

Palmoplantar Lichen Planus with Papular Oral Lesions: Case Report and a Brief Review of the Literature

Palmoplantar Liken Planus ve Papüler Oral Lezyonlar: Olgu Sunumu ve Literatürün Gözden Geçirilmesi

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ABSTRACT Lichen planus (LP) is a common inflammatory dermatosis with a chronic course which is typically characterized by small, violaceous, flat-topped polygonal papules mostly seen on the flexor aspects of the wrists, the lumbar region, and around the ankles. The etiopathogenesis has not been fully elucidated yet, however a T cell-mediated immune response to an antigenic stimulus is mostly favored. It has been proposed that LP is an outcome of an interaction between endogenous and exogenous factors. Microbial agents, metals, chemicals, and drugs are the most well-known exogenous antigenic factors in the induction or aggravation of LP. Besides the classical form, there are several other clinical variants of LP which are categorized according to morphology, configuration and distribution pattern of the lesions. Palmoplantar LP is a rare variant of LP, which is generally characterized by erythematous scaly plaques with a scaly peripheral collarette or diffuse hyperkeratotic yellow plaques similar in appearance to keratodermas. Herein, a case of toxicant-induced palmoplantar LP with typical cutaneous lesions and distinctive mucosal involvement is presented.

Key Words: Lichen planus; liken planus, oral; keratoderma, palmoplantar

ÖZET Liken planus (LP), sıklıkla el bileklerinin iç yüzü, ayak bilekleri ve lumbal bölgede görülen küçük, viyolese, düz yüzeyle, poligonale papüllerle karakterize, kronik seyirli, sık karşılaşılan inflamatuvar bir dermatozdur. Etiyopatogenezi henüz tüm yönleriyle aydınlatılmamış olmakla birlikte, antijenik bir uyaranla tetiklenen T hücre kaynaklı immünolojik bir reaksiyon olduğu en çok ileri sürülen görüştür. Ayrıca, LP'nin endojen ve eksojen faktörlerin etkileşimi nedeniyle geliştiği de bilinmektedir. Mikrobiyal ajanlar, metallere, kimyasallar ve ilaçlar LP'yi tetikleyen veya alev eden en önemli eksojen faktörlerdir. Klasik tutulumun dışında lezyonların morfolojisi ve konfigürasyonuna göre kategorize edilen çok sayıda klinik varyant da bulunmaktadır. Palmoplantar LP, sıklıkla periferik skuamli eritematöz plaklar ya da keratoderma benzeri diffüz sarı plaklarla karakterize nadir rastlanan bir LP varyantıdır. Biz de burada, toksik ajan maruziyeti sonrası ortaya çıkmış, tipik kutanöz bulgular ve dikkat çekici mukozal lezyonların eşlik ettiği bir palmoplantar LP olgusunu sunuyoruz.

Anahtar Kelimeler: Liken planus; liken planus, oral; keratoderma, palmoplantar

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Lichen planus (LP) is an inflammatory dermatosis of unknown origin that affects skin, mucous membranes, and nails. It occurs worldwide and although the exact prevalence is unknown, the frequency is between 0.5 and 1.0%. Rarely affecting children, LP is mostly a disease of middle aged persons. The clinical manifestations of LP are extremely diverse with atypical presentations and different combinations of cutaneous, mu-

cosal and appendageal lesions.^{1,2} Palmoplantar LP is one of the rare variants of LP which is known to be resistant to treatment, and usually misdiagnosed unless the other clinical features of LP is obvious.^{3,4} Here, we report a case of toxicant-induced palmoplantar LP in a 28-year-old male with pronounced mucosal lesions and characteristic cutaneous involvement with violaceous, flat-topped polygonal papules and plaques.

CASE REPORT

A 28-year-old male patient admitted to our outpatient clinic with a 6-month history of violaceous pruritic eruption. Working as a junkman, he had been exposed to an unknown chemical substance in a metal container before the eruption began. There was no family history and past history of any other diseases or medication. Upon examination, on the trunk and the extremities, it was found that there were multiple, erythematous–violaceous, polygonal papules, several millimeters in size, coalescing to form symmetrical plaques especially on the shins, volar surfaces of elbows and ankles (Figure 1). Examination also revealed slightly scaly, yellow, hard hyperkeratotic papules clustered to form bilaterally symmetrical plaques on the middle and lower portion of the palms and the thenar eminences (Figure 2). It was noticed that the papules, some of which have scaly peripheral collarette, extended from inner aspect of the ankles to the plantar arches. The lesions with collarettes were darker in colour with more violaceous tint and looked as if they were umbilicated (Figure 3). Oral examination demonstrated abundant amount of small white pinpoint papules almost entirely covering oral vestibule, lips, tongue, and bilateral buccal mucosa, at some points merging to form reticular striations (Figure 4). The patient also had erythematous papules on the glans penis and longitudinal ridging of fingernails (Figure 5).

