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Linear Oral Lichen Planus: A rare presentation

Mucosal irritation caused by cusps of upper and lower teeth represents a source of persistent, low grade mechanical trauma. This type of trauma can initiate and/or perpetuate oral lichen planus in a few susceptible patients. Here, we report a rare case of bilateral white linear striations in the buccal mucosa of a young patient, associated with sharp cusps. Oral lesions demonstrated a unique cuspal signature highlighting the causal role of subtle mucosal trauma in the genesis of reticular-oral lichen planus. Lesions appeared to be exacerbated by significant psychological distress experienced by the patient. Although the Koebner phenomenon is well reported in the cutaneous lichen planus literature, such presentations are relatively rare in the oral cavity. This report emphasizes the potential role of low-grade cuspal trauma/irritation in conjunction with psychological distress in the genesis of oral lichen planus.

Keywords: Oral lichen planus, Koebner phenomenon, mechanical trauma, dental cusps, psychological distress

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1. Introduction

Oral lichen planus (OLP) is an immune-mediated chronic oral mucosal disease that can present as reticular (bilateral white lesions), erosive, plaque-like, bullous and other clinical subtypes that frequently involve the buccal, lingual and gingival mucosa [1]. The clinical presentation of OLP generally includes a network of white lines *(Wickham's striae)* in a diffuse pattern engaging the entire buccal mucosa, and large portions of lateral tongue, and gingival mucosa. The etiology of OLP has been attributed to local factors such as dental plaque and calculus, and, systemic factors including medications, hepatitis C infection, autoimmune diseases, psychological stress and anxiety [1,2]. Among the predominant local factors that contribute to OLP, *contact sensitization* to dental plaque and calculus is widely accepted [3]. However, the role of *low-grade mucosal trauma* caused by dental cusps has not been extensively reported and is not fully understood. While the involvement of gingiva explains the role of dental plaque and calculus in the disease process, the occurrence of OLP in the buccal and lingual mucosa points to the contribution of dental cuspal irritation. The impact of trauma as a trigger for cutaneous lichen planus (CLP) referred to as the 'Koebner phenomenon' is well documented in the dermatology literature [3]. However, the potential of mechanical trauma elicited by dental cusps in OLP needs further validation. Here, we report a unique presentation of bilateral *reticular oral lichen planus* in the buccal mucosa of a young adult associated with cuspal irritation and psychological stress.

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2. Case report

An anxious dental intern visited our oral medicine clinic in December 2017 with the chief complaint of burning sensation especially upon intake of spicy foods. As a result, he had developed a habit of cooling down his food prior to intake. Examination revealed bilateral, linear white striae on the buccal mucosa extending up to the retro-molar area, closely associated with cuspal indentations (Figure 1). He was a never-user of tobacco and areca nut, was not on any systemic medication, had not changed tooth paste or developed new food habits in the recent past. Previous medical history was insignificant, and he did not undergo any fillings or other dental restorations. However, clinical evaluation revealed an apprehensive personality with considerable anxiety. Questions concerning psychological triggers and life-related problems revealed substantial stress and anxiety as well as sleep disorder (<5hrs/day) over the previous four year period. The clinical picture of bilateral, reticulated white lesion was pathognomonic for *oral lichen planus*. Differential diagnosis for similar unilateral lesions includes *'frictional keratosis'* if lesion is focal, and larger unilateral presentations such as this are suggestive of lichenoid reaction, closely linked to amalgam fillings, tobacco/areca nut habit, and systemic drugs. He was treated with triamcinolone acetonide (0.1% topical cream), but reported of having used it only for 1 week (2 times/day). Following this, a transient reduction in burning sensation was noticed, without any regression in clinical lesions. Seven months after initial presentation (July, 2018) his dental education was complete, financial tension settled, self-esteem improved after finding a job, and sleep quality and total sleep time also improved remarkably (>8 hrs/day). On 3rd examination (Sep,

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2018), lesions regressed almost completely, and on the 4th examination (March, 2019) complete regression of white lesions and clinical symptoms was observed. Our patient is under meticulous follow up, and has not displayed any symptoms or new oral lesions during the last few months.

