

Lichen planus of lip – Report of a rare case with review of literature

Shamimul Hasan¹

¹Department of Oral Medicine and Radiology, Faculty of Dentistry, Jamia Millia Islamia University, New Delhi, India

ABSTRACT

Lichen planus (LP) is a potentially malignant disorder with an immune-mediated etiopathogenesis. The condition frequently affects the skin, oral mucosa, skin appendages, and other mucous membranes. Oral lesions usually precede the onset of skin lesions and in majority of cases may only be presenting symptom. Isolated LP of the lip is rarely encountered in the clinical practice and is usually seen along with skin/other mucous membrane involvement. The clinical appearance poses diagnostic dilemmas and is often misinterpreted. This case report aims to highlight an interesting case of LP of the lower lip in a 50-year-old male patient. The patient presented with a diffuse erosive lesion on the lower lip bordered by white radiating striae on its inner aspect. Histopathological and immunofluorescent studies confirmed LP of the lip. Topical corticosteroids and Vaseline lip therapy were prescribed to the patient. There was considerable healing in the lip lesion during the follow-up period. However, recurrence was noted in the left buccal mucosa.

Keywords: Diagnosis, lichen planus, lichen planus lip, malignant lesion

Introduction

Lichen planus (LP) is a chronic mucocutaneous inflammatory disorder with an obscure etiopathogenesis,^[1] although immune-mediated and multifactorial basis have been proposed.^[2] LP primarily involves the skin and oral mucosa. However, other mucous membranes (genitalia, esophagus, and conjunctiva) and skin appendage areas (scalp and nails) may also be affected.^[3] Cutaneous LP lesions are usually self-limiting and may sometime cause pruritis. The oral lesions show chronicity and rarely undergo self-remission. The potentially premalignant nature of oral lesions often forms the basis for morbidity.^[4]

Predisposition of lip lesions to multiple injuries (lip biting, makeup application, or sunlight exposure) alters their clinical presentation, thus mimicking lesions of a diverse nature and posing diagnostic dilemma. Lip lesions in oral LP (OLP) usually

exhibit malignant potential, thus necessitating prompt diagnosis and management of such lesions.^[1]

Case Report

A 50-year-old male patient reported to the outpatient department with a chief complaint of ulcerations in the lower lip for the past 6 months. History reveals that the patient was asymptomatic 6 months back when he noticed itching sensation in the lower lip, followed by development of minute ulcerations in the lower lip region. For the past 2 months, he complained of burning sensation and mild dull intermittent pain in the ulcerated areas. The burning sensations aggravated on intake of hot and spicy foods. His medical history was nonsignificant, except for bidi smoking (two to three packets/day for the past 20 years). The patient has not taken any treatment for ulcerations. There was no associated history of vesicle formation in any area of oral cavity. The patient denied factitial habit of lip biting and excessive sun exposure. Physical examination revealed a diffuse erosive lesion on the lower lip extending till the mucocutaneous junction and inner aspect of lower labial mucosa. The diffuse erosive lesion roughly measured 2 × 3 cm and had irregular

Address for correspondence: Dr. Shamimul Hasan, C/O Mohd. Javed Khan, C-4, Duplex Quarters, New Sir Syed Nager, Aligarh, Uttar Pradesh, India.
E-mail: shamim0571@gmail.com

Access this article online

Quick Response Code:



Website:
www.jfmpc.com

DOI:
10.4103/jfmpc.jfmpc_24_19

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: reprints@medknow.com

How to cite this article: Hasan S. Lichen planus of lip – Report of a rare case with review of literature. J Family Med Prim Care 2019;8:1269-75.

