Jornal Brasileiro de Patologia e Medicina Laboratorial

ISSN: 1676-2444

jbpml@sbpc.org.br,adagmar.andriolo@gmail.com

Sociedade Brasileira de Patologia Clínica/Medicina Laboratorial

Galvão Barboza, Carlos Augusto; Ginani, Fernanda; Vieira Leite-Segundo, Airton; da Silva, Uoston Holder

Oral angioleiomyoma: a case report and considerations on differential diagnosis

Jornal Brasileiro de Patologia e Medicina Laboratorial, vol. 49, núm. 6, diciembre, 2013, pp. 429-432

Sociedade Brasileira de Patologia Clínica/Medicina Laboratorial

Rio de Janeiro, Brasil

Available in: http://www.redalyc.org/articulo.oa?id=393541979008

J Bras Patol Med Lab, v. 49, n. 6, p. 429-432, dezembro 2013

Oral angioleiomyoma: a case report and considerations on differential diagnosis

Angioleiomioma oral: relato de caso e considerações no diagnóstico diferencial

Carlos Augusto Galvão Barboza[1]; Fernanda Ginani[2]; Airton Vieira Leite-Segundo[3]; Uoston Holder da Silva[4]

ABSTRACT

Angioleiomyomas are benign mesenchymal tumors derived from smooth muscle, which rarely occur in the oral cavity. We report a case of an angioleiomyoma occurring in the maxillary gingiva. The lesion was painless, with a lobulated surface, fibrous in consistency, and firm upon palpation. Microscopic examination showed an encapsulated tumor mass composed of large vascular channels of varying calibers, surrounded by thick walls of irregularly arranged, spindle-shaped cells, which were immunopositive for smooth-muscle actin. It is sometimes difficult to differentiate an angioleiomyoma from other spindle-cell tumors, thus we emphasize its histological differential diagnosis.

Key words: oral mucosa; neoplasms; angioleiomyoma; maxilla.

INTRODUCTION

Leiomyomas are benign mesenchymal tumors derived from smooth muscle, which most often occur in the uterus and gastrointestinal tract. According to the World Health Organization[5], leiomyomas are divided into two categories: angioleiomyoma and leiomyoma of deep soft tissue. Angioleiomyomas are solitary forms of leiomyomas that usually occur in the subcutis and are the most common variant of leiomyomas affecting the oral cavity[12].

From a clinical point of view, angioleiomyomas are usually superficial, well-circumscribed and asymptomatic small lesions with slow growth, covered by normal mucosa, and sometimes bluish or purple in color[7, 9]. The most common oral sites for angioleiomyomas are the lips (27.46%), followed by the tongue (18.30%), cheeks and palate (15.49%), gingiva (8.45%), and mandible (5.63%)[2]. The greatest incidence is in the 40-59-year age group[4, 15]. We report a case of an angioleiomyoma occurring in the maxillary gingiva with clinical characteristics of a peripheral giant cell granuloma.

CASE REPORT

A 43-year-old man was referred to the Department of Stomatology of the Caruaru School of Dentistry for diagnosis and treatment of an exophytic asymptomatic lesion of the upper gingiva. The lesion appeared red-to-violet in color with a lobulated surface and arose in the gingiva between the maxillary right cuspid and the left central incisor from a sessile base (**Figure 1**). The lesion was painless, measured approximately 1.5 × 1 cm in diameter, fibrous in consistency, and firm upon palpation.

The lesion had been first noticed by the patient 6 months earlier after a tooth extraction, and it had grown slowly to its present size. The patient's oral hygiene was poor. Furthermore, generalized periodontitis and advanced attrition were diagnosed. The medical history was essentially noncontributory and the patient was taking no medications. A periapical radiograph revealed a well-defined radiolucent area between the 11 and the 23, suggesting mild osseous erosion consistent with cupping of the underling bone (**Figure 2**). The clinical differential diagnosis included a peripheral giant cell granuloma and a benign

First submission on 11/04/13; last submission on 09/08/13; accepted for publication on 11/08/13; published on 20/12/13

1. PhD in Oral Pathology; associate professor, Postgraduate Program in Oral Pathology, Universidade Federal do Rio Grande do Norte (UFRN), Brazil.
2. MSc in Dentistry; PhD candidate-Postgraduate Program in Oral Pathology, UFRN, Brazil.
3. PhD in Stomatology-Oral and Maxillofacial Surgeon, Hospital Regional do Agreste, Brazil.
4. PhD in Dentistry; professor and Head, Caruaru Dental School, Brazil.

