Solid variant of leiomyoma in the hard palate: A rare case report and review of literature

Venkatesh Anehosur¹, Priyanka Acharya¹, Deepthi Shetty¹, U. S. Dinesh²

¹Department of Oral and Maxillofacial Surgery, SDM Craniofacial Research Centre, SDM College of Dental Sciences and Hospital, Dharwad, Karnataka, India
²Department of Pathology, SDM College of Medical Sciences and Hospital, Dharwad, Karnataka, India

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Abstract
Leiomyoma is a circumscribed benign smooth muscle tumor with rare incidence in the head-and-neck region of about 0.065% and occurring most commonly in uterine myometrium, gastrointestinal tract, skin, and lower extremities. The lesion usually presents as an asymptomatic lesion, with slight female predilection with the World Health Organization classified into three subtypes: Solid, angioleiomyoma, and epithelioid types, with angioleiomyoma (74%) outnumbering in terms of occurrence in oral cavity. We have described a rare occurrence of solid variant of leiomyoma in hard palate along with clinical presentation, investigation, and reconstruction of defect using buccal pad of fat with a brief review of literature. Leiomyoma accounts for less than 0.5% of all benign smooth muscle tumors occurring in oral cavity. It presents as a relatively asymptomatic lesion, with confirmed diagnosis being only through histopathological and immunohistochemistry. Surgical excision of the lesion with sound safety margins remains as the treatment of choice with almost nil recurrence rate. Leiomyoma is a benign rare occurrence in oral cavity with minimal documentation of solid variant in hard palate of less than 9% and thereby diagnosis and management including reconstruction of secondary defect is of utmost importance.

Keywords: Palate, smooth muscle, solid leiomyoma

Introduction
Leiomyoma is a benign smooth muscle tumor occurring at any location, with female genital tract (95%) followed by skin (3%) and gastrointestinal and food intake tract (1.5%) being the most common site of occurrence in descending order (World Health Organization [WHO], 2013).¹ The incidence of the occurrence of leiomyoma is rare in the oral cavity, caused by the lack of smooth muscle in this region and accounts to only about 0.42% of all soft tissue lesions.²,³ In oral cavity, it frequently involves the lips, tongue, buccal mucosa, palate, and gingiva in the descending order with peak incidence of occurrence between 40 and 49 years of age and with 2:1 female predilection; however, considerable controversy exists regarding gender predilection.⁴⁻⁷ Leiomyoma presents asymptomatic in nature with definitive diagnosis solely based on histopathology and immunohistochemistry with surgical excision of lesion being modality of choice in management.⁸⁻¹⁰

We are reporting a case of solid leiomyoma which presented at rarest site in the oral cavity, i.e., hard palate with brief review of literature.

Case Report
A 26-year-old Indian female, who was a lactating mother reported to us with the chief complaints of pain and ulceration in the hard palate for 7 months. Pain was intermittent, sharp aching, and intermittent in nature which was reduced in intensity on taking analgesics. On examination, a solitary, well-defined oval ulcerative lesion was noted in the hard palate measuring approximately about 1 cm x 1.5 cm with rolled out margins. Surrounding mucosa was erythematous. Lesion was non-scrapable and tender on palpation [Figure 1].

Incisional biopsy of the lesion was performed under local anesthesia which was suggestive of leiomyoma. Radiographic evaluation (orthopantomograph) was suggestive of no palatal bone erosion [Figure 2]. Routine blood investigations and physician fitness were sought and wide excision of lesion was performed under general anesthesia [Figure 3]. Reconstruction of defect was done using local flap (buccal pad of fat) and secured using 4–0 vicryl. The specimen was sent for histopathological examination which revealed a discontinuous predominantly hyperparakeratinized to hyperorthokeratinized squamous...
epithelium of variable thickness with basal cell hyperplasia and acute inflammatory cell infiltrate in the superficial epithelium with focal microabscess formation. The underlying connective tissue was fibrocellular to fibrous with areas of hyalinization. The connective tissue revealed numerous spindle-shaped cells with blunt-ended nuclei and perinuclear vacuole cells arranged in short fascicles. Variable number of deep inflammatory cells was evident subepithelially and in perivascular location in deeper areas. Deeper tissue revealed mucous acini and ducts with chronic inflammatory cell infiltrate, nerve tissue [Figure 4]. Immunohistochemistry was done which revealed diffusely positive tumor cells smooth muscle actin (SMA) and negative for cytokeratin and L-caldesmon [Figure 5] following which final diagnosis of solid variant of leiomyoma of hard palate was given. Post-operative period was uneventful and the patient was on regular follow-up with no recurrence.

