

Lichen Planus Pigmentosus with Atypical Presentation- A Case Report

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Abstract

Lichen planus pigmentosus (LPP) is a chronic pigmentary disorder that shows diffuse or reticulated hyperpigmented, dark brown macules and patches on the sun-exposed areas such as the face, neck and other flexural folds. Clinically, it differs from classical lichen planus because LPP has a longer clinical course and manifests with dark brown macules. In case of LPP, involvement of the scalp, nail or mucosal area is rare. The histopathological findings of the lesions show an atrophic epidermis, the presence of melanophages and a vacuolar alteration of the basal cell layer with a sparse lymphohistiocytic lichenoid infiltration. Here we report a case of lichen planus pigmentosus over sun-protected areas sparing sun-exposed and flexural folds.

Key words: Lichen Planus Pigmentosus; Pigment Incontinence; Sun Protected Areas

Introduction

Lichen planus pigmentosus (LPP) was first described by Bhutani et al.¹ The lesions are small, brown to black, oval macules which can progress to form diffuse pigmented areas. The pigmentation may be diffuse, reticulate, blotchy, or perifollicular, usually symmetrical in distribution but may be found in a segmental, zosteriform, or blaschkoid pattern.²

Here we report a diffuse Lichen planus pigmentosus presenting over photo-protected sites sparing sun-exposed and flexural folds.

Case Report

A 21-year-old male, server at a restaurant, hailing from Assam, presented to the dermatology OPD with complaints of asymptomatic black discoloration mainly over the back, neck, arms, and thighs for 4 months.

Patient first noticed a brown to black asymptomatic lesion over the upper left scapular region, which gradually spread to involve the entire back, abdomen, bilateral shoulders, lateral aspect of the upper arm, buttocks, thighs, and lower legs. There was no history of preceding erythema or erythematous borders over the lesions, which is generally seen in Erythema Dyschronicum Perstans.

There was a history of occasional application of

mustard oil all over the body in the night. Patient denies any history of drug intake before the development of lesions; also no long duration of sun exposure with outdoor activities as the lesions were mainly present over the sun-protected areas sparing the sun-exposed sites and flexural folds.

On examination, multiple ill-defined diffuse hyperpigmentation tending towards symmetry over the back, bilateral shoulders, arms, buttocks, thighs and knees was seen (Figure 1a-f).

There was complete sparing of face, neck, intertriginous areas, forearm, legs, hands, feet, nails, oral, and genital mucosa.

Histopathological examination shows sparse superficial perivascular lymphocytic infiltrate with numerous melanophages within the papillary dermis. The papillary dermis is slightly thickened and shows delicate fibroplasia and mucin. The overlying epidermis shows a focal vacuolar change in the basal layer and infiltration of the interface by lymphocytes. The epidermis is flattened in places. The findings were suggestive of Lichen planus pigmentosus (Figure 2a, b).

Date of Submission: 4th May 2023

Date of Acceptance: 27th July 2023

Date of Publication: 1st October 2023

How to cite this article

Patankar A, Mane AA, Morgaonkar M. Lichen Planus Pigmentosus with Atypical Presentation-A Case Report. NJDVL 2023;21(2):28-31. <https://doi.org/10.3126/njdv1.v21i2.54637>



