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Authors

Youh, Joohyung
Iwata, Hiroaki
Kitamura, Shinya
et al.

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Lichen planus pigmentosus inversus presenting with clinical features mimicking acanthosis nigricans

Joohyung Youh MD, Hiroaki Iwata MD PhD, Shinya Kitamura MD PhD, Hideyuki Ujiie MD PhD

Affiliations: Department of Dermatology, Faculty of Medicine and Graduate School of Medicine, Hokkaido University, Sapporo, Japan

Corresponding Author: Hiroaki Iwata MD PhD, Department of Dermatology, Faculty of Medicine and Graduate School of Medicine, Hokkaido University, North 15 West 7, Kita-ku, Sapporo 060-8638, Japan, Tel: 81-11-716-7087, Email: hiriwata0927@gmail.com

Abstract

Lichen planus pigmentosus (LPP) is recognized as a rare variant of lichen planus, characterized by dermal hyperpigmentation. Specifically, a particular intertriginous variant of LPP is known as lichen planus pigmentosus inversus (LPPI). In our case, the patient presented with symmetric, hyperpigmented dark brown patches mainly in axillary areas, closely resembling the features of acanthosis nigricans (AN). The differential diagnosis considered included LPPI, AN, post-inflammatory hyperpigmentation related to contact dermatitis, symmetrical drug-related intertriginous and flexural exanthema, fixed drug eruption, and erythema dyschromicum perstans (EDP). Histopathological examination revealed the absence of hyperkeratosis and papillomatosis, typically associated with AN. Dermoscopy revealed diffuse brownish hue along with dots and globules of inconsistent size, which suggests dermal pigmentary incontinence and the likelihood of LPPI. This case illustrates the challenge in differentiating LPPI from similar flexural hyperpigmentation disorders based on the comprehensive approach including thorough history taking, clinical manifestations, histopathological analysis, and dermoscopic examination.

Keywords: acanthosis nigricans, dermoscopy, hyperpigmentation, intertriginous area, lichen planus, pigmentosus inversus

Introduction

Lichen planus pigmentosus (LPP), acanthosis nigricans (AN), and erythema dyschromicum perstans (EDP) are cutaneous disorders characterized by acquired macular hyperpigmentation. Notably, lichen planus pigmentosus inversus (LPPI) is a specific variant of LPP that predominantly affects the flexural areas [1]. Clinically, LPPI predominantly manifests as dark brown hyperpigmented macules and patches, most observed in intertriginous and flexural regions, as well as in skin folds. According to Chen S et al., among three cases of LPPI, 67% of the patients exhibited neither itching nor any other symptoms, whereas 33% experienced mild pruritus [2]. Although the underlying mechanisms of LPPI remain largely unclear, it is believed that an inflammatory lichenoid response leads to significant pigmentary incontinence. Various researchers have suggested that factors such as the hepatitis C virus, sun exposure, and contact with substances like mustard oil and nickel may predispose individuals to this condition [3]. When diagnosing mildly itchy or non-itchy brown macules and patches, a range of conditions should be considered, including post-inflammatory hyperpigmentation related to contact dermatitis, symmetrical drug-related intertriginous and flexural exanthema, AN, fixed drug eruption, and EDP [4].

Case Synopsis

A 69-year-old woman with a history of poorly controlled type two diabetes mellitus (DM) and hyperlipidemia presented with a two-year history of dark brown hyperpigmented lesions in intertriginous regions. These lesions, characterized

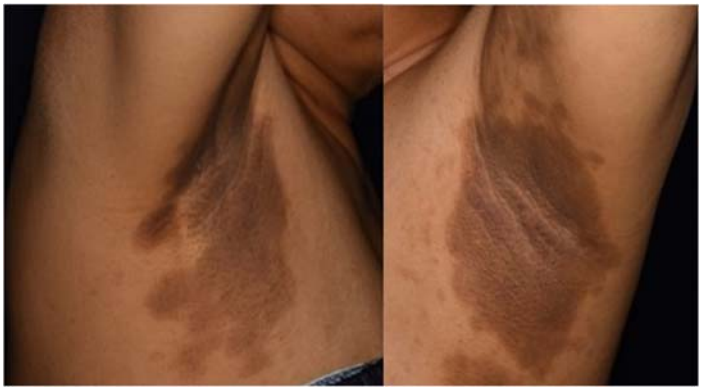

Figure 1. *Symmetrical, dark brown hyperpigmented lesions are seen in the bilateral axillae with a slightly velvety surface.*

