Clinical and Experimental Health Sciences

Case Report / *Olgu Sunumu*

Leiomyoma of Oral Cavity: Case Report and Literature Review

Oral Kavitede Leiomyom: Vaka Sunumu ve Literatür Taraması

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Abstract

Leiomyoma is a benign smooth muscle tumor that can be observed at any region but is mostly observed in the female genital tract. Although leiomyomas are uncommon in the oral cavity, they are usually observed on the lips, tongue, and palate. Here we present a case report of leiomyoma in a female patient along with the literature review. A 35-year-old Turkish female presented with a 1-year history of a well-delineated, asymptomatic, and soft mass on the vestibule alveolar mucosa of the right mandibular molar region. Under local anesthesia, excisional biopsy was performed and the specimen was sent for histopathological examination. The histopathological diagnosis was leiomyoma. Based on clinical appearance, it is very challenging to distinguish a leiomyoma from other mesenchymal-origin tumors. Histological analysis determined the final diagnosis of oral leiomyoma, and surgery was performed because it was the only option for the treatment of these lesions.

Keywords: Leiomyoma, benign tumor, oral cavity

Öz

Leiomyoma en sık kadın genital sisteminde olmak üzere herhangi bir bölgede bulunabilen benign düz kas tümörüdür. Leiomyomalar oral kavitede yaygın olmamakla beraber dil, dudak ve palatinal mukozada izlenebilir. Bu olgu sunumunun amacı leiomyoma saptanan bir bayan hastayı sunarak literatür taranmasıdır. 35 yaşında bir bayan hastada sağ mandibular molar bölgede vestibül alveol mukozasında yaklaşık bir yıldır sınırlı, asemptomatik yumuşak doku kitlesi mevcut olduğu saptanmıştır. Lokal anestezi ile eksizyonel biyopsi uygulanmış ve alınan örnek histopatolojik incelemeye gönderilmiştir. Histopatolojik tanı leiomyoma idi. Leiomyomayı diğer mezenşimal kaynaklı tümörlerden klinik olarak ayırmak oldukça zordur. Kesin tanı histolojik muayene ile olmalı ve bu lezyonların tedavisi cerrahi olarak yapılmalıdır.

Anahtar Kelimeler: Leiomyom, benign tümör, ağız kavitesi

INTRODUCTION

Leiomyomas are benign smooth muscle tumors that are frequent in the uterus, skin, and gastrointestinal tract. These can be attributed to only 0.42% of all soft tissue lesions reported in the oral cavity, caused by the lack of smooth muscle in this region. These may be observed in any age group, but the greatest prevalence of head and neck leiomyomas is seen between 40 and 49 years of age. Leiomyomas in the oral cavity appear as asymptomatic, slow-growing, submucosal masses, which are usually located on the hard palate, tongue, lips, or buccal mucosa. They are rarely observed on the floor of the mouth and gingiva (1-4).

Histologically, three types of leiomyomas are present: epithelioid leiomyoma, solid –form leiomyoma, and vascular leiomyoma (angiomyomas or angioleiomyomas) (5, 6). Solid leiomyomas are well-delineated tumors that are are introduced as connected bundles of spindle-shaped smooth muscle cells. The nuclei are elongated, pale staining, and blunt ended, and mitotic figures are rare in lesions. Because of hyperplasia of their smooth muscle coats, well-delineated vascular leiomyomas have multiple blood vessels with thickened walls (7, 8). The most recognized variant followed by solid leiomyomas in the oral cavity is vascular or angiomyoma.

Although it is mainly diagnosed by histological examination of hematoxylin and eosin (H&E)-stained tissue sections, microscopic diagnosis may sometimes be difficult because of the fact that spindle cell proliferation shares many similarities with schwannoma, neurofibroma, fibromatosis, and myofibroma. Therefore, special stains that identify collagen may be helpful in distinguishing these lesions. Immunhistochemical demonstration of actins can confirm the diagnosis (9-12).

