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Angioleiomyoma in Right Lingual Gingiva — A Case Report

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Abstract

Leiomyomas are benign soft tissue neoplasms that arise from smooth muscle. There are three distinct types of leiomyomas: piloleiomyomas, angioleiomyomas, and genital leiomyomas, which reflect the most origin of the smooth muscle tumor and correspond to the histologic or anatomic site. Angioleiomyomas, as we present here, originate from smooth muscle within the walls of arteries and veins. Although some cases are reported in the gastrointestinal tract, larynx, and brain, oral angioleiomyomas are infrequently found. Malignant transformation probably does not occur, and therefore, they do not affect mortality directly. However, careful histologic examination is still necessary to distinguish these benign lesions from their malignant counterparts due to different prognosis. Here, we present a case of angioleiomyoma found attached to the lower gingival. Clinical and histological presentation of angioleiomyomas with its treatment will be discussed in this article.

Key words: Leiomyoma, Angioleiomyoma, Malignant transformation.

Introduction

Among head and neck tumors, leiomyomas are rare and benign. Smooth muscle neoplasms are classified on the basis of histology into three subtypes: solid leiomyomas, angioleiomyomas (vascular leiomyomas), and epithelioid leiomyomas (bizarre leiomyomas or leiomyoblastomas). Most leiomyomas are found in the genital tract, while a few (0.06%) are found in the oral cavity. Generally, angioleiomyomas (representing about 64.0–66.2% of all oral leiomyomas) usually occur in the lower extremities and behave as single, firm, slow growing, and seldom painful lesions.

Case report

A 38-year-old woman was referred for treatment of a suspected sarcoma over her right lower posterior lingual gingiva. The patient complained of pain of unknown origin and about 3-weeks duration in her right lower jaw. At first, she thought it was a toothache and then sought dental treatment. However, the symptoms persisted after traditional endodontic treatment of her

right lower second molar. She was transferred to another hospital for further evaluation, where incisional biopsy was arranged. The pathologic report indicated sarcoma. One week later, she came to our department with this painful mass. She denied any history of traumatic injury, smoking, or betel nut chewing.

On physical examination, a single bulging mass (2 x 3 cm in size) over the right lower posterior lingual gingiva, and mucosa of normal appearance, were noted (Fig. 1). The lesion was firm but not tender. There was no pus discharge from the lesion, no associated palpable lymph nodes, and no bony destruction on panoramic X-ray film (Fig. 2).

Computed-tomographic scan showed a 17 x 21-mm, ill-circumscribed, nonhomogeneous solid mass in the right submandibular triangle attached to lingual surface of the mandible. Also, this study showed no definite cervical lymphadenopathy (Fig. 3). A whole body bone scan showed a focus of tracer accumulation in the right mandible.

Incisional biopsy was then performed in our out-patient department and revealed the lesion was a spindle cell tumor.

On the basis of both these radiological features and the pathological report results, surgical intervention was recommended. Under general anesthesia, a curvilinear incision was made at the base of the most superior portion of tumor lesion. The gingival mucosa was carefully elevated and dissected from this solid mass. Full exposure revealed one 2-cm firm, encapsulated tumor, which in some areas was densely attached to the lateral aspect of periosteum. This soft tissue mass was totally excised along with the adjoining areas of the mandible as one specimen. The wound was kept open and packed with iodoform gauze. After one week, the patient was trained to clean the wound by irrigating it with a syringe, following removal of the gauze. She received regular follow-up each month, and slow re-epithelization of the cavity was noted after six months. There is no local recurrence or distant metastasis to date.

Histological examination demonstrated a well-circumscribed and encapsulated nodule that consisted of spindle cells surrounding a patent lumen. A prominent feature of the nodule was abundance of vascular spaces of various sizes (Fig. 4). Some areas (both thick-walled and venous vessels) were punctuated with smooth muscle cells. Smooth muscle cells of the large vessels were arranged in orderly circumferential fashion, while other small channels were interspersed with spindle cells (Fig. 5). As in angioleiomyomas that occur elsewhere, this oral cavity tumor lacked external elastic laminae. Special immunohistochemical staining with HHF-35 demonstrated focal positive results over these spindle cells (Fig. 6). Photomicrograph of MIB-1 special staining showed less than 15% of cells had mitotic activity, which suggested the lesion was benign (Fig. 7). These histological features are compatible with a diagnosis of angioleiomyoma.

