CASE REPORT

Verruciform Xanthoma Associated With Oral Submucous Fibrosis - A Report of An Unusual Case With Literature Review

Sujoy Ghosh[1], Sunita Gupta[2], Khushboo Singh[3], Aadithya B Urs[4]

ABSTRACT

Introduction:Verruciform Xanthoma (VX) is a rare benign inflammatory mucocutaneous lesion, predominantly occurring in oral cavity. Clinically the lesion may present as papule or a single plaque showing verrucous or papillomatous mucosal growth with color varying from reddish pink to grey. Histopathological examination is essential for diagnosis and is characterized by epithelium showing papillary or verrucous growth with thin rete pegs and connective tissue papillae consisting of foam cells known as *Xanthoma cells*. Case report: we are describing a case of verruciform xanthoma on right buccal mucosa in 25 years male patient in association with oral submucous fibrous (OSMF) with its clinical and histopathological features. Conclusion: Verruciform xanthoma must be differentiated from various hyperplastic lesions, those having potential for malignant transformation through careful histological examination.

KEY WORDS

Verruciform Xanthoma, foam cells, Oral submucous fibrosis

How to cite this article: Ghosh S, Gupta S, Singh K, Urs A . Verruciform Xanthoma Associated With Oral Submucous Fibrosis - A Report of An Unusual Case With Literature Review. Int J Cont Med Res. 2014;1(2):85-88

[1]Associate Professor, [2]Professor & Head, [3]Senior Resident, Department of Oral Medicine & Radiology, [4]Professor & Head, Department of Oral Pathology, Maulana Azad Institute of Dental Sciences, New Delhi, India

Corresponding author: Dr Khushboo Singh, Senior Resident, Department of Oral Medicine & Radiology, Maulana Azad Institute of Dental Sciences, New Delhi, India

Conflict of Interest: None

INTRODUCTION

Verruciform Xanthoma (VX) is an uncommon benign inflammatory mucocutaneous lesion predominantly affecting oral cavity followed by genital mucosa. It was first described by Shafer in 1971 where he gave description of 15 cases of this condition in oral cavity and coined the term "Verruciform Xanthoma" .[1]The first report of involvement of non-oral site was described by Santa Cruz in 1979 where he reported two cases of vulva .[2] Its prevalence varies from 0.025 to 0.094% .[3] VX affects mostly adults in the 4th and 5th decades of life without sex predilection .[4] The lesions are usually asymptomatic and solitary but may be multifocal also. Clinical presentation of VX is as a papule or a plaque with color varying from reddish pink to gray and overlying mucosal surface showing verrucous or papillomatous growth. It may be misdiagnosed as various benign, premalignant, and malignant conditions. Most of the lesions of VX occur in isolated form but in many cases, it may develop in association with the other diseases like oral pemphigus vulgaris, carcinoma in situ, lichen sclerosus, solar keratoses, discoid lupus erythematosus, lichen planus, and CHILD syndrome (congenital hemidysplasia, icthyosiformerythroderma and limb defects) .[5] Final diagnosis of VX is made on the basis of histopathological examination. Histologically it is characterized by the presence of parakeratinized epithelium showing papillary or verrucous growth with thin rete ridges and connective tissue papillae extending up to the surface. The papillae characteristically consist of foam cells also called xanthoma cells. Here we describe a case report of verruciform xanthoma occurring on the right buccal mucosa in a 25–

years old male patient along with its clinical features, pathogenesis, histological features and treatment modalities discussed.

CASE REPORT

A 25 year old male patient reported to the OPD with the chief complaint of mobility of lower front teeth for the past 4 months. Medical history was non-contributory. Personal history revealed history of tobacco chewing, 6 packets per day for the past 6 years.On intraoral examination, blanching was present on the right & left buccal mucosa, hard palateandfibrous bands were palpable on right & left buccal mucosa. Also, there was a yellowish-white, nonscrapable patch present on the right buccal mucosa extending from comissure area to the retromolar region and overlying mucosal surface showing fine white papillary projections giving granular appearance (Figure 1). On palpation, it was soft to firm in consistency with well-defined margins. Hard tissue examination revealed grade 1 mobility of 31, 41 with generalized presence of calculus and extrinsic stains.

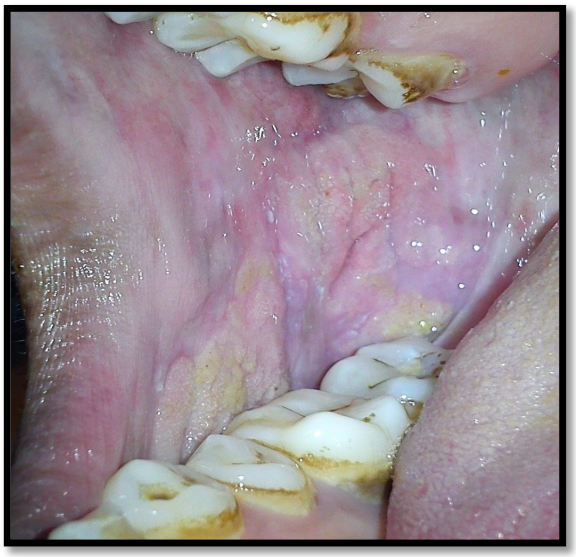

Figure 1: Clinical photograph showing whitish-yellow papillary lesion on right buccal mucosa

Based on the history and clinical findings, a provisional diagnosis of Oral submucous fibrosis with papilloma was made. Other lesions including squamous papilloma, verrucous carcinoma were considered as differential diagnosis. All blood investigation including complete blood count were within normal limits.