margins. It was surrounded by erythema and covered with areas of blood-tinged crustations. Greyish-white radiating striae were seen bordering the inner aspect of lower labial mucosa. On palpation, the lesion was mildly tender and nonindurated. There was mild diffuse swelling over the lower lip. Gentle manipulation of the normal mucosa did not induce formation of new lesions (negative Nikolsky's sign) [Figure 1a and b]. The patient's oral hygiene was poor with generalized grade I mobility in teeth. There were no signs of associated oral, ocular, cutaneous, or genital lesions. History of chronic duration of 6 months, with the clinical evidence of diffuse erosive lesion bordered by characteristic radiating white striae, and a negative Nikolsky's sign led to a provisional diagnosis of OLP of the lower lip. Oral lichenoid reaction (OLR), discoid lupus erythematosus (DLE), pemphigus vulgaris, erythema multiforme, and actinic cheilitis were considered as the most probable differential diagnosis. OLRs were ruled out as there was no history of drug intake prior to the onset of the lesion. Also, there was no clinical evidence of amalgam restoration in patient's mandibular anterior teeth. Absence of vesicle/bullae formation, skin lesions, and a negative Nikolsky's sign ruled out the possibility of pemphigus vulgaris. Erythema multiforme was excluded due to the chronicity of the lesion (6-month duration), absence of vesicle/bullae, and absence of skin lesions (target lesions). DLE was excluded as the erosive lesion remained confined to the vermillion border without blurring the sharp line of the vermilion border, an important feature of LP of the lip. Also, the irradiating white striae are much more delicate in DLE when compared with LP. After informing the patient and obtaining consent, an incisional biopsy was performed from the perilesional region for histopathological and immunofluorescence studies. Histopathological features revealed liquefaction degeneration of epithelial basal layer and inflammatory infiltrates predominated by lymphocytes and saw tooth rete pegs in the connective tissue with no evidence of dysplastic changes [Figure 2]. Absence of atypia and dysplasia histologically excluded the possibility of leukoplakia and carcinoma *in situ*. OLRs were ruled out as inflammatory infiltrates predominantly comprised lymphocytes (in contrast to OLR where plasma cells, eosinophils, and neutrophils predominate the inflammatory infiltrate). Absence of parakeratosis, keratin plugging, and perivascular infiltrates ruled out lupus erythematosus. Direct immunofluorescence showed a shaggy band of fibrinogen in the basement membrane [Figure 3]. The histopathological and immunofluorescent features were consistent with the clinical diagnosis of OLP. The patient was subjected to thorough oral prophylaxis and instructed to maintain the oral hygiene. Thereafter, the patient was prescribed topical application of low-potency steroids (kenacort 0.1% paste three to four times daily), along with Vaseline lip therapy and chewable tablet of vitamin C (tab. lymcee two times daily) for a month. The patient was reviewed after 1 month and the lesions had considerably subsided with topical steroid treatment. Lip lesions showed almost complete resolution after 2 months of therapy [Figure 4a and b]. However, recurrence was seen on the left buccal mucosa as an erythematous lesion surrounded by interlacing white striae [Figure 5]. The patient was prescribed the

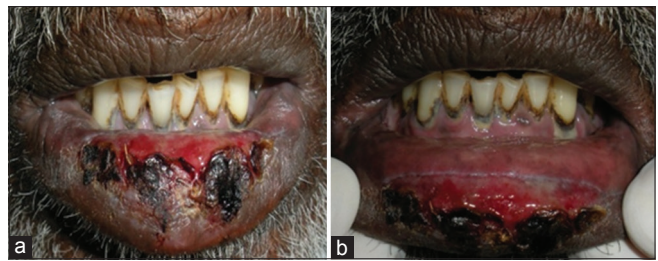


Figure 1: (a and b) Diffuse erosive lesion on lower lip covered by hemorrhagic crusts. Erosive lesion bordered by radiating white striae on the inner aspect

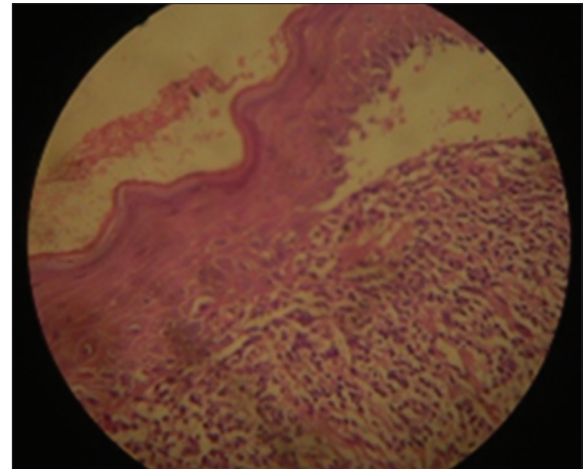


Figure 2: Histopathologic features of degeneration of epithelial basal layers and lymphocytic inflammatory infiltrates

