CASE REPORT

Lichen planus pigmentosus-inversus following Langer's lines of cleavage: a rare clinical presentation

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ABSTRACT
We present an interesting case of a patient who had a 5-month history of insidiously developing pigmented macules along skin cleavage lines over the skin of submammary and inguinal folds. There was no history of previous medication. Skin examination revealed multiple discrete and slate-grey macules distributed over the skin cleavage lines of the submammary and inguinal folds. These lesions did not coalesce, forming several, intermittent and brownish lines. Pathologically, it showed atrophic epidermis with foci of colloid bodies. A dense lymphocytic infiltration along with prominent melanophages was noticeable in the superficial dermis. The patient was diagnosed with lichen planus pigmentosus-inversus following skin cleavage lines.

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Introduction

Lichen planus pigmentosus-inversus (LPPI) is a rare variant of lichen planus characterized by mottled pigmentation of bluish, black, or grey color on the flexural folds.1 We describe a similar case of a patient whose lesions were distributed along Langer's lines of skin cleavage over the submammary and inguinal skin folds.

Case report

A 49-year-old Chinese female presented with a 5-month history of insidiously developing pigmented macules along skin cleavage lines over the submammary and inguinal skin folds. This slate-grey macule began on the submammary skin folds and then spread to the inguinal area without noticeable inflammatory changes. No drugs were taken prior to development of the eruption. She denied any infection of hepatitis B or C virus. There was no history of fever, weight loss, or exacerbation of the eruption upon sun exposure. On examination, multiple, discrete and slate-grey macules distributed along the skin cleavage lines over submammary and inguinal folds were observed. The chest, epigastric area, and upper thighs were less frequently affected. The macules were approximately 2 mm in diameter, and did not coalesce, forming several, intermittent and brownish lines. These lines obviously were unrelated to Blaschko’s lines and dermatome lines (Figure 1). The mucous and the nail were not affected. General systemic examination revealed no abnormalities. Routine laboratory tests including peripheral blood cells and liver function were all within normal limits. A biopsy specimen from the left inguinal skin lesion showed atrophic epidermis with foci of colloid bodies. In the superficial dermis, there was a dense lymphohistiocytic infiltration with prominent melanophages (Figure 2), which were in accordance with the pathological alterations in regressive lichen planus. Based on all these clinical and histological findings, the patient was diagnosed with LPPI following skin cleavage lines.

Discussion

LPPI is an uncommon type of lichen planus reported first by Pock et al1 in 2001. Subsequently, it was observed to occur in other racial and ethnic groups, and >27 cases have been reported in English literature to date.2 LPPI is most often seen in the 3rd decade to 5th decade of life. There is a slight female preponderance. The lesions seen in LPPI are mostly asymptomatic and occur most often over the axillae and groin. Other skin parts, such as the neck, abdomen, popliteal regions, inframammary folds, antecubital...
diameter, and did not coalesce, forming several, intermittent and brownish lines. However, intermittent lines over the skin cleavage, as shown in [Figure 1](#).

Langer's lines (or skin cleavage lines) are three major line forms involved. LPPI was reported to start insidiously as a small, ill-defined and oval to round macule, which coalesced to a confluent sheet of pigmentation. Its color varied from slate-blue to steel grey or brown.1,2 Occasionally, the macules were distributed in a linear, blaschkoid, reticular, blotchy, or perifollicular morphology.3,4

Histologic features that characterize LPPI vary according to its stage. A band-like infiltration of lymphocytes characterizes its earlier stage, with moderate melanophages scattered in the dermis. Additionally, there is notable vacuolar degeneration of the basal layer and colloid bodies in the epidermis. Meanwhile, the old lesion is accentuated with superficial, mild, lymphohistiocytic infiltration with frequent interstitial and perivascular melanophages. In the epidermis, it usually shows focal or absent vacuolar degeneration, few apoptotic keratinocytes, and atrophy of the epidermis with compact hyperkeratosis.4,5 The pathological findings in our case were overall in accordance with those in late-stage LPPI.

It is well known that dermatome lines, Blaschko's lines, and Langer's lines (or skin cleavage lines) are three major line forms described that cover the human body. Langer's lines were revealed in 1861 by Karl Langer (1819–1887). Langer's lines run along the natural orientation of collagen fibers and underlying muscle fibers, thus, they have a significant relevance especially to surgical techniques. In dermatology, several skin disorders have been reported to be distributed along these lines. Essentially, they are pityriasis rosea and mycosis fungoides, as well as stage two syphilis, exanthematic sarcoma idiopathicum multiplex, hemorrhagica Kaposi, creeping hair, the early stages of psoriasis vulgaris, Wolf's isotopic response, Leser-Trelat syndrome, Saurian papulosis, idiopathic eruptive macular pigmentation, and dermatosis papulosa nigra. 6-8 LPPI can be differentiated from these by its typical clinical and histological characteristics.

To date, there are no reasonable explanations for the successive distribution of a lesion over the skin cleavage lines. Some scholars believed that it might involve hematogenic dissemination of somehow activated leukocytes in its pathogenesis.6 Others suggested that Koebner's phenomenon may play a pivotal role in the pathogenesis of LPPI.9 We propose that stimulation of sweat, friction, a moist environment, or other external stimuli to the intertriginous area could be at least partially responsible. Accumulation of similar cases is necessary to further explore the profound characteristics of this rare presentation.

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**References**