Incisional biopsy was done. Histopathological section showed hyperplastic parakeratinized stratified squamous epithelium exhibiting papillary projections alternating with elongated broad reteridges coalescing with each other. The connective tissue entrapped within the coalesced reteridges showed lipid laden macrophages appearing empty associated with few dilated vessels (Figure 2 & 3). The macrophages showed PAS positivity in the cytoplasm. The underlying connective tissue is densely collagenous alternating with dilated vessels & associated with chronic inflammatory cell.

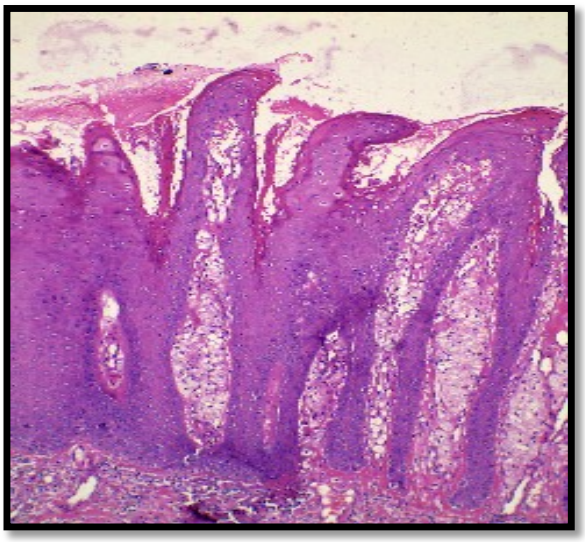

Figure 2: Microphotograph (10X power) showing epithelium exhibiting papillary projections alternating with reteridges having entrapped connective tissue

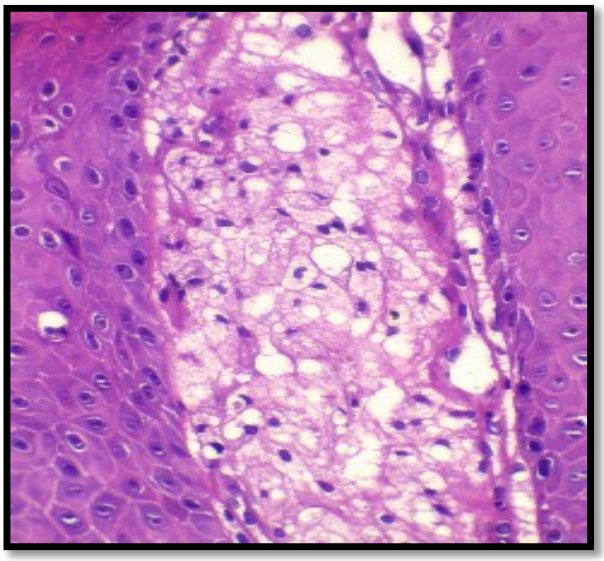

Figure 3: Microphotograph (40X power) showing lipid laden macrophages within the entrapped connective tissue

The patient was referred to the department of oral and maxillofacial surgery where excision of the lesion was done under local anaesthesia. The patient is under regular follow up and no recurrence is noted till date.

DISCUSSION

VX is a rare benign lesion, primarily of the oral mucosa that can affect skin and genital mucosa also. Approximately, 75% of all oral VX lesions occur on the masticatory mucosa of the palate, gingiva or the alveolar ridge whereas few cases have also been reported on the floor of mouth, buccal mucosa and tongue .[6,7] But present case occurred on buccal mucosa along with oral submucous fibrosis and literature search on pubmed showed that till date only one such case has been reported so far .[8]Clinically oral VX may bear a resemblance to verrucous carcinoma, squamous papilloma, verruca vulgaris, squamous cell carcinoma, or a mucosal fibroma.Clinically, majority of the squamous papillomas are found to be round in shape, whitish, and pedunculated .[9] The lesions of verruca vulgaris are circumscribed, firm, elevated papule with papillomatous, hyperkeratotic surface.[10] Verrucous carcinoma lesions are almost always large, exophytic, soft, fungating, slow growing neoplasms with a pebbly surface.[11] Whereas squamous cell carcinoma presents as an ulcer with fissuring or raised exophytic margins. It may also present as a lump, as a red lesion (erythroplakia), as a white or mixed white and red lesion, as a non-healing extraction socket or as a cervical lymph node enlargement, characterized by hardness or fixation .[12]