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Funding: None

Conflict of Interest: None

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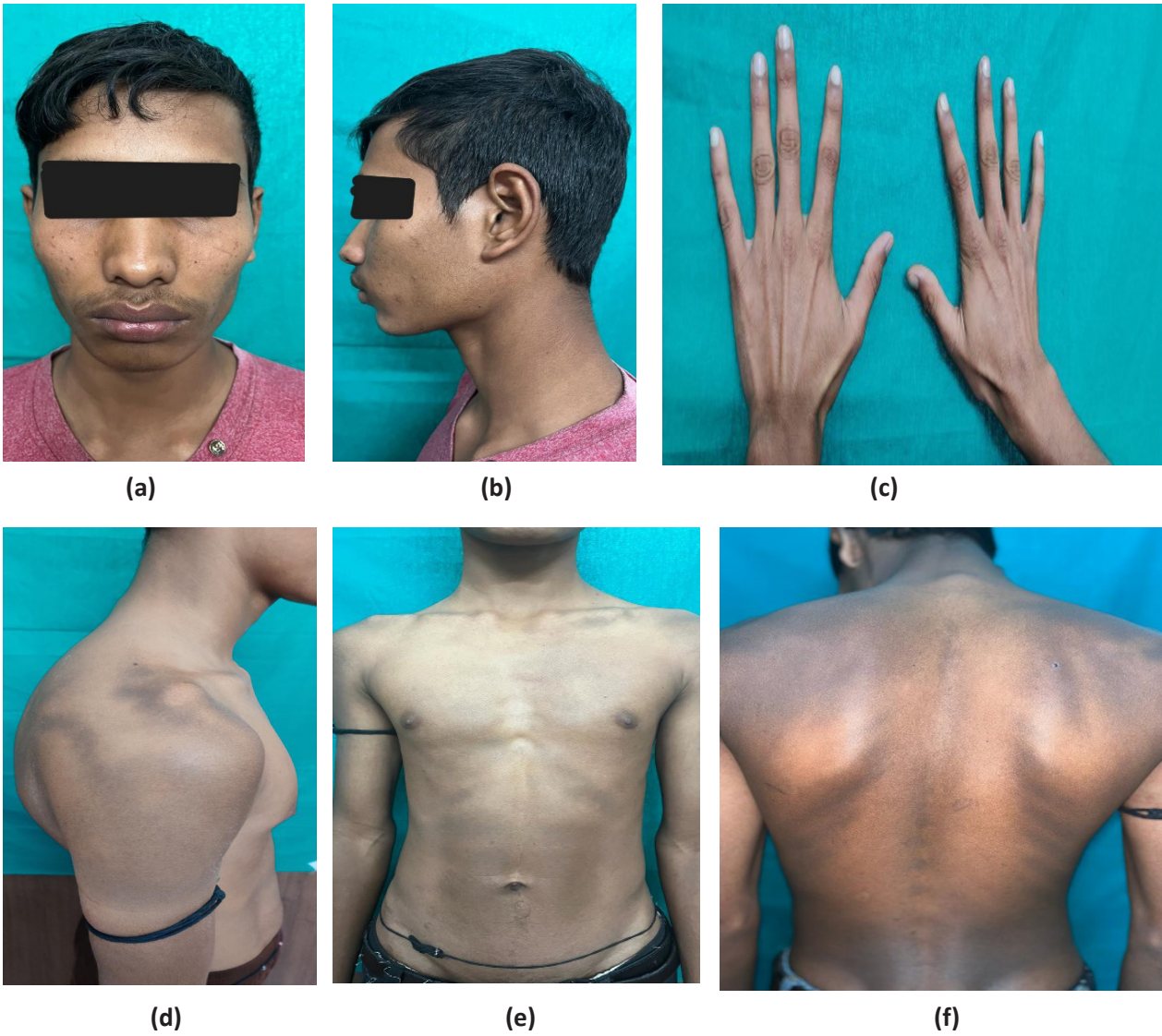


Figure 1 (a, b, c): Absence of lesions over face, neck, and hands

Figure 1 (d, e, f): Diffuse hyperpigmentation over arms, abdomen, and back

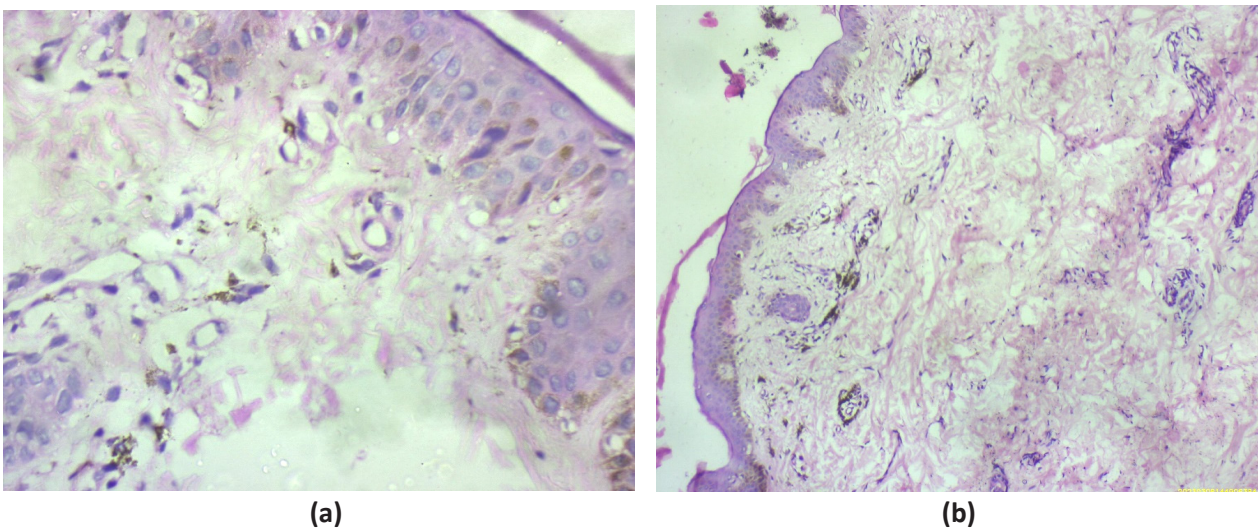


Figure 2 (a): Histopathological examination showing thinning of the epidermis, a superficial band-like lymphocytic infiltrate (40x, H and E stain)

