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## **CASE REPORT**

# **Holistic Management For Severe Oral Lichen Planus: A Case Report**

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## **ABSTRACT**

Oral lichen planus (OLP) is an immunologically mediated mucocutaneous disease associated with various factors including systemic diseases, with a higher prevalence in females than males. The majority of OLP cases are accompanied by symptoms of discomfort and/or pain. Treatment of OLP can be challenging due to its unknown aetiology and involvement of multiple contributing factors. **Objective:** Our aim is to report a long-term follow-up case of oral lichen planus with holistic management approach. This report will provide some options to clinicians who are having difficulties in managing OLP cases especially with the medications. **Case Report:** This case report discusses a female patient who suffered OLP with no definitive treatment prior to referral to us. A thorough history of her problem including psychosocial factors and dietary pattern were obtained. Clinically, the oral mucosa presented characteristics similar to erosive oral lichen planus. Patient reluctance towards pharmacotherapy due to her comorbidities was a challenge in managing her OLP. Hence, a non-pharmacotherapeutic solution was offered by considering psychology and dietary pattern. **Conclusion:** A severe oral lichen planus with ulcerative background could be managed holistically and not just through a pharmacotherapeutic approach.

**Key words:** dietary pattern, holistic therapies, oral lichen planus

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## **INTRODUCTION**

Oral lichen planus (OLP) is an idiopathic chronic mucocutaneous disease clinically characterized by a wide range of clinical manifestations including white reticular patches, plaque, papular, vesicles/bullous and erosive/ulcerative and atrophic lesions with intense symptomatology.<sup>1,2</sup> Worldwide, OLP has a prevalence of 1.01%, where the highest prevalence appears in Europe (1.43%) and the lowest is in India (0.49%), where tobacco consumption may indicate the detection of OLP.<sup>3</sup> The prevalence of OLP increases significantly and progressively among people above 40 years of age, specifically those in their 50s and 60s.<sup>3,4</sup> OLP is a risk factor for oral malignancy when it presents as an erosive lesion, mostly on the tongue and presence of aneuploidy histologically.<sup>5</sup> A thorough examination, proper diagnostic work-up, accurate treatment plan and routine follow-up visit are crucial.<sup>5-7</sup>

Some triggering or initiating factors are related to oral lichen planus even though definite aetiology remains

unknown. Some studies correlated dietary patterns with oral lichen planus including red meat, fast food, alcohol, spicy foods, and coffee.<sup>8-10</sup> Food allergies could be part of the factor and have been reported in some cases.<sup>11,12</sup> Avoidance of such types of foods could offer some preventive or relief on OLP.

On the other hand, psychological factors are vital for clinicians to consider in these patients. Depression, anxiety, and stress are well documented with oral lesions, especially oral lichen planus.<sup>13-15</sup> Studies revealed that most practitioners were using topical steroids after photodynamic therapy while only a minority of practitioners explored newer forms of therapies.<sup>16-18</sup> Topical steroids are still the pillar for an efficient therapy in the symptomatic management of OLP with equivalent effectiveness demonstrated by topical calcineurin inhibitors and retinoids. However, temporary relapse burning sensation as a side effect was found higher in calcineurin inhibitors usage.<sup>16</sup>

## OBJECTIVE

Our aim is to report a long term follow up case of severe oral lichen planus (erosive type) with holistic management. This holistic management could be considered whenever dealing with oral lichen planus cases or any oral disease at large.

## CASE REPORT

A 53-year-old Punjabi lady was referred to our clinic for further examination as well as management for severe pain and gum inflammation associated with white intraoral lesions that were initially detected 3 years before the patient's initial referral to our clinic. The patient was consulted by several dental practitioners but neither a definitive investigation nor a conclusive diagnosis was made. The patient was referred to our clinic after all forms of treatment were unsuccessful. The provisional diagnosis of chronic periodontitis from the previous hospital indicated a prescription of chlorhexidine mouthwash and full mouth scaling.

She presented with extreme pain and burning sensation in her mouth particularly at both buccal mucosa, tongue, and gingiva. The patient was barely able to open her mouth due to extreme pain. These problems affect her in many ways including self-esteem failure. Due to her problem, she had avoided any spicy and sour food. As a Punjabi, her dietary routines are bread and rice with some curry which she could not enjoy for many years. She was only able to tolerate soft or blended food since then. Her daily diet was so bland and not much of options.

Medically, she was diabetic for the past 10 years, had a history of hypercholesterolemia and previously suffered from unstable angina. She was taking several medications including metformin, insulin, atorvastatin, aspirin, beta-blocker and glyceryl-trinitrate (GTN). Her systemic diseases are stabilized with these medications and under regular follow up with the physician. The patient mentioned having hypersensitivity to augmentin and a family history of some diseases including connective tissue diseases. She was an accountant who was constantly under stress due to her work's demands. Despite being accustomed to a strict eating regime, she failed to control her coffee consumption. She drank 5 to 6 cups of coffee daily. Besides, she was a mother of two children; both of whom were adults.

