PRESENTACIÓN DE CASO

Gingival giant cell fibroma of unusual size

Fibroma gingival de células gigantes de tamaño inusual

Carlos Augusto Galvão Barboza,^I Luciana Bastos Alves,^{II} Fernanda Ginani,^{III} Cláudia Roberta Leite Vieira De Figueiredo^{IV}

I Department of Morphology. Federal University of Rio Grande do Norte-UFRN, Natal-RN, Brazil.

II Department of Dentistry. Federal University of Rio Grande do Norte-UFRN, Natal-RN, Brazil.

III Department of Oral Pathology. Federal University of Rio Grande do Norte-UFRN, Natal-RN, Brazil.

IV Department of Physiology and Pathology. Federal University of Paraíba-UFPB, João Pessoa-PB, Brazil.

RESUMEN

El fibroma de células gigantes es un tumor fibroso benigno de la mucosa bucal que típicamente se presenta como una masa asintomática sésil o pediculada generalmente menos de 1 cm de diámetro. La lesión consiste en tejido fibroso no inflamado en el que se encuentran numerosas células fusiformes y estrelladas de gran tamaño, mononucleares o multinucleadas con prominente citoplasma basófilo. El propósito de este trabajo es describir el caso de un fibroma gingival de células gigantes de tamaño inusual. Una mujer blanca de 31 años de edad se presentó al servicio dental para la evaluación de un crecimiento en la encía mandibular. El examen clínico intrabucal reveló una masa gingival exofítica de 3,0 cm x 1,5 cm situado en la encía lingual en el área de los primeros y segundos molares permanentes mandibulares del lado derecho. El diagnóstico diferencial incluyó fibroma osificante periférico, granuloma periférico de células gigantes y fibroma de células gigantes. Se realizó la escisión quirúrgica completa de la lesión y el diagnóstico definitivo fue de fibroma de células gigantes. No se han observado complicaciones o recurrencia de la lesión después de 4 años de seguimiento. Aunque los fibromas de células gigantes son lesiones benignas en las que la escisión quirúrgica simple es curativa, es muy importante que los profesionales dentales y médicos reconozcan la necesidad de un diagnóstico preciso en vista de la frecuencia de aparición.

Palabras clave: encía; mucosa bucal; fibroma de células gigantes; diagnóstico.

ABSTRACT

Giant cell fibroma is a benign oral fibrous tumor and it is typically an asymptomatic sessile or pedunculated mass that is usually less than 1 cm in diameter. The lesion consists of uninflamed fibrous tissue in which there are numerous large uninucleated or multinucleated spindle- and stellate-shaped cells with prominent basophilic cytoplasm. The purpose of this paper is to report a case of a gingival giant cell fibroma of abnormal size. A 31-year-old white woman was referred to the dental service for evaluation of a growth on the mandibular gingival. The intraoral examination revealed a 3.0×1.5 cm exophytic gingival mass located in the lingual gingiva of the right mandibular permanent first and second molars. The differential diagnosis included peripheral ossifying fibroma, peripheral giant cell granuloma, and giant cell fibroma. Complete surgical excision of the lesion was performed and the diagnosis of giant cell fibroma was made. No complications or recurrence of the lesion have been noted after 4 years of follow-up. Although giant cell fibromas are benign lesions in which simple surgical excision is curative, it is very important that dental and medical professionals recognize it in light of the frequency of occurrence and the need for a precise diagnosis.

Key words: Gingiva; mouth mucosa; giant cell fibroma; diagnosis.

