

Oral Verrucous Hyperplasia with Dysplasia – A Case Report.

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ABSTRACT

Introduction: Verrucous Papillary Lesions (VPLs) clinically present themselves as an exophytic growth seen commonly as grey-white in color. A dominant part of the favorable VPLs have viral etiology and incorporate commonly occurring squamous papilloma alongside verruca vulgaris, central epithelial hyperplasia, and condyloma. Current comprehension of possibly malignant VPLs is astounding and is basically ascribed to the utilization of confounding and unacceptable terminology. Verrucous Hyperplasia (VH) of the oral mucosa is a relatively intricate entity possessing paradoxical features making it difficult to diagnose when compared to other verruco-papillary lesions such as Verrucous Carcinoma (VC).

Case Presentation: This case report depicts the presentation of Oral verrucous hyperplasia along with dysplasia seen in buccal mucosa of a 46-year-old patient.

Management and Prognosis: Surgical excision was performed and ensured that margins were included in the excision to avoid any recurrence of the lesion.

Conclusion: The most dependable approach to isolate these substances on routine hematoxylin-eosin stained tissue areas is to perceive the exophytic growth patterns of oral verrucous hyperplasia from the combined exophytic and endophytic growth patterns related with verrucous carcinoma. Furthermore, investigations on this are required using Immunohistochemical methods.

Keywords: Dysplasia, Oral verrucous Hyperplasia, Verrucous Carcinoma, Verrucous Papillary lesions.

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INTRODUCTION

Verrucous Papillary Lesion (VPL) of the oral cavity are a heterogeneous group of lesions with OVH advocating itself to be a challenging entity to choose from a spectrum of benign, potentially malignant and frankly malignant lesions. OVH (Oral Verrucous Hyperplasia) is an elementary step towards OVC (Oral Verrucous Carcinoma), which then transforms to SCC (Squamous Cell Carcinoma) in a later stage. These elements can be recognized by the absence of intrusive development in Verrucous Hyperplasia (VH) cases; as such, it is essential to include a margin with adequate depth when performing a biopsy of the epithelium of the lesion. OVH was first described by Ackermann and is named after him as Ackermann's tumor.¹ Albeit the close relationship between OVH and OVC, the distinction between the two lesions remains enigmatic when it comes to distinguish them clinically. The clinical appearance of these lesions has not by and large been well described as much accentuation has been laid on the verrucous/exophytic nature of these lesions with little consideration being given to color variation. In addition, these lesions are viewed as clinically indistinct from verrucous carcinomas which are commonly white or grayish white in color. Buccal mucosa (57.89%) is found to be most commonly involved followed by tongue, gin-

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giva, alveolar mucosa, soft palate in decreasing order.² Therefore, a meticulous approach by means of standardised clinical and histological criteria was executed to differentiate OVH from other verrucous and papillary lesions of oral mucosa.

CASE REPORT

A 46-year-old male patient presented to the Outpatient Department of D.Y.Patil Dental College, Navi Mumbai, India in 2019 with a growth on the left buccal mucosa (Fig. 1). Six months prior to the presentation, the patient noticed a painless ulcer in the same location. There was no report of local trauma and the patient presented no parafunctional habits. The patient's medical, family and social history was non-contributory.

On clinical examination, a whitish pink sessile oral mass of approximately 2.5 x 2 x 1 cm was observed in the left region with a warty/pebbly superficial surface and well-defined margin. It was firm in consistency and non-tender on palpation. No evidence of discharge was observed on the surface or surrounding mucosa. An extraoral examination revealed no significant facial asymmetry and no palpable lymph nodes. Clinical diagnosis of Verrucous Hyperplasia was made and an incisional biopsy was carried out under local anaesthesia. Haematoxylin and eosin stained sections showed an exophytic parakeratinized stratified squamous epithelium proliferating in the form of papillary projections showing keratin plugging. The underlying connective tissue stroma showed moderate chronic inflammatory cell infiltrate, adipose tissue and blood vessels (Fig. 2). Surgical excision was done and excisional biopsy revealed few areas with dysplastic features. As a result of these features, the diagnosis was made of Verrucous Hyperplasia with Mild to Moderate Focal Dysplasia. A 6-month follow-up revealed no recurrence of the lesion (Fig. 3).

DISCUSSION

Murrah and Batsakis carefully hold the term OVH to be utilised histopathologically and considered it to be a forerunner of OVC.³ The term OVH could be applied clinically and histopathologically only to mass type and that plaque type is clinically named Oral Verruciform Leukoplakia.⁴ The scholars in the Asian Provincial Meeting propounded the expression "EVH" (Exophytic Verrucous Hyperplasia) to signify the microscopic diagnosis of OVH on clinical basis and formulated histological rules for EVH.⁵

According to the observations made, it is evident that verrucous carcinoma may develop from verrucous hyperplasia. In the present case report, OVH was seen as a fungating exophytic mass

with verrucous appearance which was whitish pink in color. Slootweg et al. reported that the presence of keratin plugging in the centre of epithelial invaginations is a histological hallmark of VC (Verrucous Carcinoma) as suggested by Shafer.^{6,7} Common age of presentation is during the 4th decade of life rather than the 6th decade as quoted by Shear and Pindborg J J (1980). In a clinicopathological study conducted by Vinay et al in 2011, it was noted that males predominated females in a ratio of 2:1, contradicting many other studies which said otherwise. Buccal mucosa (57.89%) was found to be most commonly involved followed by tongue, gingiva, alveolar mucosa, soft palate in decreasing order.² Current case report also showed epithelium with papillary projections exhibiting keratin plugging.

