BULLOUS LICHEN PLANUS IN A 5-YEAR-OLD CHILD: AN UNCOMMON PEDIATRIC VARIANT

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Abbreviation **LP** = lichen planus.

Case report. A 5-year-old boy, with no significant medical history, presented with a pruritic cutaneous eruption on the elbows. There was no history of recent infections, vaccinations, or drug exposure, nor a family history of lichen planus (LP). About one month after the onset of the eruption, painful tense blisters appeared on the palms and soles, severely impairing his ability to walk and use his hands.

Physical examination revealed multiple tense bullae on the palmar and plantar surfaces, some containing turbid fluid and arising on an erythematous base (Fig. 1). In addition, numerous flat, polygonal, violaceous, symmetric papules measuring a few millimeters in diameter coalesced into plaques predominantly over the elbows and knees. Examination of the lips revealed cheilitis with a delicate whitish reticular pattern. There was no scalp or nail involvement, and no other mucosal lesions were identified.

Dermoscopic examination of the keratotic papules revealed characteristic Wickham striae. A similar whitish "fern-leaf" pattern was observed on the lips (Fig. 2), further supporting the diagnosis of lichen.

Upon presentation, the appearance of palmoplantar vesicles had initially raised suspicion of a vesicular bullous eczematous eruption, particularly dyshidrotic eczema. However, the presence of violaceous keratotic papules on the elbows and knees, together with characteristic Wickham striae on dermoscopy, strongly directed the diagnosis toward lichen planus. Nevertheless, the associated tense bullae broadened the differential diagnosis to include other subepidermal vesiculobullous disorders of childhood.

Routine laboratory tests showed no abnormalities. HBsAg and anti-HCV antibodies were negative. Two skin biopsies were performed to characterize the different morphological components of the eruption. Histopathological examination of a keratotic papule from the elbow showed orthokeratotic hyperkeratosis, acanthosis, basal cell vacuolization with apoptotic keratinocytes, and a band-like dense inflammatory infiltrate at the dermal-epidermal junction, confirming lichen planus. A second biopsy obtained from a bulla demonstrated extensive subepidermal cleavage; the bulla cavity contained lymphocytes, melanophages, and eosinophils, the roof showed focal ischemic necrosis, and the underlying dermis contained a predominantly lymphohistiocytic infiltrate (Fig. 3).

The presence of a tense subepidermal bulla with eosinophils prompted the consideration of lichen planus pemphigoides as a further differential diagnosis. Direct immunofluorescence on perilesional skin was therefore performed, which was negative for IgG, IgA, IgM, and C3, with no linear deposition of immunoreagents along the basement membrane zone.

Overall, the clinical morphology, Wickham striae on dermoscopy, characteristic lichenoid histopathology, subepidermal blister formation, and absence of linear immunoreagents in the basement membrane zone confirmed the diagnosis of bullous lichen planus.

The child was treated with topical corticosteroids alone, tapered gradually over three months. Blister formation ceased, and the lichenoid lesions progressively regressed. Complete clinical resolution



Fig. 1



Fig. 2

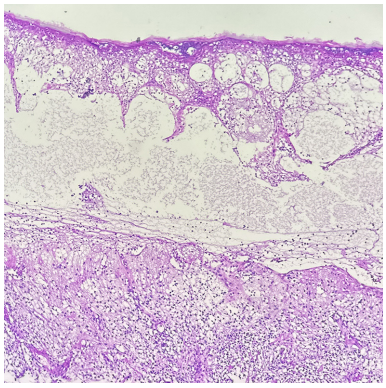


Fig. 3

Fig. 1, 2, 3: Bullous lichen planus with palmar plantar blisters (Fig. 1). Dermoscopy (Fig. 2) of the lip showing a fern-leaf whitish reticular pattern. The histological examination of a blister shows (Fig. 3, H&E x10) dermoepidermal cleavage containing lymphocytes, melanophages and eosinophils.

was achieved within six months, without recurrence of blistering and without the need for systemic therapy.

Discussion. Lichen planus is rare in the pediatric age and encompasses a broad clinical spectrum. Classic cutaneous LP is the predominant phenotype, followed by the eruptive, hypertrophic, and linear variants, whereas mucosal, nail, and scalp involvement occurs less frequently (1, 2, 7). Bullous LP lies at the rarest end of this spectrum and has been documented in children primarily through single case reports (3–6).