Correspondencia: Carlos Augusto Galvão Barboza. Department of Morphology. Federal University of Rio Grande do Norte. Av. Senador Salgado Filho, 3000, Lagoa Nova, Natal-RN, Brazil. Correo electrónico: cbarboza@cb.ufrn.br

INTRODUCTION

Giant cell fibromas (GCFs) are benign fibrous lesions of the oral mucosa, first described by Weathers, Callihan¹ (1974), that represent 5 % of all fibrous proliferations submitted for biopsy.2 No significant gender predilection exists, but the lesions predominantly occur in the first 3 decades of life and show a marked predisposition for the mandibular gingiva.^{2,3} Clinically, GCFs present as asymptomatic, small, pendunculated lesions and usually have a bosselated or being surface, nodular commonly regarded as papillomas.³ Histologically, the lesion is distinctive consisting of uninflamed fibrous tissue in which there are numerous large spindle- and stellateshaped cells with prominent basophilic cytoplasm. These giant cells may be multinucleated and dendritic.⁴ Conservative surgical excision is the treatment of choice and recurrences are rare.⁵ As the primary site of occurrence is on the gingival tissue, GCF is a disease of special interest to periodontologists. We report a case of an abnormally large GCF and provide a brief review of this notable oral tumor.

CASE REPORT



Fig. 1. Clinical aspect of the lesion: exophitic gingival mass lingual to the mandibular right permanent first and second molars, with an irregular and bosselated surface.

A 31-year-old white woman was referred to the School of Dentistry at the Federal University of Rio Grande do Norte for evaluation of a growth on the mandibular gingiva. The intraoral examination revealed a 3.0×1.5 cm exophytic mass located in the lingual gingiva of the right mandibular permanent first and second molars. The lesion had an irregular and bosselated surface and the base was pedunculated with normal mucosal coloration (Fig. 1).

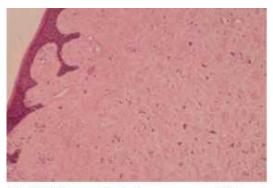


Fig. 2. Microscopic features: compact fibrous connective tissue with numerous large spindle-and stellate-shaped mononuclear and multinucleated cells, covered by a stratified squamous epithelium with thin, papillary projections (H/E, original magnification x 100).

The patient had poor oral hygiene and was not taking any medication. There were no systemic diseases and her medical history was noncontributory. The lesion was first noticed by the patient 3 years earlier and it had slowly grown to its present size. Neither radiopacities within the lesion nor radiolucency of the underlying bone were observed from the periapical radiographic evaluation. The differential diagnosis included peripheral ossifying fibroma, peripheral giant cell granuloma, and GCF. Routine blood tests were found normal. Complete surgical excision of the lesion was performed using a number 11 scalpel blade under local anesthesia and strict aseptic protocol and the specimen was submitted for histopathologic analysis. Calculus was not associated to neighbor teeth so that treatment of their surface was not necessary. Tissue examination under optical microscopy revealed a lesion composed of mature and compact fibrous connective tissue with numerous large spindle- and stellate-shaped mononuclear cells, and some multinucleated cells covered by a stratified squamous epithelium with thin, papillary projections (Fig. 2). The stellate-shaped giant cells had hyperchromatic nuclei, while the cytoplasm was well-demarcated and the frequently had a dendritic-like cells process (Fig. 3). Areas of inflammation were rarely noted. The diagnosis of GCF was made. No complications or recurrence

of the lesion have been noted after 4 years of follow-up.

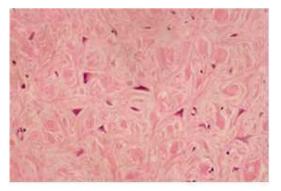


Fig. 3. Stellate-shaped giant cells with hyperchromatic nuclei and well-demarcated cytoplasm (H/E, original magnification x 200).

DISCUSSION

Oral fibrous lesions containing stellate and multinucleated giant cells were described as an entity by Weathers, Callihan1 (1974), who applied the term "giant cell fibroma" to oral mucosal tumors that had previously been diagnosed as either fibrous hyperplasia, fibromas, or fibroepithelial polyps. They believed that there were sufficient distinctive clinical and histologic features to warrant separation and reclassification as a separate clinical entity.

