Dermatology Reports

https://www.pagepress.org/journals/index.php/dr/index

eISSN 2036-7406

Please cite this article as:

Almutairi M, Nagshabandi KN, Alsergani R, Alsalem S. Yellow urticaria as a rare dermatological manifestation of biliary obstruction: a case report. *Dermatol Rep 2025 [Epub Ahead of Print] doi: 10.4081/dr.2025.10248*

Submitted 03/01/25 - Accepted 22/02/25

Yellow urticaria as a rare dermatological manifestation of biliary obstruction: a case report

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Key words: urticaria; yellow urticaria; bilirubin; hyperbilirubinemia; biliary obstruction.

Conflict of interest: the authors declare no potential conflict of interest.

Ethics approval and consent to participate: ethical approval is not required for this study in accordance with local or national guidelines.

Consent for publication: written informed consent was obtained from the patient for publication of the details of his medical case and any accompanying images.

Availability of data and materials: all data generated or analyzed during this study are included in this article. Further inquiries can be directed to the corresponding author.

Abstract

Urticaria is a prevalent inflammatory dermatological condition with a global estimated lifetime prevalence of up to 20%. Yellow urticaria is not a distinct medical condition; rather, it refers to the manifestation of yellow-colored wheals that appear in certain individuals with elevated serum bilirubin levels. This excess bilirubin accumulates in various tissues, including the conjunctiva and skin. The occurrence of yellow urticaria is notably rare and uncommon. In this report, we present a case of yellow-colored urticaria in a patient diagnosed with obstructive biliary stones, complemented by a concise review of previously documented cases in the literature.

Introduction

Urticaria is a prevalent inflammatory dermatological condition with a global lifetime prevalence estimated to be as high as 20%.[1] It is a widely encountered condition clinically characterized acutely by the emergence of wheals, angioedema, or a combination of the two, lasting up to 6 weeks, whereas chronic urticaria is typified by recurrent lesions persisting for more than 6 weeks.[2] A wheal is defined by localized superficial skin swelling, typically encircled by red erythema (flare), accompanied by itching or occasionally a burning sensation. These wheals are transient, returning to normal skin appearance within a timeframe ranging from 30 minutes to 24 hours. Urticaria may be caused by a variety of factors, usually stemming from the activation and degranulation of dermal mast cells, leading to the release of histamine and additional mediators.[2]

Yellow urticaria does not denote a separate disease; instead, it denotes the appearance of yellow-hued wheals observed in specific individuals with heightened levels of serum bilirubin, leading to its *surplus* accumulation in diverse tissues such as the conjunctiva and skin.[3] Other factors contributing to yellow skin discoloration encompass carotenemia, certain medications, and inherited conditions such as Gilbert syndrome.[3] The instance of yellow urticaria is uncommon and rare. Herein, we report a case of yellow-colored urticaria in a patient with obstructive biliary stones.

Case Report

A 24-year-old Saudi male with a known history of bronchial asthma presented at the emergency department at our university hospital complaining of severe epigastric pain following a fatty meal. He reported experiencing symptoms of nausea, vomiting, pale stools, and dark urine. Physical examination revealed an unwell patient with tenderness upon palpation of the abdomen and yellow sclera. The patient's laboratory results revealed significantly elevated levels of alanine aminotransferase (ALT) at

350 U/L (normal range: 7-56 U/L) and aspartate transaminase (AST) at 185 U/L (normal range: 10-40 U/L). Alkaline phosphatase (ALP) was at 145 U/L (normal range: 44-147 U/L). Total bilirubin was markedly elevated at 116 μmol/L (normal range: 3-21 μmol/L), with direct (conjugated) bilirubin also significantly raised at 87 μmol/L (normal range: <5 μmol/L). These findings were consistent with obstructive jaundice, likely caused by biliary obstruction secondary to gallstones, as supported by the clinical presentation and imaging findings. Moreover, the patient presented with notable skin manifestations. Dermatology was consulted regarding his rash. Upon cutaneous examination, multiple urticarial lesions with a yellowish hue and edematous borders were noted over the abdomen and trunk, with positive appreciation of dermatographism (Figure 1). He also reported experiencing generalized pruritus. A skin punch biopsy was obtained for further evaluation. The management plan consisted of 2nd-generation antihistamine (loratadine) 10 mg once daily. Due to the severity of the patient's condition, the patient was then admitted to the surgical ward for further monitoring and management. An endoscopic retrograde cholangiopancreatography (ERCP) was performed for stone removal, aiming to relieve the obstruction in the bile duct.

Histopathological evaluation of the obtained skin lesion demonstrated an unremarkable epidermis with concomitant dermal edema along with predominantly eosinophilic infiltration within perivascular and interstitial spaces (Figure 2). Following the procedure, the yellow urticarial skin lesions disappeared entirely nearly 24 hours after ERCP. A week post-admission, the patient developed acute pancreatitis and was transferred to the intensive care unit, where he underwent a second ERCP and was stabilized. Two weeks after the successful removal of the obstructing stones, the patient underwent an interval laparoscopic cholecystectomy to prevent recurrent biliary complications.

