Glossal vascular leiomyoma

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CLINICAL PHOTOGRAPH

Glossal vascular leiomyoma

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Vascular leiomyoma is an uncommon benign tumor, not premalignant, composed of smooth muscle cell and vascular endothelium that occurs usually in the uterus or the esophagus. Oral vascular leiomyoma is infrequently found. In a review of 562 cases of vascular leiomyoma (uterine and visceral tumors were excluded) by Hachisuga and colleagues, only 15 cases or 2.7% were seen in the oral cavity.[1] When only the oral region is considered, vascular leiomyoma of the tongue accounted for 9.2% of cases.[2] In other words, glossal vascular leiomyoma is extremely rare with the pathogenesis remaining obscure. In this article, a vascular leiomyoma of the tongue in a 41-year-old male is described.

A 41-year-old man was referred by his physician to the Department of Otolaryngology at Taiwan Adventist Hospital for treatment of a mass under his tongue, which had been present for more than 10 years although remaining painless. The patient reported biting the mass while eating few weeks ago; thereafter, the mass had slightly increased in size and had become painful.

Physical examination revealed a firm, mildly tender mass measuring 2 × 2 cm in diameter on the left ventral surface of the tongue (Fig 1). Overlying oral mucosa was intact, with the rest of the head and neck unremarkable following a thorough systemic examination.

With the impression being one of a benign, localized tongue tumor, the patient underwent an excisional biopsy under general anesthesia. The mass was excised completely along with part of the normal, neighboring tongue muscle. The wound was closed primarily.

Grossly, the tumor was well circumscribed with a whorled, white-gray cut surface. On histologic examination, the tumor was composed of interlacing bands of smooth muscle, apparently arising from a thick vascular wall, without mitosis (Fig 2). Masson's trichrome stain was positive. These features were consistent with vascular leiomyoma. The postoperative period was uneventful, and no evidence of recurrence was present 1 year after surgery.

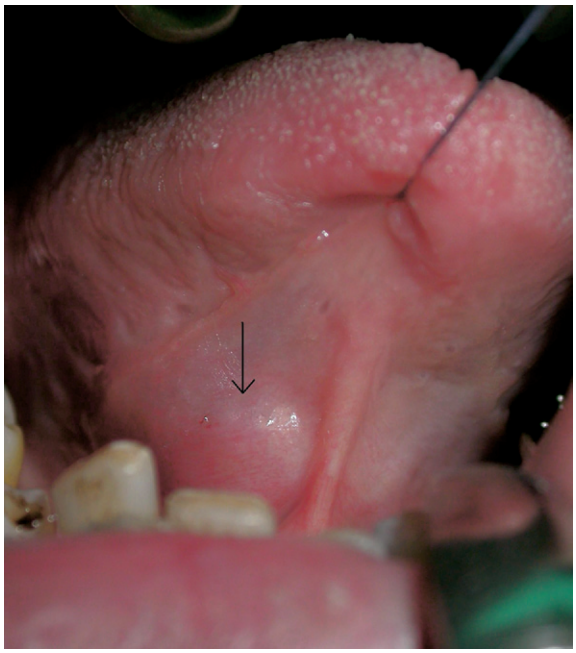

Figure 1 Preoperative view of the tumor on the left ventral surface of patient's tongue, with overlying oral mucosa intact.

DISCUSSION

In 95% of cases, leiomyomas most commonly arise from the smooth muscle of the female genital tract. Vascular

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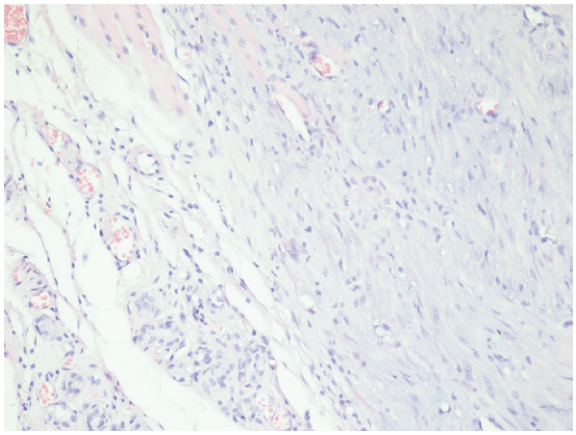

Figure 2 The histology section shows interlacing bands of uniform, elongated, spindle-shaped cells with "cigar-shaped" nuclei. Nuclear atypia was absent and mitosis was rare. Areas of striated muscle cell and glandular tissue were also noted (H&E; original magnification 400×).

leiomyomas are an uncommon histologic variant of leiomyoma and rarely occur in the oral cavity. Vascular leiomyomas arising in the tongue are even more uncommon. A review of 76,412 oral biopsies by Brooks and colleagues revealed 12 cases of oral vascular leiomyoma, representing 0.016% of all specimens.[2] Among these 12 cases of oral vascular leiomyoma, only 1 arose in the tongue. In a further analysis of 109 cases of oral vascular leiomyoma, Brooks and colleagues found 12 lesions of their own in combination with 97 additional cases identified from an English-language literature review; the tongue ranked third in the frequent site of occurrence.

Glossal vascular leiomyomas are believed to arise from the tunica media of blood vessels within the tongue. Other origins, like the circumvallate papillae, ductus lingualis, and heterotopic embryonal muscle tissue have also been proposed.[3] Unlike their uterine counterparts, estrogen plays no definitive role in the pathogenesis of glossal vascular leiomyomas.[4]

Although most oral vascular leiomyomas are described as painless and manifested a slow growth rate, those arising in the tongue more likely will be symptomatic. Glossal vascular leiomyomas may be associated with pain,[2,4] periodic numbness,[4] and even congenital airway obstruction.[3]

Differential diagnoses of glossal vascular leiomyomas include reactive and neoplastic lesions of mesenchymal origin, salivary gland lesions, and soft tissue cysts. Nevertheless, surgical excision with histologic study is the only way to make the definitive diagnosis of glossal vascular leiomyomas. Histologic study of glossal vascular leiomyomas typically reveals interlacing bands of uniform, elongated, spindle-shaped cells, whose nuclei usually have blunt ends (cigar-shaped nuclei). Nuclear atypia is absent; mitosis is rare. In questionable cases, immunohistochemical study is needed and is considered a reliable method for definitive diagnosis.[5]

Treatment of the oral vascular leiomyoma is surgical excision, and the cure rate is high. However, glossal vascular leiomyomas are located submucosally or embedded deep within tongue musculature and may not be well encapsulated, making the possibility of recurrence more likely. Complete surgical excision with adequate margin is thus advocated.[3]

CONCLUSION

Glossal vascular leiomyomas are rare smooth muscle tumors with a limited degree of morbidity. However, surgical excision, with careful histologic assessment, is an imperative for both treatment and a definitive diagnosis. Glossal vascular leiomyomas more likely are symptomatic, with a higher rate of recurrence, compared with their oral counterparts.

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