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ORIGINAL ARTICLE

Erosive palmoplantar lichen planus: clinical and histopathological insights from an ethnic population of North-East India

Líquen plano palmo-plantar erosivo: perceções clínicas e histopatológicas de uma população étnica do nordeste da Índia

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Abstract

Objectives: Palmoplantar lichen planus (LP) is an uncommon disorder often posing a diagnostic dilemma, particularly in the absence of characteristic LP lesions elsewhere in the body, but also because of its varied atypical presentations. The erosive variant of palmoplantar LP is an even rarer entity that has been described only in small case reports. Therefore, this study aimed to describe the clinico-pathological spectrum of this variant of palmoplantar LP. **Methods:** A retrospective study was conducted in a tertiary care hospital in North East India from 2013-2023, where records of all cases diagnosed as erosive palmoplantar LP were included for descriptive analysis. **Results:** A total of 16 patients were included, including seven females and nine males with a median age of 15 years (interquartile range: 12-30), with a predominance of pediatric patients (n = 10; 62.5%). All patients had plantar involvement, half had nail involvement, and only 2 patients (12.5%) had typical LP lesions elsewhere, but with no mucosal lesions or scarring alopecia. Histopathological findings included band like inflammatory infiltrate at the dermo-epidermal junction and basal cell degeneration, respectively, in 100% (n = 16) and 93.75% (n = 15) of the cases. **Conclusion:** This study presents the largest number of patients with a clinico-histopathological diagnosis of erosive plantar LP particularly from a single indigenous tribal population in North East India, with a male and pediatric preponderance, for which we have no explanation.

Keywords: Anonychia. Erosive. Lichen planus. Palmoplantar.

Resumo

Objetivos: O líquen plano palmo-plantar é uma doença incomum, que representa frequentemente um dilema diagnóstico, não só pela ausência de lesões características de líquen plano (LP) noutras partes do corpo, mas também pelas suas apresentações atípicas. A variante erosiva do LP plano palmoplantar é uma entidade ainda mais rara, descrita apenas em séries curtas ou relatos de casos. Este estudo teve como objetivo descrever o espectro clínico-patológico do LP erosivo palmoplantar. **Métodos:** Este foi um estudo retrospetivo realizado num hospital terciário no nordeste da Índia entre 2013 e 2023, onde os registos de todos os casos diagnosticados de LP palmoplantar erosivo foram incluídos para análise descritiva. **Resultados:** Foram incluídos um total de 16 doentes, com mediana de idade de 15 anos, com predomínio dos doentes da população pediátrica em 62.5% (n = 10). Todos os doentes apresentavam envolvimento plantar, metade afetação ungueal e apenas dois tinham lesões típicas de LP, mas sem lesões da mucosa oral ou alopécia cicatricial. Os achados histopatológicos

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incluíram infiltrado inflamatório em faixa na junção dermoepidérmica e degeneração das células basais em 100% (n = 16) e 93,75% (n = 15), respetivamente. **Conclusão:** Este estudo demonstra o maior número de doentes com diagnóstico clínico-histopatológico de LP plantar erosivo, particularmente de uma única população tribal indígena no nordeste da Índia, com preponderância masculina e afetando frequentemente a população pediátrica.

Palavras-chave: Anoníquia. Erosivo. Líquen plano. Palmo-plantar.

Introduction

The first description of erosive palmoplantar lichen planus (LP) was made in 1966 by Cram et al.[1] as a rare variant of LP presenting as a painful bullous eruption that progresses to chronic and debilitating erythematous ulcerated plaques affecting the soles, that can sometimes be preceded by hypertrophic and bullous plantar lesions[1]. In the few published small series and case reports, additional sites were involved, including the oral and/or genital mucosa and the nail apparatus, in the latter with atrophy and spontaneous shedding of the nail plate, seldom with characteristic LP lesions over other body sites[1-3]. Cicatricial alopecia of the scalp was commonly associated[1,3]. Whether this condition is a separate entity, with its isolated involvement of the soles, or it falls within the spectrum of Graham Little Syndrome due to its association with cicatricial alopecia which is still unknown[4]. As the clinical picture can be varied, histopathology plays a pivotal role in confirming diagnosis in such patients.