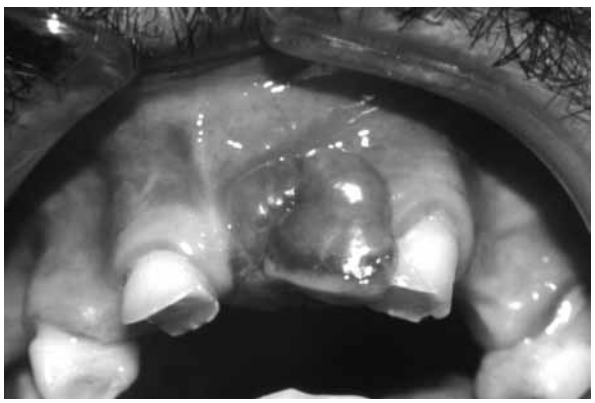

FIGURE 1 – *Intraoral view showing a violet, lobulated, sessile, and well-circumscribed nodule in the upper gingiva*

mesenchymal neoplasm. Surgical excision of the lesion was performed, and moderate bleeding was noted and controlled with pressure on the area.

Microscopic examination showed a tumor mass encapsulated by a fibrous tissue of varying texture and thickness, and covered by a normal keratinizing stratified squamous epithelium. The lesion was composed of large vascular channels of varying caliber, surrounded by thick walls of irregularly arranged, spindle-shaped cells (**Figure 3A**). The tumor cells were large, with indistinct cell margins, scant and faintly eosinophilic cytoplasm as well as elongated nuclei with rounded edges (**Figure 3B**). Whorled bundles of muscle fibers were sometimes fused with the vessel walls. Positive immunostaining for smooth-muscle actin and desmin were observed in the tumor cells (**Figures 3C** and **E**), whereas CD-34 was positive in the endothelium (**Figure 3D**). S-100 staining was negative in the tumor cells (**Figure 3F**).

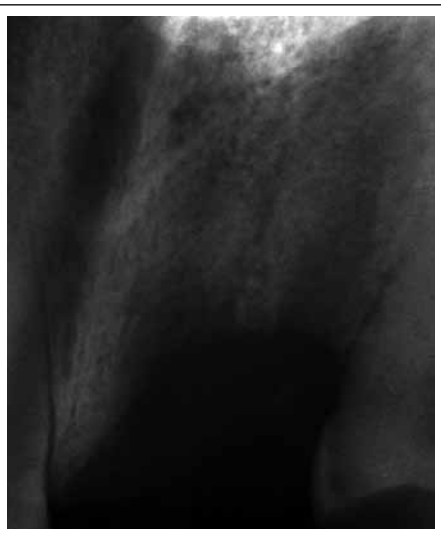

FIGURE 2 – *Periapical radiograph revealing a well-delineated radiolucent area suggestive of bone erosion*

A diagnosis of angioleiomyoma was established. No complications were observed after a one-month follow-up (**Figure 4**) and there was no recurrence after a 14-month follow-up.

