

## ANGIOMYOMA OF THE HARD PALATE - REPORT OF A RARE CASE

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### Abstract

Angiomyoma is defined as a painful, benign subcutaneous or deep dermal tumor composed of mature smooth muscle bundles that are surrounded and interlaced by vascular channels. Smooth muscle neoplasms can be classified into solid leiomyomas, angioleiomyomas (vascular leiomyomas) and epithelioid leiomyomas (leiomyoblastomas). Among these types, the vascular leiomyoma is the most common subtype in the oral cavity. It may appear at any age with the greatest incidence in the 4th and 5th decades of life. The common manifestation is a slow-growing, asymptomatic, submucosal mass. The diagnosis is only possible through histopathological examination requiring special staining. The treatment of choice is the surgical excision and no recurrence is usually seen, though malignant transformation of these tumors has been reported. Hence a thorough examination, knowledge and follow-up must be warranted in this soft tissue tumor. In this article, we present a rare case of an angiomyoma in the right side of the hard palate with description of its clinical, histological and immunohistochemical characteristics.

Keywords: Angiomyoma - hard palate - smooth muscle tumor - SMA.

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## ANGIOMYOME DU PALAIS DUR – À PROPOS D'UN CAS RARE

### Résumé

L'angiomyome est défini comme étant une tumeur bénigne douloureuse, sous-cutanée ou profonde, composée de faisceaux musculaires lisses matures qui sont entourés et entrelacés par des canaux vasculaires. Les néoplasmes du muscle lisse peuvent être classés en léiomyomes solides, en angioléiomyomes (léiomyomes vasculaires) et en léiomyomes épithélioïdes (léiomyoblastomes). Parmi ces types, le léiomyome vasculaire est le sous-type le plus observé au niveau de la cavité buccale. Il peut apparaître à tout âge avec la plus grande incidence dans la 4ème et la 5ème décennie de la vie. La manifestation commune est une masse sous-muqueuse, asymptomatique, à croissance lente. Le diagnostic n'est possible que grâce à l'examen histopathologique nécessitant une coloration spéciale. Le traitement de choix est l'exérèse chirurgicale; aucune récurrence n'est généralement considérée, bien que la transformation maligne de ces tumeurs ait été décrite. Ainsi, un examen approfondi, la connaissance et le suivi doivent être de règle lors de la prise en charge de cette tumeur des tissus mous. Dans cet article, nous présentons un cas rare d'un angiomyome survenant du côté droit de la voûte palatine avec la description de ses caractéristiques clinique, histologique et immunohistochimique.

Mots-clés: angiomyome – tumeur du muscle lisse.

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## Introduction

Angiomyomas are composed of multiple vessels that are surrounded by thickened smooth muscle layer [1]. It has been proposed that pericyte, a mesenchymal-like cell associated with the walls of small blood vessels play an important role in the pathogenesis of this tumor [2]. The most frequent site of predilection of angiomyoma is the skin of lower extremities; this tumor is rarely found in the oral cavity [1, 2]. Lips, palate, buccal mucosa and tongue are the reported sites of its occurrence in the oral region.

Tumors occur in middle and older ages and are somewhat more frequent in men. Since only a few series of leiomyomas of the head and neck have been reported in the literature, the gender prevalence cannot be confirmed. Some of these intraoral tumors have shown local recurrences [3]. Since only a few cases of intraoral angiomyoma have been reported, the exact clinical behavior and malignant transformation of intraoral angiomyoma still remain uncertain. Hence, in this article we report a case of angiomyoma of the hard palate.

## Case presentation

A 57 years-old female patient presented with a chief complaint of swelling in relation to the right palatal alveolar region since seven months. The patient had no relevant medical history. On examination, a pinkish red well-defined swelling, of size 2 x 2 cm in diameter was seen in relation to the right palatal alveolar region from the distal aspect of tooth # 14 to the mesial aspect of tooth # 16 (Fig. 1). The swelling was soft in consistency and non-tender on palpation. The underlying bone showed no evidence of erosion on radiographic examination (Fig. 2). The lesion was provisionally diagnosed as a minor salivary gland adenoma. The differential diagnosis considered was pleomorphic adenoma, mucoepidermoid carcinoma and adenoid cystic carcinoma. In order to rule out these salivary gland malignancies



Fig. 1: Swelling in the right side of the hard palate.



Fig. 2: The occlusal x-ray showed no evidence of erosion.

and to plan the further management, an incisional biopsy was performed. As a pre-surgical protocol, the patient underwent routine blood examination like blood sugar, HB, TC, BT prior to biopsy and the results were within the normal limits. Histopathologically, proliferation of numerous thickened blood vessels was observed with an endothelial cell lining. Smooth muscle cells of the blood vessels were arranged in orderly circumferential fashion. No evidence of malignancy such as mitosis, necrosis, cellular atypia and pleomorphism were evident (Fig. 3). Van-Geison staining was positive at the smooth muscle layer showing the muscular nature of the lesion (Fig. 4). The immunohistochemical study on the tumor showed that the cells were positive for smooth muscle actin (SMA) (Fig. 5).

Based on the histopathological and the immune-histochemical findings, the lesion was diagnosed as an angiomyoma. The patient went to a tertiary health care center for further treatment which mainly consisted of complete surgical excision of the tumor and further follow-up.

## Discussion

Angiomyomas are leiomyomas of vascular smooth muscle origin and

are rare in the oral cavity [4]. Oral leiomyomas were usually seen in the 4th decade of life with equal sex prevalence [5]. The most common site of presentation was the lip (27%) with the least frequent presentation seen in the mandible.