This study intends to characterize better the clinical and histopathological characteristics of ulcerative palmoplantar LP.

Methods

This study was a retrospective observational analysis conducted over a 10-year period (2013-2023) in the department of dermatology at a tertiary care teaching hospital in Northeast India. Approval was obtained from the Institutional Ethics Committee (NEIGR/IEC/M11/F14/2023).

The hospital's dermatology database and histopathology records were systematically reviewed to identify all patients with a clinico-histopathological diagnosis of the erosive variant of palmoplantar LP. The inclusion criteria involved histopathologically confirmed cases of LP with erosive lesions limited to the palms and/or soles. Patients with incomplete records were excluded.

For each eligible patient, demographic details (age and sex), clinical characteristics (site of involvement, duration of disease, presence of nail changes, and concurrent mucocutaneous LP lesions elsewhere), and histopathological findings were extracted from medical records. Histopathology slides were reviewed when available to document specific features including type and distribution of inflammatory infiltrate, basal cell degeneration, hypergranulosis, presence of Civatte bodies, and any additional epidermal or dermal changes.

Data were compiled and analyzed descriptively. Continuous variables were summarized using median and range, while categorical variables were expressed as frequencies and percentages.

Results

A total of 16 patients whose biopsy finding was consistent with erosive variant of plantar LP were included in the study. There were nine males and seven females (F:M ratio 1:1.28) with a median age of 15 years (Interquartile range 12-30), varying between 9 and 62 years, with the majority of pediatric patients (n = 10; 62.5 %). A positive family history was observed in 2 siblings (12.50%) and all the patients belonged to one ethnic tribe of Meghalaya. Although the exact census data for the specific tribe is unavailable, community notes indicate that marriage outside the tribe is uncommon, however without any documented consanguinity. Disease duration exceeded 1 year in all cases.

All cases revealed involvement of the heels, with painful hyperkeratotic plaques with ulcerations (Figs. 1A and B), and a common history of heel rubbing with a pumice stone. None of the patients had palmar involvement.

Nail involvement occurred in 8 patients (50%), with toe nails affected in all the eight patients while finger nails were additionally affected in four individuals. All ten toe nails units were involved in 2 out of 8 patients (25%), five to nine toe nail units were involved in 4 (50%), and 2 patients (25 %) had < 5 toe nail units affected. Out of the four patients who had finger nail involvement, two patients had 1-2 units affected, one had 7, and the

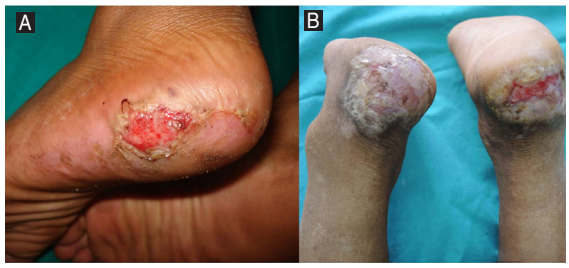

Figure 1. A and **B:** hyperkeratotic plaques with ulcerations over the heels.

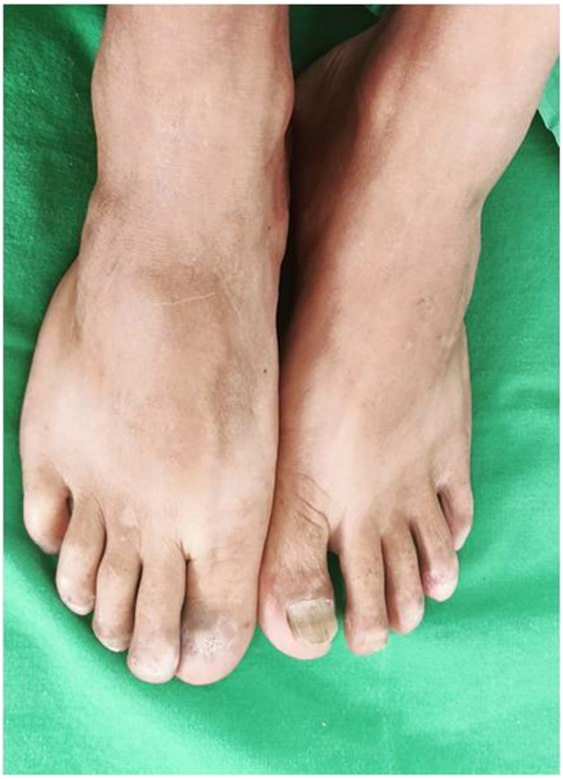

Figure 2. Partial anonychia of toe nails.