The clinical presentation can be misleading when blister formation dominates the eruption. In our patient, the most striking finding consisted of tense and painful blisters on the palms and soles, which initially raised suspicion of a vesicular bullous eczematous eruption, such as dyshidrotic eczema. Examination of the entire skin surface proved decisive: the violaceous keratotic papules on the elbows and knees, Wickham striae on dermoscopy, and the reticular pattern on the lips indicated an underlying lichenoid disorder. This illustrates an important point: when bullae constitute the most conspicuous manifestation, the most diagnostically suggestive lesions may be located far from the sites of blister formation. Palmoplantar involvement is in itself a recognized, albeit uncommon, manifestation of childhood LP (8) and should be considered part of a broader clinical spectrum rather than a defining feature of bullous LP.

Bullous eruptions associated with LP have historically been subdivided into two entities. In true bullous LP, blister formation results directly from severe lichenoid interface damage: cytotoxic T-lymphocyte-mediated damage to basal keratinocytes produces vacuolization and apoptosis, progressively weakening the dermo-epidermal junction. Small clefts develop and, when basal damage becomes sufficiently intense, coalesce into a distinct subepidermal bulla, typically within or adjacent to preexisting lichenoid lesions (5, 6, 9). By contrast, lichen planus pemphigoides is an autoimmune subepidermal bullous dermatosis in which autoantibodies are directed against basement membrane zone antigens, particularly BP180/type XVII collagen, and bullae may arise either on clinically normal skin or on lichenoid lesions (9, 10).

In our patient, performing biopsies of two morphologically distinct lesions proved particularly informative: the biopsy of the papule established the underlying lichenoid disease, whereas the biopsy

of the bulla documented the mechanism of blister formation itself. The presence of eosinophils within the bulla cavity is noteworthy, as an eosinophil-rich subepidermal bulla commonly raises suspicion of an autoimmune bullous dermatosis. Eosinophils should not, however, be interpreted in isolation when the clinical and histopathological picture strongly supports LP.

Direct immunofluorescence provides decisive complementary information. In lichen planus pemphigoides, it typically demonstrates linear deposition of IgG and/or C3 along the basement membrane zone, whereas bullous LP lacks this continuous linear deposition (6, 9, 10). In our patient, the absence of any linear deposition of IgG, IgA, IgM, or C3, in the context of clinically and histologically characteristic LP, confirmed the diagnosis of bullous LP. Nevertheless, because anti-BP180 serology and indirect immunofluorescence on salt-split skin were not available, the diagnosis relied on the overall clinicopathological and immunopathological picture rather than on the negative direct immunofluorescence study alone. Other subepidermal blistering disorders of childhood—namely linear IgA dermatosis and, less commonly, bullous pemphigoides and epidermolysis bullosa acquisita—were considered, but the characteristic lichenoid papules and their histopathology provided a strong diagnostic clue toward LP.

Pediatric bullous LP remains poorly documented, and treatment is not standardized. Reported approaches include topical and systemic corticosteroids, phototherapy, dapsone, and retinoids depending on disease extent and severity (3–6). More broadly, in childhood LP, topical corticosteroids remain the standard first-line therapy for localized disease, whereas systemic treatment or phototherapy is reserved for extensive or refractory presentations (1, 7, 11).

Of particular relevance is the case described by Arsov et al., who reported a 5-year-old boy with disseminated violaceous papules with a flat surface and blisters developing on the hands and feet, featuring characteristic LP histology, subepidermal bullae, and negative direct immunofluorescence. That patient had widespread disease and required systemic prednisone, narrowband UVB phototherapy, and topical agents, achieving remission after approximately 2.5 months (5). Our patient, by contrast, presented with more limited disease and achieved complete remission with topical corticosteroids alone, tapered over three months, with resolution at six months and no recurrence. The similar age of the two children confirms that bullous LP can occur in early childhood, whereas the differing course illustrates that the striking appearance of tense bullae does not necessarily imply the need for systemic therapy: an initial topical approach may be sufficient in limited disease, reserving systemic treatment for extensive, rapidly progressive, or refractory presentations.

The long-term prognosis of childhood LP is not fully characterized, though most children with classic LP recover within approximately six months, often with residual post-inflammatory pigmentation (2). Specific data for bullous LP are even scarcer; the complete healing and absence of recurrence in our patient are reassuring, but long-term monitoring remains appropriate.

Conclusion. Bullous lichen planus is a rare manifestation of an already uncommon pediatric inflammatory dermatosis. Its recognition depends on identifying the underlying lichenoid process, even when blister formation dominates the clinical picture. Careful clinical examination, dermoscopy, histopathological evaluation of representative lichenoid and bullous lesions, and direct immunofluorescence provide complementary diagnostic information and enable the distinction of true bullous LP from lichen planus pemphigoides.

Conflicts of interest

The authors declare that they have no conflicts of interest.

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