

Sebaceous adenoma in the submandibular gland

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Sebaceous adenoma is a rather rare developmental anomaly that has been described mainly in the dermatologic literature. The significance of cutaneous sebaceous adenoma as indicative of internal malignancy in Muir-Torre syndrome is known. Here we report a case of a patient with sebaceous adenoma of the submandibular gland, which differs from previous intrasalivary gland cases associated with the parotid gland.

CASE REPORT

An otherwise healthy 38-year-old woman sought treatment for a right-sided progressively enlarging submandibular mass of 3 years' duration. No significant personal or family history of malignancy was noted.

The mass was initially measured as 2 × 1 cm. Three years later, on palpation, the mass was over the right submandibular area and was measured as 4 × 3 cm. It was soft and movable without tenderness or local skin change. No abnormality was found in the upper aerodigestive tract. A computed tomography neck scan demonstrated a soft tissue mass of about 35 mm in diameter in the right submandibular gland, with small areas of fat density (Fig 1A).

The patient underwent excision of the mass, which was a gray-tan, multilobular cystic tumor (3.5 × 3 cm), and removal included submandibular gland tissue. Microscopically, the tumor was composed of cystic spaces lined with stratified squamous epithelium in which lobules of sebaceous gland were identified (Fig 1B). Solid clusters of sebaceous glands in inflammatory salivary parenchyma were found. There was no evidence of malignancy. The diagnosis of sebaceous adenoma was made.

DISCUSSION

Sebaceous adenoma is an uncommon benign organoid tumor composed of proliferating, incompletely differentiated sebaceous glands. It occurs principally at sites in which the concentration of sebaceous glands is especially dense, such as

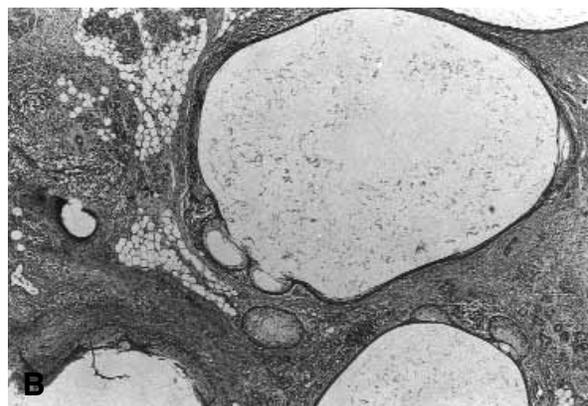
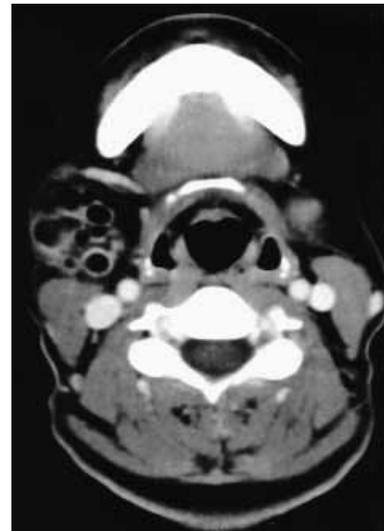


Fig 1. A, Computed tomography scan shows a 35-mm soft tissue mass in the right submandibular gland, with small areas of fatty density. **B,** Photomicrograph shows sebaceous adenoma of the submandibular gland. Multilobular cystic spaces are lined with stratified squamous epithelium in which lobules of sebaceous gland were identified (hematoxylin and eosin stain, original magnification × 40).

the face and scalp. These tumors vary from a small nodule to an ill-defined plaque and are usually tan-yellow. Most lesions do not exceed 1 cm in diameter, with a common range of 0.1 to 9 cm.¹ They usually arise in older individuals and in men slightly more often than in women. Pain or tenderness is not uncommon, whereas ulceration is rarely found. Clinically, cutaneous sebaceous adenoma is often misdiagnosed as basal cell carcinoma.

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In an extensive search of the literature on tumors of the sebaceous glands, cutaneous sebaceous adenoma was mentioned more commonly and was defined as a component of Muir-Torre syndrome,² which is an autosomal dominant genodermatosis characterized by at least a single sebaceous gland tumor and an internal malignancy such as colorectal or genitourinary carcinoma. However, sebaceous gland tissue is also found in salivary glands, especially the parotid gland.³ Rawson and Horn⁴ first described 2 tumors of the parotid gland with a sebaceous gland component.

Biopsy for histologic examination is usually necessary to reach a definite diagnosis. Clinically, cutaneous sebaceous adenoma should be differentially diagnosed from sebaceous hyperplasia, sebaceous epithelioma, sebaceous carcinoma, histiocytoma, and xanthoma. Computed tomography scanning may be performed for larger intrasalivary gland sebaceous adenomas, which could present with a clinical picture of dermoid tumor, teratoma, lipoma, or liposarcoma due to its solid, cystic, or keratoacanthoma-like morphologic patterns. Treatment is adequate excision. Compared with colorectal or genitourinary carcinoma in the general population, the clinical course of the same cancers in Muir-Torre's syndrome is relatively benign due to the low-grade malignancy.⁵

Of submandibular neoplasms, pleomorphic adenoma ranks first, as in the parotid gland. Sebaceous adenomas are found in normal sebaceous glands that originate from the blind ends of intralobular ducts in otherwise normal salivary glands. Patients with such benign salivary gland neoplasms usually

present with an asymptomatic, slowly enlarging mass. Pain related to a salivary gland neoplasm in and of itself does not confer a higher risk of malignancy, because pain may be caused by associated infection, hemorrhage, or cystic enlargement of the gland itself. Computed tomography scanning and magnetic resonance imaging can provide a better understanding of the extent of the tumor, whether intraglandular or extraglandular, before surgery.

We report here what we believe to be a unique, intrasubmandibular gland sebaceous adenoma arising from the salivary gland duct. No associated internal malignancy was found, compared with cutaneous sebaceous adenoma of Muir-Torre's syndrome.

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