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CASE REPORT

Leiomyoma with intracytoplasmic inclusion bodies in the oral cavity: an uncommon histological finding

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Abstract:

Leiomyomas are benign tumors that arise from the smooth musculature and can occur in any part of the body where these muscles are present. However, leiomyomas of the oral cavity are rare because of the scarcity of smooth muscles at this site. We present a case of leiomyoma involving the tongue which exhibited a histological finding that is extremely rare in oral leiomyomas. Immunohistochemical analysis revealed positivity for smooth muscle actin (alfa - SMA, desmin) and negativity for S - 100, CD- 68 and CD34 protein. The main clinical and histopathological features of leiomyomas, as well as their differential diagnosis, were reviewed. Intracytoplasmic inclusion bodies have been found in uterine leiomyomas, in atypical (bizarre) leiomyomas and, occasionally, in epithelioid leiomyomas and leiomyosarcomas. However, a search of the main literature databases revealed no description of these inclusions in oral leiomyomas.

Keywords: Leiomyoma; Inclusion Bodies; Mouth

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INTRODUCTION

Leiomyomas are tumors of smooth muscle origin which are common in the uterus (95%), skin (3%) and gastrointestinal tract (1.5%). Less than 1% of these tumors are found in the head and neck region[1,2]. Leiomyomas of the oral cavity are rare, accounting for only 0.065% of all leiomyomas, since smooth muscle is scare at this site[1,3-5]. Oral leiomyomas have been shown to arise from vascular smooth muscle and from the excretory ducts of the salivary glands[3,6].

Clinically, oral leiomyoma appears as a solitary nodular mass found mainly in the tongue, lips, palate and cheek mucosa[7]. The tumor generally affects patients in their 4th and 5th decades of life, with a slight female predilection, and manifests as a slow-growing and asymptomatic lesion[2].

Leiomyoma is histologically characterized by spindle-shaped cells arranged in interwoven fascicles. The cells contain elongated or spindle-shaped, blunt-ended nuclei[8]. The World Health Organization (2005) defines three histological subtypes of leiomyoma: solid leiomyoma, angiomyoma (vascular leiomyoma), and the rare form of epithelioid leiomyoma (leioblastoma/atypical leiomyoma)[4,5].

Intracytoplasmic inclusion bodies, which consist of aggregates of intermediate filaments, are a known characteristic of different neoplasms. The types of these filaments vary in different lesions[9,10]. Aggregated actin filaments are typically seen in leiomyomas, these inclusions have been described in cases of leiomyoma of the urinary bladder, intracerebral leiomyoma, leiomyoma of the gastrointestinal tract, and atypical uterine leiomyoma[9-14]. However, these inclusions have not yet been described in leiomyomas of the oral cavity. Therefore, the objective of this study was to report a case of intraoral leiomyoma with intracytoplasmic inclusion bodies and to conduct a comprehensive literature review on the clinical and histopathological characteristics, differential diagnosis and treatment of this entity, comparing the data found with the present case.

CASE REPORT

A 35-year-old white woman sought the Specialized Dental Service with a lesion in the oral cavity. No alteration was identified upon extraoral examination. Intraoral examination revealed an exophytic, pedunculated, asymptomatic mass on the ventral surface of the tongue. The lesion had the same color as the surrounding mucosa, surface smooth, resilient consistency and measured approximately 0.5 cm. The patient reported a 2-year history of the mass. On the basis of the clinical findings, the diagnostic hypothesis was fibroma. An excisional biopsy of the lesion was thus performed. On gross examination, the lesion was firm and well-demarcated, with a whitish to reddish surface. Histopathological examination showed the presence of a benign mass of muscle origin characterized by intense proliferation of mainly spindle-shaped cells with blunt-ended nuclei arranged in interwoven bundles (Figure 1A,B). Furthermore, numerous cells containing intracytoplasmic inclusions were found scattered among the neoplastic cells. These cells exhibited abundant eosinophilic cytoplasm, excentric nuclei, and paranuclear intracytoplasmic inclusions, conferring a rhabdoid appearance (Figure 1B).

