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Oral verruciform xanthoma - A rare case report

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Abstract

Oral verruciform xanthoma is a rare lesion with predilection for oral mucosa having very few publications in the literature. It is an innocuous lesion, reddish in colour chiefly affecting the masticatory mucosa occasionally showing hyper keratosis microscopically. This particular case shows Oral Verruciform Xanthoma in lining mucosa which is extremely rare. Our case had papillary projections with hyper keratosis and many foam cells limited to the papillary connective tissue. Clinically this lesion may resemble pre-neoplastic or neoplastic lesions so to rule out the confirm diagnosis to do a biopsy is mandatory.

Keywords: Oral verruciform xanthoma, foam cells, hyperkeratosis, papilloma, squamous cell carcinoma.

Introduction

Verruciform Xanthoma was first described by Shafer in 1971 on the oral mucosa as an uncommon benign lesion that has a verruciform (wart-like) appearance but may also appear polypoid, papillomatous or sessile.[1] The term xanthoma is derived from the Greek word Xanthos, meaning yellow. Xanthoma is described as an irregular

yellow patch or nodule caused by deposition of lipids mostly in reticulo-endothelial cell. [2]

Oral Verruciform Xanthoma (OVX) presents itself as an asymptomatic, solitary, sharply demarcated lesion, having papillary, granular or verrucous surface, located commonly on the oral masticatory mucosa (gingiva, palate) however sites like floor of the mouth, lining mucosa & tongue are rarely affected. [3] Lesion usually is of red or pink color. It is commonly seen in males with the most being in 5th to 7th decade of life. Most OVX have been misdiagnosed clinically as papilloma and occasionally as verrucous carcinomas or Squamous Cell Carcinoma. [4] Histopathologic ally OVX shows papillary or verrucous proliferation of hyper parakeratotic squamous epithelium, and aggregation of “foamy or xanthoma cells” within the connective tissue. The xanthoma cells are macrophages of varying size with eccentrically placed nuclei; they are mostly confined to the connective tissue papillae. [3]

The aim of present study is to report a case of OVX affecting lining mucosa (buccal mucosa), reinforcing the importance of considering this benign lesion in the differential diagnosis of papillary lesions of oral cavity.

Case Report

A 54-year-old male patient presented with a white lesion on right buccal vestibule since 1month.There was no associated pain, difficulty in mouth opening and mastication. He had a history of smoking since 15 years and later he quit the habit since 1 year. He also had a habit of keeping gutkha in right mandibular buccal vestibule for past 2years. There was no associated medical history. There was no history of trauma. On intra oral examination a well circumscribed, sessile, papillomatous white keratotic lesion of approximate size 1.5cm X 2.5cmwas present on right mandibular buccal vestibule. It was irregular in shape with slightly raised borders and surrounding mucosa showed slight erythema along with some white keratotic flecks [Fig. 1]. On palpation lesion was non-tender, non-scrapable, slightly firm in consistency and no induration was present. Based on clinical findings a provisional diagnosis of Leukoplakia was made. Excised specimen was sent for histopathological examination.

The H&E-stained section showed Para keratinized stratified squamous epithelium with long elongated and uniform rete ridges with parakeratotic plugging [Fig. 2-3]. Few areas in epithelium showed mild acanthosis. Large vacuolated foam cells (xanthoma cells) of various sizes with eccentrically placed nuclei were seen confined to connective tissue papillae [Fig. 4]. The underlying connective tissue stroma showed a moderate chronic inflammatory cell reaction. Based on these findings the diagnosis of verruciform xanthoma was given. Postoperative follow up showed no sign of recurrence.

Figures

Fig. 1: White Papillary lesion on right buccal vestibule

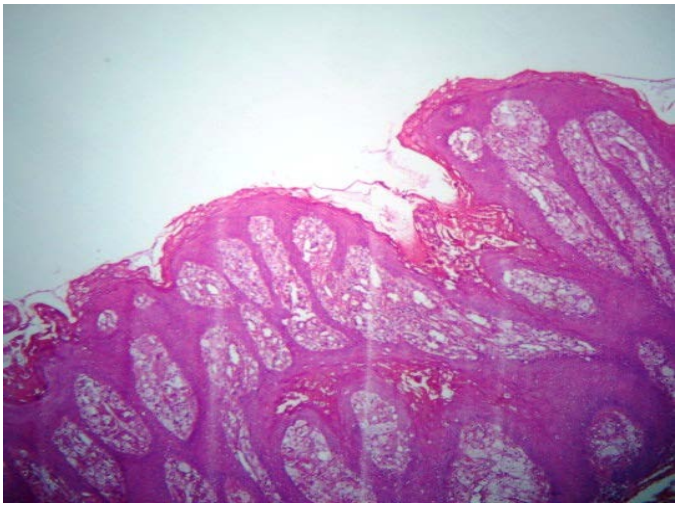

Fig 2: Pap illary or verrucous proliferation of squamous epithelium associated with hyper papa keratosis. (Scanner view)

