diagnosed. In our case, the ranula seemed to appear and then recur in the oral floor in relationship to the acinic cell carcinoma in the duct of the sublingual gland. However, it cannot be established whether the acinic cell carcinoma invoked the mucous cyst or each lesion individually occurred, because this mucous cyst was not lined by epithelium. In conclusion, the pathological diagnosis was beneficial in clarifying the reason for the recurrence of this benign lesion.

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REFERENCES

- 1) Abrams, A.M., Cornyn, J., Scofield, H.H. and Hansen, L.S. (1965). Acinic cell adenocarcinoma of the major salivary glands; A clinicopathologic study of 77 cases. *Cancer* **19**, 1145-1162.

- 2) Baden, E. and Wallen, N.G. (1965). Acinous cell carcinoma of oral floor of mouth. *J Oral Surg* **23**, 163–168.
- 3) du Toit, D.F. and Heydenrych, J.J. (1992). Acinic cell carcinoma of the sublingual gland. *Afr Med J* **82**, 373.
- 4) Eneroth, C.M., Jakobsson, P.A. and Blanck, C. (1996). Acinic cell carcinoma of the parotid gland. *Cancer* **19**, 1761–1772.
- 5) Gorlin, R.J. and Goldman, M.H. (1970). Acinic cell carcinoma. In *Thomas's Oral Pathology*. Vol. 2, 6th ed., pp. 1024–1026, CV Mosby Company, St. Louis.
- 6) Gorlin, R.J. and Chaudhry, A. (1957). Acinic cell tumor of the major and minor salivary glands. *J Oral Surg* **15**, 304–306.
- 7) Levin, J.M., Robinson, D.W. and Lin, F. (1975). Acinic cell carcinoma; Collective review including bilateral cases. *Arch Surg* **110**, 64–68.
- 8) Rankow, R.M. and Mignogna, F. (1969). Cancer of the sublingual salivary gland. *Am J Surg* **118**, 790–795.
- 9) Seifert, G. (1991). Histological typing of salivary gland tumours. In *International Histological Classification of Tumour*. 2nd ed., World Health Organization, Springer-Verlag, Berlin.
- 10) Spiro, R.H., Huvos, A.G. and Srong, E.W. (1978). Acinic cell carcinoma of salivary origin; A clinicopathologic study of 67 cases. *Cancer* **41**, 924–935.
- 11) Suzuki, H. and Henderson, R. (1968). Acinic cell carcinoma of the sublingual gland. *Arch Otolaryngol* **87**, 146–149.
- 12) van den Akker, H.P., Busemann-Sokole, E. and Becker, A.E. (1981). Acinic cell carcinoma of the sublingual gland. Scintigraphy in pre-operative evaluation. *Int J Oral Surg* **10**, 363–366.
- 13) Wahlberg, P., Anderson, H., Biorklund, A., Moller, T. and Perfekt, R. (2002). Carcinoma of the parotid and submandibular glands—A study of survival in 2465 patients. *Oral Oncol* **38**, 706–713.

Reprint requests to:

Dr. Kenichi Matsuzaka
 Department of Clinical Pathophysiology,
 Tokyo Dental College,
 1-2-2, Masago, Mihama-ku,
 Chiba 261-8502, Japan
 Tel: +81-43-270-3581
 Fax: +81-43-270-3583
 e-mail: matsuzak@tdc.ac.jp