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Serkan Demirkan,
Özgür Gündüz,
Tuba Devrim*

Lichen Planus Pigmentosus-Inversus: A Report of Two Cases

Liken Planus Pigmentosus-İnversus: İki Olgu Sunumu

Abstract

Lichen planus pigmentosus (LPP)-inversus has been identified in a few East European cases primarily intertriginous areas such as axilla and groin which is not contacting with sunlight, and involvement in other skin areas was rarely detected. Because the lesions are similar to LPP in clinical and histological appearance, this condition, which is characterized by hyperpigmented, brown macules or plaques, is called LPP-inversus. Until today, actinic LP, linear LP, zosteriform, and LP pigmentosus clinical subtypes of lichen planus have been described. LPP-inversus is a rare form of LP and so far, there have been few case reports. LPP-inversus has been reported mostly in Caucasians and Asians, and axilla was the most involved body region. In a small proportion (approximately 10%) of the cases, classical LP or LPP lesions may be located outside the flexural areas. So far, about 50 cases have been reported; also we presented two new cases that we diagnosed.

Keywords: Lichen planus, inversus, pigment

Öz

Liken planus pigmentosus (LPP)-inversus Doğu Avrupalı birkaç olguda primer olarak aksilla ve kasık gibi güneş görmeyen intertriginöz alanlarda tanımlanmış olup, diğer deri bölgelerinde tutulum ise nadiren saptanmıştır. Lezyonlar klinik ve histolojik görünüm olarak LPP'ye benzediğinden dolayı, hiperpigmente, kahverengi makül veya plaklarla karakterize bu durum LPP-inversus olarak adlandırılmıştır. Liken planusun günümüze kadar aktinik LP, lineer LP, zosteriform, LP pigmentosus klinik alt tipleri tanımlanmıştır. LPP-inversus; LP'nin nadir bir formudur ve çok az vaka bildirimi olmuştur. Beyaz ırk ve Asyalılarda en fazla bildirilmiş olup aksilla olgularda en fazla tutulan bölge olmuştur. Olguların az bir kısmında (yaklaşık %10) klasik LP veya LPP lezyonları fleksural alanların dışında bulunabilmektedir. Şu ana kadar yaklaşık 50 olgu bildirimi olmuştur; biz de tanı koyduğumuz iki yeni olguyu sunmayı uygun bulduk.

Anahtar kelimeler: Liken planus, inversus, pigment

Kırıkkale University Faculty of Medicine, Department of Dermatology, Kırıkkale, Turkey

*Kırıkkale University Faculty of Medicine, Department of Medical Pathology, Kırıkkale, Turkey

Correspondence/ Yazışma Adresi:
Serkan Demirkan, Kırıkkale University Faculty of Medicine, Department of Dermatology, Kırıkkale, Turkey
E-mail: serkan.demirkan@yahoo.com.tr
ORCID-ID: orcid.org/0000-0002-3960-3891
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Introduction

Lichen planus (LP) inversus was primarily diagnozed by Pock et al., (1) on sunless intertriginous areas such as axilla and inguen in some East European patients, and its occurance was rarely detected on the other areas of skin. Since lesions were similar to LP pigmentosus (LPP) in clinical and histological a small portion of the cases (approximately 10%), classical LP or LPP lesions could be available out of flexural areas (1). Because the lesions are similar to LPP in clinical and histological appearance, this condition, which is characterized by hyperpigmented, brown macules or plaques, is called LPP inversus. Actinic LP, linear LP, zosteriform and LPP clinical subtypes of LP have been described. LPP-inversus is a rare form of LP and there have been few case reports (1,2). In a small number of cases (approximately 10%), LPP lesions may be located outside the flexural areas (3). Pock et al., (1) reported in their first statements

that large lesions had a tendency of linear or angular configuration (3,4).

This condition is generally asymptomatic but there may be occasionally a mild pruritus. There is no occurance on mucosa, hairy skin and nails. While some cases can regress without any treatment within a few weeks, some cases can continue for years (1,3). Approximately 50 cases have been reported to date and we present two new cases of LPP-inversus.

Case 1

A 63-year-old man was admitted with 2 months history of lesions on his body. The patient had no treatment, no known disease and no medication. There were macules and plaques on the lateral body sides, back and arm median faces, some with annular shape, some with erythematous, some with hyperpigmented, 1-5 cm in diameter (Figure 1). There were no pathological findings in the mucous membranes, scalp and nails. Dermoscopic examination revealed diffuse brown patches (Figure 2). Skin biopsy revealed interstitial lymphocyte in superficial dermis, melanophages in superficial dermis, infrequent lymphocytes in deep dermis and focal lymphocytic infiltration near skin attachments (Figure 3). The patient did not benefit from topical steroid and tacrolimus treatments.