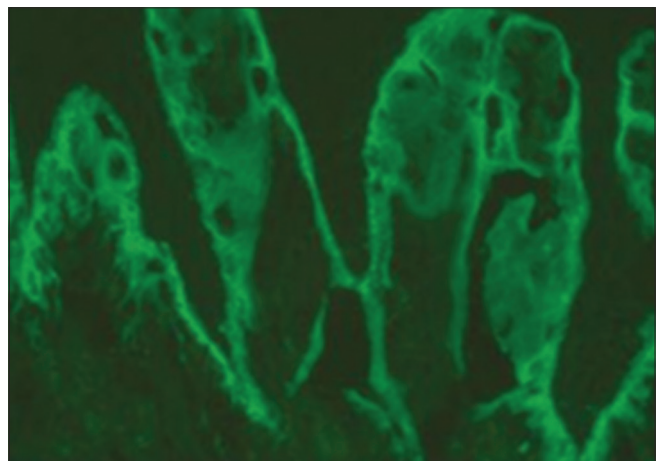


Figure 3: Immunofluorescence showing shaggy band of fibrinogen at the basement membrane

same therapy for recurrent lesions. Unfortunately, the patient did not report for further follow-up visits.

Discussion

LP is a chronic mucocutaneous disease of the stratified squamous epithelium. Mucous membranes of the oral and genital region, skin, scalp, and nails are the frequently affected sites.^[5] Prevalence rate is 0.1%–4% of the general population, most often in

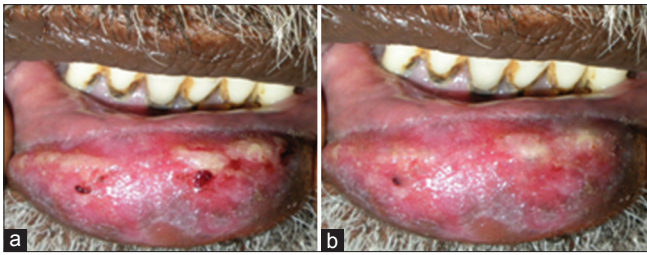


Figure 4: (a and b) Considerable resolution of the lesion after 1 month. Almost completely healed lesion after 2 months

perimenopausal women. The condition has a predilection for individuals in the age range of 30–60 years, although individuals of any age may be affected.^[6]

The exact etiology is still not completely elucidated, and multifactorial etiopathogenesis has been suggested. The autoimmune nature of LP has long been proposed, and CD8+ T lymphocytes bring about the apoptosis of the keratinocytes and result in damage to the epithelial basal cell layer. Allergic reaction to dental restorative agents (amalgam, gold), chronic irritants such as sharp cusp of tooth and ill-fitting prosthesis (Koebner phenomenon) and hepatitis B or C virus (HCV) infection seems to be the most likely causes of OLP. Genetic factors, lifestyle, and psychological factors (stress and anxiety) may also play a significant role in the pathogenesis of OLP. Lichenoid drug reaction may be caused due to many drugs (nonsteroidal anti-inflammatory drugs and angiotensin-converting enzyme drugs).^[7]

Our patient was a 50-year-old male who had a history of smoking (two packets/day) for the past 20 years. However, no other contributory factors could be elicited.

According to Andreason, OLP lesions can be classified into six different forms: reticular, papular, plaque-like, atrophic, ulcerative, and bullous.^[8] Recent classifications group OLP lesions into reticular (reticular, papular, plaque-like), erythematous (atrophic), and erosive (ulcerative, bullous) forms.^[9] Some authors have limited the forms to reticular (reticular, plaque-like) and erosive (atrophic, bullous, and ulcerative).^[10]

LP has a site predilection for buccal mucosa, followed by tongue (mainly the dorsum), gingiva, labial mucosa, and vermillion of the lower lip.^[11,12] LP isolated to a single oral site other than the gingiva is also unusual, although occasional cases of isolated lesions on the lips^[13] or tongue have been reported.^[8]

Our patient showed an isolated diffuse erosive lesion, 2 × 3 cm in diameter on the lower lip region, extending up to the mucocutaneous junction and on its inner aspect with white interlacing striae. Erosive lesion was covered with hemorrhagic crusts and erythema, and gentle manipulation of the normal mucosa elicited a negative Nikolsky's sign.

Only a few case reports and case series of LP of lips have been published so far [Table 1].



Figure 5: Recurrence of the lesion in the left buccal mucosa

The essential clinical characteristics usually comprise symmetrical, bilateral, and reticular lesions.^[14] Band-like lymphocytic infiltrates in the connective tissue area, hyperkeratosis with ortho- and/or parakeratosis, liquefaction degeneration of the epithelial basal layer, and absence of epithelial dysplasia constitute the characteristic histopathological features.^[15]

In the reported case, the histopathological features were characteristic for LP and aided in histological differential diagnosis with leukoplakia, invasive carcinoma, and other forms of cheilitis.

