



VERRUCOUS XANTHOMA OF TONGUE– AN UNUSUAL CLINICAL AND SITE PRESENTATION

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ABSTRACT

Verrucous xanthoma is a rare, benign, asymptomatic entity in the oral cavity with most cases occurring in the gingiva. Non-specific clinico-radiological features, makes the diagnosis a challenging task. We describe an exceptionally rare and unusual case of verrucous xanthoma of tongue arising in a middle aged chronic smoker. The sessile, papillomatous growth measured 1.5 x 1.5 cm. Clinical differential diagnosis of a verrucous lesion was considered and diagnosis was confirmed on histopathological examination, supported by immunohistochemical positivity for CD 68. Owing to paucity of literature, awareness of this uncommon entity is essential amongst pathologists and clinicians. Classical histopathological features along with clinical correlation is imperative for a decisive diagnosis.

KEYWORDS : benign; oral; verrucous; xanthoma

INTRODUCTION

Verrucous xanthoma is a rare entity in the oral cavity with most cases occurring in the gingiva. [1] It is a benign and asymptomatic mucocutaneous lesion, the pathophysiology of which still remains elusive.

Owing to non-specific clinico-radiological features, diagnosis of this uncommon lesion in the oral cavity is a challenging task. Classical histopathological features along with clinical correlation is imperative for a decisive diagnosis.

Keeping in mind the paucity of literature, we describe a rare case of verrucous xanthoma of tongue arising in a middle aged man. The diagnosis was confirmed on histopathological examination and supported by immunohistochemistry.

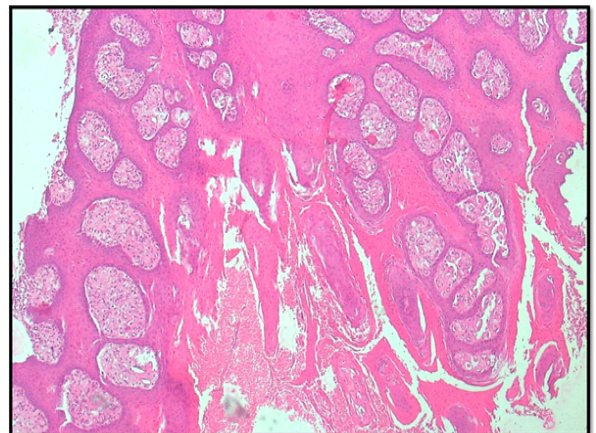
CASE REPORT

A 44 years old male patient with a swelling over his lower lip. The swelling was noticed 6 month back measuring 1.5 x 1.5 cm. He was a chronic smoker (10 pack years). There was history of local trauma. Local examination revealed a sessile, papillomatous growth on the lower lip measuring 1.5 x 1.5 cm. Oral hygiene was poor and teeth were tobacco stained. All the routine investigations including complete haemogram, blood sugar, lipid profile, liver function tests, kidney function tests and urine analysis were within normal limits. With a clinical differential diagnoses of papilloma versus verrucous lesion, an excisional biopsy was performed.

We received an excisional biopsy in Department of Pathology and histological examination revealed stratified squamous epithelium lined tissue showing epitheliomatous hyperplasia and papillomatosis (Figure 1). No dysplasia was noted.

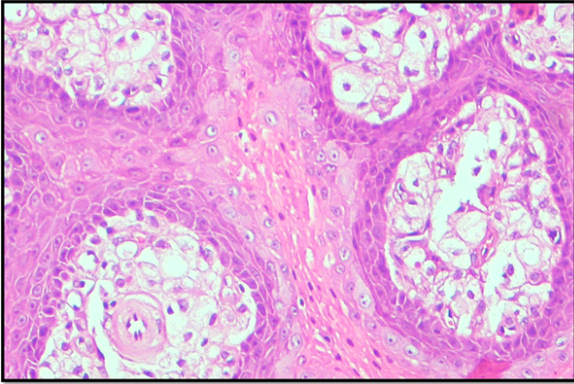
Figure 1: Photomicrograph showing stratified squamous epithelium lined tissue showing epitheliomatous hyperplasia and

papillomatosis. The subepithelium shows numerous foam cells (H&E, 200X)



The subepithelium showed xanthoma cells having moderate to abundant amount of foamy cytoplasm and eccentric nucleus (Figure 2). In some of the foamy cells the nucleus was centrally placed. Admixed with these foamy cells were chronic inflammatory cells comprising predominantly of lymphocytes. These xanthoma cells/ foamy cells showed immunohistochemical positivity for Cd68.

Figure 2: Photomicrograph shows presence of foam cells in the subepithelium having moderate to abundant amount of foamy cytoplasm and eccentric nucleus (H&E, 400X)



A final histopathological diagnosis of verrucous xanthoma was rendered. Post-operative period of the patient was uneventful. The patient was followed up regularly and is healthy at one year follow-up.

DISCUSSION

Oral verruciform xanthoma was first described by Shafer in 1971 as a reactive lesion. Since then it has been reported in the oral cavity most commonly involving the gingiva. The prevalence of OVX is unknown, a relative frequency rate of 0.025% to 0.05% is considered in the literature. [2] Albeit rare, scattered case reports involving hard palate, tongue, buccal mucosa, floor of the mouth, and soft palate are also on record. [3] Our patient showed uncommon localisation of tongue. An extensive literature search revealed approximately 40 cases of verrucous xanthoma of tongue. The patients reported an age range of 2-80 years with a slight female preponderance.

The etiopathogenesis of this lesion remains unclear. Ide F et al suggested possible oral aetiological agents, such as a mechanical stimuli, tobacco, alcohol, drugs, allergic substances, foodstuffs, and dental materials. [4] However reaction response to trauma remains the most accepted explanation. [2] Epithelial breakdown following trauma elicits an inflammatory response comprising predominantly of neutrophils along with accumulation of epithelial breakdown products. The latter comprising predominantly of lipid is scavenged by macrophages which are seen histopathologically as foam cells. Contrary to skin xanthomas, oral counterparts are not associated with metabolic disturbances of lipids, OVXs are not related to any generalized disease. [5]

Furthermore, most investigators have not found any evidence for the presence of human papilloma virus in these lesions. Association with other inflammatory conditions including pemphigus vulgaris, lichen planus, discoid lupus erythematosus, warty dyskeratoma, dystrophic epidermolysis bullosa, and seborrheic keratosis is on record. [6]

Histopathologically, verrucous xanthoma shows three patterns i.e verrucous (most common), flat, and papillary (least common). [7, 8] The diagnostic feature remains presence of lipid laden foam cells which show immuno-reactivity for CD 68. This further confirms the monocytic/ macrophage lineage of origin of these cells.

The treatment of choice is complete surgical excision of the lesion with excellent prognosis and no recurrence. However three cases of oral verrucous xanthoma involving the hard palate have shown recurrence.

To conclude, verrucous xanthoma of tongue is rare. Clinical diagnosis of this rare lesion at an uncommon site is challenging. Albeit rare, it must be kept in mind as a differential diagnosis by the clinicians and pathologists while dealing with an intra-oral verrucous lesion.

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