Oral Verruciform Xanthoma- The Shape – Shifter

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Abstract: **Verruciform xanthoma (VX) is an uncommon benign mucocutaneous lesion with unknown etiology. VX occurs predominantly in oral cavity which also occasionally affects skin and genital mucosa. It was first reported in oral cavity in 1971. Clinically it present as a sessile or pedunculated growth which is usually asymptomatic in nature. The definite diagnosis is always made during a histologic examination. Histologically it is characterized by presence of Para keratinized epithelium showing papillary or verrucous growth. The papilla consists of aggregates of lipid laden foam cells called Xanthoma cells. Here we describe the case report of Verruciform Xanthoma occurring on the mandibular alveolar mucosa in a 58 year old male patient mimicking a papillary lesion.**

Keywords: **Oral verruciform xanthoma, Foam cells, Xanthoma cells, Papilloma, Oral cavity**

INTRODUCTION

Oral verruciform Xanthoma (OVX) is a benign mucocutaneous uncertain lesion of the oral cavity which was first described by Shafer but which may also affect skin and other mucous membranes.[1]. It occurs most commonly around 40- 60 years of age with a slightly higher incidence in males [1].Its aetiology is unknown, but it is believed to be a reactive lesion, possibly a response to trauma or chronic inflammation [5]. Clinically, the lesion appears as a papillary or verrucous appearance which may lead to a misdiagnosis of papilloma. It appear as small , slow growing pink or whitened lesion which is either sessile or pedunculated and it is mostly asymptomatic in nature.[6]. Microscopically, the characteristic feature is epithelial proliferation and accumulation of foamy macrophages [8].

CASE REPORT

A 58 year old male patient reported to the department of Oral medicine and radiology with a chief complaint of pain in the lower front tooth region for past 1 week. The patient had no relevant medical history and was non- smoker. Extra oral examination showed no obvious abnormality. Intra oral examination shows blackish discolouration involving the mesio

proximal surface of crown and root in relation to 33, which was sensitivity on probing and tender on percussion. Incidentally on inspection a sessile growth of size 0.5 x 0.5 cm which was pinkish in colour present in the alveolar mucosa in relation to partially edentulous 32 tooth region. Clinically, the surface of the lesion appear as multiple small papillary projections with no evidence of ulceration. On palpation it was soft in consistency with mild tenderness. Bleeding on probing was evident. [Figure:1]

Figure 1: Intra oral Examination showed a growth in alveolar mucosa in relation to partially edentulous 32 tooth region

Moderate supra gingival calculus and stains with generalized apical migration of marginal gingiva from CEJ was present.

The chief complaint was provisional diagnosed as apical periodontitis in grossly decayed 33. The lesion was provisional diagnosed as Squamous Papilloma.

Clinical Photographs were taken and excisional biopsy was performed. The excised specimen was given to Department of oral Pathology for histopathological examination.

On gross examination, 2 bits of soft tissue, which were greyish white in colour, measuring 8x8x4mm and 6x6x4mm in size respectively.[Figure :2]

Figure 2: Gross imaging of 2 bits of soft tissue

On histological examination, H and E stained section shows papilloferous proliferation of surface epithelium in association with an inflamed fibro vascular connective tissue exhibiting diffuse mixed inflammatory cell infiltrate. The surface epithelium in the papilloferous projections are atrophic and connective tissue papillae exhibits foamy macrophages.[Figure: 3 A and B] Based on these histopathological features final diagnosis of verruciform xanthoma was made.

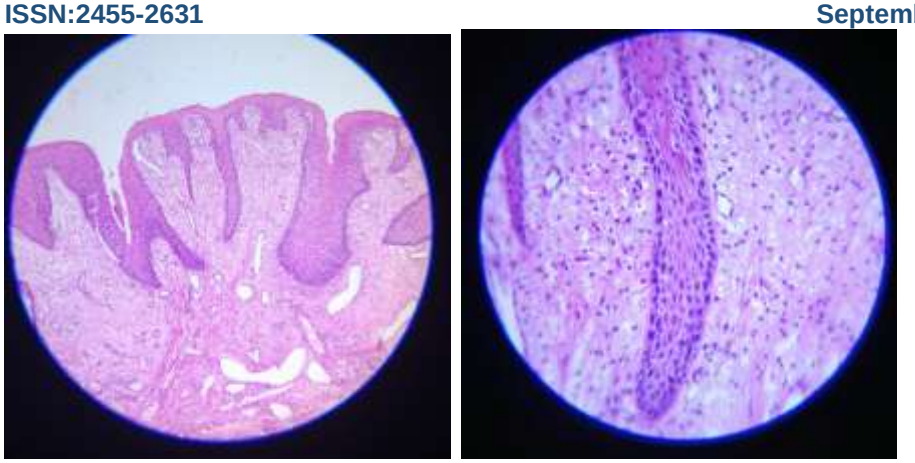

Figure 3: (A) H and E – stained section showing elongated rete pegs. (B) H and E stained section showing presence of large xanthoma cells with foamy macrophages

OUTCOME AND FOLLOW UP

Excisional biopsy was performed, the tooth 33 was extracted and the 6 month review and follow up shows no evidence of recurrence.

