Case Reports in Dermatology

Case Rep Dermatol 2024;16:226–233
DOI: 10.1159/000541695
Received: June 26, 2024
Accepted: September 24, 2024
Published online: October 24, 2024

Case Series

Syringotropic Lichen Planus: An Unusual Presentation of a Common Dermatosis – A Report of 2 Cases

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Keywords
Lichen planus · Mycosis fungoides · Syringotropic lichen planus

Abstract
Introduction: Lichen planus (LP) is a chronic inflammatory dermatosis that causes plaques and itchy papules on the skin, as well as erosion and ulcers in the mucous membranes. LP is characterized by a dense dermal T-cell infiltration. Perieccrine inflammation can be seen in a variety of dermatoses, but genuine lymphocyte permeation of the secretory coil or lymphocytic syringotropism is a rare sign that is typically seen in mycosis fungoides. ***Case Presentation:*** In this study, we present 2 uncommon instances of lymphocytic syringotropism in LP. Histopathological examination revealed dense T-cell infiltration and lymphocytic involvement of eccrine glands, confirming syringotropism. ***Conclusion:*** Lymphocytic syringotropism is an uncommon finding in LP. Its presence broadens the histopathological spectrum of LP and highlights the need to differentiate it from lymphoproliferative disorders like mycosis fungoides.

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Introduction

Lichen planus (LP) is an inflammatory skin disease with no identified cause that mostly affects adults of any sex and ethnicity. Skin, mucous membranes, hair follicles, and nails are all negatively affected by LP [1, 2].

The "six P's" (planar, pruritic, plaques, purple, papules, and polygonal) are frequently used to characterize the appearance of cutaneous LP, which is usually seen as a papulosquamous eruption of different sizes [3].

LP is considered to be responsible for 0.4%–1.2% of all dermatology referrals and characteristics, with an estimated incidence of 1% [4]. To confirm the diagnosis, histopathological examination is useful [5]. CD8+ T cells play a significant part in the basal layer's destruction in the histopathology of LP, which is characterized by band-like lymphocytic infiltration [6].

The eccrine gland is composed of the intraepidermal duct (acrosyringium), the intradermal duct, and the secretory gland (the secretory coil) [7]. True invasion of the secretory coil by inflammatory cells (syringotropism) is a very uncommon finding, despite the fact that perieccrine inflammatory cells can be seen in a variety of dermatoses [8, 9]. Syringotropic mycosis fungoidesis an uncommon form of the disease in which syringotropism is typically present in histology [10]. However, we report 2 cases of LP with syringotropic feature in histology which is an uncommon finding.

Case Reports

Case 1

A 29-year-old man with no prior medical history came to our clinic with a 6-month history of pigmented linear itching macules. On his first visit to a medical practitioner, he was treated with Clotrimazole cream for tinea versicolor, but there was no improvement and the lesions worsened.

The lesions were on the left groin and thigh and extended to the back of the knee. Over time, pigmented macules appeared on the right thigh, back, and belly (Fig. 1a–c). Our clinical diagnosis was linear pigmented LP, and to confirm the diagnosis, biopsy was obtained.

Histology revealed a variable degree of lichenoid infiltration, localized hypergranulosis, and basal vacuolar degeneration with Civatte bodies. The papillary dermis exhibited prominent pigmentary incontinence with many melanin-containing macrophages (Fig. 2a, b). The eccrine glands were shown to be significantly involved at the level of the secretory coil, with lymphocytes permeating the epithelial layer (Fig. 2c).

The lymphocytes lacked atypical architectural or cytological features. Hair follicle involvement with the downward extension of lymphocytic infiltration and associated basilar vacuolar change and scattered apoptotic keratinocytes was noted (Fig. 2d).