Discussion

Angioleiomyomas are more infrequent in the oral cavity than the extremities. The most frequent site of occurrence is the lip, especially the lower lip, [1] followed by the buccal mucosa and tongue. A comprehensive literature search identified less than 20 cases of gingival angioleiomyomas [2]. The etiology of vascular tumor is uncertain, but it may be related to trauma, infection, hormones, and arteriovenous malformations. Breakneck, in 1996, suggested that angioleiomyoma could arise from cells associated

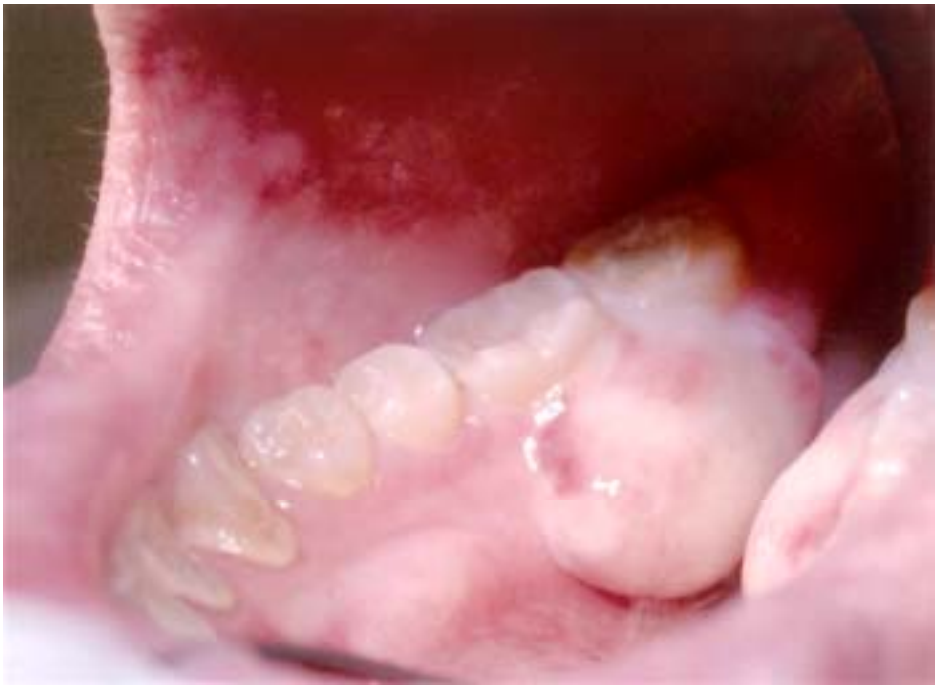

Fig. 1. Bulging out mass 2×3 cm in size over right lower posterior lingual gingival with normal mucosal appearance.

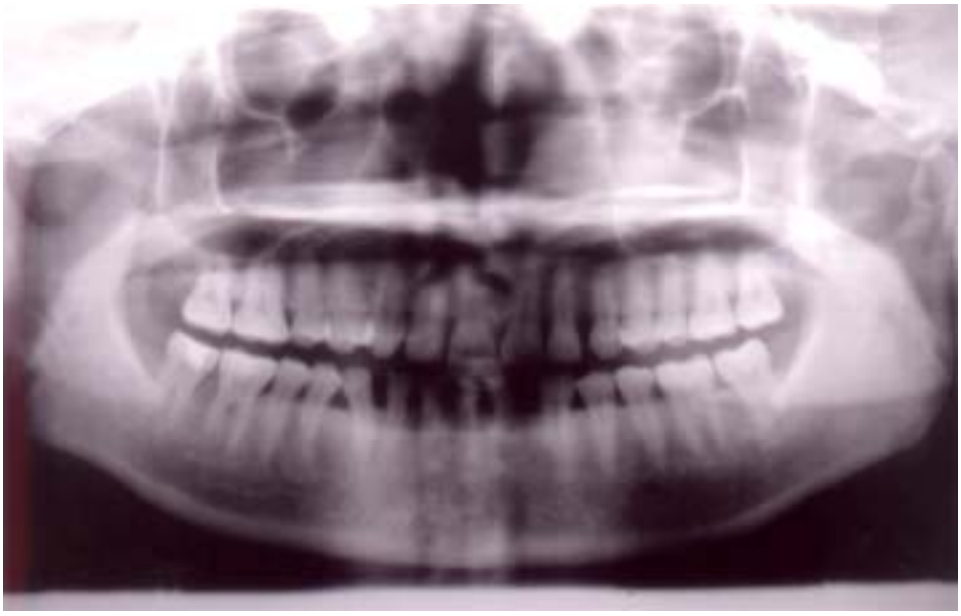

Fig. 2. There is no obvious bony destruction on panoramic film over right side posterior mandible.