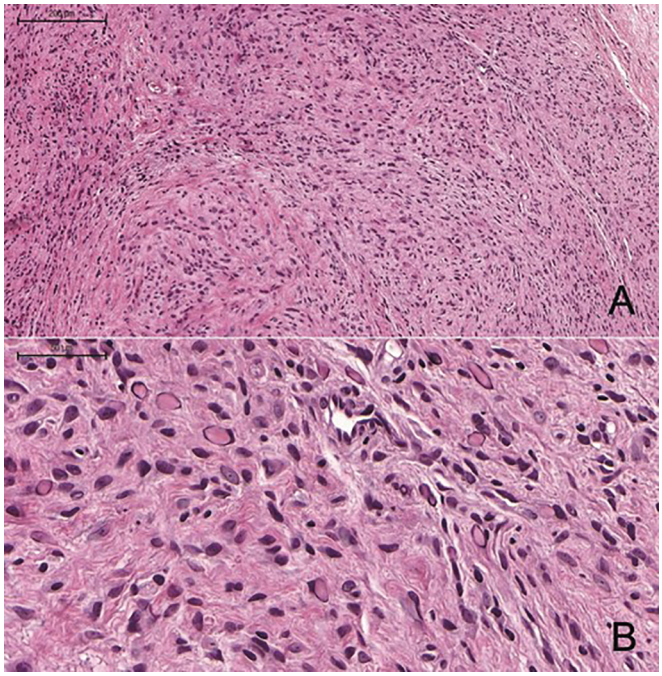

Figure 1. (A) Hematoxylin and eosin stained section demonstrating intense proliferation of spindle cells in the majority, with blunt ended nuclei arranged in interwoven bundles (slide view - 200 µm). (B) Higher magnification exhibits scattered among the neoplastic cells numerous cells with intracytoplasmic inclusions (slide view - 50 µm).

In view of the uncommon histopathological presentation for leiomyomas of the oral cavity, immunohistochemistry was performed with different markers for diagnostic confirmation. The results showed intense positive cellular staining for smooth muscle actin (α-SMA) and desmin (Figure 2A,B,C); negative staining for protein S-100, CD68 and CD34 (Figure 3A,B,C)

