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CASE REPORT

Oral Proliferative Verrucous Leukoplakia: A Case Report

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Abstract

Oral Proliferative Verrucous Leukoplakia (OPVL) is a rarely occurring malignant lesion of oral mucosa. At times it is difficult to diagnose and differentiate from verrucous carcinoma and leukoplakic patches but it is clinically more aggressive than these lesions. This case report deals with a case of OPVL with its detailed clinical and histopathological features along with the surgical procedure for treatment. (2020, Vol. 04; Issue 01: Page 56 - 60)

Keywords: Oral proliferative verrucous leukoplakia, Verrucous, White patch.

Introduction

In 2007, Warnakulasuriya et al. defined "leukoplakia" as "a white plaque with an increasing questionable oral cancer risk after excluding other known diseases and disorders that do not increase the risk" (1). In 1985, Oral Proliferative Verrucous Leu- koplakia (OPVL) was first described by Hansen et al. as a "rare potentially malig- nant lesion of the oral mucosa" (2). Ac- cording to the latest WHO nomenclature, OPVL has been assigned a new terminol- ogy of "potentially malignant lesions" as it is neither a delimited lesion nor condition

(3). Based on certain characteristics such as clinical criteria, geographic location, in- traoral sites and cultural habits, the prev- alence of OPVL ranges from 3-5% and the malignant transformation rate has a range of 0.13% to 17.5% (2).

OPVL lesions grow progressively as well as persistently and present specific charac- teristics, mainly a more aggressive biolog- ical behavior when compared to other forms of leukoplakia, which is expressed by high tendency to recur and chance of malignant transformation (2, 4). Initially, OPVL appears as a whitish patch of hyper- keratosis which eventually converts into a multifocal disease with exophytic and pro- liferative features, indistinguishable from verrucous carcinoma (4-7). We hereby pre- sent a case of Proliferative verrucous leu- koplakia along with elaborate specific di- agnostic criteria, differential diagnosis and treatment.

Case report

A patient reported to Department of Oral Medicine and Radiology, aged 33years with the chief complaint of a white growth in right cheek region since 1 year. Growth was initially small and gradually increased to attain the present size. Growth is asso- ciated with pain and bleeding during mas- tication. He gave history of a similar lesion 10 years back that had been successfully operated. The patient has a habit of pan and gutkha chewing, 7-8 times per day and cigarette smoking, 10 cigarettes per day since 15 years.

On extraoral examination, right subman- dibular lymph node was palpable measur- ing approximately 1 cm in diameter and it was soft and movable (Fig 1A).

Fig 1: A- Extraoral view, B- Intraoral view

Intraoral examination revealed a well de- fined hyperkeratotic exophytic growth on the right buccal mucosa, measuring ap- proximately 6 cm x 8 cm in diameter, ex- tending antero-posteriorly from right la- bial commissural area to 1 cm behind the retromolar pad region and supero-inferi- orly from the deepest portion of the upper buccal vestibule to the deepest portion of the lower buccal vestibule. The overlying surface showed small finger-like projec- tions. On palpation, all inspectory findings were confirmed, the surface was rough in texture, soft and leathery in consistency, tender and non-scrapable (Fig 1B).

Based on the clinical appearance, a provi- sional diagnosis of proliferative verrucous leukoplakia along with a differential diag- nosis of verrucous carcinoma and papil- loma were given.

Blood investigations were done and or- thopantomogram was taken to rule out any bony involvement. Then incisional bi- opsy was done. Histopathological exami- nation revealed orthokeratinized stratified squamous epithelium with moderate acanthosis, intact basement membrane and few areas of parakeratin plugging. The lamina propria showed moderate infiltra- tion of chronic inflammatory cells in colla- genous stroma suggestive of verrucous ep- ithelial hyperplasia with hyperor- thokeratosis (Fig 2). Thus, a final diagno- sis of proliferative verrucous leukoplakia was given.

