

Sebaceous adenoma of the submandibular gland

A case report

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The authors report a case of sebaceous adenoma of the submandibular gland and discuss the clinical and histologic characteristics of this rare neoplasm. (*Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1996;82:200-3)

Sebaceous adenomas of salivary gland origin are extremely rare tumors, comprising 0.1% of all salivary gland neoplasms and less than 0.5% of salivary adenomas.¹ Cutaneous sebaceous adenomas present as slow-growing painless firm masses; they appear predominantly during the sixth and seventh decades of life² as solitary lesions of the face or scalp.³ Rulon and Helwig⁴, in a large study of cutaneous sebaceous neoplasms, reported their occurrence mainly on the nose, cheek, or scalp with a slow rate of tumor growth in 80% of cases.

Histologically, sebaceous adenomas are benign neoplasms basically consisting of sebaceous cells arranged in nests with minimal atypia or pleomorphism and no tendency to invade local structures.⁵ They are typically circumscribed, and may be solid or cystic.⁶ Those arising in salivary gland tissue preferentially affect the parotid gland.⁷

The treatment consists of surgery, and no recurrence is observed if they are properly excised.⁵

CASE REPORT

A 44-year-old white man came to the Stomatology Department of A.C. Camargo Hospital in August 1993, with a large growth that he had had for 4 years in the floor of the mouth. The patient reported that 3 years earlier an attempt had been made to remove the lesion. Because the lesion persisted, he sought medical care at another hospital at which time a needle aspiration biopsy was interpreted as a salivary gland malignant neoplasm, and an incisional biopsy was diagnosed as hemangioma. The patient then decided to look for another institution and came to our hospital. It was not possible for us to evaluate the earlier histologic material.

Physical examination revealed a patient in good health



Fig. 1. Clinical aspect of lesion located on right side of floor of mouth appears as movable mass covered by normal mucosa.

with a slight facial asymmetry because of a large growth in the right submandibular region. Intraoral examination revealed a movable firm painless submucosal mass, covered with normal mucosa and measuring 5 cm in diameter on the right side of the floor of the mouth (Fig. 1). Tongue movements were preserved.

Tentative clinical diagnoses of sialolithiasis, benign neoplasm of mesenchymal origin, and salivary gland neoplasm were made. An occlusal radiograph of the floor of the mouth was taken, excluding the presence of salivary calculus. Salivary gland scintigraphy disclosed functional involvement of the submandibular gland.

With the patient under a general anesthesia, the lesion was removed completely via an intraoral approach. The gross specimen consisted of a firm, encapsulated, well-delimited, nodular mass, measuring 4.5 × 4.0 × 3.2 cm (Fig. 2). The surface had a fasciculated and lobulated appearance and was yellowish-white in color.

Histologic examination revealed sebaceous cells arranged in nests showing marked variation in size (Fig. 3). A few duct-like structures were set in a fibrous stroma; these cells exhibited focal atypia and pleomorphism (Fig. 4). Areas with oncocytic metaplasia (Fig. 4), histiocytes, and foreign body giant cells were also noted. The mucicar-

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mine stain demonstrated focal positivity in some cells of the ductal component but not in the sebaceous cells; this led to a diagnosis of sebaceous adenoma.

After a follow-up of almost 2 years, the patient exhibits no sign of recurrence.

DISCUSSION

Sebaceous elements are commonly found in the parotid⁸⁻¹⁰ and submandibular glands⁸ and are also observed in normal salivary tissue adjacent to benign or malignant tumors^{7, 8, 10} and inflammatory lesions.⁷ They were first described by Hamperl⁷ in the submandibular gland of four patients and in the parotid gland of one patient. In a study of 133 parotid tumors, Meza-Chavez¹⁰ found sebaceous differentiation in 33 (24.8%) glands in the adjacent normal parotid gland. Since these reports, several cases of sebaceous gland-containing salivary gland tumors and intrasalivary gland sebaceous gland adenomas of the major salivary glands have been documented.¹¹⁻¹²

The origin of sebaceous differentiation in salivary glands is unknown. Rawson and Horn¹³ suggested that, because of the ectodermal origin of parotid glands, it would not be surprising that the ducts of this gland could differentiate into structures resembling sebaceous elements. However, this theory does not explain the occurrence of sebaceous elements in the submandibular gland, which is of endodermal origin.¹⁴ Sebaceous differentiation may result from a metaplastic process as a result of ductal obstruction from a tumor or inflammatory process, it may be congenital in origin, or it may naturally develop later in life.⁵ Most authors agree with the last theory. The rare occurrence of sebaceous components in salivary glands of children before puberty⁸ argues against the congenital presence of sebaceous differentiation. The prevalence of sebaceous components increases markedly after puberty, and a frequency of 10% to 20% is reported in young and middle-aged adults. Possibly the same factors that activate sebaceous glands in the skin during puberty do the same in the salivary glands.