other had all 10 nail units affected. Complete or partial anonychia occurred in all the eight patients (Fig. 2). Dystrophic nail changes and periungual scaly plaque were the second most common finding (n = 5; 62.5%), followed by pterygium in 4 (50%) (Fig. 3A and B), longitudinal ridging and nail splitting in 3 each (37.5 %), periungual erosions in 2 (25%), and Beau's line in one (12.5%). One patient presented with 20 nail dystrophy (Fig. 4) and another with syndactyly of the toes secondary erosive lesions and scarring, giving the foot a characteristic mitten appearance (Fig. 5).

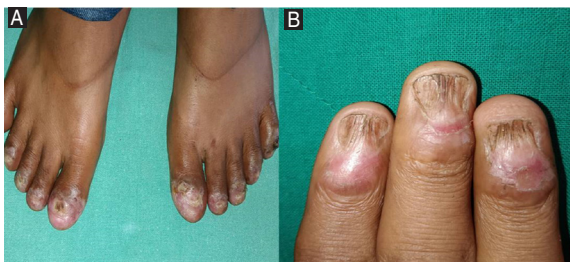

Figure 3. A and **B**: dystrophic nail changes and periungual scaly plaques involving the toe nails and dorsal pterygium involving the finger nails.

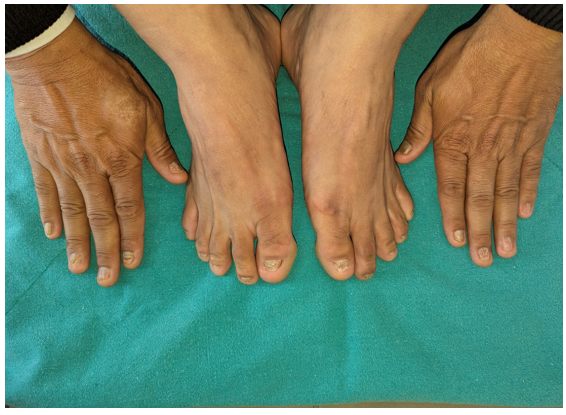

Figure 4. Twenty nail dystrophy.

One patient had LP on the dorsum of the fingers, but none of the patients had scarring alopecia of the scalp or mucosal involvement.

Histopathological findings were consistent with erosive LP in all the individuals, as shown in figure 6. Most prominent epidermal changes included basal cell degeneration observed in 93.75% (n = 15), followed by hyperkeratosis in 62.5% (n = 10), irregular acanthosis in 50% (n = 8), and hypergranulosis in 43.75% (n = 7). Dermal changes revealed characteristic band like lymphocytic inflammatory infiltrate along the dermo-epidermal junction in all cases and perivascular inflammation in 31.25% (n = 5). Civatte bodies and saw toothing of rete ridges were observed in 4 cases (25%), whereas melanophages were noted in 3 (18.75%). Atypical features included parakeratosis and a neutrophilic inflammatory infiltrate in two cases each. There was no evidence of malignancy on histopathological evaluation.

Hepatitis serology was not systematically performed in this retrospective study. Where available, results were non-reactive.

