

## Sebaceous adenoma of the submandibular gland : a case report

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**Abstract:** Sebaceous adenoma of the salivary gland is a rare tumor comprising 0.1% of all salivary gland neoplasms and less than 0.5% of salivary adenomas. Histologically, sebaceous adenomas are benign neoplasms consisting of sebaceous cells arranged in nests forming acinar and duct-like structures. Oncocytic metaplasia may also occur in some areas. We describe a case of sebaceous adenoma in the submandibular gland. Under a presumptive diagnosis of sialadenitis/sialolithiasis, the patient was administered multiple courses of antibiotics; however, these were not effective. Excisional biopsy resulted in a diagnosis of sebaceous adenoma. A 1-year follow-up showed no recurrence. (J Oral Sci 51, 641-644, 2009)

Keywords: sebaceous adenoma; submandibular gland; oncocytic metaplasia.

### Introduction

Sebaceous adenoma is a rare benign tumor that accounts for 0.1% of all salivary gland neoplasms and slightly less than 0.5% of all salivary adenomas (1). This tumor is composed of cells showing sebaceous differentiation without cellular atypia. The following criteria are established for diagnosis of sebaceous adenoma: a well-

circumscribed tumor composed of differentiated sebaceous lobules with irregularity of size and shape; lobules containing varying proportions of both mature sebaceous cells and small germinal cells that may be arranged in an irregular pattern; and no dilatation of excretory ducts on serial section (2,3). The most common site for this tumor is the parotid gland (4). This article presents a case of sebaceous adenoma arising in the submandibular gland of a 60-year-old man.

### Case Report

In January 2007, a 60-year-old apparently healthy man was admitted to the Gandhi Surgery Clinic, Tehran, Iran with a 1-year history of swelling in the right submandibular region. Intraoral examination showed a painless mass on floor of the mouth that was covered by normal mucosa. A clinical diagnosis of sialadenitis/sialolithiasis was made. Multiple courses of antibiotics were prescribed, including cloxacillin 250 mg, three times a day, but they were not effective. The patient therefore underwent an excisional biopsy via an extra-oral approach under general anesthesia.

Biopsy tissue was sent to the Department of Oral and Maxillofacial Pathology, Mashhad Faculty of Dentistry for definitive diagnosis. The gross specimen was composed of a yellowish-gray mass which was soft and elastic and measured 44 × 31 × 18 mm. Histologic examination of biopsy tissue showed benign neoplastic proliferation of epithelial cells in the salivary gland surrounded by a fibrous capsule. The proliferating epithelial cells showed cystic and solid patterns, and were contained in a dense fibrous stroma (Figs. 1 and 2). Tumor cells showed sebaceous differentiation (Fig. 3). Papillary cystic areas with oncocytic metaplasia and eosinophilic substances were also noted

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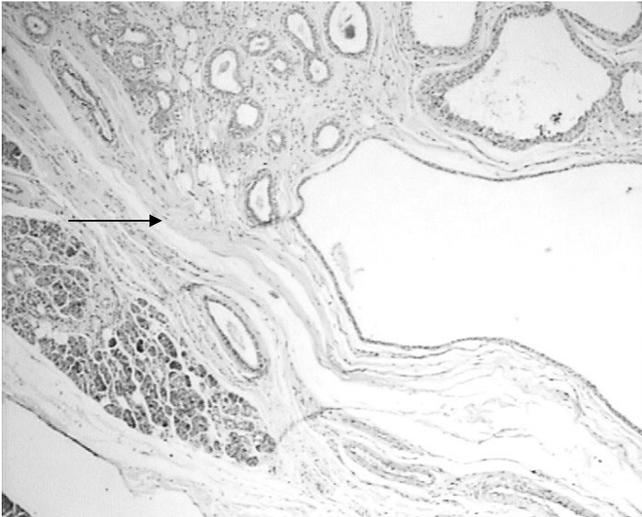


Fig. 1 Fibrous capsule around the neoplasm (arrow). (H-E staining, original magnification  $\times 100$ )