DISCUSSION

Verruciform Xanthoma is a hyperplastic condition of the epithelium of the mouth, skin and genitalia, with a characteristic accumulation of lipid- laden histiocytes beneath the epithelium. Shafer described 15 cases of this condition in the oral cavity and coined the term 'verruciform xanthoma'.[4] The term xanthoma is derived from the Greek word xanthos, meaning yellow. Originally, xanthoma was used to define a yellowish, slightly raised or flat lesion occurring on the skin, propounding an underlying illness. Oftentimes, lipid material accumulates in reticuloendothelial cells in various sites of the body. The phenomenon of xanthoma occurs beneath the epithelial surface, thus giving a yellow tan hue to the cutaneous lesions.[7] The first report of involvement of an extra–oral site was described by Santa Cruz in 1979 where he reported two cases of VX in the vulva. [4]

Although verruciform xanthoma is a papillary lesion, human papillomavirus does not appear to play a role in its pathogenesis. The etiology is not well defined, but it can be because of the damage to the squamous cells due to trauma, irritation or infection, which can cause increased epithelial turnover leading to the disease.[8] The epithelial breakdown leads to an inflammatory response and a subsequent release of lipid material from the degenerated cells.[6]

OVX occurs in in the 4th to 6th decade of life ,with a female-to-male ratio of 1:1.6 and the ratio reverses to 1:0.8 after the age of 50 years.[7] Almost half of the intra-oral lesions occur on gingiva (57.4%), followed by the tongue (10.3%), hard palate (7.1%), buccal or vestibular mucosa (6.7%), floor of the mouth (4.6%) and soft palate (3.2%) and lip. In the present case, the mandibular alveolar mucosa is involved, which is a relatively uncommon site for its occurrence.[2]

The lesion appear as a slow growing, well-demarcated, soft, asymptomatic, and are smaller than 2 cm in greatest diameter.[2] It more often occurs as a solitary lesion, although multiple lesions have occasionally been reported. The colour ranges from white to red with/without a yellow tinge and the surface of the lesion can be verrucous, papillary or cauliflower like, and flat[1]. In the present case the lesion resembles small papillary (finger like) projections from mandibular anterior alveolar mucosa.

Histologically, OVX shows three patterns: Verrucous, Papillary and Flat[2]. It is characterized by a squamous epithelial surface of varying morphology covered with parakeratin, showing elongated rete pegs of relatively uniform depth. 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. [8]The most pathognomonic feature of this lesion is the presence of large swollen “foam cells” or xanthoma cells, which fill the connective tissue papillae between the rete pegs. The rete pegs are elongated, uniform, and thin, with deep central keratinized clefts and keratin plugs. These are numerous large macrophages with foamy cytoplasm, which typically are confined to the connective tissue papillae between the elongated rete-ridges.[9] The cytoplasm of the foam cells contains tiny PAS-positive granule. The nuclei are small, round and eccentrically placed. However, no evidence of dysplasia.[8]

In this case, the patient was 55- year old male with an asymptomatic , small multiple papillary projections seen at the alveolar mucosa in relation to partially edentulous 32 tooth region. The left canine was grossly decayed. Bleeding on probing was present. The clinical features led the clinician for the provisional diagnosis of squamous papilloma. Then Excisional biopsy was performed. The general histopathologic appearance of this case shows demonstrates a broad mixed inflammatory cell infiltration and papilloferous surface epithelium growth in conjunction with fibrovascular connective tissue inflammation. The

papilloferous projections' surface epithelium is atrophic, and the connective tissue papillae show foamy macrophages.

DIFFERENTIAL DIAGNOSIS

Due to the nonspecific clinical aspect of oral verruciform xanthoma (OVX), the clinical differential diagnosis usually includes lesions with similar characteristics especially the rough surface, such as verruga vulgaris, verrucous leukoplakia, and verrucous carcinoma[9]

Histologically, the differential diagnoses included those related to the squamous proliferation (eg, squamous dysplasia, human papillomavirus, squamous cell carcinoma, fungal infections) and diagnoses related to the foamy macrophages (xanthoma, granulomatous conditions, eg, sarcoidosis). [9]

TREATMENT

OVX is a benign mucosal swelling and excision is usually curative, although recurrence has, rarely, been reported.

CONCLUSION

Verruciform Xanthoma is a rare muco-cutaneous lesion which is usually harmless with non- diagnostic clinical appeaeance. Histologically it is characterized by presence of foam cells within the connective tissue papillae. In conclusion, Verruciform Xanthoma (VX) is a localized lesion whose correct diagnosis requires surgical excision and careful histological examination. Treatment of VX consists of a simple surgical excision and the prognosis is excellent.

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