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Case Report

Spontaneous retroperitoneal bilious collection: A case report

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الملخص

الورم الصفراوي هو تراكم مغلف للصفراء في البطن. يأتي الورم الصفراوي بشكل عام بعد انفتاح القناة الصفراوية. كانت الحالة السريرية لهذا التقرير هي امرأة تبلغ من العمر 75 عاما تعاني من اليرقان والحمى وآلام في البطن والخمول وفقدان الشهية وقد تم إدخالها إلى قسم الطوارئ. أظهر "تصوير الطرق الصفراوية والبنكرياسية بالتنظير الداخلي بالطريق الراجع" تمددا شديدا منتشرا للقناة الصفراوية المشتركة والقناة الكبدية الشائعة مع عيوب الملء. تم استخراج حجر واحد وبعض الحمأة والقيح بعد الضرب بالبالون. تم إجراء وضع دعامة بلاستيكية للقناة الصفراوية المشتركة. أظهر الفحص بالموجات فوق الصوتية للبطن وجود حصوة في القناة الصفراوية المشتركة وتجمع سوائل في المنطقة المحيطة بالكلية. بتوجيه من الموجات فوق الصوتية، تم إدخال أنبوب تصريف داخل الورم الصفراوي خلف الصفاق. كشف الفحص بالموجات فوق الصوتية التسلسلية أن حجم الورم الصفراوي انخفض تدريجيا. يعتبر ثقب القناة الصفراوية الذي يتميز بتجمع السائل خلف الصفاق حالة نادرة للغاية ويمكن أن تكون قاتلة. تم استخدام التصوير بالموجات فوق الصوتية والتصوير المقطعي وتصوير الطرق الصفراوية والبنكرياسية بالتنظير الداخلي بالطريق الراجع لتشخيص الحالة. يشمل العلاج منع التسرب وتصريف الصفراء.

الكلمات المفتاحية: الورم الصفراوي؛ الحيز خلف الصفاق؛ تراكم العصارة الصفراوية؛ القناة الصفراوية المشتركة؛ تصوير الطرق الصفراوية والبنكرياسية

Abstract

Biloma refers to the encapsulated accumulation of bile in the abdomen. Bilomas generally occur after bile duct disruption. The clinical case of the present report was a 75 Y/O woman with jaundice, fever, abdominal pain, lethargy, and anorexia who had been admitted to the emergency department. Endoscopic retrograde cholangiopancreatography (ERCP) showed severe diffuse dilation of the common bile duct (CBD) and common hepatic duct (CHD) with filling defects. One stone and some sludge and pus were extracted after balloon swiping. Plastic CBD stent placement was performed. An abdominal ultrasound scan showed a stone in the CBD and fluid collection in the right perirenal space. Under ultrasound guidance, a drainage tube was inserted into the retroperitoneal biloma. A serial ultrasound scan revealed a gradual decrease in the size of the biloma.

Perforation of the bile duct, which is characterised by the collection of retroperitoneal fluid, is considered an extremely rare condition that can be fatal. Ultrasonography, computed tomography, and endoscopic retrograde cholangiopancreatography were used for the diagnosis. The treatment includes prevention of leakage and bile drainage.

Keywords: Bile accumulation; Biloma; Cholangiopancreatography; Endoscopic retrograde; Hepatic duct, Common; Retroperitoneum

Introduction

Biloma refers to the encapsulated accumulation of bile in the abdomen. Bilomas generally occur after bile duct disruption. The existing literature shows that the main causes

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Table 1: Lab tests result on admission.

Lab tests	Results	References	Unit
Leukocytes	15	4–10.5	$10^3/\mu l$
Neutrophils	90.8	54–62	%
Haemoglobin	12.1	12.5–16	g/dl
Platelets	172	150–400	$10^3/\mu l$
Albumin	2.9	3.5–5.2	g/dl
Amylase	38	Up to 90	Iu/L
Total bilirubin	3.59	0.3–1.2	mg/dl
Direct bilirubin	2.35	Up to 0.4	mg/dl
Indirect bilirubin	1.24	0.2–0.8	mg/dl
Alkaline phosphatase	249	Up to 240	IU/l
Aspartate aminotransferase	13	Up to 31	IU/l
Alanine aminotransferase	64	Up to 31	IU/l
Prothrombin time	15.9	12–14	sec
Partial thromboplastin time	44.4	25–38	sec
C-reactive protein	61.4	0–10: Neg	mg/L

of this problem include iatrogenic (following hepatobiliary surgery), trauma, or problems due to choledocholithiasis.[1]

Retroperitoneal accumulation of bile and non-traumatic rupture of the biliary tract, which is spontaneous, is an extremely special condition. Impulsive perforation of the bile duct is not common among adults, particularly in cases that accompany retroperitoneal biloma. In this study, a patient with retroperitoneal biloma due to cholangitis and choledocholithiasis is reported.

