

Case Report

Angioleiomyoma in the Retromolar Trigone Region

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Abstract

Leiomyoma is a benign soft tumor of smooth muscle origin which is rarely found in the oral cavity. The diagnosis of leiomyoma is fundamentally based on its histopathologic characteristics requiring specific staining methods and careful microscopic analysis due to the high possibility of misdiagnosis. The present paper describes a case report of recurrent oral vascular leiomyoma in the retromolar trigone region and performs a brief literature review. Specific staining methods were performed such as Masson's trichrome and immunohistochemistry for smooth muscle actin. Prognosis of oral leiomyoma is excellent.

Keywords: Leiomyoma. Mouth neoplasms.

Introduction

Leiomyoma is a benign tumor originating in smooth muscle tissue, commonly found in the uterus, gastrointestinal tract and skin. It rarely occurs in the oral cavity (0.0065%) due to the limited amount of this type of musculature in the region.¹ According to the literature, its most frequent location in the oral cavity is the lips, palate, tongue and jugal mucosa.²⁻⁴ It may appear at any age, generally presenting as an asymptomatic, occasionally painful, slow-growth mass. The diagnosis of leiomyoma is fundamentally based on its histopathologic characteristics, requiring specific stains to differentiate it from other tumor lesions.^{1,5}

According to Barnes et al.,⁶ there are three histological subtypes for leiomyoma: vascular leiomyoma (angioleiomyoma), solid leiomyoma and epithelioid leiomyoma (leiomyoblastoma). The former is the most frequent in the oral cavity, with a prevalence of 74 percent.

The aim of the present study is to report a case of recurrent vascular leiomyoma in the retromolar trigone region and perform a brief literature review.

Case Report

A 32-year-old female patient sought the oromaxillofacial surgery service due to a three-and-a-half year history of an increase in volume in the retromolar region. During the patient history, she reported having undergone the removal of an identical lesion four years earlier. Upon physical examination, an asymptomatic tumor of approximately 1.0X1.5cm was observed in the retromolar trigone region. The lesion was movable and firm to the touch, with coloration resembling the adjacent mucosa. The initial diagnostic hypothesis was adenoma. Surgical removal with a safety margin was then proposed. The specimen was fixed in 10% formaldehyde and sent for anatomopathologic examination.

For microscopic examination, the pieces were stained with HE, which evidenced strands of dense fibrous cellularized conjunctive tissue, streaked with small,

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twisting blood vessels and the presence of a mild, chronic inflammatory infiltrate. Strands of smooth muscle formed a thick, circumscribed structure, bordered by transversal cuts also of muscle tissue. Muscle fibers at times took on an undulated appearance, organized in strands that intermingled with strands of collagen fibers, hindering adequate delineation between the stroma and tumor parenchyma. The lesion was well defined (Figures 1 and 2) and neoplasm cells presented no characteristics of malignancy.

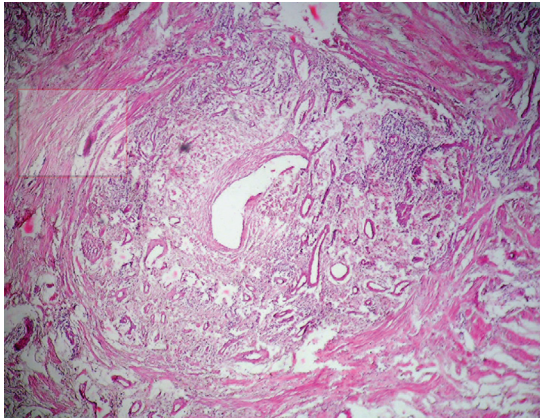


Figure 1 - Photomicrography of the lesion. Strands of smooth muscle form a circumscribed structure, bordered by transversal cuts also of muscle tissue. The muscle fibers are organized in strands that intermingled with collagen fibers. The lesion was well defined and the neoplasm cells presented no characteristics of malignancy (H.E. 40X).

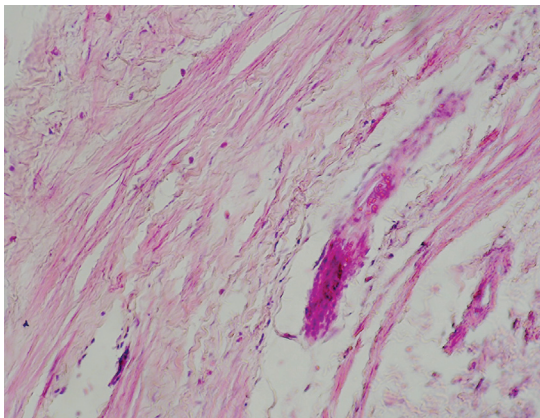


Figure 2 - Higher magnification from the selected picture of the figure 1. It shows the muscle fibers on an undulated appearance forming a circumscribing structure (H.E. 200X).