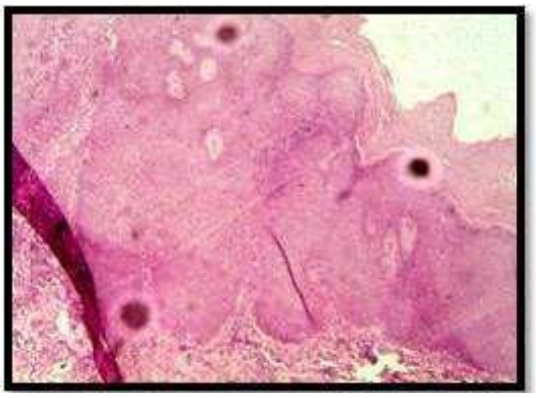

Fig 2: Histopathological view (10X)

Subsequent to histopathological diagno- sis, the entire lesion along with 2cm of normal mucosa surrounding it was surgi- cally excised (Fig 3 and Fig 4). The defect was covered with an allograft taken from the thigh region (Fig 5). The area was su- tured and the tissue was sent for further histopathological re-evaluation, which confirmed the previous histopathological diagnosis. Further, follow-up was done to evaluate post-operative healing (Fig 6).

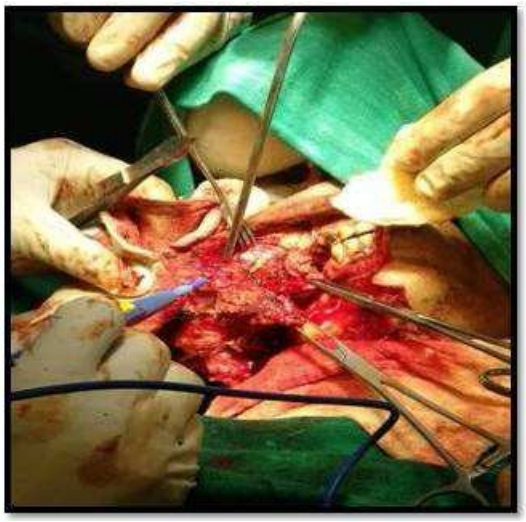

Fig 3: Surgical excision of the lesion

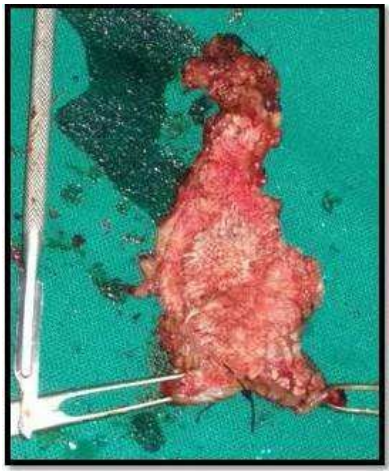

Fig 4: Surgically removed lesion

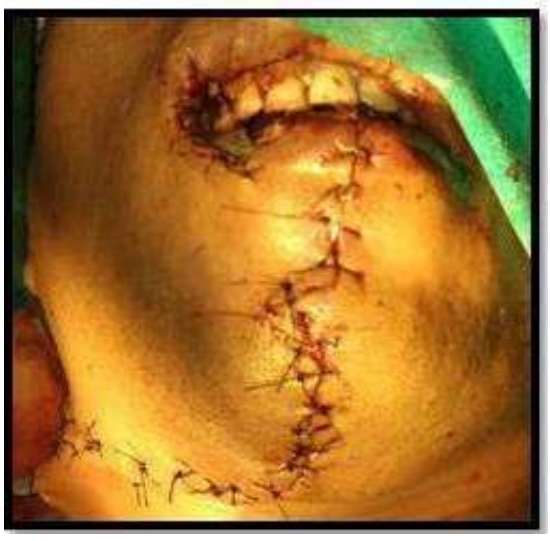

Fig 5: Suturing done after giving Allograft

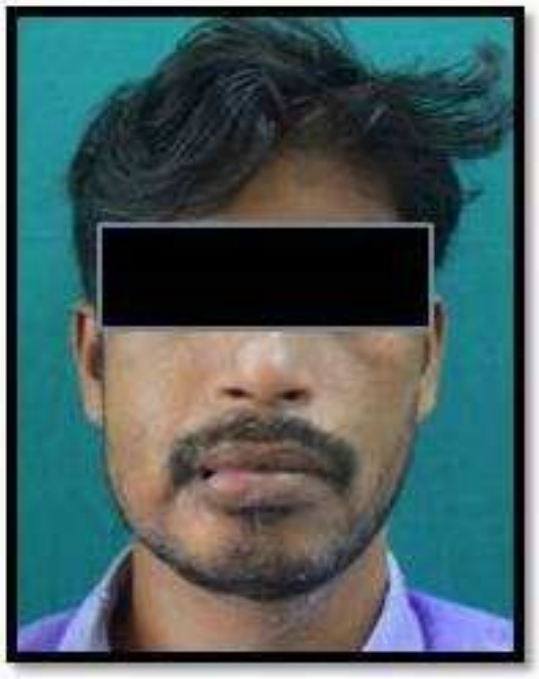

Fig 6: Post operative profile after 2 months

Discussion

The term “Proliferative verrucous leu- koplakia” has a meaning of its own. Prolif- erative refers to persistent, diffuse, pro- gressive and multifocal. Verrucous means warty, verrucal, exophytic keratotic lesion. Leukoplakia, as we have discussed earlier is a white keratotic patch (7).

Be it any mucosal site, verrucous hyper- plasia is mostly diagnosed in patients at 6th to 8th decades (8). It has a strong pre- dilection for women, having a 1:4 male to female ratio but no racial preference (4). Buccal mucosa and tongue are the most common sites followed by alveolar mu- cosa, gingiva, palate and lips (9). The true and false vocal cords are laryngeal areas of predilection (8). Some studies have shown that almost all lesions occur bilat- erally, mainly affecting the lower alveolar ridge and the buccal mucosa (10).

The lesions are slow-growing yet persis- tent which are resistant to almost all

forms of treatment. While the lesion devel- ops, certain erythematous and/or verru- cous areas are commonly found which oc- casionally progress to verrucous carci- noma or squamous cell carcinoma. In the early stages, the lesions clinically appear as homogenous leukoplakia with no un- pleasant findings on biopsy. Over an ex- tended period of time, the lesions become diffuse, show multifocal warty projections, and at times develop an erythematous component. Eventually, these lesions demonstrate regional and distant metas- tasis (9).