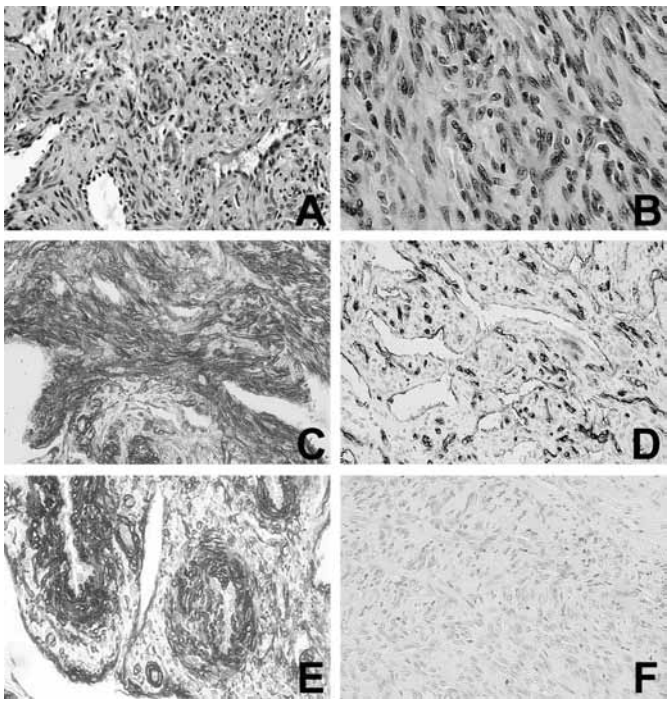

FIGURE 3 – *Microscopic features of the angioleiomyoma*

A: smooth muscle proliferation around slit-like endothelium-lined vascular spaces (hematoxylin-eosin; original magnification 100×); B: large tumor cells with indistinct cell margins and faintly eosinophilic cytoplasm, showing elongated nuclei with rounded edges (hematoxylin-eosin; original magnification 200×); C: immunostaining for smooth-muscle actin showing strong positivity in the proliferating spindle-shaped cells, which form thick vessel walls (streptavidin-biotin technique; original magnification 100×); D: CD34 immunoreactivity is observed in the endothelial cells, demonstrating the high grade of vascularization of the lesion (streptavidin-biotin technique; original magnification 100×); E: neoplastic cells with positive immunostaining for desmin (streptavidin-biotin technique; original magnification 100×); F: neoplastic cells negative for S-100 (streptavidin-biotin technique; original magnification 100×).

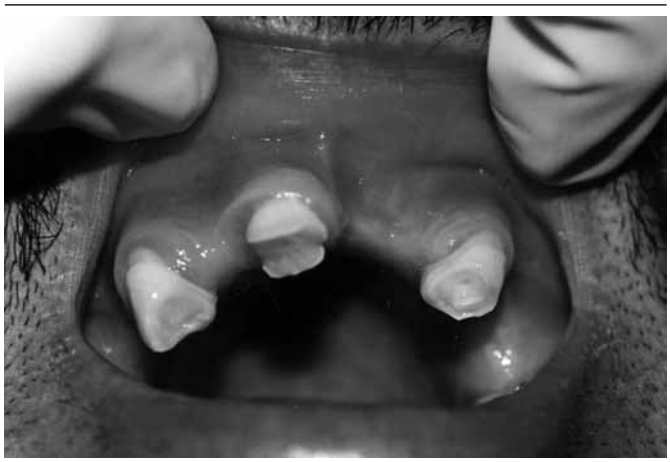

FIGURE 4 – *Intraoral view 30 days after the surgical resection of the lesion*

DISCUSSION

Angioleiomyomas are rare tumors in the oral cavity, representing only 0.016% to 0.065% of all leiomyomas[3, 8, 16], due to the paucity of smooth muscle in this region. Most authors agree that the tumor arises from the tunica media of small vessels, but other sources have been suggested, such as the smooth muscle of the circumvallate papilla of the tongue, arteriovenous anastomoses, and from ectopic smooth muscle associated with aberrant hair follicles, particularly in the cheek[1, 7, 12, 13, 15]. We believe that the gingival angioleiomyoma probably originated from the smooth muscle of blood vessels.

When found in the oral mucosa, angioleiomyomas most commonly affect the lips, followed by the tongue, cheeks, and palate[2]. Angioleiomyomas of the gingiva are very rare, accounting for only 0.9% of the oral angioleiomyomas reported in the English literature[3]. In the current case, a peripheral giant cell granuloma was most strongly suspected preoperatively because of the location, color, absence of symptoms, and progression of the lesion.

