

Case Report

Unilateral Blaschkoid Lichen Planus - A Rare Presentation of a Common Dermatological Entity

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Abstract

Lichen planus (Greek leichen, "tree moss"; Latin planus, "flat") is a unique, immunologic mediated inflammatory disorder, that affects skin, mucous membranes, nail & hair. It is a disease of unknown etiology and is characterized by purple, polygonal, pruritic, flat topped papules.

Linear lesions in lichen planus can be due to isomorphic phenomenon or due to mosaicism, where the lesion follow Blaschko`s lines. The latter are unique lines which may be followed by other conditions like epidermal nevi, psoriasis, lichen striatus, Darrier`s disease and porokeratosis to name a few^{2,3}.

Here we report an interesting presentation of Blaschkoid lichen planus in association with atopy, in a 22 year old woman.

Key Words: Blaschko`s lines, Blaschkoid lichen planus, Unilateral lichen planus

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Introduction

Lichen planus (LP) was first described by Erasmus Wilson in 1869. It is characterized by purple, polygonal, pruritic, papular eruption of unknown etiology affecting skin, mucous membranes and the nails¹.

It can range from the generalized form to the much localized form. Less commonly, the skin lesion in lichen planus may have a linear arrangement. This may be due to trauma (Koebner phenomenon) or due to their tendency to follow the lines of Blaschko. The latter is known as Blaschkoid lichen planus, and is seen in only 0.5% of patients².

Case report

A 22 year old lady presented with complaints of itchy raised skin lesions over her left upper chest, upper arm and the second webspace, over the past two months. There was no history of preceding trauma at the site of the lesions. She gave a history of similar lesions when she was around 10 years of age. The lesions had first appeared over the medial aspect of her left upper arm and left side of chest and subsided two months after the onset. At present, she complained of recurrence of lesions at the same sites over the past two months [Fig 1,2,3]. She was completely asymptomatic between the two episodes.

Past History

There was past history of Varicella zoster, one year back but she recovered without any dermatological sequelae. She had a history of allergic rhinitis. There was no past history of jaundice or history of any dental procedures. She was not on any chronic medication.

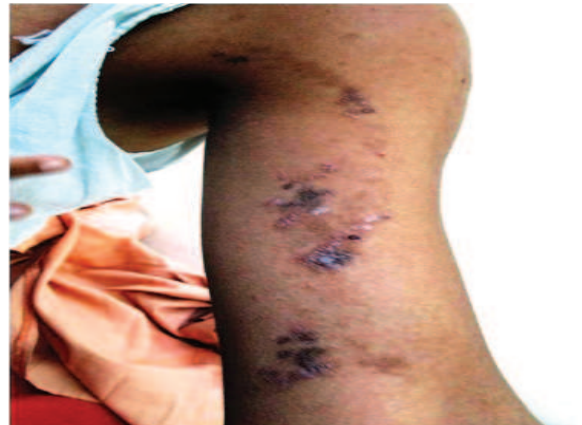


Fig 1 - Well defined multiple violaceous flat topped papules, reticulate plaques with minimal scales and hyperpigmented atrophic macules observed over the medial aspect of (L) upper arm. Intervening areas show normal skin.



Fig 2 - Similar multiple well defined violaceous flat topped papules with minimal scaling observed over the (L) upper chest over 2nd intercostal space



Fig 3 - Flat topped papules with minimal scaling observed over the interdigital space between the (L) index finger and middle finger.

With the above clinical findings, the following differential diagnoses were considered :-

- Linear Lichen Planus
- Lichen Striatus
- Inflammatory Linear Verrucous Epidermal Nevus (ILVEN)
- Blaschkoid Lichen Planus

Routine blood investigations, urine routine, liver and kidney function tests were within normal limits. Serology for HIV and VDRL were non-reactive. HBsAg and HCV were negative.

Four millimeter punch biopsy was taken from the papule on the medial aspect of left upper arm and sent for histopathology. It revealed hyperkeratotic epidermis with hypergranulosis, acanthosis and basal cell vacuolation with melanin incontinence. Dermis showed Civatte bodies with Max Joseph space. Dense band like lymphocytic infiltrate in papillary dermis with melanophages was noted (Fig 4).

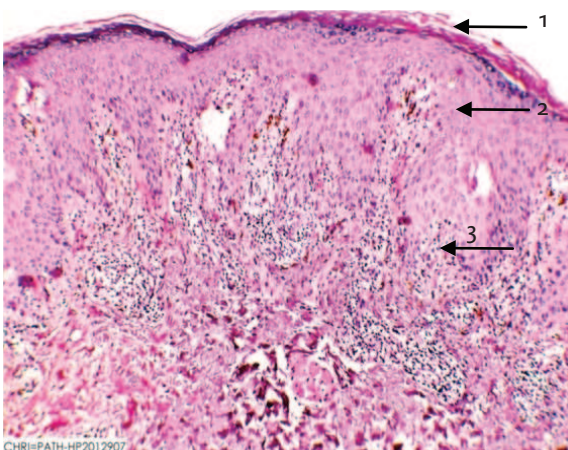


Fig 4 - Histopathology showing hyperkeratotic epidermis (1) hypergranulosis (2) acanthosis and basal cell vacuolation. Dense band like lymphocytic infiltrate (3) in papillary dermis with melanophages noted.

With the above clinical and histopathological correlation, the diagnosis of Unilateral Blaschkoid Lichen Planus was arrived at.

She was managed with Topical corticosteroid (0.1% Mometasone), oral antihistamines (T.hydroxyzine 10mg) and liquid paraffin for topical application for one month.

On follow up, the lesions regressed leaving a post inflammatory hyperpigmentation.

Discussion

The lines of Blaschko were first described and drawn by Alfred Blaschko, in 1901³. In contrast to dermatomes, these lines form a V-shape over the spine, an S-shape on the lateral and anterior aspect of the trunk, and an inverted U-shape from the breast area onto the upper arm. On the extremities, they follow a perpendicular direction and on the abdomen, they form whorls⁴.

The embryological basis of distribution pattern of these lines is so far an enigma². Blaschko lines represent a form of 'mosaicism', where two or more genetically distinct cell populations are present in an individual derived from a single zygote^{2,3}. Blaschko's lines do not correspond to any known nervous, vascular or lymphatic structures⁵. These lines represent distribution of autonomic motor-visceral afferents or stretching of the skin during embryogenesis. The lines of Blaschko may be followed by certain X-linked, congenital and inflammatory skin disorders^{6,7}.

The course of Blaschkoid LP is usually benign and self-limiting⁸. Dermatoses which may be confused with this type of lichen planus are those that present linearly or follow the lines of Blaschko like nevi, psoriasis, lichen striatus, Darrier`s disease and those dermatoses which have a dermatomal distribution like Herpes zoster^{9, 10}.

The features in favour of a true Blaschkoid lichen planus in this patient were the absence of preceding trauma or illnesses, characteristic clinical morphology and histopathology. Other interesting findings that were observed were the unilateral distribution of the lesions and the association with atopy.

Conclusion

This case of Blaschkoid lichen planus is reported for its uniqueness and rarity with regard to its unilateral distribution and its association with atopy.

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Tale of Two Cities

It was the best of times, it was the worst of times,
It was the age of wisdom, it was the age of foolishness,
It was the epoch of belief, it was the epoch of incredulity,
It was the season of light, it was the season of darkness,
It was the spring of hope, it was the winter of despair,
We had everything before us, we had nothing before us,
We were all going direct to Heaven, we were all going direct the other way-
In short, the period was so far like the present period.

- Charles Dickens