

Exanthematous lichen planus in a child and *Mycoplasma pneumoniae*: a case report and literature review

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Abstract

Lichen planus (LP) is a chronic inflammatory disease of the skin and mucous membranes. The disease usually affects adults and is only rarely encountered in children. Typically, skin lesions include violaceous, polygonal, flat papules and plaques, affecting predilection sites such as the wrists, ankles, and lower back. However, clinical presentation can be heterogeneous and is often atypical in children. Various precipitating factors are known to play an important role in the pathogenesis of lichen planus, some of which may also be coincidental. LP occurring after an infection with *Mycoplasma pneumoniae* is a rare occurrence. We present the case of a 13-year-old boy with pruritic papular skin lesions on the extremities and trunk. In view of the clinical and histopathological findings, LP exanthematicus was diagnosed. To the best of our knowledge, our case is the first of pediatric exanthematous LP after *M. pneumoniae* infection that has been reported so far.

Keywords: lichen planus, children, exanthematous lichen planus, *Mycoplasma pneumoniae*

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Introduction

Lichen planus (LP) is an uncommon chronic inflammatory mucocutaneous disease first described by Erasmus Wilson in 1869 (1). According to limited data, cutaneous lichen planus is estimated to occur in less than 1% of the population, with the highest incidence between ages 30 and 60 (2). It is only rarely encountered in children, constituting only 1% to 4% of all cases (3, 4). Although LP is usually sporadic, a familial form has been reported in 1% to 4.3% of childhood LP series (5).

Clinically, LP has a heterogeneous presentation, with varying cutaneous, mucosal, and appendageal manifestations. Classic LP presents with pruritic, violaceous, polygonal, flat-topped papules with Wickham's striae predominantly over the flexor aspects of the limbs and mucosae (3). In addition to the classic presentation of LP, multiple other clinical presentations of cutaneous disease have been described. LP variants include hypertrophic, actinic, annular, atrophic, pigmentosus, inverse, bullous, and ulcerative, as well as palmoplantar and perforating. LP can also occur on the scalp as lichen planopilaris. Other manifestations may present as nail, ocular, genital, esophageal, or otic variants. The most common pattern in pediatric patients is the classic form (3, 6). Generalized LP (exanthematous LP, eruptive LP) presents with violaceous flat-topped papules and plaques developing into a generalized infiltrated exanthem of the skin. The eruptive form is seen in 16% of pediatric patients with LP. Those patients are more likely to have a more severe disease course (3).

The etiology of lichen planus is not fully understood. An immune-mediated mechanism involving activated T cells, particularly CD8+ T cells directed against basal keratinocytes, has been proposed (7). Various precipitating factors are known to play an important role in the pathogenesis of LP. The association of hepatitis C virus with LP is controversial, although numerous studies found a statistically significant association between them (8, 9). Cutaneous LP occurrence has rarely been linked to other infections. *Mycoplasma pneumoniae* is an obligate human pathogen

that mostly causes community-acquired pneumonia, although it can be associated with a wide variety of extrapulmonary manifestations (10). It is very rarely involved in disorders of the skin, especially in children. Mucocutaneous manifestations following *M. pneumoniae* infection (MPI) are mostly the result of immune-mediated damage caused by cross-reacting antibodies or immune complexes. It can present as erythema nodosum, cutaneous leukocytoclastic vasculitis, erythema multiforme, Stevens–Johnson syndrome (SJS), toxic epidermal necrolysis (TEN), Fuchs syndrome, or subcorneal pustular dermatosis (11). Rarely, LP can occur after MPI (12–15) and presents as a classic form of LP or as a unilateral widespread form of LP (12, 13).

Case report

We describe the case of an otherwise healthy 13-year-old boy, who presented to our dermatology department due to a 2-month history of an intensely itchy rash. He had no history of concomitant drug intake, vaccination, or other putative trigger factors. Apart from a slightly sore throat before the onset of skin changes, the history for preceding infections was negative. The family history was negative for dermatologic diseases.

The clinical examination revealed partially coalescing polygonal violaceous to hyperpigmented papules and plaques covered with whitish-gray scale. These lesions were distributed symmetrically, affecting the upper (Fig. 1) and lower extremities (Fig. 2) as well as large areas of the trunk (Fig. 3). The scalp, mucous membranes, and nails were not involved. Physical examination revealed normal vital signs, and his pulmonary and cardiovascular examination were also unremarkable.