Case 2

A 48-year-old woman presented with a 6-month history of discoloration in the bilateral inframamarian regions. She had antifungal and antibiotic treatments and had no benefit from these treatments. There were hyperpigmented macules with a diameter of 1-4 cm in the bilateral inframamarian regions (Figure 4). There was no finding of wood light examination. No mycotic element was detected in direct mycotic examination with potassium hydroxide. There were no pathological findings in the mucous membranes, scalp and nails. Dermoscopic examination revealed diffuse brown patches (Figure 5). Histopathological examination revealed interstitial lymphocyte infiltration rare melanophages in superficial dermis (Figure 6). The patient did not benefit from topical steroids and tacrolimus treatments.

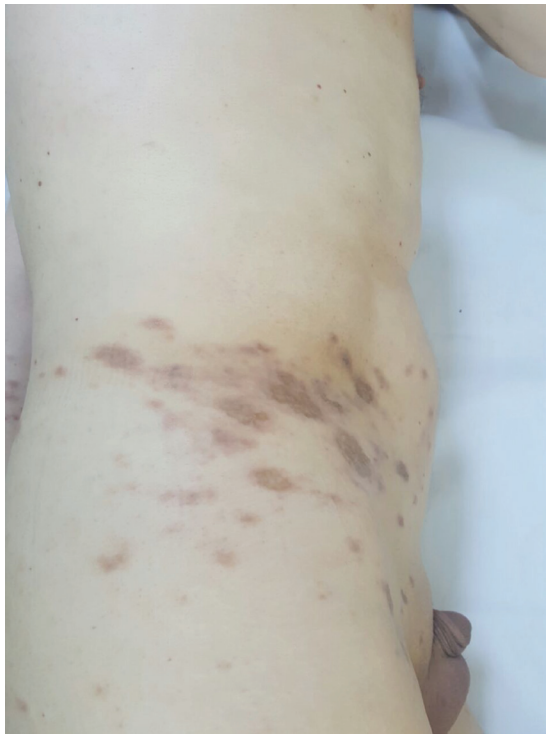

Figure 1. Macules and plaques on the lateral body sides, back and arm median faces, some with annular shape, some with erythematous, some with hyperpigmented, 1-5 cm in diameter

Discussion

Uyar and Sivrikoz (5) reported that 8.9% of 161 patients having LPP had it on their axilla, 6.5% of them had it on friction areas of their and 3.2% of them had it on inguinal areaswith a frequent impact on popliteal area. In 2001, Pock et al., (1) namedthis version of LPP which histopathologically had the characteristics of LPP and specifically occured on the intertriginous areas of skin as LPP-inversus. Together with our

Figure 2. Diffuse brown patches dermoscopic examination

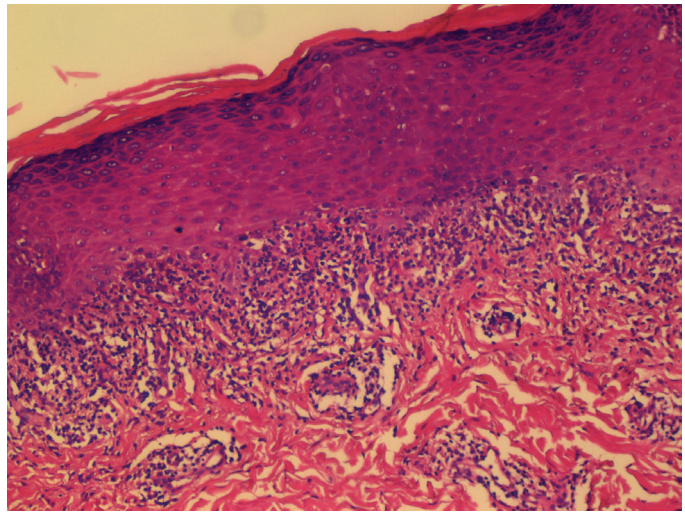

Figure 3. Interstitial lymphocyte in superficial dermis, melanophages in superficial dermis, infrequent lymphocytes in deep dermis and focal lymphocytic infiltration near skin attachments (H and E, ×100)

cases, 11 cases from Turkey and 52 cases from the world have been reported so far. 14 of these patients (26.9%) are male while 28 of them (73.1%) are female (5-9). It can be inferred from this information that it is much more frequent among females ($p<0.001$).

LPP-inversus is characterized by well limited macules which may be a little symptomatic and change colour from purple to brown by clinically affecting intertriginous areas. Some cases which are similar to our case occuring with lesions in annular plaques have been reported in literature so far. It is also reported that it occurs on external auditory canal and antitragus (3,4,6,10).