During her initial visit to our clinic, her intraoral examination findings showed a wide distribution of white patches on her buccal mucosa bilaterally including regions of the upper and lower gingiva (Figures 1 and 2). These white patches were in the background of ulcerated/erythematous mucosa. We



**Figure 1.** Right buccal mucosa shows erythematous mucosa surrounded by whitish lesion



**Figure 2.** Left buccal mucosa shows more intense erythematous region with some ulcerated areas

also observed the frothy saliva as a sign of dry mouth condition, presumably a side effect of her medications. The patient was not wearing any dental prosthesis and had undergone regular scaling procedures with her dentist from the previous hospital. The patient takes up to 20 minutes to brush her teeth (in a gentle manner) daily with an automatic, soft bristle toothbrush.

She showed slight dryness of the skin without any other significant findings. Eyes were moist without any evidence of dryness. Skin and lips were dry, but there were no abnormalities in the temporomandibular joint (TMJ) and cervical lymph nodes were non-palpable. Clinically, the presentation of her mouth condition gave the impression of an oral lichenoid reaction based on some medications consumed by the patient. Psychologically, we found patient showed some signs of depression correlated with her ongoing chronic oral problem.

During the first consultation, we tried to establish a diagnosis for her to be able to commence treatment. Due to her history as well as intraoral and extraoral findings, we believed that she could have either oral lichen planus (erosive type) or an oral lichenoid reaction (associated with medications). A biopsy was planned but was put on hold due to her complaint of extreme pain. The aim of treatment at this point was to stabilize her oral conditions so she would be able to tolerate a better diet regime. Before that, we determined several risk factors that potentially contributed to the development of her condition including polypharmacy, her pre-existing diseases, heavy coffee consumption, dry mouth, depression, stress (work-related) and family history of various systemic diseases. We were also understood that she was strongly opposed on taking additional drugs due to her existing polypharmacy. This was our challenge to manage her as a chronic disease sufferer. Therefore, we have developed a treatment approach by considering all the risk factors, her concerns, and preferences. Holistic management was used to achieve certain goals with her permission. The first goal was mainly aimed at relieving her oral dryness problem as it is considered the main contributor to her other problems. The strategy used included the reduction of coffee consumption to one cup daily since caffeine had become the diuretic agent which makes her mouth dry even more; to consume more frequent plain water but in smaller portions; and consumption of moist food only. Her daily diet shall include a variety of vegetables (steamed or boiled are preferred) and fruit juice to be consumed with a straw to avoid direct contact with her fragile oral mucosa. This is because fruit juices are relatively acidic despite being nutritious.

The second goal was to reduce her stress which she mentioned during the consultation. We proposed that she could consider a break or a long holiday to have some time off for herself. We have convinced her about the importance of giving herself a break. We have also presented some case samples from other patients to enhance and deepen her understanding of the disease. The third goal was to reduce other contributing factors to her oral problems. Her oral hygiene was fairly controlled due to her fear of further worsening the pain in her mouth. We introduced her to an alternative to regular brushing; instead of her previous method, she was encouraged to switch to the sponge/gauze method. The sponge or gauze is soaked with a chlorhexidine mouthwash before being wiped on the teeth. As mentioned earlier, she was reluctant with new oral medication, thus we suggested for her to use a topical steroid instead which she could tolerate; she was prescribed betamethasone dispropionate 0.1% ointment to be applied at the area where she feels most discomfort or pain.

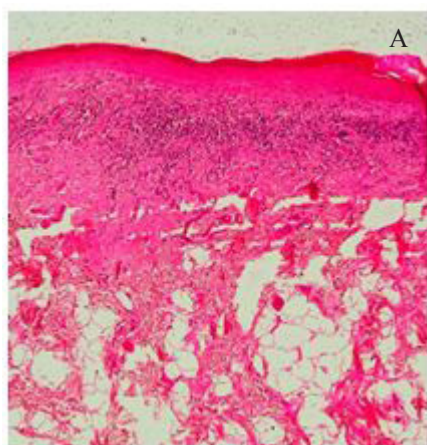
After 2 weeks, she came for her first review appointment, presented with a remarkable improved

condition. She only had minor pain and slight discomfort on the left cheek. During the intraoral examination, we observed a very small erythematous lesion on the left lower buccal vestibule and slightly whitish striae on the right buccal mucosa. Her gingiva appeared to be slightly erythematous but showed significant improvement when compared to the previous visit. Her tongue appeared to be slightly dry but has generally improved since the last visit. After clinical observation, we concluded that she was a very compliant and cooperative patient; a patient deemed ideal for healthcare practitioners to work with. At the end of her first review appointment, she was suggested to continue maintaining the practice using those three goals within this holistic management. She was also introduced to several over-the-counter (OTC) products for oral dryness. She agreed to experiment with the recommended products to restore her oral moisture to an optimal level. The patient was advised to follow up after one month and to have a biopsy done when necessary.

