PUNCTATE KERATOTIC ACRAL LICHEN PLANUS: AN UNUSUAL AND RARE VARIANT OF LICHEN PLANUS

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Summary

Introduction: *Acral lichen planus (ALP) is an uncommon variant of LP, that can sometimes be difficult to diagnose, due to atypical clinical and dermoscopy features and resistance to different therapeutical means. We report the case of a female patient diagnosed with punctate keratotic lichen planus.*

Case report: *A 49-year-old female patient presented to the Dermatology Clinic of Mures Clinical County Hospital for multiple, severely pruritic, flat and yellowih-coloured papules confined in hyperkeratotic plaques located bilaterally on the palms and soles, in evolution for three months. Clinical, dermoscopical, and histopathological examinations further established the diagnosis of punctate keratotic lichen planus. Combined treatment with systemic corticotherapy and anti-H1 antihistamines, topical heparine gel 500 UI/g, twice a day and narrow-band UVB phototherapy was started with significant improvement at two months follow-up.*

Conclusions: *Keratotic punctate ALP is a very rare and uncommon variant of LP. Patients with punctate keratotic LP might benefit from treatment with topical heparine.*

Keywords: *lichen planus, heparine, keratotic lichen planus.*

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Introduction

Lichen planus (LP) is an inflammatory disorder, with a heterogenous morphological pattern and that can affect patients' skin, mucosas, and nails. With a complex and incompletely elucidated ethiopathogeneis, LP may be due to and can lead to impaired quality of life for affected subjects. Acral LP (ALP) is an uncommon variant of LP, that can sometimes be difficult to diagnose, due to atypical clinical and dermoscopy features and resistance to different therapeutical means. Herein, we report the case of a female patient diagnosed with an even rarer variant of ALP, namely punctate keratotic lichen planus.

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Case report

We report the case of a 49-year-old female patient presenting to the Dermatology Clinic of Mures Clinical County Hospital with a papulo-squamous rash in evolution for 3 months. Various home remedies were previously used, with no result. Clinical examination revealed multiple, severely pruritic, flat, and yellowish-coloured papules confined in hyperkeratotic plaques located bilaterally on the palms and soles (as seen in Figure 1). Some of the lesions presented with a central, punctate crateriform depression (Figure 2). Itch severity was appreciated on the visual analogue scale at 8/10. Similar, scarce small-sized lesions were noted on the volar surface of the forearms. No mucous or nail lesions were identified.

Laboratory investigations identified leucocyturia (75 Leu/µl) with no other associated comorbidities. Dermoscopy examination identified an erythematous background, with yellow dots and thick scales. No Wickham striae were seen (Figure 3). A 6 mm diameter punch biopsy was performed from one of the lesions located on

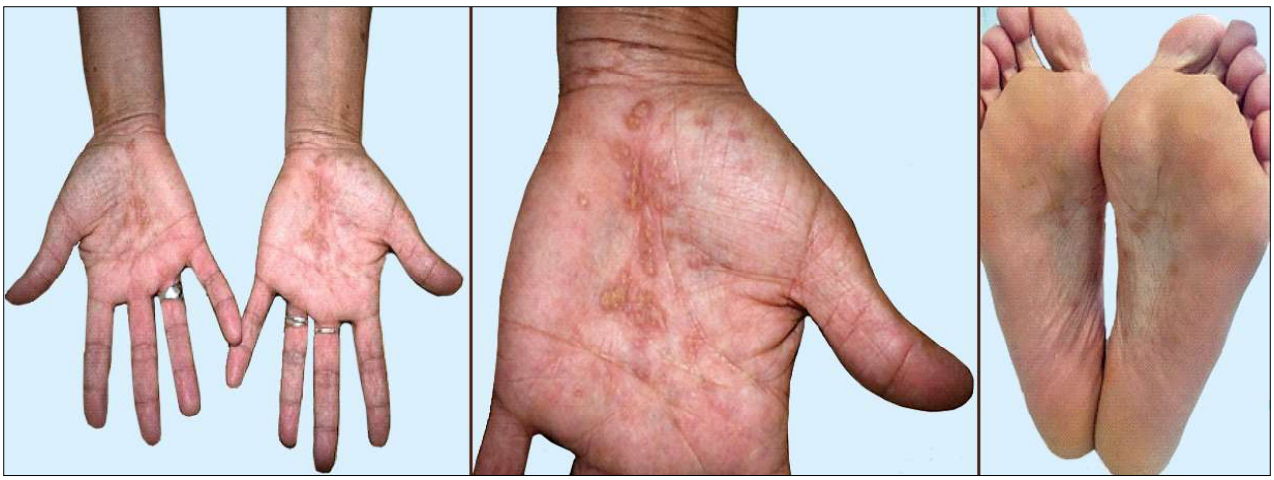


Figure 1. Clinical picture at diagnosis.

