A Case of Inflammatory Vitiligo

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Vitiligo is common dermatologic disorder, but inflammatory vitiligo is a rare occurrence that is characterized by erythematous rim at the periphery of a patch of hypopigmented skin. Since the initial description of inflammatory vitiligo, only about 20 cases have been identified via a review of the literature. We herein report the first Korean case of inflammatory vitiligo developed in an 56-year-old male. (Ann Dermatol (Seoul) 19(2) 96~98, 2007)

Key Words: Inflammatory vitiligo

INTRODUCTION

The classic description of inflammatory vitiligo is an erythematous rim at the periphery of a patch of hypopigmented skin[1]. Although vitiligo is a common pigmentary disorder, there are few published reports of inflammatory vitiligo. Since the initial description of inflammatory vitiligo[2], only about 20 cases have been identified via a review of the literature. Moreover, there have been no reports of this condition in Korea.

The diagnosis of inflammatory vitiligo is not easy, especially when the typical erythematous margin is not prominent and a skin biopsy shows dense, superficial, lymphocytic infiltrates. It can be misdiagnosed as another skin disease, such as hypopigmented mycosis fungoides (MF)[3], atrophic lichen planus[2], fungal infection or parapsoriasis[4].

We herein report the first Korean case of inflammatory vitiligo.

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CASE REPORT

A 56-year-old male resented with a 1-year history of hypopigmented lesions on the trunk and extremities. On physical examination, there were variable sized hypopigmented patches on the buttocks and trunk, both under natural light and by Wood' lamp examination (Fig. 1a). On the thigh, there were several centrally-hypopigmented patches with a discrete, elevated, scaly, erythematous rim (Fig. 1b). The patient described that at first, the lesions on the abdomen and buttocks also showed an elevated, scaly, erythematous rim but the lesions had then subsequently turned into completely depigmented patches. There was no significant past medical or family history. Intermittent mild itching was noted. A KOH preparation of scales from the thigh and ankle showed negative findings. Laboratory evaluation including a complete blood count, ANA, and

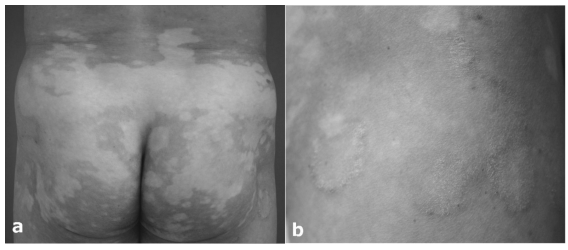

Fig. 1. (a) Variable-sized, hypopigmented patches with scalloped borders on the buttocks, (b) Several centrally-hypopigmented patches with a discrete, elevated, scaly erythematous rim on the thigh.

VDRL were within normal limits. Given the loss of pigment on the buttocks and trunk, inflammatory vitiligo was the leading diagnosis. However, the differential diagnosis included hypopigmented mycosis fungoides. Three biopsy specimens were obtained from the thigh, one each from the normally-pigmented skin, the erythematous border, and the depigmented skin. Histopathologically, the biopsy specimen from the erythematous border showed mild acanthosis, mild spongiosis and focal vacuolar alteration of the basal layer associated with exocytosis and superficial papillary and perivascular lymphocytic infiltrate (Fig. 2a). No atypical lymphocytes were noted. An immunohistochemical stain for melanocytes, NKI-beteb, showed a markedly-decreased number of melanocytes in the epidermis and in some foci, they were completely absent (Fig. 2b). Fontana-Masson staining for melanin revealed diminished melanin pigment. CD3 staining revealed a large number of T cells, and they were mainly concentrated along the basal layer. A biopsy specimen from depigmented skin showed a very sparse superficial perivascular lymphocytic infiltrate (Fig. 2c). NKI-beteb staining showed a complete absence of melanocytes in the epidermis (Fig. 2d). Fontana-Masson staining revealed no melanin pigment in the basal cell layer. Histological examination from normal skin showed no histopathological abnormalities (Fig. 2e). NKI-beteb staining revealed a normal number and distribution of melanocytes (Fig. 2f).

