

CASE REPORT

Common Presentation of Uncommon Lesion

¹Radhika MB, ²Lalita J Thambiah, ³Paremala K, ³Sudhakara M, ⁴Umashankar DN, ⁴Mahesh Kumar R

¹Professor and Head, Department of Oral Pathology, Krishnadevaraya College of Dental Sciences, Bengaluru, Karnataka, India

²Professor, Department of Oral Pathology, Krishnadevaraya College of Dental Sciences, Bengaluru, Karnataka, India

³Senior Lecturer, Department of Oral Pathology, Krishnadevaraya College of Dental Sciences, Bengaluru, Karnataka, India

⁴Reader, Department of Oral and Maxillofacial Surgery, Krishnadevaraya College of Dental Sciences, Bengaluru, Karnataka, India

Correspondence: Lalita J Thambiah, Professor, Department of Oral Pathology, Krishnadevaraya College of Dental Sciences Via Yelahanka, Sir MVIT Campus, Bengaluru-562157, Karnataka, India, Phone: 9448052427, e-mail: ljthambiah@gmail.com

ABSTRACT

Angioleiomyomas are rarely seen in the oral cavity. We present here an asymptomatic swelling discovered during routine intraoral examination. On excision and further histological examination it was diagnosed as an angioleiomyoma. There was no recurrence following excision.

Keywords: Angioleiomyoma, Leiomyomas, Palatal swelling.

INTRODUCTION

Palatal swellings are commonly seen in the oral cavity associated with dental caries, periodontal diseases, reactive phenomena, minor salivary gland pathologies, etc. Occasional mesenchymal lesions also present themselves as palatal swellings uncommonly in the oral cavity. One such lesion is a leiomyoma, which is rare due to the limited amount of smooth muscles in the oral cavity. Leiomyoma is a benign smooth muscle tumor, most commonly found associated with the uterus, gastrointestinal tract or skin, but is found rarely in the oral cavity because of the paucity of smooth muscles.¹ These account for 0.42% of all soft tissue neoplasms reported in the oral cavity.²

In the oral cavity, leiomyomas occur at any age with a mean age of 41 years and with a slight male predilection.² When the tumor occurs within the oral cavity, it is most commonly found as a soft tissue lesion on the lips, palate, tongue or gingiva. Intraosseous oral leiomyomas are even rarer than those found in oral soft tissues and excretory ducts of salivary glands. We present here a relatively uncommon case of angioleiomyoma of the palate.

CASE REPORT

A 48-year-old male patient presented to the outpatient department of Krishnadevaraya College of Dental Sciences for pain in the upper molar tooth since 2 weeks. On examination, he was found to have a painful carious right maxillary first molar. His oral hygiene was poor with periodontal pocket in relation to 16 and 17. He was also found to have an asymptomatic round, sessile swelling on the right side of the hard palate 1cm away from the midline in the region of 15 and 16. The swelling started two months back and had grown to

the present size. The swelling measured about 1.0 × 1.5 cm, was soft in consistency and pale pink in color. The surface of the lesion was smooth, nonulcerated and did not blanch on pressure (Fig. 1). He had no history of trauma, pain and discharge in relation to lesion in the palate. Aspiration ruled out a vascular lesion. Radiograph neither show any periapical and periodontal involvement in relation to teeth in the area nor any underlying bony change. A provisional diagnosis of a peripheral fibrous lesion was made. The lesion was excised under local anesthesia as a routine out-patient procedure, and the specimen was received for histopathological diagnosis.

HISTOPATHOLOGY

On macroscopic examination, the soft tissue mass was measuring 1.0 × 1.5 cm in dimension, soft in consistency, pale brown in color, and was cut into two halves (Fig. 2). The cut surface showed a peripheral condensation with slightly cellular core. The tissue was processed and sections were stained with hematoxylin and eosin (H&E), and subsequently with Mallory's phosphotungstic acid hematoxylin (PTAH).

Microscopically, the H&E stained sections showed a well-circumscribed non-encapsulated lesion in the deeper connective tissue. Plump oval or spindle-shaped cells were seen arranged in a streaming pattern and seem to form whorls originating from small vascular areas and separated by collagen bands. Small-sized arteries appeared to be consumed by these spindle cells. These cells had eosinophilic cytoplasm with prominent nuclei, which were oval and slender band shaped. No mitotic figures were seen. Perinuclear vacuolization was also seen in the tumor cells (Figs 3 and 4). The lesion was diagnosed as an angioleiomyoma and Mallory's PTAH staining was done.

