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T. Arif,[a,*] M. Adil,[b] S. Suhail Amin,[b] M. Alam[c]

[a] *Departamento de Dermatología, ETS y Lepra, Ellahi Medicare, Kashmir, India*

[b] *Departamento de Dermatología, ETS y Lepra, Facultad de Medicina Jawaharlal Nehru (JNMC), Universidad Aligarh Muslim (AMU), Aligarh, India*
[c] *Departamento of Dermatología, Facultad de Medicina Jawaharlal Nehru (JNMC), Universidad Aligarh Muslim (AMU), Aligarh, India*

* Corresponding author.
E-mail address: dr_tasleem_arif@yahoo.com (T. Arif).

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Circumscribed Plantar Hypokeratosis☆

Hipoqueratosis plantar circunscrita

To the Editor:

Circumscribed palmoplantar hypokeratosis (CPH) is a rare disease characterized by focal thinning of the stratum corneum. It manifests as a round, well-defined, depressed, erythematous lesion with a scaly border, generally located on the palms. It is generally asymptomatic and predominantly affects middle-aged or elderly women. We report 3 cases of CPH on the sole of the foot, a rare site for this disease.

Case 1. A 74-year-old diabetic man who visited our department with an asymptomatic erythematous lesion on the sole of the left foot; the lesion had well defined borders and had grown slowly over the previous 7 years (Fig. 1A) . Dermatoscopy revealed a stepped scaly border, an erythematous base with punctate vessels, regular white spots occasionally surrounded by the punctate vessels, and thin white lines (Fig. 1B). Clinical and dermatoscopic data were compatible with CPH. The patient refused a biopsy and did not want to undergo treatment.

Case 2. An 82-year-old woman with no relevant past history visited our department with a mildly pruritic lesion on the inside surface of the right foot that had grown slowly over the previous 4 years. Physical examination revealed a lesion measuring 12 × 10 mm with well-defined borders and a depressed erythematous center. Dermatoscopy

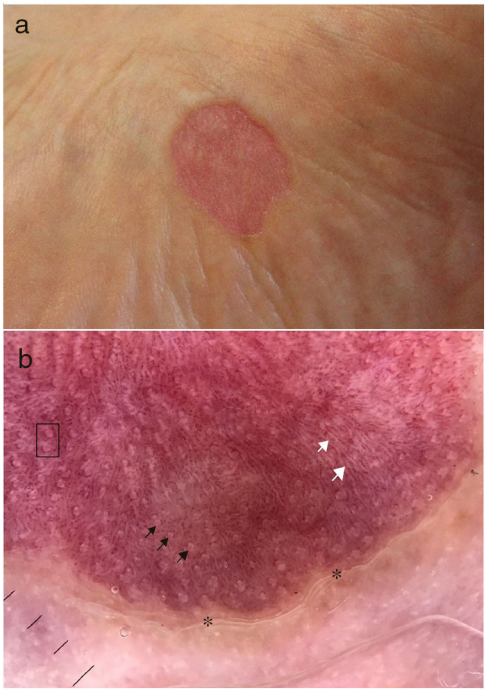

Figure 1 Case 1. A, Clinical image. B, Dermatoscopic image showing the stepped scaly border (asterisk), regular white spots (white arrows) on an erythematous background with punctate vessels, some of which are distributed around the white spots (black square).

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Table 1 Review of Cases of Circumscribed Plantar Hypokeratosis Published to Date, With Clinical Characteristics, Treatments Used, and Response to Treatment.

No.	Authors and Date	Age	Sex	Sole	Treatments	Clinical Course
1	Pérez et al.,[1] 2002	68	Female	Left	Several	No change
2	Rütten et al.,[3] 2004	64	Female	Left	None	NI
3	Berk et al.,[3] 2007	75	Female	Left	5-fluorouracil topical Calcipotriol ointment Clobetasol ointment	No change No change No change
4	Berk et al.,[3] 2007	49	Female	Left	Fluocinolone cream Excision	No change Complete resolution
5	Barry et al.,[3] 2008	59	Male	Right	NI	NI
6	Tanioka et al.,[3] 2009	62	Male	Right	Topical corticosteroids Antibiotics	No change No change
7	Cribier et al.,[3] 2009	65	Male	Left	None	No change
8	Kanitakis et al.,[3] 2011	56	Female	Right	NI	NI
9	Arbesman et al.,[3] 2012	10	Male	Left	NI	NI
10	Santamarina-Albertos et al.,[3] 2013	2	Male	Right	Calcipotriol topical	No change
11	Batalla et al.,[3] 2013	17	Male	Right	Calcipotriol topical	Complete resolution
12	Mitkov et al.,[3] 2014	73	Male	Right	Mometasone topical Tacrolimus topical Pimecrolimus topical 5-fluorouracil topical Cryotherapy	No change No change No change No change No change
13	Pinos-León et al.,[3] 2014	55	Female	Left	Topical corticosteroids Topical antimycotics	No change No change
14	Ramos-Garibay et al.,[3] 2016	69	Female	Left	Calcipotriol topical	No change
15	Ramos-Garibay et al.,[3] 2016	71	Female	Left	None	No change
16	Nazzaro et al.,[4] 2016	80	Male	Left	NI	NI
17	Aranguren-López et al., 2018	74	Male	Left	None	NI
18	Aranguren-López et al., 2018	82	Female	Right	Topical betamethasone dipropionate and calcipotriol	Slight improvement
19	Aranguren-López et al., 2018	69	Female	Left	Cryotherapy	Complete resolution

Abbreviation: NI indicates not indicated.

revealed similar findings to the previous case. The biopsy showed a sudden reduction in the thickness of the stratum corneum, with hypergranulosis and diffuse areas of parakeratosis with no evidence of a parakeratotic column. CPH was diagnosed and the patient was treated with betamethasone dipropionate and calcipotriol, which produced a slight improvement. Treatment was suspended after a few weeks owing to irritation. The patient refused other treatments.