Histologically, we found a lesion composed of spindle-shaped cells presenting elongated nuclei with rounded ends, which were closely compacted with a fibrous stroma surrounding large vascular channels. These findings corroborated the reports in the literature regarding the typical microscopic appearance of an angioleiomyoma, which is a well-defined nodule of smooth muscle tissue punctuated with thick-walled vessels with a partially patent lumen[1, 3, 6, 13, 17-19]. Typically, the inner layers of smooth muscle of the vessels are arranged in an orderly circumferential fashion, whereas the outer layers spin or swirl away from the vessel, merging within the less well-ordered peripheral muscle fibers[19].

It is sometimes difficult to differentiate an angioleiomyoma from other spindle-cell tumors. Several oral tumors should be included in the histopathological differential diagnosis[16], namely schwannomas, neurofibromas, fibromatoses, hemangiopericytomas, hemangioendotheliomas, well-diferentiated leiomyosarcomas, fibrosarcomas, inflammatory myofibroblastic tumors, and malignant schwannomas[11]. To that end, immunohistochemistry is a valuable, precise, and reliable method for establishing a definitive diagnosis[14, 15].

In the present case, the tumor cells were positive for smooth muscle actin (SMA) and desmin, whereas the endothelium exhibited immunoreactivity for CD-34. S-100 protein was not identified in the tumor cells. Monoclonal antibody to SMA reacts with the muscle cells of vessels and it is useful for excluding a diagnosis of schwannoma, fibromatosis, fibrosarcoma, and malignant schwannoma since those tumor cells are negative for SMA. Hemangiopericytomas and hemangioendotheliomas are also negative for SMA inasmuch as these lesions do not have a smooth muscle component in their vessel walls[17], although their endothelium is generally positive for CD-34. This antibody is useful to define the degree of vascularization of the lesion.

Myopericytomas show histological and immunohistochemical overlap with angioleiomyomas, including SMA expression. However, only angioleiomyomas coexpress SMA and desmin, which reflects an advanced smooth muscle differentiation[10]. Leiomyoblasts are negative for S-100[13] and CD-34 antibodies. On the other hand, tumor cells of schwannomas, neurofibromas, and malignant schwannomas usually show strong staining for S-100[19]. The number of mitotic figures, anaplasia, and bizarre cell forms are regarded as features that may aid in the distinction between leiomyomas and low-grade leiomyosarcomas[15].

Angioleiomyomas are benign tumors that cause minor problems. Local surgical excision with an adequate margin of unaffected tissue is the suitable therapy[9, 19]. The postoperative prognosis is generally good and this tumor rarely recurs[1]. Only a few cases of recurrence are reported[3] and they are probably related to the incomplete resection of the lesion[9].

RESUMO

Angioleiomiomas são tumores mesenquimais benignos derivados de músculo liso que raramente ocorrem na cavidade oral. Relatamos um caso de angioleiomioma na gengiva maxilar. A lesão era indolor, com superfície lobulada, consistência fibrosa e firme à palpação. O exame histológico mostrou massa tumoral encapsulada, composta por grandes canais vasculares de diferentes calibres, circundados por paredes espessas de células fusiformes dispostas de forma irregular, as quais se apresentavam imunopositivas para actina de músculo liso. A distinção entre angioleiomioma e outros tumores de células fusiformes algumas vezes é difícil e, por essa razão, enfatizamos seu diagnóstico diferencial histológico.

Unitermos: *mucosa oral; neoplasias; angioleiomioma; maxila.*

REFERENCES

1. ANASTASSOV, G. E.; VAN DAMME, P. A. Angioleiomyoma of the upper lip: report of a case. *Int J Oral Maxillofac Surg*, v. 24, n. 1, p. 301-2, 1995.

2. BADEN, E.; DOYLE, J. L.; LEDERMAN, D. A. 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,* v. 30, n. 1, p. 1-7, 1994.

