

A Case of Eosinophilic Pustular Folliculitis

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We report a case of eosinophilic pustular folliculitis in a 6-month-old male infant who had pruritic, tiny, erythematous papulopustules on his scalp and eosinophilia in the peripheral blood. Histopathologic examination revealed an acute follicular and perifollicular inflammatory infiltrate with abundant eosinophils. The patient responded to systemic and topical corticosteroid and dapsone. (Ann Dermatol 13(1) 52~54, 2001).

Key Words : Eosinophilic pustular folliculitis

Eosinophilic pustular folliculitis (EPF) is a cutaneous inflammatory follicular disorder of unknown etiology. It was first described in three Japanese men by Ofuji et al.¹ in 1970. The disease is characterized by recurrent crops of pruritic follicular papulopustules on the scalp, trunk, and extremities, sparing the hands, feet, and mucous membranes. Microscopically, there is degeneration and disintegration of follicular epithelial cells resulting in spongiotic vesicle formation in the outer root sheath, associated with infiltration of eosinophils and some mononuclear cells².

We report herein a case of EPF, which developed in infancy and well controlled by dapsone.

CASE REPORT

A 6-month-old male infant, weighing 7-kilogram, visited our clinic because of a pruritic papulopustular eruption on the scalp for one month.

Other family and personal history were irrelevant.

On physical examination, clusters of 1- to 2-mm sized pruritic papulopustules and crusted lesions were present on the scalp (Fig. 1). Direct fresh KOH examination did not reveal fungal structures. Fungal and bacterial cultures of the pustules yielded no organisms. His peripheral white blood cell count was 10,100/BB with 25% eosinophils. However, no abnormalities were seen in liver and renal function test, urine analysis, stool examination and c-reactive protein.

A biopsy specimen taken from the lesion revealed edema, spongiosis, destruction of follicle by infiltration of lymphocytes and numerous eosinophils (Fig. 2), subcorneal pustule and dermal inflammatory infiltrate consisting of lymphocytes, neutrophils and numerous eosinophils (Fig. 3). A periodic acid-Schiff (PAS) stain for fungi gave negative results.

We treated him with an application of a topical corticosteroid ointment and administration of prednisolone, 0.5mg/kg/day, and antihistamine. Partial improvement was obtained with this therapy. Because of aggravation in the next month, dapsone, 2mg/kg/day, was begun with improvement. After this therapy, the lesions became completely clear and follow up eosinophil count revealed normal range. When the drug was discontinued, the rash did not recur over the following 6 months.

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Fig. 1. Clusters of 1- to 2-mm sized papulopustules and crusted lesions were present on the scalp.

DISCUSSION

Eosinophilic pustular folliculitis (EPF) is a cutaneous inflammatory follicular disorder of unknown etiology. It was first proposed as a distinct clinical entity in 1970 by Ofuji and coworkers¹ who described three patients with recurrent crops of pruritic follicular papulopustules. Lesions were distributed most commonly on the face, chest, back, and extensor surfaces of the upper arms. The hands, feet, and mucous membranes were spared.

The pathogenesis of EPF is obscure and most theories on the pathogenesis of EPF have evoked immunologic mechanisms in the initiation of lesions. Takematsu and coworkers identified an eosinophil chemotactic factor in stratum corneum extract from palmoplantar lesions of an adult with EPF^{2,5}. Autoantibodies directed against the intercellular substance of the lower epidermis⁶ or the cytoplasm of basal cells of the epidermis and the outer sheath of hair follicles have been proposed to be etiologic in EPF⁷. Sebaceous gland activity or hypersensitivity reaction followed by liberation of major basic protein have also been postulated to be etiologic factors^{5,9}.

Infants with EPF make up less than 10% of all cases reported. In infants, the lesions are most prominent in the scalp. Also, the classic annular and polycyclic appearance with centrifugal enlargement has been lacking in infants³. Microscopically, adults have shown an eosinophilic infiltrate that invades sebaceous glands and the outer root sheath of hair follicles, often leading to spongiosis in

Fig. 2. The biopsy specimen from the papule of the scalp showed edema, spongiosis, destruction of follicle by infiltration of lymphocytes and numerous eosinophils (H & E stain, $\times 100$).

Fig. 3. Subcorneal pustule and dermal inflammatory infiltrate consisting of lymphocytes, neutrophils and numerous eosinophils (H & E stain, $\times 100$).

the outer root sheath³. The eosinophilic infiltrate in most infants, however, has been perifollicular, without spongiosis in the outer root sheath⁴. The differences between EPF in infancy or children and adults appeared in the literatures^{3,4,8} are summarized in Table 1.

Laboratory investigations in EPF usually show normal results, except for the frequent leukocytosis

Table 1. Differences between EPF in infancy or children and adults appeared in the literatures^{3,4,8}.

Criteria	Infancy or Children	Adults
Papulopustules on the scalp	+	—
Urticarial papules, erythema multiforme-like lesions	+	—
Pruritus	++	+
Annular, polycyclic papulopustules	—	+
Involvement of seborrheic areas	—	+
Spongiosis in the outer root sheath of hair follicle	—	+
+, present		
—, absent		

with eosinophilia. Our patient demonstrated clinically EPF and leukocytosis was noted, with a white blood cell count of 10,100/mm³ with 25% eosinophils, without significant clinical manifestations of infections.

Histologically, there is degeneration and disintegration of follicular epithelial cells resulting in spongiotic vesicle formation in the outer root sheath, associated with infiltration of eosinophils and some mononuclear cells².

The differential diagnosis includes erythema toxicum neonatorum, infantile acropustulosis, pustular folliculitis, transient neonatal pustular melanosis, subcorneal pustular dermatosis, incontinentia pigmenti, dermatitis herpetiformis, insect bite, impetigo, and Kimura's disease must be considered⁸. The combination of the specific clinical, laboratory, and histologic features determines the diagnosis of EPF.

Response of EPF to therapy has been variable and no one specific treatment has surfaced as the therapy of choice. Therapeutic agents have been used in EPF including topical and oral steroid^{1,5}, dapsone⁵, oxyphenbutazone, aspirin, colchicine, minocycline⁹, isotretinoin¹⁰, and oral and topical indomethacin^{2,11}. In our patient, we treated him with an application of a topical corticosteroid ointment and administration of systemic steroid and antihistamine. Partial improvement was obtained with this therapy. Because of aggravation in the next month, dapsone, 2mg/kg/day, was begun with improvement. After this therapy, the lesions became completely clear and follow up eosinophil counts revealed normal range.

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