Eosinophilic Pustular Folliculitis (Ofuji Disease) Manifested as Pustules on the Palms and Soles

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Eosinophilic pustular folliculitis (EPF) is clinically characterized by eosinophilic follicular papulo-pustules that form annular configurations on the face, trunk, and extremities. We describe a case of a patient whose EPF manifested on the soles of the feet, an area that normally does not contain hair follicles. The patient experienced a dramatic therapeutic response to indomethacin.

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osinophilic pustular folliculitis (EPF) was first described by Ofuji et al¹ in 1970. EPF is characterized clinically by the recurrence of corpus pruritic follicular papulopustules that tend to form annular configurations on the face, trunk, and extremities. The characteristic histopathologic feature of EPF is an eosinophilic, leukocyte-rich folliculitis with neutrophils and mononuclear cells.

Before or after the follicular lesions appear on the face or extremities, pustules occasionally develop on the palm of the hand or sole of the foot, where hair follicles are normally nonexistent.^{2,3} On clinical examination, the pustules resemble pustulosis palmaris et plantaris (PPP). In previous reports in our hospital, these lesions were sometimes limited to the palm and sole. These cases were thought to be related to EPF and were subsequently diagnosed as EPF.

Histologic examination of the lesions of the sole and the palm in EPF reveals pustules infiltrated with eosinophilic leukocytes; the eosinophilic infiltration is not found in PPP. The histologic evidence led us



Figure 1. Many irregularly shaped pustules on the soles of the feet. Note the hyperkeratosis in the healing lesion.

to consider that the pustules on the palm and sole in EPF did not reflect a combination of EPF and PPP but were a symptom of EPF.

Because pustules in EPF occur in areas where hair follicles do not exist, some authors have proposed that the term *eosinophilic pustulosis* or *eosinophilic pustular dermatosis* would be more suitable for this entity.^{4,5}

We describe a case of EPF (Ofuji disease) that manifested as pustules on the palms and soles and summarize 6 similar cases reported at our hospital.

Case Report

A 33-year-old man presented with pustules on the soles of the feet. Four months later, the pustules had spread to involve the interdigital area of the foot, trunk, and extremities, and he was referred to our hospital.

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Visual examination of the soles revealed numerous pustules, each with a red halo, 1 to 3 mm, round to irregular, and slightly elevated (Figure 1). Many follicular papules the size of a half grain of rice were scattered within an egg-sized pigmentation area on the trunk and extremities. An annular configuration was not obvious (Figure 2).

Histologic examination of the pustules on the soles revealed subcorneal multilocular pustules, which were filled with neutrophils and eosinophilic leukocytes. Mild intracellular edema was observed around the pustules. Emigration by eosinophilic leukocytes to the epidermis and infiltration into the upper dermis also were detected (Figure 3). Histologic examination of trunk lesions showed spongiosis of the follicles infiltrated by lymphocytes and eosinophilic leukocytes (Figure 4).

A diagnosis of EPF was made based on typical clinical and histologic findings, and indomethacin treatment was prescribed. All eruptions cleared with oral administration of indomethacin 75 mg per day for 4 weeks. The eruption was subsequently controlled with oral administration of minocycline and antiallergic drugs for 5 months.

The Table summarizes the clinical histories of 6 male patients treated at our hospital who were diagnosed with EPF based on the appearance of typical eosinophilic pustules of the follicles on the face, trunk, and extremities.

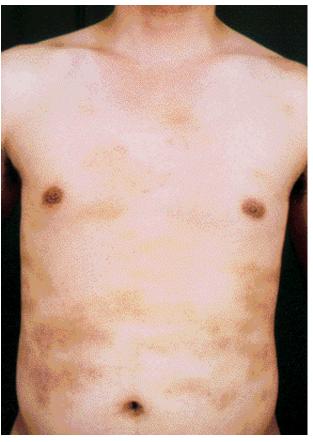


Figure 2. Red follicular papules the size of half grains of rice scattered over the trunk.

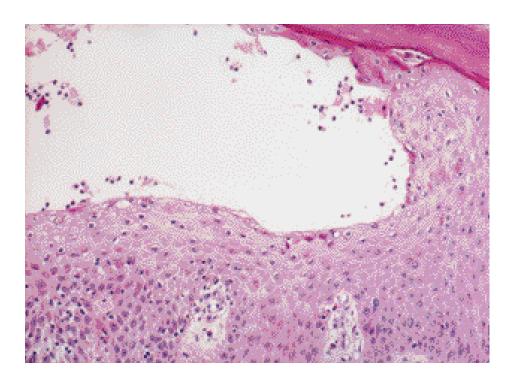


Figure 3. Subcorneal pustules on the sole filled with neutrophils and eosinophilic leukocytes (H&E, original magnification ×100).

