

Verruciform xanthoma of the upper anterior gingiva

Janderson Teixeira Rodrigues ¹
Juliana de Noronha Santos
Netto ²
Águida Maria Menezes Aguiar
Miranda ²
Simone de Macedo Amaral ²
Fábio Ramôa Pires ^{1,3}

Abstract:

Introduction: Verruciform xanthoma (VX) is a reactive oral mucosal lesion that can resemble other papillary conditions. **Objective:** To report a case of gingival VX clinically mimicking other papillary swellings. **Case report:** A 66-year-old female presented for evaluation of a painless papillary growth in the gingival margin of the upper right central incisor. Clinical diagnosis included papillary gingival hyperplasia and VX. An excisional biopsy was done and histology revealed a proliferative surface epithelium with papillary projections and CD68+ xanthomatous cells on the upper papillary superficial connective tissue. Diagnosis was VX and the patient remains in clinical follow-up with no signs of recurrence for 4 months. **Conclusion:** VX should be considered in the differential diagnosis of gingival papillary swellings.

Keywords: Gingiva; Xanthomatosis; Mouth Mucosa; Mouth Diseases.

¹ Estácio de Sá University, Post-graduation program in Dentistry - Rio de Janeiro - RJ - Brasil

² Brazilian Dental Association, Stomatology - Rio de Janeiro - RJ - Brasil

³ State University of Rio de Janeiro, Oral Pathology - Rio de Janeiro - RJ - Brasil

Correspondence to:

Fábio Ramôa Pires.

E-mail: ramoafop@yahoo.com

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INTRODUCTION

Verruciform xanthoma (VX) is an uncommon reactive papillary exophytic lesion first described in 1971¹. Clinicians usually do not include VX in the differential diagnosis of papillary growths affecting the oral mucosa and, as it can resemble infectious, reactive and potentially malignant/malignant disorders, it is advisable that this condition should be recognized and included as a differential diagnosis in this situation. The aim of the present study is to report a VX affecting the gingiva calling attention to the importance of considering this entity in the differential diagnosis of oral papillary swellings.

CASE REPORT

A 66-year-old Afro-American female was referred for evaluation of a painless papillary growth on the anterior gingiva lasting 15 days. Medical history included controlled systemic hypertension managed with hydrochlorotiazide and hydralazine. Oral examination revealed a 1,2 x 0,6 cm painless firm exophytic papillary slightly reddish growth in the free buccal gingival margin of the upper right central incisor (Fig. 1A).



Figure 1. (A) Exophytic papillary reddish growth in the free buccal gingival margin of the upper right central incisor. Comparison of the initial clinical picture (B) and the clinical aspect of the region 4 months after surgical removal of the lesion (C).

Clinical differential diagnosis included papillary gingival hyperplasia, condiloma acuminatum and VX. An excisional biopsy was performed under local anesthesia and histological analysis of the 5 µm hematoxylin and eosin stained histological slides showed a proliferative parakeratinized stratified squamous epithelium with papillary surface and long and thin projections to the adjacent connective tissue (Fig. 2A and 2B). Xanthomatous cells with large vesicular and granular cytoplasm occupied the papillary superficial connective tissue (Fig. 2B and 2C).

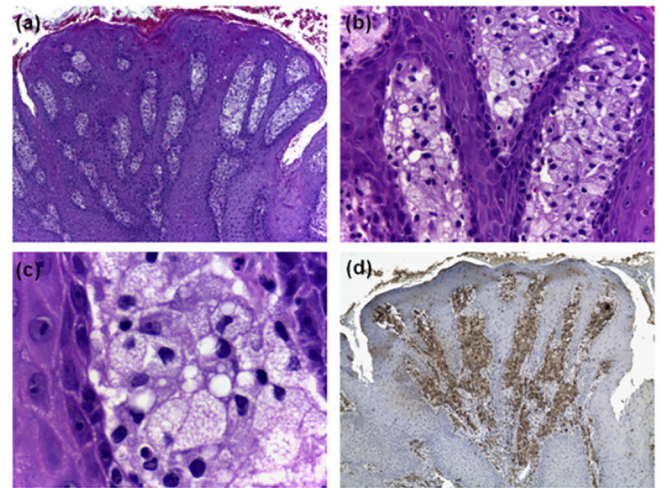


Figure 2. (A) Histological picture of the lesion showing the keratinization, epithelial hyperplasia and the papillary surface (HE, 100x). (B) Xanthomatous cells occupying the superficial connective tissue close to the elongated rete ridges (HE, 400x). (C) Detail of the xanthomatous cells showing the granular vacuolated cytoplasm (HE, 1000x). (D) Immunoperoxidase staining of CD68 on the xanthomatous cells (same area as Figure 1A) (Immunoperoxidase, 100x).

