Inflammatory Linear Verrucous Epidermal Naevus Arising on a Burn Scar

Sir,

Inflammatory linear verrucous epidermal naevus (ILVEN) is a dermatosis that is usually present at birth or appear during the first few years of life though it may have its onset as late as the 40s or 50s (1, 2). It comprises characteristically pruritic erythematous verrucous papules in linear array which are refractory to treatment (2, 3). We report here a patient who developed typical pruritic skin lesions 3 months after an electric burn. Skin lesions began at the burn scar on the dorsum of the right hand and spread to the normal skin and burn scar on the forearm, shoulder, anterior chest and buttock ipsilaterally. Histology showed psoriasiform epidermal hyperplasia with areas of orthokeratosis and a prominent granular layer alternating with cup-like areas of hypogranulosis and overlying parakeratosis. The burns were the triggering factor in the pathogenesis of this case. Treatment with calcipotriol ointment, twice daily for 5 months, induced marked improvement.

CASE REPORT

A 51-year-old Korean man had been suffering from a strongly pruritic skin eruption since July 1989. He was burned on his right hand, back and buttocks in April 1989. Physical examination revealed burn scars involving an area approximately 5 or 6 cm in diameter on the dorsum of both hands and extensive areas on the back and buttocks. On clinical examination there were linear hyperkeratotic, confluent verrucous papules on the burn scar of the dorsum of the right hand and on normal skin and burn scars on the right upper extremity (Fig. 1), anterior chest and buttock. However there were no skin lesions on the burn scars on the left hand, back and buttock. He stated that his skin lesions began on his right hand and gradually spread to the other areas described above. He did not have any other medical problems and routine laboratory tests including complete blood cell counts, liver function test and urine analysis were normal. There was no personal or family history of psoriasis. Biopsy specimen obtained from the skin lesion showed regular epidermal acanthosis, alternating parakeratosis and orthokeratosis. In the upper dermis there was a chronic inflammatory infiltrate. The diagnosis of ILVEN was established on the clinical features and histological findings. The skin lesions were first treated with etretinate 10 mg 3 times daily and topical corticosteroids. However, since liver function test showed elevated serum transglutaminase (SGOT/SGPT) and triglyceride level after 1 month of treatment, etretinate was discontinued. The patient received prolonged topical treatment with corticosteroid and liquid nitrogen but the skin lesions waxed and waned with treatment until May 1997. Treatment with calcipotriol 50 μg/g ointment, twice daily was started. After 5 months there was marked improvement (Fig. 2).

Fig. 1. Linear hyperkeratotic, confluent verrucous papules on the burn scar on the dorsum of the right hand and normal skin and burn scars on the right upper extremity.

Fig. 2. Marked improvement of the skin lesions was noticed after 5 months’ treatment with calcipotriol ointment.
DISCUSSION

The cutaneous sequelae of burns are usually temporary loss of pigment, hypertrophic scar and keloid. Infrequently skin neoplasia such as squamous cell carcinoma, basal cell carcinoma, malignant melanoma and sarcoma develop within the burn scar. Because ILVEN was developed after a burn in our patient, it is suggested that the possible mechanism for the development of ILVEN may have been that the burn acted as a simple trauma or induced an impairment of the immune function which was responsible for the onset of ILVEN. ILVEN is typically resistant to various therapies including corticosteroids, tar, dithranol and cryotherapy (3, 4). The skin lesions in our patient also responded poorly to various therapies. There are several reports that topical calcipotriol is an effective and safe therapy for the treatment of ILVEN (4, 5). Our patient was treated with topical calcipotriol twice a day for 5 months and showed marked improvement.

REFERENCES


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Anti-centromere Antibodies in 2 Patients with Discoid Lupus Erythematosus and No Signs of Systemic Sclerosis

Sir, Patients with typical discoid lupus erythematosus (DLE) only rarely have associated systemic sclerosis (SS) (1, 2) and there are no reports of them having anticitromere antibodies (ACA) in the absence of SS signs.

CASE REPORTS

Case 1
A 36-year-old woman presented with a 6-year history of DLE of the scalp, face and ears. She had developed Raynaud’s phenomenon at the age of 20 years. In 1979 she had only speckle-patterned IgG ANA at a final titre of 1/160. She was treated with chloroquine and the DLE lesions cleared and were then controlled every 2 years. In 1996, she developed centromere-patterned ANA and antimitochondrial antibodies at a final titre of 1/1280. All other routine and immunological findings were within normal limits. No signs of sclerosis have been observed so far.

Case 2
A 56-year-old woman was seen in 1984 with a 40-year history of DLE of the scalp, face and ears. She had speckle-patterned IgG ANA at a final titre of 1/40 and low total complementaemia (CH50 <200) without immunocomplexes. Treatment with chloroquine was successful during relapses. The cutaneous lesions were silent for 2 years, but the ANA titre progressively rose, acquiring in 1996 a centromeric pattern at a final titre of 1/2560. Raynaud’s phenomenon and other signs of SS were absent.

DISCUSSION

ACA are usually present in patients with a limited form of SS and relatively good prognosis. They have also been reported occasionally in patients with systemic lupus erythematosus, rheumatoid arthritis, Raynaud’s disease, Sjögren syndrome, primary biliary cirrhosis, immune haemolytic anaemia and thrombocytopenic purpura (3). They have also been found at low titre in healthy subjects and in patients with non-autoimmune diseases (4).

In our 2 patients, ACA developed long after the onset of DLE and had a high titre. Very rarely have patients been observed with DLE/SS combination and even more rarely with a serology reminiscent of SS. ACA was found in only 1 of the 6 DLE/SS cases described by Sasaki & Nakajima (1). As in all such patients in whom DLE preceded SS, ACA may be regarded as a sign of impending SS development. Whether this is the case in our 2 patients remains to be seen.

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