Dermoscopy also showed round Wickham striae secondary to wedge-shaped hypergranulosis at the center of hair follicles and acrosyringium in all cutaneous lesions. Our diagnosis, based on clinicopathologic correlation, was LP pigmentosus with significant lymphocytic syringotropism and hair follicle involvement. This patient was treated with local steroids and a calcineurin inhibitor (pimecrolimus), with positive effects after 6 months (Fig. 3).

Case 2

A 20-year-old woman presented to our clinic with the major symptom of pruritus papules that grew slowly on her lower back (Fig. 4a, b), and our clinical diagnosis was hypertrophic LP or lichen simplex chronicus. Biopsy and dermoscopy were done for diagnosis. Histology

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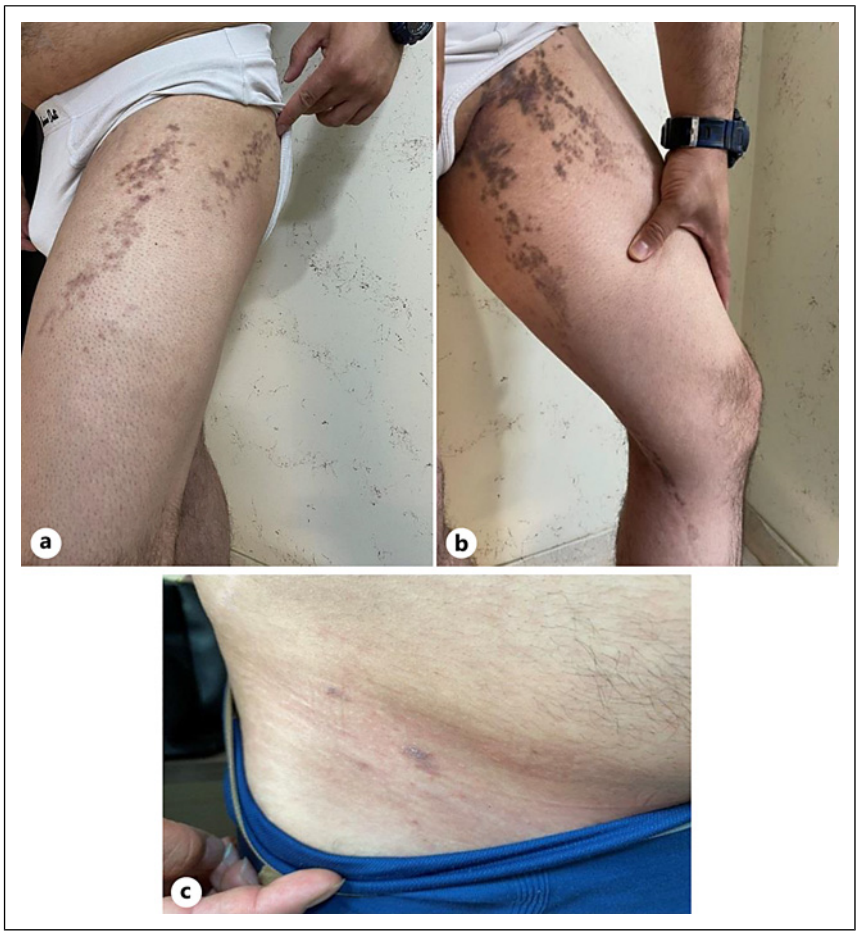

Fig. 1. Clinical presentation of itchy, pigmented macules before treatment.

revealed patchy lichenoid infiltration with a few civatte bodies in dermo-epidermal junction and melanophages in papillary dermis. The secretory portion of the eccrine glands was found to be heavily involved in lymphocyte infiltration. There were no abnormal architectural or cytological features in the lymphocytes (Fig. 5a–d).

There was a good response to treatment after 5 months, which included topical steroids and topical calcineurin inhibitor. Based on the clinicopathologic correlation, consultation with two dermatopathologists, and data review, this case was diagnosed with LP with lymphocytic syringotropism.