Laboratory studies including complete blood count and differential, erythrocyte sedimentation rate, serum chemistry profile, urinalysis, thyroid panel were within normal limits. Serologic tests for hepatitis B, C, and syphilis were negative. Patch testing with the European standard series of aller-

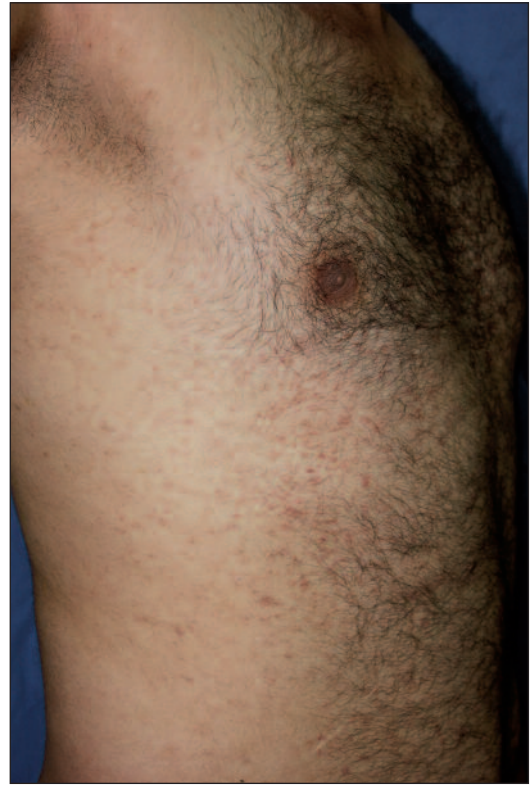


FIGURE 1: Multiple, 1-5 mm, erythematous to violaceous, plane-topped papules evident on the trunk.

(See color figure at <http://www.turkiyeklinikleri.com/journal/dermatoloji-dergisi/1300-0330/>)



FIGURE 2: Symmetric, yellowish, hyperkeratotic plaques with a violaceous rim on the midpalm and wrists accompanied by violaceous, polygonal papules on the flexor aspects of the forearms.

(See color figure at <http://www.turkiyeklinikleri.com/journal/dermatoloji-dergisi/1300-0330/>)

gens showed a weak positive response (+1) for the sesquiterpene lactone mix at 48 and 72 hours and a doubtful reaction to para-tertiary-butylphenol (PTBP)-formaldehyde resin at 48 hours. Histopathological examination of the skin biopsy



FIGURE 3: Multiple, erythematous papules some of which are with a violaceous hue and scaly peripheral collarettes located predominantly on the inner aspects of the ankles and plantar arches.

(See color figure at <http://www.turkiyeklinikleri.com/journal/dermatoloji-dergisi/1300-0330/>)



FIGURE 4: Numerous pinpoint white papules in the oral cavity.

(See color figure at <http://www.turkiyeklinikleri.com/journal/dermatoloji-dergisi/1300-0330/>)

from plantar region revealed marked compact hyperkeratosis, acanthosis, hypergranulosis, superficial band-like lymphocytic infiltration with hydropic degeneration of the basal layer (Figures 6, 7). In addition histopathological examination of the skin biopsy from erythematous–violaceous papules on the volar surface of the left ankle showed apoptotic keratinocytes in the basal layer of the epidermis. The basal damage was associated with a band-like infiltrate of lymphocytes in the papillary dermis and there was variable melanin incontinence (Figures 8, 9). As a result, a diagnosis of LP was made both with clinical and histopatholog-



FIGURE 5: Longitudinal ridging of fingernails.

