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A rare case of a vascular benign tumor in oral cavity – Case report

Um caso raro de tumor benigno vascular na cavidade oral – Relato de caso

Un caso raro de tumor benigno vascular en la cavidad oral – Reporte de caso

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Thayná Melo de Lima Morais
ORCID: https://orcid.org/0000-0002-2997-8016
University of Taubate, Brazil
E-mail: moraistml@gmail.com
Dárcio Kitakawa
ORCID: https://orcid.org/0000-0002-4321-5306
University Center of Braz Cubas, Brazil
E-mail: dkitakawa@yahoo.com
Felipe da Silva Peralta
ORCID: https://orcid.org/0000-0002-1664-4658
University Educational Society of Santa Catarina, Brazil
E-mail: felipe.periodontia@hotmail.com
Jussara Maria Gonçalves
ORCID: https://orcid.org/0000-0003-4030-9398
University Educational Society of Santa Catarina, Brazil
University of the Region of Joinville, Brazil
E-mail: jussaramariagoncalves@yahoo.com.br
Lineu Perrone Júnior
ORCID: https://orcid.org/0000-0002-1594-857X
Municipal Hospital Professor Doctor Alípio Corrêa Netto – Ermelino Matarazzo, Brazil
E-mail: lineu.perrone@gmail.com
Luis Felipe das Chagas e Silva de Carvalho
ORCID: https://orcid.org/0000-0003-1063-4624
University of Taubate, Brazil
University Center of Braz Cubas, Brazil
E-mail: luisfelipecarvalho@hotmail.com

Abstract
Leiomyomas are benign tumors arising from smooth muscle and can be classified as solid, epithelioid, and vascular leiomyoma (angioleiomyoma). Angioleiomyoma frequently occurs on the skin. Due to the lack of smooth muscle in the oral cavity, this tumor is rarely located in the mouth. This study aims to present a rare case of a 36-year-old woman with a painless submucosal nodule in the left cheek mucosa, measuring approximately 2 cm x 1 cm, with 5-years of evolution. Morphological and histochemical examination evidenced the proliferation of smooth muscle around the blood vessels of various calibers. The diagnosis of angioleiomyoma was confirmed, the tumor was surgically removed, and the patient had no further complications after 9-months of follow-up. The diagnosis of oral angioleiomyoma is challenging since this tumor is rare and can mimic or transform into malignancy. For this reason, histological examination in association with immunohistochemical is invaluable to establishing an accurate diagnosis and delivering suitable treatment.
Keywords: Angioleiomyoma; Vascular leiomyoma; Benign neoplasm; Neoplasms muscle tissue.

Resumo
Leiomiomas são tumores benignos que se originam a partir do músculo liso e podem ser classificados como leiomioma sólido, epitelioide e vascular (angioleiomioma). O angioleiomioma ocorre frequentemente na pele. Devido à ausência de músculo liso na cavidade oral, esse tumor raramente é localizado na boca. Este estudo tem como objetivo apresentar um caso raro de uma mulher de 36 anos com um nódulo submucoso, indolor, na mucosa jugal esquerda, medindo aproximadamente 2 cm x 1 cm, com 5 anos de evolução. O exame morfológico e histoquímico evidenciou a proliferação de músculo liso ao redor dos vasos sanguíneos de vários calibres. O diagnóstico de angioleiomioma foi confirmado, o tumor foi removido cirurgicamente e a paciente não apresentou mais complicações após 9 meses de acompanhamento. O diagnóstico de angioleiomioma oral é desafiador, pois esse tumor é raro e pode mimetizar ou se transformar em malignidade. Por esse motivo, o exame histológico em associação com a imuno-histoquímica é inestimável para estabelecer um diagnóstico preciso e fornecer tratamento adequado.
Palavras-chave: Angioliomioma; Liomioma vascular; Neoplasia benigna; Neoplasias de tecido muscular.

Resumen
Los leiomiomas son tumores benignos que se originan del músculo liso y pueden clasificarse como leiomioma sólido, epitelioide y vascular (angioleiomioma). El angioleiomioma aparece con frecuencia en la piel. Debido a la ausencia de músculo liso en la cavidad oral, este tumor rara vez se localiza en la boca. Este estudio tuvo como objetivo presentar un caso raro de una mujer de 36 años con un nódulo submucoso, indoloro, en la mucosa yugal esquerda, midiendo aproximadamente 2 cm x 1 cm, con 5 años de evolución. El examen morfológico e histoquímico evidencia la proliferación de músculo liso en relación a los vasos sanguíneos de varios calibres. El diagnóstico de angioleiomioma se confirmó, el tumor fue extirpado quirúrgicamente y el paciente no presentó más complicaciones después de 9 meses de acompañamiento. El diagnóstico de angioleiomioma oral es desafiante, porque el tumor es raro y puede mimetizarse o transformarse en maligno. Por esta razón, el examen histológico en asociación con la inmunohistoquímica es inestimable para establecer un diagnóstico preciso y proporcionar un tratamiento adecuado.
Palabras clave: Angioleiomioma; Leiomioma vascular; Neoplasia benigna; Neoplasias de tejido muscular.

