

# Three Cases of Eosinophilic Pustular Folliculitis Without Eosinophilia

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**We present three cases of eosinophilic pustular folliculitis without eosinophilia in a 19-year-old female, a 27-year-old male, and a 51-year-old male. Multiple erythematous plaques with papules and pustules were observed on the face in two patients and on the face, trunk, and extremities in the third patient. Histopathologic findings showed hair follicles and the sebaceous glands infiltrated with mixed inflammatory cells composed of eosinophils and neutrophils, characteristic of eosinophilic pustular folliculitis. However, peripheral blood eosinophilia was not present in any of our three cases. (Ann Dermatol 3:(1) 80-83, 1991)**

*Key Words:* Eosinophilia, Eosinophilic pustular folliculitis, Ofuji's disease

Eosinophilic pustular folliculitis (EPF), first described by Ofuji et al in 1970<sup>1</sup>, is a dermatosis characterized by remissions and exacerbations of erythematous plaques with follicular papules and pustules generally affecting the face, the trunk and the upper extremities. The typical histopathologic picture is that of an infiltrate of eosinophils and neutrophils within hair follicles and sebaceous glands. Eosinophilia and moderate leukocytosis in the peripheral blood is characteristic. The diagnosis is made on the basis of the distinctive clinical, histopathologic and laboratory findings.

Most cases of EPF have been reported in Japan and a few cases have been reported in Europe<sup>2</sup> and the United States. In Korea, four cases have been reported up to present<sup>3, 4, 5</sup>. We report here three cases of EPF which did not show the characteristic peripheral eosinophilia.

## REPORT OF CASES

### Case 1

A 19-year-old female presented with a 5 month history of erythematous plaques with papules and pustules localized to the face.

The cutaneous eruptions began on the left cheek as crops of small erythematous follicular papules and changed to pustules in a few days. As the papulopustules increased gradually in size and number, they expanded centrifugally to form annular plaques with new eruptions predominant at the periphery. The erythematous plaques were elevated with densely packed pustules at the periphery. The erythematous plaques were elevated with densely packed pustules at the periphery and had a tendency toward central clearing (Fig. 1). The lesions resolved with residual pigmentation and new lesions appeared in the contralateral area and chin. The patient was asymptomatic except for mild itching at the beginning of the severe exacerbation. She denied seasonal variation of the lesions or exacerbation by sunlight.

Laboratory studies showed an eosinophil count of 126/mm<sup>3</sup> with white blood cell count of 6,800/mm<sup>3</sup>.

Histopathologic examination of a biopsy from

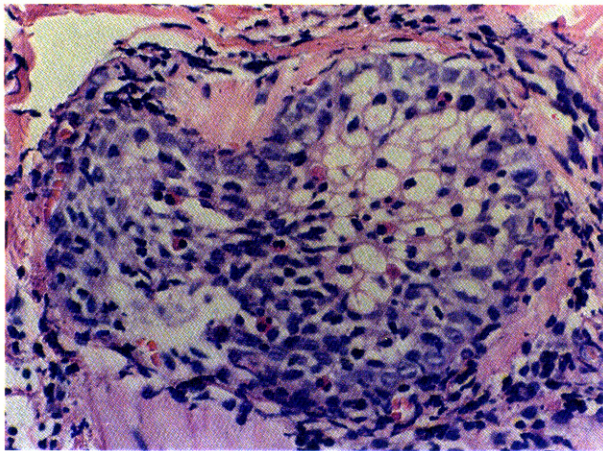
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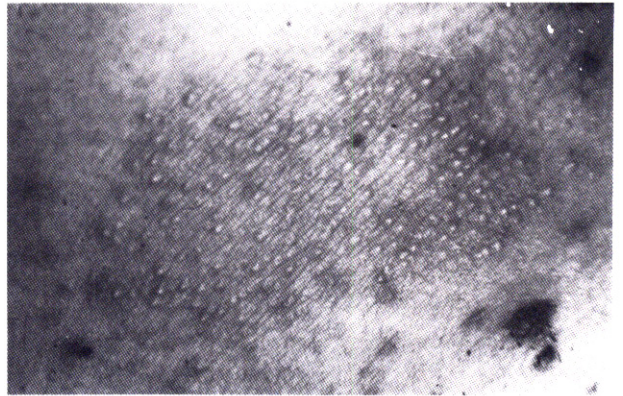
**Fig. 1.** (case 1). A fairly well defined indurated erythematous plaque studded with multiple papulopustules on the left cheek.



**Fig. 2.** (case 1). Sebaceous gland infiltrated with many eosinophils (H & E stain,  $\times 200$ ).

the left cheek revealed sebaceous glands infiltrated with mixed inflammatory cells composed of mainly eosinophils, and a few mononuclear cells (Fig. 2). No fungal hyphae were demonstrated on PAS stained material.

Since the clinical examination and histopathologic findings were consistent with the diagnosis of EPF, antihistamines, dapsone (50mg/day) and topical corticosteroid were prescribed. The skin lesions showed some improvement, but exacer-



**Fig. 3.** (case 2). Erythematous, annular plaque with pustules on the trunk.



**Fig. 4.** (case 3). Intraepidermal abscess filled with predominantly eosinophils and a few neutrophils (H & E stain,  $\times 100$ ).

bated after medication was discontinued.

