



Oral Verruciform Xanthoma: A Case Report

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Verruciform xanthoma is a benign growth found on the oral mucosa and occasionally may affect the skin. Clinically, verruciform xanthoma is an asymptomatic lesion. Depending on the amount of keratin present on the surface of the oral mucosa, it can appear as a reddish-gray papillary, flat or a slightly raised, rough lesion. The verrucous structure and exophytic properties of these lesions are similar to those of malignant lesions; therefore, histopathological assessment is mandatory for definitive diagnosis. Herein, we report a case of oral verruciform xanthoma on the lower right labial vestibule and to confirm the diagnosis immunohistochemistry (IHC) was done which showed positive CD 68 macrophages.

Keywords: Xanthoma; immunohistochemistry; CD 68.

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1. INTRODUCTION

Verruciform xanthoma [VX] is an uncommon benign lesion primarily of the oral mucosa with an incidence rate of 0.025-0.05% of all the pathology cases. In 1971, Shafer first described the entity "Verruciform Xanthoma" [1]. The etiopathogenesis of verruciform xanthoma is unknown, though been identified in several parts of the body. Most frequently encountered sites for oral lesion are gingiva, alveolus and hard palate [2]. It is most commonly presented with a verrucous appearance, however in some instances it may appear polypoid, papillomatous, or sessile. It occurs as a small (0.2–2 cm), solitary, asymptomatic, slow growing, white or yellowish red lesion with no sex predilection [3,4]. Histopathological examination is the gold standard for the diagnosis of verruciform xanthomas. Microscopically, these lesions are characterized by the presence of parakeratinized stratified squamous epithelium having papillary or verrucous growth with connective tissue papillae extending up to the surface. The papillae characteristically contain foam cells called xanthoma cells [5]. The treatment of the VX lesion involves local surgical excision and recurrence is rare [4]. The aim of this report is to present a rare case of oral verruciform xanthoma on the lower right labial vestibule of a 52-year-old male along its clinical and pathologic features and treatment modalities.

2. CASE REPORT

A 52 year-old male patient presented with the chief complaint of a painless growth on the right labial vestibule for 4-5 months (Fig. 1). He had a

habit of tobacco chewing for the past 15 years. On clinical examination, whitish pink, exophytic lesion, measuring 1x 1 cm approx. in size, was found over lower right labial mucosa. The lesion was asymptomatic and soft in consistency. Lymph nodes were not palpable. A provisional diagnosis of verrucous hyperplasia was made. After clinical examination an excisional biopsy was done to rule out malignancy. Grossing examination showed verrucous projections with pebbly surfaces (Fig. 2). On histopathological examination, the hematoxylin and eosin (H&E) stained sections showed parakeratinized stratified squamous epithelium with underlying fibro cellular connective tissue stroma. Epithelium showed varying degrees of exophytic proliferation with thin rete ridges and entrapped connective tissue core. Mild dysplastic features such as nuclear hyperchromatism and increased nuclear cytoplasmic ratio were evident. Adjacent papillary connective tissue showed presence of large foam cells. Mild degree of chronic inflammatory cell infiltrate and vascularity was evident throughout connective tissues (Figs. 3 and 4). Deeper section showed salivary gland acini, transverse section of muscle fibers and nerve bundles. Overall features suggestive of Verrucous hyperplasia with mild dysplasia. To eliminate Verruciform Xanthoma, Periodic Acid Schiff (PAS) and IHC staining was performed. PAS staining was found to be negative. The immunohistochemical staining for CD68 was positive for the foamy macrophages. All the foam cells were strongly stained with anti-macrophage antibodies (Figs. 5 and 6). Surgical excision was done under local anesthesia. Postoperative check-up showed no sign of recurrence.