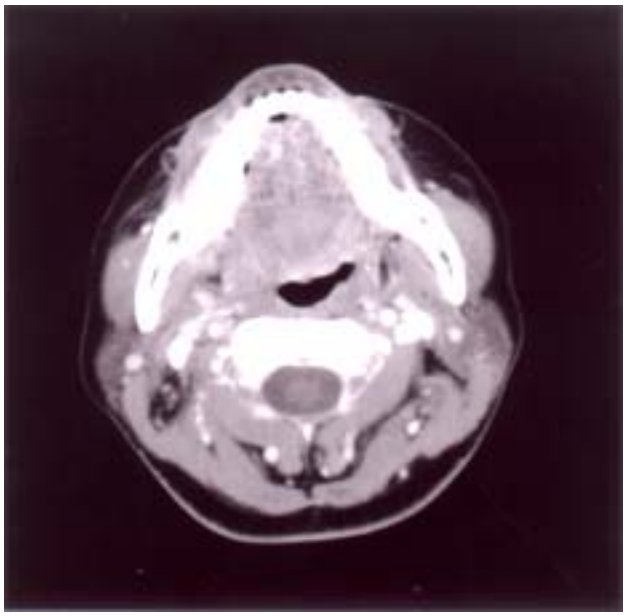

Fig. 3. Computed-tomographic scan examination (horizontal section) showed 17×21 mm, ill-circumscribed, nonhomogeneous solid mass in the right submandibular triangle attached to lingual surface of mandible. There was no definite cervical lymphadenopathy in this study.

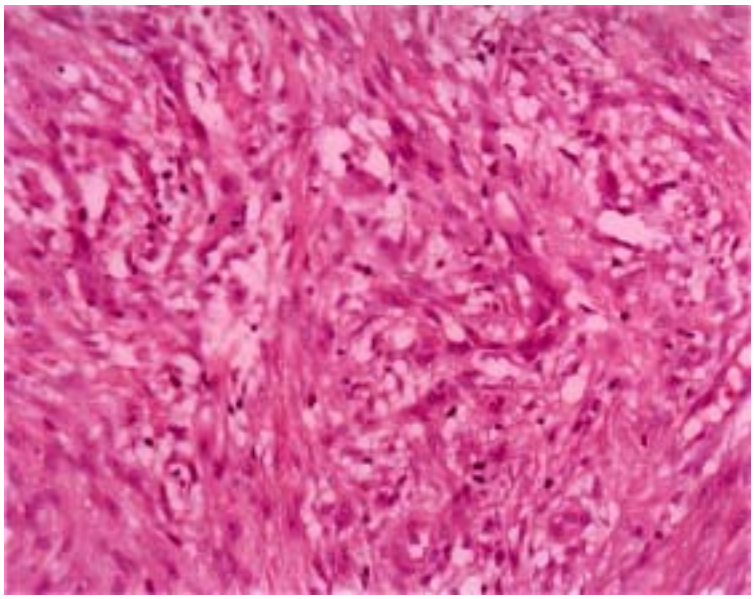

Fig. 4. Low power field of histological examination shows proliferation of spindle-shape cells interspersed among multiple vascular spaces of varied size in submucosal connective tissue of the gingival. (hematoxylin-eosin stain; original magnification×100)

