Solitary fibrous tumor of the floor of the mouth: Case report and review of the literature

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Abstract
Solitary fibrous tumor is an uncommon spindle cell neoplasm that is believed to be of mesenchymal origin. Rarely does it originate in the oral cavity, and only 1 case of this lesion involving the floor of the mouth has been previously reported. We describe a new case of solitary fibrous tumor arising from the soft tissues of the floor of the mouth.

Introduction
Solitary fibrous tumor is a rare spindle cell neoplasm that is believed to be of mesenchymal origin. The first report of this tumor was published in 1931 by Klemperer and Rabin, who described a lesion that had arisen from the pleura. Since then, extrapleural lesions have been reported in a wide variety of head and neck sites, including the orbit, nose, paranasal sinuses, thyroid gland, and salivary gland. Within the oral cavity, the most common sites are the buccal mucosa, tongue, and lip. Until now, only 1 case of solitary fibrous tumor involving the floor of the oral cavity has been documented in the literature. In this article, we present a new case of a solitary fibrous tumor that arose in the soft tissues of the floor of the mouth. We also review the literature on solitary fibrous tumors of the oral cavity, and we discuss the clinical presentation, diagnosis, and histopathologic and immunohistochemical characteristics of this rare lesion.

Case report
A 35-year-old woman presented to the Department of Otolaryngology–Head and Neck Surgery at South Infirmary–Victoria Hospital with a 6-month history of a slowly enlarging mass in the floor of the oral cavity. Two months earlier, she had begun to experience dysarthria and difficulty masticating. She had no history of similar swelling, oral cavity surgery, trauma to the area, or smoking.

Clinical examination revealed the presence of a firm, nontender, 3 × 4-cm submucosal mass on the right side of the floor of the mouth. Egress of saliva from Wharton’s ducts was normal, and findings on a regional examination were unremarkable. Computed tomography (CT) showed a well-circumscribed heterogenous lesion cephalad to the mylohyoid muscle (figure 1).

With the patient under general anesthesia, excision biopsy was performed. Gross inspection revealed that the specimen was a well-circumscribed ovoid lesion with a fibrous pseudocapsule. The cut surface had a homogenous gray-white color without any recognizable gross pattern. The specimen was sent for histopathologic and immunohistochemical evaluation. Microscopically, the tumor was made up of uniform spindle cells with separating bands of collagen (figure 2, A). In addition, marked variations of cellularity were noted with alternating collagenous, cellular, and focal myxoid areas. These appearances were consistent with a so-called “patternless pattern.” In some areas, there was vague nuclear palisading. The cells had an indistinct cytoplasm, oval nuclei, and inconspicuous nucleoli. Mitotic figures were rare. Hemangiopericytoma-like areas were noted, where staghorn-type vessels demonstrated perivascular hyalinization (figure 2, B).

Immunohistochemical analysis showed strong spindle cell cytoplasmic positivity for CD34, strong cytoplasmic staining for CD99, and strong focal nuclear staining for Bcl-2 (figure 3). Reticulin staining failed to detect a continuous intercellular network staining pattern. The tumor was immunonegative for S-100 protein, cytokeratin, and desmin. This analysis confirmed the diagnosis of a solitary fibrous tumor.

At the 5-month follow-up, the patient remained well and showed no signs of recurrence.

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Discussion
Based on initial reports of solitary fibrous tumors, some authors postulated that these tumors had a mesothelial origin because they had been noted to arise only from mesothelium-lined surfaces. Since then, however, these tumors have also been noted in extrapleural and extraserosal sites, a finding that supports the theory that they have a mesenchymal cell origin.

To the best of our knowledge, the literature contains only 40 cases of a solitary fibrous tumor involving the oral cavity, including our case. Of these, the tumor site in 37 patients is known. The most common site is the buccal mucosa (n = 21 [56.8%]), followed by the tongue (n = 6 [16.2%]) and the lip (n = 3 [8.1%]). Other documented sites of involvement include the soft and hard palates, the mental regions in the oral cavity, and the gingival, mandibular, and sublingual glands. The nonsalivary soft tissue of the floor of the mouth has not previously been described as a site of origin of a solitary fibrous tumor. Of the 37 cases in which the patient’s sex was recorded, 23 of these tumors (62.2%) occurred in women. Patients’ ages ranged from 20 to 83 years (mean: 52.2).

Patients with a solitary fibrous tumor of the oral cavity typically present with a slowly growing nontender mass. Most of these patients are asymptomatic, but when symptoms do occur, they vary according to the site and size of the lesion. Dysarthria and abnormal mastication, which occurred in our patient, have been noted by others.

Because solitary fibrous tumor is so rare, the clinical diagnosis is difficult. Lesions that arise from the floor of the mouth—such as dermoid and epidermoid cysts, salivary gland tumors, fibromas, myofibromas, neurofibromas, ranulas, hemangiopericytomas, and desmoid tumors—should all be considered in the differential diagnosis. Confirmation of the diagnosis requires a full histopathologic and immunohistochemical analysis.

The classic histologic appearance of a solitary fibrous tumor is characterized by a variable, patternless architecture of spindle cells with areas of hypocellularity and adjacent hypercellular regions. These spindle cells are intermingled with abundant collagen fibers that form tenuous fascicles. The tumor tends to be well vascularized and contain hemangiopericytoma-like areas. Immunohistochemical staining for CD34 and Bcl-2 is consistently positive. CD99 positivity is common but more variable. Immunonegative staining for S-100 protein, cytokeratin, desmin, actin, and myogenin is a fairly constant feature of solitary fibrous tumor. The microscopic and immunohistochemical profile in the case presented herein was consistent with a solitary fibrous tumor.

When distinguishing solitary fibrous tumor from other tumors, the most difficult pathologic distinction to make...
is between it and hemangiopericytoma because their morphologic and immunohistochemical profiles are similar.\textsuperscript{6,9,12} Hemangiopericytomas are more cellular and homogenous lesions that contain less collagen than do solitary fibrous tumors. CD34 is reported to be more focal and less constant in hemangiopericytomas. Other lesions to be included in the histologic differential diagnosis are solitary myofibroma, fibrous histiocytoma, desmoid tumor, and spindle cell lipoma.\textsuperscript{13}

In conclusion, soft-tissue fibromas of the floor of the mouth are rare. Diagnosis requires a full histopathologic and immunohistochemical evaluation. The clinician should include solitary fibrous tumor in the differential diagnosis of tumors that arise from the soft tissues of the floor of the mouth.

References