

Sebaceous adenoma in the retromolar region: report of a case with a review of the English literature

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Abstract. This paper reports a rare case of sebaceous adenoma on the right mandibular retromolar mucosa in a 73-year-old Japanese man, with a review of the English literature of sebaceous adenomas of salivary gland origin. A painless and yellowish polypoid lesion in the retromolar mucosa was histologically a relatively well-circumscribed neoplastic mass composed of well-differentiated sebaceous cells with cystic and duct-like structures, and was considered to be a true sebaceous gland neoplasm arising from the minor salivary gland tissue.

Key words: sebaceous adenoma; retromolar region.

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Introduction

Sebaceous adenoma of salivary gland origin is a rare, benign epithelial tumour composed of cells showing sebaceous differentiation without cellular atypia^{3,6}. This tumour shows typically well-circumscribed solid or cystic form^{3,6}. Sebaceous differentiation in the parotid and submandibular glands are relatively common^{14,16}, and sebaceous adenoma the most commonly involves the parotid glands^{9–11}. Because sebaceous glands are commonly found in the oral mucosa⁴, it is difficult to confirm a salivary origin in sebaceous adenomas from minor salivary glands^{12,13}. This article presents a rare case of sebaceous adenoma on the mandibular retromolar mucosa, with a review of the English literature of sebaceous adenomas of salivary gland origin.

Case report

In July 2001, a 73-year-old Japanese man was referred by general practitioner

of dentistry for treatment of a lesion in the right mandibular retromolar mucosa. The patient had noticed a gradual increase in mucosal swelling over the 5 months prior to his admission.

The medical history was significant for prostatic hypertrophy and bilateral emphyema. Intraoral examination showed painless and yellowish polypoid swelling in the right retromolar region of

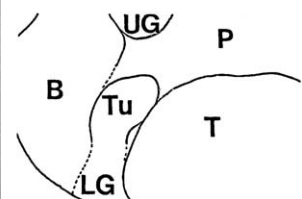


Fig. 1. Intraoral view showing a polypoid swelling (Tu) in the right retromolar region of lower gingiva (LG). (T: tongue; B: Buccal mucosa; P: palate; UG: upper gingiva).

lower gingiva (Fig. 1). There was no evidence of cervical lymphadenopathy. The clinical diagnosis was lipoma. Under local infiltration anaesthesia, the lesion was totally excised with a surrounding margin of normal tissue and microscopically examined. A 6 months follow-up revealed no recurrence.

Microscopically, the hemisected specimen showed well-circumscribed tumour mass covered with thinned stratified squamous epithelium (Fig. 2A). The tumour was composed of irregularly proliferating nests of well-differentiated sebaceous cells with cystic and duct-like structures (Fig. 2B). Small salivary ducts adjacent to the tumour showed dilation and sebaceous differentiation (Fig. 2C). Sebaceous and squamous tumour cells were negative for mucin staining. Immunohistochemically, these tumour cells showed positive reactivity for cytokeratin and epithelial membrane antigen (EMA), and were not reactive with vimentin, s-100 protein or smooth muscle actin (SMA).

Discussion

Sebaceous adenoma of salivary gland origin is a rare tumour that accounts for 0.1% of all salivary tumours and slightly less than 0.5% of salivary adenomas⁶. Nineteen cases are found in the English literature^{1,2,5,7-10,12,13,15,17-19} and present case, and are listed in Table 1. The mean age of patients with these tumours is 61.4 years old with a range of 36 to 74, and gender information is available for 15 patients: nine were men and six were women. Seven and two cases involved the parotid gland and submandibular gland, respectively, and five and three cases were located in the buccal mucosa and mandibular mucosa, respectively. Most patients are asymptomatic and note slowly growing mass^{1,2,5,7,8,10,12,13,15,17,18}.

These tumours show well-circumscribed masses varied in colour from pinkish-white to yellow, and ranged in size from 10 to 30 mm in diameter. The present case revealed as a yellowish polypoid lesion on the mandibular retromolar mucosa. Generally, treatment is total excision of tumour or extirpation including normal salivary glands^{6,10,11}. Follow-up information ranged from 1 week to 16 years is available, and no recurrence has been recorded^{1,2,5,7,8,10,12,13,15,17,18}. Our case showed no recurrence for 6 months after total tumour excision.

Microscopically, sebaceous adenoma is at least partially encapsulated, and tumour cells reveal both squamous and sebaceous differentiation in variable proportions^{3,6}. In the present case, the tumour was well-circumscribed by fibrous capsule, and most tumour cells showed sebaceous differentiation. This tumour is proven to occasionally include mucous-containing cells or oncocytic cells^{1,8,10,12,13,15,17,18}. The present

tumour didn't react with mucous staining, and oncocytic metaplasia was not recognized. The immunohistochemical findings of the present tumour indicated epithelial characteristics but not myoepithelial differentiation. Sebaceous lymphadenoma is a distinctive variant of sebaceous adenoma with well-differentiated sebaceous cells lying in a stroma of lymphocytes with or without lymphoid follicles^{3,6,10,11}. Sebaceous

