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Case Report

Ulcerative Lichen Planus with an Unusual Lichenoid Infiltrate Masquerading as Secondary Syphilis

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Abstract

Ulcerative Lichen Planus (LP), also known as erosive lichen planus, is an variant of LP characterized by chronic, painful and cicatricial bullae and ulcerations. This form of LP most commonly involves mucosal surfaces and sometimes palmoplantar surfaces.

Ulcerative LP less commonly involves areas such as the scalp, nails and anogenital regions. This variant differs from the normal variant due to its chronicity and disabling symptoms. We describe a unique case of a 24-year old African American male who presents with painful and pruritic shallow ulcerations of the bilateral palms, nailbeds, lower legs and glans penis. Clinically, the lesions on this patient appear to share features with the lesions seen in secondary syphilis. After extensive workup, including clinical and histological examination and process of elimination, the patient's lesions were attributed to a diagnosis of ulcerative LP. The reasons for presenting this case are two-fold. The first reason is for clinical interest and to remind clinicians to keep ulcerative LP as a consideration in the differential diagnosis when evaluating ulcerative eruptions located on palmoplantar and anogenital regions. The second reason is for histopathological interest and to remind pathologists that when analyzing histology to keep in mind the unique histopathologic findings that can be seen with ulcerative LP.

Keywords: Ulcerative Lichen Planus; Anogenital Regions; Ulcerations

Introduction

Ulcerative Lichen Planus (LP) is a variant of lichen planus characterized by chronic, painful erosions, bullae and ulcerations most commonly located on the mucous membranes. Ulcerative lesions also occur on the palmoplantar surfaces, particularly on the soles. This variant is more common in female patients despite the fact that palmoplantar LP is more common in males.

Ulcerative LP clinically manifests as intense pain, typically described by patients as a "burning pain". These lesions may result in therapy-resistant ulcers and lead to chronic atrophic scarring or the development of squamous cell carcinoma within these lesions [1]. Histologically, the ulcerative type resembles the other forms of LP with degeneration of the basal layer of the epidermis and a lymphocytic infiltrate obscuring the dermoepidermal junction, as well as sparse scattered plasma cells. Other important histologic features include hyperkeratosis without parakeratosis and focal increases in the granular cell layer.

Syphilis is a chronic, multistage disease caused by *Treponema pallidum* that is acquired via sexual contact with active primary or secondary lesions. Syphilis can present in one of four varying stages. Syphilis is often referred to as "the great mimicker" because lesion characteristics vary to a considerable degree. The second stage of syphilis typically presents as a disseminated disease

characterized by constitutional symptoms, a rash and condyloma lata. The rash of secondary syphilis consists of papules or patches covering the trunk and extremities, as well as the palms and soles. Condyloma lata refers to the broad, white, wart-like lesions that accompany the rash and typically occur on mucosal membranes. We report a unique case of a 24-year old African American male who presented with shallow ulcerations, which clinically and histologically appeared to share features with the lesions seen in secondary syphilis.

Case Report

A 24-year-old African American male presented with a 6-month history of intensely pruritic and painful ulcerations involving the fingers, fingernail beds, palms, glans penis and lower legs. His past medical history was significant for Hodgkin's lymphoma, which had been treated two years earlier, with no evidence of recurrence since treatment. He was not taking any medications at the time of presentation.

On physical examination, the patient was afebrile and denied systemic symptoms on review of systems. Cutaneous examination revealed well-demarcated, round to oval, shallow ulcerations measuring 1-2 cm on the bilateral palms, nail beds, lower legs and glans penis (Fig. 1-3). Additionally, the right heel demonstrated a larger, deep, weeping ulceration measuring 5 × 3 cm (not shown).

Based on the clinical presentation, the differential diagnosis included syphilis, primary HIV infection, ulcerative Lichen Planus (LP), lichenoid drug eruption, pityriasis rosea, viral exanthem, infectious etiologies, paraneoplastic pemphigus, plasma cell dyscrasia, recurrent Hodgkin's lymphoma and ulcerative Herpes Simplex Virus (HSV). Secondary syphilis was the primary concern at initial presentation due to the anatomical distribution of the lesions.

Several involved sites were sampled using 8-mm punch biopsies for histopathologic evaluation. Laboratory studies included darkfield microscopy, Warthin-Starry staining, Rapid Plasma Reagin (RPR), fluorescent treponemal antibody absorption (FTA-ABS), HIV testing, Tzanck preparation, Purified Protein Derivative (PPD) and wound cultures for bacteria, HSV, fungi and mycobacteria. Additional testing evaluated autoimmune disease and possible recurrence of Hodgkin's lymphoma. Initial empirical treatment consisted of two courses of acyclovir for presumed HSV infection and three intramuscular doses of penicillin for possible syphilis.