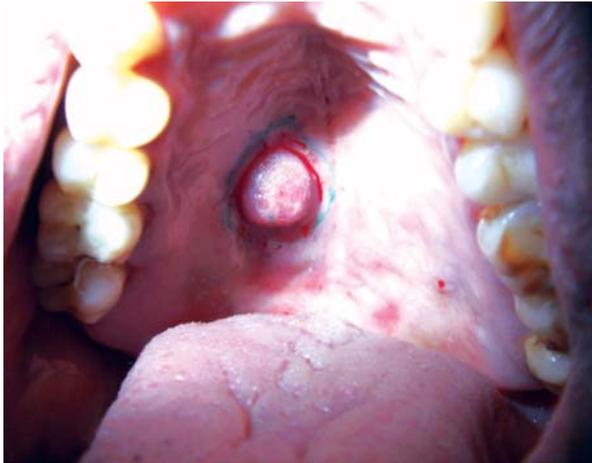


Fig. 1: Clinical picture showing a palatal swelling

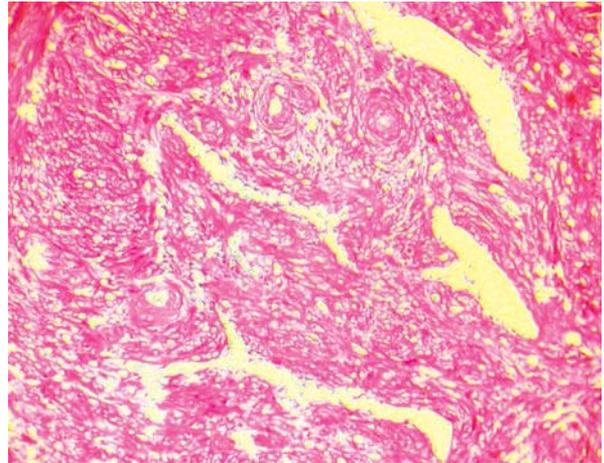


Fig. 4: Photomicrograph showing arteries appearing as consumed by the spindle cells. H&E $\times 200$



Fig. 2: The cut surfaces of the lesion

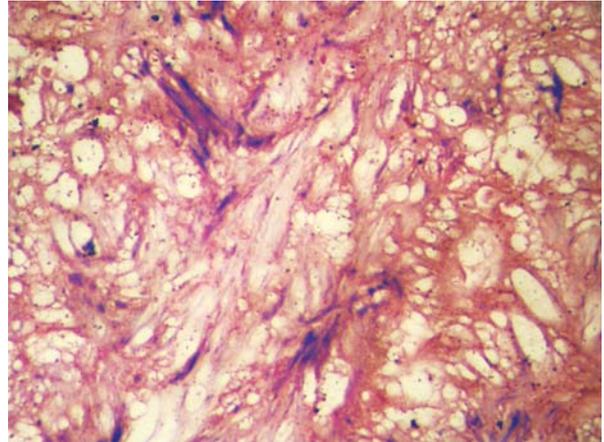


Fig. 5: Photomicrograph showing the cells with dark blue staining intracytoplasmic fibrils. Mallory's PTAH $\times 400$

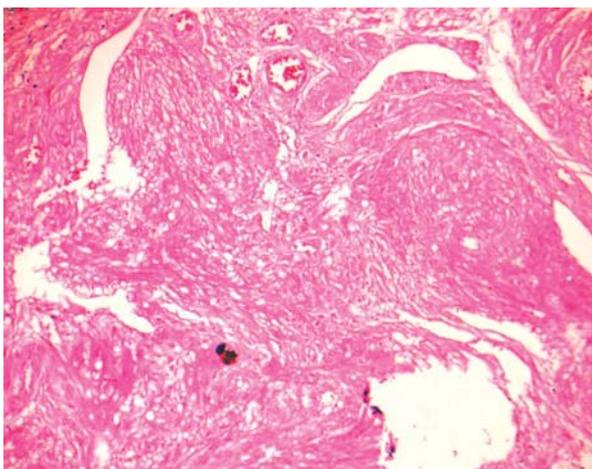


Fig. 3: Photomicrograph showing plump or spindle shaped cells in a streaming pattern. H&E $\times 200$

In Mallory's PTAH stained sections, the tumor cells showed distinct dark blue stained intracytoplasmic fibrillary extensions (Fig. 5).

DISCUSSION

Palatal swellings are a common presentation of a plethora of causes. The differential diagnoses for such swellings include periapical or periodontal conditions, reactive fibrous lesions, minor salivary gland conditions and mesenchymal lesions. Benign smooth muscle tumors are uncommon in the palate. The first case of soft tissue oral leiomyoma was reported by Blanc in 1884.³ Since then, there have been many reports of soft tissue oral leiomyoma in the literature.

The World Health Organization classifies leiomyoma histologically into three categories: Leiomyoma (solid

leiomyoma), angiomyoma (vascular leiomyoma) and epithelioid leiomyoma (leiomyoblastoma).⁴

Leiomyomas are more frequently seen in women, except for those that occur in the oral cavity, which has a male predominance.^{1,5} These develop later in life usually between 4th and 6th decades of life as a solitary lesion.