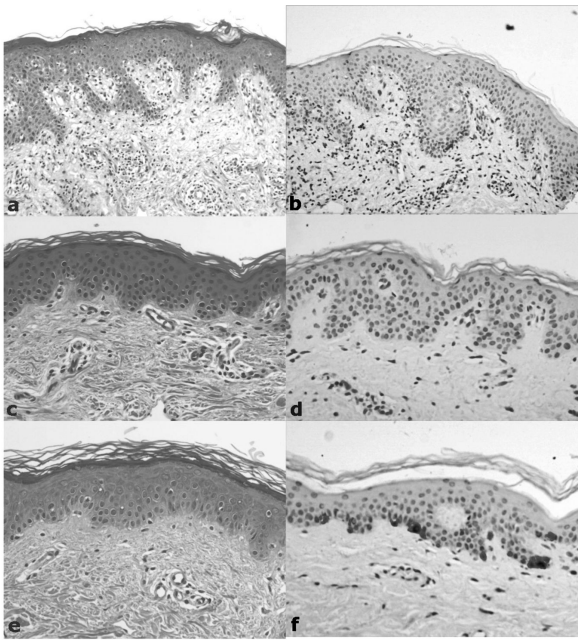

Fig. 2. (a) Erythematous border showed exocytosis, focal vacuolar alteration of the basal layer and superficial inflammatory infiltrates, (b) Staining for melanocytes of the erythematous border showed a markedly- decreased number of melanocytes in the epidermis, and in some foci, they were completely absent, (c) Depigmented skin showed very sparse, superficial perivascular infiltrates, (d) Staining for melanocytes of depigmented skin showed a complete absence of melanocytes in the epidermis, (e) Histological examination from normal skin showed no histopathological abnormalities, (f) Staining for melanocytes of normal skin revealed a normal number and distribution of the melanocytes (a,c,e: H & E; original magnifications: × 200, b,d,f: NKI- beteb stain; original magnifications: × 200).

DISCUSSION

Although vitiligo is a common pigmentary disorder, inflammatory vitiligo is an unusual occurrence. Since the initial description of inflammatory vitiligo[2], only about 20 cases have been identified via a review of the literature. Among them, 3 reports were of inflammatory vitiligo with erythematous, scaly plaques, as in our patient[4-6].

This unique form of vitiligo is also considered to be an accentuated reaction of vitiligo, and provides evidence for the involvement of cellular immunity in the pathogenesis of vitiligo[7]. The pathogenesis of vitiligo is still a matter of debate, but the autoimmune hypothesis is considered to be the major factor, and both humoral and cellular immunity has been considered. Histologically, an infiltrate of skin-homing cytotoxic T-cells expressing granzyme/perforin is often found close to the remaining melanocytes in marginal skin from progressing lesions of generalized vitiligo[8]. An histology and immunophenotypic study of vitiligo on inflammatory-raised erythema was reported by LePoole et al[7] and the erythematous margin in our case showed acanthosis, focal vacuolar alteration of the basal layer associated with exocytosis and superficial papillary and perivascular lymphocytic infiltrate, which mainly consisted of T-cells, and a decrease in the number of melanocytes. These results suggest that our case might be an accentuated form of vitiligo and that T cell-medicated cytotoxicity may be involved in its pathogenesis. Histopathologic findings

could rule out atrophic lichen planus and parapsoriasis. The main risk in the diagnosis of inflammatory vitiligo is to rule out a hypopigmented variant of MF. In our case, the erythematous raised border surrounding hypopigmented patches, the loss of melanocytes, exocytosis with spongiosis and absence of atypical lymphocytes favoured the diagnosis of inflammatory vitiligo.

To conclude, we present the first case of inflammatory vitiligo in Korea.

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