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Case Report

A 17-year-old male presenting with multiple hyperpigmented macules over right side of chest in zosteriform pattern

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ABSTRACT

Lichen planus pigmentosus (LPP) is a variant of lichen planus. It is characterized by small black, brown macules, on sun exposed areas like face, neck, upper extremities and trunk. Lesions are insidious in onset with gradually progressive course. Few variants have been reported like LPP inversus, localized LPP, linear LPP, zosteriform LPP and LPP of oral mucosa etc. Here, we present a case of zosteriform LPP in a young adolescent male, which is a rare presentation.

1. Introduction

Lichen planus pigmentosus is a variant of lichen planus, first described by Bhutani et al. in 1974.[1] This disease of unknown etiology runs an insidious and prolonged course and is characterized by macules of dark brown color.[2] The hyperpigmentation is usually bilateral and symmetric on sun-exposed areas and flexural folds in a diffuse pattern.[3] The disease may involve the mucosa but spares the scalp and nails. A retrospective study of 124 patients with lichen planus pigmentosus found that nearly 90% had involvement of the face and neck, suggesting that sun exposure plays a role in the pathogenesis.[3] LPP along Blaschko's lines is also known as linear LPP if it affects the limbs or the face, or zosteriform LPP if the trunk is affected.[4] Only two cases of zosteriform LPP are reported in the past.[4,5]

2. Case History

A 17-year-old boy, presented with multiple dark colored macules, which were asymptomatic over right side of chest for the last 2 years. Lesions were initially smaller in size, which gradually progressed over a period of 2 years. There was no history of prior lesions like vesicle, papule, pustule etc. No history of any allergen exposure or drugs was elicited from the patient.

On examination, multiple hyperpigmented round to oval macules with irregular and poorly defined borders were present on lateral aspect of right side of chest involving T3-T6 dermatomal segment. Lesions were arranged in zosteriform pattern [Figures 1 and 2]. There was no overlying hypertrichosis. The hair, nails and mucosae were normal. On the basis of clinical examination, Becker nevus, lichen planus pigmentosus and progressive cribriform and zosteriform hyperpigmentation were kept as differential diagnosis. Routine investigations were normal. Hepatitis C serology was negative.

A punch biopsy with a 4 mm punch was taken from the lesion. Histopathology findings on hematoxylin and eosin (H&E) staining showed sparse superficial perivascular lymphocytic infiltrate with pigmentary incontinence and numerous melanophages within papillary dermis. Overlying epidermis showed focal vacuolar changes in the basal layer. These findings were consistent with lichen planus pigmentosus-active lesion [Figures 3 and 4].

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Patient was advised clobetasol propionate 0.03% cream for daily application, but subsequently he was lost to follow-up.

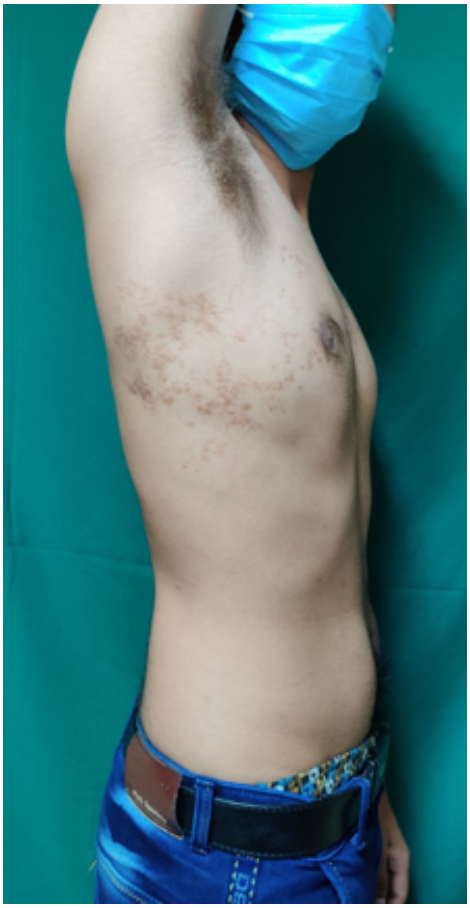

Fig. 1: Clinical photograph showing multiple ill-defined macules in a zosteriform distribution on T3-T6 thoracic region.

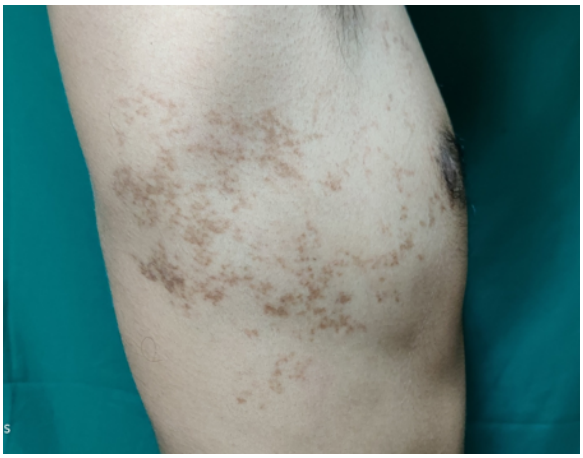

Fig. 2: Clinical photograph showing multiple ill-defined macules in a zosteriform distribution on T3-T6 thoracic region.