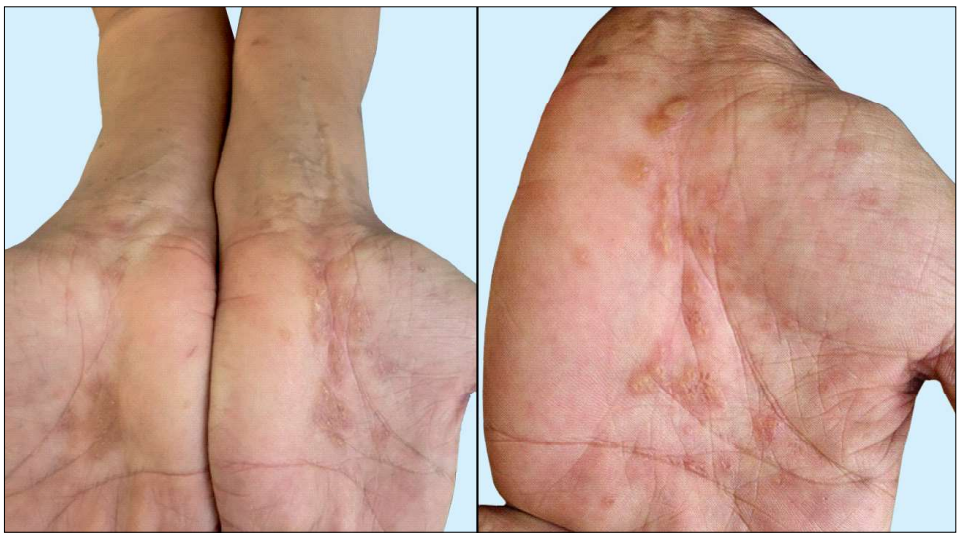


Figure 2. Central, punctate crateriform depressions of the lesions.

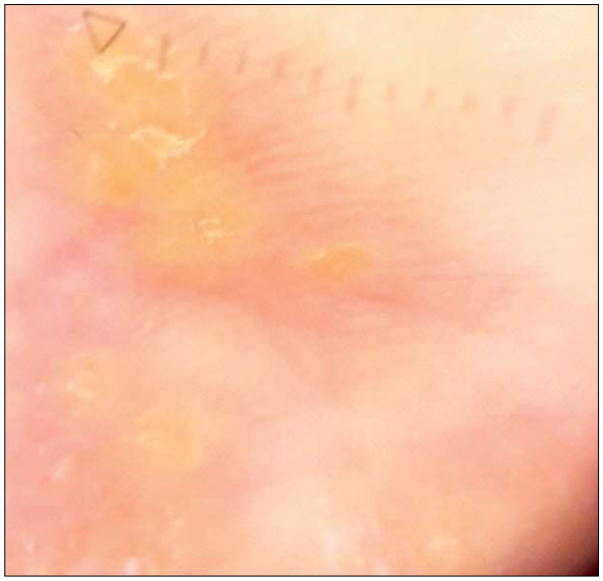
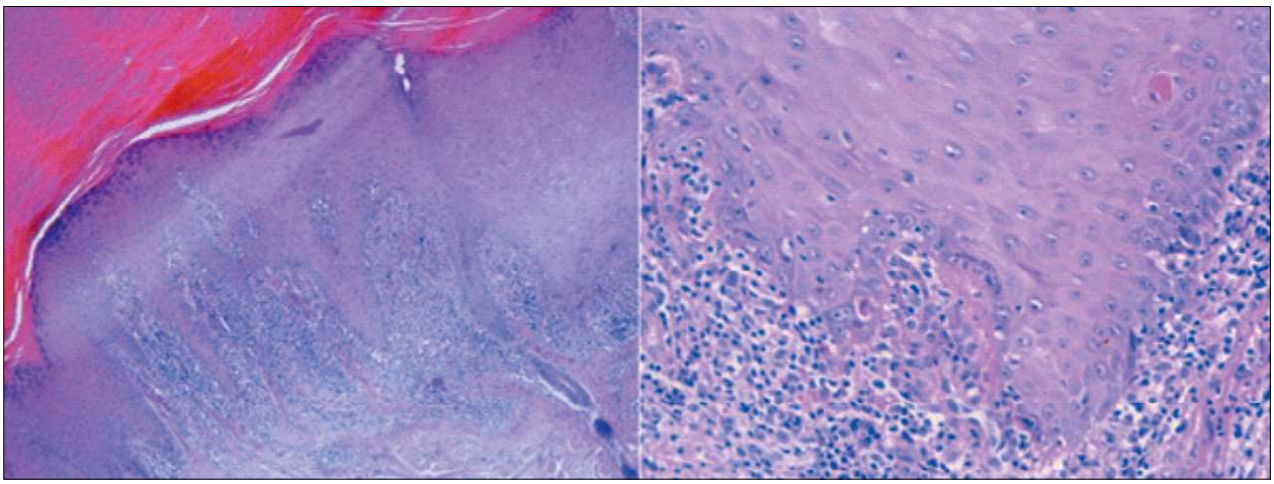