Histopathologic examination revealed hyperkeratosis, focal hypergranulosis, irregular acanthosis and a characteristic "saw-toothing" of the epidermis with vacuolar degeneration. A dense, band-like lymphoplasmacytic inflammatory infiltrate was present in the dermis, obscuring the dermoepidermal junction and extending into the deep dermis in a perivascular and periadnexal distribution (Fig. 4-6).

Darkfield microscopy, Warthin-Starry staining, RPR and FTA-ABS testing were negative, effectively ruling out syphilis. HIV testing, Tzanck preparation, PPD and wound cultures for bacteria, HSV, fungi and mycobacteria were also negative. The Antinuclear Antibody (ANA) titer was 1:40, making autoimmune disease unlikely. The patient's wife tested negative for syphilis with nonreactive RPR and FTA-ABS results. Immunohistochemical staining for light chain restriction and immunofixation electrophoresis ruled out plasma cell dyscrasia. Bone marrow aspiration and lymph node biopsies showed no evidence of recurrent Hodgkin's lymphoma. Indirect immunofluorescence excluded paraneoplastic pemphigus. A diagnosis of ulcerative lichen planus was made based on histopathologic findings and exclusion of other conditions. At a two-month follow-up visit, no clinical improvement was observed. The patient was subsequently started on an oral course of prednisone, resulting in greater than 50% healing of the lesions.

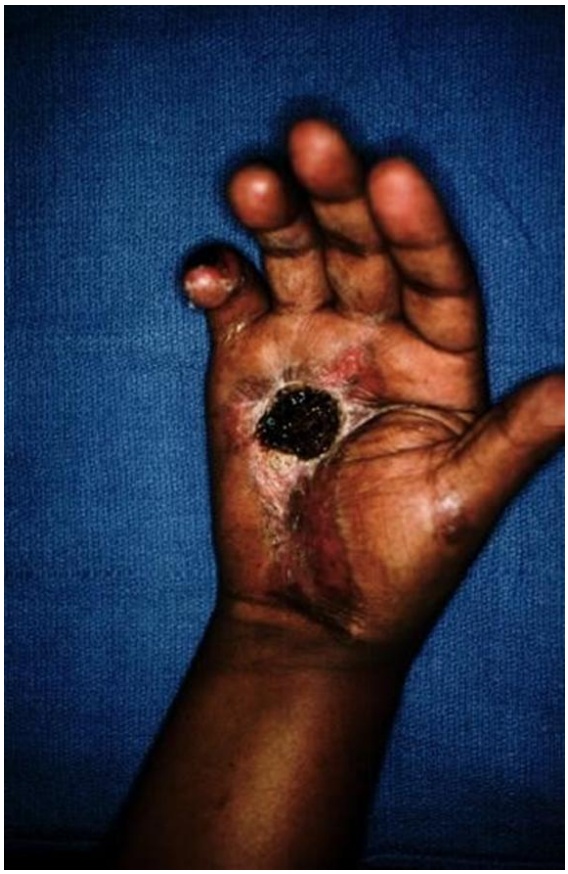

Figure 1: Well-circumscribed 2-cm oval ulceration of the right palmar hand.

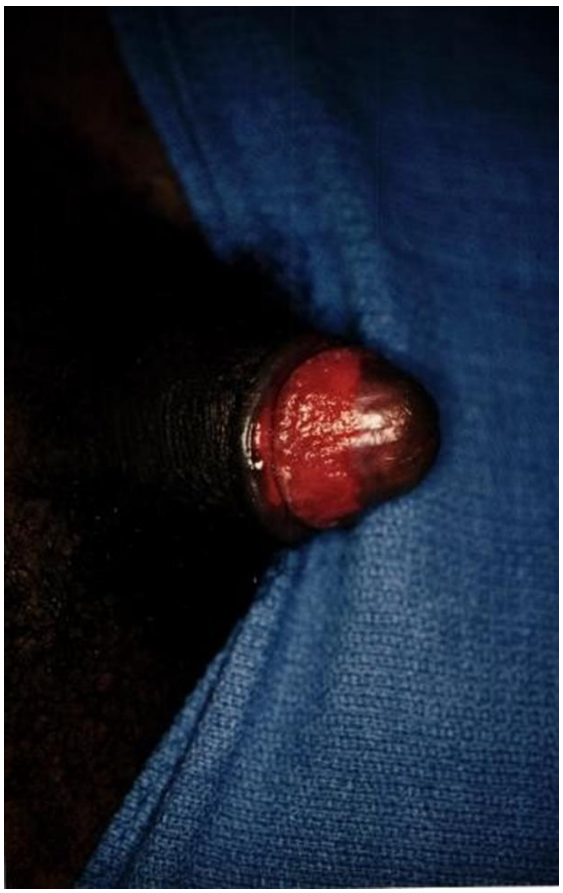

Figure 2: Well-demarcated pigmented flat topped papule overlying the tip of glans penis.