Discussion

Leiomyoma is defined as an “circumscribed benign, often cutaneous tumor composed of intersecting bundles of mature smooth muscle cells”(WHO).[4,5] It usually presents as an asymptomatic lesion with literature suggestive of slight female predilection with peak incidence in the 4th–5th decade of life. With uterine myometrium and lower extremities outnumbering sites of occurrence, head-and-neck regions account for about 0.065% with lips (27.5%), followed by tongue (18.3%), buccal mucosa (15.5%), and gingiva (8.5%) being common sites of presentation.[5]

Literature dates back to first presentation in head-and-neck region, at the base of the tongue by Blanc in 1884 to present with very few cases being reported till date. The origin of the lesion is attributed to three areas with smooth muscle in oral cavity (Scouts): Tunica media of blood vessels, ductus lingualis, and circumvallate papilla.[8-10]

Leiomyoma usually presents as an asymptomatic small, solitary, gradually progressing lesion, with color of the lesion

Figure 1: Pre-operative: Solitary ulcerative growth over palate measuring about 1.5 cm in greatest dimension

Figure 2: Intraoperative: Excised lesion from hard palate in toto

Figure 3: 128 slice 1 mm MDCT coronal section illustrating no palatal erosion

Figure 4: Hematoxylin and eosin staining (×40 magnification) demonstrating connective tissue with numerous spindle-shaped cells with blunt-ended nuclei and perinuclear vacuole with spindle cells arranged in short fascicles
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Similar to the adjacent mucosa with rare association of secondary manifestations such as ulceration, pain, and teeth mobility. The WHO has histologically distinguished the lesion into three variants: Solid (25%), angioleiomyoma (74%), and only one case of an epithelioid leiomyoma.\(^5\) Angioleiomyoma was further classified into three histological subtypes (Morimoto,1973): Capillary, venous, and cavernous with capillary form the most common characterized by closely compacted smooth muscle and many small, slit-like vascular channels followed by venous form with thick vascular channels and well-defined muscular walls and cavernous form with presentation as dilated vascular channels and scanty smooth muscle.\(^6,7\) Vascular leiomyomas often exhibit as a blue or red discoloration which are usually tender or painful which in comparison to our case presented as an ulcerative, tender, and slowly progressive lesion over the palate.

With occult nature of clinical presentation, leiomyoma often poses a diagnostic difficulty, in terms of differentiating from commonly occurring other mesenchymal tumors (fibroma, neurofibroma, and lipoma), salivary gland neoplasm (mucocele and pleomorphic adenoma), vascular tumors (lymphangioma, hemangioma, and pyogenic granuloma), and soft tissue cysts such as dermoid cysts with definitive diagnosis mainly based on histological and immunohistochemistry.\(^6,8,9\) Various special stains are used to delineate collagen fibers and muscle fibers including hematoxylin-eosin staining, Mason’s trichome [Figure 6], van Geison’s stain, Mallory’s phosphotungstic acid, and immunohistochemical markers specific for smooth muscle types including SMA, desmin, vimentin, and cytokeratin.\(^6,8,9\)

Solid leiomyoma comprises interlacing bundles of smooth muscle fibers interspersed by varying amounts of fibrous connective tissue devoid of vascular smooth muscle. Whorled pattern of fibers is noted in fascicular arrangement in varying planes with typically spindle-shaped nucleus with blunt ends with uncommon mitotic figures.\(^5,6,8\) Angiosarcoma is well-delineated lesions with thickened walls and multiple tortuous blood vessels. Intertwining bundles of smooth muscle may be found between the vessels, intermixed with adipose tissue. Epithelioid leiomyoma presents as an acidophilic cytoplasm and round or polygonal cells with rare smooth muscle fibers.\(^10\)

The treatment of choice is surgical excision with adequate safety margins which is noted to have good prognosis with low recurrence rate and incidence of malignant transformation.

### Conclusion

Leiomyoma accounts for <0.5% of all benign smooth muscle tumors occurring in oral cavity. It presents as a relatively asymptomatic lesion, with confirmed diagnosis being only through histopathological and immunohistochemistry. Surgical excision of the lesion with sound safety margins remains as the treatment of choice with almost no recurrence rate.

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