

A case of cutaneous focal bilirubin deposition in a man with jaundice

Sir,

Spontaneous appearance of a green pigmented cutaneous rash is an unusual clinical finding and it may present in certain medical scenarios such as chloroma, second stage of well's syndrome, copper deposition in the hair and nails and pseudomonas infection.¹ However, such kind of unusual green pigmentation on the palms and soles also happens to be associated with underlying hyperbilirubinemia. In the literature, till now very few cases have been reported.¹⁻⁸

A 65-year-old male presented with a 10 days history of an asymptomatic progressive skin rash on his bilateral palms and soles. Patient complained of progressive jaundice and flatulence for about 2 months and was recently diagnosed with cholangiocarcinoma. There was no fever, sweating or any other symptoms. Physical examination revealed multiple, discrete, green to black, 1–2 mm pin point macules mainly on the palms and soles, especially on the volar and lateral aspects of the fingers and dorsomedial aspects of the toes [Figures 1 and 2]. The green punctae were distributed

along the dermatoglyphic ridges. Patient denied any recent use of topical or oral medications. He also denied any contact with greenish dyes or substances. Histopathological examination of the lesional skin biopsy indicated hyperkeratosis, parakeratosis and deposition of a pink amorphous substance within the stratum corneum [Figures 3 and 4]. The gram stain, potassium hydroxide examination and hall stain (if positive indicates a yellow hue) were negative. Tissue cultures for bacteria and fungus were also negative. Liver function test indicated elevated levels of total serum bilirubin (20.1 mg/dl). Patient underwent an elective pylorus



Figure 1: Multiple, discrete, green to black, pin point macular rash on the palms



Figure 2: Multiple, green to black pin point macules on the soles

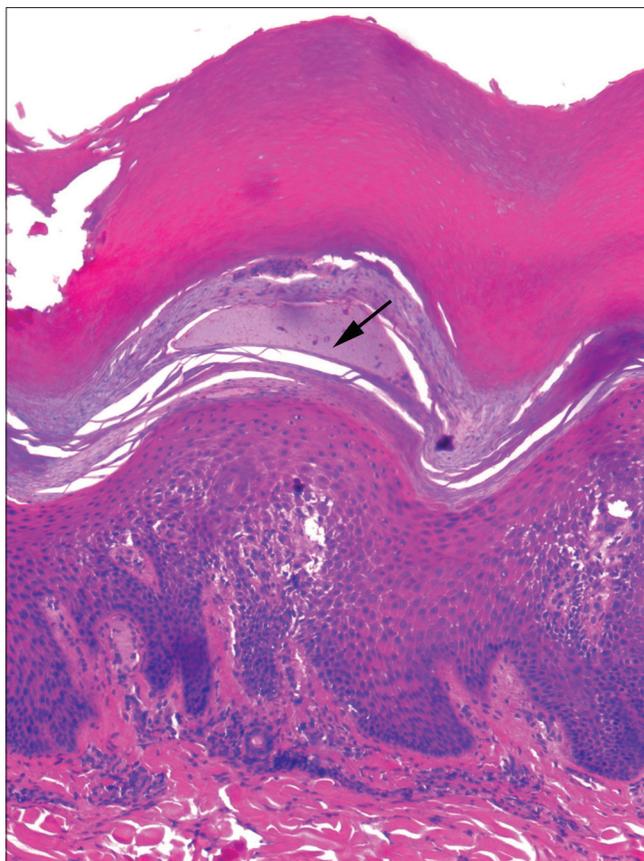


Figure 3: Hyperkeratosis and amorphous pink material in the stratum corneum consistent with bilirubin (arrow) (H and E, $\times 100$)

sparing pancreaticoduodenectomy. Two months after the surgery, the total serum bilirubin levels decreased to 0.9 mg/dl and were within the normal range (0.3–1.1 mg/dl). No further recurrence of the skin lesions was observed since then. According to the clinical and laboratory findings, a diagnosis of cutaneous focal bilirubin deposition was made.