Figure 2 (b): Basal layer vacuolization, lymphocytic infiltration and pigmentation incontinence in superficial dermis (100x, H and E stain)

Discussion

Lichen planus pigmentosus (LPP) is a chronic inflammatory pigmentary disorder essentially seen in adults after the age of 30. It occurs in both sexes but shows a female preponderance. It has been reported to occur predominantly in people with darker skin.¹ Several agents have been reported to act as predisposing factors. The occurrence in exposed areas in many patients has led to the proposition that sunlight may be a principal etiological agent. Another is mustard oil, which contains the potential photosensitizer allyl thiocyanate, and amla oil, where photosensitivity may be caused by fragrances, cosmetic agents such as kumkum, hair dyes, etc. Abnormalities in T lymphocyte functions have also been implicated.²⁻⁶ Morphological variants previously described include diffuse, reticular, blotchy, perifollicular, segmental, zosteriform and linear.⁷⁻¹¹ Another variant is LPP-

inversus, described in 2001 by Pock et al., defining it as a variant of LPP limited to intertriginous and flexural regions, sparing sun-exposed areas.^{12,13} Dermoscopic findings show discrete brownish to bluish-grey dots, globules, blotches, rods and white lines against a diffuse brownish background. Brown indicates epidermal pigmentation, while the grey and blue dots indicate melanin incontinence in the papillary and reticular dermis.^{14,15} The histologic findings of LPP show hyperkeratosis and atrophy of the epidermis with vacuolar degeneration of the basal layer. A perivascular lymphohistiocytic infiltration and pigmentary incontinence in the dermis are also noted.^{3,7,9} The most common differential diagnosis of LPP include idiopathic eruptive macular pigmentation, erythema dyschromicum perstans, Riehl’s melanosis, ochronosis, hori nevus, fixed drug reaction, and post inflammatory hyperpigmentation.^{16,17}

Table 1: Differential diagnosis of Lichen planus pigmentosus

Differential diagnosis	Comment
Idiopathic eruptive macular pigmentation	Occurs in younger patients. Brown to grey macules start in the middle area of the trunk and then spread to proximal areas of the limbs. Histopathology reveals pigmentation of the basal layer with mild perivascular inflammatory infiltrate. It is an epidermal hypermelanosis and does not show significant melanophages in the papillary dermis. ^{16,17,18}
Ashy dermatosis/erythema dyschromicum perstans	EDP presents with blue-grey, regularly shaped, hyperpigmented macules compared with dark brown, irregularly shaped, and ill-defined hyperpigmented macules in LPP with erythematous raised active borders. Histopathological evaluation reveals superficial dermal melanin and melanophages in LPP, whereas EDP usually has deep dermal melanophages, giving rise to the characteristic brown-grey color in LPP and the bluish-grey hue in EDP. ^{16, 19,20}
Riehl’s melanosis/pigmented contact dermatitis	It is characterized by facial hyperpigmentation, most pronounced on the forehead and in the zygomatic and/or temporal region. A correlation with clinical history of contactants and positive patch testing is necessary. ^{16,17}
Ochronosis	History of hydroquinone use at high concentration for a prolonged period, most commonly on the face. Usually does not affect neck and flexural areas. ¹⁶
Hori nevus	Bluish-grey or dark brown 2–5 mm macules caused by dermal melanocytes affecting the cheeks, temples, or forehead. Biopsy confirms diagnosis. ¹⁶
Fixed drug reaction	Round, initially erythematous macules with history of medication intake prior to onset. ¹⁶
Post inflammatory pigmentation	Previous history of dermatosis which leaves pigmentation as it subsides. ¹⁶

Conclusion

This case exhibits LPP over atypical sites, including a wide region of grey-brown patches occurring on the

photo-protected sites sparing the sun-exposed and intertriginous areas showing characteristic histologic features of Lichen planus pigmentosus.