Figure 3. Dermoscopy image. Erythema, yellow dots and scales. No identified Wickham striae.



a **b**

Figure 4. Histopathological examination (5X HE, 10X). Orthokeratotic hyperkeratosis, hypergranulosis, band-like lymphocytic infiltrate.

the right palm with consequent histopathological examination. Hematoxilin-eosin stain examination (Figure 4) identified orthokeratotic hyperkeratosis, hypergranulosis, and epidermal hyperplasia with a saw-tooth appearance. A band-like lymphocytic infiltrate with macrophages, lymphocytes, and rare plasmocytes was seen in the superficial dermis, further establishing the diagnosis of punctate keratotic lichen planus.

Combined treatment with systemic corticotherapy (oral Prednisone, b.i.d 0.5 mg/kg for ten days with consequent tapering) and anti-H1 antihistamines (Chlorpheniramine 4 mg, once daily, at night), topical heparine gel 500 UI/g b.i.d, and narrow-band (nb) UVB phototherapy was started. Two-months check-up identified a marked regression of the palmoplantar lesions with a strongly alleviated itch (VAS=2), as seen in Figure 5.

Figure 5. Clinical picture at two months follow-up.

Discussions

Acral, palmoplantar lichen planus is an uncommon entity within the lichen planus spectrum. Five types of acral lichen planus were described, namely eczematous, punctate keratotic, ulcerated, lichenoid, and psoriasiform, the latter being the most common [1]. According to Sanchez-Perez et al. [2], ALP may be encountered in up to 26% of LP cases and seems to be defined by distinctive epidemiological, clinical, dermoscopical, histological, and therapeutical features.

Unlike other forms of LP, ALP mainly affects middle-aged males, between the third and fifth decade. Our patient does not fit the epidemiological data available for ALP. Typical polygonal violaceous papules, regarded as pathognomonic lesions of LP, are generally not seen in ALP. Moreover, in this particular form, the lesions mainly affect the lateral margins of the fingers and hand and seem to spare the fingertips [3,4]. However, in our case, the lesions were primarly located between the thenar palmar and the distal palmar crease.

Histologically, ALP presents with a thick stratum lucidum, which further reflects clinically in the absence of Wickham's striae [5,6]. As in our case, this may be due to impossible observation of the localized granular layer thickening due to the thickness of the horny layer.

Up to one third of ALP may become generalized, between one and four months from onset. Generalized, secondary lesions usually exhibit typical LP features, unlike the palmoplantar localization [5]. A proper follow-up of these patients proves therefore to be mandatory.

Differential diagnoses should take into account lichen nitidus, psoriasis, calluses, hyperkeratotic eczema, granuloma annulare, and secondary syphilis. In our case, the severe pruritus presented by the patient, negative serological investigations, and histopathological examinations were essential clues for the positive diagnosis.

Topical and systemic corticotherapy are the mainstay of treatment. Various agents, such as acitretin [7], methotrexate [8], cyclosporine. nb-UVB phototherapy, and psoralen and ultraviolet A therapy (PUVA) proved to be effective. Moreover, Yasar et. al [9] reported the successful treatment of ALP with 12 courses of subcutaneous injections of 3 mg enoxaparine. As for enoxaparine use in LP, it was first successfully administered by Hodak et. al [10] to 10 patients with generalized LP. Enoxaparine, a low-molecular-weight heparin, inhibits the activity of the heparanase released by the T cell lymphocytes. As such, the extracellular matrix is inhibited, as well as the production of pro-inflammatory cytokines, especially TNF-α [10], due to the disaccharide structure of the enoxaparine.

In our case, we hypothesized that topical heparin may exhibit the same therapeutical effects on ALP lesions as systemic enoxaparine. Therefore, the patient was started, as previously

mentioned, on topical heparine gel with subsequent diminishing in pruritus intensity and an obvious flattening of the lesions. Two-month follow-up identified the definite positive effect of the treatment of choice (systemic corticosteroids and topical heparine), and the patient is further carefully monitored in our clinic. To the best of our knowledge, this is the first case reporting the usefulness of topical heparine gel in treating LP.

Conclusions

Keratotic punctate ALP is a very rare and uncommon variant of LP. Defined by clinically and dermoscopically atypical lesions, it can present with a prolonged course of disease, possible generalization, and resistance to treatment. Patients with punctate keratotic LP should be carefully monitored, while topical heparine gel might be an useful therapeutical option for such cases.

Ethics statement: *Patients' rights were respected according to the Declaration of Helsinki. The patient gave her consent for the publication of her case, by obiding the GDPR cases. The reported case was not previosuly published, nor is it under consideration for publication in another journal.*

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Conflict of interest
NONE DECLARED

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