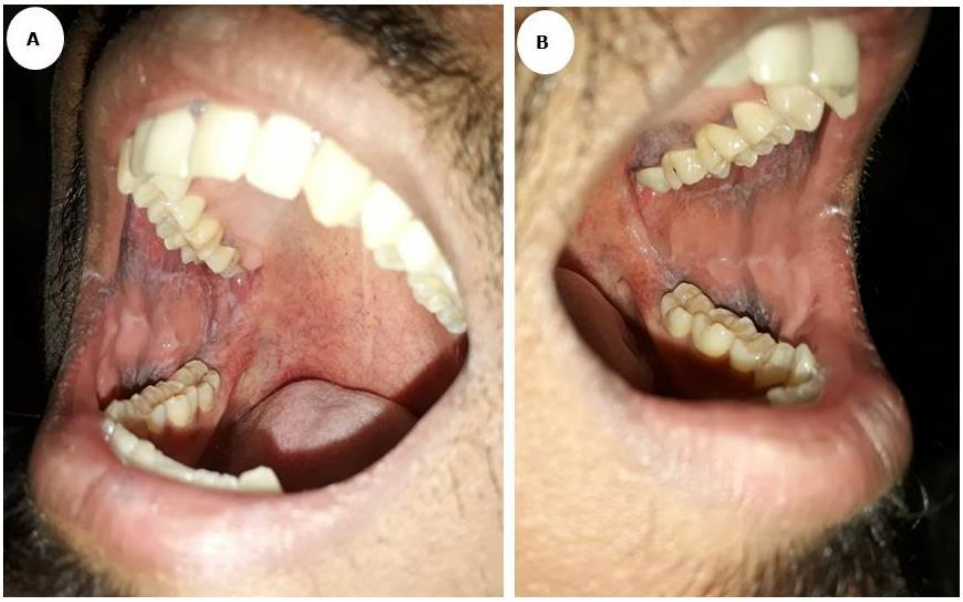

Figure 1: Bilateral reticular oral lichen planus in close proximity with dental cusps associated with hyperpigmentation. The lesions follow the cuspal pattern, and spared all other oral sub-sites implying the direct involvement of chronic low-grade mechanical trauma caused by dental cusps. Careful examination showed relatively sharper cusps in all teeth, indicating a persistent, low grade mechanical trauma.

3. Discussion

Trauma caused by upper and lower dental cusps is an important trigger in the inception and/or precipitation of OLP. For this reason, buccal and lingual mucosa are frequently involved in OLP [1]. In the present case, the bilateral buccal mucosal involvement implicates cuspal trauma. Common oral conditions like OLP and pemphigus first occur on the buccal mucosa, and can thus serve as models to understand the role of mechanical irritation [3]. Recent studies highlight the role of *neurogenic mechanisms* underlying OLP. In these

studies, higher expression of *sensitive ion channels* such as Transient Receptor Potential Ankyrin-1 (TRPA1), and transient receptor potential vanilloid type-1 (TRPV1) have been reported in OLP [4, 5]. These ion channels are known to respond to *'mechanical' and 'chemical' stimuli*, and may contribute to exacerbation of OLP through the Koebner phenomenon [4]. In this regard, documentation of the site of initial onset of OLP lesions and pattern of progression may provide valuable insight into the dynamics of disease. Furthermore, mechanical trauma is well known to induce oral carcinogenesis in sub-sites like tongue and buccal mucosa [1, 6, 7]; whether malignant transformation in buccal and tongue OLP cases is related to mechanical trauma induced by dental cusps needs further investigation. Although dental cuspal irritation was obvious in the present case, treatment to manipulate the healthy morphology of dental cusps could not be justified and was therefore not initiated.

In the present case, a rapid decline in disease symptoms was observed following recovery from psychological and emotional distress. Although common in OLP patients, clinicians often do not delve into the emotional and psychological histories of patients. This scenario of cortisol being an important biomarker in OLP is suggestive of psycho-somatization [8]. Although small studies disagree on the neuroendocrine etiology, elevation in cortisol was identified in nearly 55% studies in a recent systematic review [9]. *Poor 'quality-of-sleep'* is another common observation in OLP patients that was also noticed in the current patient. Sleep disturbances can be due to psychological factors like stress, anxiety and depression that could result from financial and other life-related issues [10]. It is certainly important to screen for such root level triggers, and a detailed history can actually provide this piece-of-

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information, as documented in the present report. Given the limited literature, additional investigation into the role of sleep disturbances in OLP is warranted.

Consistent with the present report, severity-of-symptoms have been correlated with anxious and depressive symptoms in symptomatic-reticular OLP [11]. Accumulated clinical evidence strongly links *symptomatic-reticular OLP to psychological morbidity*. Interestingly, in a recent study, both OLP and oral lichenoid lesions showed a relationship between disease severity and anxiety levels [12]. Our report revealed subtle dynamics between psychological distress and sleep disturbances with disease- severity over a long review period (~18 months). It is clear that *psychological distress precedes OLP* and propels its genesis. In extensive erosive-OLP and in symptomatic-reticular-OLP cases however, disease associated psychological distress may further exacerbate the severity of the condition. Therefore, in both reticular and erosive-OLP cases, psychological issues should be considered as a potential beginning event, and therapies targeting the mind-body complex may be useful besides traditional pharmacological therapy (eg: topical corticosteroids) [13]. Clinicians must promote life-style modification, work-life balance, and periodic relaxation for all OLP patients, as these can truly arrest disease progression and may even induce regression.

4. Conclusion

The present report depicts a rare case of linear reticular-oral lichen planus. The presence of severe psychological distress along with OLP in a unique cuspal pattern points to the possible link between local traumatic events (Koebner phenomenon) and psychological triggers. From this standpoint, we assume that patients with psychological distress may possess a sensitized

oral mucosa, susceptible to low-grade trauma from dental cusps. This unique report explains the orchestration of stress and anxiety, sleep problems, and local events like trauma from dental cusps in the pathogenesis of OLP.

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