Because sebaceous differentiation in salivary glands is a common finding, sebaceous neoplasms could be expected to occur in salivary glands. However, the incidence of primary salivary gland sebaceous tumors is very low compared with the normal frequency of sebaceous gland tissue within the salivary glands. This fact suggests that the pathogenetic mechanisms of salivary gland sebaceous differentiation and salivary gland sebaceous tumors are quite different.⁷

The criteria for establishing the diagnosis of sebaceous adenoma are as follows³: (1) a sharply circumscribed lesion with an organoid pattern; (2) irregular-

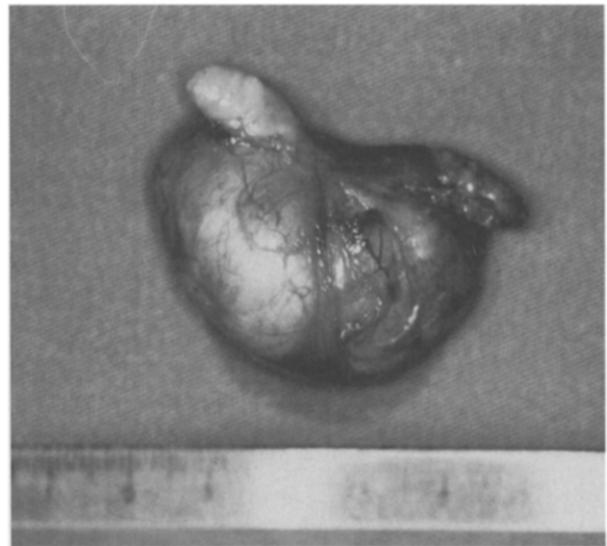


Fig. 2. Macroscopic view of surgical specimen shows well-demarcated nodular structure.

ity of size and shape of the sebaceous lobules; (3) appearance of both mature sebaceous cells and small germinal cells that may be arranged in an irregular pattern; and (4) lack of a dilated excretory duct or a common excretory duct after serial sections.

The microscopic description of our case satisfies the above criteria. Rulon and Helwig⁴ have established these criteria for cutaneous sebaceous adenomas and also use them to differentiate this neoplasm from sebaceous hyperplasia. In addition, Lever and Schaumberg-Lever² note that in sebaceous hyperplasia the lobules are grouped around a central wide duct that empties on the surface.

In a review of the literature, Gnepp⁷ found reports of 18 cases of sebaceous adenoma with an age distribution that ranged from 22 to 90 years. Most of the tumors occurred in men. The principal location was the parotid gland followed by the buccal mucosa. The present case is probably the third described in the floor of the mouth in the region of the submandibular gland. According to Daley,¹⁵ there have been eight lesions reported as intraoral sebaceous adenomas in the English-language literature, but of the seven non-salivary lesions, only four meet the microscopic criteria for a diagnosis of sebaceous adenoma.

In the literature, tumors varied in size from 0.4 to 3.0 cm in diameter; they were commonly encapsulated or sharply circumscribed.⁷ The lesion of the case reported here measured 4.5 cm, probably the largest lesion described.

Many tumors are microcystic and are composed predominantly of sebaceous glands that vary markedly in size and tortuosity. The neoplasms are em-