Immunofluorescence shows a linear pattern of fibrin and shaggy fibrinogen deposits at the epithelial basement membrane or cytooid bodies (Russell bodies), or both, in the absence of deposition of fibrinogen.^[16] Direct immunofluorescence features in the present case showed a shaggy band of fibrinogen at the basement membrane.

There is no definitive treatment protocol for OLP. The therapy mainly aims at improvement of symptoms and regular observation of dysplastic changes. Topical glucocorticoids usually form the mainstay of treatment for erosive OLP, although systemic and intralesional steroids are also in use.^[17] Use of orabase should be exclusively on moist intraoral sites, and topical steroids in orabase vehicle should be avoided for lip lesions.^[18]

Topical and systemic immunosuppressive agents, such as retinoids, cyclosporine, and tacrolimus, may also be used. Thalidomide and psoralen–UV-A are reserved for the recalcitrant disease.^[17]

Yu *et al.* reported the role of oral traditional Chinese medications and topical wet dressing in management of isolated lip LP.^[19] Surgical management, including cryosurgery and carbon dioxide (CO₂) laser, has been performed on OLP lesions. As LP is an inflammatory condition prone for recurrence, and therefore, surgical excision is not considered as the first-choice treatment of LP.^[20]

Table 1: Review of literature: Case reports of isolated lichen planus of lips/series											
Author	Year	No. of patients	Age/sex	Site	Features	Clinical type	Cutaneous/other mucosal/skin appendage involvement	Systemic pathologies	Diagnosis	Management	Results
Whittle CH	1937	01	69/M	Lower lip	Irritation	Plaque (appeared as leukoplakic patch)	Genital mucosa	NA	LPL?	Mercury, arsenic, X-rays	Stable
Altman J	1961	NA	NA	NA	NA	NA	NA	NA	LPL	NA	NA
Piamphongsant T.	1978	02	NA	Lower lip	NA	NA	NA	Lupus erythematosus	LPL + LES	NA	NA
Harland CC	1992	01	23/M	Lower lip + buccal mucosa	NA	Nodular	Skin involvement	H/O smoking 10 years back	OLP	Topical steroid	Recurrence as SCC, vermilionectomy and radiotherapy for SCC
Itin PH.	1995	01	44/M	Lower lip	Edematous lip with erosions and crusting	Erosive	No	NA	LPL	Acitretin + steroids + sunscreen	Resolved lesions
Allan SJ	1996	01	51/M	Lower lip	Itching and scales	Reticular	No	No	LPL	Steroids	Resolved lesions
Isogai Z.	1997	01	54/M	Lower lip	Pain	Erythematous	Skin+nails	NA	LPL	NA	NA
De Argila D	1997	01	51/F	Lower lip	NA	NA	No	NA	LPL	Chloroquine phosphate	Resolved lesions
Melato M	2000	01	NA	Upper lip	NA	NA	Morphea on upper lip	Vitiligo	LPL + morphea	NA	NA
Demitsu T	2000	01	62/F	Lower	NA	erosive	NA	NA	LPL	Steroid-resistant case treated with cyclosporine	Resolved lesion in 4 weeks
Cecchi R	2002	01	45/M	Lower lip	Swollen lip with burning	Erythematous/ulcerated	No	No	LPL	Steroid	Resolved lesion, lichenoid papule after 4 months
Chiang CT	2002	01	36/F	Lower lip	Painful ulcers	Erythematous	No	No	LPL + superficial mycosis	Unsatisfactory results from ketoconazole	Resolved lesions in 3 weeks, recurred 1 week after therapy
Yu TC	2003	01	44/M	Lower lip	Lip edema with burning pain	Erosive	No	Diet-controlled hypertension	LPL	Topical steroids	Resolved lesions in 6 weeks
Donovan JC	2005	01	51/M	Lip	Pain	Erosive	NA	Hep.-C-positive	LPL	Steroid refractory LP treated with 0.1% tacrolimus	Resolved lesions within 2 weeks of therapy, no recurrence after a year's follow-up

Contid...

Table 1: Contd...

