

An Acral Pityriasis Lichenoides Chronica Case Presenting as Plantar Keratoderma

Plantar Keratoderma ile Prezente Olan Bir Akral Pitriyazis Likenoides Kronika Olgusu

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ABSTRACT Pityriasis lichenoides chronica (PLC) is a rare papulosquamous dermatosis with a relapsing and remitting course. It has been proposed that PLC is a clonal T cell lymphoproliferative disease triggered by an antigenic stimulus. Although the infectious agents and drugs are the possible etiopathological factors, the etiology of PLC still remains unclear. PLC is usually characterized by recurrent crops of erythematous-brown papules predominantly localized on trunk and proximal extremities. Atypical presentations of PLC with unusual sites of the involvement have been described rarely so far. Notably, a significant number of these presentations are acral or segmental variants. Here, to our knowledge, we report the first case of acral PLC presenting as plantar keratoderma.

Key Words: Pityriasis lichenoides; keratoderma, palmoplantar

ÖZET Pitriyazis likenoides kronika (PLK), remisyon ve alevlenmelerle seyreden, nadir rastlanan papüloskuamöz bir dermatozdur. PLK'nın antijenik bir uyaranla tetiklenen klonal T- hücre kaynaklı lenfoproliferatif bir hastalık olduğu düşünülmektedir. Her ne kadar enfeksiyöz ajanlar ve ilaçlar muhtemel etiopatolojik faktörler olsa da, PLK'nın etiolojisi henüz tüm yönleriyle aydınlatılmamıştır. PLK genellikle gövde ve proksimal ekstremitelerde yerleşen, tekrarlayan eritematöz-kahverengi papüllerle karakterizedir. Günümüze kadar beklenmedik lokalizasyonlarda ortaya çıkan atipik PLK olgusu nadiren tanımlanmıştır. Kayda değer bir şekilde, bu olguların önemli bir kısmı akral ya da segmental varyantlardır. Biz de burada bildiğimiz kadarıyla, literatürdeki ilk plantar keratoderma ile prezente olan akral PLK olgusunu sunuyoruz.

Anahtar Kelimeler: Pitriyazis likenoides; keratoderma, palmoplantar

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First described by Neisser and Jadassohn in 1894, pityriasis lichenoides (PL) is a spectrum of uncommon papulosquamous dermatoses with unknown etiology.^{1,2} PL encompasses a clinical spectrum between pityriasis lichenoides et varioliformis acuta (PLEVA) and pityriasis lichenoides chronica (PLC). Also known as Mucha-Habermann disease, PLEVA is characterized by an acute eruption of erythematous papules which soon evolve into polymorphic lesions such as vesicles, pustules, and necrotic ulcers. Leaving varioliform scars, PLEVA usually heals within weeks to months. On the other hand, PLC, which is the chronic form, distinguished by recurrent crops of lichenoid papules that may last even several years. The typical lesion of PLC is a small erythematous-brown papule

with adherent mica-like scale. As each lesion evolve independently, lesions in all stages of progression are found next to each other predominantly on trunk and extremities in both PLEVA and PLC.³⁻⁷ Here, we report an atypical presentation of PLC which was presented as plantar keratoderma.

CASE REPORT

A 51-year-old woman was admitted to our outpatient clinic with a several- months' history of pruritus on both palms and soles. There was no family history and past history of any other diseases or medication. Upon dermatological examination we observed mild erythema, multiple fissures and hyperkeratosis involving the entire plantar surfaces (Figure 1). Examination also revealed mild palmar erythema and palmar hyperlinearity (Figure 2). Laboratory studies including serum biochemical analysis, complete blood count with differential, urinalysis, chest radiography, abdominal ultrasound scan, hormonal studies, antinuclear antibody, and thyroid autoantibodies, fecal occult blood test, and specific tumor markers were within normal limits. Serological tests for human immunodeficiency virus and syphilis were negative. Skin scrapings for potassium hydroxide (KOH) examination did not reveal any fungal hyphae. All causes of acquired palmoplantar keratoderma were excluded on clinical and laboratory evaluation. Besides, histopathological examination of the plantar lesions demonstrated parakeratosis, orthokeratosis, irregular epidermal acanthosis, necrotic keratinocytes, mild lymphocytic exocytosis, focal erythrocyte extravasation and lymphohistiocytic mononuclear cell infiltrate in the upper dermis (Figures 3-5). Accordingly a diagnosis of PLC which was presented as plantar keratoderma was made and treatment with topical keratolytics and corticosteroids was started. Unfortunately, the patient lost to follow up.