Surgical excision of lesions appears to be the best choice of treatment. Moreover, recurrence of the lesion is unexpected (1, 2). Here we present a case report of leiomyoma in a female patient along with the literature review.

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CASE PRESENTATION

A 35-year-old Turkish female presented with a 1-year history of a well-delineated asymptomatic mobile mass on the lingual alveolar mucosa of the right mandibular molar region. The medical and family history was unremarkable. Intraoral examination revealed a compressible, soft, mucosal colored mass (Figure 1). Under local anesthetics, an excisional biopsy was performed, and the specimen was sent for histopathological examination at the Department of Oncologic Cytology and Tumor Pathology, Institute of Oncology, İstanbul University (Figure 2a, b). This study was performed according to the guidelines of the Declaration of Helsinki concerning ethical principles for medical research involving human subjects, and written informed consent was obtained from the patient.

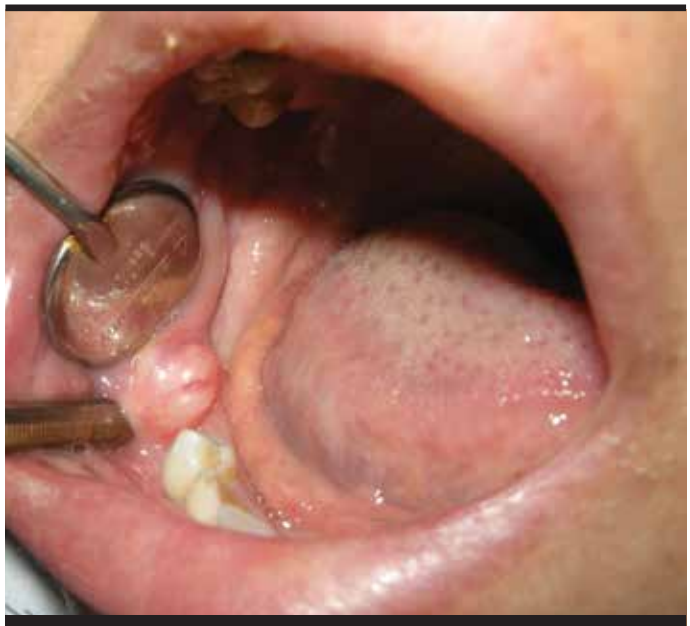

Figure 1. Clinical presentation of mucosal colored mass

Histopathological features were as follows: the surface epithelium displayed a normal maturation pattern and a well-delineated tumor constituting of spindle-shaped smooth muscle cells. These cells had elongated, fusiform nuclei (Figure 3, 4). In addition, the cells were aligned in sheets of well-delineated cell outlines. Mitotic figures were absent. Immunohistochemical stains for desmin and smooth muscle actin (SMA) revealed positivity within the lesional cells (Figure 5, 6) and a low Ki-67 percentage in tumor cells (Figure 7). The histopathological diagnosis was leiomyoma. At 5-month follow-up, there was no recurrence.

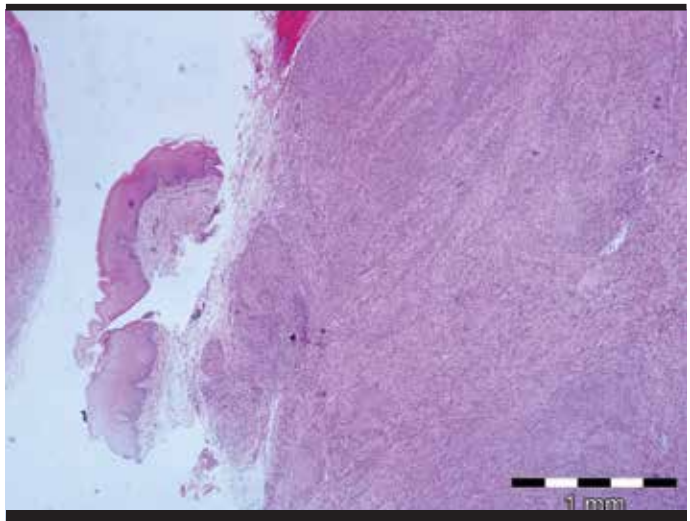

Figure 3. The surface epithelium exhibited a normal maturation pattern and a well-circumscribed tumor with fusiform cells (H&Ex40)