Angiomyomas are benign smooth muscle neoplasms classified into leiomyomas (solid leiomyomas), angiomyomas (vascular leiomyomas), and epitheloid leiomyoma (leiomyoblastoma) [6]. A rare variant of angiomyomas known as myxoid angiomyoma has also been reported by Holder et al. [7].

Oral leiomyomas are believed to be derived from vascular smooth muscle [4, 8] and the division between angiomyoma and leiomyoma is based on the degree of vascularity. Pericytes which line the walls of the blood vessels have been found to play an important role in angiomyomas. Pericytes have a lineage to get differentiated into smooth muscle cells which emphasizes their role in the pathogenesis of this tumor. Pericytes can represent an intermediate phenotype between fibroblasts and vascular smooth muscle cells (VSMC). Various stimuli can induce the differentiation of the pericytes into VSMC. Therefore, pericytes are said to be progenitors for VSMC in angiomyomas [9].

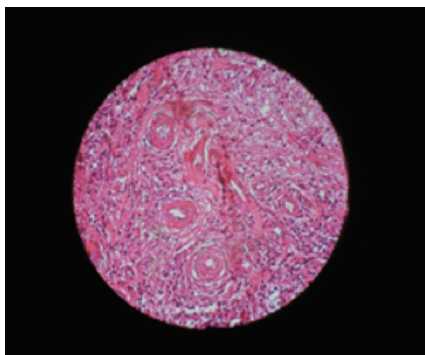


Fig 3: Numerous vascular channels with smooth muscles arranged in orderly circumferential fashion. (H and E staining; 10x magnification).

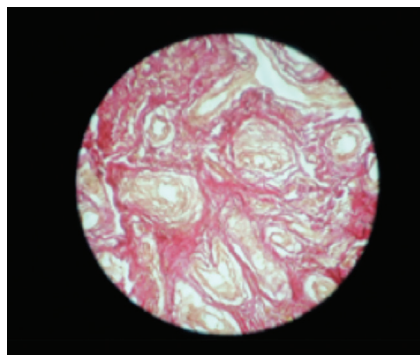


Fig. 4: Van Geison staining positivity for smooth muscle layer surrounding the vascular channels (10x magnification).

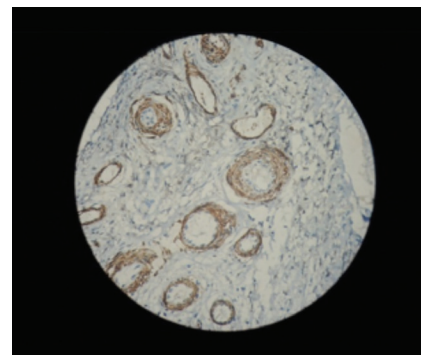


Fig. 5: Immunohistochemistry showing increase in SMA positive cells in the perivascular area.

In our case, we used the immunohistochemical marker smooth muscle actin (SMA) and the tumor cells were found to be positive to SMA. Van Geison special staining was done to confirm the presence of vascular smooth muscles in the tumor.

Oral angiomyomas are usually surgically excised; recurrence is extremely rare. Few cases of malignant transformation of angiomyoma have been reported [10].

## Conclusion

Intraoral angiomyoma is a rare entity. Even though it is often diagnosed clinically as a benign lesion, malignant transformation has been reported. Hence, the clinician should be aware of pathological features and behavior of this lesion. He should take into account the differential diagnosis of tumors and similar lesions in the soft tissues of the oral cavity and perioral

region in order to ensure prompt diagnosis, appropriate management and follow-up for these patients. Since very few cases of intraoral angiomyoma are reported, the exact biological behavior of lesion is yet to be ascertained.

## References

1. Al-Amad SH, Angel C, O'Grady JF, McCullough MJ. Angioleiomyoma on the hard palate. *Oral Oncol Extra* 2006;42:244-6.
2. Nehls V, Denzer K, Drenckhahn D. Pericyte involvement in capillary sprouting during angiogenesis in situ. *Cell Tissue Res* 1992;270(3):469-74.
3. Hung KF, Yan AH, Kao SY, Chang CS. Angioleiomyoma in right lingual gingiva. A case report. *Chin J Oral Maxillofac Surg* 2005;16:179-87.
4. Gonzalez Sanchez MA, Colorado BM, Berini AL, Gay EC. Leiomyoma of the hard palate: a case report. *Med Oral Patol Oral Cir Bucal* 2007;12(3):E221-E224.
5. Leung KW, Wong DY, Li WY. Oral leiomyoma: case report. *J Oral Maxillofac Surg* 199;48(7):735-8.
6. 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;30:1-7.
7. Holder R Jr, Dellinger TM, Krolls SO, Hill WJ, Livingston HM, Alemar GO. Myxoid angiomyoma of the hard palate: A case report. *Ear Nose Throat J* 2001;80:872-5.
8. Lloria-Benet M, Bagán JV, Lloria de Miguel E, Borja-Morant A, Alonso S. Oral leiomyoma: a case report. *Med Oral* 2003 May-Jul;8(3):215-9.
9. Gerhardt H, Betsholtz C. Endothelial-pericyte interactions in angiogenesis. *Cell Tissue Res* 2003;314(1):15-23.
10. Brooks J, Nikitakis N, Goodman N, Levy B. Clinicopathologic characterization of oral angioleiomyomas. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology* 2002;2:221-227.