Case presentation

A 75 years old woman was admitted to the emergency department with abdominal pain that had begun 72 h before admission. Along with pain, jaundice, fever, lethargy, and anorexia were observed. The patient's medical history showed Hypertension and open cholecystectomy 15 years prior. Examination of the patient revealed distention of the patient's abdomen. In addition, the right upper quadrant of the abdomen and the epigastric region were tender. In the abdomen, no rebound tenderness or guarding was observed. The patient had a fever of 38.9 °C.

Lab test results are presented in Table 1. The rest of the laboratory test results were normal.

An ultrasound scan of the abdomen revealed fluid accumulation in the right perirenal space, dilatation of intrahepatic and extrahepatic bile ducts, and sludge and a stone in the CBD (Figure 1).

Computed tomography revealed fluid collection in the anterior and posterior pararenal spaces on the right side, as well as dilatation of the biliary tract (Figure 2).

Endoscopic retrograde cholangiopancreatography (ERCP) revealed severe diffuse dilation of the common hepatic duct (CHD) and common bile duct (CBD) containing filling defects. A sphincterotomy was performed. After balloon sweeping, one stone was extracted, and some sludge and pus were extracted. Plastic CBD stent placement was performed.

In percutaneous aspiration of the retroperitoneal fluid, which was carried out by ultrasound, a bilious fluid was found to be negative for bacteria. Regarding the diagnosis of retroperitoneal biloma, re-ERCP was performed, which revealed diffuse dilation of the bile ducts. The previous CBD stent was removed. No obvious leakage was observed. Because of the symptoms of cholangitis, a plastic stent was placed. A drainage tube was inserted into the retroperitoneal biloma under ultrasound guidance. The drainage output was bilious coloured. In serial ultrasound scans, the size of the biloma decreased gradually. Despite the removal of the drainage tube, a follow-up ultrasound scan after one month showed no fluid collection in the abdomen.

Discussion

A biloma refers to an accumulation of bile that occurs in the exterior parts of the biliary system and in the abdominal area and may be extrahepatic or intrahepatic. The most prevalent causes of biloma are hepatobiliary interventions and laparoscopic cholecystectomy, after which choledocholithiasis, abdominal trauma, and biliary dilatation following biliary stricture are observed.[2] Most bilomas are iatrogenic or traumatic.[3] Spontaneous biloma is not common, and most of them are observed with choledocholithiasis or cholangiocarcinoma.[4]

The major perforation site of the biliary tract is the GB, and the cause is usually cholecystitis and cholecystolithiasis.[5] The CBD was another perforation site. Bile duct perforation

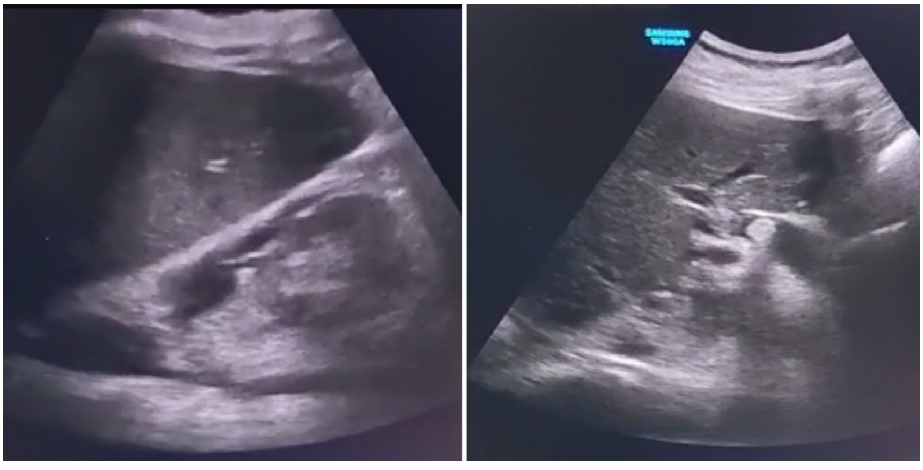

Figure 1: Fluid collection in right perirenal space and dilated CBD containing a stone.