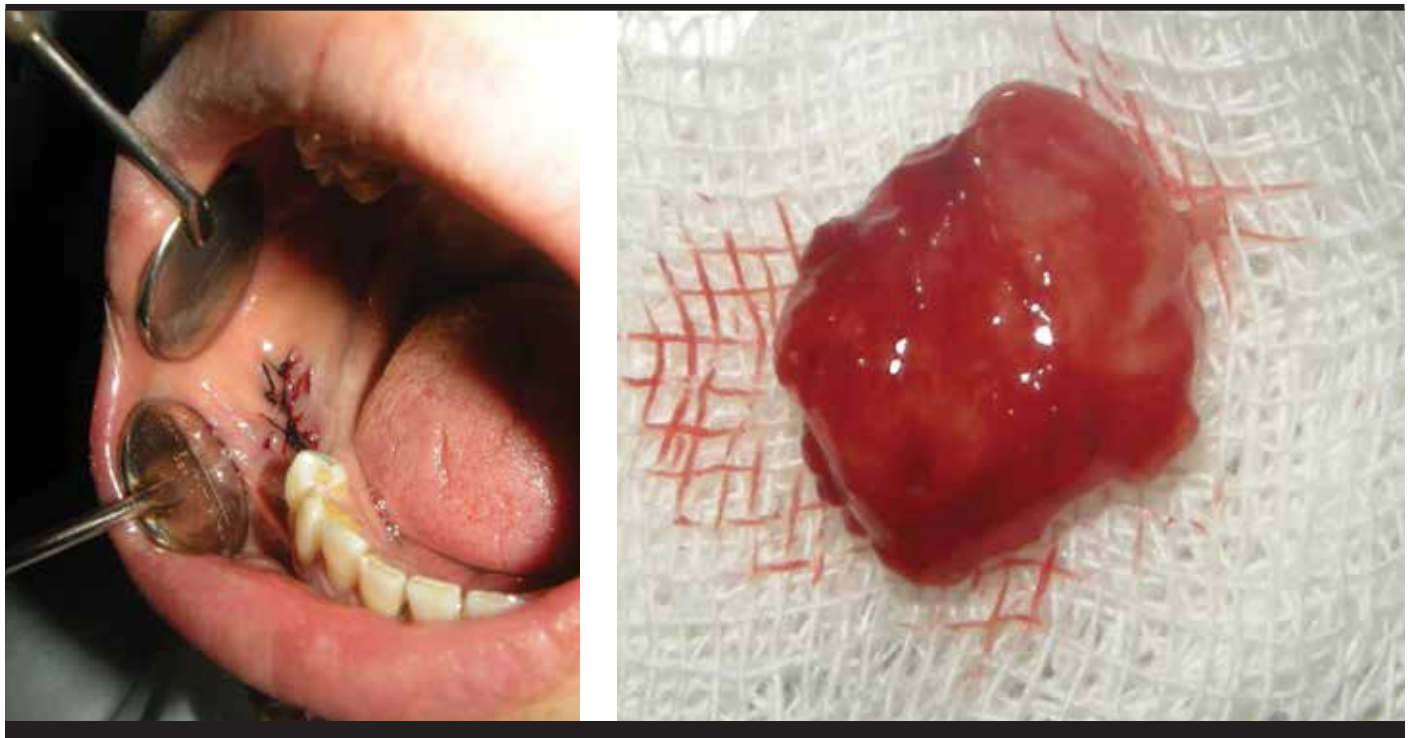

Figure 2. a, b. (a) Suture of the wound, (b) lesion removed by excisional biopsy

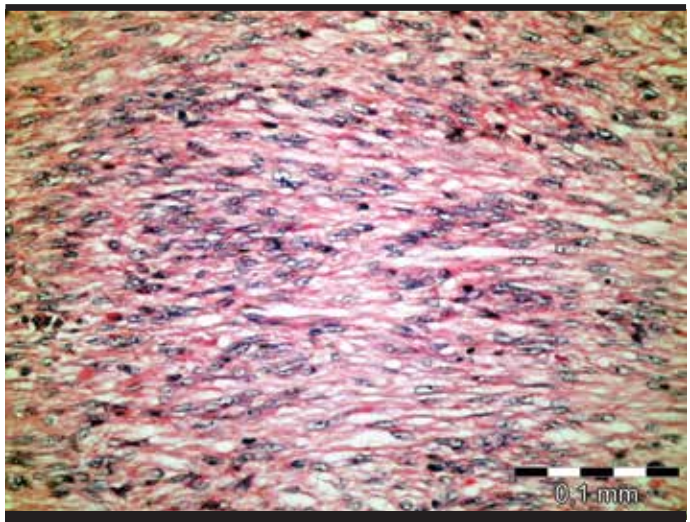

Figure 4. Bundles of small fusiform cells (H&Ex400)

