Verruciform xanthoma of the lip: A rarity

Treville Pereira, Subraj Shetty, Shashibhushan Dodal, Avinash Tamgadge

Department of Oral and Maxillofacial Pathology and Microbiology, DY Patil School of Dentistry, Navi Mumbai, Maharashtra, India

ABSTRACT

Verruciform xanthoma (VX) is an uncommon, benign, asymptomatic mucocutaneous lesion of an unknown etiopathology. It usually presents as a papule or single plaque with a verrucous or a papillomatous surface and pale yellowish to red in color. It occurs primarily on the masticatory mucosa of middle-aged individuals. We report a case of VX on the lip of a 59-year-old man. Histopathologically, VX is diagnosed by the presence of foam cells in the papillary region of the connective tissue. Differentiating a verrucous carcinoma from VX is important, especially in small superficial lesions, which may lead to inappropriate and excessive surgical intervention. Treatment of VX consists of simple surgical excision and recurrence is rare.

Key words: Foam cells, lip, verruciform xanthoma

INTRODUCTION

Verruciform xanthoma (VX) was first reported by Shafer in 1971 as an uncommon benign lesion of the oral mucosa that resembles a virus-induced papilloma but has an unknown etiology and is of uncertain nature.[1] The most frequently encountered site is the gingival margin, followed by the alveolus, hard palate, and floor of the mouth.[2] Extraorally, VX has been reported in the vulval region.[3]

VXs are asymptomatic, slow growing lesions, red to yellowish in color, granular or verrucous in appearance, essentially being sessile or pedunculated.[4] The clinical appearance of a VX is not diagnostic; and diagnosis is almost always done by histologic examination. Histologically, it is characterized by parakeratosis, papillomatosis, and an aggregate of foam cells in the submucosal stroma without any epidermal atypia.[1,2] Clinically, VX needs to be differentiated from papilloma, verrucous carcinoma, and sometimes squamous cell carcinoma. Most often, the affected sites are in the epithelia that are subjected to trauma or irritation. Three different architectural appearances have been seen under the light microscope: (1) Verrucous appearance, (2) papillary or cauliflower architecture, and (3) slightly raised or flat lesion.[5] The characteristic microscopic feature is the accumulation of lipid-laden macrophages in the connective tissue papillae between the epithelial ridges.[6] The treatment of a VX lesion involves local surgical excision and recurrences are rare.[7]

CASE REPORT

A 59-year-old male was referred for evaluation of an asymptomatic gingival swelling on the lower lip of eight months duration. Clinical examination revealed a pink, sessile, papillomatous growth, measuring approximately a 0.5 × 1.0 cm, on the lower lip [Figure 1]. He gave a history of smoking beedis and chewing tobacco three to four times/day since the past 45 years, due to which the all his teeth were stained. There was no report of local trauma. The patient's medical, family, and social history was noncontributory. Extraoral examination revealed no other associated lesions. Routine investigations were carried out, which included a complete hemogram, urine analysis, lipid profile, thyroid, liver, kidney function tests, and blood sugar levels. The results of all the tests were within normal range. With a differential diagnosis of

Access this article online
Website: www.idoj.in
DOI: 10.4103/2229-5178.182356
Quick Response Code:

Address for correspondence:
Dr. Treville Pereira, Department of Oral and Maxillofacial Pathology and Microbiology, DY Patil School of Dentistry, Sector 7, Nerul, Navi Mumbai - 400 706, Maharashtra, India.
E-mail: trevillepereira@gmail.com

Cite this article as: Pereira T, Shetty S, Dodal S, Tamgadge A. Verruciform xanthoma of the lip: A rarity. Indian Dermatol Online J 2016;7:180-2.

papilloma and verrucous carcinoma, an excisional biopsy was performed under local anesthesia.

Hematoxylin and eosin–stained sections showed a parakeratined stratified squamous epithelium with papillary projections. The papillary portion of the underlying connective tissue had large swollen cells with clear-to-eosinophilic granular cytoplasm and eccentrically placed nuclei. These large swollen cells are called "xanthoma cells" or foamy histiocytes. Mild-to-moderate inflammatory cell infiltrate consisting mainly of lymphocytes were observed in the deeper portion of the connective tissue [Figure 2]. Immunohistochemically, the xanthoma cells showed a strong cytoplasmic positivity for CD68 [Figures 3 and 4]. Based on clinical, histopathological, and immunohistochemical findings, the case was diagnosed as a verruciform xanthoma.