Characteristics of Eosinophilic Pustular Folliculitis Manifested as Pustules of the Sole and Palm in 6 Male Patients

Age,	Initial Site	Distribution of Lesions	Histologic Features of Sole Lesions	Treatment	Treatment Response	Ap	ration After opearance of Initial Lesion
43	Sole, interdigital area of foot	Face, palm, interdigital area of hand	Psoriatic, intradermal infiltration with eosinophilic leukocytes	Steroid oint- ment and antiallergic drugs for 1 y	Not effective	Lost to follow-up	3 mo
66	Sole, palm, interdigital area of foot	None	Intraepithelial pustules infiltrated with eosinophilic leukocytes and neutrophils	Indomethacin 75 mg	Effective for 2 wk	Treatment with indo- methacin 50 mg for 3 y	
50	Sole	Face, extremities, palm	Not done	Indomethacin 75 mg	Effective for 4 wk	Controlled with indomethacin for 4 wk. Lost to follow-up	9 y
30	Sole	Face, trunk, extremities	Intraepithelial pustules infiltrated with eosinophilic leukocytes and neutrophils	Indomethacin 100 mg and minocycline 200 mg	Effective for 2 mo	No recurrence with indo- methacin and minocycline for 1 y	19 mo
45	Sole	Trunk, extremities	Not done	Naproxen 300 mg and diphenyl sulphone 75 mg	Effective for 1 mo	Controlled with naproxen and diphenyl sulphone for 5 y. Lost to follow-up	1 y
33	Sole	Trunk, extremities, interdigital area of foot	Intraepithelial pustules infiltrated with eosinophilic leukocytes and neutrophils	Indomethacin 75 mg and minocycline 100 mg	Effective for 1 mo	Controlled with minocycline for 5 mo. Lost to follow-up	1 4 mo

Comment

EPF lesions are not always confined to hair follicles. In addition to the typical locations on the face, trunk, and extremities, these lesions also have been reported to occur on the palm and the sole.² Aoyama and Tagami³ reported that the skin lesions began on the palms, soles, or both in 8% of their EPF patients (18 male and 3 female). In the Japanese literature, 18% of EPF patients exhibited

pustules on the sole and palm. In a few cases, all the pustules were palmoplantar lesions.⁶

The clinical appearance of the pustules on the sole and palm is similar to that of the lesions in PPP. If the lesions are limited to the sole and palm, either EPF or PPP is a possible diagnosis. The appearance of typical follicular lesions on the face, trunk, or extremities rules out the diagnosis of PPP and confirms the diagnosis of EPF. Saruta and

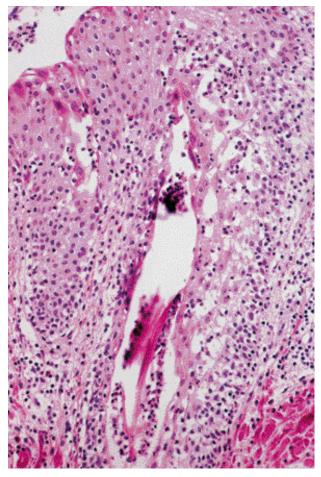


Figure 4. Spongiosis of the follicular keratinocytes infiltrated with lymphocytes and eosinophils on the trunk (H&E, original magnification ×100).

Nakamizo⁷ reported some clinical characteristics of EPF lesions that differentiate them from PPP. EPF lesions are larger than PPP lesions, and they tend to merge and display irregular margins. The surfaces of EPF lesions are erosive, and the pustules tend to be elevated and to invade interdigital areas. The clinical findings of the case reported here (pustules on the soles and palms) are similar to those of PPP.

Histologic examination of EPF lesions reveals multilocular, subcorneal, and intraepidermal pustules filled with numerous eosinophilic leukocytes and neutrophils. Other histologic features include perivascular infiltration of eosinophilic leukocytes into the upper dermis and their emigration to the epidermis. In contrast, subcorneal monolocular neutrophilic pustules are indicative of PPP. In our patient's case, the presence of subcorneal, intraepidermal, multilocular pustules containing numerous

eosinophilic leukocytes and neutrophils supported the diagnosis of EPF.

The causes of EPF remain unknown; some cases have been associated with fungal or bacterial infection or immunologic disorders,⁸ in particular, HIV infection.⁹ Three cases of EPF associated with drugs also have been described.¹⁰

Systemic treatment with the cyclooxygenase-inhibitor indomethacin and other anti-inflammatory drugs provides produces dramatic improvement in patients with EPE.^{11,12}

In conclusion, the palmoplantar lesions of EPF and PPP are difficult to differentiate by clinical differences alone; however, histologic examination can reveal the eosinophilic pustules in the epidermis that characterize EPF. The efficacy of oral indomethacin treatment is useful in the diagnosis of cases of EPF with palmoplantar lesions.

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