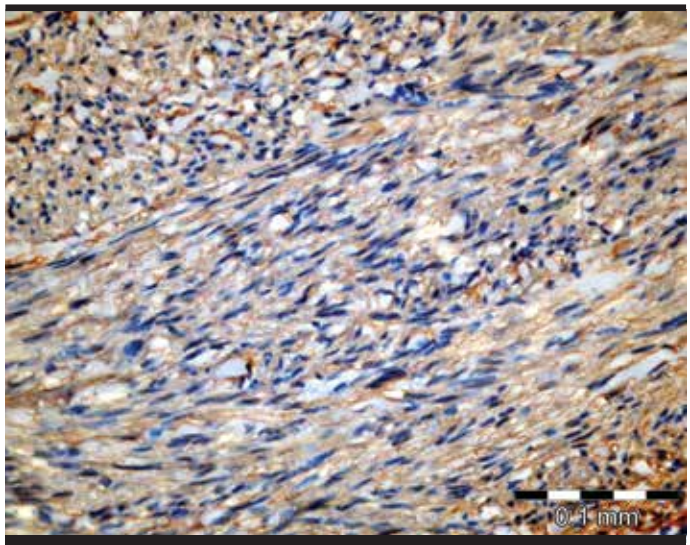

Figure 5. Lesional tissue showing diffuse for smooth muscle actin (SMAx400)

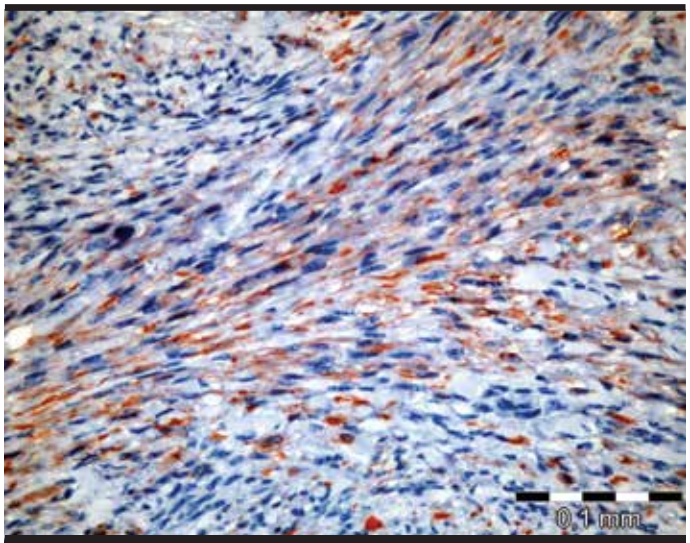

Figure 6. Tumor cells with positive for desmin (Desminx400)