Discussion

We described 2 cases of syringotrophic LP with conventional pathologic changes of LP and prominent lymphocytic involvement of eccrine glands at the level of secretory coil and hair follicle. Several interface dermatitides have distinct atypical adnexotropic variants in which the inflammation affects skin adnexa including the hair follicle and the sweat gland [11].

Adnexotropic varieties of interface dermatitides were described in a review by Stephan et al. [11]. The 2 groups of interface dermatitis in this study were those with distinct clinicopathologic adnexotropic variants and those with microscopic adnexotropic features. The following interface

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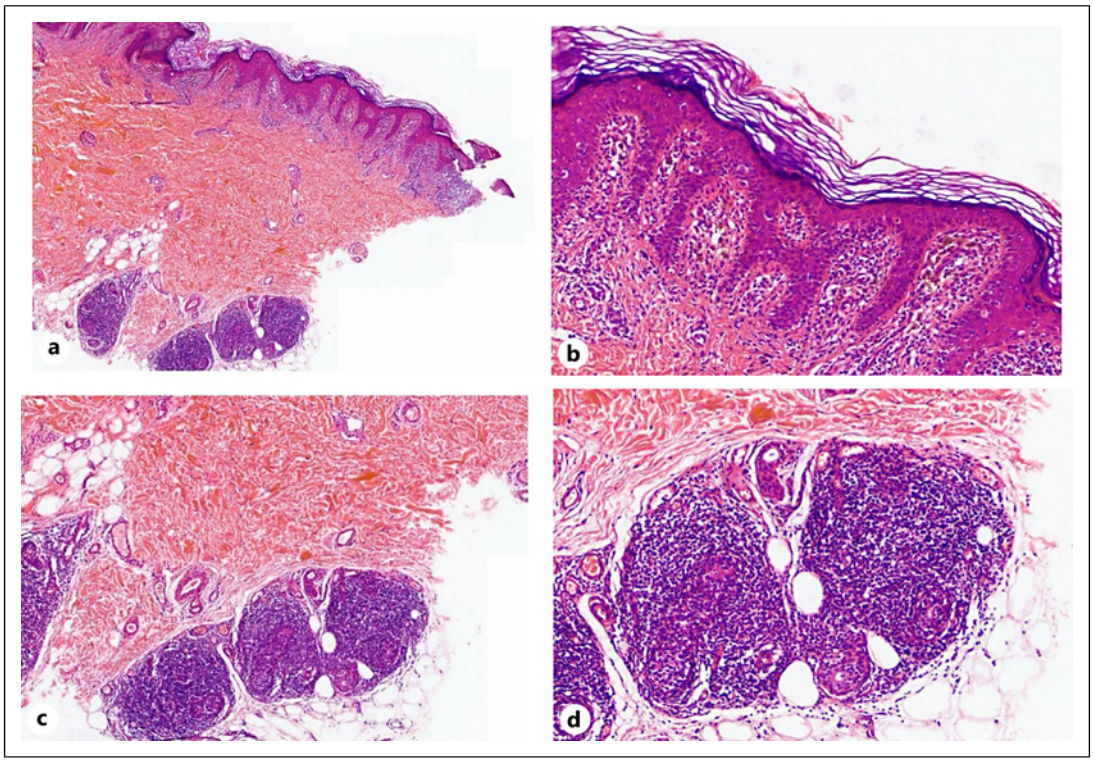

Fig. 2. H&E-stained biopsy samples from lesions. Basilar vacuolar alteration and a few scattered apoptotic keratinocytes. The pigmentary incontinence and melanin containing macrophages were prevalent in the papillary dermis: ×10 (**a**) and ×40 (**b**). Lymphocyte permeation of epithelial cells, with eccrine gland involvement at the level of the secretory coil: ×20 (**c**) and ×100 (**d**).

dermatitis diseases have distinct adnexotropic variants: LP (including follicular LP, lichen follicularis tumidus, and syringotropic LP), cutaneous graft-versus-host disease, lichen nitidus, lichen sclerosus et atrophicus, and lichenoid drug eruptions. Included among the interface dermatitides with microscopic adnexotropic features are connective tissue disease, lichen striatus (LS), pityriasis lichenoides, keratosis lichenoides chronica, and lichen aureus.