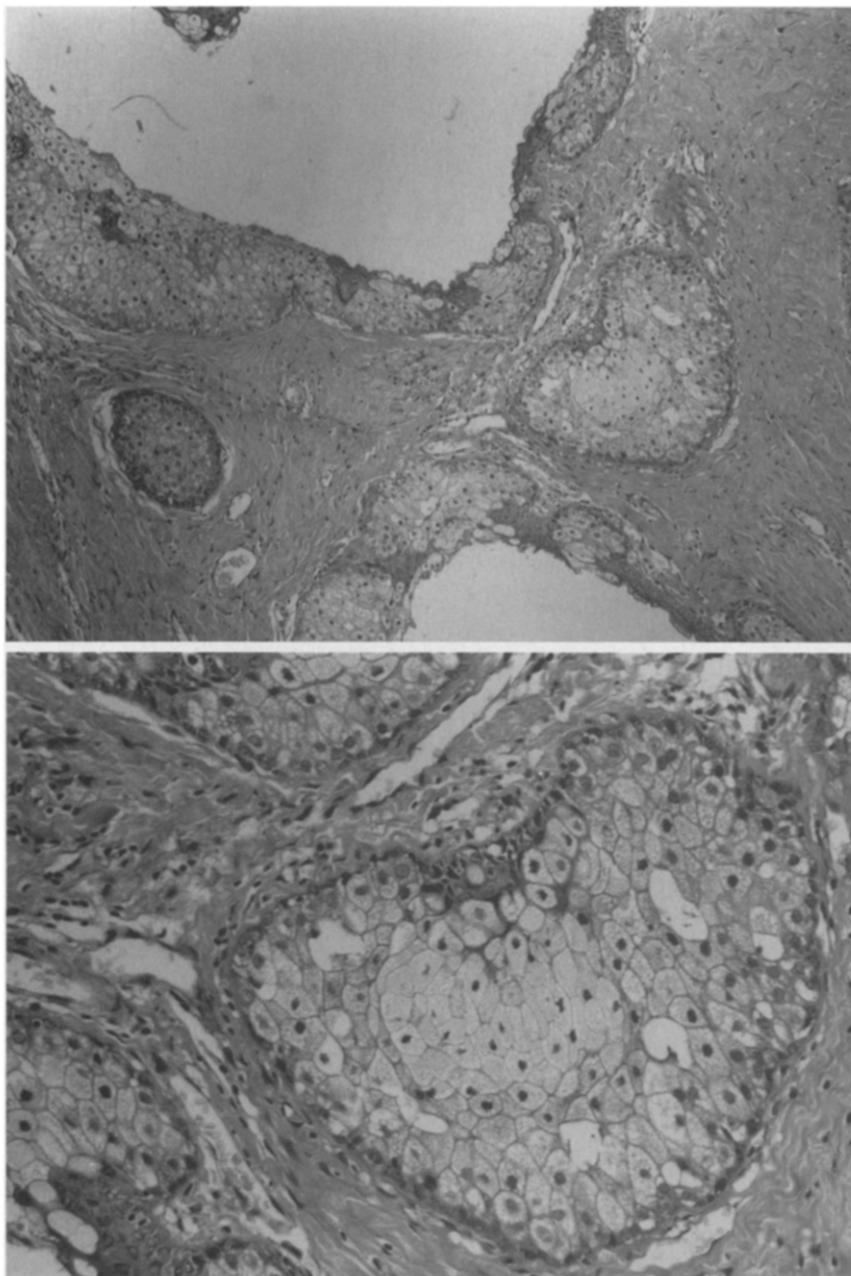


Fig. 3. **A**, Sebaceous adenoma from submandibular gland demonstrates individual irregular sebaceous glands in a fibrous stroma. (Hematoxylin-eosin stain; original magnification $\times 18$.) **B**, high power view of same tumor demonstrates nests of sebaceous cells with minimal atypia. (Hematoxylin-eosin stain; original magnification $\times 200$.)

bedded in a fibrotic stroma and in some cases exhibit oncocytic metaplasia.⁷ The precise origin of oncocytes and the cause of the oncocytic change are not known. Possibly these oncocytes represent the trans-differentiation of the cells of an aging organ or an adaptation to cell damage. Musebeck¹⁶ suggests that the transformation could be induced by factors causing a disturbance of respiration and oxidizing phosphory-

lation compensated by an increase of energy supplying factors.

Several tumors demonstrate focal positivity for mucicarmine staining in ductal components but not in sebaceous cells. The finding of sebaceous tumors with a mucin-producing component and of a mucin-producing carcinoma emphasizes the pluripotential nature of the salivary gland ductal elements. This

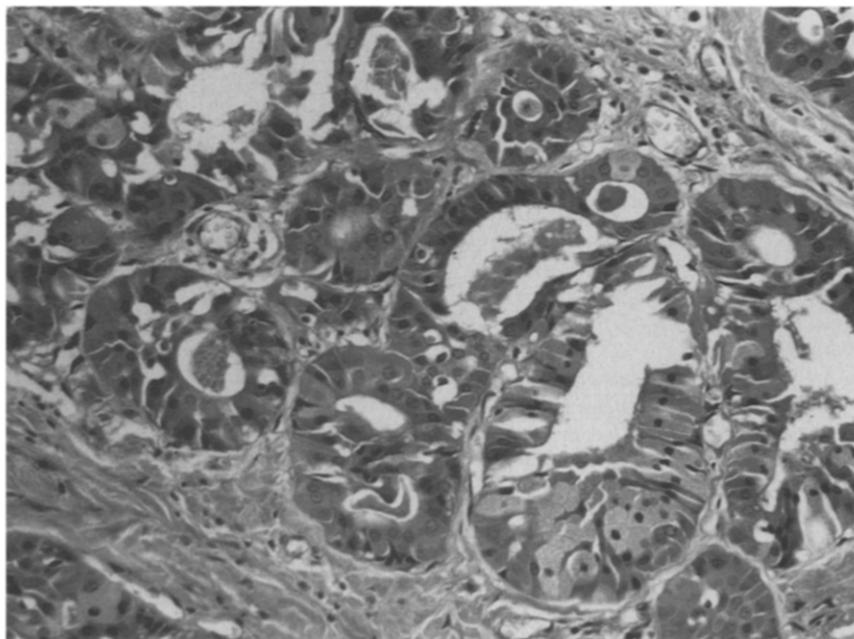


Fig. 4. Sebaceous adenoma exhibits marked oncocyctic metaplasia.

must be carefully evaluated to determine if the sebaceous elements are benign or malignant.⁷

Surgical removal with a normal tissue margin is the most effective therapy and was applied in the present case with good results. These lesions infrequently recur as shown in the literature.^{5,7}

Recently, several authors^{2,5,7,17} have emphasized the association of sebaceous neoplasms of the skin with an increased risk of developing visceral, especially colonic, carcinomas. The literature also describes a case of sebaceous adenoma associated with a prostatic carcinoma;⁵ In our case there was no association with any other tumor, but the patient is still being followed up despite the low risk of local recurrence or of the occurrence of another kind of neoplasm.

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