A few histological stages have been sug- gested by Hansen et al. (1985) in the con- tinuum of PVL with intermediate, so that the disease may have any of these appear- ances during its course. They are as fol- lows: Grade 0: Normal mucosa; Grade 2: Hyperkeratosis (clinical leukoplakia); Grade 4: Verrucous hyperplasia; Grade 6: Verrucous carcinoma; Grade 8: Papillary squamous cell carcinoma; Grade 10: Less well-differentiated squamous cell carci- noma

Batsakis et al. (1999) reduced the number of histological stages to four with interme- diates:- Grade 0: Clinical flat leukoplakia without dysplasia; Grade 2: Verrucous hy- perplasia; Grade 4: Verrucous carcinoma; Grade 6: Conventional squamous cell car- cinoma with intermediates (9).

Based on the recent classification of histo- logical stages by Batsakis et al., our case belongs to the Grade 2 category of Verru- cous hyperplasia.

PVL, due to its progressive and aggressive nature is said to be resistant to a variety of therapies, thus making the manage- ment difficult and poor prognosis. Treat- ment modalities including carbon dioxide laser, radiation, topical bleomycin solu- tion, oral retinoids, beta-carotene and sys- temic chemotherapy have all failed in achieving permanent cure (9). Methi- sopriol has shown some efficacy in HPV- induced lesions as it inhibits the viral ri- bonucleic acid synthesis and replication and stimulates the antiviral cell-mediated reactions. Laser ablation and topical pho- todynamic therapy may also prove to be useful in few cases. The most successful treatment modality is total excision of the lesion with free surgical margins and life- long follow-up (3, 9, 10).

Conclusion

Though OPVL is a rare and highly aggres- sive form among all types of leukoplakia, it's diagnostic criteria help us to delineate the possible beneficial outcome of disease progression as well as implement thera- peutic approach to get rid of malignant transformation.

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