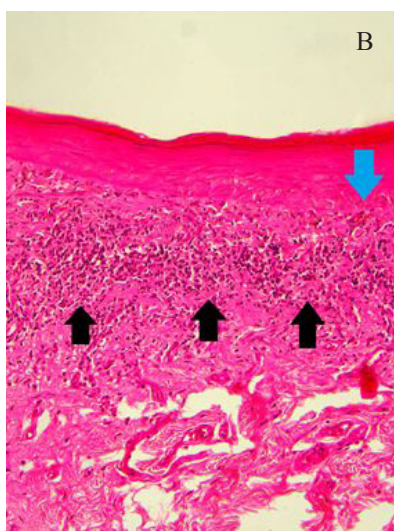
The patient returned after a month with promising newfound confidence; exuding happiness and cheerfulness that were absent during her initial visits to our clinic. The patient was no longer complaining regarding her mouth by the third visit. The patient was able to reduce their coffee intake to one cup per day and was able to expand her food options. The patient was doing well with OTC products. Intraorally, we found no evidence of any white lesion nor erythematous lesion on labial mucosa, tongue, the floor of the mouth, palate, and oropharynx. There were some vague white patches at the posterior right and left buccal mucosa extending to the gingiva from the molar to the first premolar. These white patches were also found at the right and left sides of the upper posterior gingiva. Again, upon these remarkably improved conditions, we asked her to maintain her current practice with oral hygiene, diet and topical steroid (if necessary). She was still complaining of stress at work; we kindly reminded her of our initial suggestion of going on a break or holiday. Eight months later, the patient visited us after a vacation in Europe. She was recharged and energetic. Her miserable past was history. The patient had been following the regime imposed by us which aided in the successful maintenance of her improved condition. We successfully convinced her to do an oral biopsy and informed consent was obtained.

An incisional biopsy was performed under local anaesthesia. Two sections of soft tissue samples were sent to the lab. In a biopsy report, the specimen showed marginally disintegrated oral mucosa, moderate lymphocytic infiltration, focal pigmentary incontinence within the lamina propria, surfaced by hyperplastic stratified squamous epithelium with thickened basal lamina. Some of the histopathologic features are shown in Figures 3a and 3b for the specimen. The oral





**Figure 3A.** Band-like lymphocyte infiltration within the superficial connective tissues (H&E, 20X)



**Figure 3B.** Lymphocyte infiltration occurs just below the epithelium/superficial aspect of the connective tissue (Black arrows), Basal degeneration vaguely seen (Blue arrow) (H&E, 40X)



**Figure 4. A and B.** Recent follow-up shows her right and left buccal mucosa is in stable condition with no symptoms

pathologist interpretation of the findings was an oral lichenoid reaction or oral lichen planus. The patient regularly visits our clinic every 6 months and remains compliant with continuing the clinician-imposed regime. Her oral lichen planus remains stable until his last appointment with us about 6 months ago. A

clinical photo was taken at that appointment as depicted in Figure 3. As for now, we continually having her for follow-ups in our clinic for the past 10 years and she is still doing great with minimal flare-ups of the symptoms. She was happy with her current conditions and is content with being able to continue with her daily activities and socializing.

## DISCUSSION

Oral lichen planus (OLP) is a noninfectious, symptomatic, chronic inflammatory T-cell mediated disease that affects the oral mucosal stratified squamous epithelium and the underlying lamina propria with or without additional skin lesions and rare spontaneous remission.<sup>19-21</sup> A high prevalence of OLP was found in non-Asian countries, among women and individuals over the age of 40 or those in their 50s and 60s. OLP signature lesions are symmetrical and bilateral, and the buccal mucosa is frequently affected.<sup>4,22</sup>

OLP diagnosis is established either by clinical examination only or by both clinical and histopathologic examinations. Direct immunofluorescence (DIF) in the histopathological examination is used as an adjunct to the above method of diagnostic work up and to rule out specific autoimmune diseases such as pemphigus and pemphigoid. Histopathologic features of OLP and OLR are similar with suggestions of certain discriminatory features by some authors. Topical corticosteroids are the treatment of choice for OLP although several other medications have been studied including retinoids, tacrolimus, cyclosporine, anti-inflammatory agents which specifically target T-lymphocytes, CO<sub>2</sub> laser and photodynamic therapy.<sup>23,24</sup> It has been studied that a single treatment of photodynamic therapy with topical methyl 5-aminolevulinat presented long-term improvement for OLP cases.<sup>25,26</sup> Lichenoid histology feature or clinical appearance can be performed in oral dysplasia, which may mimic other potentially premalignant features.