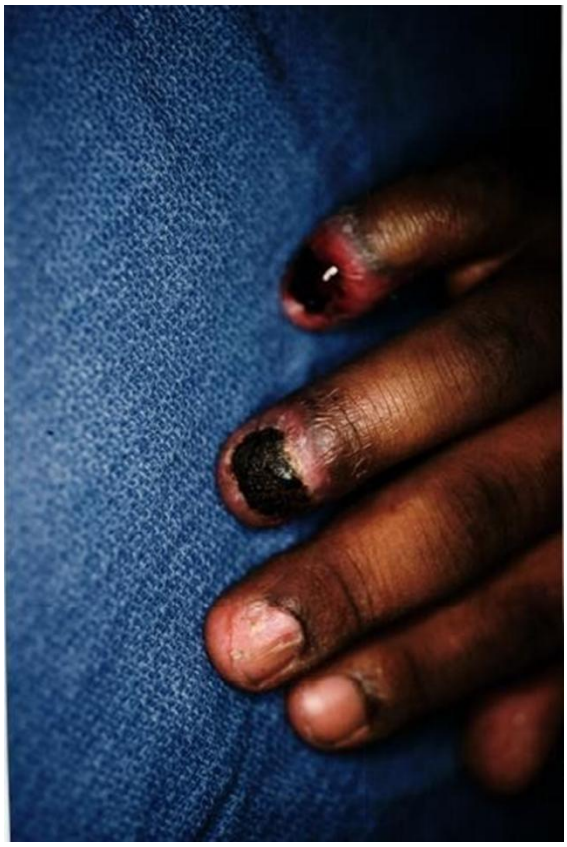

Figure 3: Destruction of nail fold and nail bed of the right fourth and fifth digit due to crusted ulcerations and scarring.

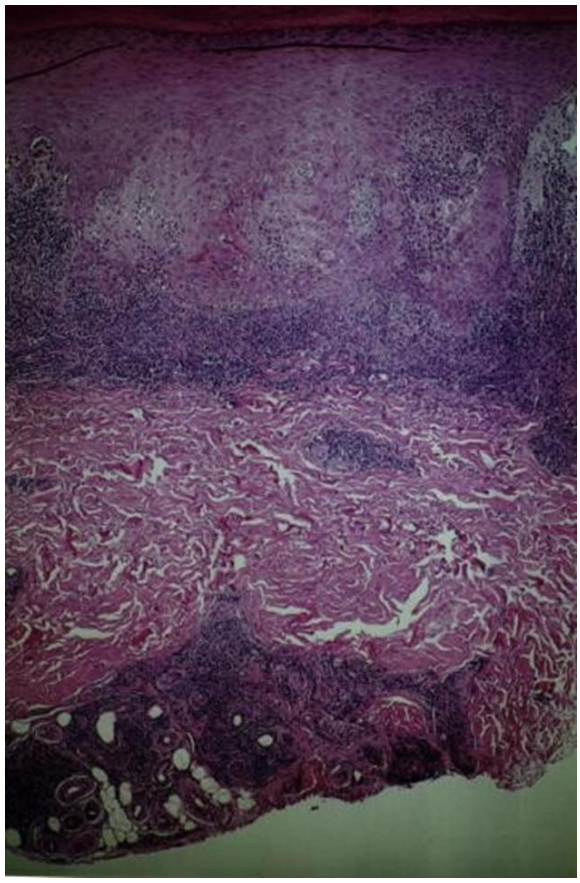

Figure 4: (H&E, 40X magnification) Histology shows epidermal irregular acanthosis with a saw-tooth pattern, hyperkeratosis, parakeratosis and hypergranulosis.