Immunohistochemical analysis showed that these cells were CD68 positive (Fig. 2D), confirming their macrophage origin, and final diagnosis was VX. Post-surgical recovery was uneventful and the patient remains in clinical follow-up without any signs of local recurrence for 6 months (Fig. 1B and 1C).

The authors declare that they have followed the Declaration of Helsinki and all ethical requirements.

DISCUSSION

Most VX affect the oral cavity but some cases have been reported in the skin and other mucosal surfaces. Clinical presentation of oral VX usually includes painless isolated reddish to whitish exophytic papillary growths measuring less than 2.0 cm mostly affecting adult males²⁻⁵. The most common oral locations are the gingiva, alveolar mucosa and palate³⁻⁵ and, although most

VX are isolated lesions, some multiple cases affecting the oral cavity, especially the gingiva, have been reported⁶.

Although the clinical picture is suggestive, few clinicians have raised the suspicion of VX during treatment planning of papillary exophytic lesions. Differential diagnosis should include mainly squamous papilloma, verruca vulgaris, spongiotic gingival hyperplasia, verrucous hyperplasia and early verrucous carcinoma and papillary squamous cell carcinoma^{2-4,7,8}. As final diagnosis is based on histological analysis, suspicion of VX when dealing with the differential diagnosis of oral papillary lesions is advisable for adequate treatment.

Pathogenesis of VX is still unknown but local trauma is considered the major etiological factor. The condition has been reported as secondary to other diseases associated with epithelial integrity disruption, such as graft versus host disease, pemphigus vulgaris and lichen planus^{4,8}. Histologically VX is characterized by the presence of epithelial proliferation, acanthosis, parakeratosis and elongated rete ridges, associated with a papillary/verrucous surface of the stratified squamous surface epithelium with the crypts plugged with parakeratin^{2,9,10}.

Xanthomatous CD68+/cathepsin B+ macrophages with a large vesicular to granular lipid-rich cytoplasm and a rounded central or displaced nucleus occupy the connective tissue papillae among the rete ridges^{3,9}. Treatment of VX is established with conservative surgical excision and no recurrences are expected after complete removal^{2,4}.

In conclusion, VX should be included in the differential diagnosis of oral gingival papillary swellings for adequate treatment planning and proper management.

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REFERENCES

1. Shafer WG. Verruciform xanthoma. *Oral Surg Oral Med Oral Pathol.* 1971;3:784-9.
2. Takehana S, Kameyama Y, Fukaya M, Kawai T. Verruciform xanthoma of the gingiva: report of three cases. *J Oral Maxillofac Surg.* 1989;47:1079-81.
3. Oliveira PT, Jaeger RG, Cabral LA, Carvalho YR, Costa AL, Jaeger MM. Verruciform xanthoma of the oral mucosa. Report of four cases and a review of the literature. *Oral Oncol.* 2001;37:326-31.
4. Philipsen HP, Reichart PA, Takata T, Ogawa I. Verruciform xanthoma--biological profile of 282 oral lesions based on a literature survey with nine new cases from Japan. *Oral Oncol.* 2003;39:325-36.
5. Ide F, Obara K, Yamada H, Mishima K, Saito I, Kusama K. Cellular basis of verruciform xanthoma: immunohistochemical and ultrastructural characterization. *Oral Dis.* 2008;14:150-7.
6. Qi Y, Sun Q, Yang P, Song A. A case of multiple verruciform xanthoma in gingiva. *Brit J Oral Maxillofac Surg.* 2014;52:e1-3.
7. Yu CH, Tsai TC, Wang JT, Liu BY, Wang YP, Sun A, et al. Oral verruciform xanthoma: a clinicopathologic study of 15 cases. *J Formos Med Assoc.* 2007;106:141-7.
8. Shahrabi Farahani S, Treister NS, Khan Z, Woo SB. Oral verruciform xanthoma associated with chronic graft-versus-host disease: a report of five cases and a review of the literature. *Head Neck Pathol.* 2011;5:193-8.
9. de Andrade BA, Agostini M, Pires FR, Rumayor A, Carlos R, de Almeida OP, et al. Oral verruciform xanthoma: a clinicopathologic and immunohistochemical study of 20 cases. *J Cutan Pathol.* 2015;42:489-95.
10. Nowparast B, Howell FV, Rick GM. Verruciform xanthoma. A clinicopathologic review and report of fifty-four cases. *Oral Surg Oral Med Oral Pathol.* 1981;51:619-25.