Oral leiomyomas are also classified as vascular leiomyomas, solid pattern leiomyomas and epithelioid leiomyomas.⁴

The solid pattern leiomyoma is more often an unencapsulated lesion and a less well-defined mass. Histologically, the smooth muscle bundles here show an interweaving pattern and the stroma is variable.^{5,6} Occasionally, the collagenous stroma is so extensive that it can obscure the true nature of the lesions.⁴

Angioleiomyoma is a solitary form of leiomyoma that usually occurs in the connective tissue and composed of numerous smooth muscle bundles with thick-walled blood vessels and interspersed fibrous connective tissue. Stout designated them as vascular leiomyomas to contrast them with cutaneous leiomyomas that has thin-walled vessels.^{7,8} This variant of leiomyoma is most frequently seen in the oral cavity with 75% of all cases corresponding to this histological type. Since the pathogenesis is unclear, some consider them as vascular hamartomas wherein the smooth muscle cells proliferates within a hemangioma, which would produce an angioleiomyoma. This further proliferates toward a simple leiomyoma.⁹ Smooth muscle is sparse in the oral cavity, however this region is rich in blood vessels. Therefore, it has been proposed that the tunica media layer of the blood vessels may be the origin of oral vascular leiomyoma. Lips are the most common site followed by tongue, cheek, palate and gingiva. They can also arise from the excretory ducts of salivary gland.

The angioleiomyomas have a characteristic histological appearance with abundant thick-walled muscular vascular channels with intervening fibrous or occasionally myxoid connective tissue.¹⁰ It is characterized by a well-localized proliferation of mesenchymal cells that are fusiform in shape with eosinophilic cytoplasm and elongated basophilic nuclei that show tapered ends. The centrally located nuclei develop deep indentations during contraction. The cells are arranged in fascicles in which the nuclei are staggered.

The vascular spaces lined by a single layer of endothelial cells are a constant feature in vascular leiomyoma. It is not uncommon to observe vascular leiomyomas with a heterogenic cell population, and two or more different histological patterns could be interlaced in the same lesion. In this particular case, fusiform and epithelial-like cell areas were observed.

In our case, we used Mallory's PTAH stain as against the use of Van Gilsan and Masson's trichrome stains to prevent false-positives.^{11,12}

CONCLUSIONS

Clinically, palatal swellings are not an uncommon presentation of various odontogenic, periodontal and reactive fibrous lesion, but are relatively rare in mesenchymal lesions. One such uncommon lesion is angioleiomyoma, a benign smooth muscle tumor. An angioleiomyoma of the oral cavity is a relatively rare one and here we present a case of angioleiomyoma of the palate, which presented itself as an innocuous painless swelling, and was diagnosed during routine oral examination. Angioleiomyoma should be included in the differential diagnosis of oral soft tissue lesions. Here, we have used Mallory's PTAH to visualize the intracytoplasmic myofibrils and to confirm our diagnosis.

Oral angioleiomyomas do not tend to recur following excision. In the case presented here, after 4 years and 2 months of follow-up, the patient has had no recurrence.

Long-standing leiomyomas can undergo malignant changes. Hence, all leiomyomas should always be viewed with caution, and follow-up is essential even though the benign nature of such lesions is well-documented.

ACKNOWLEDGMENTS

We would like to thank Mr Samuel Rathna Raju and Mrs Sunita S, Laboratory technicians, Krishnadevaraya College of Dental Sciences, Bengaluru for their technical work.

REFERENCES

1. Farman AG, Kay S. Oral leiomyosarcomas: Report of a case and review of the literature pertaining to smooth muscle tumours of the oral cavity. *Oral Surg* 1977;43:402-09.
2. Blanc E. Travaux originaux, *Gaz Hebd Med Chir* 1884;21: 611-13.
3. Baden E, Doyle JL, Lederman DA. Leiomyoma of the oral cavity: A light microscopic and immunohistochemical study with review of the literature from 1884 to 1992. *Eur J Cancer B Oral Oncol* 1994;30B:1-7.
4. Enzinger FM, Lattes R, Torloni H. Histological typing of soft tissue tumours, Geneva, World Health Organization 1969; 30-31.
5. Cherrick HM, Dunlap CL, King OH. Leiomyomas of the oral cavity: Review of the literature and clinicopathologic study of seven new cases. *Oral Surg* 1973;35:54-66.
6. Napier SS, Devine JC, Rennie JS, et al. Unusual leiomyomatous hamartoma of the hard palate: A case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1996;82:305-07.
7. Stout AP. Solitary cutaneous and subcutaneous leiomyomas. *Am J Cancer* 1937;29:435-69.
8. Ide F, Mishima K, Yamada H, Saito I, Horie N, Shimoyama T, et al. Perivascular myoid tumors of the oral region: A clinicopathologic reevaluation of 35 cases. *J Oral Pathol Med* 2008;37:43-49.

9. Duhig JT, Ayer JP. Vascular leiomyomas: A study of sixty-one cases. *Arch Pathol Lab Med* 1959;68:424-30.
10. Savage NW, Adkins KF, Young WG, Chapman PJ. Oral vascular leiomyoma: Review of the literature and report of two cases. *Aust Dent J* 1983;283:46-51.
11. Damm DD, Neville BW. Oral leiomyomas. *Oral Surg, Oral Med, Oral Pathol* 1979;47:343-48.
12. Chang JY, Kessler H. Masson trichrome stain helps differentiate myofibroma from smooth muscle lesions in the head and neck region. *J Formos Med Assoc* 2008;107:767-73.