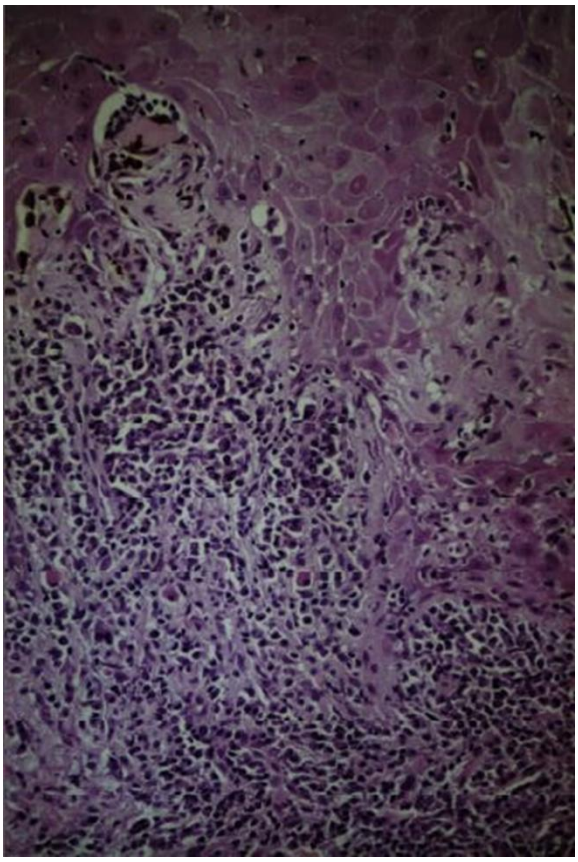

Figure 5: (H&E, 200X magnification) At higher magnification, there is epidermal spongiosis with vacuolar degeneration of the basal cell layer and individually necrotic keratinocytes. The inflammatory cell infiltrate is very rich in plasma cells with occasional Russell bodies.

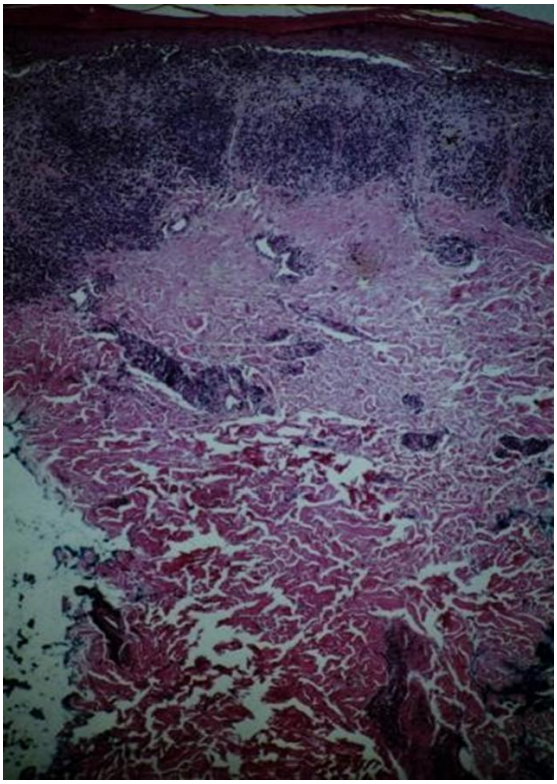

Figure 6: (H&E, 40X magnifications) There is a band-like dense lymphoplasmacytic inflammatory cell infiltrate in the dermis abating the dermoepidermal junction and extending to the deep dermis in a perivascular and periadnexal distribution.

Discussion

This case is diagnostically challenging due to the unusual clinical distribution of lesions. Cutaneous manifestations of ulcerative LP may closely resemble those of secondary syphilis, particularly when involving the palms and soles. Syphilitic lesions can present in multiple morphologic forms, including macular, papular, follicular, lichenoid, vesicular, pseudovesicular or psoriasiform patterns. Consequently, syphilis must be rigorously excluded to prevent serious complications associated with untreated infection. While anogenital involvement is common in syphilis, ulcerative LP rarely presents in this region. Therefore, a comprehensive diagnostic evaluation is essential to establish an accurate diagnosis and guide appropriate treatment. Close follow-up is warranted in patients with ulcerative LP due to its chronic course and the potential risk of squamous cell carcinoma developing within chronic lesions.

Histologically, ulcerative LP is typically characterized by a lymphocytic infiltrate with few or absent plasma cells. However, several other dermatologic and infectious conditions including syphilis, plasmacytoma, necrobiosis lipoidica, leishmaniasis and deep fungal infections may share similar features. Rare case reports describe ulcerative LP with a prominent plasma cell-rich lichenoid infiltrate. These cases have primarily involved older patients with nail involvement [2-6].

Ulcerative LP has been reported in association with immune dysfunction and various malignancies, including small-cleaved cell lymphoma, non-Hodgkin's lymphoma, Castleman's disease, metastatic adenocarcinoma and malignant fibrous histiocytoma. It has been proposed that ulcerative LP may represent an autoimmune reaction resulting from cross-reactivity between tumor antigens and cutaneous or mucosal structures. Cytotoxic T lymphocytes sensitized to tumor antigens may attack cross-reactive skin antigens. Notably, tumor-associated lichen planus has been reported to persist even after complete remission of the underlying malignancy.

Conclusion

Ulcerative LP can present unconventionally. The clinical and histopathological features may vary from the standard presentation. Thus, this case illustrates an important concept that there should be a high index of suspicion for ulcerative lichen planus when evaluating ulcerative lesions of the palmoplantar and anogenital areas.

Conflict of Interest

The authors declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

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None

Data Availability Statement

Not applicable.

Ethical Statement

The project did not meet the definition of human subject research under the purview of the IRB according to federal regulations and therefore, was exempt.

Informed Consent Statement

Informed consent was taken for this study.

Authors' Contributions

All authors contributed equally to this paper.

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