Conventional LP alterations are typically restricted to acrosyringia epidermal hypergranulosis [12]. In addition to the common LP pathologic features, perieccrine lymphocytic inflammation has been found, with lymphocyte infiltration into the eccrine coils in syringotropic type [13].

It is critical to recognize that LP can exhibit syringotropism. Perieccrine inflammation with syringotropic lymphocytes is also a common pathologic feature of syringotropic MF, which can lead to misdiagnosis. MF is the most frequent type of cutaneous T-cell lymphoma, with infiltration of eccrine glands as an identifiable characteristic in histology in rare cases [14]. On pathological and clinical grounds, MF cannot be considered in either example of this study as dermo-epidermal junctions exhibit the common appearance of LP.

Mazzeo and colleagues [13] also described a case of syringotrophic LP. However, real lymphocytic syringotropism is a feature of mycosis fungoides (syringotropic MF), but lymphocyte penetration of the secretory coil was observed in both studies.

Lichen planopilaris (LPP) or follicular LP is characterized by localized inflammation that occurs mostly at the level of the follicular infundibulum and isthmus [15]. There is no evidence of perieccrine chronic inflammation in LPP. Consequently, our situations cannot be considered as an LPP variety.

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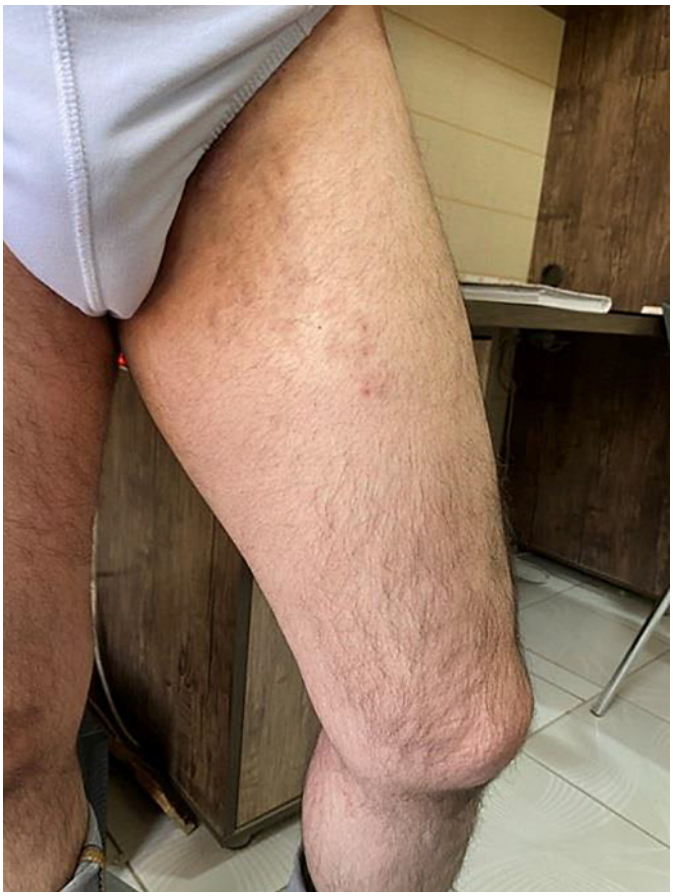

Fig. 3. Clinical features after treatment with topical steroid and topical calcineurin inhibitor.

There has been a report of an unusual type of LP in which only the eccrine tissues are inflamed. However, unlike our cases, lichenoid changes were limited to the acrosyringium (acrosyringeal LP) [16].