Author	Year	No. of patients	Age/sex	Site	Features	Clinical type	Cutaneous/other mucosal/skin appendage involvement	Systemic pathologies	Diagnosis	Management	Results
Schichinohe R	2006	02	64/M 68/M	Lower lip+buccal mucosa Upper lip + buccal mucosa	Pain	Erosive	No	NA	OLP	Tacrolimus	Resolved lesions, No recurrences reported
Van Tuyll SAM	2007	01	74/F	Lower lip + buccal mucosa	Burning + bleeding	Bullous	Skin involved	No	OLP	Topical retinoids + steroids	Resolved lesions
Petruzzi M	2007	10	NA	Lower + upper lip	Erosion with hemorrhagic crusts	Erosive	NA	Five patients were Hep.-C-positive	LPL	Steroids + tocopherol	Eight patients showed complete resolution
Johnson H	2008	01	42/F	Lower lip	Dryness + peeling	Erosive	No	No	LPL	Tacrolimus	Stable
De Moraes PC	2011	01	07/F	Upper lip	NA	NA	NA	NA	OLP	Topical + intralesional steroids	Resolved lesion: lichenoid lesion after 3 years
Gencoglan G	2011	04	56/M, 61/M, 65/M, 22/M	Lower lip	Painful erosion/erythematous plaques	Erosive/erythematous	No	H/O smoking in one patient	LPL	Imiquimod cream	Lesion recurred in one case
Sugashima Y	2012	01	32/F	Lower + upper lip	Asymptomatic erythematous macule	Annular	No	Zinc allergy	LPL	Tacrolimus	Resolved lesion
Holmukhe S	2012	01	40/M	Lower lip	Asymptomatic annular lesion	Annular	No	No	LPL	Tacrolimus	No follow-up
Domingues E	2012	01	44/M	Lower lip	Painful hemorrhagic crusting	Erosive	No	Following treatment with imiquimod cream	LPL	Steroid	Resolved lesion
SamalDK	2015	01	52/M	Lower lip	Asymptomatic	Reticular	No	NA	LPL	Surgical excision	No recurrence on follow-up
Choi E	2017	01	62/F	Lower lip	Recurrent ulcerations	Reticular	Skin changes (lentiginous + wrinkles)	Precipitated by sun exposure	LPL	Topical tacrolimus + steroids + Vaseline lip therapy	No recurrences after 2-year follow-up
Hasan S	2017	01	50/M	Lower lip	Burning in the ulcerated region	Erosive	No	H/O smoking	LPL	Steroids + Vaseline therapy	Resolved lesion: recurrence seen

OLP: Oral lichen planus; SCC: Squamous cell carcinoma; LPL: Lichen planus Lip; LES: Lupus erythematosus; H/O: History of; NA: Not applicable

Our patient was prescribed topical application of low-potency steroid ((kenacort 0.1% paste three to four times daily), along with Vaseline lip therapy and chewable tablet of vitamin C (tab. lymcee two times daily) for a month. Lip lesions had considerably subsided with topical steroid treatment; however, recurrence on the left buccal mucosa was noted after 2 months of follow-up. The patient was lost for subsequent follow-up visits.

LP is associated with various comorbidities including diabetes mellitus, metabolic syndrome, dyslipidemia (predisposing factor for cardiovascular diseases), thyroid dysfunction (hypothyroidism), and HCV infection. Hence, patients with OLP require special attention from health professionals and should be thoroughly screened for early detection of these ailments, thus preventing complications and prolonged disabilities.^[21]

The predisposition to oral squamous cell carcinoma (OSCC) is the most dreaded complication of OLP.^[22] The first case of OLP with malignant changes was reported by Hallopeau in 1910.^[23] The predisposition of malignant transformation ranges between 0.4% and 5%,^[24] and about 1.1% of patients with OLP with a high frequency of smoking, alcohol abuse, and HCV infection have a propensity to develop OSCC.^[25] Our patient did not show any signs of malignant changes during the 2-month follow-up period.

Conclusion

Lip involvement is an uncommon and unusual site of presentation of OLP. Early and correct diagnosis and effective management together with periodic follow-up is mandatory to alleviate the pain and symptoms. This might also be a significant preventive protocol for development of squamous cell carcinoma in lip LP lesions.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

