

Bullous Type Lichen Planus: A Rare Case Report

Büllöz Tip Liken Planus: Nadir Bir Olgu Sunumu

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ABSTRACT

Objective: Oral bullous lichen planus (BLP) is an uncommon form of lichen planus that affects just 1% of the oral mucosa. It is seen in oral mucous, palate, buccal mucous, and occasionally in the tongue. The purpose of this case report is the importance of including lichen planus in the differential diagnosis of leukoplakia-like lesions.

Case: The presentation included lateral parts of the tongue, sublingual, and hyperkeratotic areas on the cheek, as well as asymptomatic bullous lichen planus phenomena, clinical and histological characteristics that have been present for 5 years. Systemic corticosteroid was given to the patient who had no skin symptoms.

Conclusion: Burning sensation, the BLP's common clinical symptom, which is infrequently observed in the oral mucosa, may not be visible in all patients. In addition, BLP can be confused with leukoplakia when observed in plaque form. Histopathological examination is mandatory for definitive diagnosis in terms of malignant potential.

Keywords: Bullous lichen planus, leukoplakia, white lesion

Alınış / Received: 10.12.2021 Kabul / Accepted: 08.08.2022 Online Yayınlama / Published Online: 31.08.2022



ÖZET

Amaç: Oral büllöz liken planus (BLP), liken planusun nadir görülen bir formu olup, oral mukozada %1 oranında görülmektedir. Oral mukozada damak, bukkal mukoza ve nadiren dilde görülür. Bu olgu sunumun amacı lökoplaki benzeri görülen lezyonların ayırcı tanısında, liken planusun da bulundurulması gerekliliğinin önemidir.

Olgu Raporu: Bu sunumda 5 yıldır var olan, dilin lateral kısımları, dil altı ve yanakta hiperkeratotik alanlar şeklinde izlenen asemptomatik büllöz liken planus olgusu, klinik ve histopatolojik özellikleri sunuldu. Herhangi bir cilt bulgusu olmayan hastaya sistemik kortikosteroid tedavisi başlandı.

Sonuç: Oral mukozada çok nadir olarak izlenen BLP'un yaygın klinik belirtisi olan yanma hissi her hastada görülmeyebilir. Ayrıca BLP plak formunda izlendiğinde lökoplaki ile karıştırılabilir. Malignite potansiyeli açısından kesin tanı için histopatolojik inceleme zorunlu olup, bu sayede doğru tanıya gitme süreci hızlandırılmalıdır.

Anahtar Kelimeler: Büllöz Liken Planus, lökoplaki, beyaz lezyon



1. Introduction

Lichen planus is a dermatological disease that affects skin and mucous membrane, usually seen in middle-aged individual. [1] The global prevalence is estimated to be between 0.5% and 2%. [1] It can be observed in genital region, eyes, esophageal mucosa, nails, skin with hair, and oral region. [3] Oral lichen planus (OLP) is characterized by bilateral white lines or plaques on the buccal mucosa, language, or gums, and is more common in women by the ratio of 1.4:1. [2] Clinical manifestations of the OLP include reticular, atrophic, papular, erosive (ulcerative), bullous, and plaque, which can develop alone or in combination. [3] The most commonly monitored reticular OLP is seen as interconnecting white lines (Wickham lines) in the bilateral posterior buccal mucosa, but it can also be detected on the dorsal and lateral surfaces of the tongue, the teeth, and the vermillion edge. [4] On the dorsum of the tongue and buccal mucosa, the plaque form is more common. [4] Erosive and atrophic OLP are characterized by erythematous patches surrounded by thin white lines, as well as pain that is aggravated by spicy or hot foods. [4,5]

A bullous lichen planus is one of the uncommon types of lichen planus (BLP). [6] It can be seen on the hands, feet, dorsal face of the body, and most commonly the legs. Compared to other kinds of lichen planus, it can be found at a rate of 1% in oral mucosa. Bilateral lesions are rather common. The lesions have a clinical appearance as an erythematous area or solid bulla surrounded by white lines. [9] Lesions can cause a burning sensation as well as pain. Due to the small number of cases documented in the literature, the etiology and pathogenesis of BLP are yet unknown. [6,7, 9]

2. Case

A 65-year-old healthy male has applied for a normal clinical checkup at an oral and maxillofacial radiology. The patient's oral examination revealed white hyperkeratotic lesions in the right-hand tongue laterals (Figure 1/A), which were approximately 5 cm x 2 cm in size, and the left cheek mucosa (Figure 1/B). A 3 cm x 1 cm ulcerated lesion at the junction of the sublingual and floor of the mouth (Figure 1/C) was also seen.