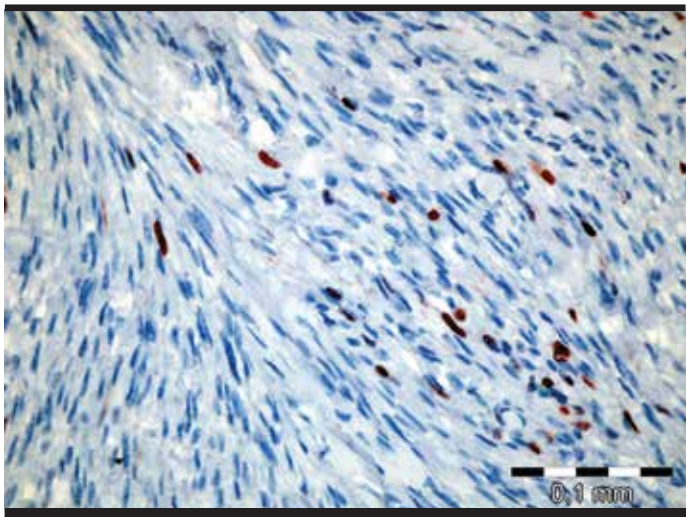

Figure 7. Low Ki-67 percentage of tumor cells (Ki-67x400)

DISCUSSION

Oral leiomyomas are rare benign tumors of the oral cavity that are hypothesized to originate from excretory ducts of salivary glands and vascular smooth muscles (5). There are histological variations (clear cell, epithelioid, granular cell, and myxoid changes) and secondary degenerative changes (hyaline degeneration, cystic change, myxoid degeneration, infection, necrosis, and calcification) in leiomyoma of the oral cavity and head and neck region. It is thought that these changes result from deficient blood supply, leading to a replacement of muscle fibers by collagen, hyaline material, calcium, mucopolysaccharide, or a combination of these factors (13, 14).

The World Health Organization classifies leiomyoma in soft tissue tumors (WHO 2002) that arise from smooth muscle. Three types of leiomyoma are categorized by WHO. The most frequent form of leiomyoma is angiomyoma (74%), which is followed by solid leiomyomas (25%). Only one case of epitheloid leiomyoma has been reported in the literature (9-11).

Oral leiomyomas represent 0.4% of all soft tissue lesions in the oral cavity. The most frequently affected area is the lips (27.5%), followed in a descending order by the tongue (18.3%), buccal mucosa (15.5%), palate (15.5%), and gingiva (8.5%) (1-3, 7, 15). In this Case, the site was vestibular mucosa of the right mandibular molar region.

Histological examination of the case revealed that the tumor was in a solid form and it was a rare type of tumor for the oral cavity. Compared with the vascular type, solid leiomyomas are smaller. Histologically, solid leiomyoma contains interlaced bundles of spindle-shaped or stellate smooth muscle cells with blunt-ended, elongated, pale-staining nuclei: lesions are rather cellular; yet, the packs of tumor cells are separated by collagenic strands. All the histological characteristics of the lesion were observed in our histological examination (9).

Immunohistochemical demonstration of actins can confirm the diagnosis. Actin is a small cytoplasmic filament that has contractile properties. The six actin isotypes differentiate between striated muscle, smooth muscle, and nonmuscle cells. Antismooth muscle actin

generally provides good intensity and sensitivity for the detection of leiomyoma. Staining for desmin is less reliable because it is positive in about two -thirds of cases. In our case, immunohistochemical analysis demonstrated the expression of smooth muscle actin and desmin within the tumor cells.

The absolute treatment for benign smooth muscle tumors is mainly surgical excision. Surgery has been performed as the only treatment option in all leiomyomas reported in the literature (1-3). There are no reports of lesion recurrence after total excision, and this indicates the need for cautious and complete excision of the lesion. Under local anesthesia, total excision of the mass was performed in our case. At 5-months follow-up, the patient did not present with any complications and recurrences.

CONCLUSION

The characteristics of leiomyoma may be similar to those of various lesions in clinical and histological examinations. Therefore, dental practitioners and pathologists should be aware of differential diagnosis.

Informed Consent: Written informed consent was obtained from patient who participated in this study.

Peer-review: Externally peer-reviewed.

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