Marked Hyperkeratosis of the Soles in Keratitis-Ichthyosis-Deafness Syndrome: Treatment With Hydrocolloid Dressing

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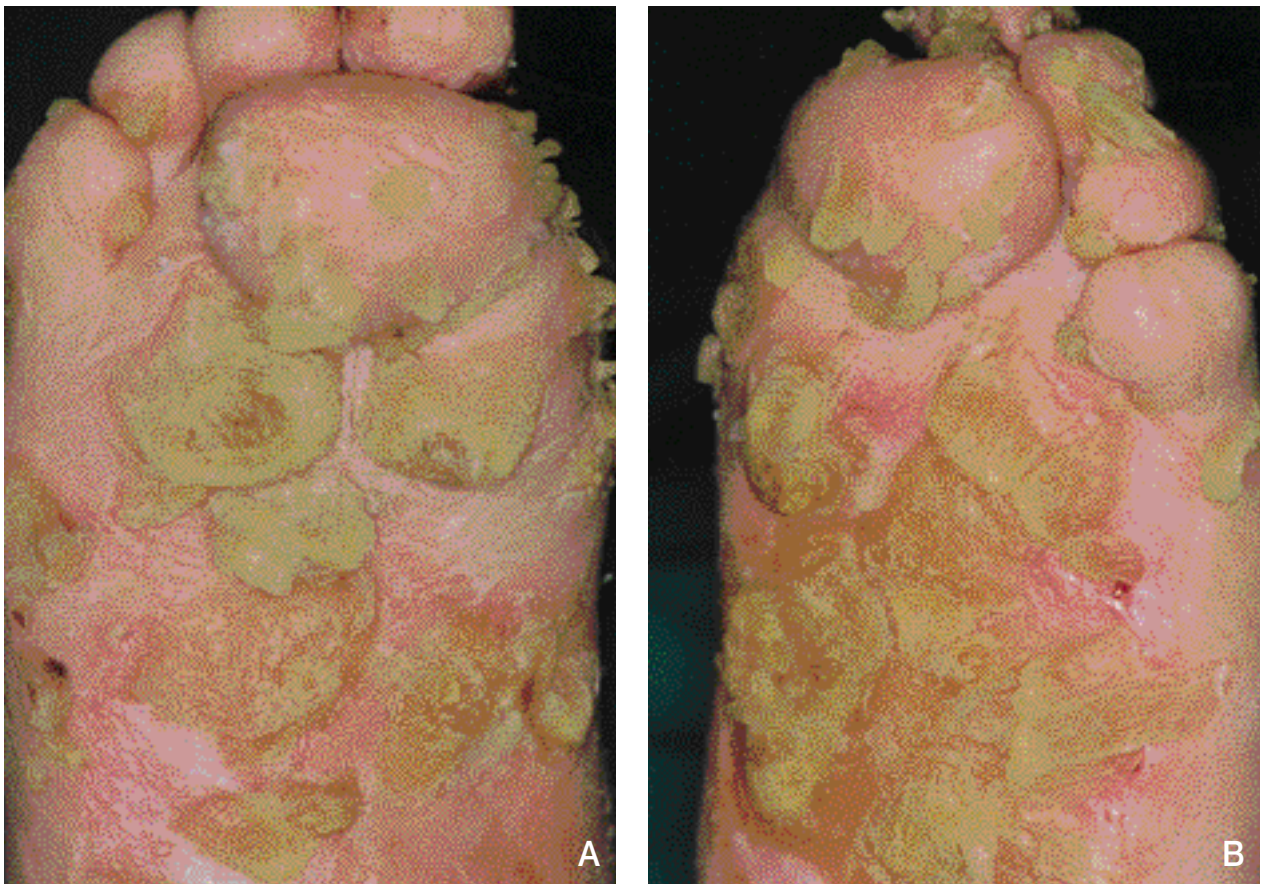

Figure 1. Yellow, granite-hard, thick crusts about 5-mm thick were attached to the soles (A and B).

Keratitis-ichthyosis-deafness (KID) syndrome is a rare congenital disorder that exhibits marked hyperkeratosis of the skin. We successfully treated cutaneous lesions on the soles of a patient with KID syndrome using hydrocolloid dressing.

