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LETTER TO THE EDITOR

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Papuloerythroderma occurring in a patient with myelodysplastic syndrome

Dear Editor,

Papuloerythroderma (PE) is a clinical entity proposed by Ofuji et al[1] in 1984. While PE was originally described in possible association with internal malignancies,[1] cutaneous T-cell lymphoma occasionally manifests PE,[2] and drugs may induce PE as an adverse effect.[1,3] We present a case of PE possibly associated with myelodysplastic syndrome (MDS).

An 85-year-old Japanese man was referred to us with a 2 month history of pruritic erythema on the trunk and extremities. He was treated with topical corticosteroids and tacrolimus and oral antihistamines without therapeutic effects. Two and half years prior to our examination, the patient was diagnosed as having MDS. He was first noticed to have mild anemia and a high number of immature granulocytes on routine blood examination. Bone marrow biopsy revealed hyperplastic hematologic cells and trisomy of chromosome 8.

On our initial examination, the patient had red solid papules, some of which coalesced into plaques, on the trunk and extremities. One month later, the eruption extended to whole trunk, sparing abdominal creases and forming deck-chair sign (Figure 1A). Simultaneously, he developed uneven hyperkeratosis on the bilateral palms (Figure 1B) and soles. A biopsy specimen from the femoral lesion showed acanthosis of the epidermis and perivascular infiltration of lymphocytes intermingled with eosinophils, but not immature myeloid cells, in the upper dermis (Figure 1C). Immunohistochemically, $CD4^+$ T cells dominantly infiltrated. Gene rearrangement of T-cell receptor Cβ1 was negative with the lesional skin specimen. On blood examination, there were leukocytosis (29 440 per mL) with neutrophilia (76%) and eosinophilia (7%), but not lymphocytosis, mild anemia (hemoglobin, 13.4 g/dL), and normal count platelets. High serum IgE level (2734 U/mL; normal range, <233) was observed. Serum protein fractions were normal, and anti-human T-cell lym-

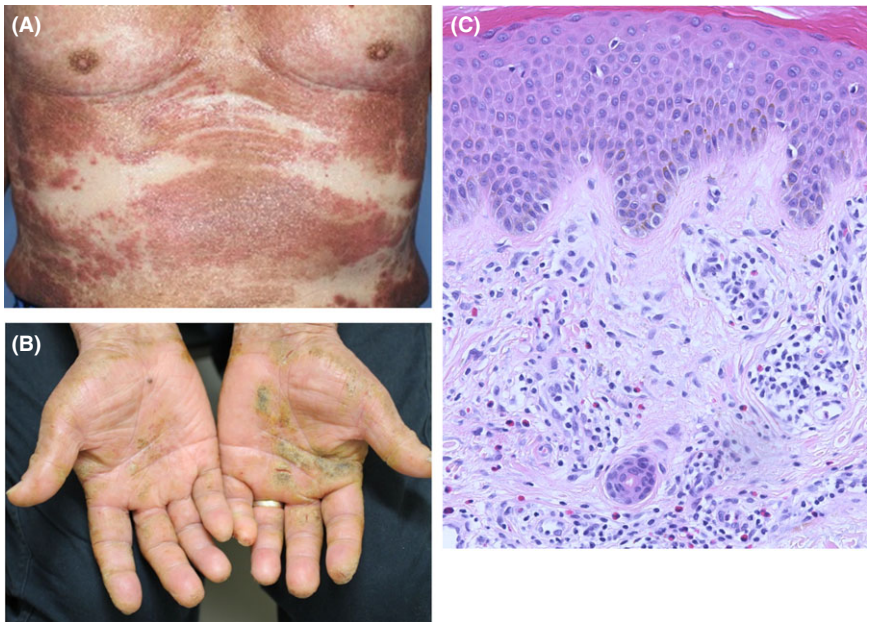

FIGURE 1 Skin manifestations and histopathological findings. A, PE on the chest and abdomen, with deck-chair sign; B, Palmar hyperkeratosis; C, Histopathology, showing perivascular lymphohistiocytic infiltration intermingled with eosinophils (hematoxylin and eosin staining, original magnification ×200)

photropic virus-1 antibody was negative. Lymphocyte stimulation test to the daily taken oral drugs was negative, and the eruption was not improved even after withdrawal of the drugs. No finding suggestive of visceral malignant tumors was seen with blood tumor markers or contrast-enhanced computer tomography. Based on these findings, we diagnosed his eruption as PE possibly associated with MDS.

We initiated oral administration of cyclosporine (200 mg daily), which improved the eruption on the trunk, leaving pigmentation, and the palmoplantar hyperkeratosis remarkably. However, cyclosporine discontinued because of elevation of his blood pressure. As the skin lesions immediately recurred, oral prednisolone (20 mg daily) started, which currently alleviated the eruption.

There have been some cases of PE associated with hematological neoplasms, including both lymphocytic and myeloid leukemia/lymphoma. In cases of lymphocytic malignancies, PE is mostly a direct manifestation induced by malignant lymphocytes as represented by mycosis fungoides and Sézary syndrome.[2] On the other hand, myeloid leukemia is an indirect condition because malignant myeloid cells do not infiltrate in the lesional skin. To our knowledge, only one case of PE with MDS was documented.[4] In that case, cyclosporine and prednisolone were therapeutically effective, but MDS evolved into acute myeloid leukemia thereafter.[5] It should be kept in mind that such development might take place in our patient.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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