Received: 7 October 2020 | Revised: 22 October 2020 | Accepted: 29 October 2020

DOI: 10.1002/cia2.12147

CORRESPONDENCE

Journal of Cutaneous Immunology and Allergy

WILEY

A case of eosinophilic pustular folliculitis presenting as papuloerythroderma of Ofuji-like eruption

Dear Editor,

A 70-year-old Japanese man presented with a 7 month history of disseminated pruritic red-to-brown papules on the face, trunk, and extremities and scaly erythema on the soles (Figure 1A). He had no medical history. Previous treatments of minocycline 200 mg/day and betamethasone butyrate propionate ointment had been ineffective. A skin biopsy from a follicular red papule on the upper forearms showed inflammatory infiltrates of lymphocytes and eosinophils around the blood vessels in the upper dermis and in the hair follicles and sebaceous glands (Figure 1B-D). A laboratory examination revealed normal blood eosinophil counts (250/μL), elevated serum levels of immunoglobulin E (IgE; 269 IU/mL) and thymus and activation-regulated chemokine (TARC; 6127 pg/mL). Serological tests for HIV and human T-lymphotropic virus type I were negative. Under a diagnosis of eosinophilic pustular folliculitis (EPF), treatments with oral indomethacin farnesyl (IMF), which is a prodrug of indomethacin, and topical tacrolimus on the face were started. Topical tacrolimus was effective, but neither IMF nor topical corticosteroid improved the cutaneous symptoms. The papules gradually expanded and subsequently coalesced into large erythematous plaques, and several sterile pustules developed on the palms and soles (Figure 1E,F). The erythematous lesions spared

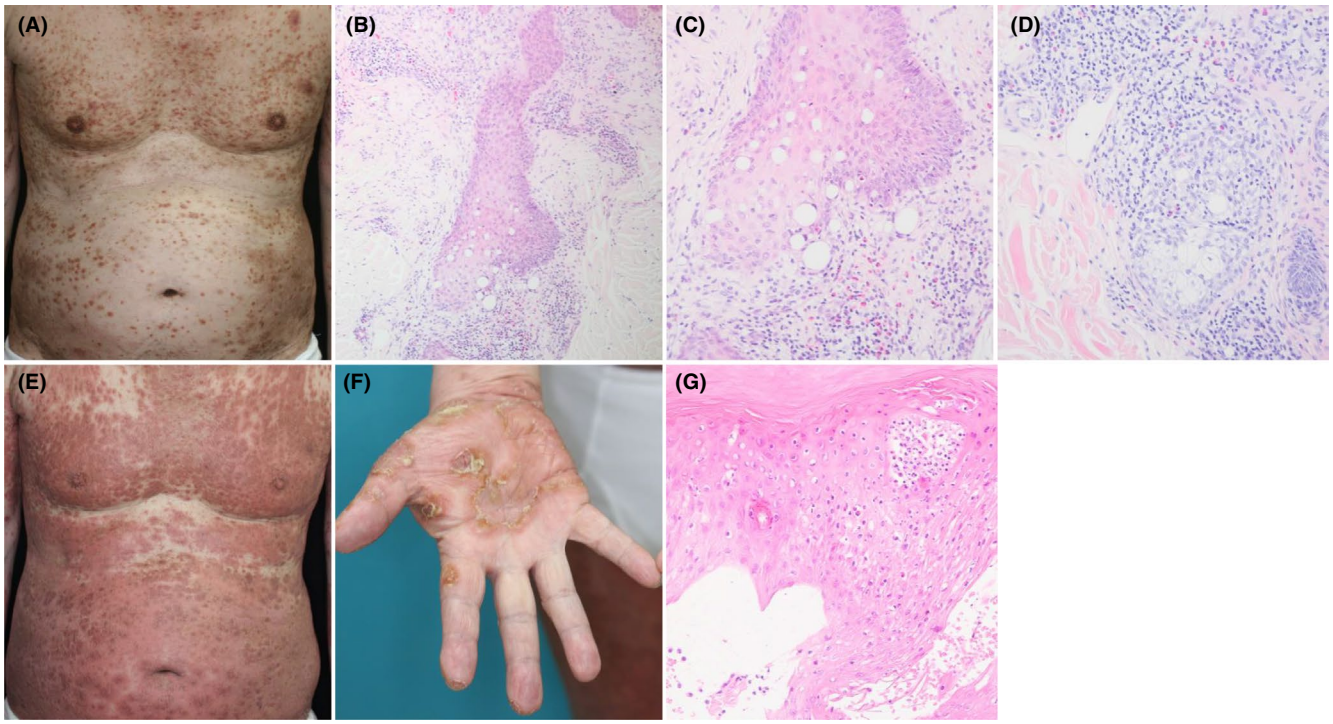

FIGURE 1 (A) Disseminated pruritic red-to-brown papules on the trunk. (B-D) Inflammatory infiltrates of lymphocytes and eosinophils around the blood vessels in the dermis (B) and in the hair follicles (C) and the sebaceous glands (D) (hematoxylin–eosin [HE], original magnifications, B ×100; C ×200; D ×200). (E) Erythroderma-like lesions which spared the skin folds (deck-chair sign) on the abdomen. (F) Pustular lesions on the right palm. (G) Multilocular pustules containing neutrophils and a few eosinophils (HE, original magnifications, ×200)