Discussion

This case study highlights the occurrence of yellow-colored urticaria in a patient with obstructive biliary stones, shedding light on a rare and unusual manifestation of urticaria associated with elevated serum bilirubin levels. The presentation of urticaria with yellow-hued wheals is often overlooked or misdiagnosed due to its rarity. Moreover, the association between the emergence of yellow wheals and bilirubin levels lacks clarity. While many patients displaying yellow wheals also present with hyperbilirubinemia and icteric sclera, there have been documented instances of individuals with high bilirubin levels who did not experience yellow wheals, instead exhibiting ordinary wheals without yellow discoloration.[4]

Yellow urticaria is a less frequently encountered form of urticaria, and its etiology remains elusive. It may manifest alongside hepatic and biliary disorders with hyperbilirubinemia frequently identified, often accompanied by jaundiced sclera.[2] Some researchers propose that heightened serum bile acid levels might directly stimulate mast cell degranulation, initiating urticaria. Conversely, others contend that the antioxidant and anti-inflammatory properties of bilirubin could potentially mitigate urticaria symptoms.[5,6] It is also suggested that the yellow discoloration observed in the disease may be attributed to bilirubin having an affinity for binding with elastin in the dermis, resulting in a yellowish hue similar to icteric sclera. Elastin, abundant in the sclera, typically appears yellow when bilirubin levels exceed 3 mg/dL.[7]

Antihistamines represent the primary therapeutic approach for managing yellow urticaria, mirroring the standard treatment for typical non-yellowish urticaria, along with corticosteroids. [2] Identifying and avoiding potential triggers of urticaria is crucial for management. These triggers include physical factors such as dermographism, cold urticaria, pressure urticaria, and others, as well as dietary elements like eggs, milk, soy, and certain medications like aspirin and ACE inhibitors. Environmental factors such as infections, smoking, and pollen should also be considered. Avoiding these triggers can significantly enhance patients' quality of life.[8,9]

This case highlights the intricate interplay between systemic conditions such as obstructive jaundice and its dermatological manifestations. The resolution of yellow urticarial lesions after ERCP suggests a direct correlation between the biliary obstruction and this dermatological manifestation. Through prompt diagnosis and appropriate intervention, his symptoms were effectively managed, emphasizing the importance of a multidisciplinary approach in the care of patients with complex medical presentations.

Conclusions

Yellow-colored urticaria is a rare variant of urticaria. This case underscores the importance of recognizing and investigating this uncommon presentation of urticaria. Based on our experience, wheals of yellowish hue serve as a clinical indicator of underlying internal disease, highlighting the necessity for healthcare practitioners to be vigilant and knowledgeable about this atypical manifestation of urticaria.

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Figure 1. A,B) Physical exam revealed several urticarial lesions with a yellowish hue and swollen edges across the abdomen and trunk, **C)** accompanied by a noticeable presence of dermatographism.

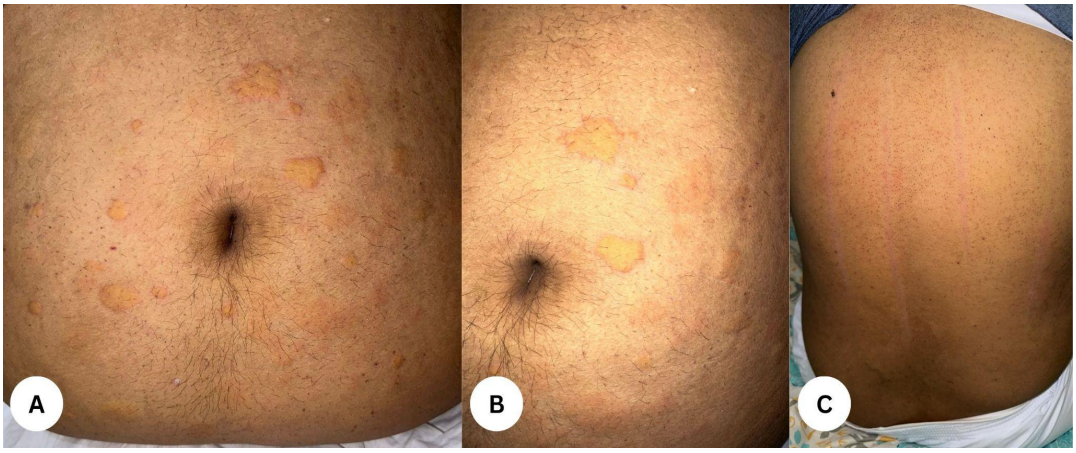

Figure 2. A) Histopathological evaluation of the skin punch biopsy revealed an unremarkable epidermis with underlying dermal edema (H&E stain, original magnification x200), and **B)** a higher power of the dermis shows perivascular and interstitial inflammatory cells, mainly eosinophils (H&E stain, original magnification x400).