To allow a better morphological evaluation of the specimen, histological cuts were stained with Masson's trichrome stain and revealed fragments of benign neoplasm tissue originating in vascular smooth muscle, in which the red coloration referred to the muscle layer and the blue coloration referred to collagen fibers). Figures 3 and 4 show the results of Masson's trichrome staining. Immunohistochemical examination was performed for the smooth muscle anti-actin antibody. Neoplasm cells exhibited a strong, positive reaction, which confirmed the diagnosis of leiomyoma (Figures 5 and 6).

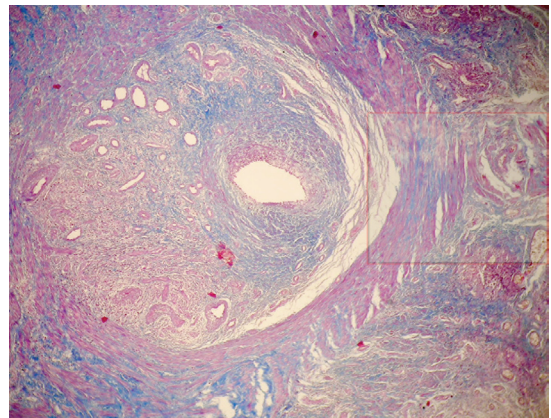


Figure 3 - Photomicrography exhibiting the neoplasm aspect with intermingling smooth muscle cells in red and collagen fibers in blue (Masson's trichrome, 40X).

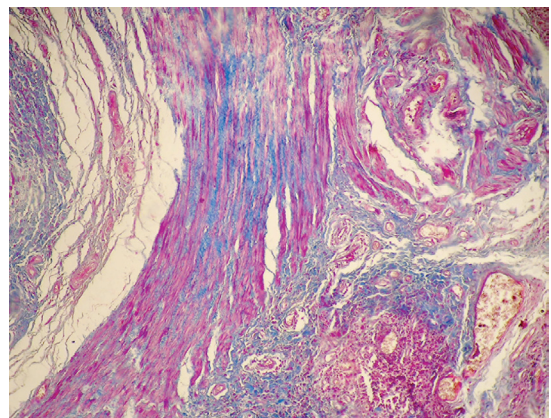


Figure 4 - Higher magnification of the selected picture in figure 3 (Masson's trichrome, 200X).

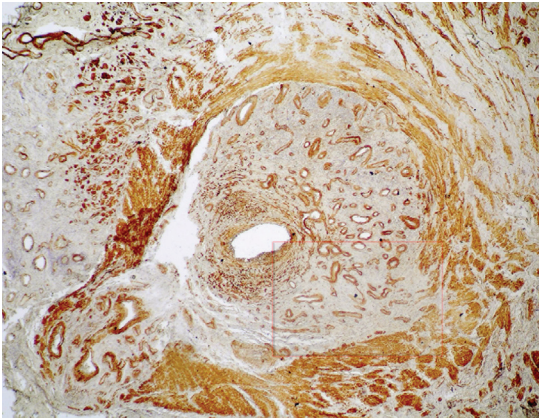


Figure 5 – Immunohistochemical study showing positivity for smooth muscle actin (40X).

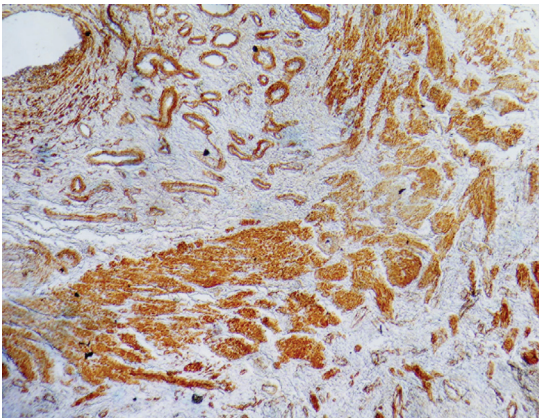


Figure 6 – Higher magnification of figure 5 (100X).

Discussion

The first case of oral leiomyoma oral was described in 1884. Other cases have since been added to the literature.¹ Table 1 displays cases of oral cavity leiomyoma reported in the international literature, partially obtained from a survey on Medline (www.pubmed.com) and Lilacs (www.bireme.br) databases and partially obtained from older, non-indexed case reports.

There was no preference regarding gender in the literature from 1969 to 2007 and adults were more frequently affected. However, Baden et al.⁷ state that the oral leiomyomas studied in their series occurred more commonly in men (54%), on average affecting individuals between 39 and 41 years of age.

The classic literature describes the lips as the

Table 1 - Cases of oral cavity leiomyoma reported in the international literature.