Spontaneous appearance of a greenish pigmented rash on the palms and soles in patients with hyperbilirubinemia is a rare entity. Kanzaki and Tsuda were the first to report it in two patients, one with non-A, non-B acute hepatitis and the other recovering from a bypass surgery.² Over the years, similar rash has been reported in patients suffering from cholecystitis, choledocholithiasis, cholangiocarcinoma, adenocarcinoma of the gall bladder, biliary atresia, alcoholic liver cirrhosis and invasive duodenal adenocarcinoma.^{1,3–8} Usually, it is a greenish yellow pigmented rash with black-brown pin-point macules or papules or vesicles usually distributed along the dermatoglyphic ridges where eccrine sweat glands open. Moreover, in all of the reported cases, the rash was located only on the palms and soles, the location where eccrine glands are abundant.^{1–8} Our patient also had strikingly similar features on his palms and soles along with marked hyperbilirubinemia secondary to cholangiocarcinoma. The transient association of a green pigmentation with underlying marked hyperbilirubinemia and its resolution on correction of the serum abnormality supports the deposition of bilirubin and/or metabolites into the skin as the probable mechanism for the clinical presentation.³ In 1992, Kanzaki and Tsuda regarded this eruption as a variant of eccrine chromhidrosis (a condition characterized

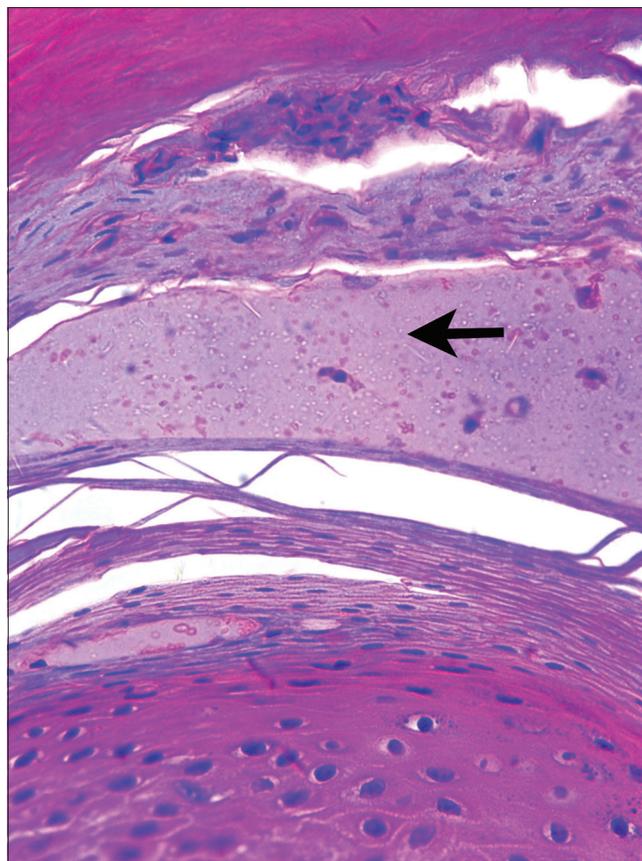


Figure 4: The presence of amorphous pink material in the stratum corneum consistent with bilirubin (arrow) (H and E, $\times 400$)

by excretion of water soluble pigment from certain dyes or drugs through the eccrine sweat glands). They hypothesized that direct water soluble bilirubin is excreted into the stratum corneum through the eccrine glands eventually where it gets oxidized to biliverdin.^{2,3} Fever and perspiration can also be considered as contributing factors but may not be present in all cases.¹ However, the special staining for bilirubin and/or metabolites did not yield any significant outcome. As reported in the literature, only two cases have shown positive results for bile stain.^{1,4} This may be due to the unavailability of optimal techniques to detect the presence of cutaneous bilirubin metabolites. Apart from the cutaneous deposition, bilirubin metabolites also tend to get deposited in the hard dental tissues secondary to hyperbilirubinemia during dentition. This contributes to the characteristic greenish discoloration of the permanent and deciduous teeth in individuals with congenital biliary atresia.⁹

Cutaneous focal bilirubin deposition is a very rare entity since not all patients with hyperbilirubinemia present with such a phenomenon. Diagnosis is usually based on the classic clinical and histopathological findings. However, certain medical scenarios such as chromhidrosis and pseudochromhidrosis need to be ruled out. We would like to report a case of cutaneous focal bilirubin deposition and would also like to highlight the fact that this condition can be considered as the cutaneous sign of underlying hepatobiliary disease.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their

consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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