Pleomorphic Adenoma of the Sublingual Salivary Gland: An Unusual Diagnostic Challenge

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Introduction

Salivary gland tumors are rare, representing 3.5–10% of all head and neck tumors. Neoplasms of the sublingual salivary glands constitute 0.3–5.2% of all epithelial salivary gland tumors and approximately 1.5% of all carcinomas of major salivary glands [1, 2]. The great majority, approximately 80–90% of the tumors that arise from the sublingual gland, are malignant [3, 4], whereas benign tumors such as pleomorphic adenoma are rarely reported [5–7]. In this case we describe an asymptomatic pleomorphic adenoma of the sublingual salivary gland.

Case Report

An 80-year-old patient was referred to the Department of Oral and Maxillofacial Surgery, School of Dentistry, Aristotle University of Thessaloniki, Greece, complaining of a painless swelling located in the floor of the mouth, causing minor discomfort. The duration of the swelling was 4 months and it was progressively enlarging. His medical history was free of any local or systemic diseases.
The intraoral examination and palpation revealed a firm, solid-elastic, movable, tumorous lesion, measuring 2 × 3 cm covered by normal mucosa located in the anterior part of the floor of the mouth beside the excretory duct of the left submandibular gland (fig. 1). Computed-tomography (CT) images showed a distinct radiolucent, homogeneous lesion extending anteriorly in the area where the left sublingual gland (sublingual sulcus) normally lies (fig. 2). Under local anesthesia, a complete surgical enucleation of the lobular tumor with an additional zone of adjacent normal tissue was performed intraorally. Histological examination showed a large number of epithelial/myoepithelial-type neoplastic cells with low mitotic activity forming sheets, solid islands (arrowhead) and duct-like structures (arrow). Small amount of myxoid stroma containing spindle-shaped and plasmacytoid cells (arrowhead), among neoplastic cell structures (arrow). The tumor (small arrow) was almost completely separated from adjacent mucosa and minor salivary glands (arrowhead) by a fibrous capsule (large arrow).

**Discussion**

Sublingual salivary gland tumors are rare compared with other major glands with a reported ratio of 1 case of sublingual tumor to 100 parotid tumors [1] or less [7]. The great majority of these tumors are malignant and few benign cases of pleomorphic adenomas, myoepitheliomas and oncocytomas have been described [7–9]. Sublingual benign tumors present as asymptomatic swelling of the floor of the mouth under the tongue, causing discomfort and difficulty in dental prosthesis retention [10], and may be incidentally discovered by a dentist, as happened in our case.

The differential diagnosis of sublingual gland tumors may include other salivary gland tumors of submandibular/minor salivary glands, or even other non-salivary gland tumors like lymphomas, non-neoplastic entities of salivary glands such as ranulas, retention cysts, sialoli-
thiasis, sialadenitis, and cystic lesions of the floor of the mouth such as dermoid cysts.

Early total surgical resection of the sublingual gland and its neoplastic mass in normal margins for benign tumors is the treatment of choice to avoid recurrences. The excision may possibly be accompanied by selective neck dissection and radiotherapy for malignancies. The histologic examination of the lesions is necessary not only to establish the diagnosis but also for the better management of the surrounding tissues.

Depending on the tumor size and location, the total resection may become quite complex especially for malignancies due to the close proximity of the sublingual gland to the inner cortex of the mandible, the submandibular salivary gland and its duct, the lingual vessels and nerve, and the hypoglossal nerve [8, 10]. In our case, clinical and CT imaging features indicated a close-to-surface, circumscribed, easily accessible tumor, and thus an intraoral total resection of the lesion was performed.

Conclusions

This case showed that sublingual salivary gland tumors, including pleomorphic adenoma, should be included in the differential diagnosis of masses located in the floor of the mouth with minimal or no symptoms.

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