A study on sweat gland anomalies was also performed. The sweat glands were abnormal in 9/12 (75%) of the LP patients. Lymphocytic infiltration was observed in 5 cases and only 1 case involved the proximal duct [17]. In contrast to our cases, those in this study involve the glands more frequently in the ducts and acrosyringium than the secretory part. However, the secretory components in our cases largely constituted lymphocyte invasion and infiltration, and a syringotrophic appearance was obvious.

LS is a rare, self-limiting eruption of linear papules which follow the Blaschko lines. Significant lymphoid infiltrates may be observed surrounding the deep secretory portion of eccrine glands in LS [18]. Deep and superficial lymphocyte infiltration of eccrine glands is reported in LS. However, it is more common in children with linear and erythematous lesions [9]. Blaschkitis is an interfacing dermatosis that usually occurs in adults and clinically follows the line of blaschko and histopathology similar to LS [19].

Our cases could be distinct from LS and Blaschkitis both clinically and histologically. In case 1, Even though the main lesion has a linear view, LS, and blaskitis cannot be considered clinically since the patient also has pigmented lesions on other parts of the body and because the lesions were pigmented from the moment they first appeared. As a result, the diagnosis of LP pigmentosus is supported by clinical, pathological, and dermoscopic evidences.

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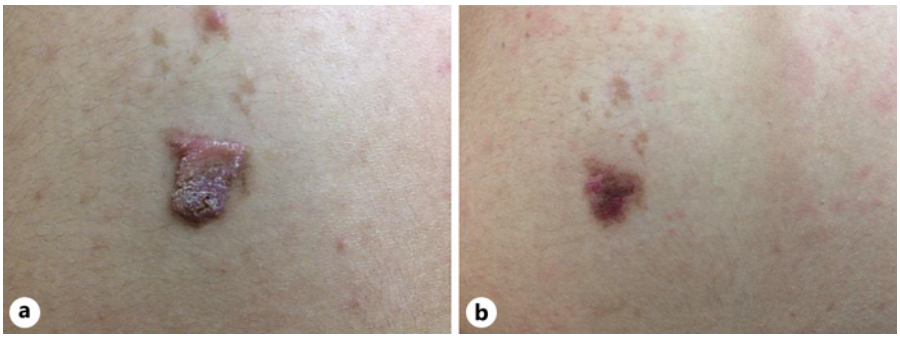

Fig. 4. Clinical presentation before (**a**) and after (**b**) treatment.

