

A Case of Eosinophilic Folliculitis after Allogenic Bone Marrow Transplantation in Acute Myelogenous Leukemia

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Eosinophilic folliculitis(EF) is regarded as a variant of eosinophilic pustular folliculitis (EPF), because it has a few distinctive clinical features different from those of EPF. EF is generally associated with systemic disorders, such as acquired immunodeficiency syndrome (AIDS) and hematologic malignancies.

We have recently experienced a case of EF occurring in a 40 year-old male patient treated with allogenic bone marrow transplantation(BMT) for acute myelogenous leukemia(AML) and achieved a good clinical outcome after a short course of systemic corticosteroid therapy. The immunologic aberration resulting from systemic diseases may play a role in the development of EF. (*Ann Dermatol* 13(4) 235~238, 2001).

Key Words : Eosinophilic folliculitis, Acute myelogenous leukemia(AML), Allogenic bone marrow transplantation(BMT)

Eosinophilic pustular folliculitis(EPF) is a rare, pruritic, papulo-pustular dermatosis of unknown origin, which was first described by Ise and Ofuji in Japan¹ and later in Europe and America as an idiopathic disease^{2,3}. Its association with systemic disorders was not known, until some cases were observed in patients affected by the acquired immunodeficiency syndrome(AIDS)^{4,5}. Because this HIV-associated EPF has distinctive clinical features, some authors described this variant of EPF as eosinophilic folliculitis(EF)⁶.

Recently, we have experienced a rare case of EF occurring in a patient treated with allogenic bone marrow transplantation(BMT) for acute myeloge-

nous leukemia(AML) and achieved a good clinical outcome after a short course of systemic corticosteroid therapy. The association of an EF with an AML has already been reported in the literature and this is the first report of EF in Korean literature.

CASE REPORT

A 40 year-old male patient with acute myelogenous leukemia(AML)/M4 was admitted to our unit with moderately pruritic, multiple, erythematous papules on the chest, back, and both arms(Fig. 1A, 1B). Following 2 cycles of chemotherapy with cytosine arbinoside and idarubicin, and pre-conditioning with total body irradiation(TBI) and melphalan, allogenic bone marrow transplantation was performed. Methotrexate and cyclosporin A were given to prevent graft-versus-host reaction. Twenty weeks after BMT, he developed these itchy skin eruptions which consisted of follicular and nonfollicular papules and plaques. Interviews with the patient revealed no past illnesses except a hematologic problem. Family history revealed nothing remarkable.

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Fig. 1. (A) Multiple, erythematous, follicular and non-follicular papules on the trunk and arms. (B) Close-up view of the right arm.

Fig. 2. (A) Biopsy specimen taken from the chest shows a marked mixed inflammatory cell infiltration located in perifollicular and perivascular area(H & E, $\times 40$). (B) Higher power view showing numerous eosinophils within the inflammatory cells(H & E, $\times 400$).

Laboratory findings and physical examinations, including complete blood count, blood chemistry, urinalysis, and serological test, were within the normal range except relative eosinophilia. The proportion of eosinophils in the peripheral blood

was 5.1%. His HIV serology was negative. Infectious causes were ruled out on the basis of negative results on the bacterial and fungal cultures.

Histopathological findings from the chest and right arm revealed a marked mixed inflammatory

infiltration consisting of eosinophils and lymphocytes which were around and inside hair follicles (Fig. 2A, 2B). Associated follicular spongiosis and focal destruction of the follicular structures were also present.

Treatment with oral antibiotics and topical corticosteroid was unsuccessful, but he responded well to a short course of oral prednisolone 20mg daily. With the clinical improvement of skin rashes, serum eosinophilia also normalized. But the lesions frequently recurred when the medication was discontinued. With the above observations, we diagnosed the case as eosinophilic folliculitis, a variant of eosinophilic pustular folliculitis. We are following up on the patient with intermittent courses of systemic corticosteroid therapy.