(See color figure at <http://www.turkiyeklinikleri.com/journal/dermatoloji-dergisi/1300-0330/>)

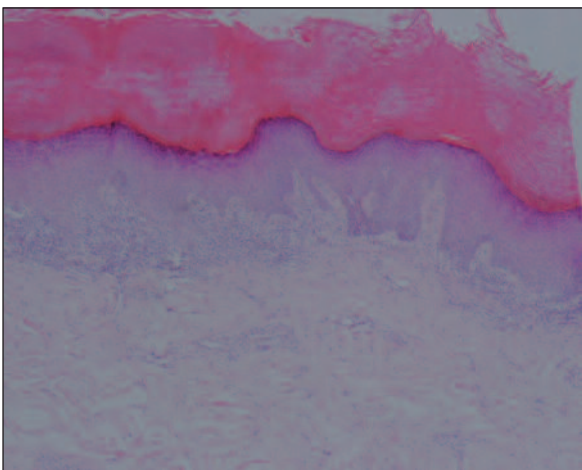


FIGURE 6: Marked compact hyperkeratosis, acanthosis, hypergranulosis and superficial band-like lymphocytic infiltrate (H&Ex40).

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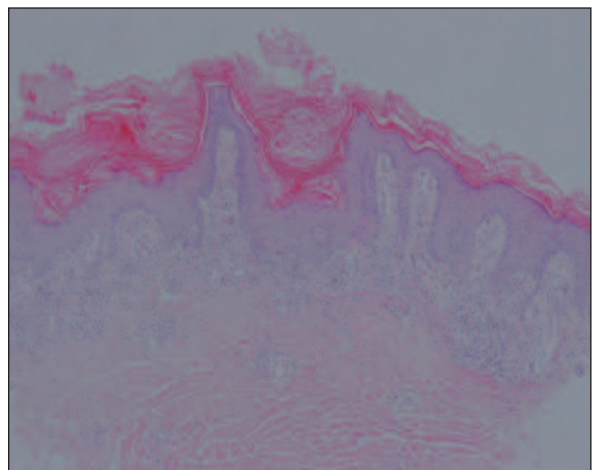


FIGURE 7: Hyperkeratosis, acanthosis, hypergranulosis, band-like infiltrate of lymphocytes in the papillary dermis and there was variable melanin incontinence (H&Ex40).

(See color figure at <http://www.turkiyeklinikleri.com/journal/dermatoloji-dergisi/1300-0330/>)

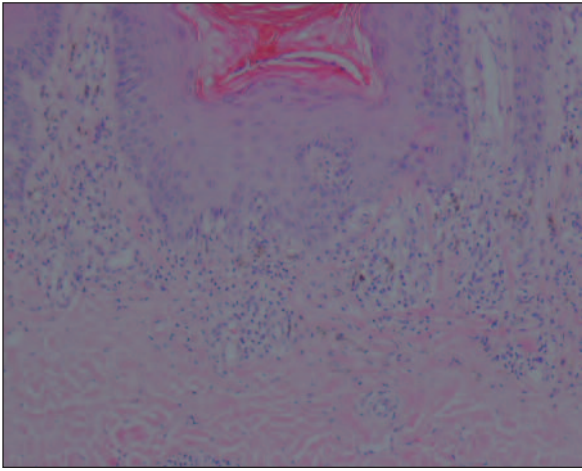


FIGURE 8: Interface dermatitis, Civatte (Colloid) bodies (H&E \times 100).
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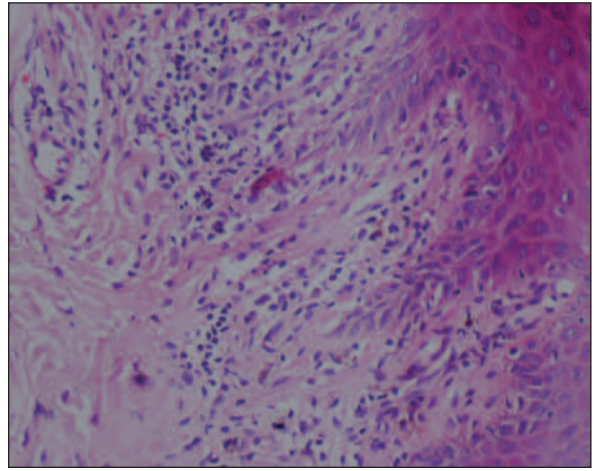


FIGURE 9: Interface dermatitis with Colloid bodies and melanophages (H&E \times 200).
(See color figure at <http://www.turkiyeklinikleri.com/journal/dermatoloji-dergisi/1300-0330/>)

ical findings. The patient was given a therapy of 0,5 mg/kg/day of oral methylprednisolone, topical keratolytics, and emollients. As the patient showed clinical improvement, oral methylprednisolone was tapered gradually and discontinued within 1 month, instead topical corticosteroids were added to the treatment regimen. After intermittent application of topical corticosteroids and keratolytics for 3 months, the patient had a near full improvement of the lesions.