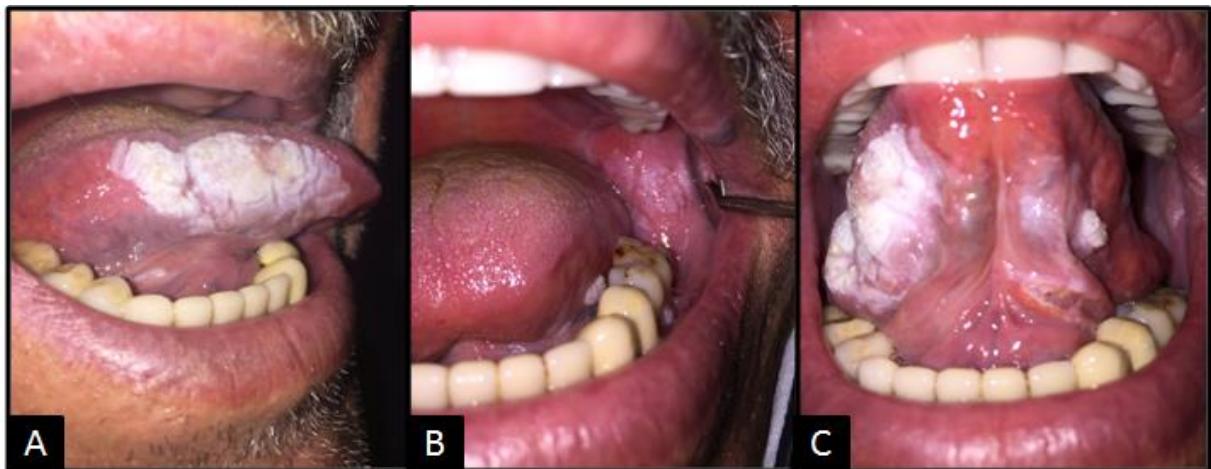


Figure 1: A view of the patient's tongue lesions and buccal mucous.

These lesions which look like plaque form have been learned to exist for around 5 years. There were no skin symptoms or stories of smoking or alcohol in the patient with asymptomatic lesions. A double-incision biopsy was performed on the patient. Due to extensive decomposition limits and lichenoid infiltrations observed during the histological examination (Figure 2/A, B), the lesion was diagnosed with oral BLP.