Case 3 A 69-year-old woman with no relevant past history visited our department with an asymptomatic, erythematous, depressed lesion on the arch of the left foot, with scaly border, which had appeared 10 years earlier (Fig. 2A) . The dermatoscopy and histology findings were similar to the previous cases, confirming the diagnosis of CPH. The patient was treated with tacrolimus ointment 0.1% for 4 months with little improvement. Cryotherapy with liquid nitrogen was then used and the lesion resolved completely (Fig. 2B).

CPH is a benign entity of unknown etiology that was first described in 2002 by Pérez et al.[1] It is thought to be due to an abnormal keratinocyte clone that does not differentiate into palmoplantar epidermis, leading to marked thinning of the stratum corneum in comparison to the surrounding normal skin.[2] Most of the cases described had no past history of trauma.

CPH is a rare disease, with approximately 100 cases reported in the literature.[3] More than 60% of these cases were located on the thenar prominence, on the palm or back of the hand, on the sole of the foot, and on the medial surface of the foot. These last 2 locations are rare and account for less than 15% of reported cases of CPH.[3,4] A review of the literature found 16 published case of CPH on the sole of the foot[1,3,4] (Table 1).

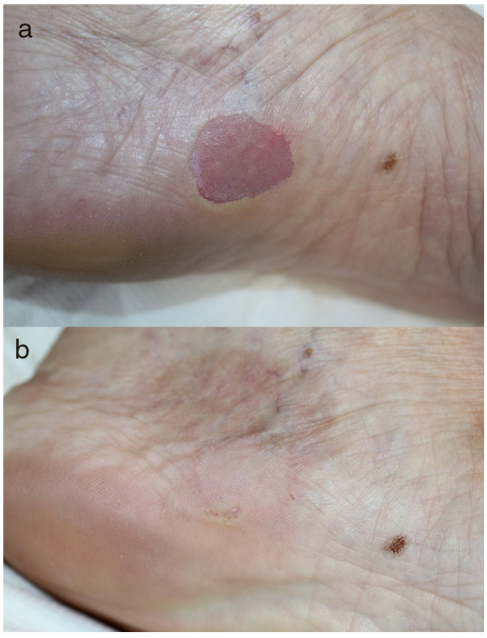

Figure 2 Caso 3. A, Lesion before cryotherapy. B, The same lesion 6 months after cryotherapy.

The histopathology of CPH is characteristic and confirms the diagnosis.[2] A sudden interruption can be observed between the normal dermis and that of the lesion, which shows marked hypokeratosis and mild hypogranulosis. No parakeratotic column can be seen at the periphery of the lesion. This differentiates the disease from porokeratosis of Mibelli, the most clinically similar entity, which must be included in the differential diagnosis.[2,5]

Dermatoscopy is very useful for diagnosing this dermatosis. Typical findings are peripheral stepped desquamation (corresponding to the abrupt limit between the normal and hypokeratotic epidermis), a uniform erythematous base with small punctate vessels (reflection of the congestive capillaries in the underlying dermis, which become more visible due to the epidermal thinning) and regularly distributed white spots (the pores of the acrosyringia).[5] White lines on the erythematous base,5 have been recently described, which correspond to the epidermal folds. The only dermatoscopy of palmoplantar hypokeratosis on the sole of the foot published to date[4] describes for the first time distribution of the punctate vessels around the acrosyringia. These recent findings were also observed in our patients.

No effective treatment for CPH exists. In some of the reported cases,[6] and in one of our patients, cryotherapy achieved complete remission of the lesions. Topical calcipotriol and 5-fluorouracil have also proven effective in some cases.[2]

In conclusion, we report 3 cases of CPH on the sole of the foot, a rare site for this rare disease, which is probably underdiagnosed due to its asymptomatic nature. We describe the dermatoscopic characteristics of the entity, which are very useful for diagnosis, and highlight the white lines found in one of our patients, which have been reported recently. We also highlight the utility of cryotherapy, which produced complete remission of the lesion in 1 case.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

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I. Aranguren-López,[a,*] S. Vildósola-Esturo,[a]
I. Arias-Camisón,[b] A. López-Pestaña[a]

[a] *Servicio de Dermatología, Hospital Universitario Donostia, San Sebastián, Guipúzcoa, España*
[b] *Servicio de Anatomía Patológica, Clínica de la Asunción, Tolosa, Guipúzcoa, España*

* Corresponding author.
E-mail address: INIGO.ARANGURENLOPEZ@osakidetza.eus (I. Aranguren-López).

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