

CASE REPORT

Acute generalized lichen planus with diffuse palmoplantar involvement in a 4-year old child: A case report and review

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ABSTRACT

Lichen planus (LP), is a unique chronic inflammatory disease affecting the skin, mucous membranes, nails, palms, soles, and scalp with varying clinical presentations. Childhood LP is rare constituting 1-4% of all LP cases seen as reported in various studies. Childhood LP is reported to be more common among Asian and Middle East region, compared to Europe, North America or Africa. A number of treatment options are available to treat a patient with LP. These include topical steroids and topical calcineurin inhibitors (tacrolimus and pimecrolimus), oral steroids in daily, or weekly pulse form, methotrexate, azathioprine, mycophenolate mofetil, dapsone and thalidomide and retinoids (acitretin, etretinate), phototherapy (UVB and PUVA). We report here a 4-year-old child with generalized eruptive lichen planus having diffuse palmoplantar involvement that responded very well to oral corticosteroids with no side effects.

CASE REPORT

A 4-year-old boy was brought by his parents with history of sudden onset of a severely itchy generalized skin eruption of violaceous to dark colored papules and plaques involving his trunk and limbs of 1 month's duration. There was no history of any constitutional or systemic symptoms, drug intake or recent vaccination prior to the appearance of the lesions. The child had recently arrived from India for the first time into Kuwait. There was no family history of similar lesions in the family. The child had been treated with topical steroid cream hydrocortisone 1% and oral antihistamines by the general practitioner with no relief in his signs and symptoms. General physical examination revealed a healthy male child, visibly distressed from scratching continuously, with a generalized skin eruption consisting of violaceous to dark colored, discrete as well as coalescing papules and plaques with overlying whitish streaky scales (Wick-

ham's striae) on his trunk (Fig. 1,2), and limbs (Fig. 3). The palms and soles were affected diffusely by skin colored to yellowish hyperkeratotic papules (Fig. 4,5,6) giving appearance of a diffuse keratoderma. Face, nails, scalp, oral, genital and eye mucosae were not involved. A clinical diagnoses of generalized lichen planus was made.



Fig. 1 Violaceous papules and plaques on trunk and limbs of a 4-year old child.

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Fig. 2 Violaceous papules and plaques on anterior aspect of trunk and limbs of a 4-year old child.



Fig. 3 Violaceous papules and plaques on the back of trunk and limbs of a 4-year old child.



Fig. 4 Violaceous flat papules on dorsal aspect of the both hands.



Fig. 5 Diffuse involvement of yellowish hyperkeratotic papules of both palms producing keratoderma like appearance.

His routine base line investigations including CBC, ESR, blood sugar, LFT, RFT and G6PD levels were all normal. Serology for HBV and HCV was negative. A skin biopsy from a lesion on the back showed features of Lichen Planus (Fig. 7,8).

The child was treated with oral syrup dexamethasone (0.5 mg/5 ml) three times a day with topical hydrocortisone (1.0%) cream application twice a day, syrup chlorpheniramine 2.5 ml three times a day and emollient cream application twice a day. Within 2-weeks the severe itching had stopped, and most of the lesions flattened leaving behind dark hyperpigmentation. The dose of the oral steroid was tapered to 0.5 mg/5ml to twice a day for next 2 weeks and then then stopped over next six weeks. There had been no fresh crops and all the lesions had flattened completely leaving hyperpigmentation. There were no side effects. The child gained 2 kg of weight during the treatment period. The child is being followed every 4 weeks for any recurrence or side effects.

DISCUSSION

Lichen planus (LP), is a unique chronic inflammatory disease affecting the skin, mucous membranes, nails, palms, soles, and scalp with varying clinical presentations. Classic LP is characterized by polygonal, purple, pruritic, flat papules and plaques occurring over the flexor/volar aspects of extremities. A number of variants are recognized according to the site and morphology of the lesions. These include lichen planus ruber, hypertrophic, annular, atrophic, lichen planus pigmentosus, exanthematous/generalized, linear, oral, bullous and lichen planus pemphigoides.¹



Fig. 6 Coalescing yellow hyperkeratotic papules on both the soles (less extensive than the palms).



Fig. 7 H&E showing features typical of lichen planus.

