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Schwannoma arising from the sublingual gland

Hiroyuki Okada, DDS, PhD[a,*], Shigeo Tanaka, DDS, PhD[b], Hideto Tajima, DDS, PhD[b], Yoshiaki Akimoto, DDS, PhD[b], Takashi Kaneda, DDS, PhD[c], Hirotsugu Yamamoto, DDS, PhD[a]

[a]*Department of Oral Pathology, Nihon University School of Dentistry at Matsudo, Chiba 2718587, Japan*
[b]*Department of Oral Surgery, Nihon University School of Dentistry at Matsudo, Chiba 2718587, Japan*
[c]*Department of Radiology, Nihon University School of Dentistry at Matsudo, Chiba 2718587, Japan*

Abstract Sublingual gland tumors, especially mesenchymal tumors, are extremely rare. We describe the first reported case of schwannoma arising from the sublingual gland with details of the histopathologic and immunohistochemical features. A 70-year-old woman developed a painless swelling on the floor of the mouth. The excised material was sublingual gland tissue with an ovoid, grayish-yellow solid tumorous mass at the cut surface. The tumor was composed of proliferated spindle-shaped tumor cells exhibiting palisading patterns. In the center of the tumor, a small salivary gland component was recognized. Immunohistochemically, the tumor cells were strongly positive for S-100 protein but negative for neurofilament protein. The Ki-67 labeling index was 4.58. The clear presence of a remnant sublingual gland lobule in the present tumor provided convincing evidence that it was a schwannoma arising from the sublingual gland and thus the first of its type to be reported.

Keywords: Schwannoma; Sublingual gland; Salivary gland tumor; Mesenchymal tumor; Ki-67

1. Introduction

Salivary gland tumors are uncommon, representing 3% to 10% of all neoplasms of the head and neck region, and 70% of them involve the parotid gland [1,2]. Sublingual gland tumors are very rare, accounting for only 0.3% to 1% of all epithelial salivary gland tumors, and most are malignant, with the most common histologic types being adenoid cystic carcinoma and mucoepidermoid carcinoma [1,3]. Mesenchymal tumors, excluding hematopoietic neoplasms, account for only 2 to 5% of salivary gland tumors [4]. More than 85% of soft tissue tumors arise in the parotid, and more than 10% involve the submandibular gland [4]. Therefore, mesenchymal tumors of the sublingual gland are extremely rare, and to our knowledge, there have been few reports of mesenchymal tumors with microscopic proof of their sublingual gland origin [5,6].

Here we describe the first reported case of schwannoma arising from the sublingual gland with details of histopathologic and immunohistochemical features.

2. Case report

A 70-year-old woman was referred to our hospital because of a painless swelling on the floor of the mouth. She had not noticed the swelling, but her private dentist pointed it out and referred her to our hospital. On her first visit, laboratory tests showed no abnormality. Anamnesis revealed that she had been receiving medication for hypertension for 3 years.

Oral examination revealed a well-defined, elastic, hard mass 40-mm in diameter on the left side of the floor of the mouth. Magnetic resonance imaging revealed a well-defined, partly heterogeneous, ovoid mass showing low intensity on T1- (Fig. 1) and low-to-intermediate intensity on T2-weighted images adjacent to and behind the ablated and compressed left sublingual gland. On computed tomography, the mass showed slightly lower intensity in

* Corresponding author. Department of Oral Pathology, Nihon University School of Dentistry at Matsudo, 2-870-1, Sakaecho-Nishi, Matsudo, Chiba 2718587, Japan. Tel.: +81 47 360 9335; fax: +81 47 360 9335.
E-mail address: okada.hiroyuki@nihon-u.ac.jp (H. Okada).

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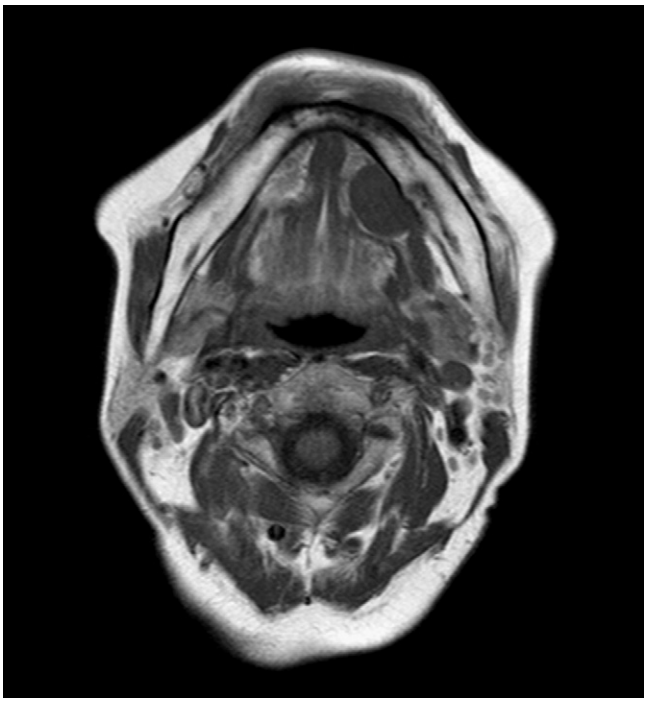

Fig. 1. T1-weighted axial magnetic resonance imaging shows a well-defined ovoid mass in contact with the left submandibular gland.

comparison with muscle. The clinical and imaging diagnosis was a sublingual gland tumor, possibly malignant.