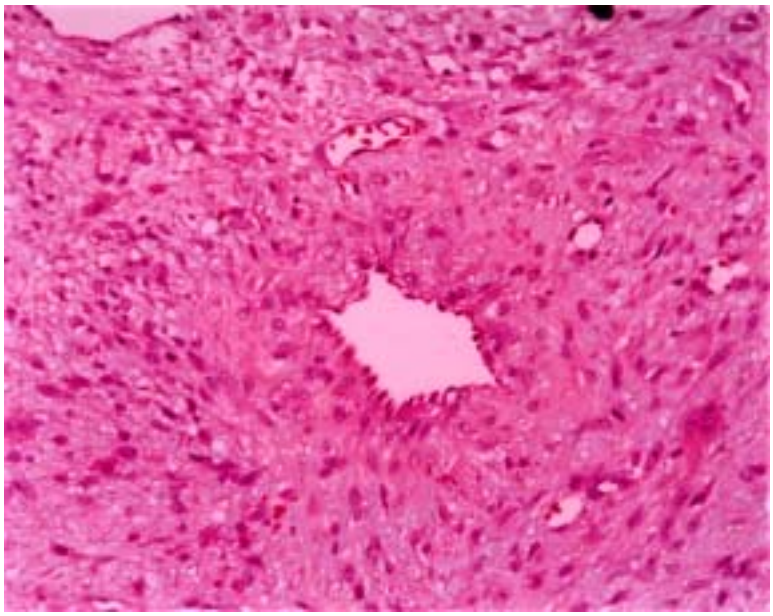

Fig. 5. Histologic micrograph showed thickness of blood vessel walls composed with smooth muscle cells in a whirling pattern. Fasicles of spindle cells with eosinophilic cytoplasm and spindled shape vesicular nuclei are also noted. Atypical change of the spindle cells and mitotic figure in the persistent tumor tissue are not found. (hematoxylin-eosin stain; original magnification×100)

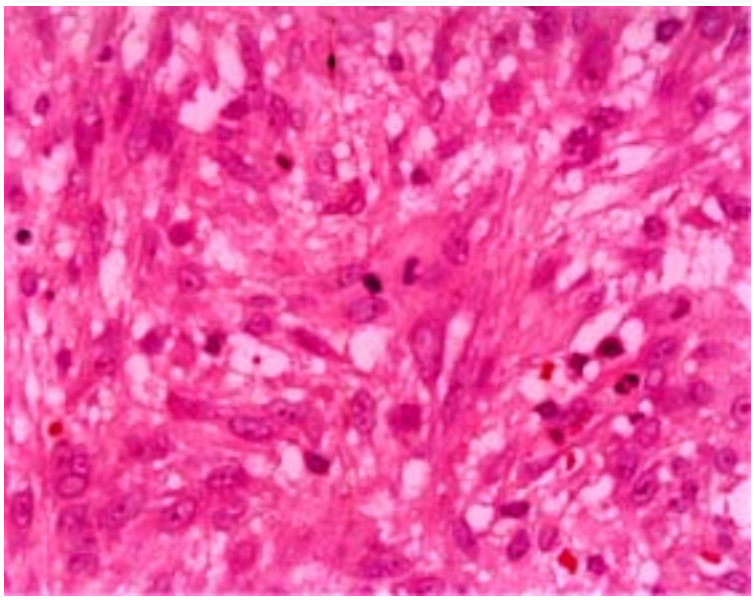

Fig. 6. Special immunohistochemical stains of HHF-35 demonstrated focal positive results over these spindle cells. (HHF-35 immunostain; original magnification×400)

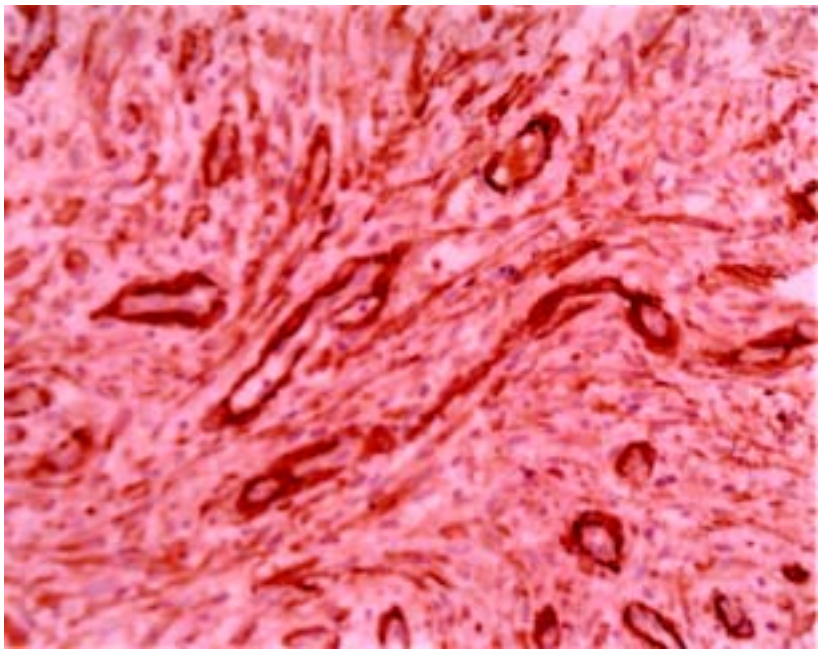

Fig. 7. Photomicrograph of MIB-1 special staining showed about 15% of mitotic activity rate. (MIB-1 stain; original magnification×200)

with tumor capillaries that differentiate into smooth muscle cells. More recent research has identified some chromosome abnormalities using genome mapping or DNA screening techniques in patients with angioleiomyomas. Nevertheless, as in most neoplasms, the angioleiomyoma formation mechanism remains obscure.