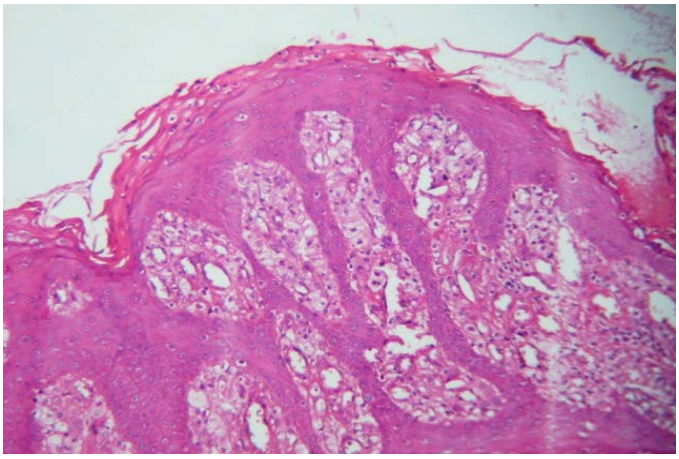

Fig 3: Numerous foamy cells confined to papillary connective tissue. (10X view)

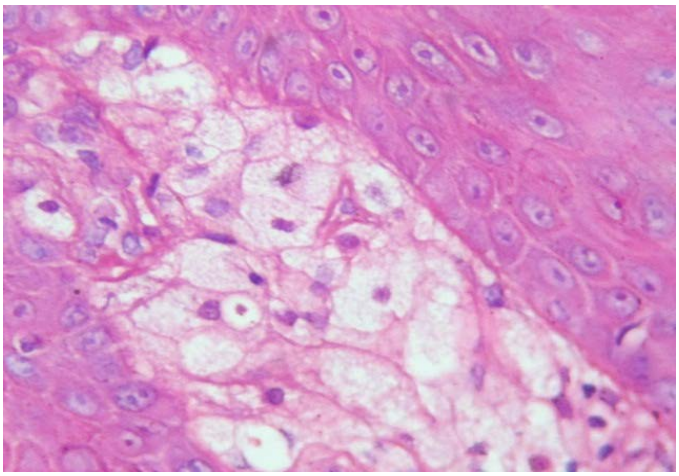

Fig 4: Path gnomic Xanthoma cells confined to papillary connective tissue. (40X view)

Discussion

Verruciform xanthoma is an uncommon lesion that usually occurs on the oral mucosa and is of unknown etiology and uncertain nature. Apart from the oral cavity, lesion affects genital mucosa like vulva, penis and on the skin of thigh and perineum. It is considered to be reactive process rather than a true neoplasm. [1] Philipsen et al reviewed that its more common in males with female: male ratio 1:1.4 with an age range of 16-87 years with mean age of 40-50 years. Majority of the cases were found on the masticatory mucosa followed by floor of the mouth, lower lip, soft palate and labial mucosa. [5]

OVX occurs as sessile or pedunculated solitary lesion with a rough or pebbly surface either normal or reddish in color but sometimes may also appear pale or hyperkeratotic. It may be small or large with size ranging from 2mm to 4cm in diameter. [1] The margins are sharply demarcated and is slightly raised above the surrounding oral mucosa. The center of the lesion may be depressed, Cuphead or crateriform with or without ulceration. Now parast et al. described verruciform xanthoma into three different types.

1. A warty or verrucous appearance
2. A papillary or cauliflower architecture
3. A slightly raised or flat lesion. [2]

Most of the lesions are asymptomatic except when localized to areas where trauma occurs by toothbrushing in which case tenderness, pain and slight bleeding may occur. Symptoms may also be present if the OVX is associated with other oral conditions such as leuoplakia, pemphigus vulgaris, focal acantholytic dyseratosis, apthae etc. The majority of cases are detected during routine oral examination. Underlying bone in OVX does not show any significant radiographic changes. [5]

Etio-pathogenesis of OVX is still not clear. It does not seem to be associated with systemic abnormality in lipid

metabolism though reactive, immunological, infectious and genetic mechanism has been considered. [4] van der Waal et al suggested it to be a reactive process initiated by inflammatory reaction rather than a true neoplasm. [6] The origin of foam cells is still not clear and was thought to be of epithelial/neural/dermal-dendritic in nature. Immunohistochemical studies suggest that foam cells originate from macrophages because of their strong immunoreactivity to CD-68 antibody. [7] Zegarelli *et al*. proposed that local trauma causes epithelial entrapment which initiates inflammatory reaction. This leads to epithelial breakdown and release of lipid substance which is engulfed by macrophages. This theory was acceptable as OVX is most often found on the masticatory mucosa which is subjected to trauma and subsequent inflammatory reaction. [8]

Histopathologic ally OVX shows Para keratinized stratified squamous epithelium showing elongated rete pegs of comparatively uniform depth. [5] The characteristic finding is the presence of foam cells or lipid laden macrophages in connective tissue papillae limited to the papillary lamina propria. Xanthoma cells distinguishes the lesion from squamous papilloma, verruca vulgaris, fibroma and verrucous carcinoma, squamous cell carcinoma. [9]

OVX can be treated by topical steroids, carbon-dioxide laser, cryotherapy, wire loop electro-sectioning and radiotherapy but complete surgical excision is still the main choice of surgeons. [10] The lesion does not recur after complete surgical excision and in our patient also no recurrence was reported.

Conclusion

Knowledge of the most frequent lesions and their prevalence is of utmost importance in the process of diagnosis. OVX, since 1997 is classified by the WHO as benign lesions that resemble microscopically oral squamous cell carcinoma and oral verrucous carcinoma. The presence of xanthoma cells is the only diagnostic criteria by which we can rule out verrucous carcinoma and prevent overtreatment and excessive surgery. If correctly diagnosed, OVX is treated simply by total excision, without recurrence. But if misdiagnosed as a malignancy, overtreatment with aggressive surgery is the main danger that a surgeon will have to face.

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