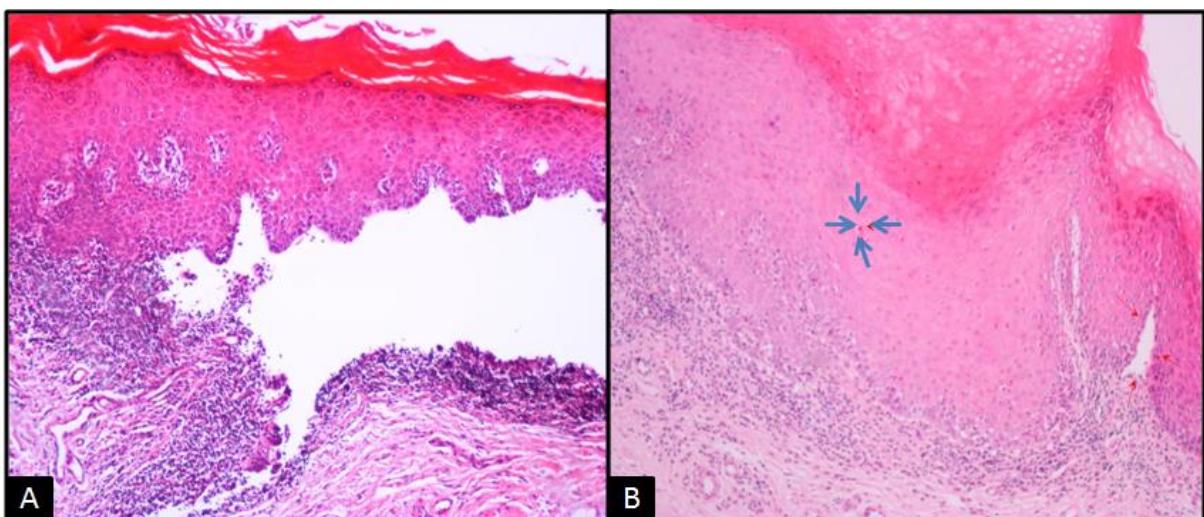


Figure 2: Oral lichen planus. A. Biopathal produced from tongue laterals showing heavy liquid penetration (HEX10) at a wide degradation limit under epithelial conditions. B. Sublingual bioptotic liquid penetration under epithelial (HEX10) conditions (blue arrows: Apoptotic objects)

The patient was consulted to a dermatological clinic, where Prednol 4 mg (2x2) and Mofeccept 500 mg were prescribed (1x1). The gastrointestinal department recommended an HBV test every three months for the patient who had negative Elisa and hepatitis testing.

3.Discussion

The bullous form of lichen planus, which is usually visible as Wickham lines in reticular shape, is extremely unusual [6], and there are only a few case reports in the literature as far as we know. [6,7,9] Oral BLP presents as small bullae and vesicle that can rupture easily, exposing an ulcerated and painful surface when ruptured. Lichen planus plaques, like reticular OLP, can be confused with leucoplakia [4] and are often asymptomatic. [10] Lichen planus that is atrophic/erosive, ulcerative, or bullous frequently has oral symptoms that need to be treated. [4] Many people with BLP have described experiencing a burning sensation. [9,11] In addition, there are two forms of BLP: familial and

non-familial BLP. [12] A long illness course, extensive rashes, frequent nail involvement, and a younger patient group are all present in the familial variant. [12] There was no complaint of burning in the tongue, and the patient was fully asymptomatic, despite the existence of white plaque lesions on the tongue and an ulcerated lesion at the junction of the sublingual and floor of the mouth. It was discovered that no other family members had identical lesions. The histological investigation revealed extensive separation zones under the epithelium and lichenoid infiltration, which led to the diagnosis of BLP.

In the differential diagnosis of BLP, subepidermal bullous disorders, particularly lichen planus pemphigoid, should be evaluated. [14] The degenerative feature between the epidermal and basal layers in classical lichen planus distinguishes BLP lesions from lichen planus pemphigoid. Furthermore, oral lichenoid responses can be triggered by a variety of dental materials (such as amalgam, composite, metals). [15] In most cases, the repair heals quickly after it has been removed. [16] Smoking raises the risk of lichen planus tumors, which the World Health Organization classifies as a premalignant lesion. [16] The chances of developing a tumor in the buccal mucosa, where OLP is most common, are minimal, but they are high at the tongue edges. [16] Oral cancer risks, particularly those related with the erosive and atrophic forms, should be decreased. [4] Follow-up is required for reticular and other asymptomatic forms of OLP. There is no need for treatment if there is no ulceration; however, follow-up is encouraged. [4] However, treatment is essential if there are symptoms and a risk of malignancy. [4] Topical steroids with a high potency are used to treat BLP. Oral betamethasone is used to treat oral BLP that is moderate to severe. In circumstances where local therapies have failed, systemic corticosteroids are utilized. [17] Because of the plaque-like appearance of the lesions in the patient who did not smoke, leukoplakia with a high first malignant transformation [18] was deemed a preliminary diagnosis in this case. Because no drugs or prostheses were used, a lichenoid reaction was ruled out of the differential diagnosis. Lichen planus was included in the differential diagnosis because of the bilateral and multifocal lesions. Despite the fact that the patient was asymptomatic, she was referred to a dermatologist due to the danger of premalignancy due to lesions on the lateral edges of the tongue and the floor of the mouth. The patient was given systemic corticosteroid medication and was monitored for 7 months.