the skin folds on the abdomen and covered up to 80% of the total body surface area. The cutaneous presentation resembled that of papuloerythroderma of Ofuji (PED) except for the palmoplantar pustular lesions. A skin biopsy from a pustular lesion on the right palm showed multilocular pustules containing neutrophils and a few eosinophils (Figure 1G), which was compatible with EPF.[1] A laboratory examination again revealed increased blood eosinophil counts (2686/μL) and highly elevated serum levels of IgE (1809 IU/mL) and TARC (48,414 pg/mL).[2] Internal and hematological malignancy was excluded by examinations with computed tomography (CT) of the chest and abdomen, gastroscopy, colonoscopy, and bone marrow aspiration. Treatment with oral cyclosporine 200 mg/day significantly improved the cutaneous symptoms as well as the laboratory abnormalities. The cyclosporine dose was gradually tapered and discontinued without recurrence of cutaneous symptoms.

The diagnosis of EPF is sometimes challenging, as EPF may show atypical clinical presentations or share clinical appearances or histopathological findings with other diseases.[2,3] The high response rates of oral indomethacin therapy can facilitate the diagnosis of EPF, although the efficiency of IMF is relatively low, as in our case[3].

Papuloerythroderma is characterized by a pruritic erythroderma-like eruption formed by coalescing flat-topped red-to-brown papules that spare the skin folds (deck-chair sign).[4] While some characteristic features of PED, including pruritic erythroderma-like eruption with deck-chair sign, peripheral eosinophilia, and elevated serum levels of IgE and TARC,[4,5] were observed, our patient also showed resistance to topical corticosteroid therapy, nonbacterial palmoplantar pustular lesions and histologically eosinophil infiltration in the pilosebaceous units, which are the main findings of EPF.[1,3]

To our knowledge, this is the first case of EPF that clinically mimicked PED. If topical corticosteroid-resistant PED-like eruption is observed, it is necessary to take EPF into consideration and utilize serial sections to assess the presence of eosinophil infiltration in pilosebaceous units.

DECLARATION SECTION

Approval of the research protocol: N/A.
Informed Consent: Written informed consent was obtained from the patients.
Registry and the Registration No. of the study/trial: N/A.
Animal Studies: N/A.

KEYWORDS

cyclosporine, eosinophilic pustular folliculitis, indomethacin farnesyl, papuloerythroderma of Ofuji, TARC

CONFLICT OF INTEREST

Dr. Manabu Fujimoto is the Editor in Chief for the Journal of Cutaneous Immunology and Allergy. Management of the peer review process, and all editorial decision-aking, for this article was undertaken by an Associate Editor.

Aya Maekawa
Yukinobu Nakagawa
Kasumi Kakuda
Eiji Kiyohara
Mari Wataya-Kaneda
Manabu Fujimoto

Department of Dermatology, Course of Integrated Medicine, Graduate School of Medicine, Osaka University, Osaka, Japan

Correspondence
Yukinobu Nakagawa, Department of Dermatology, Course of Integrated Medicine, Graduate School of Medicine, Osaka University, 2-2 Yamadaoka, Suita, Osaka 565-0871, Japan.
Email: nakagawa@derma.med.osaka-u.ac.jp

ORCID

Yukinobu Nakagawa https://orcid.org/0000-0002-8247-0385
Eiji Kiyohara https://orcid.org/0000-0002-0436-4296
Manabu Fujimoto https://orcid.org/0000-0002-3062-4872

REFERENCES

1. Takematsu H, Nakamura K, Igarashi M, Tagami H. Eosinophilic pustular folliculitis. Report of two cases with a review of the Japanese literature. Arch Dermatol. 1985;121(7):917–20.
2. Murayama T, Nakamura K, Tsuchida T. Eosinophilic pustular folliculitis with extensive distribution: correlation of serum TARC levels and peripheral blood eosinophil numbers. Int J Dermatol. 2015;54(9):1071–4.
3. Nomura T, Katoh M, Yamamoto Y, Miyachi Y, Kabashima K. Eosinophilic pustular folliculitis: a proposal of diagnostic and therapeutic algorithms. J Dermatol. 2016;43(11):1301–6.
4. Torchia D, Miteva M, Hu S, Cohen C, Romanelli P. ,Papuloerythroderma 2009: two new cases and systematic review of the worldwide literature 25 years after its identification by Ofuji et al. Dermatology. 2010;220(4):311–20.
5. Yamamura R, Murao K, Kubo Y. High serum thymus and activation regulated chemokine levels in papuloerythroderma of Ofuji. J Dermatol. 2017;44(2):214–5.

How to cite this article: Maekawa A, Nakagawa Y, Kakuda K, Kiyohara E, Wataya-Kaneda M, Fujimoto M. A case of eosinophilic pustular folliculitis presenting as papuloerythroderma of Ofuji-like eruption. *J Cutan Immunol Allerg*. 2021;4:37–38. https://doi.org/10.1002/cia2.12147