- Nuzzolo P, Celentano A, Bucci P, Adamo D, Ruoppo E, Leuci S, *et al.* Lichen planus of the lips: An intermediate disease between the skin and mucosa? Retrospective clinical study and review of the literature. *Int J Dermatol* 2016;55:473-81.
- Moger G, Thippanna CK, Kenchappa M, Puttalingaiah VD. Erosive oral lichen planus with cutaneous involvement in a 7-year-old girl: A rare case report. *J Indian Soc Pedod Prev Dent* 2013;31:197-200.
- Nogueira PA, Carneiro S, Ramos-e-Silva M. Oral lichen planus: An update on its pathogenesis. *Int J Dermatol* 2015;54:1005-10.
- Carbone M, Arduino PG, Carrozzo M, Gandolfo S, Argiolas MR, Bertolusso G, *et al.* Course of oral lichen planus: A retrospective study of 808 northern Italian patients. *Oral Dis* 2009;15:235-43.
- Canto AM, Müller H, Freitas RR, Santos PS. Oral lichen planus (OLP): Clinical and complementary diagnosis. *An Bras Dermatol* 2010;85:669-75.
- Zakrzewska JM, Chan ES, Thornhill MH. A systematic review of placebo-controlled randomized clinical trials of treatments used in oral lichen planus. *Br J Dermatol* 2005;153:336-41.
- Chatterjee K, Bhattacharya S, Mukherjee CG, Mazumdar A. A retrospective study of oral lichen planus in paediatric population. *J Oral Maxillofac Pathol* 2012;16:363-7.
- Andreason JO. OLP: A clinical evaluation of 115 cases. *Oral Surg Oral Med Oral Pathol* 1968;25:31-41.
- Eisen D. The clinical manifestations and treatment of OLP. *Dermatol Clin* 2003;21:79-89.
- Edwards PC, Kelsh R. OLP: Clinical presentation and management. *J Can Dent Assoc* 2002;68:494-9.
- Eisen D. The clinical features, malignant potential and systemic associations of oral lichen planus: A study of 723 patients. *J Am Acad Dermatol* 2002;46:207-14.
- Silverman SJr, Gorsky M, Lozada-Nur F. A prospective follow-up study of 570 patients with oral lichen planus: Persistence, remission, and malignant association. *Oral Surg Oral Med Oral Pathol* 1985;60:3034.
- Allan SJ, Buxton PK. Isolated lichen planus of the lip. *Br J Dermatol* 1996;135:145-6.
- Mattsson U, Jontell M, Holmstrup P. Oral lichen planus and malignant transformation: Is a recall of patients justified? *Crit Rev Oral Biol Med* 2002;13:390-6.
- Cassol-Spanemberg J, Rodríguez-de Rivera-Campillo ME, Otero-Rey EM, Estrugo-Devesa A, Jané-Salas E, López-López J. Oral lichen planus and its relationship with systemic diseases. A review of evidence. *J Clin Exp Dent* 2018;10:938-44.
- Helander SD, Rogers RS III. The sensitivity and specificity of direct immunofluorescence testing in disorders of mucous membranes. *J Am Acad Dermatol* 1994;30:65-75.
- Torti DC, Jorizzo JL, McCarty MA. Oral lichen planus. A case series with emphasis on therapy. *Arch Dermatol* 2007;143:511-5.
- Nico MM, Fernandes JD, Lourenço SV. Lichen planus affecting the lips. *J Clin Exp Dermatol Res* 2015;6:306:1-4.
- Yu F, Xu N, Zhao B, Ren X, Zhang F. Successful treatment of isolated oral lichen planus on lower lip with traditional Chinese medicine and topical wet dressing: A case report. *Medicine (Baltimore)* 2018;97:50.
- Moraes MD, Matos FRD, Pereira JDS, Medeiros AMCD, Silveira EJDD. Oral lichen planus: Two case reports in male patients. *Rev Odonto Ciênc* 2010;25:208-12.
- Kumar SA, KrishnamRaju PV, Gopal K, Rao TN. Comorbidities in lichen planus: A case-control study in Indian patients.

- Indian Dermatol Online J 2019;10:34-7.
22. Chaiyarit P, Ma N, Hiraku Y, Pinlaor S, Yongvanit P, Jintakanon D, *et al.* Nitrate and oxidative DNA damage in oral lichen planus in relation to human oral carcinogenesis. *Cancer Sci* 2005;96:553-9.
 23. Yu TC, Kelly SC, Weinberg JM, Scheinfeld NS. Isolated lichen planus of the lower lip. *Cutis* 2003;71:210-2.
 24. Van der Meij EH, Schepman KP, Smeele LE, van derWal JE, Bezemer PD, van der Waal I. A review of the recent literature regarding malignant transformation of oral lichen planus. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1999;88:307-10.
 25. Aghbari SMH, Abushouk AI, Attia A, Elmaraezy A, Menshawy A, Ahmed MS, *et al.* Malignant transformation of oral lichen planus and oral lichenoid lesions: A meta-analysis of 20095 patient data. *Oral Oncol* 2017;68:92-102.