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Keratitis-ichthyosis-deafness (KID) syndrome is a rare, congenital ichthyosiform disorder associated with neurosensory deafness and vascularizing keratitis. Various stages of erythrokeratotic scaling plaques develop over the skin during a person's first year of life. The palms and soles

exhibit spiny hyperkeratosis and keratoderma.[1] We used hydrocolloid dressing (HCD) to treat marked hyperkeratosis of the soles that interfered with normal walking in a patient with KID syndrome.

Case Report

Our patient was a 21-year-old Japanese man who was diagnosed with KID syndrome at the age of 2 years.[2] He was treated with etretinate 20 mg but continued to exhibit spiny hyperkeratosis and keratoderma over the skin, especially on the soles (Figure 1). The patient walked slowly to reduce the pain associated with these stony hard crusts. The crusts were too hard and thick to cut with scissors, and attempts to remove them induced bleeding easily. Keratolytic agents, including topical urea 10% ointment and vitamin D_3 ointment, did not relieve the symptoms.

We applied HCD to the patient's soles and changed the dressing twice a week for 4 weeks. Most of the thick crusts became softened and were easily removed (Figure 2). After treatment, the patient reported no more pain and the ability to walk comfortably. The patient has been successful on a maintenance regimen of topical vitamin D_3 ointment once a day and occasional HCD monotherapy (about once a month for several days).

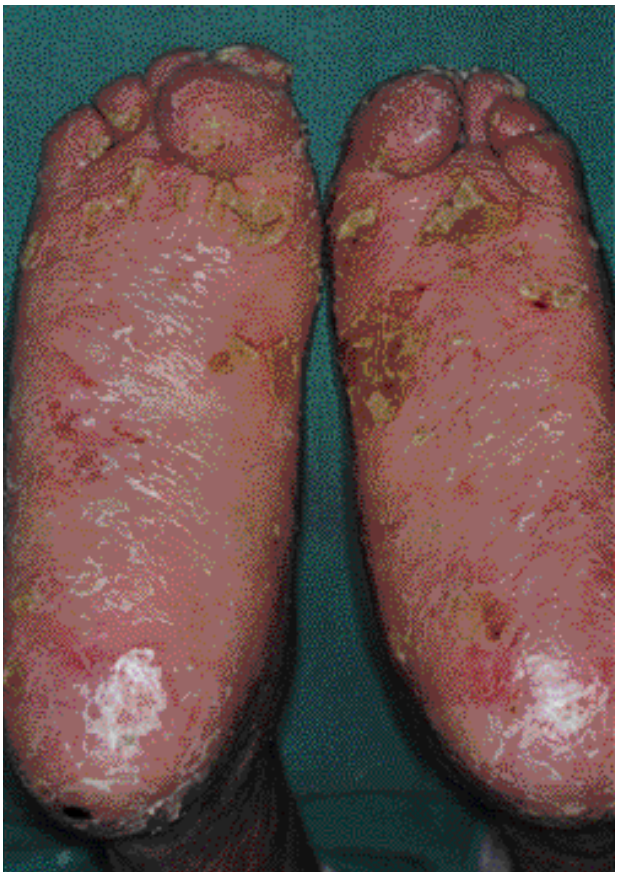

Figure 2. The soles after treatment with HCD, which softened most of the thick crusts and enabled their removal.

Comment

The keratolytic agents that treat hyperkeratosis in KID syndrome only provide symptomatic relief. The effectiveness of retinoids is not constant, and retinoids should be used with caution because they can exacerbate keratitis.[1] In our patient, oral retinoids and topical urea ointment did not reduce the thick crusts. Although the effect of topical calcipotriol on congenital ichthyoses has been reported,[3] this agent was not effective in our patient; however, it was useful as maintenance therapy after initial treatment with HCD.

The clinical efficacy of HCD in the treatment of plaque psoriasis has been reported.[4] HCD monotherapy induced slight improvement of erythema, but it markedly improved scaling and induration. Although the precise mechanism of action of HCD in psoriasis is still unknown, a mechanical effect caused by stripping away the scales is thought to be important.[4]

We used HCD to remove the masses of scales and crusts on the soles of our patient. The therapy showed remarkable effects without any adverse events. HCD may have protected the lesion from pressure or friction while providing moisture to the dry crusts. These effects may soften the hard crusts of the soles, which consequently can be stripped away, as in psoriasis. We believe that HCD could be an effective therapeutic option for hyperkeratosis in KID syndrome.

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