Fig. 1. Whitish pink exophytic growth on right labial mucosa of lower lip



Fig. 2. Grossing of the specimen showing prominent crypts resembling pebbly surfaces (Stereo microscope)

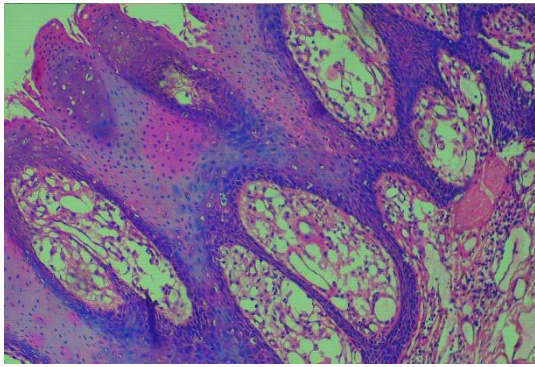


Fig. 3. Photomicrograph showing papillary projections with thin epithelial rete ridges with connective tissue papillae having numerous xanthoma cells (H&E Stain, 10X)

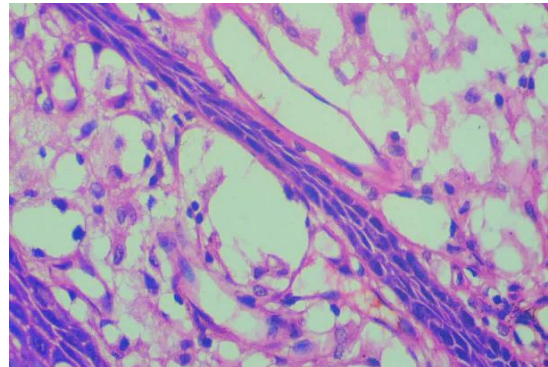


Fig. 4. Photomicrograph showing presence of xanthoma cells (H&E stain, 40X)

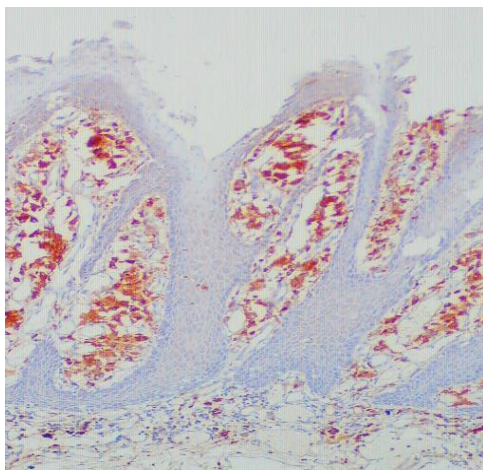


Fig. 5. Photomicrograph showing foam cells with strong immunoreactivity to antibody CD68 (IHC stain, 10X)

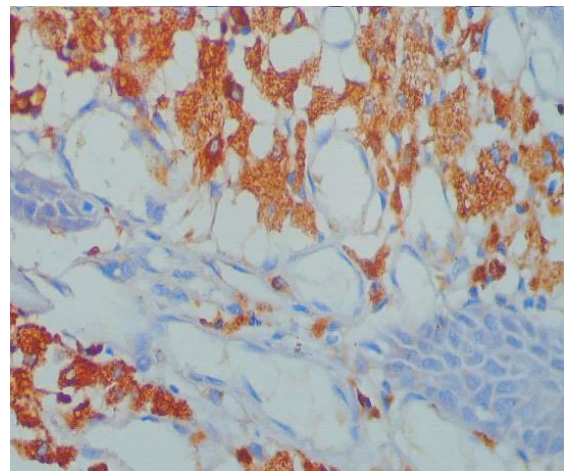


Fig. 6. Photomicrograph showing foamy histiocytes with strong cytoplasmic CD68 immunostaining (IHC stain, 40X)

3. DISCUSSION

VX is a benign epithelial lesion that irrespective of intra or extraoral development can simulate benign and malignant lesions causing diagnostic dilemmas. Extraoral verruciform xanthoma was first described on vulva by Santa Cruz and Martin [6]. The most common intraoral sites of its occurrence are gingiva, alveolar mucosa and hard palate [2]. The etiology still remains obscure. It can be because of the damage to the squamous cells due to trauma, local irritation or infection, which can cause epithelial entrapment and increased epithelial turnover leading to the disease. The epithelial degeneration leads to an inflammatory response and releases lipid material from the degenerated cells [4]. The damaged and degenerating keratinocytes that

move downwards into the papillary dermis and submucosal region are engulfed by dendritic cells, which develop into foam cells [1].