DISCUSSION

According to distribution of the lesions, PL has been classified into three main types as diffuse, central and peripheral forms.⁸ In general, PLC has



FIGURE 1: Mild erythema, multiple fissures and hyperkeratosis on left plantar surface.

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

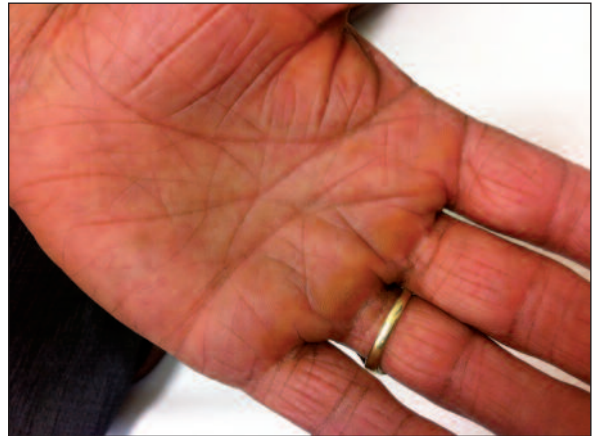


FIGURE 2: Mild erythema and palmar hyperlinearity on left palmar area.

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

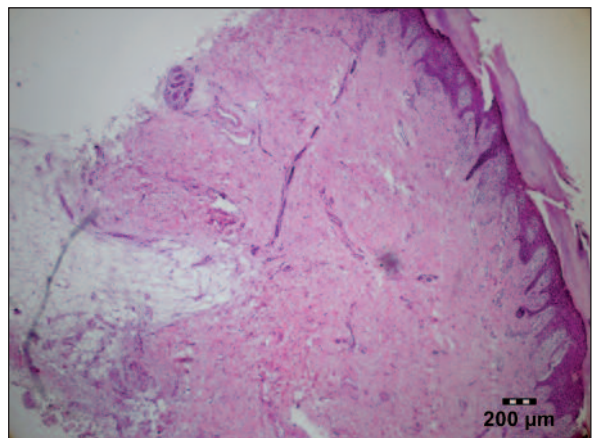


FIGURE 3: Prominent parakeratosis, irregular epidermal acanthosis, perivascular band-like inflammatory infiltrate in the upper dermis (H&E, x40).

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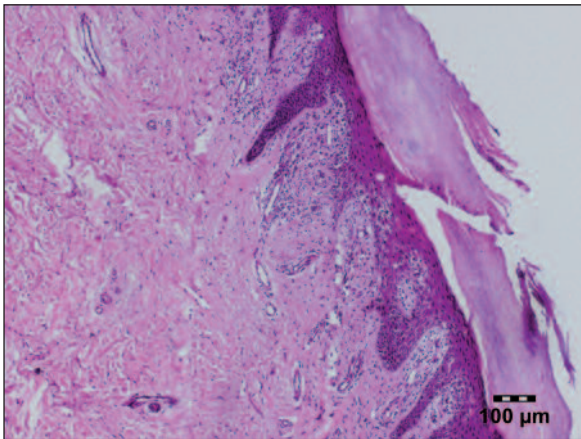


FIGURE 4: Prominent orthokeratosis, mild epidermal spongiosis, necrotic keratinocytes, irregular acanthosis, mononuclear cell infiltration in the upper dermis (H&E, x100).