Additionally, another unusual lesion such as proliferative verrucous leukoplakia can frequently mimic oral lichen planus clinically, a differential diagnosis should be obtained through thorough clinical and histopathological examinations.<sup>27</sup> OLP and OLR tend to have one or more unknown antigens involvement. The OLP and OLR could be determined by the appearance of hyperkeratotic striation, varying degrees of mucosal inflammation from mild erythema to severe widespread ulceration, and a band-like infiltrate of mononuclear inflammatory cells including activated T lymphocytes, macrophages, and dendritic cells.<sup>28</sup>

The current cohort study found that nearly all patients with OLP were symptomatic at the initial visit, with location, type, gender, steroid use, and

medical conditions being predictors of symptomatic OLP. Despite improvement in OLP symptoms in most patients over time, 25% of patients who are asymptomatic at the initial visit present with symptomatic OLP at a future visit.<sup>29</sup>

Several contributing factors trigger OLP which can be classified as local and systemic factors including the usage of certain medications under specific drug groups, autoimmune diseases, diabetes, and hepatitis C virus infection.<sup>19,30</sup> Specific and non-specific antigen mechanisms had been implicated in the pathogenesis of OLP. Many studies have been trying to find an association between OLP and psychological states such as anxiety, depression and sleeping disturbances.<sup>31</sup> Other studies revealed an association between dyslipidemia and chronic inflammation in patients with lichen planus. They also stated that lipid levels test in a patient with lichen planus might be beneficial to detect individuals at risk and start preventive treatment against the development of cardiovascular disease.<sup>28,32,33</sup>

These findings correlated with our case and confirmed physiological disorders, as well as systemic diseases, are predisposing factors of OLP, given that our patient had hypercholesterolemia and unstable angina. A study done by Oczko et al.<sup>26</sup> suggested some possible associations between the signs, severity, and duration of symptoms of OLP, as well as the complaints and psychological correlation between anxiety and depression.<sup>34</sup> The mean age of OLP patients was around 59.9 years, which is similar to observations that the disease occurs in adult patients between 40 and 70 years old. Women were 80% of the studied patients.<sup>34</sup> Our case was comparable with Oczko et al.<sup>26</sup> since our patient's age is a 53-year-old female who was in a stressful working environment.<sup>34</sup>

Other studies showed an incidence of anxiety and depression in patients with OLP which negatively impacted patient's quality of life by impairing the patients' physical state, liveliness, mental health, stress level and social life.<sup>13,15,35</sup> Some researchers suggested that associated psychological treatment may be crucial in the follow-up of these kinds of patients.<sup>35-37</sup> In our present case, anamnesis showed how the patient was depressed after experiencing a long period of pain and oral problem paired with an arduous working environment. Based on our findings, we advised the patient to reduce stress at work and to take a break which she did. Moreover, the outcome was very positive and has improved the patient's quality of life remarkably.

Aside from continuously motivating and inculcating oral health care to the patient, several other aspects should also be considered when handling patients diagnosed with OLP.<sup>38</sup> A psychological approach, a patient-centred approach, and an established

sense of trust between patient and clinician should simultaneously be imposed along with the patient's understanding to improve the quality of life.<sup>39,40</sup> Interferences that improve individual protective factors may be highly advantageous in reducing stress and depression in some cases of severe OLP diseases.<sup>36</sup>

Holistic assessment is a vital part of providing high-quality overall health care concentrating not only on the disease itself but also the urgency of putting connection, caring attitude, communication, and control into each respective patient that is treated. Holistic assessment is a vital part of providing overall high-quality healthcare that simultaneously focuses on the disease itself, a caring attitude expressed by clinicians, good communication and proper patient control.<sup>41,42</sup> A holistic assessment demands to be considered in forefront of the clinician mind while reviewing all necessary aspects including the social, physical, psychological, and spiritual well-being of the patient.<sup>41,43</sup>

## CONCLUSION

Oral lichen planus regardless of its severity or type could be managed holistically and not just through a pharmacotherapeutic approach. Clinicians must acknowledge the necessity to recognize all initiating or contributing factors to be able to deal with the disease process. Pharmacotherapeutic therapy Dietary patterns and psychological aspects shall be considered too. Therefore, holistic management could be the best option for patients who suffer from chronic diseases with multiple contributing factors.

## CONFLICT OF INTEREST

The authors declare that there were no conflicts of interest related to this case report.

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