Oral mucosal angioleiomyomas characteristically range in size from 2 mm to 10 mm. They usually present as small, round or elevated sessile, normal colored nodules that vary in firmness. Rough or irregularly circumscribed tumors are seldom encountered. Ulceration is also rare, although acute inflammatory change has sometimes been reported, as in our case [3]. Although the most frequent subjective complaints of patients with angioleiomyoma of the extremities are pain or tenderness or both, patients with head and neck tumors seldom complain of pain [4]. Because angioleiomyoma is usually slow-growing, in a superficial location, and asymptomatic, the average preoperative period is months to years.

Accurate diagnosis before surgery without prior positive incisional biopsy is usually difficult, even though several diagnostic tools could be used [5]. Clinical differential diagnosis should include mesenchymal tumor (pyogenic granuloma, fibroma, and lipoma), epithelial or salivary gland originating from benign tumor, hemangioma, or malignant tumor (sarcoma and leukemia) [6]. A retrospective review of the literature from 1988 to 2001 found 21 tumors in 21 patients with head and neck angioleiomyoma, which were well-encapsulated and without extracapsular extension. Though, preoperative embolization of the tumor can potentially reduce perioperative bleeding, complete removal of angioleiomyoma has been achieved without massive blood loss in most

cases. Therefore, simple surgical excision along the tumor margin or capsule is always the treatment of choice. Wide excision with significant margin is usually not necessary. Recurrence is rare [7], except in cases of incomplete excision or deep-seated tumor. Regardless of the pathological type, about 5% recurrence rates are documented and the regrowth can usually be removed with ease without further recurrence. Although ultrasound or color flow imaging will show that the tumor is a vascular entity, massive bleeding during surgery is infrequent.

Histologically, angioleiomyomas contain dilated vascular spaces with an endothelial cell lining and smooth muscle bundles arranged in a more concentric fashion. The smooth muscle tumor cells are generally well-differentiated, cigar-shaped cells with blunt ends [8]. Angioleiomyomas together with other lesions such as myofibromas or neurofibromas, have similar microscopic appearances. However, special stains specific for smooth muscle actin are useful in distinguishing smooth muscle from collagen, both of which are pink in hematoxylin-eosin staining. Smooth muscle is stained dark red by Masson trichome, red by aniline blue, and yellow by Van Gieson's [8].

In conclusion, oral angioleiomyomas are benign smooth muscle tumors. Because these tumors have only a limited degree of associated morbidity, careful differentiation of these lesions from other malignant tumors such as leiomyosarcoma before treatment is necessary and important [9]. Although the number of mitotic figures defining malignant change has not been established, there is one case report that shows an angioleiomyoma can occur in association with a leiomyosarcoma. [10] This suggests there is a potential for malignant conversion, and therefore regular follow-up of patients with the above findings is always suggested. [11]

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下顎舌側牙齦之血管平滑肌瘤—病例報告

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摘 要

平滑肌瘤(leiomyoma)為一種源由於肌肉細胞所形成之良性軟組織腫瘤，在三種平滑肌瘤中，衍生於動靜脈血管之血管平滑肌瘤(angioleiomyoma)的原發性口腔病變亦是少見。根據文獻統計，口腔平滑肌瘤約只佔所有腫瘤之0.06%，其中約66%屬於血管平滑肌瘤。臨床表現多呈現為單一且緩慢生長之型態，極少疼痛。早期惡性轉變機率低，亦少致死。診確定斷須由特殊之鏡下表徵，包含增生肌肉細胞群，圍繞著擴大之血管腔。本病例報告一中年女性，求診時為下顎齒齦區之慢速增生牙齦腫塊，經入院詳察後診斷為血管平滑肌瘤。本文將就血管平滑肌瘤之臨床表現、病理組織表徵及治療方式作一討論。

關鍵語：血管平滑肌瘤，惡性轉變，牙齦腫塊。

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