by slightly velvety surfaces and dark hyperpigmentation, were symmetrically distributed on both sides of the axillae (**Figure 1**). There was no erosion or ulceration, and she reported no pain or itching in these affected regions. Additionally, linear hyperpigmented lesions were observed on her extremities. She had not received any dermatological treatments for these cutaneous lesions. Laboratory tests revealed an elevated HbA1c level of 8.5% and triglycerides at 208mg/dl. Thyroid hormone and adrenocorticotropic hormone levels were within normal ranges.

For histopathological assessment, a 4mm punch biopsy was performed on a hyperpigmented lesion in the axillary area. The biopsy showed epidermal atrophy and mild inflammatory infiltrates in the upper dermis. Notably absent were hyperkeratosis and papillomatosis, typically characteristic of AN. High-magnification microscopy revealed vacuolar alterations in the basement membrane zone and the presence of melanophages in the upper dermis.

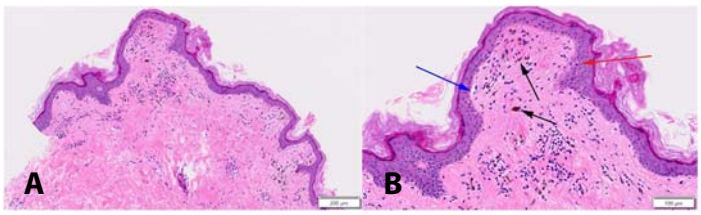

Figure 2. *H&E histopathology **A)** There was no significant hyperpigmentation of the basal cell layer. Lymphocytic infiltration, associated with vacuolar changes, and melanophages in the upper dermis were noted, 40×. **B)** Higher magnification reveals slight vacuolar changes at the basement membrane zone (blue arrow) and melanophages in the upper dermis (black arrows). A necrotic keratocyte is also noted (red arrow), 100×.*

There was no significant hyperpigmentation in the basal cell layer. In some areas, lymphocytic infiltration, associated with vacuolar changes, was noted (**Figure 2**). Dermoscopic examination of the hyperpigmented axillary lesions showed a structureless brownish area along with dark-brown dots and globules (**Figure 3**).

Considering the clinical presentation and histopathological findings, the condition was diagnosed as LPPI. Treatment commenced with the application of topical tacrolimus on the lesions on one side and a potent topical corticosteroid on the other side. Both treatments yielded slight improvements; however, the lesions treated with tacrolimus showed more significant improvement.

Case Discussion

In our case, because the patient had a history of poorly controlled type two DM, we initially considered the likelihood of AN, which has been reported to have a relationship with DM. In addition, we considered the likelihood of post-inflammatory hyperpigmentation related to contact dermatitis, fixed drug eruption and symmetrical drug-related intertriginous and flexural exanthema. However, the patient denied any events of contact dermatitis and there was no history of medication changes around the time the cutaneous lesion appeared.

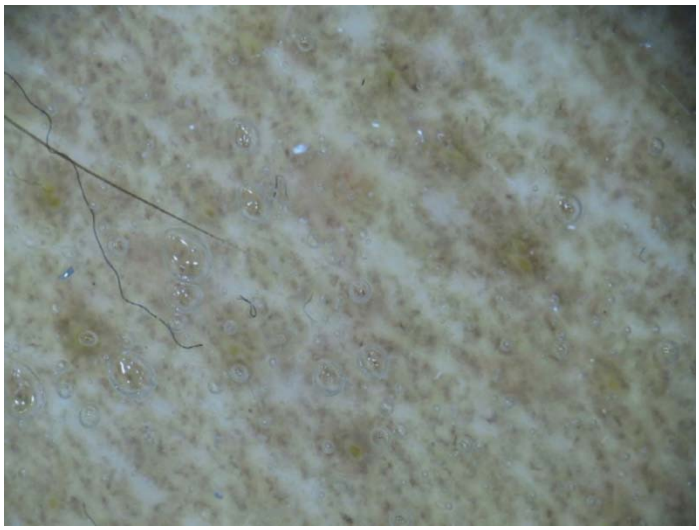

Figure 3. *Dermoscopy shows a diffuse, structureless, brownish area along with dark brown dots and globules which are inconsistent in size.*