DISCUSSION

Palmoplantar LP is a rare variant of LP, which is most commonly seen in middle-aged men. Unlike to other manifestations, it usually creates a clinical dilemma to diagnose palmoplantar LP, if the other characteristic features of LP is not distinguishable.⁵⁻⁹ Palmoplantar LP may manifest in several clinical presentations, including hyperkeratotic keratoderma-like plaques,^{4,5} ulcerative lesions,^{9,10} vesicle-like papules,^{3,5} umbilicated papules,¹¹ diffuse palmar hyperpigmentation,¹² erythematous scaly plaques,⁵ punctate keratosis¹³ and acrosyringial variant.¹⁴ Actually, variants of palmoplantar LP, with other clinical manifestations of LP may appear contemporaneously in an individual.^{3-5,9,15,16} There are case reports indicating hyperkeratotic palmoplantar plaques accompanying oral manifestations and typical cutaneous lesions of LP.^{4,15} Likewise, plantar

ulcero-erosive lesions of LP have been described with longitudinal melanonychia, reticular oral LP, and post inflammatory hyperpigmentation.^{9,16}

Up to date the most comprehensive investigation regarding palmoplantar LP has been held by Sanchez-Peres et al.⁵ Although it can't be figured out which combinations of clinical presentations their patients had displayed, they reported that simultaneous appearance rate of LP in the palmoplantar region and elsewhere was 26%. In this study the palmoplantar lesions were classified according to prevailing morphological patterns, as the erythematous scaly variant was the most common type observed, occurring in three-quarters of the patients, and the hyperkeratotic pattern was the second most prevalent one, described in the remaining one-quarter. In two of the patients with hyperkeratotic variant, lesions were arranged in a diffuse fashion forming plaques identical to keratodermas, besides one of them had concomitant vesicle-like papular lesions.⁵ Gündüz et al. also reported a patient with white reticular plaques on bilateral buccal mucosa and palmoplantar LP with hyperkeratotic papules, some of which looked alike vesicles.³ However, Sanchez-Peres et al. could not exclude the possibility of coexistence of atopic dermatitis and LP, since the patient had a personal and family history of atopic dermatitis and the vesicular lesions were located on the sides of the fingers

and the palms.⁵ On the other hand, in the case of Gündüz et al.'s the vesicle-like lesions were actually LP papules which were also histopathologically confirmed.³

Our case not only represents the unique combination of erythematous scaly and hyperkeratotic patterns of palmoplantar LP, but also displays papular type oral LP. Papular type oral LP is rarely seen and usually coexists with other variants of oral LP. Indeed these minute whitish papules most generally appear transitionally in the initial phase of the disease.¹⁷⁻¹⁹ As we checked through the literature, we have noticed that palmoplantar LP is infrequent, furthermore different patterns occurring together is unlikely, reported almost exclusively for vesicle-like papules accompanying hyperkeratotic variant.^{3,5} In addition, our literature search did not reveal any case report with papular type oral LP, which is already an uncommon variety, concurrent with palmoplantar LP. Hence, we think that our case is an exceptional example of palmoplantar

LP. Nevertheless, we could not identify what was the etiological factor to initiate LP in our patient. Although the patch testing showed positive response for the sesquiterpene lactone mix, contact hypersensitivity to sesquiterpene lactone mix have not yet been shown to induce LP. As we trust the patient's history, we presume that the unknown chemical agent or the metallic substance he had been exposed must be the initiating factor. In addition, according to the Naranjo Adverse Drug Reaction (ADR) Probability Scale, which assesses the likelihood of a substance-related adverse reaction,²⁰ an ADR due to sesquiterpene lactone mix in this case was doubtful.

Based upon our case report, we discussed the clinical features and associations of palmoplantar LP. To our knowledge, this is the first case report with papular oral lesions, typical cutaneous manifestations and concurrent presentation of both the first and second most common patterns of palmoplantar LP.

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