Idiopathic Recurrent Palmoplantar Hidradenitis in Children

Report of 22 Cases

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Background: Idiopathic recurrent palmoplantar hidradenitis (IRPH) is characterized by tender erythematous plaques and nodules on the soles and, less often, the palms of young patients. To date, 10 cases of IRPH have been documented in the literature.

Observations: We describe 22 pediatric patients with characteristic cutaneous and histologic findings of IRPH. Their mean age was 6 years (age range, 1.5-15 years). The onset of the disease clustered in 2 peaks, in autumn and spring. All patients had complete resolution of their lesions within 3 weeks, in 16 cases without any treatment. Ten of the 22 patients experienced more than 1 episode of IRPH.

Conclusions: Our study represents the largest number of cases of IRPH collected to date. Based on our data, the clinical picture is so characteristic that a histopathologic examination may not be necessary in every case. We suggest that there is a seasonality in the occurrence of this always benign and limited disease.

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PATIENTS AND METHODS

EPIDEMIOLOGICAL AND LABORATORY FINDINGS

Fifteen of our 22 patients were female, revealing an overall female-male ratio of 2.1:1. The mean patient age was 6 years (age range, 1.5-15 years) (Figure 1). Two patients were sisters who suffered from IRPH at the same time. The onset of the disease in our patients clustered in 2 peaks, in autumn and spring (Figure 2), suggesting seasonality.

Laboratory tests, which were performed in 21 of 22 cases, revealed elevated white blood cell counts (14-20×10⁹/L), erythrocyte sedimentation rates (24-28 mm/h; reference range, 0-10 mm/h), and serum C-reactive protein levels (11-12 mg/L; reference range, 0-10 mg/L) in 3 patients. The results of other routine blood chemistry tests were within normal limits.

CLINICAL SPECTRUM

The occurrence of multiple, painful, erythematous macules and indurated nodules ranging from 0.5 to 3 cm in diameter on the palms and soles of otherwise healthy children is the clinical hallmark of IRPH (Figure 3). Of 22 patients, 21 had exclusively tender plantar involvement, resulting in debilitation. The lesions were unilateral in 2 children. Bilateral, tender, palmar macules and nodules appeared in 50% of the group. An elevated body temperature (<38°C) was noted in 2 girls. All patients had complete resolution of the lesions (duration, 2-21 days after onset [median duration, 8 days]); 6 were treated with topical nonsteroidal anti-inflammatory agents, and 16 were not treated. Ten of our 22 patients experienced more than 1 episode of their eruption.

HISTOLOGIC FINDINGS

The microscopic findings in all patients (n=22) with IRPH were very similar. Each specimen showed a nodular infiltrate consisting predominantly of neutrophils that was localized to the eccrine apparatus, particularly to the coils (Figure 4), with slight extension to the periglandular tissue. In the middle and deep dermis, there was a mild, mixed perivascular infiltrate without the typical changes of a leukocytoclastic vasculitis. Stains were negative for microorganisms, including bacteria and fungi, in all cases.

The findings of histopathologic examination, including distinctly focal, nodular, neutrophilic infiltrates around the eccrine sweat units, confirm the clinical diagnosis of IRPH, but the examination may not be necessary if the findings of medical history, as well as the clinical and laboratory findings (infections?), are characteristic.

The pathogenesis of IRPH is unknown. Based on our clinical and histologic observations, we believe that mechanical and/or thermal trauma can lead to a rupture of the palmar plantar eccrine glands. The release of glandular secretions into the surrounding tissue may activate a cytokine cascade (eg, tumor necrosis factor α, granulocyte colony-stimulating factor, and interleukin 8) that is capable of attracting neutrophils that can result in a transient inflammation clinically represented by tender, erythematous palmar plantar papules and nodules. Therefore, the high relapse rate observed in our patients is not surprising.

The majority of our patients did not need any distinct therapy. If the disease causes pain or low-grade fe-
Figure 3. Classic lesions of idiopathic recurrent palmoplantar hidradenitis. A, B, and C, Tender, erythematous macules and nodules on the plantar aspects of the feet. D, Painful, erythematous nodules on the palms.

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<th>Differential Diagnosis of Idiopathic Recurrent Palmoplantar Hidradenitis</th>
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<tr>
<td><strong>Diagnosis</strong></td>
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<tr>
<td>Traumatic plantar urticaria</td>
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<td>Erythema multiforme</td>
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<td>Erythema nodosum</td>
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<td>Sweet syndrome</td>
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<td>Behçet disease</td>
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<td>Chilblains</td>
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*ESR indicates erythrocyte sedimentation rate; CRP, C-reactive protein.
ver, treatment with nonsteroidal anti-inflammatory drugs is helpful. We do not see the necessity for further treatment or diagnostic procedures in this probably common and, in every case, harmless benign disease. On the other hand, if no improvement occurs within 1 or 2 weeks after the onset of the tender, erythematous palmoplantar nodules in young patients, histopathologic evaluation is necessary to rule out clinically similar diseases (Table).

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REFERENCES