In the present case, the lesion was 3 cm in its largest diameter which is somewhat unusual as the vast majority of lesions are less than 1 cm in diameter, with an average size less than 0.5 cm.1,3,6 A slight female preponderance has been reported,3,6 whereas others observed no significant gender predilection.1,2,7 GCFs occur most often among young people, with a peak incidence in the 2nd decade of life.1-3 It is found more frequently on the gingiva, followed by the tongue and the buccal mucosa or palate.^{1,3, 8-11}

The clinical and histopathologic characteristics of the excised lesion were with in agreement the existing descriptions of GCFs in the literature; specifically, a bosselated, peduncunlated exophytic mass largely composed of fibrous connective tissue loosely arranged with a prominent vascular element, especially in the subepithelial zone. The most characteristic histologic feature is the presence of large spindle-shaped or more often stellate-shaped cells. These cells are usually mononuclear but multinucleated cells can also be present. The cytoplasm of these cells is welldemarcated and occasional dendritic processes are observed. The cellular boundaries appear to be separated from the surrounding collagen fibers in areas, and some of the cells contain melanin granules.¹² The stellate-shaped cells are most prominent just beneath the epithelium and are less common or absent in the central portion of the lesion.^{3,5}

Different origins of stellate giant cells have been suggested, such as the melanocytic lineage,³ but other studies have ruled out this possibility since these cells were found to be negative for antibodies against protein S100.2,4 Langerhans cells were considered to represent these giant cells, but were excluded due to the fact that no Birbeck granules could be identified on ultrastructural studies.¹³

An endothelial or myofibroblastic origin is unlikely given the negative reaction for alpha-smooth muscle actin, and also the possibility that these cells are derived from the macrophage-monocyte lineage is not supported by the negative staining for CD68, LCA, and HLA-DR.2,4 In the study of Souza et al.13 (2004), stellate giant immunolabeling cells exhibited for vimentin and were negative for muscle actin (HHF-35), CD68, and factor XIIIa. According to the authors, the most plausible hypothesis is that these cells are indeed derived from a fibroblastic lineage.

Weathers, Callihan1 (1974) suggested that the giant cells observed in GCFs were formed by the fusion of fibroblast-like mononuclear cells. Mighell et al.¹⁴ (1996) also proposed that giant cells result from the fusion of fibroblasts; a fact explaining the presence of these cells in other fibrous lesions, as observed in the present study for both fibrous hyperplasias and fibroepithelial polyps.

Analyzing the possible interaction of stellate giant cells with mast cells, Santos (2011) al.15 studied еt the immunohistochemical expression of mast cell tryptase in GCF of the oral mucosa and the possible interaction of these cells with stellate giant cells, as well as their role in fibrosis and tumor progression. They concluded that although mast cells were less numerous in CGFs, these cells exhibited a significant interaction with stellate giant cells present in these

tumors. Furthermore, the involvement of mast cells in the induction of fibrosis and modulation of endothelial cell function in CGFs has been suggested.

Various fibrous lesions of the skin and the mucous membranes have been shown to contain stellated and multinucleated cells of presumed fibroblastic origin.3,16 Thus, the presence of these cells is not unique for GCFs and therefore the lesions are likely to be reactive or hamartomatous rather than true neoplasms.2 Because of the innocuous nature of the lesion, simple surgical excision appears to be the treatment of choice and the lesions are rarely recurrent.^{3,17,18}

Although GCFs are benign lesions in which simple surgical excision is curative, it is very important that dental and medical professionals recognize it in light of the frequency of occurrence and the need for making a precise diagnosis and for proper treatment planning, even in tumors with abnormal and excessive size as reported in this case.

Conflicto de intereses

Los autores no declaran conflictos de intereses.