Author	Year	N	Location
McGowan, Jones ⁸	1969	1	Lower lip
Cherrick et al. ⁹	1973	7	Oral cavity*
Gutmann et al. ¹⁰	1974	2	Hard palate/ lower lip
Farman ¹¹	1975	5	Oral cavity*
Damm and Neville ¹²	1979	3	Oral cavity*
Davis ¹³	1980	1	Palate
Hemani et al. ¹⁴	1983	1	Palate
Hachisuga et al. ¹⁵	1984	15	Oral cavity*
Svane et al. ¹⁶	1985	1	Hard Palate
Birman et al. ¹⁷	1986	2	Upper lip
Baden et al. ⁷	1994	3	Oral cavity*
Burkes ¹⁸	1995	1	Mandible/ intraosseous Tongue
Álvarez et al. ³	2001	1	Palate
Holder et al. ¹⁹	2001	1	Palate
Brooks et al. ²⁰	2002	12	Oral cavity*
Reis et al. ²¹	2002	1	Tongue
Lloria-Benet et al. ¹	2003	1	Jugal mucosa
Liang et al. ²²	2003	1	Mandible/ intraosseous Palate
Pahwa et al. ²³	2004	1	Lip
Pitukkirojronnakorn et al. ²⁴	2005	1	Tongue
Félix et al. ²	2006	1	Oral cavity*
Bhattacharyya et al. ²⁵	2006	2	Mandible/ intraosseous Retromolar trigone
Suresh et al. ²⁶	2007	1	Palate
Rey et al. ⁵	2007	1	
Sánchez et al. ²⁷	2007	1	

*location not informed

most common location. However, the authors studied identified a preference for the palate, followed by the tongue. The scarcity of detailed data in the articles used for this review, especially in the series reports, renders impossible the determination of a preferential location for the lesion.

The case presented here did not show any relevant symptomatology. The patient presented discomfort caused by the increase in lesion volume, but without pain. The literature describes oral leiomyoma as an asymptomatic entity of slow growth,^{1,4,8} although Felix et al.³ report a case of painful leiomyoma in the tongue.

Stout²⁸ states that due to the limited amount of smooth muscle in the oral cavity, it likely originates in the walls of small blood vessels/arterioles. Glas²⁹ cites the embryological remains of the lingual duct and Jaeger et al.³⁰ cite undifferentiated mesenchymal cells as probable factors involved in the etiology.

Although the World Health Organization, Enzinger et al.³¹ classifies leiomyoma into three histological subtypes, Holder et al.¹⁹ report an angiomyoma subtype with myxoid degeneration. Moreover, Bhattacharyya et al.²⁵ documented a histological variation of granular cell

leiomyoma.

According to the majority of authors in the present review, the most common histological type is angioleiomyoma. According to Duhing e Ayer,³² angioleiomyoma represents a progressive maturation that begins as hemangioma and evolves into a solid leiomyoma through cell proliferation of the smooth musculature of hemangioma vessels.

Tumor cells are normally fusiform with a nucleus that has coarse margins, which commonly involves blood vessels forming concentric layers in the perivascular region. A fibrillary stroma may be mistaken as collagen. Therefore, Masson's trichrome stain is an appropriate method of differentiation, as it stains muscle fibers and collagen differently, as can be seen in Figures 3 and 4.

These histopathologic aspects may hamper the differential diagnosis with other lesions of mesenchymal origin^{1,6,32} such as fibroma, neurofibroma, schwannoma and low-grade leiomyosarcoma. For such cases, immunohistochemical reactions are indispensable; leiomyoma is positive for smooth muscle actin, specific muscle actin, HHF 35, desmin and myoglobin, whereas it is negative for cytokeratin AE1/AE3 and S100 7.³³

In a study by Baden et al.⁷ on six cases initially diagnosed by HE alone, later immunohistochemical analysis only confirmed the diagnosis of leiomyoma in three cases. Bhattacharyya et al.²⁵ found that immunohistochemical analysis was essential in differentiating leiomyoma from a squamous-cell tumor. In the immunohistochemical analysis of the case reported herein, the specimen demonstrated an intense positive reaction to smooth muscle actin, principally in neoplasm cells circumjacent to blood vessel walls. Such a finding was essential to the final diagnosis of the lesion, as the arrangement of collagen and muscle fibers very often take on a fusiform or spiral shape, suggesting a lesion of neural origin when based solely on routine staining.

Anatomopathologic analysis of such soft tissue lesions associated to special staining methods is a diagnostic resource of considerable importance, as this lesion clinically requires a differential diagnosis to distinguish it from a number of other lesions, including fibroma, lipoma, salivary gland neoplasm, vascular tumors such as lymphangioma or hemangioma, as well as malignant pathologies such as leiomyosarcoma.²⁷ As with the present case report, Rey et al.⁵ suggested adenoma - a pathology originating in the lesser salivary glands - as the primary diagnostic hypothesis.

The first treatment option for leiomyoma is surgical excision with a safety margin.²¹ According to Rey et al.,⁵ cases of recurrence have been reported, despite not being expected. Recurrence is due to incomplete removal, which likely occurred in the case described herein.

Recurrence may also occur in cases of leiomyosarcoma with a low degree of malignity mistakenly diagnosed as its corresponding benign tumor.

Conclusion

Leiomyoma originating in the oral cavity is rare and requires careful microscopic analysis due to the high possibility of misdiagnosis. Special staining methods are indispensable for favoring a reliable diagnosis and consequently lead to an adequate treatment planning. The aim of the present study is to contribute to the understanding of this pathologic entity for which there is yet little discussion in the literature.

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