4. Conclusion

Burning sensation, which is a common clinical symptom of BLP, which is observed very rarely in the oral mucosa, may not be seen in every patient. In the differential diagnosis of plaque-like lesions, lichen planus should also be considered. A definitive diagnosis requires a histopathological study, and the process of determining the accurate diagnosis should be accelerated.

References

- [1] Schlosser, B. J. 2010. Lichen planus and lichenoid reactions of the oral mucosa. *Dermatologic therapy*, 23(3), 251-267.
- [2] Chainani-Wu, N., Silverman Jr, S., Lozada-Nur, F., Mayer, P., & Watson, JJ. 2001. Oral lichen planus: patient profile, disease progression and treatment responses. *J Am Dent Assoc*, 132 (7), 901-909.
- [3] McCartan, BE., Healy, C. 2008. The reported prevalence of oral lichen planus: a review and critique. *J Oral Pathol Med.*, 37 (8), 447-453.
- [4] Mollaoglu N. 2000. Oral lichen planus: a review. *British Journal of Oral and Maxillofacial Surgery*, 38,370–377.
- [5] Gunhan Ö. ed. 2015. *Oral Maxillofacial Pathology*. Quintessence Publishing, İstanbul, 461s.
- [6] Liakopoulou, A., Rallis, E. 2017. Bullous lichen planus—a review. *Journal of dermatological case reports*, 11(1), 1.
- [7] Babu, A., Chellawamy, S., Muthukumar, S., Pandey, B., Jayaraj, M., Francis, S. 2019. Bullous lichen planus: Case report and review. *Journal of pharmacy & bioallied sciences*, 11(Suppl 2), 499s.
- [8] Karthikeyan, K., Jeevankumar, B., Thappa, DM. 2003. Bullous lichen planus of the glans penis. *Dermatol Online J*, 9(5):31.

- [9] Shirasuna, K. 2014. Oral lichen planus: Malignant potential and diagnosis. *Oral science international*, 11(1), 1-7.
- [10] Huber, M. A. 2010. White oral lesions, actinic cheilitis and leukoplakia: confusions in terminology and definition: facts and controversies. *Clinics in dermatology*, 28(3), 262-268.
- [11] Patil, A., Prasad S., Ashok L., Sujatha GP. 2012. Oral bullous lichen planus: Case report and review of management. *Contemp Clin Dent.*, 3(3):344-8.
- [12] Huang C., Yan X., Yang L., Zhang J., Tian J., Li J., Wang C., Tu Y. 2007. A retrospective and comparative study of familial and non-familial bullous lichen planus. *J Huazhong Univ Sci Technolog Med Sci.*, 27(3):336-8.
- [13] Cheng, YSL., Gould, A., Kurago, Z., Fantasia, J., Muller, S. 2016. Diagnosis of oral lichen planus: a position paper of the American Academy of Oral and Maxillofacial Pathology. *Oral Surg Oral Med Oral Pathol Oral Radiol*, 122(3):332-54
- [14] Gawkrodger, DJ., Stavropoulos, PG., McLaren, KM., Buxton, PK. 1989. Bullous lichen planus and lichen planus pemphigoides--clinicopathological comparisons. *Clin Exp Dermatol*, 14(2):150-3.
- [15] Ismail, S. B., Kumar, S. K., Zain, R. B. 2007. Oral lichen planus and lichenoid reactions: etiopathogenesis, diagnosis, management and malignant transformation. *Journal of oral science*, 49(2), 89-106.
- [16] Regezi J. A, Sciubba J. J., Jordan R. C. K. ed. 2017. *Oral Pathology: Clinical Pathologic Correlations*, 7th, completely revised and enlarged edition. Elsevier Health Sciences, St. Louis. 402s.
- [17] Asch, S., Goldenberg, G. 2011. Systemic treatment of cutaneous lichen planus: an update. *Cutis*, 87(3):129-34.
- [18] Srivastava, VK. 2014. To Study the Prevalence of Premalignancies in Teenagers having Betel, Gutkha, Khaini, Tobacco Chewing, Beedi and Ganja Smoking Habit and Their Association with Social Class and Education Status. *Int J Clin Pediatr Dent*, 7(2):86-92.