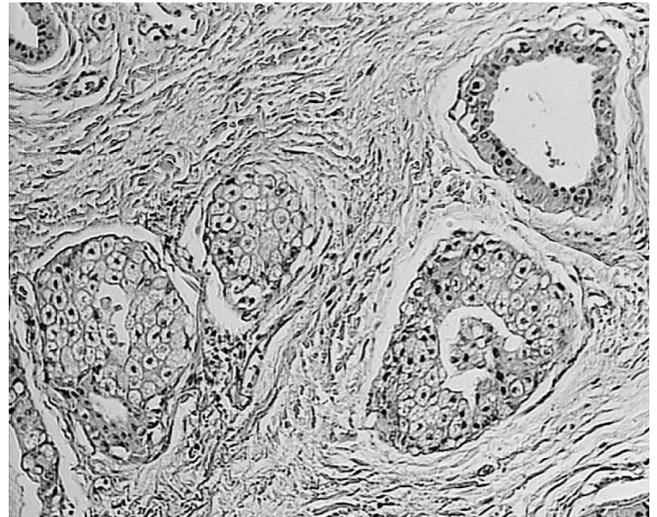


Fig. 3 Sebaceous differentiation in the submandibular gland. (H-E staining, original magnification  $\times 100$ )

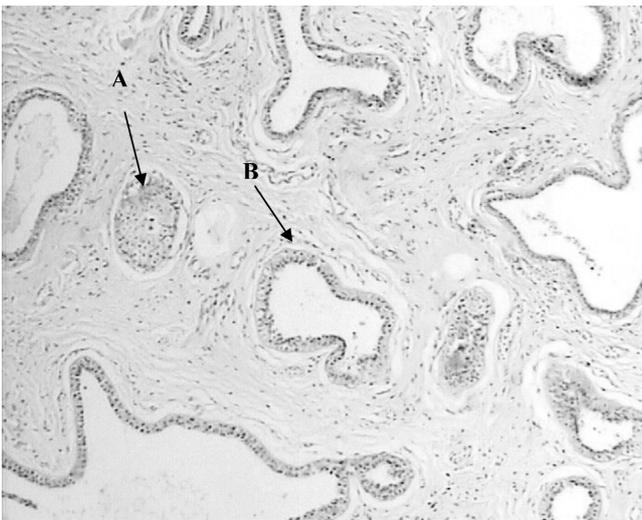


Fig. 2 Benign neoplastic proliferation of salivary gland epithelial cells in a dense fibrous stroma. A: solid and B: cystic area. (H-E staining, original magnification  $\times 40$ )

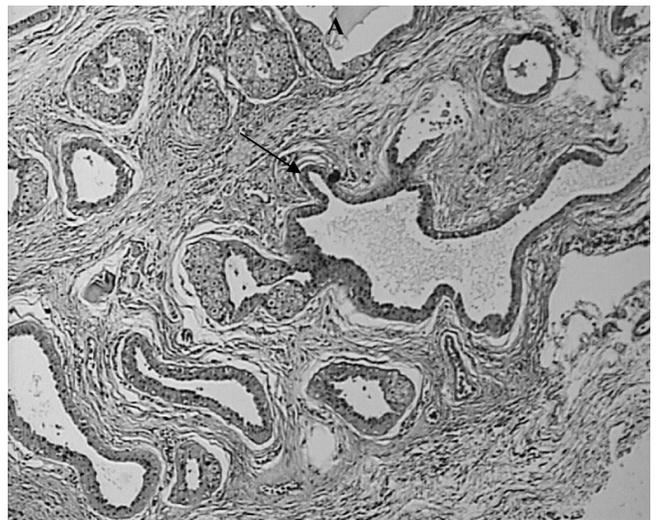


Fig. 4 Papillary cystic areas with oncocytic metaplasia (arrow) and eosinophilic substances (A) (H-E staining, original magnification  $\times 40$ )

(Fig. 4). In some areas, sebaceous differentiation coexisted with oncocytic metaplasia (Fig. 5). A diagnosis of sebaceous adenoma was made. A 1-year follow-up after tumor excision showed no recurrence.

### Discussion

Sebaceous adenoma of the submandibular gland is a very rare benign neoplasm (3). Although salivary glands commonly contain sebaceous elements, it is rare for these to undergo neoplasia (5).

Sebaceous adenomas of salivary gland origin may be cystic or solid; they are usually well circumscribed and sharply demarcated from surrounding tissue (1,2). The mean age of patients is 61.4 years and men are more commonly affected than women (1,4). These lesions are rare in adult patients and have been reported only twice in children (6).

