

Localized Abdominal Wall Bile Staining Due To Retroperitoneal Bile Leak

Francisco Allegue¹, Lidia Pérez-Pérez¹, María Teresa Maza¹, José Antonio Hermo² and Ander Zulaica¹

Departments of ¹Dermatology and ²Gastroenterology, Complejo Hospitalario Universitario de Vigo, C/ Porriño 5, ES-36209 Vigo, Spain.

E-mail: fallegue@mun-do-r.com

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Sir,

Xanthoderma is any yellow to yellow-orange macular discolouration of the skin. Jaundice and carotenoderma are the primary differential diagnosis in patients with xanthoderma due to a high serum concentration of bilirubin. There are other less common causes of both generalized and localized xanthoderma. Jaundice is generalized skin pigmentation due to high serum concentration of bilirubin, but serum levels below 3 mg/dl may present as conjunctival icterus alone (1). We report here a case of localized yellow pigmentation in the loins, which clinically evoked Grey Turner's sign, a brownish to greenish colouration of the loins.

CASE REPORT

A 70-year-old woman with diabetes and hypertension with a medical history of cholecystectomy was admitted to the gastroenterology ward of our hospital for a choledocolitiasis. She underwent an endoscopic retrograde cholangiopancreatography (ERCP), which showed multiple filling defects in the common bile duct. An endoscopic sphincterectomy was performed and the gallstones were extracted. Severe abdominal pain developed shortly after this procedure. Blood biochemistry showed an increased serum amylase level (2247 UI/ml, normal range 20–115 UI/ml), and gamma glutamyl transferase (172 UI/l, normal range 10–43 UI/l), whereas haemogram, bilirubin, glutamic-oxaloacetic transaminase, glutamic-pyruvic transaminase, lactate dehydrogenase and alkaline phosphatase were within normal ranges, initially suggesting a post-ERCP pancreatitis, which was managed with analgesics and bowel rest. However, computed tomography (CT) scan showed retroperitoneal fluid, mostly in the anterior and right posterior pararenal spaces, with higher density in the latter, extending to the posterior wall muscles. No signs of pancreatitis were evident.

The patient progressively worsened and striking yellowish macules on her loins, more intense in the right side, were noticed 5 days after the ERCP. Those macules were painless, well defined and distributed in a similar fashion as that of the Grey Turner's sign, but they were predominantly yellow (Fig. 1). No changes in colour were noticeable on the periumbilical area. The colour of the sclerae and the rest of the body surface were normal. The serum bilirubin was normal. A CT scan was subsequently performed, showing gas in the area adjacent to the papilla of Vater, which suggested a



Fig. 1. Yellow staining in the right loin.

post-ERCP duodenal perforation, and two large retroperitoneal fluid collections extending into the pelvis, suggesting recent retroperitoneal bleeding.

The patient underwent an urgent surgical intervention, which revealed a large retroperitoneal haematoma affecting the left perirenal area and the right parietocolic ligament. Duodenal perforation was detected and duodenal diversion was performed. The patient continued to improve after surgery and a trans-Kehr cholangiography was performed before she was discharged showing a normal flow into the duodenum without leak.

The patient's yellow macules progressively faded within 4 days, but no other colour than yellow was observed as such cases of haemoglobin degradation. No new yellow macules or bruising were evident in the following days, despite the fact that remnants of the retroperitoneal haematoma were observed in the image studies (CT) performed 1.5 months after discharge.

DISCUSSION

Non-traumatic abdominal wall ecchymoses, though rarely seen, are frequently mentioned in the medical literature as a rare clinical sign of retroperitoneal haemorrhage. Extravasated blood in the retroperitoneal space may spread throughout various anatomical planes and directions to produce subcutaneous ecchymosis. Bruising of the periumbilical region (Cullen's sign) and flanks (Grey Turner's sign) are subcutaneous manifestations of retroperitoneal haemorrhage, often due to severe acute pancreatitis. However, in acute pancreatitis their overall incidence is around 3% and their

presence usually predicts the development of severe complications during the course of the disease, with a mortality rate of 37% (2, 3). Bryant's sign (scrotal ecchymosis), Fox's sign (upper outer thigh bruising) and Stabler's sign (pubis and groin bruising) are additional eponymous signs of retroperitoneal bleeding (4–6).

The mechanisms underlying the appearance of these signs have only recently been clarified with a better understanding of the embryonic development of the different layers of the retroperitoneum. Fluid collections originating in the retroperitoneal space may exit the space by entering dissectible planes that result from the embryological fusion of dorsal mesenteries (7). Thus Grey Turner's sign is due to a retroperitoneal bleeding through the posterior pararenal space, between the two laminae of the posterior renal fascia and the lateral border of the quadratus lumborum muscle and thereafter to the subcutaneous tissues by means of a defect in the fascia of the flank (2, 7–9). Several conditions have been described mimicking Grey Turner's sign, like bacterial peritonitis due to *Pseudomonas aeruginosa* (10) and retroperitoneal necrotizing fasciitis (11). Our patient manifested bilateral flank yellow discolouration, reminiscent of the pattern of ecchymosis seen in cases of Grey Turner's sign, although the pure yellow colour in our patient's loins is quite different from the dirty green discolouration of Grey Turner's sign. We think it was due to a subcutaneous deposit of bile secondary to a duodenal post-ERCP perforation rather than being the result of the haemoglobin degradation, although at laparotomy bile-stained fluid was not found, probably because massive posterior bleeding obscured it.

Other retroperitoneal fluid collections, such as bile, can act as blood, remaining localized in the retroperitoneal space (12) or following natural pathways in the abdominal cavity, reaching the skin and then inducing the development of yellowish macules, in a similar manner as that described in the Grey Turner's sign or other signs of retroperitoneal haemorrhage. Retroperitoneal bile leak is due to rupture of the bile tree or a duodenum perforation. Blunt abdominal trauma, peptic ulcer disease and endoscopic sphincterotomy, as in the present case, are the major causes of duodenal perforation and consequently collections of pathological gas, bile and pancreatic juice may occur in this space and also may exit as described in retroperitoneal bleeding (7). Because the retroperitoneal fascial planes traverse the midline, interfascial fluid collections can spread from the abdominal retroperitoneum across the midline, thus explaining the fact of bilateral bruising in retroperitoneal haemorrhage or, as in our case, the bilateral yellow staining (7).

Localized staining of the flanks due to bile is an exceptional finding and has scarcely been reported in

the literature (13, 14). In these cases additional yellow staining of the scrotum (13), as a pseudo-Bryant's sign, the groins (14) and pubis (13), mimicking Stabler's sign, and thighs (14), a modified Fox's sign, were described. Moreover, before Cullen's description in 1918 of the ecchymoses in the periumbilical area, Ransohoff described in 1906 a localized yellow umbilical stain in a patient with a ruptured common bile duct (15). Those findings support the similar behaviour of pathological retroperitoneal fluid collections and being aware of them is of critical importance, as their presence can reveal an unknown bile leak, or bleeding, with the consequent need for urgent surgery.

The authors declare no conflict of interest.

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