Acanthosis nigricans is characterized by symmetric hyperpigmentation and coarse or velvety plaques in intertriginous sites, features that are also common in LPPI [5]. Despite the similarity in clinical manifestations, histological findings distinctly differ between the two diseases. In AN, common observations include papillomatous acanthosis, hyperkeratosis, and hyperpigmentation of the basal cell layer [6]. The histological characteristics of LPPI have been reported to include epidermal hyperplasia with orthokeratotic hyperkeratosis and hypergranulosis, necrotic keratinocytes, a band-like lymphocytic infiltration in the upper dermis, vacuolar changes in the basal layer, and melanophages in the dermis [7]. In the current case, although epidermal hyperplasia and vacuolar changes are less pronounced, necrotic keratinocytes, band-like lymphocytic inflammatory infiltrates, and pigment incontinence were noted. The presence of dermal melanophages and epidermal atrophy in this case led to the exclusion of AN.

Subsequently, we considered LPPI and EDP. In LPPI, histology may resemble that of EDP, characterized by the presence of dermal melanophages, either with or without interface dermatitis [8]. In EDP, interface dermatitis may be present but is not necessary for diagnosis. Differentiating between these conditions based solely on histopathologic findings is very challenging [8]. Given the histopathological similarities between LPPI and EDP, differentiation was based on clinical features. According to a report reviewing 68 cases of EDP in Korea, EDP typically affects the trunk (69.1%), neck (39.7%), upper extremities (38.2%), face (32.4%) and lower extremities (23.5%), [9]. However, in the above report and through literature searches on PubMed, there is only one case of EDP affecting the intertriginous regions. [4, 8, 9]. Clinically, the characteristic skin lesion of EDP is known as the eruption of multiple blue-gray macules or patches that sometimes have erythematous and slightly elevated borders. In contrast, the cutaneous lesion of LPPI is known for its brownish color [4, 9].

In the current case, dark brown hyperpigmented patches without elevated borders were observed, distributed primarily in the intertriginous regions, along with linear cutaneous lesions on the extremities. Based on these differences in clinical characteristics, including the distribution and color of the cutaneous lesions, EDP was excluded from the diagnosis.

Dermoscopy, traditionally used in diagnosing cutaneous neoplasms, is now increasingly recognized as a valuable tool for evaluating cutaneous inflammatory conditions [4]. Several reports have described the dermoscopic findings of LPPI [4, 10-12]. In both previous cases and in our current case, a diffuse, structureless, brownish area along with dark-brown dots and globules was observed. These patterns are thought to be related to dermal melanophages [11]. Errichetti E et al. suggest that EDP can be differentiated from LPP through dermoscopic characteristics such as a predominantly blue/gray hue and more consistent size and arrangement of dots. In LPP, the dots tend to be brownish and larger, whereas in EDP, the dots are smaller, gray-blue, and appear against a bluish background. The uniform gray color observed in gross and dermoscopic examinations of EDP is believed to relate to the deeper dermal pigment deposition. In contrast, the brownish and larger dots in LPP are considered to be related to the relatively superficial dermal pigmentary incontinence following the interface dermatitis [13]. Because LPPI is an intertriginous variant of LPP, it is believed that the dermoscopic findings of LPP could be used to differentiate LPPI from EDP. In the present case, brownish and relatively large, inconsistently-sized dots were observed in the dermoscopic examination; these findings also suggest the likelihood of LPPI.

Conclusion

We present a patient with LPPI with clinical features resembling those of AN. Although the cutaneous manifestations of LPPI and AN are very similar, the histopathological findings and the depths of the lesions are markedly different. This similarity in cutaneous manifestations posed a significant challenge, making it difficult for dermatologists to formulate a differential diagnosis through visual

inspection alone. In this case, the histopathological findings and the clinical features, including color and distribution of the cutaneous lesions, were instrumental in differentiating LPPI from AN and EDP. Additionally, the dermoscopic findings played a role as a valuable diagnostic tool in distinguishing between hyperpigmentation disorders in this case. This report underscores the importance of a comprehensive evaluation, encompassing thorough history taking, color and distribution of cutaneous lesions, histopathological analysis, and dermoscopic assessment, in the evaluation of intertriginous, hyperpigmented lesions.

Potential conflicts of interest

The authors declare no conflicts of interest.

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