A rare case of Verruciform Xanthoma

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ABSTRACT:
Verruciform Xanthoma is an uncommon benign lesion. The most common presentation occurs in the oral cavity; however, it has also been described in other sites, especially the penis. The case report describes a patient presenting with a small asymptomatic sessile growth in lower anterior gingiva. The subsequent patient management, histopathological and Immunohistochemical diagnosis and a review for the same is discussed.

INTRODUCTION
Verruciform Xanthoma was first described by Shafer in 1971 [1]. It occurs mainly in the oral mucosa. Extra oral involvements have been reported on the vulva, scrotum, penis, anal region and extremities [2]. In 1981, Kraemer et al reported the first verruciform xanthoma case arising on the penis [3]. Verruciform xanthomas of the oral cavity are usually encountered in the middle aged persons. There is no sex predilection [4]. Most cases of Verruciform Xanthoma have been misdiagnosed solely on clinical basis, and therefore histopathological diagnosis is necessary [2].

CASE REPORT
A 30 yrs old male patient reported to Dept. of Oral Medicine and Radiology at Dr. D.Y. Patil Dental College & Hospital, Pimpri, Pune, with a chief complaint of a small asymptomatic growth in lower anterior region and bleeding from gums in the same region since 2 months. No past dental history of any similar complaints. Patient had no relevant medical history.

CLINICAL FEATURES
On intra oral examination, a small approximately 0.5 x 0.5 cm, sessile, cauliflower like growth was seen in the lower anterior gingival region in between two central incisors (Fig 1). The growth was pinkish red in color. Heavy stains and calculus deposits were noted and there was gingival recession and gingival inflammation seen with same region. There was a grade one mobility with both the central incisors and interdental spacing seen between centrals (31 and 41) and left central and lateral incisors (31 and 32).

After history and intra oral examination, provisional diagnosis of Oral Papilloma was established.

Fig 1 - Small approx. 0.5 x 0.5 cm, sessile cauliflower like growth seen in the lower anterior gingival region in between two central incisors.
RADIOGRAPHIC FEATURES

Intra Oral Periapical radiograph of lower anterior region reveals severe horizontal bone loss and radiopaque interproximal calculus (Fig 2).

Fig 2 – Intra Oral Radiograph

DIFFERENTIAL DIAGNOSIS

1. Pyogenic granuloma
2. Peripheral Giant cell granuloma
3. Common Wart
4. Condyloma acuminatum
5. Focal epithelial hyperplasia
6. Peripheral ossifying fibroma
7. Eosinophilic granuloma

FINAL DIAGNOSIS

After complete oral prophylaxis, excision of the lesion was performed under local anesthesia after sensitivity test. Excision was performed in two pieces (Fig 3) and the sample was sent for histopathological examination. Histopathologic examination of hematoxylin and eosin stained slide showed, hyperparakeratotic stratified squamous epithelium without dysplasia arranged in a papillary architecture. The underlying mature connective tissue revealed presence of large foam cells in the papillary area. The cytoplasm of foam cells seemed to be granular in nature. The overall histopathological features were suggestive of Verruciform Xanthoma (Fig 4).

Immunohistochemical staining using antimacrophage antibody – CD 68 showed the foam cells as monocyte/macrophage lineage, confirming the diagnosis of a verruciform xanthoma (Fig 5 and Fig 6).

Fig 3 - Excision of the lesion in two pieces

Foam cells are strongly positive for CD-68 seen in connective tissue. Distribution of CD-68 forms a delicate network delineating small vessels.

Follow up was done after 15 days (Fig 9) and the patient was sent for further periodontal treatment.
Fig 5 – Scanner view

Fig 6 – High Power view

Fig 7 -  PAS without diastase

Fig 8 -  PAS with diastase

Fig 9 -  Follow up after 15 days
DISCUSSION

Verruciform Xanthoma is an uncommon lesion that usually occurs on the oral mucosa of middle-aged persons or on the scrotum of middle-aged to elderly Japanese men.

The most common site for Verruciform Xanthoma is the oral mucosa. Lesions that occur elsewhere usually arise on the perineum or on the skin with some predisposing factor, such as lymphedema or an epidermal nevus. It is also known as Histiocytosis Y.[4]

The pathophysiology of Verruciform Xanthoma is unknown. Many authors consider it to be a reactive process rather than a true neoplasm. Damage to the squamous cells with increased epithelial cell turnover, leading to the deposition of epithelial cell debris that is engulfed by macrophages in the corium may lead to the development of this lesion. Patients usually present with a history of an asymptomatic or tender lesion on the skin or mucosa.[6]

The clinical appearance of a Verruciform Xanthoma is not diagnostic; the diagnosis is almost always made at histologic examination. Depending on the nature of the individual lesion, Verruciform Xanthoma may clinically resemble any Verrucous, papillary, or lichenoid oral lesion, particularly any such lesion that is also hyperkeratotic. It is frequently misdiagnosed at clinical examination as a papilloma.[6]

It can occur at any site and is more frequently found on the gingiva or alveolar ridge followed by buccal mucosa, palate, floor of mouth, lip and lower mucobuccal fold; it has got normal or red in color but sometimes pale or hyperkeratotic with a rough pebbly surface and either a sessile or pedunculated base.[4]

Lesions have also been found in the oral cavity in association with lichen planus, pemphigus vulgaris, oral bullae, carcinoma in situ or frank squamous cell carcinoma; the lesions were also found in a bone marrow transplant recipient.[11]

The most striking and characteristic histologic feature of the Verruciform Xanthoma is the presence of large foam cells in the connective tissue papillae. These cells characteristically fill the entire papilla but only rarely extend beyond the base of the papilla. Most or all of the papillae are involved with these cells, which occasionally may also be seen in the epithelium (i.e., epidermis, mucosa).[6]

Ultrastructurally, most studies have concluded that the foam cells in verruciform xanthoma are fat-laden macrophages. Immunohistochemical analysis revealed these cells to be positive for CD68.

They are also periodic-acid-Schiff (PAS) positive and diastase resistant, indicating that the PAS-positive material is not glycogen.[6]

Local surgical excision is almost always curative. The prognosis is excellent after local surgical excision. Recurrence is rare.[6]

REFERENCES