DISCUSSION

Eosinophilic pustular folliculitis (EPF), or Ofuji's disease is a rare chronic dermatosis, originally described in adult Japanese in 1970⁷ but now reported occasionally in Caucasians^{2,3,8}. This condition has a peak incidence in the third decade of life and is characterized by recurrent crops of pruritic, sterile, follicular papules and pustules which coalesce to form circinate plaques. They may show central clearing with residual hyperpigmentation. Seborehmic areas, such as face, trunk, and extensor surface of the proximal part of the limbs, are usually involved but palms and soles are rarely involved. Infectious causes are ruled out on the basis of negative results on the following tests: periodic acid-Schiff and acid-fast staining, potassium hydroxide examination, bacterial and fungal cultures. For these reasons, the designations 'eosinophilic pustular dermatosis'³ and 'sterile eosinophilic pustulosis'⁹ have been suggested.

EPF was initially described in immunocompetent healthy individuals, but it also has been reported in infants¹⁰ and in patients with human immunodeficiency virus (HIV) infection^{4,5}. Rosenthal et al.⁶ have suggested that although this HIV-associated skin disorder is similar to EPF, it also has distinctive clinical features, such as the presence of pruritus, absence of annular or circinate plaques and no central clearing. Therefore, they described this variant of EPF as eosinophilic folliculitis (EF). Recently, similar cases have been reported in patients without HIV infection during therapy for

hematological malignancies¹¹⁻¹³. Bull et al.¹⁴ reported five cases of EF occurring in HIV-negative patients treated for hematological malignancies and suggested that EF is a consequence of disturbances of the immune function, not specific for HIV infection. They concluded that EF may only be a part of the spectrum of EPF or represent a whole new entity.

The pathogenesis of EF and EPF is still unknown. It is not clear whether eosinophils play a primary role in these disorders, or whether the increased infiltration of eosinophils is a part of an allergic response to an unknown stimulus. The presence of edema, pruritus, eosinophilia, elevated IgE levels, and deregulated mast cells in the lesions adjacent to the follicles suggest that the mast cells may be important in the pathogenesis of these lesions¹⁵. Furthermore, the association of some cases with immunologic alterations or diseases, such as immunodeficiencies and hematologic malignancies, suggests a possible immunopathologic event in the pathogenesis¹².

Histopathological findings of EF are similar to those of EPF. The principal histopathologic features are inflammation of the upper half of the hair follicle, sometimes with marked involvement of sebaceous gland, destruction of the hair shaft, and mixed inflammatory cell infiltrates around the hair follicle and blood vessels. Eosinophils were prominent in the inflammatory infiltrates, both inside and around the hair follicle. But the occurrence of eosinophil-rich pustules is comparatively infrequent in EF. We found the same histopathologic features mentioned above from the patient admitted to our unit.

Patients respond rapidly to oral corticosteroid therapy, which has been noted as an effective treatment¹⁶. Other agents reported to be effective include dapsone¹⁷, minocycline¹⁶, nonsteroidal anti-inflammatory drugs¹⁸, isotretinoin¹⁹, metronidazole²⁰, ultraviolet B (UVB) phototherapy²¹, indomethacin²², and interferon- γ ²³. Likewise, the skin rashes of our patient showed rapid improvement after a short course of corticosteroid therapy.

EF needs to be differentiated from pustular eruptions seen in immunosuppressed patients, such as staphylococcal folliculitis, pityrosporum folliculitis, Gram-negative folliculitis, and fungal infection¹⁴ and can be easily differentiated from these disorders on the basis of negative results of laboratory studies. In present case, the rashes were confined to

the arm and trunk, as in HIV-associated EF. This distribution is similar to EPF but unlike EPF annular or circinate plaques with central clearing or palmar/plantar lesions were not observed.

With the above observations, we finally diagnosed the case as EF that developed after allogeneic bone marrow transplantation for acute myelogenous leukemia. We suggest that the immunologic aberration resulting from systemic diseases may play a role in the development of EF and careful follow up is necessary.

