

Rare Vascular Leiomyoma of the Tongue: Case Report

Dilin Nadir Vasküler Leiomyomu

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ABSTRACT

Leiomyomas are smooth, benign muscle tumors, and although they can appear at any location in the body, the most frequent site is the uterine myometrium. A leiomyoma is very rarely located in the oral cavity, probably because of the lack of smooth muscle tissue in this region. Leiomyomas can occur at any age and typically manifest themselves as asymptomatic, slow-growing masses, occasionally with painful lesions. There is a slight male gender predominance. When a leiomyoma occurs in the oral cavity, the most common sites are the lips, palate, tongue, and oral mucosa. Due to the unspecific clinical appearance of leiomyoma diagnosis is determined by histological studies and by special specific stains. Treatment is surgical, and it is important to resect the lesion completely because of the risk of recurrence. To make a contribution to the literature, we present our case of a 23-year-old male patient with a tumor located on the base of the tongue, growing toward the oropharynx and causing dysphagia. After total resection with a diode laser, we did not encounter any complications, and at one-year follow-up, the tumor did not show any recurrence.

Keywords

*Leiomyoma; smooth muscle tumor;
tongue*

ÖZET

Leiomyom'lar vücudun her yerinde görülebilmekle birlikte en sık olarak uterus myometriyum'unda görülen benign düz kas tümörleridir. Leiomyom'lar oral kavitede düz kas dokusu az olduğundan dolayı bu bölgede oldukça nadir görülürler. Herhangi bir yaşta görülebilen bu tümörler tipik olarak asemptomatik ancak bazen ağrılı olabilen, yavaş büyüyen kitleler olarak görülürler. Erkeklerde biraz daha sıktır. Oral kavite içerisinde en sık dudak, damak, dil ve oral mukozada ortaya çıkarlar. Kendine özgü bir kliniği olmamasından dolayı, tanısı histolojik olarak preparatların özel boyalarla boyanarak incelenmesi ile konulur. Tedavisi cerrahidir ve rekürrens riski nedeniyle lezyonun tamamen eksizye edilmesi önemlidir. Literatüre katkıda bulunmak amacıyla 23 yaşında erkek hastada, dil kökünde yerleşen, orofarenkse doğru büyüyen ve disfajiye neden olan olguyu sunduk. Lezyonun diode lazer ile eksizyonu sonrası herhangi bir komplikasyon gelişmedi ve hastamızın 1 yıllık takibinde rekürrens saptanmadı.

Anahtar Sözcükler

*Dil; düz kas tümörü;
leiomyom*

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INTRODUCTION

Leiomyomas are benign mesenchymal tumors arising from smooth muscles, and in 95% of all cases, they occur in the uterine myometrium. The gastrointestinal tract and skin are less commonly affected.¹ Less than 1% of all leiomyomas are seen in the head and neck region.² Approximately 0.06% of leiomyomas are found in the oral cavity, probably due to the lack of smooth muscle tissue in this region.¹ The most common sites in the oral cavity are the lips, palate, tongue, and oral mucosa.³

According to the World Health Organization, there are three histological subtypes of leiomyoma. These are solid leiomyoma, angiomyoma (vascular leiomyoma), and a rare form of epithelioid leiomyoma (leioblastoma).⁴ Clinically, oral leiomyoma manifests itself as an asymptomatic, slow-growing submucosal nodular mass. Tumor's depth and vascularity determine the color of the lesions.⁵ While most cases are asymptomatic, there may be symptoms such as pain, teeth mobility, and difficulty in chewing.^{6,7} It is presumed that leiomyomas originate from smooth muscles in the tunica media of the blood vessel wall and in the excretory ducts of salivary glands.^{8,9} Although oral leiomyomas can appear at any age, most cases are noted in adults, with the highest prevalence in the age group of 40-59 years, and there is a slight male gender predominance.⁷ The diagnosis is determined by histological studies.⁶ At present, treatment is surgical, and it is important to resect the lesion completely because of the risk of recurrence.

We present our case of an oral vascular leiomyoma sited at the base of the tongue, describe its treatment, and review the literature.

CASE REPORT

A 23-year-old male patient, without any chronic disease or previous history of surgery, was referred to our clinic with a complaint of a mass on the tongue and a difficulty in swallowing solid foods in the last five months. During his examination, a well circumscribed, asymptomatic, dull reddish mass with lobulated contours was seen at the tongue base, and the mass was measured to be approximately 4x4x3 cm at its largest dimensions (Figure 1). Radiological investigation with MRI, using contrast, did not show any sign of invasion, and the lesion was isointense, with muscular structures on the T1 AG se-

quence; there was significant hypointensity in the center and millimetric hyperintense foci on the T2 AG sequence (Figure 2). The MRI study presumed a benign striated muscle tumor (rhabdomyoma). The tumor was biopsied, and a provisional diagnosis of irritation fibroma was made. For a definitive diagnosis as well as for treatment, the tumor was excised with safe margins, with a diode laser under general anesthesia (Figure 3). The mass was sent for histological examination, and there were no intraoperative or postoperative complications.

