Green palmoplantar vesicular eruption in a patient with hyperbilirubinemia

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Key words: dyshidrotic eczema; eccrine chromhidrosis; hyperbilirubinemia; liver disease; pompholyx.

INTRODUCTION

Green pigmentation on the palms and soles in patients with hyperbilirubinemia is a rare entity.[1-3] Eccrine chromhidrosis is an unusual form of green acral pigmentation characterized by the secretion of colored sweat from the eccrine glands.[1-3] Rarely, eccrine chromhidrosis can mimic an eczematous process leading to misdiagnosis.[1] We report a case of a patient with hyperbilirubinemia who presented with multiple deeply seated, greenish-black vesicles on the hands and feet, mainly of the palmoplantar surfaces, and pityriasis rosea on the trunk. Although the clinical presentation of the palms and soles appeared to be consistent with pompholyx, the color of the vesicles suggested other underlying pathophysiology. We suspect our patient had a variant of eccrine chromhidrosis secondary to hyperbilirubinemia presenting as greenish-black pompholyx. Here we describe the clinicopathologic features and potential pathomechanism of this unusual cutaneous presentation.

CASE REPORT

A 28-year-old Hispanic man with alcoholic cirrhosis presented with a 10-day history of a pruritic eruption that began on the trunk and gradually spread to involve all extremities. He was not aware of any contact allergens and denied preceding fever or hyperhidrosis. He also denied a medical history of skin disease such as atopy, dermatophytosis, or hand eczema. Physical examination found scleral icterus and dusky, thin plaques with central collarette of scale on the face, neck, axilla, trunk, and extremities. Hyperkeratotic plaques and deeply seated vesicles with greenish-black hue were noted to involve the dorsal and ventral aspects of the bilateral hands and feet. These lesions were especially accentuated along the dermatoglyphic lines of the palmoplantar surfaces (Fig 1, *A* and *B*). Laboratory testing found a total bilirubin level of 28.9 mg/dL (reference, <1.2 mg/dL) with a direct component of 17.7 mg/dL (reference, <0.3 mg/dL). Abnormal levels of aspartate aminotransferase at 78 IU/L (reference, <35 IU/L), alanine aminotransferase at 42 IU/L (reference, <40 IU/L), and alkaline phosphate at 231 IU/L (reference, 0-100 IU/L) were also detected. Punch biopsy of the right palm found psoriasiform epidermal acanthosis, intraepidermal spongiosis, and hyperkeratosis with sparse perivascular lymphocytic inflammation (Fig 2, *A*). A dilated eccrine duct with amorphous pink material within the epidermis was also noted (Fig 2, *B*). Gram, Periodic acid–Schiff, and iron stains were negative. A diagnosis of pompholyx was rendered. Punch biopsies from the right side of the chest and right thigh were in keeping with pityriasis rosea. Topical triamcinolone 0.1% ointment twice daily was initiated, and the patient was noted to improve over the course of 1 week. Unfortunately, he was lost to follow-up so long-term response could not be assessed.

DISCUSSION

The spontaneous appearance of green discoloration of the hair, skin, and nails is unusual.[4] Differential diagnoses include granulocytic sarcoma, poisoning (such as by arsenic), acute pancreatitis (Grey Turner and Cullen signs with the appearance of ecchymotic patches on the abdomen), eccrine chromhidrosis (which may cause yellow, blue, black, or green discoloration), and chronic liver disease. Hair and nails can become green from the

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Funding sources: None.
Conflicts of interest: None declared.
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JAAD Case Reports 2017;3:273-5.
2352-5126

http://dx.doi.org/10.1016/j.jdcr.2017.05.001

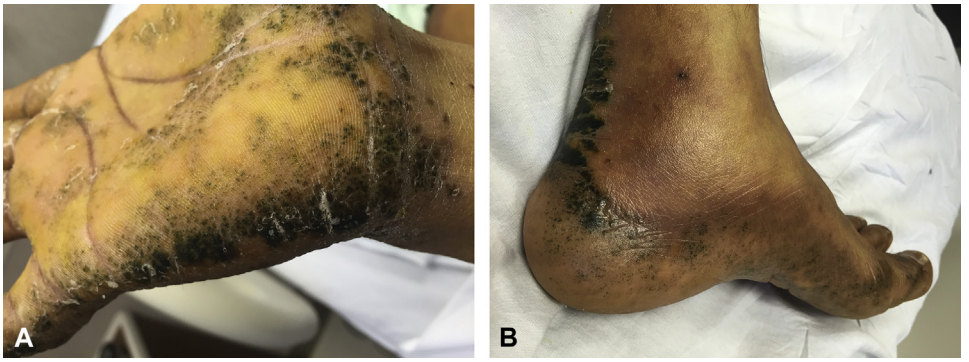

Fig 1. Multiple deeply seated vesicles with greenish-black hue with accentuation of the dermatoglyphic lines on the palmar aspect of the right hand (**A**) and the medial aspect of the left foot (**B**).