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

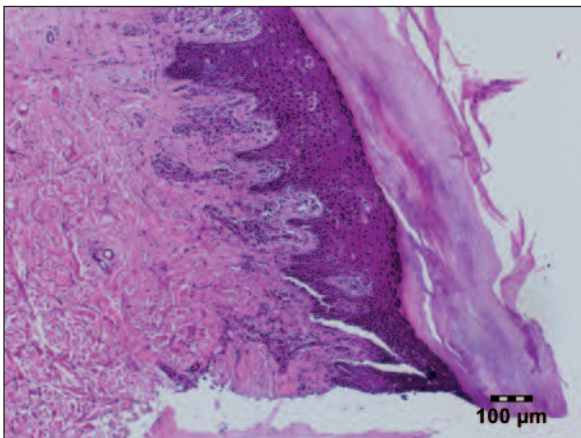


FIGURE 5: Prominent orthokeratosis, mild epidermal spongiosis, necrotic keratinocytes, mild lymphocytic exocytosis, irregular acanthosis, focal erythrocyte extravasation in the upper dermis and lymphohistiocytic mononuclear cell infiltrate (H&E, x200).

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a diffuse distribution affecting both trunk and proximal extremities.^{4,7-10} Pure acral involvement is occasional and has been described only in a few case reports up to date.¹¹⁻¹³ In 2002 *Kossard* reported two patients with PL, one of whom had polymorphic scaly papules over the dorsum of right foot, and the other had numerous recurrent keratotic papules restricted to bilateral lower distal extremities.¹² Of note, although the histopathological examinations of the lesional skin biopsies of both

patients were consistent with PL, one biopsy of the second patient demonstrated features of syringolymphoid hyperplasia. Since PL had been described as a T-cell premalignant disorder and syringolymphoid hyperplasia had been equated to syringotropic mycosis fungoides, thus both are associated with cutaneous T-cell lymphocytosis, *Kossard* has suggested that finding of syringolymphoid hyperplasia raises the suspicion of a relationship between acral PL and syringolymphoid hyperplasia.¹²

In 2010, Halbesleben et al. also reported a case of localized acral PLC with an asymptomatic eruption of sharply demarcated erythematous papules with overlying scale on the dorsal left foot and lower leg.¹³ Exhibiting typical clinical and histopathological features of PLC, the patient was a remarkable presentation of localized acral PLC.¹³ Besides acral localisation, palmoplantar involvement of PLC is extraordinary. *Chung et al.* described a case of PLC involving palmoplantar areas, mimicking palmoplantar syphilid.¹¹ As the eruption firstly developed on the palmoplantar areas and then spread to the distal parts of the extremities, the initial diagnosis was syphilis, however with serological and histopathological evaluation syphilis was excluded. The case was also unique with characteristic clinical and histopathological findings of PLC involving palmoplantar areas.¹¹

We assume that our case is an exceptional example of acral PLC. Despite the fact the pruriginous symmetrical hyperkeratotic plantar plaques have favoured a diagnosis of plantar keratoderma, histopathological features of the lesions were consistent with PLC. Contrary to our case, except the case mimicking palmoplantar syphilid, in all the reported cases clinical signs were correlated with histopathological findings supporting the diagnosis of PLC.¹¹⁻¹³ Indeed, instead of the characteristic copper-coloured syphilitic lesions, this patient demonstrated reddish brown papules covered by micaceous scales which is typical for PLC.^{11,14} Although syphilis is in the differential diagnosis, clinical picture is quite evident to make a diagnosis of PLC. On the other hand, on dermatological examination there is no doubt that our patient displays

features of plantar keratoderma. However, we could not explain the particular discrepancy between the nature of the dermatosis and localisation. Environmental and immunological factors might have been interacted with the genetic background

of the patient. Acral PLC is rare, moreover acral PLC affecting palmoplantar areas is more unusual and cases reported in the literature is so limited. To our knowledge, we report the first case of plantar PLC presenting as plantar keratoderma.

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