References

- Bhutani LK, Bedi TR, Pandhi RK, Nayak NC. Lichen planus pigmentosus. *Dermatologica* 1974;149:43–50. <https://doi.org/10.1159/000251470>
- Ghosh A, Coondoo A. Lichen planus pigmentosus: The controversial consensus. *Indian journal of dermatology*. 2016;61(5):482–6. <https://doi.org/10.4103/0019-5154.190108>
- Kanwar AJ, Dogra S, Handa S, Parsad D, Radotra BD. A study of 124 Indian patients with lichen planus pigmentosus. *Clin Exp Dermatol*. 2003;28:481–5. <https://doi.org/10.1046/j.1365-2230.2003.01367.x>
- Rieder E, Kaplan J, Kamino H, Sanchez M, Pomeranz MK. Lichen planus pigmentosus. *Dermatol Online J*. 2013;19(12):20713. PMID: 24365004
- Vachiramon V, Suchonwanit P, Thadanipon K. Bilateral linear lichen planus pigmentosus associated with Hepatitis C virus infection. *Case Rep Dermatol*. 2010;2(3):169–172. <https://doi.org/10.1159/000320775>
- Gupta D, Thappa DM. Dermatoses due to Indian cultural practices. *Indian J Dermatol*. 2015;60(1):3–12. <https://doi.org/10.4103/0019-5154.147778>
- 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(5): 535–40. <https://doi.org/10.1111/j.1468-3083.2009.03460.x>
- Kumar YH, Babu AR. Segmental lichen planus pigmentosus: An unusual presentation. *Indian Dermatol Online J*. 2014;5(2):157–9. <https://doi.org/10.4103/2229-5178.131087>
- Cho S, Whang KK. Lichen planus pigmentosus presenting in zosteriform pattern. *J Dermatol*. 1997;24(3):193–7. <https://doi.org/10.1111/j.1346-8138.1997.tb02771.x>
- Hong S, Shin JH, Kang HY. Two cases of lichen planus pigmentosus presenting with a linear pattern. *J Korean Med Sci*. 2004;19(1):152–4. <https://doi.org/10.3346/jkms.2004.19.1.152>
- Vineet R, Sumit S, Garg VK, Nita K. Lichen planus pigmentosus in linear and zosteriform pattern along the lines of Blaschko. *Dermatol Online J*. 2015;21(10):13030/qt4rk2w3rm. PMID: 26632802
- Pock L, Jelínková L, Drlík L, Abrahámová S, Vojtechovská S, Sezemská D, et al. Lichen planus pigmentosus-inversus. *J Eur Acad Dermatol Venereol*. 2001;15(5): 452–4. <https://doi.org/10.1046/j.1468-3083.2001.00347.x>
- Robles-Méndez JC, Rizo-Frías P, Herz-Ruelas ME, Pandya AG, Ocampo Candiani J. Lichen planus pigmentosus and its variants: review and update. *Int J Dermatol*. 2018;57(5):505–14. <https://doi.org/10.1111/ijd.13806>
- Murzaku EC, Bronsnick T, Rao BK. Axillary lichen planus pigmentosus-inversus: dermoscopic clues of a rare entity. Diagnosis: lichen planus pigmentosus (LPP). *J Am Acad Dermatol*. 2014;71(4): e119–20. <https://doi.org/10.1016/j.jaad.2014.01.881>
- Sonthalia S, Errichetti E, Kaliyadan F, Jha AK, Lallas A. Dermoscopy of lichen planus pigmentosus in Indian patients—Pitfalls to avoid. *Indian J Dermatol Venereol Leprol*. 2018;84(3):311–13. https://doi.org/10.4103/ijdv.IJDVL_40_18
- Kumarasinghe SPW, Pandya A, Chandran V, Rodrigues M, Dlova NC, Kang HY, et al. A global consensus statement on ashy dermatosis, erythema dyschromicum perstans, lichen planus pigmentosus, idiopathic eruptive macular pigmentation, and Riehl's melanosis. *Int J Dermatol*. 2019;58(3):263–72. <https://doi.org/10.1111/ijd.14189>
- Robles-Méndez JC, Rizo-Frías P, Herz-Ruelas ME, Pandya AG, Ocampo Candiani J. Lichen planus pigmentosus and its variants: review and update. *Int J Dermatol*. 2018;57(5):505–14. <https://doi.org/10.1111/ijd.13806>
- Joshi R. Idiopathic eruptive macular pigmentation in an Indian male. *Indian Dermatol Online J*. 2018;9(1):64–5. https://doi.org/10.4103/idoj.IDOJ_298_17
- Leung N, Oliveira M, Selim MA, McKinley-Grant L, Lesesky E. Erythema dyschromicum perstans: a case report and systematic review of histologic presentation and treatment. *Int J Womens Dermatol*. 2018;4(4):216–22. <https://doi.org/10.1016/j.ijwd.2018.08.003>
- Rodrigues M, Pandya AG, Bekkenk M, Parsad D, Kumarasinghe SP. Current understanding of lichen planus pigmentosus, erythema dyschromicum perstans (ashy dermatosis), and idiopathic eruptive macular pigmentation. *Pigment Int*. 2019;6(1):4–8. https://doi.org/10.4103/Pigmentinternational.Pigmentinternational_18_18