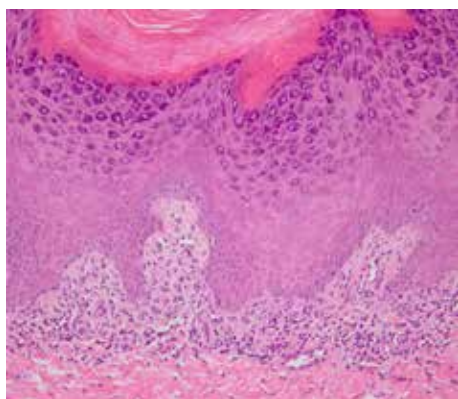


Fig. 8 Close up view of H & E picture of LP in a child.

Histologically, a dense, band-like lymphocytic infiltrate is seen underlying an acanthotic epidermis with hypergranulosis, apoptosis, and liquefactive degeneration of the basal cell layer. LP is considered to be the prototype of all lichenoid dermatoses. The exact etiology of LP is not known but it is considered to be an autoimmune disease although the triggering antigen is not yet recognized. The disorder has been asso-

ciated with multiple environmental exposures, including viral infections, medications, vaccinations and dental restorative materials.²

LP is a common dermatosis in adults constituting 1.0 to 2.5% of all new patients seen in dermatology out patient departments, although exact incidence in general population is not known.² Childhood LP is rare constituting 1-4% of all LP cases seen as reported in various studies.³ Childhood LP is reported to be more common among Asian and Middle East region,⁴⁻⁹ compared to Europe,¹⁰⁻¹¹ North America¹² or Africa.¹³

The average age of LP is around 7 years from different series with age range of 2 to 17 years. Although all the clinical types of LP seen in children are similar to those seen in adults, there are differences in incidence of various subtypes. Linear, and generalized, eruptive or exanthematous types are more commonly seen in children than in adults.¹⁴ Mucosal involvement although seen in children is much less common compared to adults. Recently few series (6 and 8 patients each) of oral LP among children (age range: 7-17 years) have been reported.¹⁵⁻¹⁶ Hyperkeratotic palmoplantar involvement is very rare among pediatric cases of LP.^{4,17} Lesions on the palms and soles do not show the characteristic purple color and Wickham's striae and instead appear yellowish because of hyperkeratosis that masks the hypergranulosis. Lichen planus pemphigoides (LPP) is a very rare variant of LP among children.¹⁸ Other rare variants of LP reported in children are actinic lichen planus¹⁹ and lichen planopilaris.²⁰ The clinical, histopathological and immunofluorescence features of LPP are similar in children and adults.

A number of treatment options are available to

treat a patient with LP.^{21,22} These include topical steroids and topical calcineurin inhibitors (tacrolimus and pimecrolimus),²³ oral steroids in daily,²⁴ or weekly pulse form,^{25,26} methotrexate,²⁷ azathioprine,²⁸ mycophenolate mofetil,²⁹ dapsone³⁰ and thalidomide³¹ and retinoids (acitretin, etretinate), phototherapy (UVB and PUVA).²⁰ The points to be considered in children are age of the child, the extent and number of lesions and the duration of the disease. Localized LP or LP with few scattered lesions is easily treated by potent topical steroids such as fluorinated steroids or clobetasol propionate. There is a case report where a child with generalized exanthematous LP was treated successfully with potent topical steroids. Generalized LP is treated with oral steroids 2.0 mg betamethasone or 20 mg prednisolone equivalent daily over 4 weeks when control is usually achieved followed by tapering by 0.5 mg every month as described by Pasricha.²² Oral steroids in weekly pulsed (5 mg betamethasone or dexamethasone approximately equivalent of 50 mg Prednisolone on 2 consecutive days every week for 4 to 6 weeks and then tapered over next few months depending upon clinical response avoids long term side effects of daily steroid use.^{23,24} Other systemic treatments may be needed in severe, generalized, erosive or resistant cases. Childhood actinic LP has been treated successfully with antimalarial hydroxychloroquine in 4 children.³⁰ Methotrexate, azathioprine, acitretin and mycophenolate mofetil are useful agents although requiring thorough pre-treatment screening and regular clinical and laboratory follow up especially in children. Generalized eruptive LP in children usually resolves rapidly within 6 to 8 weeks. Post inflam-

matory hyperpigmentation takes few months to subside.

CONCLUSION

We report here a 4-year-old child with generalized eruptive lichen planus having diffuse palmoplantar involvement that responded very well to oral corticosteroids with no side effects. A brief review of lichen planus in children with treatment options is presented.

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