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CLINICAL CASE

Spontaneous retroperitoneal biloma: A case report☆

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KEYWORDS
Bile duct diseases;
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Abstract
Background: Non-traumatic spontaneous rupture of the biliary tract and retroperitoneal accumulation (retroperitoneal biloma) is an extremely rare condition.
Clinical case: A 57 year-old woman with no known biliary disease, started with intense pain in the right abdomen 30 days prior to consultation. She also had jaundice (4+). The initial hepatobiliary ultrasound reported choledocholithiasis and retroperitoneal fluid collection, which was confused with a peri-renal abscess. Guided puncture was performed and the presence of bile was evident. Dilation of the bile duct was observed in the computed tomography. The patient underwent laparotomy to correct both conditions.
Conclusion: The retroperitoneal biloma, also called choleretroperitoneum, is of multifactorial origin. Clinical presentation is non-specific, with diffuse abdominal distension and pain in all patients. The diagnosis is made based on ultrasonography and computed tomography, and can even be diagnosed intra-operatively. The treatment is based on liquid bile drainage and correction of the leak.

PALABRAS CLAVE
Enfermedades de los conductos biliares;
Colección de líquido;
Retroperitoneal;
Biloma

Biloma retroperitoneal espontáneo: reporte de un caso

Resumen
Antecedentes: La rotura espontánea no traumática de las vías biliares y su acumulación en retroperitoneo (biloma retroperitoneal) es una condición infrecuente.

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Caso clínico: Mujer de 57 años de edad, no conocida como portadora de enfermedad biliar, que inició padecimiento 30 días previos a consulta, con dolor intenso en hemiabdomen derecho y posteriormente ictericia (4+). En el ultrasonido hepatobiliar inicial se reportó: coledocolitiasis y colección de líquido retroperitoneal, que se confundió con un absceso perirrenal roto. Se realizó punción guiada, con la cual se evidenció presencia de bilis. En la tomografía axial computada fue evidente la dilatación de la vía biliar. La paciente fue intervenida mediante una laparotomía para resolver ambas dolencias.
Conclusión: El biloma retroperitoneal, también llamado coleretroperitoneo, es de etiología multifactorial. Su presentación clínica es inespecífica y cursa con distención y dolor abdominal difuso en todos los pacientes. Su diagnóstico se realiza a base de ultrasonografía y tomografía axial computada, e incluso puede ser diagnosticado en el transoperatorio. Su tratamiento se basa en el drenaje del líquido biliar y en la corrección de la fuga.

Background

The term biloma refers to a collection of liquid bile outside the biliary tract, which may or may not be encapsulated.[1–3] The condition is associated with rupture of the biliary tract due to trauma, abdominal surgery, duodenal diverticula, or biliary disease, and very occasionally presents spontaneously.[3] Non-traumatic spontaneous rupture of the bile ducts can occur extrahepatically or intrahepatically. It can manifest as an intraperitoneal or retroperitoneal collection; the latter does not compromise the peritoneal cavity, as occurred with our patient.

According to Cólović and Persić, the word biloma was first used in 1979 by Gould and Patel.[4] However, reports have been found dating back to the 19th century,[3] although it is rare and there are few cases in the literature. We present a clinical case and review of the subject.

Clinical case

A 57-year-old woman, whose clinical symptoms started with intense pungent-type pain, irradiating towards the right iliac fossa and lumbar region, of 30 days' onset. She was managed with proton pump inhibitors, and analgesics. However, the pain worsened, the patient had little tolerance to food, and her general condition deteriorated. Three days earlier the patient had started with jaundice, acholia and choluria, which is when we became involved in the case.

The patient was not known to be a carrier of biliary disease, and had a history of hysteroopherectomy for benign conditions; 2 pregnancies, 2 births; smoker, up to 10 cigarettes per day, no detectable pathological history or relevant family history.

On physical examination we found: normal vital signs, conjunctival jaundice (4+), normally hydrated oral mucosa. Uncompromised cardiopulmonary system, distended abdomen, resistance and induration in the right hemiabdomen, pain on superficial palpation, dullness to percussion and bowel sounds present and normal.

The laboratory tests on admission reported: leukocytes $11.42 \times 10^3/\mu l$, neutrophils $9.6 \times 10^3/\mu l$, haemoglobin 12.59 g/dl, platelets $738.1 \times 10^3/\mu l$, glucose 103 mg/dl, cholesterol 330 mg/dl, LDL 336 mg/dl, albumin 2.9 g/dl, globulin 4.2 g/dl, total bilirubin 13.5 mg/dl, direct bilirubin 9.9 mg/dl and indirect bilirubin 3.6 mg/dl, alkaline phosphatase 1.029 IU/l, gamma-glutamyl transpeptidase 1.090 IU/l, aspartate aminotransferase 260 IU/l and alanine aminotransferase 316 IU/l. Prothrombin time 15.6 s, partial thromboplastin time 31.4 s (amylase and lipase were not performed). General urine test with 4 mg/dl bilirubin. The remainder of the results were within normal limits.

Abdominal ultrasound reported choledocolithiasis and perirenal abscess ruptured into the right paracolic gutter.

On arrival at hospital the patient underwent an abdominal ultrasound-guided aspiration of the retroperitoneal fluid, a bile-like fluid was obtained that was sent to the laboratory for culture and gram staining, and was negative for bacteria.

Computed axial tomography revealed marked dilation of the intra and extrahepatic biliary tract, without identifying the gallbladder; in addition, fluid was observed in the anterior and posterior pararenal area (Figs. 1–3).

Retrograde endoscopic cholangiopancreatography revealed dilation of the extrapancreatic and common hepatic bile duct, with a large stone inside it. No leakage of contrast was observed and neither could the gallbladder be seen. Due to the large size of the stone, we did not attempt to extract it via this approach. We placed a 10fr biliary endoprosthesis, 10 cm in length, to prevent cholangitis (Fig. 4).

A laparotomy was then performed via a right subcostal incision and we approached the peritoneal cavity. We incised the right parieto-colic gutter and the hepatic angle of the colon, to approach the retroperitoneum, from which we drained a clear, greenish fluid. The space that contained the fluid was dissected towards the right iliac fossa and psoas, respecting the pancreatic space.