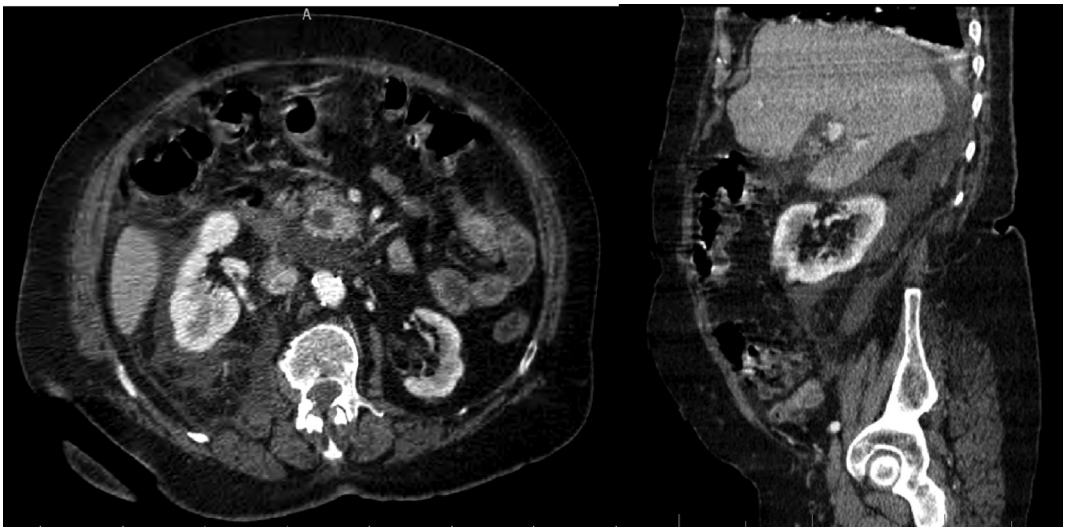

Figure 2: Retroperitoneal fluid collection.

is shown as either localised fluid accumulation or general biliary peritonitis.[6]

The reason for bile leakage may be the retroperitoneal portion.[7] One pathophysiological cause is a perforation in the gallbladder. This adheres to the parietal peritoneum through a chronic inflammatory process. When this happens, communication exists between the retroperitoneum and the biliary tract.[8]

As it is known, the most common cause of spontaneous biloma is choledocholithiasis.[4,9] On the other hand, the secondary cause of spontaneous bile duct perforation may be due to the erosion of gallstones. Increased intraductal pressure may also be due to obstruction of the distal bile duct (due to a stone, mass, or spasm of the ode sphincter), thrombosis of a vessel supplying the bile duct wall, cholangitis, regurgitation of pancreatic secretions into the bile duct, choledochocele, and acute pancreatitis.[10–12]

In the present case, a stone existed in the CBD; however, no obvious bile leak was observed on ERCP. Ultrasound and computed tomography (CT) scan showed no choledochal cysts and no evidence of pancreatitis. Perforation of the biliary tract may be caused by choledocholithiasis or cholangitis.

In terms of incidence, there is no difference between females and males; however, the condition is usually observed in the age period of the 60s and 70s. The presentation is nonspecific, with abdominal pain, usually in the right upper quadrant. Fever may accompany abdominal distension and jaundice.[13] In the present case, a 75 Y/O woman with pain in the right upper quadrant of the abdomen and fever with jaundice was admitted.

Blood tests may reveal an increase in CRP and leukocyte counts and liver function tests. In the present case, the CRP level was elevated to 61.4 mg/L and liver function tests were disrupted. Gram-negative bacteraemia may be revealed by blood cultures.

Biloma may be detected by magnetic resonance imaging, ultrasound, or CT scan. Abdominal ultrasound is the first imaging modality that leads to the suspicion of a choleretroperitoneum, because it identifies perirenal and retroperitoneal fluid accumulation, which may be misunderstood as a renal abscess. In most cases, dilatation of the bile ducts is observed, and[3,14] retroperitoneal bile will be revealed by needle aspiration of the collection. In the present case, a diagnosis of biloma was made after percutaneous aspiration.

ERCP can help identify an active leak that can also be therapeutically intervened.[15] Identifying the leakage site will also be difficult because, in most cases, it will have closed.[16] Endoscopic treatment in cases where it is suitable includes sphincterotomy with stone extraction to lower biliary pressure. As another possible treatment, a stent can be placed in more distal lesions, since this results in a reduction in the pressure gradient into the duodenum and simplifies the flow of bile. This also releases obstruction from lesions that tighten the biliary tree.

In this case, due to the patient's cholangitis, imipenem was prescribed empirically. Also, apart from external drainage, biliary stent was inserted. Non-surgical intervention before definitive biliary surgery is considered the first choice for treating biloma, as done in this case.

Conclusion

Perforation of the bile duct, which is characterised by the collection of retroperitoneal fluid, is considered an extremely rare condition, which can be fatal. Biloma should be considered in the differential diagnosis of retroperitoneal fluid collection without an air bubble.

The diagnosis and treatment of damages to the biliary tract are considered a challenge. Luckily, the process of diagnosis and treatment has been improved by new technologies, and many patients are treated without sequelae. In this case, after non-surgical interventions, the size of the biloma on serial ultrasound scans decreased gradually and disappeared eventually.

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Conflict of interest

The authors have no conflict of interest to declare.

Ethical approval

The patient was aware and content that her clinical data would be used for publication purposes and provided her consent.

Consent

Informed consent was obtained from the patient for this case report publication and any accompanying images.

Authors contributions

SAMY conceived and designed the analysis; SAMY and GM collected the data; MR and GM contributed data and wrote the paper. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

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