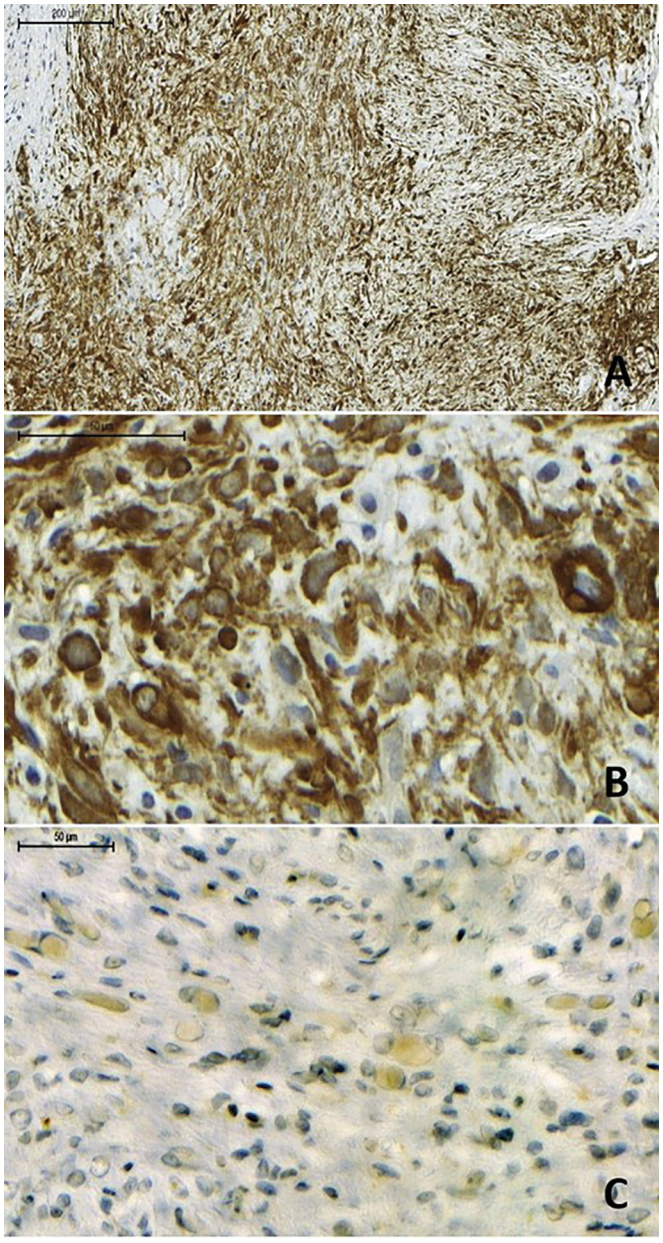

Figure 2. (A) Immunohistochemical expression of smooth muscle actin in tumor cells confirm¬ing the muscle origin (slide view - 200 µm). (B) Cells with intracytoplasmic inclusion bodies positive for SMA (slide view - 50 µm) (C) Cells with intracytoplasmic inclusion bodies positive for desmin (slide view - 50 µm).

confirming the muscle origin of the tumor. The cells containing intracytoplasmic inclusion bodies were positive for α-SMA (Figure 2B), with this staining being stronger at the periphery of the inclusions than in the center. The final diagnosis was leiomyoma with intracytoplasmic inclusion bodies. The patient continues under follow-up and has been tumor free for 2 years.

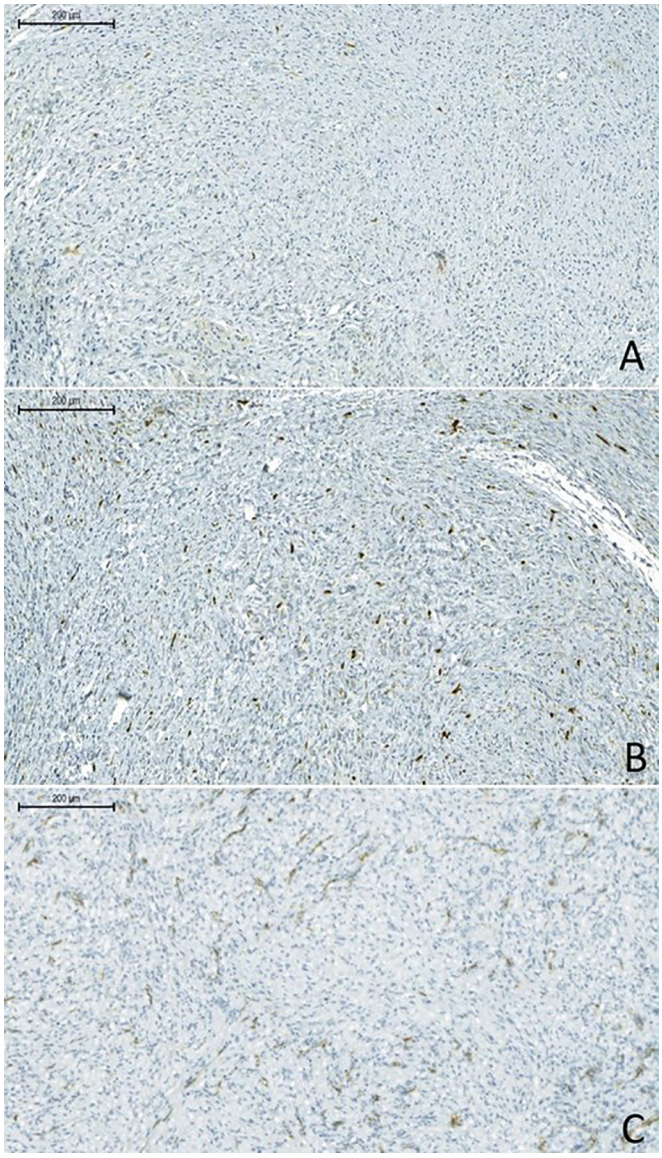

Figure 3. (A) Immunohistochemical expression negative for S-100in tumor cells(slide view - 200 µm). (B) Immunohistochemical expression negative for CD- 68 proteinin tumor cells(slide view - 50 µm). (C) Immunohistochemical expression negative for CD34 proteinin tumor cells(slide view - 50 µm).