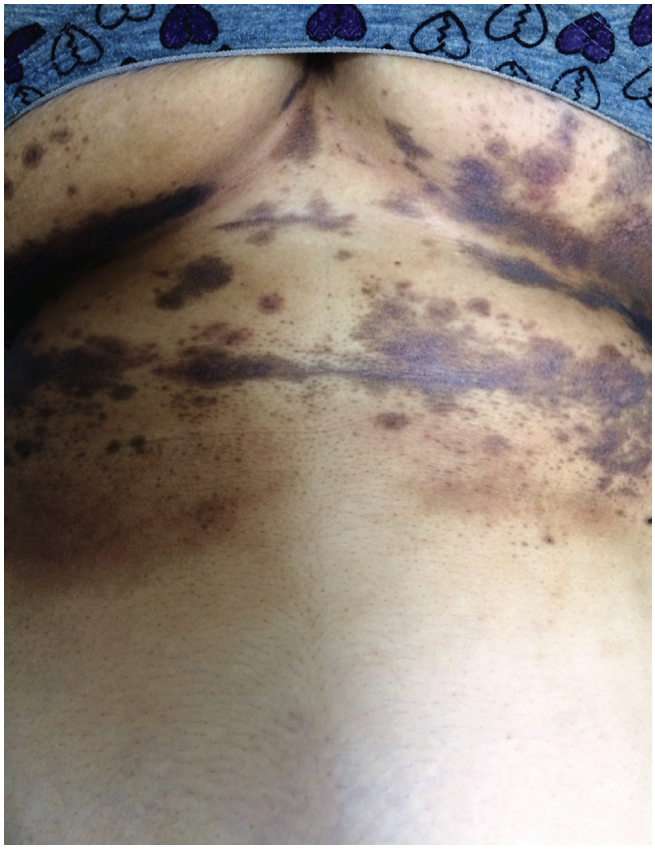

Figure 4. Hyperpigmented macules with a diameter of 1-4 cm in the bilateral inframamarian regions

Figure 5. Diffuse brown patches dermoscopic examination

Very few patients with LPP-inversus also have typical PL plaques on areas which are not intertriginous as in our first case. While LPP lesions generally occur on sun-exposed areas, LPP-inversus lesions generally occur on sunless areas as in the case of our patient. In patients with LPP-inversus, there is no occurence on mucosas, hairy skin and nails as in our case. There is also no relationship determined between LPP-inversus and viral hepatitis, infections, drugs or neoplastic illnesses. Since it occurs on intertriginous areasand observed in 2 patients who gets dressed tightly,it is reported that friction can have an impact on the occurence of the lesions (11,12).

In dermoscopy, it is reported that the appearance of multiple granular stains can be an indicator of poor prognosis, as well as the fact that lesions are long term and there can be no respose to treatment (7). But there were no findings without diffused brown patches in dermoscopy examination on our two cases. There was no findings like this without hyperpigmented macula in our two cases.

In histopathology, it is reported in literature that there can be both regressive type lichenoid reaction as in our cases and significant lichenoid reaction in biopsy specimens taken from the same patient. Lichenoid infiltration, which includes lymphocyte andhystiocyte in various dominance on ortokeratotic atrophic epidermis, is observed in the specimens. There are macrophages which include pigment incontinence and melanin on superficial dermis. While it is accepted that early-onset intensive lichenoid reaction leads to epidermoid atrophy and significant pigment incontinence, there is no epidermoid hyperplasia observed as it is available in LP. Pigmentation which is observed on deeper dermis, contrary to ashy dermatosis, and which leads to a change of bluish-gray colour through Tyndall impact is available on superficial dermiş (1,13,14). Skin biopsy showed lymphocytic infiltrate, macrophages that containes melanin, and pigment incontinence especially

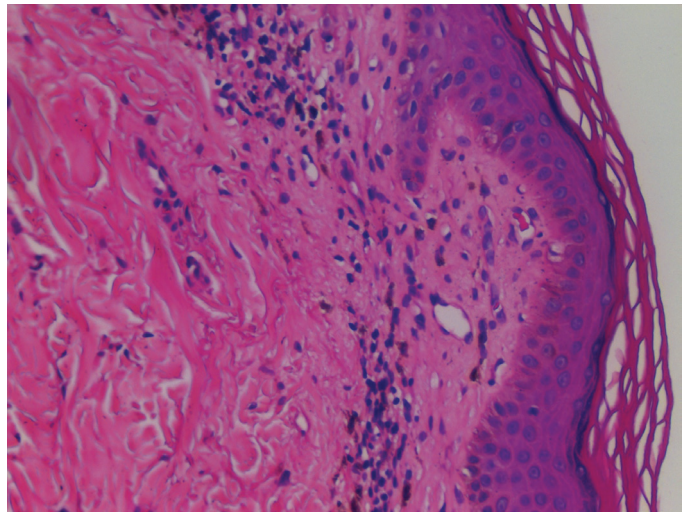

Figure 6. Interstitial lymphocyte infiltration rare melanophages in superficial dermis (H and E, ×400)

in the superficial dermis in our cases. Accumulation of immunoglobulin and complement which is observed in classical LP and determined through immunofluorescent staining has not been reported in literature. In our case, accumulation of immunoglobulin and complement through immunofluorescent has been researched but there is no accumulation determined too.