These tumors are composed of sebaceous cell nests forming acinar and duct-like structures in a fibrous stroma. They often contain areas of squamous differentiation with

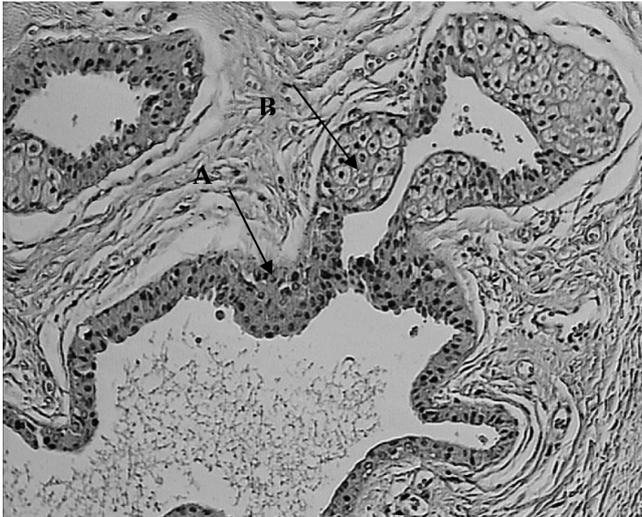


Fig. 5 Both oncocytic metaplasia (A) and sebaceous differentiation (B) are seen in this figure. (H-E staining, original magnification  $\times 100$ )

minimal atypia, minimal pleomorphism, and no tendency to invade local structures. They are immunoreactive to cytokeratin and epithelial membrane antigen (EMA) (1,4).

In some cases, oncocytic metaplasia can be observed. The exact origin of oncocytes and the cause of oncocytic change are unknown, although this process may represent cellular transdifferentiation in an aging organ or adaptation to cell damage (3). In the present case oncocytic metaplasia was predominant. Oncocytic metaplasia in submandibular sebaceous adenoma was also reported by Riberio (3).

The most detailed information on these sebaceous neoplasms is provided by Gnepp and Branon (7). They reported 21 cases of salivary gland sebaceous neoplasms, only one of which was sebaceous adenoma of the submandibular gland. Riberio (3) reported a case of submandibular sebaceous adenoma and he believed that it was the third described in this location. Liu (8) later reported another case in the submandibular gland, so the present case is probably the fifth case of submandibular sebaceous adenoma in the English-language literature.

Sebaceous adenomas are composed of sebaceous lobules branching from a centrally located wide sebaceous duct, with appearance of both mature sebaceous cells and undifferentiated basaloid germinative cells which may be arranged in an irregular pattern (2,4). These lesions must be differentiated from sebaceous hyperplasia, a condition in which a single enlarged sebaceous gland consists of numerous well-differentiated lobules associated with a dilated duct and one or more layers of peripheral germinative cells (9).

In addition, sebaceous adenoma should be included in

the differential diagnosis of sebaceous lymphadenoma, sebaceous lymphadenocarcinoma, and sebaceous carcinoma. In contrast to sebaceous adenoma, sebaceous lymphadenoma consists of well-differentiated sebaceous cells lying in a stroma of lymphocytes with or without follicles (4). Sebaceous lymphadenocarcinoma is an extremely rare malignant tumor that presents with carcinomatous transformation of sebaceous lymphadenoma (1). Sebaceous carcinoma differs from sebaceous adenoma by the presence of cellular pleomorphism, cytologic atypia, and invasiveness (1,4). In the present case, lymphoid stroma and cytological malignancy were not observed, leading to a diagnosis of sebaceous adenoma. In addition, some salivary gland tumors including mixed tumors, carcinomas ex mixed tumors, mucoepidermoid carcinomas, and oncocytomas may have foci of sebaceous differentiation (1).

The etiology of sebaceous differentiation in the salivary gland is unknown. In the major salivary glands, the phenomenon of ductal transformation into sebaceous glandular elements is thought to account for the presence of sebaceous gland-like structures (5). It is possible that the same factors that activate sebaceous glands in the skin during puberty also activate those in the salivary glands (10). Treatment consists of complete surgical excision. These tumors will not recur if they are adequately excised (1,5).

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