DISCUSSION

VX is a rare lesion of unknown etiology, accounting for 0.025%–0.095% of all cases in which oral biopsy was performed.[8] Most consider it to be a reactive process rather than a true neoplasm. Local irritation or trauma has been proposed to play a role in the pathogenesis. In the present case, beedi smoking may have played a role as a local irritant. In 1971, Shafer first reported cases of VX of the oral cavity.[1]

These lesions are reported mostly in males, with a male-to-female ratio of 1.1:1. However, the ratio reverses after the age of 50 years in favor of females, when the male-to-female ratio is 0.8:1. In the order of frequency, the gingival and the hard palate are the most commonly affected sites followed by the buccal mucosa, tongue, and the floor of the mouth. The soft palate and the lip are rarely affected.[2] In the present case, the lesion was noticed on the lower lip of a 59-year-old male patient.

VX tends to occur as an isolated lesion, but it can develop in association with other diseases, including snuff dipper's keratosis, pemphigus vulgaris, dystrophic epidermolysis

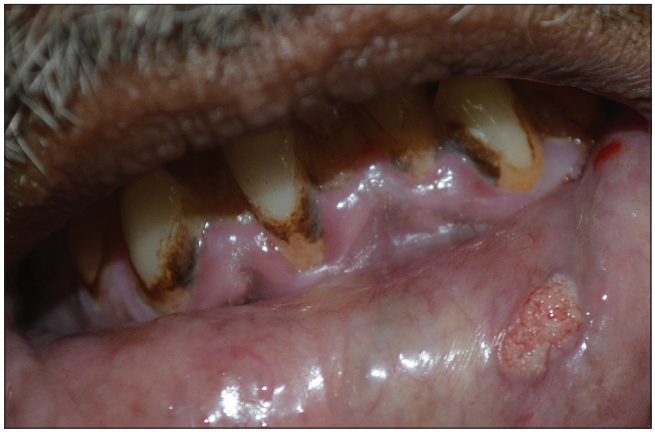

Figure 1: Photograph showing a growth on the lower lip

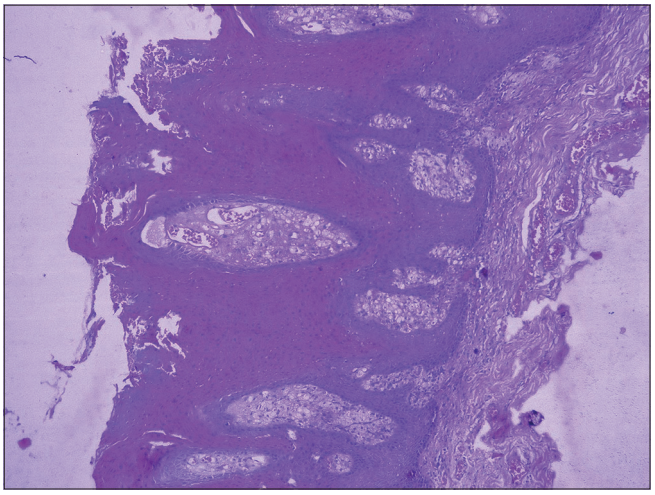

Figure 2: Photomicrograph showing papillary projections lined with a thin epithelium and core of connective tissue extending into the epithelium. (H and E stain, ×10)

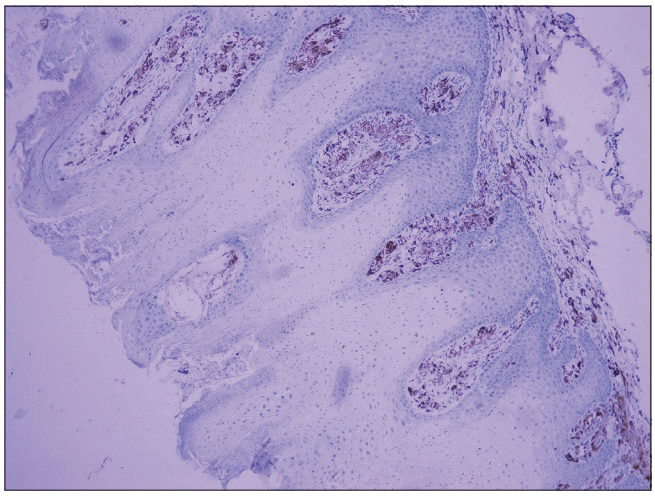

Figure 3: Foam cells showing strong cytoplasmic CD68 immunostaining. The epithelial cells were negative. (Immunohistochemical staining, ×10)