BIBLIOGRAPHIC REFERENCES

1. Weathers DR, Callihan MD. Giant-cell fibroma. Oral Surg Oral Med Oral Pathol. 1974;37(3):374-84.

2. Houston GD. The giant cell fibroma: a review of 464 cases. Oral Surg Oral Med Oral Pathol. 1982;53(6):582-7.

3. Magnusson BC, Rasmusson LG. The giant cell fibroma: a review of 103 cases with imunohistochemical findings. Acta Odontol Scand. 1995;53(5):293-6.

4. Odell EW, Lock C, Lombardi TL. Phenotypic characterization of stellate and giant cells in giant cell fibroma by imunohistochemistry. J Oral Pathol Med. 1994;23(6):284-7.

5. Swan RH. Giant cell fibroma: a case presentation and review. J Periodontol. 1988;59(5):338-40.

6. Barboza CAG, Ginani F, Carezato TT, Sousa SCOMD. Fibroma de células gigantes: análise epidemiológica de 309 casos. RPG: Rev Pós Grad. 2009;16(2):81-4.

7. Nikitakis NG, Emmanouil D, Maroulakos MP, Angelopoulou MV. Giant cell fibroma in children: report of two cases and literature review. J Oral Maxillofac Res. 2013;4(1):e5. doi: 10.5037/jomr.2013.4105.

8. Jimson S, Jimson S. Giant cell fibroma: a case report with immunohistochemical markers. J Clin Diagn Res. 2013;7(12):3079-80. doi: 10.7860/JCDR/2013/6476.3859.

9. Sabarinath B, Sivaramakrishnan M, Sivapathasundharam B. Giant cell fibroma: a clinicopathological study. J Oral Maxillofac Pathol. 2012;16(3):359-62. doi: 10.4103/0973-029X.102485.

10. Campos MS, Domaneschi C, Nunes FD, Weinfeld I. Giant cell fibroma of the maxillary gingiva in children: a case report. J Dent Child. 2010;77(3):174-6.

11. Datar UV, Mohan BC, Hallikerimath S, Angadi P, Kale A, Mane D. Clinicopathologic study of a series of giant cell fibroma using picrosirius red polarizing microscopy technique. Arch Iran Med. 2014;17(11):746-9. doi: 0141711/AIM.005.

12. Sonalika WG, Sahu A, Deogade SC, Gupta P, Naitam D, Chansoria H, Agarwal J, Katoch S. Giant cell fibroma of tongue: understanding the nature of an unusual histopathological entity. Case Rep Dent. 2014;2014:864512. doi: 10.1155/2014/864512.

13. Souza LB, Andrade ES, Miguel MC, Freitas RA, Pinto LP. Origin of stellate giant cells in oral fibrous lesions determined by immunohistochemical expression of vimentin, HHF-35, CD68 and factor XIIIa. Pathology. 2004;36(4):316-20.

14. Mighell AJ, Robinson PA, Hume WJ. PCNA and Ki-67 immunoreactivity in multinucleated cells of giant cell fibroma and peripheral giant cells granuloma. J Oral Pathol Med. 1996;25(5):193-9.

15. Santos PP, Nonaka CF, Pinto LP, de Souza LB. Immunohistochemical expression of mast cell tryptase in giant cell fibroma and inflammatory fibrous hyperplasia of the oral mucosa. Arch Oral Biol. 2011;56(3):231-7. doi: 10.1016/j.archoralbio.2010.09.020.

16. Uloopi KS, Vinay C, Deepika A, Sekhar RC, Raghu D, Ramesh T. Pediatric giant cell fibroma: an unusual case report. Pediatr Dent. 2012;34(7):503-5.

17. Vergotine RJ. A giant cell fibroma and focal fibrous hyperplasia in a young child: a case report. Case Rep Dent. 2012;2012:370242. doi: 10.1155/2012/370242.

18. Shapira M, Akrish S. A 6-year-old girl with a lesion on the tongue. Giant cell fibroma of tongue. Pediatr Ann. 2011;40(2):71-4. doi: 10.3928/00904481-20110117-05.

Recibido: 21 de marzo de 2014. Aprobado: 30 de marzo de 2015.