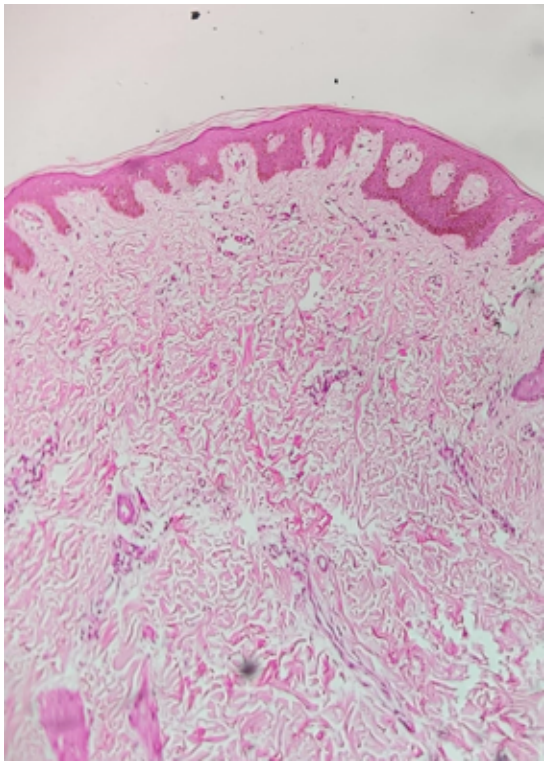

Fig. 3: Papillary dermis shows sparse superficial perivascular lymphocytic infiltrate with pigmentary incontinence and numerous melanophages. [H and E, 10x]

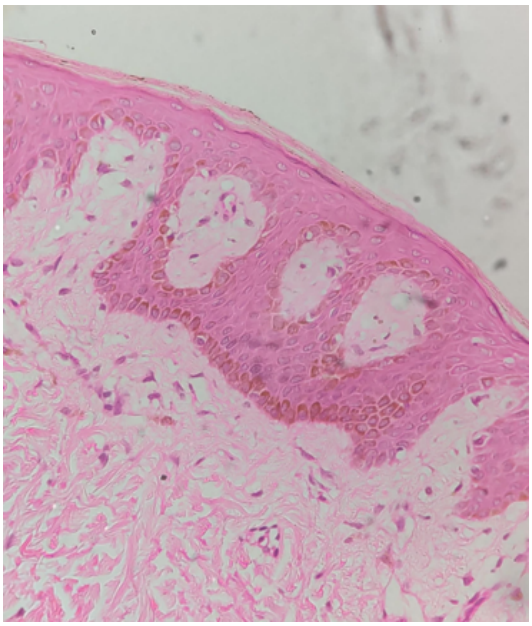

Fig. 4: Histopathology shows vacuolar degeneration of basement membrane along with hypermelanosis. [H and E, 40x]

3. Discussion

LPP usually affects patients with fitzpatrik skin type III to VI, with type IV being the most commonly affected. It has been commonly observed in India, Latin America, Asia, and Africa, and is rare in Caucasians. In the majority of patients, it presents between the 3rd and 5th decades of life, but it has been found in individuals 13–67 years of age.[6] It occurs in both sexes with slight female preponderance.[3] LPP is characterized by a symmetrical distribution of dark brown to gray or gray-blue, round or oval macules with poorly-defined borders, which gradually enlarge and coalesce to form larger lesions. The lesions are usually asymptomatic but present with mild itching in 27–62% of patients.[3] Morphological variants described in order of frequency from highest to lowest are diffuse, reticular, blotchy, perifollicular and annular, all of which may coexist in the same patient.[6]

It may be associated with other disorders such as hepatitis C virus induced liver disease, endocrinopathies, and autoimmune diseases as well as other variants of LP and its sequelae.[6]

In India, LPP has been linked to topical use and consumption of mustard oil containing a photosensitizer (allyl-thiocyanate), amla oil, henna, hair dye, cold cream, and environmental pollution.[6] Our patient did not give any history of application of any such oil or creams.

Zosteriform LPP is type of linear LPP present over trunk. It may be unilateral or bilateral, distributed along one or many Blaschko's lines.[7] Susceptibility to developing a variant of LPP may be explained during embryogenesis through mosaicisms, which determine cell populations with different immunological and antigenic properties.[4] It has also been associated with hepatitis C virus.

Pathogenesis is unknown, may be an altered cellular immune response mediated by T lymphocytes, in which CD8+ T lymphocytes recognize and attack epidermal keratinocytes, causing intense pigmentary incontinence. The lichenoid infiltrate is predominantly composed of CD8+ T lymphocytes, which are responsible for the cytotoxic response against keratinocytes.[6]

Histopathology characteristically shows vacuolar degeneration of the epidermal basal cell layer, band-like lichenoid or perivascular lymphocytic infiltrates in the papillary dermis, superficial pigmentary incontinence, melanophages along with less common findings such as hyperkeratosis and epidermal atrophy. The histopathology in our patient was suggestive of LPP.

LPP is usually recalcitrant to treatment. Topical treatment includes medium to high potency corticosteroids, tacrolimus and skin lightening creams, of that tacrolimus is most commonly used topical treatments. Systemic treatment includes corticosteroids in pulse doses or continuous administration with gradual tapering, as well as dapsone. Isotretinoin has been recently reported as a promising therapy with a relatively better side effect profile.[6] The disease does not respond well to existing treatment options.

4. Conclusion

We present a rare case of unilateral zosteriform lichen planus pigmentosus, involving T3-T6 thoracic dermatomal segment in a 17-year-boy.

5. Source of Funding

None.

6. Conflicts of Interest

There is no conflict of interest.

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