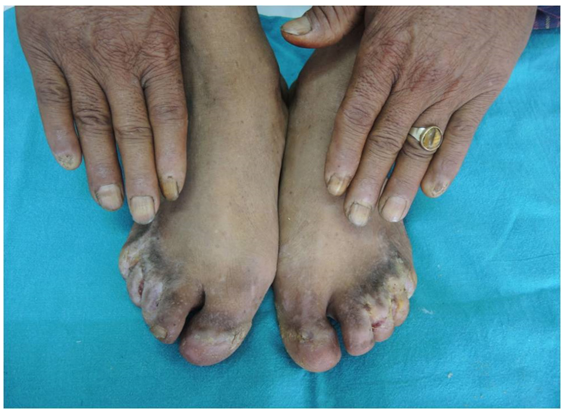

Figure 5. Syndactyly with mitten appearance of toe nails.

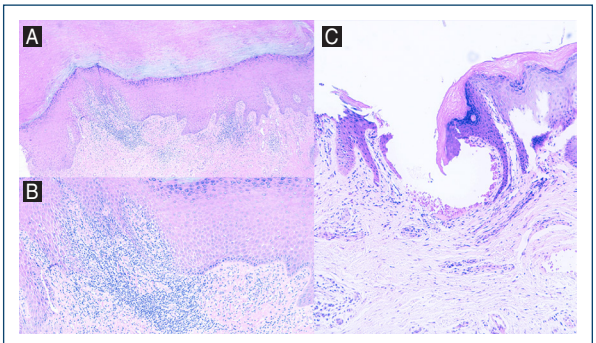

Figure 6. A: low-power view showing epidermis with hyperkeratosis, hypergranulosis, acanthosis with saw-toothing of rete-ridges, and dense lymphocytic infiltrate hugging the dermoepidermal junction (H&E stain, ×4). **B:** high-power view showing dense lymphocytic infiltrate obscuring the dermoepidermal junction. Also seen Max Joseph space between epidermis and papillary dermis (H&E stain, ×20). **C:** a case of erosive lichen planus showing ulceration of the epithelium with adjacent irregular acanthosis, Max Joseph space, and lymphocytic infiltrate in the upper dermis (H&E, ×20).

This was a retrospective study, and structured follow-up was beyond the scope of the study. All patients had a disease duration exceeding 1 year at presentation. The lesions exhibited a chronic, slowly progressive course with intermittent symptom fluctuation. Topical corticosteroids were initiated for all patients at their first visit, and no systemic therapy had been administered before presentation. Despite the chronicity, none of the cases showed histopathological evidence of malignant transformation.

Discussion

Since the first description of erosive palmoplantar LP by Cram et al., in 1966[1], there is limited information regarding the clinical and demographic details of this entity, mostly by publication of isolated case reports and small case series. Our study is the first to highlight the clinical-histopathological characteristics in a series of 16 patients all from one of the indigenous tribes in Meghalaya, in the Northeast India.

On reviewing and comparing with the literature, a single study was conducted in 2018 by Chessa et al.[5] in Italy which included seven patients with a mean age of 49 years with this erosive form of LP with nail unit involvement. In contrast with these and many other published cases where adults predominate, our study revealed a median age of 15 years with a predominant pediatric population. However, gender distribution was similar and the M:F ratio with males accounting for 56.25% of the subjects was comparable in ours and Chessa's study, although more female involvement was referred in previous case reports[6-8]. In addition, our study demonstrated a positive family history in two sisters, which has never been reported before, but no parents or other first-degree relatives give a similar history, even though there was a common ethnic background to all patients in our study (the tribe of Meghalaya). We cannot exclude a common genetic or environmental influence on the unexpected high number of cases in this tribe. As marriage outside the tribe is uncommon, the genetic pool is relatively restricted, which may predispose to inherited susceptibility. Moreover, habitual rubbing of the heels with a pumice stone, a common cultural practice in this community, may act as a mechanical trigger for lesion development on acral skin. All patients had a disease duration exceeding 1 year at presentation, underscoring the chronic and recalcitrant nature of plantar erosive LP. However, these associations remain speculative and require further investigation through larger, prospective, and molecular studies.

Heels were the only common site of involvement in our study but sole involvement is seldom reported in other studies[9,10]. Neville JA reported palm and scrotal involvement in a man, which we did not find[11].

Nail changes were present in half of the subjects, involving particularly the toe nails, with either complete or partial anonychia of all or a few nail units and nail dystrophy with periungual scaly plaque and pterygium.