DISCUSSION

Leiomyomas are benign smooth muscle tumors commonly found in the uterus, skin and gastrointestinal tract. Their occurrence in the oral cavity is extremely rare. Most cases affecting the oral cavity are of the solid or vascular type and account for approximately 75% of all oral leiomyomas[5,15]. Vascular leiomyoma is the most common in the oral cavity since, although smooth muscles are scarce, this region is rich in blood vessels[4,7].

The origin of leiomyomas is uncertain; however, it is believed that these tumors arise from the tunica media of blood vessels which consists of smooth muscles, in addition to other possible sources such as the circumvallate papillae of the tongue, lingual ducts and heterotopic embryonic tissue[16].

Oral leiomyomas can occur at any age, but are more prevalent in the 4th and 5th decades of life, and show a slight predilection for females. The sites most commonly affected are the lips, tongue, hard and soft palate, and cheek mucosa[6,17]. In the present case, the leiomyoma occurred in the tongue of a 35-year-old woman.

The clinical differential diagnosis of leiomyoma includes different oral tumors, either benign such as fibroma, myofibroma, neurofibroma, lipoma and schwannoma, or malignant such as leiomyosarcoma[4,5]. Leiomyomas exhibit clinical features that are similar to those of these tumors, but not specific, a fact that makes the differential diagnosis difficult[2,4,18]. Therefore, only accurate histopathological analysis can define the characteristics of this tumor[5,19].

According to the World Health Organization, leiomyoma can be divided into three types: angioleiomyoma, solid leiomyoma, and leiomyoblastoma. The most common type in the oral cavity is angioleiomyoma, accounting for 74% of all cases, followed by solid leiomyoma (25%), the most common type affecting the tongue[18].

In the present case, histopathological analysis identified the solid type, which is rare in the oral cavity. Solid leiomyomas are generally smaller than the vascular type[4] and are histologically characterized by interwoven bundles of spindle-shaped/stellate or elongated smooth muscle cells with blunt ends and pale nuclei. The tumors are highly cellularized and sometimes contain lobules of tumor cells separated by fibrous septa and a fibrous or myxoid stroma[4,20]. All of these histological features were observed in the present case.

Intracytoplasmic inclusion bodies are a known characteristic of different neoplasms and consist of aggregates of filaments that vary among different tumors. The types of these filaments vary in different lesions, being filaments of actin in leiomiomas[9,10]. Thus, the finding of these structures makes the present case unique, which is the first case of intracytoplasmic inclusion bodies in oral leiomyoma described in the literature. The filamentous components of muscle cells in leiomyomas comprise myofilaments and intermediate filaments[10,21].

These inclusion bodies generally show strong positive staining for α-SMA (predominantly at the periphery) and desmin, and moderate staining for vimentin and, in some cases, for cytokeratin. In the case of the last two markers, staining is usually found in the center[10]. In the present study, immunohistochemical detection of anti-α-SMA, anit-S-100 and anti-CD68 antibodies revealed positive staining for α-SMA, which was stronger at the periphery of the inclusions than in the center.

Regardless of the histopathological findings, surgery has been the only option for the treatment of all leiomyomas described in the literature. There are no reports of recurrence after complete excision[4,5]. The patient studied here was submitted to complete resection of the mass and showed no complications or recurrence after 2 years of follow-up.

FINAL COMMENTS

The present case is unique because of the peculiarity of its histopathological findings, particularly the presence of intracytoplasmic inclusion bodies, a feature not yet reported for leiomyomas of the oral cavity. Within this context, immunohistochemistry was found to be an important complementary tool.

CONFLICT OF INTEREST

The authors declare that they have no conflicts of interest.