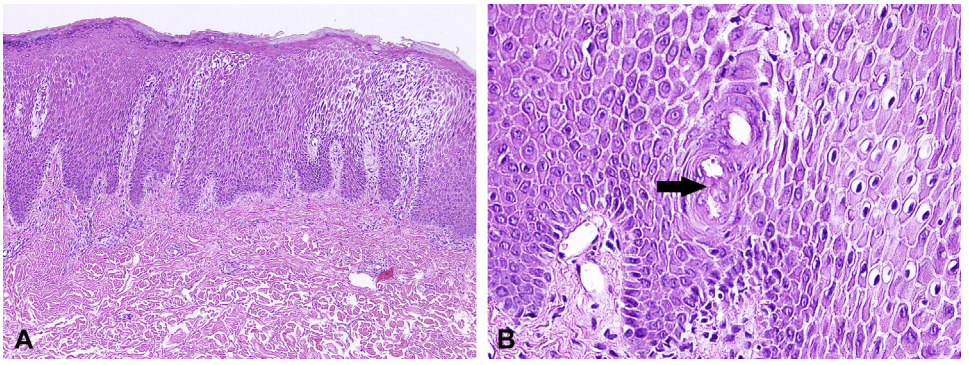

Fig 2. **A**, Hyperkeratosis, spongiosis, and psoriasiform hyperplasia with sparse inflammation. **B**, Dilated intraepidermal eccrine duct with associated amorphous pink material (*arrow*). (**A** and **B**, Hematoxylin-eosin stain; original magnifications: **A**, ×20; **B**, ×80.)

deposition of copper or from *Pseudomonas* infection. A greenish-black discoloration of the palms and soles, as seen in our case, is also rare.[1-3]

The etiology of pompholyx is unknown, but associations with atopy and contact sensitivity have been reported.[5,6] A pompholyx-type presentation with intensely pruritic flesh-colored vesicles on the palms and soles has also been observed in pityriasis rosea.[7] We describe an unusual case of pompholyx presenting as greenish-black palmoplantar vesicles in a patient with pityriasis rosea on the trunk and hyperbilirubinemia. Although pompholyx is a spongiotic dermatitis, the greenish discoloration of the palmoplantar vesicles seen in our patient cannot be explained by spongiotic changes alone. We hypothesize that this condition occurs because increased water-soluble bilirubin is delivered to the intraepidermal sweat glands of patients with hyperbilirubinemia, where it is deposited into the stratum corneum.[4,8] Inflammatory, spongiotic vesicles may induce rupture of neighboring sweat ducts containing high bilirubin, resulting in bilious-appearing vesicles. The green color occurs because brown-colored bilirubin is oxidized to green-colored biliverdin.[4,8]

In any patient with hyperbilirubinemia and palmoplantar vesicles, the differential diagnosis should include eccrine chromhidrosis, a rare condition in which pigment from dyes or medications are excreted via the eccrine sweat glands.[1-3] Histopathology of eccrine chromhidrosis finds hyperkeratosis, diffuse acanthosis, and increased number and size of intraepidermal eccrine ducts surrounded by homogenous eosinophilic materials, which can sometimes be seen in the stratum corneum.[1-3] In addition to these features, intraepidermal spongiosis and vesiculation can be seen when eccrine chromhidrosis presents as pompholyx,[1] as seen in our case. To the best of our knowledge, eccrine chromhidrosis has been reported in 3 patients,[1-3] and an eccrine chromhidrosislike presentation has been reported in 4 patients[4,6,8]; all patients had hyperbilirubinemia secondary to either liver disease, cholelithiasis, or cancerous processes. It is possible that in our case, because the green vesicles were located on the palms and soles, where the eccrine sweat gland density is highest, and along the dermatoglyphic ridges, our patient's presentation may actually be a

form of eccrine chromhidrosis. This possibility is supported by proposed pathophysiology that direct bilirubin is water soluble and may serve as the pigment or stain that is necessary in the pathologic basis of chromhidrosis.[1,4]

Eccrine chromhidrosis is a very rare manifestation of hyperbilirubinemia that can present as pompholyx, complicating diagnosis. The presence of hyperbilirubinemia and greenish-black discoloration of the palmoplantar eruption can aid in proper diagnosis. Our case is unusual in that our patient presented with concomitant pityriasis rosea, which, to our knowledge, has not been reported. Additional case reports are needed to elucidate this unusual disease presentation further.

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