Ide et al. suggested some possible oral etiologic agents such as wet microenvironment, periodontal pathogens, mechanical stimuli, tobacco, alcohol, drugs, and sensitizing or allergic substances of foodstuffs and dental materials. It may be associated with localized inflammation, Immunologic factors and viral aetiologies have been also suggested .[7]

Histologically, VX shows three patterns: verrucous (most common), flat, and papillary (least common) .[13] It is characterized by the presence of parakeratinized epithelium showing papillary or verrucous growth with thin rete ridges and connective tissue papillae extending up to the surface. The papillae characteristically consists of foam cells also called xanthoma cells (xanthos = yellow).These xanthoma cells contain lipid as well as periodic acid Schiff (PAS) positive, diastase-resistant granules. There is a controversy over the exact origin of xanthoma cells but they are said to be a descent of monocytes/macrophages. However, there is no evidence of dysplasia. Macrophages are responsible for the initiation of process. As the epithelium degenerates due to local trauma, there is accumulation of epithelial breakdown products which induce inflammatory response and subsequent release of lipid material through the epithelium that finally is scavenged by the macrophages. They also suggested a local irritant as the initiator of the process .[1]Nowparast et al suggested that foam cells may be responsible for verrucous and papillary architecture which affects the nutrition and metabolism of the epithelial cells, leading to the hyperkeratotic change.[13]

In most of the cases VX tends to occur as an isolated lesion but in many cases the lesion developed in association with the other diseases like oral pemphigus vulgaris, carcinoma in situ, lichen sclerosus, solar keratoses, discoid lupus erythematosus, lichen planus and congenital hemidysplasia with ichthyosiformerythroderma and limb defects (CHILD syndrome)[5] , though in the present case it is found in concomitant association with oral submucous fibrosis which is an very unusual and rare finding. Excision is usually the treatment of choice and VX usually doesn't recur .[1] In the present case too there was no recurrence of the lesion reported after excision. However, there are no conclusive evidences exists in the literature regarding the malignant transformation of VX.

CONCLUSION

VX is a localized chronic reactive lesion that must be differentiated from various hyperplastic lesions whose correct diagnosis requires careful histological examination and surgical excision. Although histopathologically it still poses a challenge to the understanding of its pathogensis. Also emphasis should be done to investigate if any associations with other epithelial and skin

lesions especially, those having potential for carcinogenesis.

REFERENCES

1. Shafer WG. Verruciform xanthoma. Oral Surg Oral Med Oral Pathol. 1971; 31:784-9.
2. Santa Cruz DJ, Martin SA. Verruciform xantohoma of the vulva: Report of two cases. Am J ClinPathol. 1979;71:224-8.
3. Farahani SS, Treister NS, Khan Z, Woo SB. Oral verruciform xanthoma associated with chronic graft-versus host disease: a report of five cases and a review of the literature.Head and Neck Pathology. 2011; 5:193–8.
4. Philipsen HP, Reichart PA, Takata T,Ogawa I. . Verruciform xanthoma-biological profile of 282 oral lesions based on a literature survey with nine new cases from Japan. Oral Oncol. 2003; 39:325–36.
5. Kunal S, Alka DK, Seema H. Verruciform xanthoma: Report of two cases and review on pathogenesis. JOMFP.2008;12:41-4.
6. Bhalerao S, Bhat P, Chhabra R, Tamgadge A. Verruciform xanthoma of buccal mucosa: A case report with review of literature. ContempClin Dent. 2012; 3:S257–59.
7. Ide F, Obara K, Yamada H, Mishima K, Saito I, Kusama K. Cellular basis of verruciform xanthoma: Immunohistochemical and ultrastructural characterization. Oral Dis. 2008;14:150–7.
8. Yu CH, Tsai TC, Wang JT, Liu BY, Wang YP, Sun A, et al. Oral verruciform xanthoma: a clinicopathologic study of 15 cases. J Formos Med Assoc. 2007;106:141-7.
9. Carneiro TE, Marinho SA, Verli FD, Mesquita AT, Lima NL, Miranda JL. Oral squamous papilloma: clinical, histologic and immunohistochemical analyses. Journal of Oral Science. 2009; 51:367-72.
10. Vela DD, Sharma R, Durgesh NB. Extensive mucocutaneous verruca vulgaris in a non–immunocompromised patient. Int J Clinic Paediatric Dent. 2011; 4:65-8.
11. Walvekar RR, Chaukar DA, Deshpande MS, Pai PS, Chaturvedi P, Kakade A, et al. Verrucous carcinoma of the oral cavity: A clinical and pathological study of 101 cases. Oral Oncology. 2009; 45: 47– 51.
12. Markopoulos AK. Current Aspects on Oral Squamous Cell Carcinoma. Open Dent J. 2012; 6:126-30.
13. Nowparast B, Howell FV, Rick GM. Verruciform xanthoma: A clinicopathologic reviewand report of fifty four cases, Oral Surg Oral Med Oral Pathol. 1980; 51:619-25.