REFERENCES

1. Campelo VE, Neves MC, Nakanishi M, Voegels RL. Nasal cavity vascular leiomyoma: a case report and literature review. Braz J Otorhinolaryngol. 2008;74:147-50.
2. Veeresh M, Sudhakara M, Girish G, Naik C. Leiomyoma: A rare tumor in the head and neck and oral cavity: Report of 3 cases with review. J Oral Maxillofac Pathol. 2013;17:281-7.
3. Lloria-Benet M, Bagán JV, Lloria de Miguel E, Borja-Morant A, Alonso S. Oral leiomyoma: a case report. Med Oral. 2003;8:215-9.
4. Gianluca S, Marini R, Tonoli F, Cristalli MP. Leiomyoma of oral cavity: case report and literature review. Ann Stomatol (Roma). 2011;2:9-12.
5. Alves PM, Novaes MM, Lucas Neto A, Godoy GP, Costa DA, Medeiros AMC, et al. Leiomioma vascular oral: relato de caso e estudo imunohistoquímico. Rev Cir Traumatol Buco-Maxilo--fac. 2013;13:47-52.
6. Gonzàlez Sànchez MA, Colorado Bonnin M, Berini Aytès L, Gay Escoda C. Leiomyoma of the hard palate: a case report. Med Oral Patol Oral Cir Bucal. 2007;12:E221-4.

7. Gaitan Cepeda LA, Quezada Rivera D, Tenorio Rocha F, Leyva Huerta ER, Mendez Sánchez ER. 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. 2008;13:E483-8.
8. Weiss SW, Goldblum JR. Benign tumors of smooth muscles. In: Schmitt W, ed. Enzinger and Weiss's Soft Tissue Tumors. 5th ed. St. Louis: Mosby; 2008. p. 517-44.
9. Dundr P, Povýsil C, Tvrdík D. Leiomyoma of the gastrointestinal tract with intracytoplasmic inclusion bodies. Report of three cases. Cesk Patol. 2006;42:1391-44.
10. Dundr P, Povýsil C, Tvrdík D, Mára M. Uterine leiomyoma as with inclusion bodies: an immunohistochemical and ultrastructural analysis of 12 cases. Pathol Res Pract. 2007;203:145-51.
11. Alroy J, Ucci AA, Tischler AS, Mitcheson HD. Intracytoplasmic inclusion bodies in leiomyoma of the urinary bladder. Ultrastruct Pathol. 1983;5:89-91.
12. Lin SL, Wang JS, Huang CS, Tseng HH. Primary intracerebral leiomyoma: a case with eosinophilic inclusions of actin filaments. Histopathology. 1996;28:365-9.
13. Matsukuma S, Takeo H, Ohara I, Sakai Y. Endoscopically resected colorectal leiomyomas often containing eosinophilic globules. Histopathology. 2004;45:302-3.
14. Parker RL, Young RH, Clement PB. Skeletal muscle-like and rhabdoid cell in uterine leiomyomas. Int J Gynecol Pathol. 2005;24:319-25.
15. Srinath VS, Meher R, Sabherwal A, Sharma N. Angiomyoma of soft palate - a case report. Ind J Surg. 2004;66:293-4.
16. Kotler HS, Gould NS, Gruber B. Leiomyoma of the tongue presenting as congenital airway obstruction. Int J Pediatr Otorhinolaryngol. 1994;29:139-45.
17. Luaces Rey R, Lorenzo Franco F, Gómez Oliveira G, Patiño Seijas B, Guitián D, López-Cedrún Cembranos JL. Oral leiomyoma in retro molar trigone. A case report. Med Oral Patol Oral Cir Bucal. 2007;12:E53-5.
18. Brooks JK, Nikitakis NG, Goodman NJ, Levy BA. Clinicopathologic characterization of oral angioleiomyomas. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2002;94:221-7.
19. Orsini G, Fioroni M, Rubini C, Piattelli A. Leiomyoma of the lip: report of a case. J Oral Maxillofac Surg. 2001;59:80-3.
20. Baden E, Doyle JL, Lederman DA. Leiomyoma of the oral cavity: a light microscopic and immunohistochemical study with review of the literature from 1984 to 1992. Eur J Cancer B Oral Oncol. 1994;30B:1-7.
21. Eyden BP, Hale RJ, Richmond I, Buckley CH. Cystoskeletal filaments in the smooth muscle cells of uterine leiomyomata and myometrium: an ultrastructural and immunohistochemical analysis. Virchows Arch A Pathol Anat Histopathol. 1992;420:51-8.