Because of its clinical and histopathological resemblance to human papilloma virus-induced lesions, verruciform xanthoma was believed to be caused by HPV. However, no evidence was found for the presence of HPV in these lesions [7]. Cobb et al. (1976) supported the theory that VX is inflammatory in origin, generally occurs on the masticatory mucosa while some authors interpreted that it is an immune-mediated process [8]. Verruciform Xanthoma is associated with conditions such as pemphigus vulgaris, lichen planus, discoid lupus erythematosus, warty dyskeratoma, epidermal nevus/Congenital hemidysplasia with ichthyosiform erythroderma

and limb defects syndrome, dystrophic epidermolysis bullosa and seborrheic keratosis [9].

Clinically, the lesions of VX presented a papillary aspect, pink to white color, normally ranging between 0.2 to 2 cm. Neville et al.(1980), described that VX may differ from “white and verruciform” to “red and ulcerated” [10]. It may be sessile or pedunculated and can resemble leukoplakia or squamous papilloma. Most commonly occurs in 4th–6th decade of life with equal distribution between both genders. However, V. Raphael et al. (2012) has reported that there is a slight male predilection for Verruciform Xanthoma [3,4,11]. Intraorally, the lesion is usually present on alveolar ridge, gingiva, followed by buccal mucosa, palate, floor of the mouth and lip [9,12]. In our case, whitish pink, exophytic lesion, measuring 1x 1 cm approx. in size, was found over lower right labial mucosa which was asymptomatic and soft in consistency.

Histologically, VX shows three patterns: verrucous or warty (most common), papillary or cauliflower (least common), and flat or slightly raised [13]. The papillary pattern exhibits a finger-like exophytic epithelial proliferation covering thin cores of connective tissue, whereas in the flat pattern, the lesion demonstrates “endophytic” (below the surface) growth. In the flat pattern, abundant foamy cells accumulation can be observed in the lamina propria, thus leading to the rete ridge elongation and thinning of the covered oral epithelium through compression [14]. Sometimes, there is parakeratosis of the hyperplastic epithelium. The rete pegs are thin, elongated and uniform.

The connective tissue papillae between the rete pegs are characterized by massive accumulation of large swollen foam cells known as xanthoma cells, which are restricted to the extension of the rete pegs. The cytoplasm of the foam cells contains tiny PAS-positive granule. The nuclei are small, round and eccentrically placed [15]. Still, controversy over the exact origin of these cells exists. They are suggested to be a lineage of monocytes/macrophages [14]. The lipid found in the xanthoma cells resembles the same as seen in other inflammatory reactions [9]. In our case, parakeratinized stratified squamous epithelium showed varying degrees of exophytic proliferation with thin rete ridges and entrapped connective tissue core. Mild dysplastic features such as nuclear hyperchromatism and increased

nuclear cytoplasmic ratio were evident. Adjacent papillary connective tissue showed presence of large foam cells. Also, mild degree of chronic inflammatory cell infiltrate and vascularity was evident throughout connective tissue. CD68 is a cytoplasmic marker confirming the possible role of macrophages in the formation of foam cells [9]. In our case, the foam cells showed strong CD68 immunoreactivity. The treatment of choice is complete surgical excision which is very effective with no signs of recurrence [16].

4. CONCLUSION

Verruciform Xanthoma is a rare muco-cutaneous lesion because of multifactorial chronic reactive process. It's clinical appearance is not pathognomonic. Histopathological examination and IHC marker CD68 are the paramount for accurate diagnosis. The clinicians should be aware that clinically, verruciform xanthoma may mimic malignancy. Hence, it should be considered in the differential diagnosis of commonly occurring verruco-papillary lesions in the oral cavity.

CONSENT

As per international standard or university standard, patient(s) written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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