3. BROOKS, J. K. *et al*. Clinicopathologic characterization of oral angioleiomyomas. *Oral Surg Oral Med Oral Patho Oral Radiol Endod*, v. 94, n. 2, p. 221-7, 2002.

4. CEPEDA, L. A. G. *et al.* Vascular leiomyoma of the oral cavity. Clinical, histopathological and immunohistochemical characteristics. Presentation of five cases and review of the literature. *Med Oral Patol Oral Cir Bucal*, v. 13, n. 8, p. e483-8, 2008.

5. CHRISTOPHER, D. M. *et al*. *World Health Organization pathology and genetics of tumours of soft tissue and bone*. Lyon, France: International Agency for Research on Cancer (IARC), 2002.

6. EPIVATIANOS, A.; TRIGONIDIS, G.; PAPANAYOTOU, P. Vascular leiomyoma of the oral cavity. *J Oral Maxillofac Surg*, v. 43, n. 5, p. 377-82, 1985.

7. ESGUEP, A.; SOLAR, M. Oral vascular leiomyoma: report of 5 cases and review of the literature. *J Oral Med*, v. 41, n. 2, p. 126-33, 1986.

8. GONZÁLEZ-SÁNCHEZ, M. A. *et al*. Leiomyoma of the hard palate: a case report. *Med Oral Patol Oral Cir Bucal*, v. 12, n. 3, p. e221-4, 2007.

9. GROSSMANN, S. M. C. *et al.* Anterior midline nodule of the hard palate. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*, v. 108, n. 6, p. 808-11, 2009.

10. IDE, F. *et al*. Perivascular myoid tumors of the oral region: a clinicopathologic re-evaluation of 35 cases. *J Oral Pathol Med*, v. 37, n. 1, p. 43-9, 2008.

11. KIM, Y. H. *et al*. Congenital angiomyoma of the tongue: case report. *Dentomaxillofac Radiol*, v. 39, n. 7, p. 446-8, 2010.

12. LEUNG, K. W.; WONG, D.Y.; LI, W. Y. Oral leiomyoma: case report. *J Oral Maxillofac Surg*, v. 28, n. 7, p. 735-8, 1990.

13. MAEDA, Y. *et al*. Angiomyoma of the upper lip: report of a case with electron microscopic and immunohistochemical observation. *Br J Oral Maxillofac Surg*, v. 27, n. 3, p. 236-42, 1989.

14. NONAKA, C. F.; PEREIRA, K. M.; MIGUEL, M. C. Oral vascular leiomyoma with extensive calcification areas. *Braz J Otorhinolaryngol*, v. 76, n. 4, p. 539, 2010.

15. ORSINI, G. *et al*. Leiomyoma of the lip: report of a case. *J Oral Maxillofac Surg*, v. 59, n. 1, p. 80-3, 2001.

16. REY, R. *et al.* Oral leiomyoma in retromolar trigone. A case report. *Med Oral Patol Oral Cir Bucal*, v. 12, n. 1, p. e53-5, 2007.

17. SAVAGE, N. W. *et al*. Oral vascular leiomyoma: review of the literature and report of two cases. *Aust Dent J*, v. 28, n. 6, p. 346-51, 1983.

18. TOIDA, M.; KOIZUMI, H.; SHIMOKAWA, K. Painful angiomyoma of the oral cavity: report of a case and review of the literature. *J Oral Maxillofac Surg*, v. 58, n. 4, p. 450-3, 2000.

19. WEISS, S. W.; GOLDBLUM, J. R. *Enzinger and Weiss's soft tissue tumors*. St Louis: Mosby, 2001.

MAILING ADDRESS

Carlos Augusto Galvão Barboza

Universidade Federal do Rio Grande do Norte; Departamento de Morfologia; Centro de Biociências; Av. Salgado Filho, 3000; Campus Universitário; CEP: 59072-970; Natal-RN, Brazil; e-mail: cbarboza@cb.ufrn.br.