A sum of molecular pathogenesis of LP has not been understood yet. It is thought that T-lymphocyte derived cytotoxic activity plays a role against basic keratinocytes in LPP-inversus as in classical LP (15).

What comes to mind in definitive diagnosis of LPP inversus is Dowling Degos, acanthosis nigricans, erythema dyschromicum perstans, Riehl melanosis, cutaneous drug reactions, actinic LP, candidal intertrigo, erythrasma, postinflammatory hyperpigmentation, lichenoid toxicodermatitis. These illnesses can clinically and histopathologically be distinguished from LPP-inversus.

Calcineurin inhibitors and potent topical corticosteroids have been tried but generally failed as in our two cases (16). Spontaneous remissions have been reported but there is no proved effective treatment for LPP-inversus.

Conclusion

In conclusion, the best strategy in the management of this condition is to "wait and watch".

Ethics

Informed Consent: Consent form was filled out by all participants.

Peer-review: Internally peer-reviewed.

Authorship Contributions

Concept: S.D., Ö.G., Design: S.D., Data Collection or Processing: S.D., Analysis or Interpretation: S.D., Ö.G., Literature Search: S.D., Ö.G., Writing: S.D.

Conflict of Interest: No conflict of interest was declared by the authors.

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References

1. Pock L, Jelínková L, Drlík L et al. Lichen planus pigmentosus-inversus. J Eur Acad Dermatol Venereol 2001;15:452-4.
2. Cho S, Whang KK. Lichen planus pigmentosus presenting in zosteriform pattern. J Dermatol 1997;24:193-7.
3. Jung YJ, Lee YH, Lee SY, et al. A Case of Lichen Planus Pigmentosus-inversus in a Korean Patient. Ann Dermatol 2011;23:61-3.
4. Bennàssar A, Mas A, Julià M, Iranzo P, Ferrando J. Placas anulares en grandes pliegues: cuatrocasos de liquen plano pigmentoso-inverso. Actas Dermosifiliogr 2009;100:602-5.
5. Uyar B, Sivrikoz ON. A case of lichen planus pigmentosus-inversus. Türk Derm 2012;46:160-2.
6. Mohamed M, Korbi M, Hammedi F, et al. Lichen planus pigmentosus inversus: a series of 10 Tunisian patients. Int J Dermatol 2016;55:1088-91.
7. Murzaku EC, Bronsnick T, Rao BK. Axillary lichen planus pigmentosus-inversus: Dermoscopic clues of a rare entity. J Am Acad Dermatol 2014;71:e119-20.
8. Dizen-Namdar N, Kural E, Pulat O, et al. Lichen planus pigmentosus inversus: 5 Turkish cases. JEADV 2016;30:446-556.
9. Gaertner E, Elstein W. Lichen planus pigmentosus-inversus: Case report and review of an unusual entity. Dermatol Online J 2012;18:11.
10. Nijhawan RI, Borkin MS, Wilentz SE. Case Presentation Lichen planus pigmentosus-inversus involving the post-auricular sulci. Dermatology Online Journal 2013;19:14.
11. Majima Y, Hiroaki Yagi H, Ikeda Y, et al. Two cases of lichen planus pigmentosus inversus: possible causative role of tightly fitting underclothes. EJD 2013;23:904-5.
12. Ohshima N, Shirai A, Saito I, Asahina A. Lichen planus pigmentosus-inversus occurring extensively in multiple intertriginous areas. J Dermatol 2012;39:412-4.
13. Saray Y, Güleç T, Seçkin D. Lichen Planus Pigmentosus: Report of Four Cases. Türkiye Klinikleri J Dermatol 2004;14:222-6.
14. Barros HR, Almeida JRP, Dinato SLM, et al. Lichen planus pigmentosus inversus. An Bras Dermatol. 2013;88(6 Suppl 1):146-9.
15. Shai A, Halevy S: Lichen planus and lichen planus-like eruptions: patogenesis and associated diseases. Int J Dermatol 1992;31:379-84.
16. Al-Mutairi N, El-Khalawany M. Clinicopathological characteristics of lichen planus pigmentosus and its response to tacrolimus ointment: an open label, non-randomized, prospective study. J Eur Acad Dermatol Venereol 2010;24:535-40.