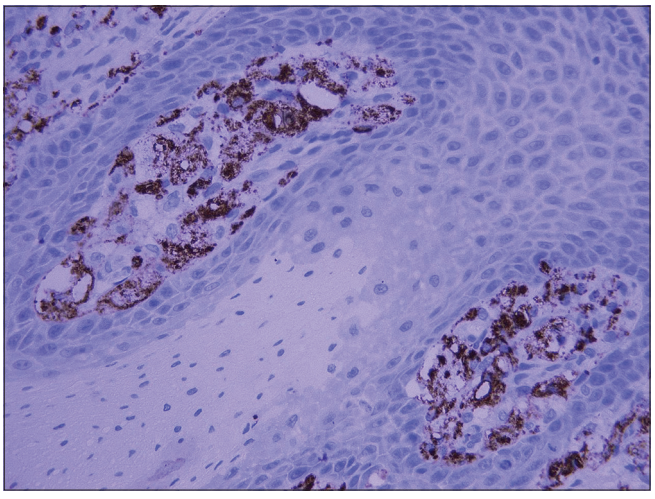

Figure 4: Xanthoma cells showing strong cytoplasmic CD68 immunostaining. (Immunohistochemical staining, ×40)

bullosa, lichen planus, carcinoma-*in situ*, discoid lupus erythematosus, and CHILD syndrome.[7]

VX, histologically is characterized by a squamous epithelial surface of varying morphological patterns covered with parakeratin and a hyperplastic epidermis without atypia.[5] The characteristic feature is the presence of foam cells that fill the dermal papillae. The origin of these foam cells is controversial. Most authors consider that foam cells are lipid-laden macrophages. The concept that these lipid-laden macrophages may accumulate due to epithelial degeneration was introduced by Zegarelli *et al*. The products of epithelial breakdown could elicit an inflammatory response with a subsequent release of the lipid material through epithelial degeneration, which is finally scavenged by the macrophages.[9]

Mostafa *et al*. suggested that the elongation of the epithelial rete ridges seen in VX is not due to the proliferation of the epithelial cells with downward growth of the retepegs, but is an illusionary effect due to the upward pushing effect of the accumulated macrophages. This may result in the thinning of the epithelium overlying the macrophages in the connective tissue.[10] Mostafa *et al*. were also the first to demonstrate the origin of VX foam cells as cells of the monocyte/macrophage lineage. They observed that xanthoma cells were positive for CD68 and vimentin but were negative for S-100 protein. They also suggested that VX may be an immunological disorder, most probably of a cell-mediated mechanism, although the exact cause could not be determined.[10] In the present case, the foam cells showed a strong CD68 immunopositivity.

Since the surface epithelium is keratinized, the color may vary from white to red. The papillary or verrucous outer appearance may be clinically misleading. A correct diagnosis is almost always made during histologic examination. Histopathologically, VX may be misdiagnosed as verrucous vulgaris, condyloma acuminatum, or verrucous carcinoma. However, verrucous carcinoma can be distinguished by the presence of an invasive epithelial proliferation, cellular atypia, and the lack of characteristic xanthoma cells.[2] Treatment of VX consists of simple surgical excision. The prognosis of VXs is excellent.

VX being a chronic reactive process with an asymptomatic clinical course may make the diagnosis challenging due to its varied clinical appearance. However, the histological features are well defined. In small surgical specimens, the presence of xanthoma cells may be missed, if one is unfamiliar with the site and existence of this lesion. Furthermore, a proper diagnosis is important since VX can occur in coexistence with other systemic diseases.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

REFERENCES

1. Shafer WG. Verruciform xanthoma. Oral Surg Oral Med Oral Pathol 1971;31:784-9.
2. Philipsen HP, Reichart PA, Takata T, Ogarva 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.
3. Santa Cruz DJ, Martin SA. Verruciform xanthoma of the vulva: Report of two cases. Am J Clin Pathol 1979;71:224-8.
4. Polonowita AD, Firth NA, Rich AM. Verruciform xanthoma and concomitant lichen planus of the oral mucosa: A report of three cases. Int J Oral Maxillofac Surg 1999;28:62-6.
5. Nowparast B, Howell FV, Rick GM. Verruciform xanthoma: A clinicopathological review and report of fifty-four cases. Oral Surg Oral Med Oral Pathol 1981;51:619-25.
6. 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 literature. Oral Oncol 2000;37:326-31.
7. Sah K, Kale AD, Hallikerimath S. Verruciform xanthoma: Report of two cases and review on pathogenesis. J Oral Maxillofac Pathol 2008;12:41-4.
8. Buchner A, Hansen LS, Merrell PW. Verruciform xanthoma of the oral mucosa: Report of five cases and review of the literature. Arch Dermatol 1981;117: 563-5.
9. Zegareth DJ, Zegarelli-Schmidt EC, Zegarelli EV. Verruciform xanthoma. Further light and electron microscopic studies, with the addition of a third case. Oral Surg Oral Med Oral Pathol 1975;40:246-56.
10. Mostafa KA, Takata T, Ogarva I, Ijuhin N, Nikai H. Verruciform xanthoma of the oral mucosa: A clinicopathological study with immunohistochemical findings relating to pathogenesis. Virchows Arch A Pathol Anat Histopathol 1993;423:243-8.