A histopathological examination performed on a lesion from the forearm showed hyperkeratosis without parakeratosis and wedge-shaped hypergranulosis in the epidermis, vacuolization of the basal layer, band-like lymphocytic infiltrate at the dermal-epidermal junction, presence of Civatte bodies, and pigment incontinence in the papillary dermis (Fig. 4). Direct immunofluorescence

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Figure 1 | Exanthematous lichen planus. Diffusely scattered violaceous papules with overlying silver scale coalescing into plaques on the upper extremities.



Figure 2 | Exanthematous lichen planus. Diffusely scattered violaceous to hyperpigmented papules coalescing into plaques on the lower extremities.



Figure 3 | Exanthematous lichen planus. Diffusely scattered violaceous to hyperpigmented papules with overlying silver scale coalescing into plaques on the trunk.

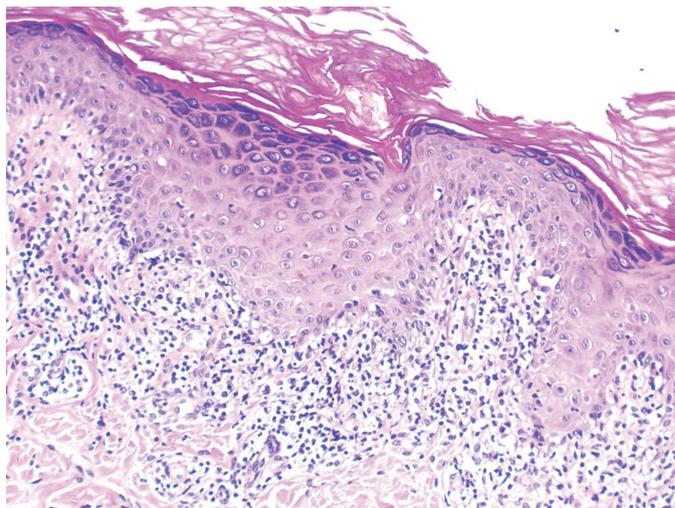


Figure 4 | Histopathological features of lichen planus.

demonstrated complement and immunoglobulins (mostly IgM) and linear fibrin deposition along the dermal–epidermal junction. In light of the clinical presentation, these findings were consistent with pediatric eruptive generalized lichen planus. Initial laboratory examination showed a normal complete blood count, C-reactive protein level, and blood chemistry panel. Erythrocyte sedimentation rate was slightly increased (18, positive > 15 mm/h). Urinalysis and results of blood test for hepatitis B and C viruses, Epstein–Barr virus (EBV), and thyroid function were within the normal range. Immunoserological evaluation of HEP 2 test, ENA panel, and complement studies showed no abnormalities. *Mycoplasma*-specific IgM antibody titer was elevated (26.4 U/ml, positive > 17 U/ml).

The skin lesions were treated with a moderate potency topical corticosteroid ointment with a good therapeutic effect. Because of a slightly sore throat before the onset of skin changes and elevated specific IgM *Mycoplasma* antibody titer, we started therapy with peroral azithromycin 500 mg daily for 3 days. The patient responded very well to local and systemic therapy and showed a significant improvement of the clinical picture and pruritus at the follow-up visit 2 weeks later.

Discussion

Lichen planus is an uncommon disorder of unknown cause that most commonly affects middle-aged adults. It is rarely seen in children, and the clinical presentation is often atypical. The classic presentation of cutaneous LP is a papulosquamous eruption characterized by the development of flat-topped violaceous papules on the skin. Often, the clinical manifestations are described as the four P's: pruritic, purple, polygonal, and papules or plaques. In addition, a network of fine, reticular white lines called Wickham striae may be seen within the skin and mucosal lesions (16). Rarely, Blaschkoid (17), zosteriform (18), and inverse distributions as well as generalized involvement have been observed. LP is said to be less common and more severe in children than in adults. The eruptive form (generalized exanthematous LP) is the second most common type of LP seen in children, as was our case. The eruptive form usually has a more severe disease course (3). The pruritus associated with cutaneous LP is often intense, as in our patient.

Asymptomatic eruptions are rare. Patients with lichen planus, similar to those with psoriasis, may exhibit the Koebner reaction, which usually occurs as a result of scratching. In exanthematous LP, the face is almost never involved. In our patient, the scalp, mucous membranes, and nails were also not involved.

