

Angioleiomyoma of the Lower Lip

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Abstract

Leiomyoma is a benign smooth muscle tumor that occurs most frequently in the uterine myometrium, gastrointestinal tract, skin, and lower extremities. Leiomyoma rarely affects the oral cavity. Angioleiomyoma (vascular leiomyoma) is a histological subtype of the leiomyoma. The diagnosis is commonly determined by histopathological studies. This case report shows a 57-year-old male patient with a lesion of the lower lip. After laser excision, hematoxylin and eosin and smooth muscle actin staining confirmed the diagnosis of angioleiomyoma.

Keywords: Angioleiomyoma, histopathology, laser, lower lip

INTRODUCTION

Leiomyoma is a benign smooth muscle tumor that can appear at any location. It most frequently appears in the female genital tract (95%) followed by the skin (3%) and the gastrointestinal tract (1.5%). Approximately 1% of leiomyoma occurs in the head and neck.^[1] Only 0.065% of the leiomyomas are diagnosed intraorally.^[2] When detected, they are typically found on the lips (lower/upper), tongue, palate (soft/hard), buccal mucosa and rarely on the gingiva, buccal or labial sulcus, floor of the mouth, and mandible.^[3] Most of the angioleiomyomas are well-defined, typically painless and slow-growing lesions, <2 cm in diameter, covered by normal mucosa, and are sometimes bluish or purple in color.^[2,3] In terms of clinical presentation, it is very difficult to differentiate a leiomyoma from other mesenchymal tumors. Diagnosis is mainly made by histopathological examination. According to the World Health Organization, leiomyomas are divided into two categories: angioleiomyoma and leiomyoma of deep soft tissue. Angioleiomyomas are solitary forms of leiomyomas that usually occur in the subcutis and are the most common variant of leiomyomas affecting the oral cavity. Surgical excision is the main treatment of leiomyoma and recurrences are rare. We report a case of an angioleiomyoma occurring in the lower left lip with clinical characteristics of a mucocele/fibroma.

CASE REPORT

A 57-year-old male patient presented to the Department of Oral and Maxillofacial Surgery, of a dental institution with

a swelling in the left lower lip region for about 6 months. Swelling was uniform in consistency and painless with no other signs of inflammation. Clinical examination revealed an exophytic, nodular, reddish-purple, well-outlined lesion measuring about 1 cm × 1 cm in diameter, located on the left lower lip [Figure 1]. The patient had no other dentoalveolar symptoms and no sign of local infection.

Surgical excision under local anesthesia with diode laser (980 nm) was done. An elliptical incision was made to fully enucleate the lesion along with the overlying mucosa [Figure 2a and b]. Antibiotic and analgesic therapy was given to the patient for 5 days. Antibacterial chlorhexidine gluconate rinse (0.12%) was prescribed for one week.

The specimen was fixed in 10% formalin solution. The histological examination made with hematoxylin and eosin [Figure 3a] and smooth muscle actin staining [Figure 3b] revealed the diagnosis of angioleiomyoma. The lesion was composed of large vascular channels of varying caliber, surrounded by thick walls of irregularly arranged, spindle-shaped cells [Figure 3a]. The spindle-shaped cells

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Figure 1: Clinical representation of lesion on lower lip

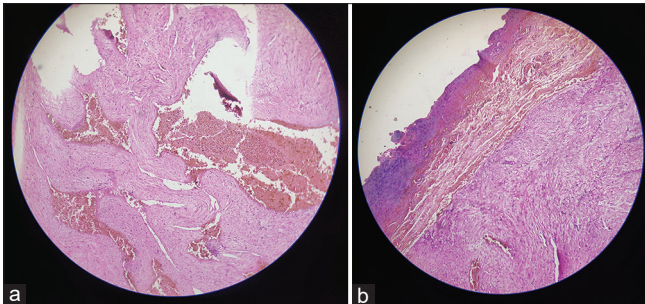


Figure 3: (a) Histopathology characteristics, (b) Histopathological view with different magnification

showed elongated blunt-ended nuclei and eosinophilic cytoplasm-forming smooth muscle bundles showing interlacing patterns with collagen fibers between dilated and slit-like vascular spaces [Figure 3b]. Whorled bundles of muscle fibers were seen fused with the vessel walls. Necrosis, atypical mitosis, and pleomorphism were not observed in the histological examination. The postoperative course of the patient was uneventful with 7 days of follow-up [Figure 4]. The tissue was completely healed and there was no sign of scar. There was no recurrence at 3 months of follow-up.

DISCUSSION

Oral leiomyomas may appear at any age, but the greatest prevalence is in individuals of 35–57 years' age with gender preference for female. In this report, the patient was a 35-year-old male. Oral cavity leiomyomas are uncommon lesions, representing 0.016%–0.065% of all leiomyomas.^[3,4] Leiomyomas are identified by their smooth muscle cell lineage and are histomorphological classified as either solid angioleiomyoma or epithelioid types. According to the World Health Organization classification of tumors of soft tissue (2002), the most frequent type is angioleiomyoma with a 74% incidence rate, followed by solid leiomyomas with a 25% incidence rate and there is only one case of an epithelioid leiomyoma described in the literature.^[5] Leiomyomas are

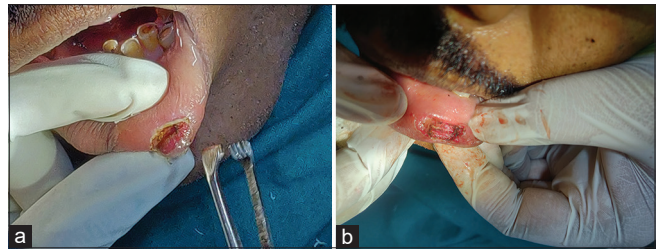


Figure 2: (a) Elliptical incision using laser was given, (b) Lower lip after excision of lesion



Figure 4: Follow up clinical presentation on day 7

rare in the oral cavity because of the absence of smooth muscle. They are typically found on the lips, tongue, palate, and buccal mucosa. Brooks *et al.*^[3] showed that the most frequently reported site was the lip (48.6%), followed by the palate (21.1%), buccal mucosa and tongue (each 9.2%), mandible (8.3%) and buccal sulcus, labial sulcus, floor of the mouth, and gingiva (each 0.9%). Although most mucosal lesions varied in size from a few millimeters to 2 cm, our patient's lesion is >2 cm. Although angioleiomyomas are vascular lesions, only 55.9% of cases appeared red, blue, or purple, the remainders were gray, white, or color of normal mucosa.^[4] Oral angioleiomyomas are generally well-defined, nodular, painless, slowly enlarging lesions. However, some central lesions can be painful. The clinical differential diagnosis relevant to angioleiomyoma usually includes other benign mesenchymal tumors (fibroma, neurofibroma, lipoma, or leiomyosarcoma), salivary gland neoplasms (mucocele and pleomorphic adenoma), vascular tumors (e.g., lymphangioma, hemangioma, and pyogenic granuloma), and soft-tissue cysts (dermoid cysts). When the tumor is located in the hard palate, adjacent to teeth, it can be confused with a periodontal lesion.^[3] The treatment of choice for oral angioleiomyomas is surgical excision. Despite vascular component, profuse bleeding during removal is rarely seen. Rarely, recurrence has been reported. Thus, it is important to obtain complete removal with safe margins to avoid recurrence. About 5% of leiomyomas show local recurrence.^[7] This has been attributed

to incomplete excision or deeply situated lesions. The case of malignant transformation has not been reported in literature.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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