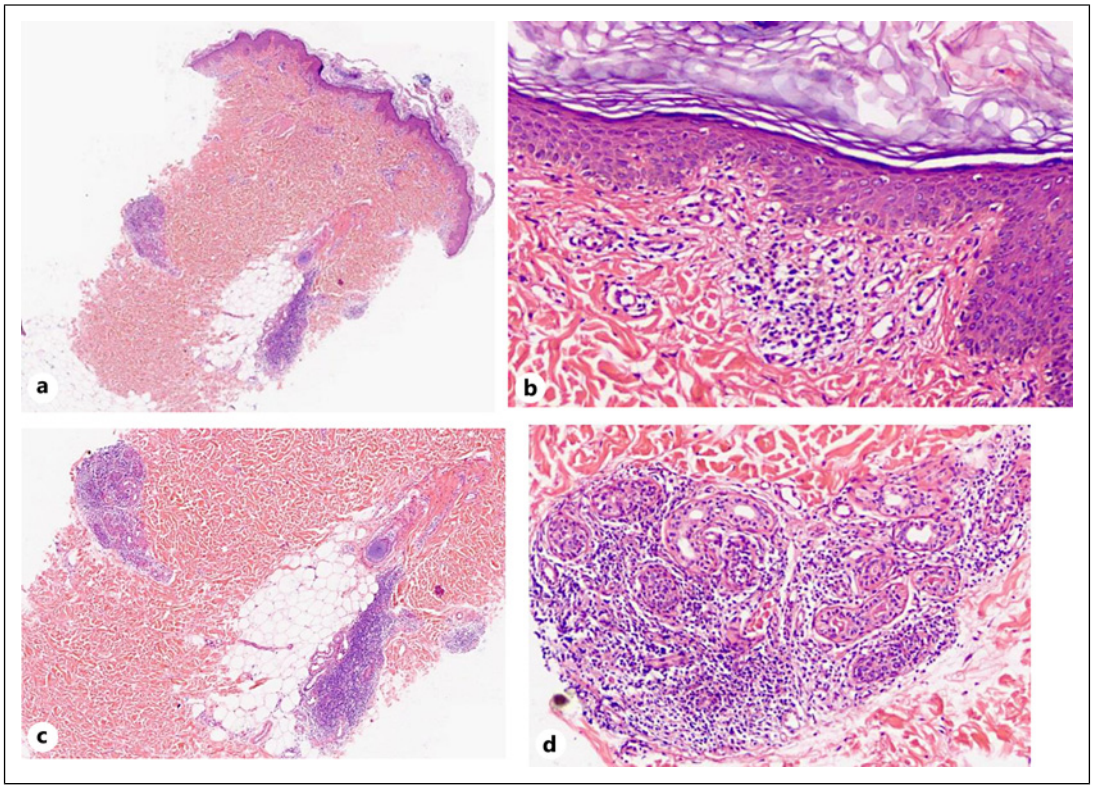

Fig. 5. H&E-stained biopsy specimens. The papillary dermis contains melanophages and Civatte bodies: ×10 (**a**) and ×100 (**b**). Significant lymphocyte infiltration of the eccrine glands (secretory parts): ×20 (**c**) and ×100 (**d**).

In case 2, the lesions are not through the Blaschko lines and are not linear, distinguishing LS and blaschkitis clinically. So, LP with syringotropism has been identified.

In the dermoscopic examination of case 1 lesions, round Wickham stria corresponded to the hair follicle and acrosyringium involvement by LP were seen. However, dermoscopic features of folliculotropic MF include comedo-like structures, follicular hyperkeratosis, and follicular accentuation [20]. Besides, dermoscopic features of LS include gray granular pigmentation accompanied by vessels, with or without whitish scales and white scar-like areas/lines which is different from the pattern seen in LP [21].

In summary, we described 2 uncommon cases of LP that showed the characteristics of lymphocytic infiltration of the sweat glands histopathologically. Syringotropic LP can be considered as a histologic imitator of various syringotropic dermatoses. To prevent being given the wrong diagnosis of these conditions, dermatologists must be aware of this uncommon finding in histology of LP.

Conclusion

Understanding how the conditions listed above can appear as follicular and eccrine inflammation can help dermatologists make the correct diagnosis and prevent misdiagnosis. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000541695).

Acknowledgments

Written consent was obtained from the patients for publication of study. We wish to thank patients for their collaboration.

Statement of Ethics

This case report was approved by the Ethics Committee of Isfahan University of Medical Sciences on February 2024. Written informed consent was obtained from the patients for publication of this case report and any accompanying images.

Conflict of Interest Statement

The authors declare that they have no competing interests.

Funding Sources

This research was conducted without any external funding or financial support. No grants, sponsorships, or third-party funding were involved in the execution or publication of this work.

Author Contributions

Dr. Fatemeh Mohaghegh was responsible for the treatment and management of the patients involved in this study. Dr. Zohre Khodashenas played a key role in gathering clinical information and relevant data for analysis. Dr. Mina Saber performed the dermoscopic examinations and provided expert consultation on the content and structure of the manuscript. Dr. Haniyeh Sohrabi contributed by writing the manuscript and coordinating the overall process of manuscript preparation. All authors have read and approved the final version of the manuscript.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material files. Further inquiries can be directed to the corresponding author. The raw data supporting the conclusions of this study will be made available by the authors to any qualified researcher without restriction.

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