The distinction between VC and VH clinically is strenuous and unfeasible. The main differences observed in the histopathological sections were the level of verrucous projections with relation to adjacent epithelium. In VH most of the hyperplastic broadened rete ridges lay above the adjacent normal epithelium while VC, on the contrary, exhibits a downward growth pattern of otherwise similar rete ridges.² As proposed by Shear and Pindborg, considerable acanthosis with broadened rete ridges causes deprivation of distant epithelial cells from blood supply and becomes oedematous and swollen. These necrotic cells undergo desquamation leaving cleft in the surface of epithelium. Verrucous projections are formed in this way in between clefts. In a later stage, both verrucous projections and clefts between them undergo keratinisation. Dysplasia is commonly seen in VH which is consistent in the findings of Shear and Pindborg (1980). For grading dysplasia, Pindborg photographic standard of epithelial dysplasia was followed.⁷ Prevailing case report revealed augmented rete ridges in the same line with the normal epithelium without much downward growth into the lamina propria. Dysplasia is commonly seen in VH which is consistent in the findings of Shear and Pindborg. Out of 19 cases, 68.42% showed dysplastic epithelium and among which moderate dysplasia (61%) was predominant than mild dysplasia (38%).⁷ Microscopic features as suggested in the contemporaneous case report also suggested few areas with dysplastic features.

Researchers involved in the study conducted in 2011, showed adjacent atrophic epithelium and juxta-epithelium hyalinisation showing features of OSMF. In a single-centred study conducted in 2019 by Desai et al reported the occurrence of EVH in the background of OSF. The term OVH should be restricted to lesions showing microscopic features of VH without dysplasia occurring in

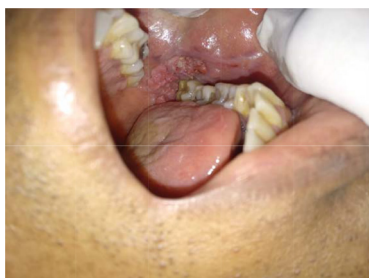
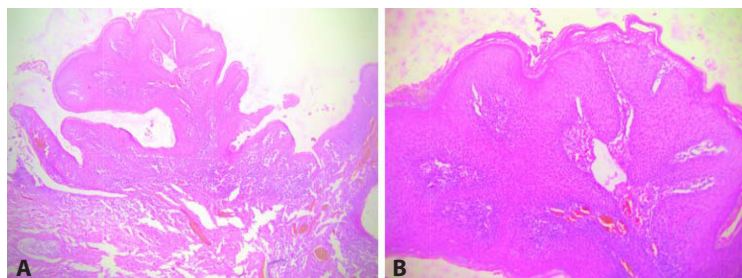


Fig. 1: whitish-pink sessile exophytic lesion on the left buccal mucosa of a 46-year-old patient.



Figs. 2A and 2B: Exophytic parakeratinized stratified squamous epithelium proliferating in the form of papillary projections showing keratin plugging. (H and E, 10x and 40x)

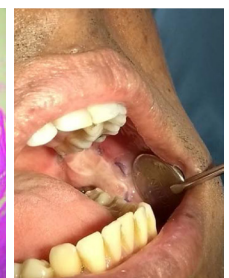


Fig. 3: The site after 6 months of follow up.

non-exophytic lesions. Similarly, the terms OVD (Oral Verrucous Dysplasia) and EVD (Exophytic Verrucous Dysplasia) should be reserved for lesions showing microscopic verrucous hyperplastic features with dysplasia in non-exophytic and exophytic lesions respectively. Thus proposed EVH and EVD are clinicopathological terms with prognostic value against EVH.⁸

The association of Human papilloma virus (HPV) was found with development of verrucous hyperplasia by Greer R D (1990).⁹ C-erb B-3 protein expression was an index of malignancy during progression from Verrucous Hyperplasia to Verrucous Carcinoma and Squamous cell carcinoma.¹⁰ Expression of P53 and EGFR levels could be used as marker while differentiating verrucous hyperplasia from verrucous carcinoma and squamous cell carcinoma.¹¹

Therefore, for evaluation of OVC and OVH an adequate biopsy sample is of utmost importance which includes a lesional margin with adequate depth. Many researches have concluded that it's always better to take an average of 3-4 biopsies before a correct diagnosis of OVH/OVC.¹² In terms of treatment, traditional method is total surgical excision, which always leads to scar formation for a large OVH lesion.¹³ The treatment modality executed in the present case was wide surgical excision of the lesion with adequate soft tissue margins to avoid recurrence. Photodynamic Therapy (PDT) is another mode of treatment option because it is well tolerated by patients and can be used repeatedly without cumulative side effects and also contributes to lesser scar formation. In a case report published by Yu-Chang and Chuan-Hang Yu, the efficacy of topical ALA-PDT protocol was tested using a laser light source combined with Cryogun Cryotherapy for an extensive OVH lesion which showed no recurrence of lesion after follow-up period of 18 months.¹⁴

CONCLUSION

OVH is a premalignant lesion which occurs rarely and requires an early intervention to increase the chances of favourable outcome. Apart from the absence of an endophytic growth pattern in OVH, the presence of dysplasia in OVH was noted in our case report. Thus, biopsies of verrucous lesions should include the adjacent normal epithelium in order to ensure correct diagnosis. Follow-up after the treatment acts as a desideratum to assess recurrence potential and clinical behaviour.

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