The histological report revealed that the tumor consisted of interlacing fascicles of spindle cells with slight eosinophilic cytoplasm and dull stained nuclei (Figure 4). The tumor was also rich in terms of small vessels, some of which were also ectatic. Edema, fibrosis, hyalinisation, calcification, and myxoid changes were seen in cross-sectional analysis. There was no necrosis, significant nuclear atypia, nor increase of cellularity. Immunohistochemical techniques were also applied, as well as monoclonal antibodies against SMA, desmin, vimentin, caldesmon, S-100 protein, myoglobin, keratin, CD34, MyoD1 protein, and EMA. Positivity for SMA, desmin, caldesmon, and vimentin was observed, and with these findings, the diagnosis of leiomyoma was

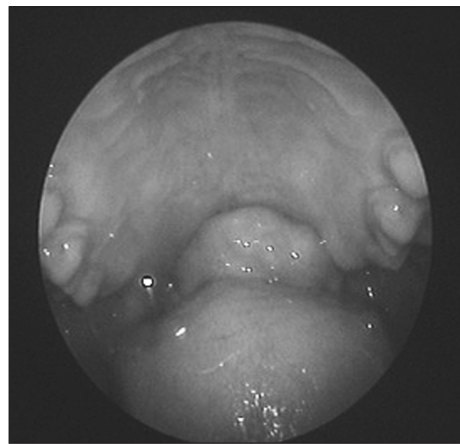


Figure 1. Endoscopic view of the leiomyoma at the base of the tongue.

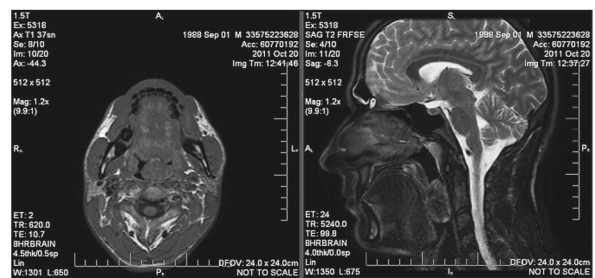


Figure 2. MRI shows the mass on the base of the tongue.

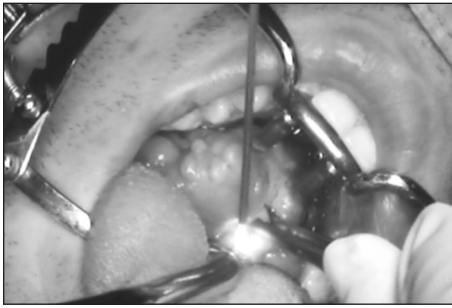


Figure 3. Intraoperative image shows the excision of the tumor with diode laser.

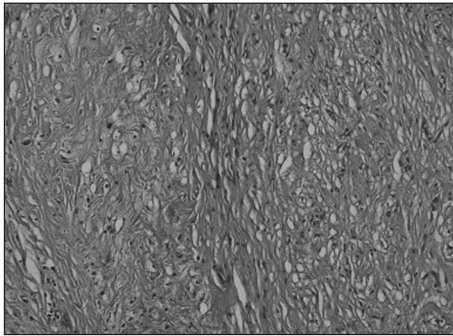


Figure 4. Interlacing fascicles of spindle cells with slight eosinophilic cytoplasm and dull stained nuclei (Hematoxylin & Eosin x 200).

confirmed (Figure 5). After a one-year follow-up, there was no sign of recurrence (Figure 6).

DISCUSSION

Leiomyoma is a benign, smooth muscle tumor that can appear in any location, with the most common sites being the female genital tract, skin, and the gastrointestinal and food intake tract.¹⁰ Since its first description in 1884, additional cases of oral leiomyoma have been reported.¹ Oral leiomyomas are very rare, and approximately 0.06% of leiomyomas are found in the oral cavity, probably due to the paucity of smooth muscle tissue in this region.^{1,11} The origin of oral leiomyomas is controversial. Although Glass⁹ considers the smooth muscles of the ductus lingualis and circumvallate papilla as a possible origin, Staut⁸ suggests the smooth muscle of the tunica media of the blood vessel wall as another source for oral leiomyomas.

Although oral leiomyomas can appear at any age, most cases are noted in adults, with the highest prevalence in the age group of 40-59 years of age. There is a slight male gender predominance.⁷ In our case, the pa-

tient was male and 23 years of age, younger than would be expected.