Considering the great number of LP variants, differential diagnoses are abundant and include psoriasis, atopic dermatitis, lichen simplex chronicus, pityriasis lichenoides, lichen sclerosus, pityriasis rosea, chronic graft-versus-host disease, and lichenoid drug eruptions (19).

In many cases, the diagnosis of LP can be established based on clinical findings. In uncertain cases, skin biopsy is recommended. In our patient, a punch biopsy was performed due to the generalized clinical presentation. Histopathology demonstrated a lichenoid dermatitis consistent with lichen planus. In view of clinical and histopathological findings, the diagnosis of pediatric eruptive generalized lichen planus was established.

There is evidence that CD8+ T cells are strongly implicated in the pathogenesis of LP (7). Upregulation of intercellular adhesion molecule-1 (ICAM-1) and cytokines associated with a Th1 immune response, such as interferon (IFN)-gamma, tumor necrosis factor (TNF)-alpha, interleukin (IL)-1 alpha, IL-6, and IL-8, may also play a role in the pathogenesis of LP (20–22).

Various authors have reported different disease associations, some of which may be coincidental. These include active hepatitis, vaccination, depression, anxiety, and dyslipidemia (23). In addition, several autoimmune diseases have been linked to LP, including vitiligo, Hashimoto thyroiditis, myasthenia gravis, and alopecia areata, as well as atopic dermatitis, lichen nitidus, metal allergy, graft-versus-host disease, and several drugs (3, 24). Triggering factors that are frequently described were all excluded in our patient.

Cutaneous LP has been rarely linked to infections (with the exception of hepatitis B and C infections), and especially to *Mycoplasma* infection. Whereas pneumonia and other respiratory presentations are caused by direct infection with *Mycoplasma*, the extrapulmonary symptoms are mediated by one of the following three mechanisms: direct infection (e.g., pericarditis, arthritis, aseptic meningitis, encephalitis, or myelitis), immune-mediated damage caused by cross-reacting antibodies or immune complexes (e.g., hemolytic anemia, conjunctivitis, iritis, uveitis, or myocarditis), and vascular occlusion either by direct infection with bacteria or by vasculitis (aortic thrombus, pancreatitis, splenic infarct, pul-

monary embolism, priapism, renal artery embolism, or thalamic necrosis) (10, 11). Skin manifestations as extrapulmonary features of MPI are often presented as an exanthematous and nonbullous rash (10). Manifestations such as erythema nodosum, erythema multiforme, SJS, TEN, Fuch's syndrome, and cutaneous leukocytoclastic vasculitis are less frequently noted (11). The onset of these extrapulmonary manifestations is reported to be quite variable in relation to pulmonary signs and symptoms. Occasionally, they are reported even in the absence of any respiratory symptoms. Rarely, LP can occur after MPI (12–15). With regard to the literature published so far, it presents as a classic form of LP or as a unilateral widespread form of LP (12, 13). To the best of our knowledge, pediatric exanthematous LP following MPI has not been described so far.

A slightly sore throat before the onset of skin changes, elevated *M. pneumoniae* antibody IgM titer, and skin changes in our patient suggests that the patient's LP could have been provoked by *Mycoplasma* infection, most likely as an immunological reaction reflecting T-cell attack toward the microorganism. The mechanisms behind these skin lesions are not completely understood, although evidence mostly suggests immune-mediated damage and an autoimmune response (10, 25). In this context, given that both CD4+ T-helper (Th)1 and CD8+ cytotoxic T (Tc)1 cells are deeply involved in the pathogenesis of LP, the activated Th1/Tc1 cells in association with the microorganism eradication might have induced the lichenoid eruption in our case (12, 13).

A major limitation of this case report is the absence of convalescent titers and PCR testing. IgM testing by itself has a specificity of only 92% compared to IgM combined with *Mycoplasma* PCR, which has a specificity of 100% and sensitivity of 98% (26). However, our patient tested negative for other possible infectious causes, such as EBV and hepatitis viruses, and only the *Mycoplasma* IgM was elevated. The positive IgM titers (in the absence of convalescent titers) suggest either acute or very recent infection (26). IgM testing, although highly sensitive (81%), can remain positive for a few weeks after an acute infection, which we assume was the case in our patient (26, 27).

Conclusions

To the best of our knowledge, this is the first case of eruptive LP in the pediatric population following MPI. Our case, along with previous reports, emphasizes the importance of considering *Mycoplasma* infection in children presenting with lichenoid lesions.

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