REFERENCES

1. Ise S, Ofuji S: Subcorneal pustular dermatosis: A follicular variant? *Arch Dermatol* 92:169-171, 1965.
2. Holst R: Eosinophilic pustular folliculitis. *Br J Dermatol* 95:661-664, 1976.
3. Orfanos CE, Sterry W: Sterile eosinophile pustulose. *Dermatologica* 157:193-205, 1978.
4. Soeprono FF, Schinella RA: Eosinophilic pustular folliculitis in patients with acquired immunodeficiency syndrome: report of three cases. *J Am Acad Dermatol* 14:1020-1022, 1986.
5. Jenkins D Jr, Fisher BK, Chalvardjian A, Adam P: Eosinophilic pustular folliculitis in a patient with AIDS. *Int Dermatol* 27:34-35, 1988.
6. Rosenthal D, LeBoit PE, Klumpp L, Berger TG: Human immunodeficiency virus-associated eosinophilic folliculitis: A unique dermatosis associated with advanced human immunodeficiency virus infection. *Arch Dermatol* 127:206-209, 1991.
7. Ofuji S, Ogino A, Horio T, et al: Eosinophilic pustular folliculitis. *Acta Derm Venereol (Stockh)* 50:195-203, 1970.
8. Jaliman HD, Phelps RG, Fleischmajer R: Eosinophilic pustular folliculitis. *J Am Acad Dermatol* 14:479-482, 1986.
9. Saruta T, Nakamizo Y: Eosinophilic pustular folliculitis: Particularly with reference to lesions on the palm of the hand and sole of the foot. *J Dermatol (Tokyo)* 7:239-246, 1980.
10. Lucty AW, Esterly NB, Heskel N, Krafchik BR, Solomon LM: Eosinophilic pustular folliculitis in infancy. *Pediatr Dermatol* 1:202-206, 1984.
11. Roger H, Souteyrand P, Bignon YJ, Fonck Y, Plagne R, Fournier JR: Pustulose folliculaire a eosinophiles revelant un limphone T de haut degre de malignite rapidment fatal. *Ann Dermatol Venereol* 115:1209-1212, 1988, Cited from reference 12.
12. Patrizi A., Lernia VD, Neri I, Gherlinzoni F: Eosinophilic pustular folliculitis (Ofuji's disease) and non-Hodgkin lymphoma. *Acta Derm Venereol (stockh)* 72:146-147, 1992.
13. Evans TRJ, Mansi JL, Bull R, et al: Eosinophilic folliculitis occurring after bone marrow autograft in a patient with non-Hodgkin's lymphoma. *Cancer* 73:2512-2514, 1994.
14. Bull RH, Harland CA, Fallowfield ME, Mortimer PS: Eosinophilic folliculitis: a self-limiting illness in patients being treated for haematological malignancy. *Br J Dermatol* 129:178-182, 1993.
15. Buchness MR, Gregory N, Lim HW, Soter NA: The role of mast cells and eosinophils of the acquired immunodeficiency syndrome. *J Invest Dermatol* 92:408, 1989.
16. Ofuji S: Eosinophilic pustular folliculitis. *Dermatology* 174:53-56, 1987.
17. Steffen C: Eosinophilic pustular folliculitis (Ofuji's disease) with response dapsone therapy. *Arch Dermatol* 121:921-923, 1985.
18. Leiferman KM: Cutaneous eosinophilic diseases. In Freedberg IM, Eisen AZ, Wolfe K, et al. (eds): *Fitzpatrick's Dermatology in general medicine*. 5th ed., McGraw-Hill, New York, 1999, pp1129-1137.
19. Berbis P, Jancovici E, Lebreuil G, Benderitter T, Dubertret L, Privat Y: Eosinophilic pustular folliculitis (Ofuji's disease): efficacy of isotretinoin. *Dermatologica* 179:214-216, 1989.
20. Smith KJ, Skelton HG, Yeager J, Wagner KF: Metronidazole for eosinophilic pustular folliculitis in human immunodeficiency virus type 1-positive patients. *Arch Dermatol* 131:1089-1091, 1995.
21. Buchness MR, Lim HW, Hatcher VA, Sanchez M, Soter NA: Eosinophilic pustular folliculitis in the acquired immunodeficiency syndrome: Treatment with ultraviolet B phototherapy. *N Eng J Med* 318:1183-1186, 1988.
22. Miyauchi T, Fujigaki H, Uehara M, Hayashi S, Hirogana M: Effect of indomethacin on eosinophilic pustular folliculitis. *Acta Dermatol Kyoto* 80:9-14, 1985.
23. Fushimi M, Tokura Y, Sachi Y, et al: Eosinophilic pustular folliculitis effectively treated with recombinant interferon- γ : suppression of interleukin 5 in peripheral blood mononuclear cell. *Br J Dermatol* 134:766-772, 1996.