Clinical and Histopathological Findings in Palmoplantar Lichen Planus Presenting as Diffuse Keratoderma

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Introduction

Palmoplantar lichen planus (PPLP) is a rare and poorly described entity which frequently does not present the typical clinicopathological features of lichen planus (LP) [1]. The protean spectrum of manifestations along with its overall rarity make PLPP a challenging clinical scenario [2].

Case Presentation

Herein, we report on a 54-year-old woman referred to our Dermatology unit for palmoplantar itching appeared two months before. Her medical history was positive for esophageal candidiasis and biopsy-confirmed vulvovaginal and oral lichen planus, successfully treated with high-potency topical steroids three years before. The patient family history

was unremarkable and extensive anamnesis failed to identify any suspected drug or potential infectious trigger. The routine laboratory tests were in normal ranges and serology for hepatitis B and C were negative, as well as VDRL and TPPA for syphilis. Upon physical examination, smooth yellowish-orange patches and plaques sparing the nail apparatus were seen on the patient soles. Similar lesions were noted over both the palms of the hands. A punch biopsy performed on the plantar surface of the right medial arch revealed hyperkeratosis, focal wedge-shaped hyper-granulosis, irregular acanthosis, saw tooth-shaped epithelial rete ridges, sporadic epidermal colloid bodies and lymphohistiocytic inflammatory infiltration in the papillary dermis (Figure 1, A-D). According to the clinical and histopathological findings, a diagnosis of PPLP presenting with diffuse keratoderma pattern was made.

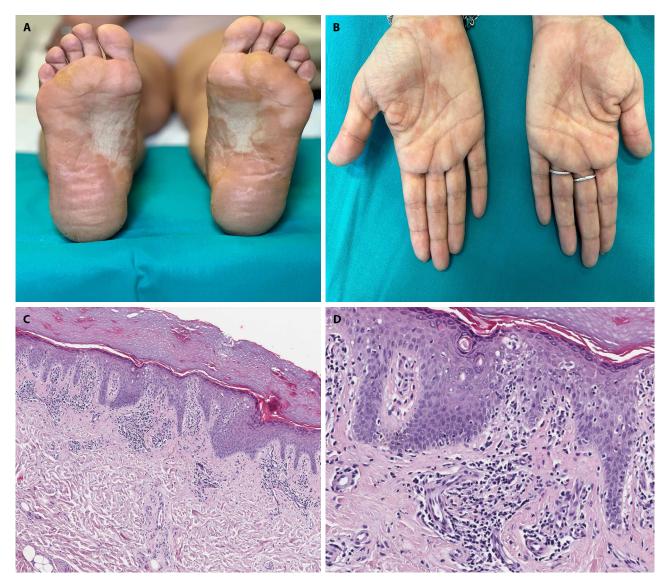


Figure 1. (A) Smooth yellowish-orange patches and plaques on the patient soles. (B) Erythematous-yellowish patches and plaques on the patient palms. (C) Hyperkeratosis, focal wedge-shaped hypergranulosis, irregular acanthosis, areas of spongiosis, and scattered epidermal colloid bodies. In the papillary dermis, lymphohistiocytic inflammatory infiltration vaguely arranged in a band-like pattern (H&E 8x). (D) Interface dermatitis with areas of focal epidermal exocytosis and lymphohistiocytic inflammatory infiltration in the papillary dermis (H&E 20x).

Discussion

To date, several morphological patterns of PPLP lesions have been described: erythematous plaques, punctate keratosis, diffuse keratoderma and ulcerated lesions [3,4], yet diffuse keratoderma presentation has been described in only few reports [1-5]. Such presentation may represent an isolated form or may be associated with a previous history of lichen planus in other sites. In a study on 18 patients with LP lesions, either predominantly or exclusively on palms and/or soles, the diffuse palmar keratoderma presentation was reported only in one patient. The histology reported compact hyperkeratosis with epidermal hyperplasia, hyper-granulosis and a band-like infiltrate of lymphocytes [5]. Moreover, Sanchez-Perez et al identified 36 patients with LP affecting the palmoplantar area, two of whom presented with a diffuse hyperkeratotic

lesions forming a palm-plantar keratoderma pattern [3]. This unreported association of PPLP with oral and genital LP provides further data on this underrecognized clinical condition, with the aim of supporting clinicians in the differential diagnosis with similar diseases, such as palmoplantar keratoderma. While banded infiltrates and parakeratosis are not consistently found in PPLP and not essential features for the diagnosis in the presence of other suggestive ones, the histological assessment remains mandatory to achieve the diagnosis [3]. The reasons why the LP presentation appears atypical only in certain cases, the frequency, clinical course, and possible molecular mechanisms underlying have yet to be established. Albeit rare, this variant should be considered when dealing with unusual LP clinical pictures, aiming at preventing diagnostic delay and at promptly performing suitable diagnostic tests, when needed.

Conclusions

In conclusion, we have reported an atypical PPLP case with diffuse keratoderma pattern associated with typical oral and genital LP. Further research is warranted to shed light on the pathological mechanisms underlying of this peculiar clinicopathological picture.

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