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CLINICAL CASE

Painful nodules on the soles in a pediatric patient: a diagnostic challenge

Nódulos plantares dolorosos tras ejercicio intenso en paciente pediátrica: un reto diagnóstico

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What do we know about the subject matter of this study?

Painful plantar nodules are a condition of unknown etiology, with well-defined clinical and pathological characteristics.

What does this study contribute to what is already known?

A very representative clinical case of this condition, which is probably under-diagnosed, that helps the understanding of this entity, easing its management and avoiding diagnostic errors that may lead to unneeded treatments.

Abstract

Introduction: Idiopathic Palmoplantar Eccrine Hidradenitis (IPPH) is a rare neutrophilic dermatosis, with painful erythematous nodules of sudden onset in the plantar or palmoplantar region, in children without other underlying diseases. **Objective**: To present a case that shows the main clinical and histological characteristics of this entity. **Clinical Case**: 11-year-old girl with a 48-hours history of painful erythematous-violaceous nodules on the right foot plant associated with fever of up to 38.2 ºC, with no history of interest except hyperhidrosis and intense exercising on previous days. Given the clinical suspicion of IPPH, a skin biopsy was performed, which showed inflammatory neutrophil infiltration around eccrine sweat glands and neutrophilic abscesses, confirming the diagnosis. Oral NSAIDs and rest were prescribed, with resolution of the lesions in 7 days. **Conclusions**: This case demonstrates the most important aspects of this entity, in many cases underdiagnosed, since it can be confused with other pathologies that occur with painful acral nodules, but have different pathogenic and therapeutic implications. To properly identify the IPPH allows preventing an unnecessary alarm, both patients and their parents, as in dermatologists and pediatricians themselves.

Keywords:
Hidradenitis; Foot Dermatoses/diagnosis; Eccrine Glands/pathology; Child

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Introduction

Idiopathic palmoplantar eccrine hidradenitis (IPPH) is a rare neutrophilic dermatosis that was first described in 1994 by Stahr et al[1] in six healthy patients, five children, and one young adult. All of them presented a clinical picture of painful plantar lesions and a histological study similar to neutrophilic eccrine hidradenitis (NEH) but with different characteristics. Since then, less than 50 cases have been reported, most of them in healthy patients aged between 2 and 15 years, with plantar and, sometimes, palmar involvement.

Its etiopathogenesis is still unclear. A higher incidence has been observed in patients with history of intense physical exercise and/or use of closed shoes during the warm season[2,3]. Some theories support that, in patients with immature glands, a mechanical or thermal trauma could produce the rupture of those glands infiltrating their content into the dermis, activating inflammatory cytokines which can attract polymorphonuclear ones[2,4,5]. Another hypothesis suggests that excessive sweating, either intrinsic or related to intense physical exercise, would initiate this inflammatory cascade[6,7]. However, laboratory studies do not provide specific data in this regard[8].

The histological findings are characteristic of the presence of intense inflammatory infiltration, predominantly neutrophilic, around the eccrine sweat glands, both in the portion of the coil, where neutrophil abscesses can also form in the adjacent tissues and in the duct, where neutrophil accumulations can also be observed inside the duct in an inconstant but a quite specific manner[8]. There were no eccrine squamous syringometaplasia or typical signs of leukocytoclastic vasculitis[2,8].

The objective of this report is to present a case that describes the main clinical and histological characteristics of IPPH in order to highlight a characteristic picture that is probably underdiagnosed.

Clinical Case

11-year-old girl seen at the Emergency Department due to a 48-hours history of painful lesions on the right foot which prevented her from walking, with fever of up to 38.2ºC and no other associated cutaneous or systemic clinical symptoms or signs.

The presence of frequent episodes of plantar hyperhidrosis and basketball practice in the previous days were the only data of interest referred by the patient. Physical examination showed erythematous-violaceous nodules on the external lateral edge and forefoot area of the right foot painful on palpation (Figure 1).

A skin biopsy was performed on one of the lesions, which showed a predominantly neutrophilic inflammatory infiltrate around the eccrine sweat glands and neutrophil abscesses near to the coil (Figure 2). With these findings, the diagnostic suspicion of IPPH was confirmed and we started treatment with oral non-steroidal anti-inflammatory drugs and rest, with gradual resolution of the lesions in 7 days, with no sequelae or recurrences after 3 years of follow-up.