1. Introduction

Leiomyoma is a benign smooth muscle neoplasm (Aitken-Saavedra, *et al.* 2018). It frequently occurs on the skin, with the extremities being the most affected site (Eley et al., 2012). Due to the lack of smooth muscle in the oral cavity, leiomyoma is rarely located in the mouth (Ranjan et al., 2014).

Morphologically, leiomyomas can be classified as solid leiomyoma, epithelioid leiomyoma (leioblastoma), and vascular leiomyoma (angioleiomyoma) (Aitken-Saavedra et al., 2018). Angioleiomyoma or angiomyoma is histologically classified into three categories: solid, venous, and cavernous (Kim et al., 2010). The most prevalent site in the oral cavity is the lips (Hassona et al., 2017) (Mehta et al., 2020) (Gueiros et al., 2011). However, other regions have been reported, such as palate (Bezerra et al., 2021), tongue (Ishikawa et al., 2016), cheek, and gingiva (Matiakis et al., 2018), among others.

Although the etiology is still uncertain, previous studies report trauma or spontaneous development as the main causes of origin in the oral cavity (Kim et al., 2010). Most angioleiomyomas present as painless and slow-growing, with the development of a well-defined lesion covered by normal mucosa (Cepeda et al., 2008). Regarding clinical presentation, angioleiomyomas in the oral cavity may mimic other benign conditions, making it very difficult to differentiate this tumor from other mesenchymal neoplasms. For this reason, the correct diagnosis is only possible after morphological and immunohistochemical analysis (Giudice et al., 2019). Therefore, this study aims to report an unusual case of angioleiomyoma of the oral cavity in a 36-year-old woman.

2. Methodology

This is a qualitative study, structured as a case report (Estrela, 2018). The main objective is to describe a rare case of oral angioleiomioma. Relevant literature was gathered about the diagnosis and treatment of this tumor to discuss this case. Therefore, a PubMed search using the term ‘‘oral angioleiomioma’’ was made to find the most relevant publications regarding this neoplasm.

This project is endorsed by the Ethics Committee and respects the guidelines and principles of CNS Resolution 466/2012, the CONEP 2018 letter, and the Declaration of Helsinki. The informed consent form was signed by the patient authorizing the use of data and images for scientific purposes.

3. Case Report

A 36-year-old woman was referred to a stomatology service complaining of an increase in volume in the left cheek mucosa with 5 years of evolution (Figure 1). Past medical history revealed nothing of note. On intraoral clinical examination, it was observed a painless submucosal nodule, smooth surface, of normal color on the left cheek mucosa, fibrous on palpation and measuring approximately 2 cm x 1 cm. Under the differential clinical diagnosis of deep mucocele, fibrous hyperplasia,

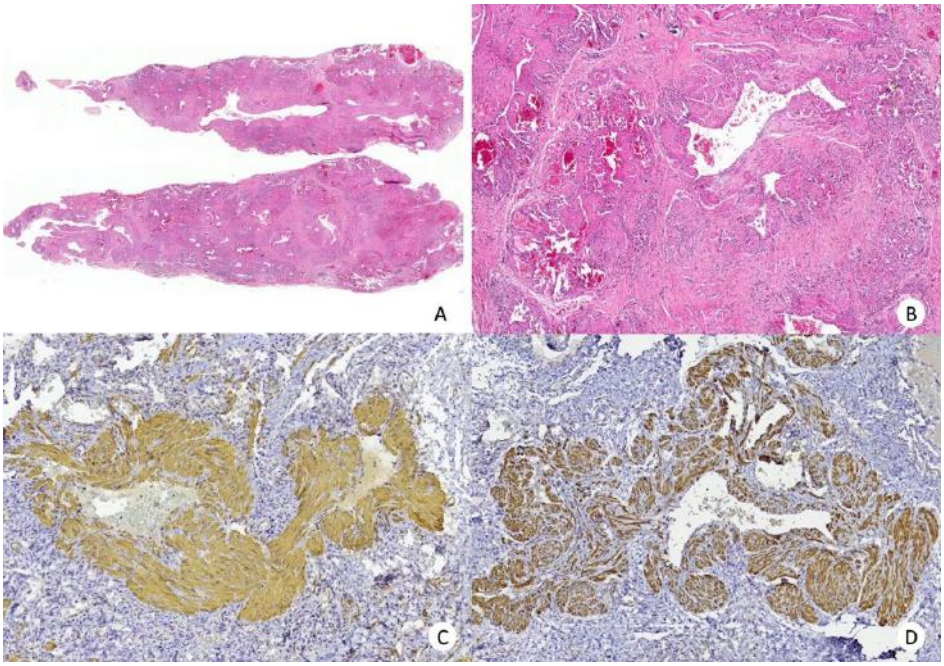
pleomorphic adenoma, and hemangioma, an ultrasound examination was requested, which showed a well-delimited mass (Figure 1).