Fine-needle aspiration cytology was performed, but the specimen was nondiagnostic. Therefore, under local anesthesia with 2% lidocaine, a biopsy sample was taken from the mass, which yielded a diagnosis of "lipomatous lesion of the sublingual gland." Excision of the lesion including the sublingual gland was performed under general anesthesia. During the operation, no involvement of nerves, such as facial nerves, with the tumor was evident.

3. Pathologic findings

3.1. Gross findings

The irregular oval-shaped excised specimen consisted of solid yellowish-brown sublingual gland tissue measuring 39 × 33 × 17 mm and included an ovoid, grayish-yellow solid tumorous mass measuring 23 × 19 mm at the cut surface (Fig. 2).

3.2. Microscopic finings

The tumor was composed of proliferated spindle-shaped cells with elongated or ovoid nuclei on a fibrous or myxomatous background, which had formed an oval mass lesion completely encapsulated by thin fibrous tissue (Fig. 3). In the cell-rich area, Antoni A–type palisading patterns were occasionally recognized (Fig. 4). There was a mild degree of nuclear pleomorphism in some areas, but no

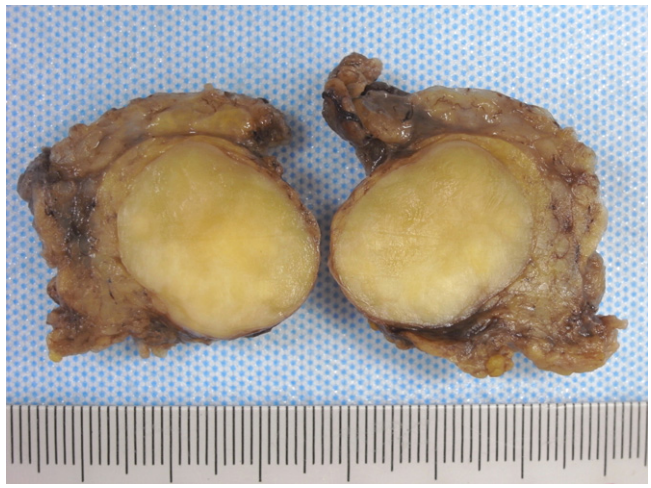

Fig. 2. Macroscopic view of the cut surface of the specimen.

mitosis was identified. At the center of the tumor, a small salivary gland component with fatty tissue, peripheral nerves, and muscular vessels without a surrounding capsule, presenting the remnants of the sublingual gland, was obvious (Fig. 5). In the stroma, mild lymphocyte infiltration and capillary proliferation were partly evident, and lymphoid tissue was observed at the tumor periphery (Fig. 3). Outside the capsule, the sublingual gland was identified (Fig. 3). Duct dilatation, basophilic or hyalinized material in the lumen, oncocytic ductal metaplasia, mild lymphocyte infiltration, and degeneration, atrophy, and necrosis of the acinus were occasionally found. The final diagnosis was schwannoma arising from the sublingual gland.

3.3. Immunohistochemical findings

The tumor cells were strongly positive for S-100 protein (S-100, polyclonal; Dako, Glostrup, Denmark) (Fig. 6),

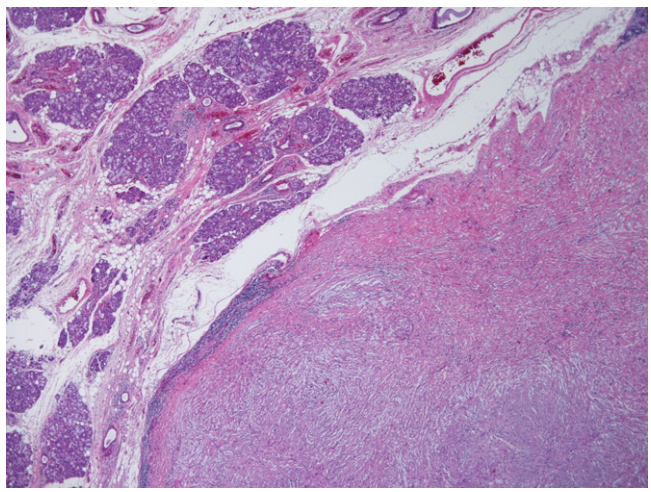

Fig. 3. Low-power view of the schwannoma, showing a tumor clearly demarcated from the adjacent sublingual gland by a thin fibrous capsule (×20).