## Case 2.

A 27-year-old male presented with an 84 month history of a pruritic eruption on the face, trunk and extremities.

The grouped, reddish papulopustules were present on erythematous patches localized to the trunk and extremities. As the papulopustules spread gradually to the periphery, they became sharply demarcated erythematous plaques, which attained a diameter of 5cm in a few days (Fig. 3). The lesions resolved with residual pigmentation. Subsequently, the similar lesions developed on the both cheeks. The lesions underwent remissions and exacerbations in spite antihistamine therapy.

Laboratory studies disclosed a white blood cell count of  $8,400/\text{mm}^3$  and an eosinophil count of  $134/\text{mm}^3$ .

The histopathology was similar to that of case 1.

There was no response to a multi drug regimen which included antihistamines, antibiotics, prednisolone (10-30mg/day), dapsone (100mg/day) and topical corticosteroid.

### Case 3.

A 51-year-old male presented with a 5 month history of erythematous plaques on the face.

He had noticed pruritic, reddish follicular erythematous papules on both cheeks and eyelids. The lesions changed to pustules and became crusted over time. As the papulopustules increased gradually in size and number, they expanded peripherally to form sharply demarcated erythematous plaques with central clearing. They resolved with residual pigmentation and new papulopustules developed in the hyperpigmented areas aside from the skin lesions, the patient had been in good health and was on no medication.

Laboratory studies showed a white blood count of  $8,100/\text{mm}^3$ , with an eosinophil count of  $45/\text{mm}^3$ .

The histopathology was similar to that of case 1.

In addition an intraepidermal abscess containing predominantly eosinophils and a few neutrophils was noted (Fig. 4.).

Antihistamine, dapsone (100-150mg/day), prednisolone (5-10mg/day) and topical corticosteroids were prescribed, with little improvement in the skin lesions.

## DISCUSSION

Eosinophilic pustular folliculitis (EPF) is a rare

condition. It was first described in 1965 as subcorneal pustular dermatosis, a follicular variant? by Ise and Ofuji<sup>6</sup>. Subsequently, Ofuji *et al*<sup>7</sup> reported three additional cases in 1970 and proposed "eosinophilic pustular folliculitis" as a new clinical entity. As the lesions were found not to be limited to the hair follicles, the terms "sterile eosinophile pustulosis" or "eosinophilic pustular dermatosis" were also proposed<sup>7,8</sup>.

Although this dermatosis occurs from infancy to old age, the peak incidence is in the third decade<sup>9</sup>, with a male to female ratio of patients is 4.8:1<sup>9</sup>. The main features of this dermatosis are recurrent pruritic, follicular and sterile papulopustules, 1 to 2 mm in diameter, which become confluent to form plaques. New lesions tend to appear at the edge of the plaques, while the older more central lesions resolve with some hyperpigmentation. The distribution is usually asymmetric, with a predilection for the face, trunk and extensor aspect of the upper extremities. Rarely, lesions of the scalp, neck, axillae, groin and legs have been described. Pustular lesions on the palms and soles occur in one fourth of the patients<sup>8</sup>, while mucous membranes, and the dorsum of hands and feet are generally spared. In infants, the scalp is the primary focus of involvement unlike the disease in adults, the lesions often do not form annular or polycyclic rings<sup>10</sup>.

The clinical presentation of EPF varies. Although there are other dermatoses presenting with follicular pustules, it is not difficult to diagnose EPF because of the distinctive histopathologic findings.

Blood eosinophilia is present in most patients<sup>1</sup>, however the eosinophil count may be within normal limits<sup>9</sup>. The peripheral eosinophil count in our cases, ranged from 45 to  $134/\text{mm}^3$  during active disease (normal range: 0-450/ $\text{mm}^3$ ).

**Table.** Summary of the cases

	Case 1	Case 2	Case 3
Age/Sex	19/F	27/M	51/M
Site	face	face, trunk and extremities	face
Duration (month)	5	84	5
WBC count ( $/\text{mm}^3$ )	6,800	8,400	8,100
Eosinophil count ( $/\text{mm}^3$ )	126	134	45

The pathogenesis of EPF is still unknown. Takematsu et al<sup>11)</sup> demonstrated that skin surface lipids from seborrheic areas of normal subjects and stratum corneum extracts from EPF lesions contained chemotactic factors for neutrophils and eosinophils. It is known that eosinophils release basic proteins which are cytotoxic to epithelium. However, the exact cause for the eosinophilic migration to hair follicles remains unclear. Cutler<sup>13)</sup> found a positive IgE RAST to the house dust mite *Dermatophagoides pteronyssinus* in a patient with EPF. Frentz et al<sup>14)</sup> suggested that EPF might occur as an early sign of infection with the Human Immunodeficiency Virus.

According to most authors the treatment of this dermatosis, with systemic or topical corticosteroids, dapsone<sup>15)</sup> or oxyphenbutazone, has varying results. It has been suggested that the effect of treatment may depend on the underlying cause of the disease in each patient. In our patients, these drug regimens resulted in some improvement or no response at all.

In summary, our 3 cases showed the typical skin manifestations and histopathologic findings of EPF without peripheral blood eosinophilia. We suggest, therefore, that the peripheral blood is not the source of the tissue eosinophilia in EPF and that an elevated eosinophil count in the peripheral blood is not essential in the diagnosis of EPF.

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