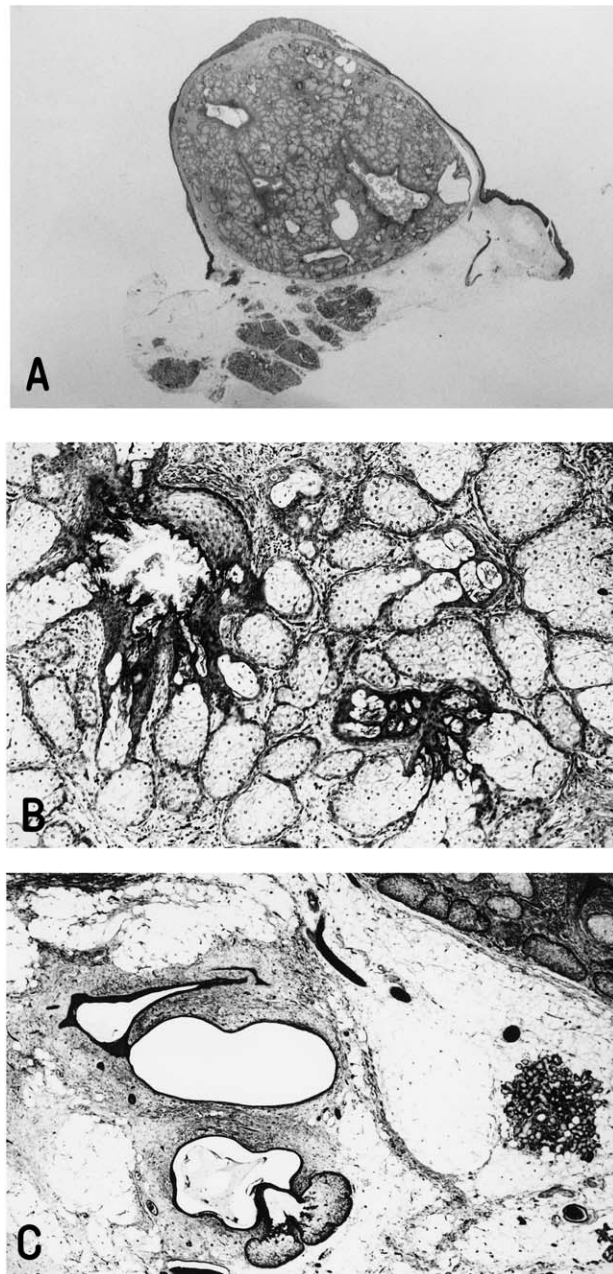


Fig. 2. Histological features. A: A relatively well-circumscribed tumour mass in elevated mucosal tissue. Underlying minor salivary glands are observed (HE, $\times 4$). B: The tumour composed of irregularly proliferating nests of well-differentiated sebaceous cells arranged in glandular configuration. Some tumour cells show squamous differentiation with marked keratinization (HE, $\times 42$). C: Some small salivary ducts adjacent to the tumour showing dilation and sebaceous differentiation (HE, $\times 14$).

Table 1. Reported cases of sebaceous adenoma of salivary gland origin

No.	Author	Reference No.	Location	Sex	Age	Clinical evidence	Follow-up
1	Foote & Frazell	9	Parotid gland	—	—	—	—
2	Albores-Saavedra & Morris	1	Submandibular gland	F	64	Pale yellow, firm, 15 mm diameter	—
3	Miller & McCrea	17	Buccal mucosa	M	65	Soft, sharply circumscribed	3 months, no recurrence
4	Epker & Henny	7	Buccal mucosa	M	45	15 mm diameter	2 years, no recurrence
5	Bab & Ulmanky	2	Parotid gland	F	57	—	Dead at 6 months of adenoid cystic carcinoma, no recurrence
6	Pieters & Seymour	19	Parotid gland	—	—	—	—
7	Lipani et al.	15	Retromolar pad	F	36	White colour, soft mass	10 years, no recurrence
8	Lipani et al.	15	Mandibular second molar	M	66	Yellowish, soft mass, 11 × 10 × 4 mm	2 years, no recurrence
9	Gnepp & Brannon	10	Parotid gland	M	42	Pink-white colour, 30 × 20 mm mass	16 years, no recurrence
10	Gnepp & Brannon	10	Parotid gland	F	71	Yellowish, granular, 14 mm diameter	1.5 years, no recurrence
11	Gnepp & Brannon	10	Submandibular gland	M	67	Bright yellow	3 years, no recurrence
12	Gnepp & Brannon	10	Parotid gland	M	74	23 mm diameter	Died with no evidence of disease at 6 years
13	Gnepp & Brannon	10	Buccal mucosa	—	—	—	—
14	Orlian et al.	18	Pterygomandibular raphe	F	74	10 mm in size, movable well circumscribed	3 months, no recurrence
15	Ferguson et al.	8	Buccal mucosa	F	65	Firm nodular lump, yellowish, painless	1.5 years, no recurrence
16	Koutlas & Yaholnitsky	13	Mandibular retromolar mucosa	M	64	Yellow, soft, fluctuant, well circumscribed	1 week, no recurrence
17	Dent et al.	5	Buccal mucosa	M	62	Yellow, firm	2 years, no recurrence
18	Iezzi et al.	12	Buccal mucosa	M	60	Elevated grey-yellowish lesion	3 years, no recurrence
19	Present case	—	Retromolar region	M	73	Polypoid swelling	6 months, no recurrence

carcinoma is a malignant counterpart of sebaceous adenoma and is composed predominantly of sebaceous cells with different degrees of nuclear atypia, pleomorphism and invasiveness¹⁰. In the present tumour, lymphoid stroma and cytological malignancy were not observed, and, therefore, these tumours were distinguished.

Sebaceous glands are prominent cutaneous adnexal components formed closely associated with hair follicles or independently¹⁴. The presence of intraoral sebaceous glands is commonly found and is accounted for on the basis of the embryology of the oral cavity⁷. About 10% of normal parotid glands and 6% of normal submandibular glands have been reported to show sebaceous differentiation¹⁴. Sebaceous glands are found in approximately 80% of normal oral mucosa and are called Fordyce's granules⁶. The present tumour was adjacent to the minor salivary glands consistent with the retromolar glands and was delineated from the neighbouring normal minor salivary glands by distinct capsular fibrous connective tissue. These histological features suggested that this tumour was a true sebaceous gland neoplasm arising from the minor salivary gland tissue.

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