Verruciform xanthoma of buccal mucosa: A case report with review of literature

Sudhir Bhalerao, Pooja Bhat, Ranit Chhabra, Avinash Tamgadge

Abstract

Verruciformxanthoma (VX) is an uncommon benign mucocutaneous lesion of unknown etiology. It appears as a papule or single plaque with verrucous or papillomatous surface and variable color from reddish pink to gray. It occurs primarily in the masticatory mucosa. Histologically, VX is characterized by the presence of parakeratinized epithelium with thin rete ridges and connective tissue papillae extending up to the surface. The papillae characteristically consist of foam cells, also called xanthoma cells. We report a case of VX in the buccal mucosa and discuss their clinical and histopathological findings.

Keywords: Buccal mucosa, foam cells, verruciform xanthoma

Introduction

Verruciformxanthoma (VX) is an uncommon benign lesion of adults and was first described by Shafer in 1971.[1] Oral VX presents as a solitary or multiple demarcated, granular lesions exhibiting a flat to papillary or verrucous surface. It can be found in masticatory mucosal sites with a predilection for the gingival margin and hard palate.[2] The characteristic microscopic feature is the accumulation of lipid-laden macrophages in the connective tissue papillae between the epithelial ridges.[3] Excision is the treatment of choice. However, no potential for malignancy has been reported to date.[4]

Case Report

A 70-year-old man reported to the department with a chief complaint of missing teeth. He gave a history of tobacco chewing four to five times a day for the last 50 years. Intraoral examination revealed a solitary pale and hyperkeratotic lesion with a rough, pebbly surface and a sessile base. It measured around 1.5×1.5 cm and was asymptomatic with no other associated lesion [Figure 1]. Clinically, the lesion was diagnosed as leukoplakia and an excisional biopsy was performed under local anesthesia.

Histopathologically, the epithelium was slightly raised with surface varying from papillomatous to verrucous, composed of parakeratinized stratified squamous epithelium [Figure 2]. The elongated epithelial ridges uniformly extended into lamina propria. Numerous foamy histiocytes or xanthoma cells were seen in the connective tissue papillae between epithelial ridges [Figure 3]. These cells showed clear-to-eosinophilic cytoplasm with eccentrically placed nuclei [Figure 4]. Mild-to-moderate degree of inflammatory cell infiltration consisting mainly of lymphocytes was observed in the stroma. Based on these histopathological features, diagnosis of VX was given.

Discussion

VX presents clinically as a papule or single plaque showing a verrucous or papillomatous surface, with color varying from reddish pink to gray. It is asymptomatic, has a low growth rate, and evolves to sizes varying from 0.2 to 2 cm.[5] There is no gender predilection and most commonly occurs in middle-aged persons with mean age of 40–50 years.[6] It can occur in any site but has a predilection for masticatory mucosa (gingival and hard palate).

Although in most cases VX tends to occur as an isolated lesion, in several cases the lesion develop in association with other diseases, including snuff dipper's keratoses, oral pemphigus vulgaris, carcinoma *in situ*, regressive dystrophic epidermolysis bullosa, lichen planus, solar keratoses, and discoid lupus erythematosus.[7,8]

Ide *et al*. suggested some possible oral etiologic agents such as wet microenvironment, periodontal pathogens, mechanical stimuli, tobacco, alcohol, drugs, and sensitizing or allergic substances of foodstuffs and dental materials. Viral infection could be associated with the lesion; however,

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studies have been done with no association found with human papillomavirus. Immune response was also related to VX pathogenesis.[9]

Histopathologically, lesions of VX may be flat or slightly raised with a papillomatous or verrucous surface composed of parakeratinized epithelial cells. Elongated epithelial rete ridges extend into the lamina propria at a uniform level or depth. The most characteristic feature is the presence of numerous foam or xanthomatous cells within lamina propria or connective tissue papillae.[5]

Discussing the epithelial hyperplasia, it is suggested that elongation of the epithelial rete ridges is illusory. It is not a downward proliferation of epithelial cells but rather results from the upward pushing effect of accumulated macrophages towards the epithelium. Thus, it reflects thinning of the epithelium overlying the macrophages in the connective tissue papillae.[4]

Xanthomas represent the accumulation of lipid-rich histiocytes (macrophages) known as foam cells. The lipids in xanthomas are primarily free and esterified cholesterol, but occasionally other sterols and even triglycerides accumulate. Due to high plasma concentration, there is subsequent permeation of lipoproteins through the walls of capillaries. The lipid is taken up by macrophages, which evolve into foam cells. Xanthomas are usually associated with disorders of lipoprotein metabolism.[2] It has also been suggested that degenerative changes in the epithelium lead to the presence of foam cells.[10]

Investigations about matrix metalloproteinase (MMP-2 and -9) and the accumulation of foam cells in VX lesions revealed that the latter is mediated partly by an immune mechanism associated with MMPs, which degrade basal membrane of the epithelium and promote a reciprocal induction between the mesenchymal and epithelial cells.[2] Ide *et al*. have suggested that under synergistic regulation of T cells, macrophage recruitment in the sub-basal mesenchyme and the lysosomal engulfment of epithelial lipids by macrophages are essential in the formation of the VX lesion.[9]

Three histological subtypes (types A, B, and C) have been described based on the texture of the surface epithelium. Lesions of the verrucous type (A) have hyperparakeratosis,

