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What is your diagnosis?

A submucosal tumor at the base of the tongue

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1. Case report

A 70-year-old female visited the ENT clinic after a mass was found at the tongue base during a routine health examination. There was no notable medical history. The patient did not suffer from dysphagia or odynophagia. A laryngoscopic examination reveal a tumor, 1 cm sized, well demarcated, at the base of the tongue without invasion or ulceration of the overlying normal mucosa (Fig. 1A). Based on our suspicion of a benign tumor arising from a minor salivary gland, excision was performed via transoral laser microsurgery. An incisional biopsy was not planned preoperatively for avoiding the spillage of tumor content. During the operation, the mass was palpated firmly, and well circumscribed without invasion of the tongue muscle. The surgical site was remained to be healed secondarily and the operative procedure was completed without any complications. The histopathologic exam revealed a definite diagnosis (Fig. 1B–D). A patient did not complain of dysphagia after surgery, and the operative site had fully recovered with normal mucosa after 3 weeks.

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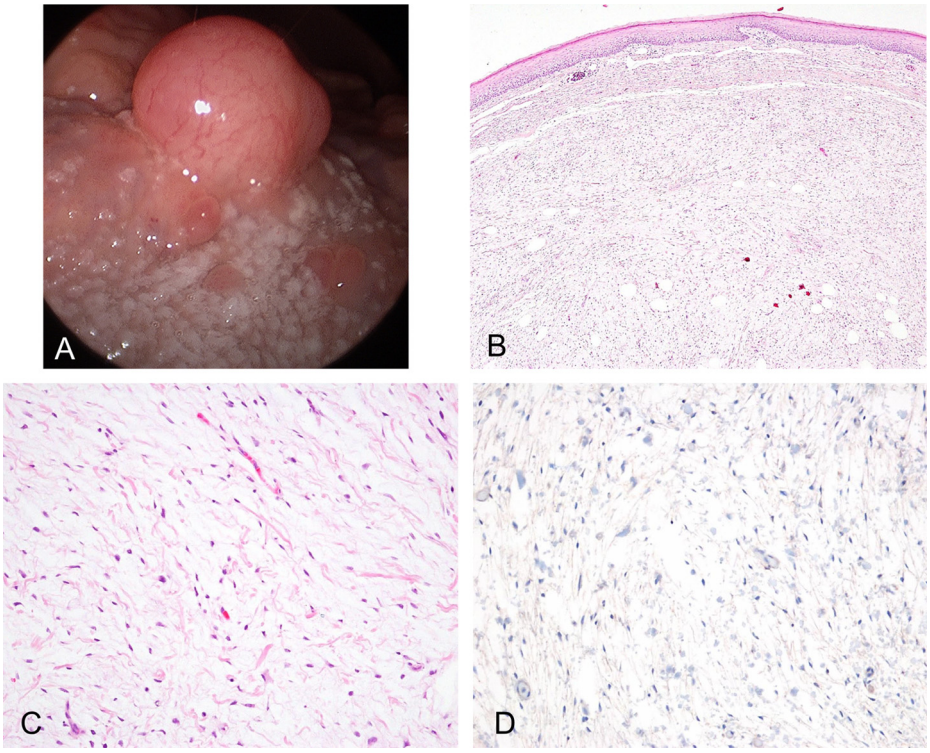

Fig. 1. A. Laryngoscopy showing submucosal tumor at the tongue base. B. Microscopic examination of the tumor (hematoxylin-eosin stain, ×40). Note the well-circumscribed mass covered by stratified squamous epithelium. C. High-power magnification (hematoxylin-eosin, ×200). Note the fibro-myxoid stroma with bipolar, fusiform or stellate shaped fibroblasts. D. Immunohistochemical stain (×200). The lesion is negative for S-100.

What is your diagnosis?

2. Answer

The histopathologic evaluation concluded on focal mucinosis. Microscopically, loose fibro-myxoid stroma with bipolar, fusiform or stellate fibroblasts was observed. Fibroblasts contained a small amount of cytoplasm and delicate fibrillate processes extending into the surrounding matrix. Immunohistochemistry did not reveal the presence of S-100 protein, so the presence of a neurogenic tumor such as a neurofibroma or Schwannoma was ruled out.

After focal mucinosis involving the oral cavity was first described in 1974 by Tomich [1], only a few cases of OFM affecting gingival tissue or the hard palate and tongue have been reported [1–3]. The pathologic term for this disease entity is site based: oral focal mucinosis (OFM). Focal mucinosis arising from the oropharynx has not previously been reported. OFM is an uncommon, benign disease with uncertain etiology, which is considered analogous to cutaneous focal mucinosis [1]. OFM is histopathologically identified by a well circumscribed lesion of myxoid stroma filled with acid glycosaminoglycan composed of hyaluronic acid, secreted by fibroblasts and surrounded by fibrous connective tissue [3]. The common approach to treating OFM is complete excision [1,4]. There has been no case of recurrence after complete excision.

One of the differential diagnoses for OFM is a nerve sheath myxoma. A nerve sheath myxoma has a more discrete border with bulky stromal cells, and consists of myxoid areas separated by fibrous connective tissue. [5]. OFM is a well-circumscribed tumor that contains myxoid materials composed of hyaluronic acid and ovoid and stellate fibroblasts in an alcianophilic stroma [1]. Immunohistochemical staining for S-100 protein, expressed by nerve sheath myxomas, can thus be helpful in differentiating these tumors from OFM [5].

Focal mucinosis was identified at the tongue base, oropharyngeal focal mucinosis could be another pathologic entity, and considered in the differential diagnosis of oropharyngeal submucosal tumors.

Disclosure of interest

The authors declare that they have no competing interest.

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