When encountered in the oral cavity, the most common sites of oral leiomyoma are the lips, palate, tongue, and oral mucosa.³ According to the World Health Organization, leiomyomas are classified under three histological subtypes. These are solid leiomyoma, angioleiomyoma (vascular leiomyoma), and a rare form of epithelioid leiomyoma (leioblastoma).⁴ The most frequent type is angioleiomyoma, which makes up 74% of all cases. Solid leiomyomas account for 25% of all cases, and there is only one reported case of epithelioid leiomyoma.¹² Solid leiomyoma is a well circumscribed tumor that consists of interlacing bundles of fusiform cells, and it is not associated with smooth vascular muscle.¹³ This type of tumor is typically normal in colour.¹⁴ Angioleiomyomas are well bordered tumors that originate from the smooth muscles of blood vessels.¹³ Between the vessels, intervening bundles of smooth muscle cells can be seen.¹⁴ Epithelioid leiomyomas consist of round or polygonal cells, and smooth muscle fibers are rarely found.¹³ The vascular leiomyoma

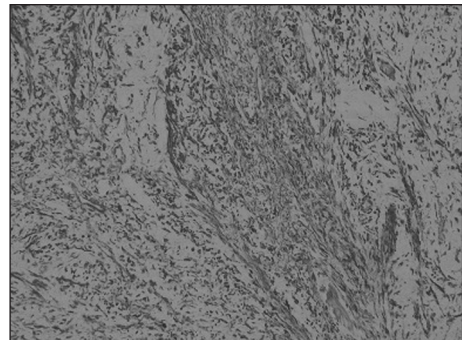


Figure 5. The bundles of spindle cells show diffuse and strong immunoreactivity for Smooth Muscle Actin (SMA).

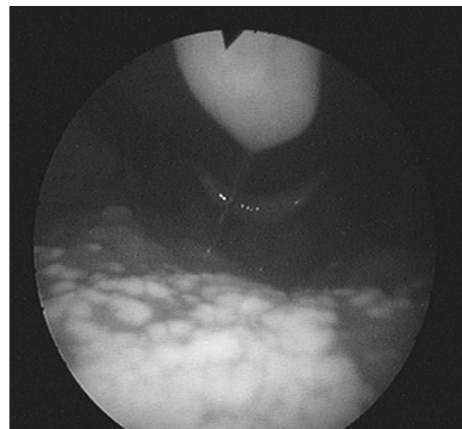


Figure 6. Endoscopic view of the base of the tongue after one year follow-up.

is the most frequent type seen in the oral cavity.^{15,16} This is explained by the scarcity of smooth muscle in the oral cavity and by the proposed origin in the tunica media of blood vessels. Histological examination revealed that our patient had vascular leiomyoma.

Clinically, oral leiomyoma manifests itself as an asymptomatic, slow growing nodular mass. Sometimes ulceration on the overlying epithelium may occur. The depth and the vascularity of the lesion determine the color of the tumor.⁵ Only 55.9% of these tumors are red, blue, or purple in color, and the rest display the appearance of normal mucosa or have a grayish tone.¹⁵ Our patient had a dull reddish lesion, and there was no ulceration on the overlying mucosa. Although most cases are asymptomatic, there may be symptoms such as pain, difficulty in chewing and swallowing, and also teeth mobility, if the mandibula is affected.^{6,7} Although dyspnea had been expected to be our patient's major complaint, because of the tumor's size and location at the base of the tongue, the only symptom was dysphagia.

The diagnosis of leiomyoma is determined by histological studies, and for a differential diagnosis, im-

munohistochemical studies are recommended.^{6,16} Due to its unspecific clinical appearance, it is difficult to differentiate a leiomyoma from other mesenchymal tumors (fibroma, lipoma, neurofibroma, leiomyosarcoma, etc.), vascular tumors (lymphangioma, hemangioma, pyogenic granuloma), or salivary gland tumors (mucocele, pleomorphic adenoma), and soft tissue cysts such as dermoid cysts which must also be excluded.^{15,17}

At present, the best treatment for oral leiomyomas is the total excision of the lesion with safety margins. Periodic examination of the patient is also important because of the risk of recurrence. We did not see any sign of recurrence during our patient's one-year follow-up.

In conclusion, we present our case of a rare vascular leiomyoma of the oral cavity at the base of the tongue, causing dysphagia. The tumor occurred in a relatively short time, namely about five months. Surprisingly, the lesion did not cause dyspnea despite its location and size. We managed to excise the tumor completely with a diode laser without any intra- or post-operative complication, and no signs of recurrence have been observed during our one-year follow-up.

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