Discusión

IPPH is a disease specific to children and young adults, clinically characterized by erythematous or erythematous-violaceous plaques and/or nodules, which are painful, of sudden onset, located in the plantar or palmoplantar regions, uni- or bilaterally[2,4,8]. Generally, this entity occurs in healthy patients with no history of other underlying diseases or recent medication use[8]; however, it also appears with fever but without other general symptoms in some cases[2,4].

When a pediatric patient presents with painful acral nodules, we must know how to recognize this picture and differentiate it from others with similar symptoms (Table 1).

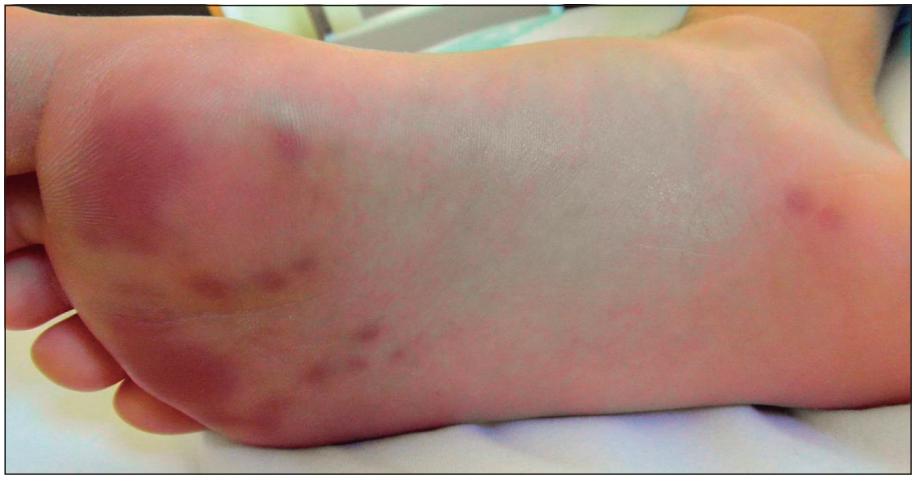

Figure 1. Erythematous-violaceous nodules on the external lateral edge and forefoot area of the right foot.

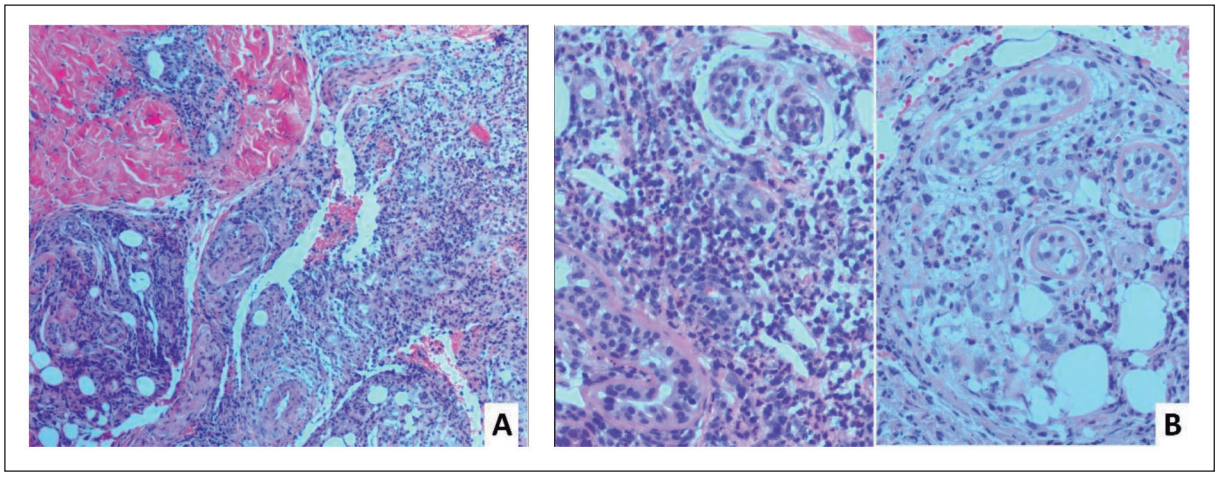

Figure 2. A) HE x20: Neutrophilic inflammatory infiltrate araound the eccrine sweat glands and neu trophil abscesses near to the coil. **B)** HE x40: inflammatory infiltration surrounding and inside the duct of glands.

