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Circumscribed palmoplantar hypokeratosis: a case report and review of the literature

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Abstract

We describe an 84-year-old man presenting with a solitary, well-circumscribed, chronic erosion of the sole. Histopathologic examination confirmed diagnosis of circumscribed palmoplantar hypokeratosis. Circumscribed palmoplantar hypokeratosis is a rare and benign condition of unknown etiology presenting as an erosion on the palms or soles. Although lesions are typically asymptomatic, the entity is important for dermatologists and providers in other specialties to recognize, especially considering a differential diagnosis that includes neoplasia.

Keywords: circumscribed palmoplantar hypokeratosis, CPHK, human immunodeficiency virus, HIV, chronic erosion, palms, soles, human papillomavirus

Introduction

Circumscribed palmoplantar hypokeratosis (CPHK) is a rare histopathologic diagnosis that became recognized by Perez et al. in 2002 [1]. Although typically benign and asymptomatic, the entity remains an important diagnostic consideration because clinical appearance can resemble a neoplastic process such as Bowen disease [2].

Case Synopsis

An 84-year-old man presented for a full body skin examination. On the right plantar foot he was found to have a well-circumscribed, erythematous erosion

with scalloped borders, approximately two centimeters in diameter. The skin immediately surrounding the lesion appeared clinically normal (**Figure 1**). On further questioning, it was determined that the lesion had been present for at least five years, was asymptomatic, and had not been bleeding, growing, or changing in appearance. Given the persistence and chronicity of the lesion, a biopsy was performed. The differential diagnosis included neoplastic etiology, such as Bowen disease, and non-neoplastic etiology, such as porokeratosis.

On histopathologic examination the final diagnosis was CPHK. The condition provides a remarkable example of clinicopathologic correlation, with a precipitous decline of the corneocyte layer at the lesional margins demonstrated under light microscopy; the stratum corneum at the transition appears frayed (**Figure 2**). The perilesional cornified



Figure 1. Solitary well-circumscribed erosion on the right plantar foot.

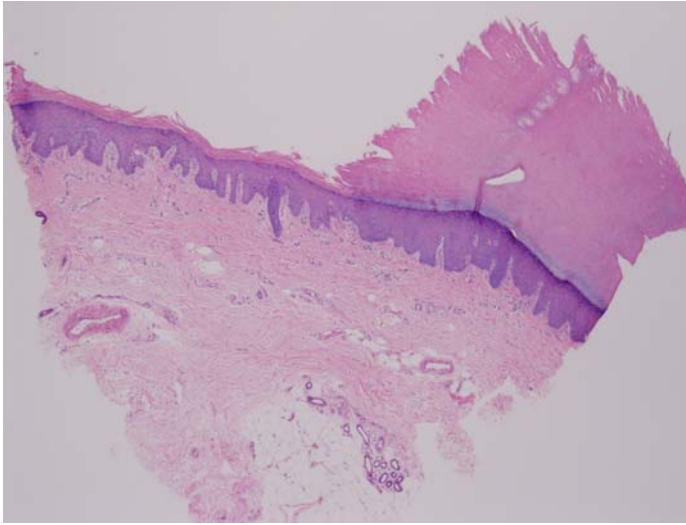


Figure 2. Histopathological examination reveals sharp demarcation with frayed stratum corneum between normal acral hyperkeratosis and the hypokeratotic erosion. H&E, 100x.

layer was found to be thick and orthokeratotic, consistent with normal acral skin, and the eroded area demonstrated marked loss of stratum corneum with only a hint of parakeratosis in the corneocyte layers that remained. The epidermis and dermis appeared normal except for a sparse presence of superficial perivascular infiltrate.

Case Discussion

There are 97 previously reported cases of CPHK in the literature. Circumscribed palmoplantar hypokeratosis is a rare and somewhat recently discovered condition that most often affects women over age 40 [3]. The classic clinical presentation is a well-circumscribed, round, depressed, erythematous patch on the palms or soles that may be confused for Bowen disease or prokeratosis of Mibelli [2]. It is typically considered benign, although there is one reported case of an actinic keratosis developing within the lesion [4]. Histologically, the lesions demonstrate a rapid decline in stratum corneum at the periphery of the lesion, with a well-demarcated

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change from perilesional normal compact orthokeratosis to lesional hypokeratosis, unremarkable epidermis and lack of inflammatory cells [2].

Human papillomavirus (HPV) has previously been suggested as one possible pathomechanism [5]. We performed HPV testing on the specimen, which was negative. In our review, only two of the 25 patients (8%) tested for HPV had a positive result [5, 6]. The low prevalence of lesional HPV detection in CPHK patients makes this an improbable etiology [7, 8].

There are a number of other hypotheses regarding the pathogenesis of CPHK. In the original case series, Perez et al. postulated that CPHK is an epidermal malformation [1]. Repeated trauma has been proposed as a potential origin and is supported by the condition's common location on the thenar and hypothenar eminence of the dominant hand as well as the plantar surfaces [9]. It has also been suggested that abnormal clonal keratinocytes are to blame [10]. Ultimately, further investigation is needed to elucidate the true pathogenesis of this disease.

Conclusion

In conclusion, CPHK is an uncommon condition with distinctive dermatopathology that characteristically presents on the palms or soles and has an uncertain pathogenesis. Herein we describe a case of CPHK that is clinically and pathologically consistent with those previously described; although HPV has been previously proposed as an etiology, our review suggests this is unlikely. Circumscribed palmoplantar hypokeratosis is an important benign entity for clinicians to recognize as the differential diagnosis includes malignant neoplasms.

Potential conflicts of interest

The authors declare no conflicts of interests.

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