Figure 1 - Submucosal nodular lesion in the left buccal mucosa.

Source: Authors (2024).

Excisional biopsy was performed and the specimen was processed. The histological examination revealed numerous blood vessels of various calibers interconnected with a proliferation of smooth muscle (Figure 2). Immunohistochemical staining for smooth muscle was performed and the tumor cells expressed diffuse and strong reactivity to SMA, confirming its proliferation. The diagnosis of angioleiomyoma was made, and the patient had no further complications after 9 months of follow-up (Figure 2).

Figure 2 - Morphological and histochemical aspect of oral cavity angioleiomyoma. (A, B) Photomicroscopy evidencing the proliferation of smooth muscle around the blood vessels (H&E, 5X and 20X). (C, D) Tumor cells express diffuse and strong reactivity to SMA (IHC, 20X).



Source: Authors (2024).

4. Discussion

Angioleiomyomas are tumors characterized by neoplastic proliferation of mature smooth muscle cells interspersed with blood vessels of different calibers (Aitken-Saavedra et al., 2018). They can occur in any region of the body, but the lower limbs followed by the head and neck are the most affected regions (Eley et al., 2012); however, angioleiomyomas are rare in the oral cavity (Ranjan et al., 2014). Several etiologic factors are suggested – hormonal changes, trauma, and venous stasis – however, the etiologic factor is unknown (Osano et al., 2015).

Clinically, oral angioleiomyomas are more common in men in the fourth decade of life (Jafarian et al., 2021), in contrast to the present case that occurred in a 36-year-old woman. Previous reports revealed occurrence in lips (Hassona et al., 2017) (Mehta et al., 2020) (Gueiros et al., 2011), buccal mucosa (Matiakis et al., 2018), palate (Bezerra et al., 2021) (Tsuji et al., 2014), tongue (Ishikawa et al., 2016), submandibular region (Wong et al., 2000), gingiva (Arpağ et al., 2016), buccal space (Inaba et al., 2015) (Kim et al., 2010), and mandible (Hamid et al., 2020). The lip is the most common site of involvement, followed by the buccal mucosa (Jafarian et al., 2021), which corroborates our case. A common finding for oral angioleiomyomas was painless swelling reported by patients. However, in rare cases painful symptoms associated with the lesion were reported (Jafarian et al., 2021) (Toida and Shimokawa, 2000). Imaging tests such as radiographs, ultrasound, magnetic resonance imaging, and computed tomography have been reported as preoperative evaluations. In this case, we requested an ultrasound which revealed a well-defined mass. Thus, based on the clinical presentation, location, and imaging exam, the differential diagnosis was a benign salivary gland lesion. However, the differential diagnosis should also include benign mesenchymal tumors such as lipoma, fibroma, neurofibroma, and vascular lesions (Jafarian et al., 2021). However, these lesions may have different imaging characteristics.

Histologically, this type of tumor is composed of a proliferation of thin and thick-walled blood vessels. The

morphological pattern of angioleiomyoma can mimic other benign vascular tumors such as hemangioma, hemangiopericytoma, hemangioendolethioma, vascular malformation, or other neurovascular hamartomas (Jafarian et al., 2021). Immunohistochemical markers specific for smooth muscle may aid in the diagnosis. In the present study, immunohistochemistry stains were positive for SMA and calponin. The diagnosis of sarcoma was excluded due to the lack of cellular pleomorphism and mitotic figures.

5. Conclusion

In conclusion, we report a case of oral angioleiomyoma of the buccal mucosa. We observed that the lips followed by the buccal mucosa are the most affected regions. In addition, oral angioleiomyomas can mimic other benign conditions, and histological examination in association with immunohistochemical staining is essential for a correct diagnosis. Due to the benign origin of the lesion, the treatment of choice is conservative surgical removal. Finally, due to the rarity of this neoplasm, it is important to continue studying this entity to expand the understanding of its pathogenesis, treatment, and prognosis.

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