This contrasts with Chessa et al. findings as all 7 patients had nail bed erosions and marked inflammation of the periungual skin[5], eventually suggesting more severe disease in these patients. In addition, two of these patients had webbing and fusion of the toes with sole while, in our study, only one of the subjects had syndactyly with mitten appearance. None of the patients in these two studies had mucosal involvement or scarring alopecia, contrasting with Jiménez-Sánchez MD et al. who reported whitish reticulate oral LP and scalp alopecia in a patient[8], Romero et al.[12] who reported oral erosive LP and Bazargan et al.[13] who reported erosive plantar LP associated with longstanding LP and erosive oral mucosal involvement. As in our series, concurrent cutaneous LP was not frequently documented.

We observed no malignancies within this 10-year period of the study, although literature reports development of malignancies over the chronic ulcerations. A case report by Thakur et al. reported the development of verrucous carcinoma over an ulcerative lesion of LP of the sole after 20 years of disease onset, suggesting disease chronicity; however, literature reporting the same is scarce[14].

Skin biopsy helps to confirm the diagnosis of erosive LP and rule out malignancy. On histopathology, typical findings of erosive LP are hyperkeratosis without parakeratosis, liquefaction degeneration of the epidermal basal layer, saw-toothed appearance of rete ridges, Civatte bodies (eosinophilic masses representing apoptotic keratinocytes), and a band-like lympho-histiocytic infiltrate at the dermo-epidermal junction. However, due to the presence of ulceration, many of these features may not be observed, as occurred in our case[15].

In our study, treatment was limited to the use of topical corticosteroids, which were initiated for all patients at their first visit to relieve pain and reduce inflammation. However, published literature highlights that management of erosive LP remains challenging due to its chronic, relapsing course, and tendency for partial response. According to a systematic review by Ho and Hantash[16], systemic corticosteroids are the most frequently used agents, offering partial to complete remission in many cases but with frequent relapse after discontinuation. Systemic retinoids such as etretinate and acitretin have shown modest benefit, particularly for mucosal disease, though their use is often limited by mucocutaneous side effects. For refractory disease, immunosuppressants including azathioprine, cyclosporine, mycophenolate mofetil, methotrexate, and thalidomide have been reported to induce improvement, with azathioprine appearing most effective among this group. Biologic therapies such as adalimumab and infliximab have been used in resistant cases, but responses are inconsistent and paradoxical lichenoid reactions have been described. Interestingly, Bacillus Calmette-Guérin therapy achieved the highest overall treatment assessment score in the review, though evidence remains limited to small series. Overall, therapeutic selection should be individualized, balancing efficacy and safety, and the risk of oncologic degeneration in long-standing erosive disease.

The present study is limited by its retrospective nature and follow-up were incomplete in several cases. Hepatitis serology and genetic testing were not systematically performed, and structured follow-up to assess treatment response or long-term outcomes was beyond the study's scope. Consequently, potential associations between genetic predisposition, environmental factors, and chronicity remain speculative and warrant validation through larger, prospective studies.

Conclusion

This study throws light on the clinical characteristics of erosive palmoplantar LP, a rare skin disease with frequent nail involvement but often without oral lesions or cicatricial alopecia, suggesting it can be considered a separate entity. Our observation points to a childhood and male preponderance, instead of female and adults preponderance previously mentioned in the literature, which may be due to some genetic or environmental factors present in this population of the Northeast India, that may justify the higher frequency and/or earlier disease onset.

Funding

None.

Conflicts of interest

None.

Ethical considerations

Protection of humans and animals. The authors declare that the procedures followed complied with the ethical standards of the responsible human experimentation committee and adhered to the World Medical Association and the Declaration of Helsinki. The

procedures were approved by the institutional Ethics Committee.

Confidentiality, informed consent, and ethical approval. The authors have followed their institution's confidentiality protocols, obtained informed consent from patients, and received approval from the Ethics Committee. The SAGER guidelines were followed according to the nature of the study.

Declaration on the use of artificial intelligence. The authors declare that no generative artificial intelligence was used in the writing of this manuscript.

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