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Case Report

Verruciform Xanthoma a Diagnostic Dilemma

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Abstract

Verruciform xanthoma is an uncommon lesion of the oral mucosa having unknown etiology. It accounts for 0.025 - 0.05% of all pathology cases. The age of occurrence varies widely from 2 to 89 years. But most oral cases occur in middle-aged persons with mean age of 40 - 50 years, and no gender predilection is apparent. Apart from the oral mucosa it affects the genital mucosa as well and is considered to be a reactive process rather than a true neoplasm. The lesion commonly has a verruciform appearance, but it may appear polypoid, papillomatous or sessile.

This is a case report of a 58-year-old male patient who was initially diagnosed as speckled leukoplakia with left buccal mucosa but upon histopathological analysis was diagnosed as verruciform xanthoma.

Keywords: *Verruciform Xanthoma; Oral Histiocytosis Y; Verruciform Xanthoma with Buccal Mucosa*

Abbreviation

VX: Verruciform Xanthoma

Introduction

Verruciform xanthoma is an uncommon lesion of the oral mucosa having unknown etiology.

It is also known as inflammatory papillary hyperplasia with foam cell response.

It was first described by Shafer in 1971. Apart from the oral mucosa it affects the genital mucosa as well and is considered to be a reactive process rather than a true neoplasm [2].

The lesion commonly has a verruciform appearance, but it may appear polypoid, papillomatous or sessile [1].

The major distinguishing feature of verruciform xanthoma is the presence of large numbers of lipid-laden foamy histiocytes. These cells are known as xanthoma cells and are limited to the connective tissue papillae in the lesion [2].

It is often misdiagnosed on clinical examination as a papilloma and that along with verruca vulgaris, verrucous carcinoma and squamous cell carcinoma can be included in the differential diagnosis [5].

Excision of complete lesion is the treatment and recurrence is rare.

Case Report

A 58-year-old male patient reported to the Department of Oral medicine and Radiology with the chief complaint of missing teeth in upper and lower back region.

Patient had a positive habit history tobacco chewing since 25 years which he kept in the right buccal vestibule.

On examination there was presence of a diffuse, yellowish pink, raised lesion on left buccal mucosa surrounded by melanosis.

It was approximately 1.5 x 3 cm in size seen extending superinferiorly from line of occlusion to depth of the vestibule and anteroposteriorly from 33 region to 37 region.

It had a papillomatous surface was nontender, non-scrapable and soft to firm on palpation.

A provisional diagnosis of speckled leukoplakia was given. Verrucous leukoplakia was considered under differential diagnosis.

The patient was advised to quit tobacco smoking habit completely. Patient was advised a complete blood count and an incisional biopsy was planned.

The excised specimen was sent for histopathological examination which revealed, accumulation of numerous large macrophages with foamy cytoplasm which were typically confined to connective tissue papillae.

Moderate chronic inflammatory cell infiltrate were noted adjacent to the epithelium along with many dilated and engorged blood vessels. Deeper region showed adipose tissue.

This picture was characteristic for verruciform xanthoma, and therefore a final diagnosis of the same was made.

Post-excision healing was uneventful.

Figure 1: *58 year old male patient.*

Figure 2: *Lesion with left buccal mucosa.*

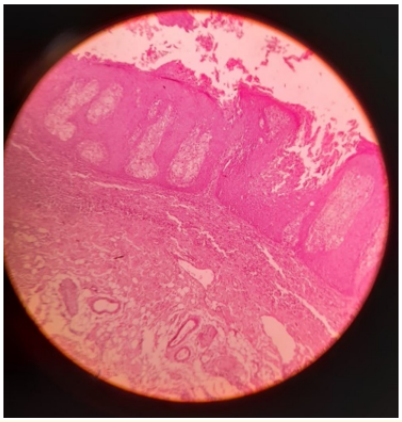

Figure 3: *Photomicrograph of biopsied tissue.*

Discussion

Verruciform xanthoma is uncommon, accounting for 0.025 - 0.05% of all pathology cases [2].

The age of occurrence varies widely from 2 to 89 years. But most oral cases occur in middle-aged persons with mean age of 40 - 50 years, and no gender predilection is apparent [2].

In the biological profile of oral verruciform xanthoma [VX], based on a worldwide literature survey of 282 cases, it was seen that oral lesions of VX were mostly seen in males, with a male-to-female ratio of 1.1: 1 [4].

Below the age of 50, the male-to-female ratio was 1.6:1; and this ratio reversed after the age of 50 in favour of females, when the male-to-female ratio was 0.8:1 [4,12].

The most common location for VX reported is masticatory mucosa [73.4%], with gingival margin accounting for 85 [49.1%] cases. Next in order of frequency were hard palate [n = 25] [of these, 17 cases were contained in the 85 cases from gingival margin]; tongue [n = 16, of which 8 were localized to the dorsum/lateral border]; buccal mucosa/vestibular fold [n = 12]; floor of the mouth [n = 10] and attached gingiva/alveolar mucosa [n = 9]; soft palate [n = 8]; lower lip [n = 5]; junction between hard and soft palate [n = 3] [4,12-15,18,19].

The size of oral VXs was usually between 0.2 and 2.0 cm [13-15,17-19] with the exception of the largest lesion measuring 4.0 × 1.5 cm documented by Graff., *et al*. [16,19].

Verruciform xanthoma outside oral mucosa is usually associated with other conditions like lymphoedema, epidermal naevi, discoid lupus erythematosus and congenital hemi dysplasia with ichthyosiform erythroderma and limb defect (CHILD) syndrome [2].

A few cases have also been reported to be associated with disorders of lipid metabolism [1,9].

The aetiology of VX is largely unknown, although a myriad of aetiological factors have been implicated. It has been attributed to trauma and inflammation, as it is more commonly seen in masticatory mucosa [3,8].

According to Zegarelli., *et al*. inflammation due to local irritant or trauma causes an increase in epithelial cell turn-over rate and degeneration. Lipid is released as degeneration by-product, which is scavenged by macrophages, resulting in formation of foam cells and hence the verruciform xanthoma lesion [9].

An immunological cause has been suggested due to the presence of Langerhans cells within the lesions [3,8].

It has also been proposed that epithelial entrapment with degeneration and accumulation of lipids leads to the formation of xanthoma cells or foam cells [9], or that the pathogenesis involves excessive accumulation of lipids, which are then gathered by macrophages [3,8].

The lipid found in the xanthoma cells is said to be the same as seen in other inflammatory reactions [9].

Hyperkeratotic or dysplastic oral lesions can undergo degenerative changes to form a verruciform xanthoma [11].

The treatment of choice is complete surgical excision which is very effective with no recurrence [1].

When managed by local surgical excision no malignant metamorphosis has been detailed [20].

Although a recurrent verruciform xanthoma of the vulva was reported of a 30-year-old woman, 8 years after the initial treatment [6].

Histopathological examination of the biopsies should be performed to distinguish VX from other verrucous lesions [1,7].

Conclusion

Although the verruciform xanthoma has an asymptomatic course it is imperative to study it's etiopathogenesis and know the actual cause of this lesion.

Conflict of Interest

No conflict of interest.

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