Table 1. Differential diagnosis of painful acral nodules

	IPPH	NEH	TPU	PEN	PHFS
Clinic	Palmoplnatar painful nodules, of sudden onset, uni-or bilatera-lly, +/- fever	Polymorphous skin rash due to fever which affects the trunk, face, and limbs	Recurrent outbreaks of plantar erythema-tous maculopapulae, pruritic or painful	Painful plantar subcu-taneous nodules with post-inflammatory hyperpigmentation +/- Fever, arthralgias and general malaise	Erythematous painful plantar nodules, sudden onset
Medical history/ associations	Healthy young patient +/- Hyperhidrosis, physical exercise	Chemotherapy (cytarabine...) BRAF inhibitors G-CSF Lymphoma/leukemia HIV	Hours after intense physical activity	Preceded by infection or drugs	6-48 hours after bathing in a hot or heated pool. Community outbreak in public pools
Etiopatho-genesis	Immature eccrine gland injury due to high temperature or mechanical factors	Direct cytotoxic effect of the secretion of chemotherapy in the eccrine glands or paraneoplastic syndrome	Physical pressure urticaria	Delayed hypersensi-tivity to a variety of antigenic stimuli	*Pseudomonas aerugi-nosa* infection
Histology	Neutrophilic abscesses around the eccrine sweat glands, both in the portion of the coil and in the ducts No vasculitis	Neutrophilic infiltra-te in the secretory portion of the eccrine glands, reticular der-mis and subcutaneous cell tissue. Eccrine squamous syringometaplasia No abscesos	Inflammatory infil-tration in the dermis, with edema, without affecting the eccrine sweat glands	Septal panniculitis without vasculitis	Similar to HPPI, except Gram stain in which the microorganism can be seen
Treatment	resolves sponta-neously in < 3 weeks	Resolves sponta-neously in 1-3 weeks. +/- Etiological treatment	Avoid triggering stimulus Oral antihistamines	Rest +/- salicylates or NSAIDs	Avoid the source of infection Antimicrobials and topical antiseptics +/- oral antibiotic

HPPI (Hidradenitis palmoplantar ecrina idiopática), HEN (hidradenitis ecrina neutrofílica), UTP (urticaria traumática plantar), ENP (eritema nodoso plantar infantil), SPCP (Síndrome de pie caliente por pseudomonas)

The following are the main differential diagnoses to consider: traumatic plantar urticaria, characterized by recurrent outbreaks of plantar erythematous maculopapulae, pruritic or painful, which appear hours after intense physical activity and its histological study shows an inflammatory infiltration in the dermis without affecting the eccrine sweat glands[2,8,9]; plantar erythema nodosum of childhood, it presents very painful nodules, generally after infection or the use of medicines, which resolve more slowly, leaving post-inflammatory hyperpigmentation and in its histology we observe the characteristic septal panniculitis typical of erythema nodosum[2,8,10]; and the pseudomonas hot-foot syndrome, which presents a clinical picture very similar to IPPH, with painful, erythematous plantar nodules of sudden appearance, but which characteristically appears as community outbreaks in children, 6 to 48 hours after bathing in a hot or heated pool contaminated by this pathogen[11].

Other entities such as erythema multiforme, perniosis, and some vasculitis and cellulitis could also be considered in the differential diagnosis[2,8].

Finally, we should know how to differentiate IPPH from NEH, a polymorphous skin rash due to fever which affects the trunk, face, and limbs. This entity was initially described in patients with hematologic malignancy receiving chemotherapy and, later, it has also been associated with other drugs, malignancies, and infections. Both present similar histology, but with different characteristics such as eccrine squamous syringometaplasia in NEH, and the presence of neutrophil abscesses near the glandular coils in IPPH[8].

Regarding the treatment, only symptomatic relief is recommended, since it is a condition that resolves spontaneously in about 3 weeks without any sequelae. It generally includes rest which may be done along with the use of non-steroidal anti-inflammatory drugs and topical corticosteroids[2,6].

Conclusion

We present a case of IPPH with a typical clinical and histological picture. Also, we provide the keys to make a proper differential diagnosis with other entities with painful acral nodules, but that present a different clinical evolution and therapeutic implications, which causes unnecessary concern, both in patients and their parents, as in the doctors themselves.

Ethical Responsibilities

Human Beings and animals protection: Disclosure the authors state that the procedures were followed according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

Data confidentiality: The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.

Rights to privacy and informed consent: The authors have obtained the informed consent of the parents (tutors) of the patients and/or subjects referred to in the article. This document is in the possession of the correspondence author.

Conflicts of Interest

Authors declare no conflict of interest regarding the present study.

Financial Disclosure

Authors state that no economic support has been associated with the present study.

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