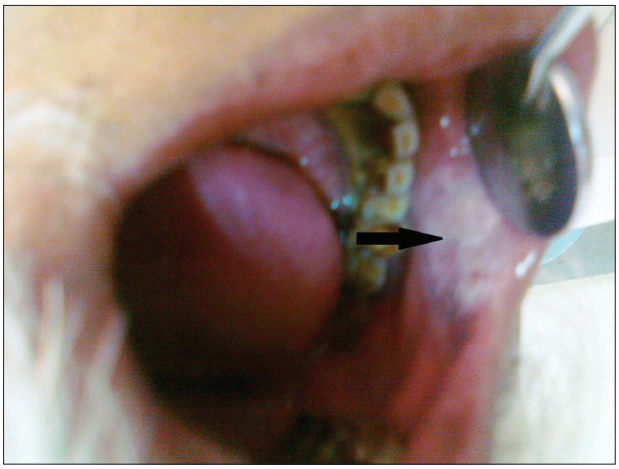

Figure 1: Greyish white lesion on the left buccal mucosa

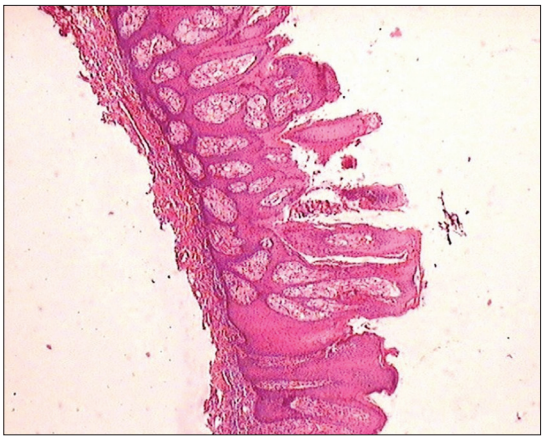

Figure 2: Papillary projections lined by epithelium consisting of core of connective tissue. (H and E) (×4)

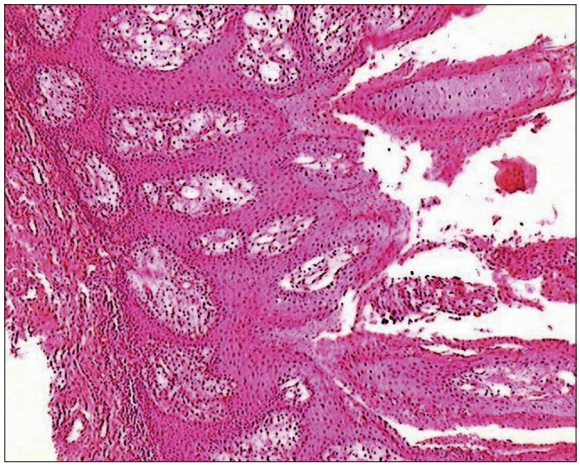

Figure 3: The lamina propria exhibit presence of foam cells, extending up to the lower border of epithelium (H and E) (×10)

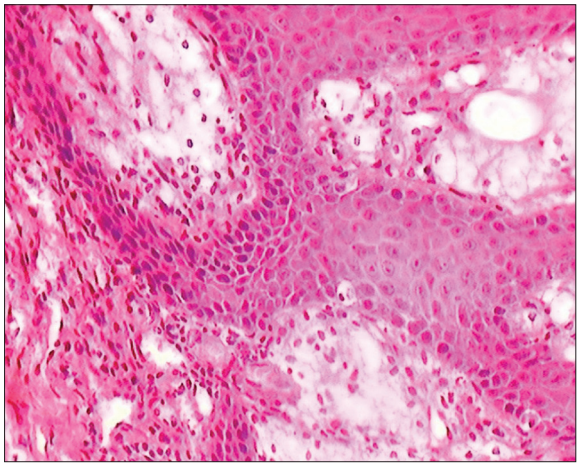

Figure 4: Foam cells showing clear-to-eosinophilic cytoplasm with eccentrically placed nuclei (H and E) (×40)

verrucous type acanthosis, and elongation of the rete ridges. The papillary form (B) has many finger-like projections composed of stratified squamous epithelium containing connective tissue cores. In the flat type (C), mild acanthosis and subtle thin parakeratosis with variable elongation of rete ridges are commonly observed.[9,11]

Because of the level of surface keratinization, its color may vary from white to red. Therefore, oral VX may resemble a squamous papilloma, verruca vulgaris, or a mucosal fibroma. These entities do not contain lipid-laden macrophages and they exhibit varying degree of koilocytic change, which are known to be absent in VX. Because of its verrucous nature, verrucous carcinoma should be also added to the list of differential diagnoses and can be distinguished by the presence of invasive epithelial proliferation, cellular atypia, and the lack of foamy histiocytic infiltrate.[2,8] VX is usually treated by a conservative excision. To date, only three cases of recurrence have been reported.[2,9]

Conclusion

VX is a multifactorial chronic reactive process. The clinical diagnosis may be challenging; however, the histological features are diagnostic and well defined. It is noteworthy that in a small and superficial biopsy, xanthoma cells may be scanty and their presence can be missed, especially if one is unfamiliar with the existence of this lesion. Its recognition and correct diagnosis is critical because VX can occur in conjunction with systemic and cutaneous inflammatory diseases; further clinical assessment for these is required.

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