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Retroperitoneal Biloma due to Spontaneous Perforation of the Left Hepatic Duct

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
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Conflict of interest: None declared

Patient: Male, 82
Final Diagnosis: Retroperitoneal biloma due to spontaneous perforation of the left hepatic duct
Symptoms: Abdominal pain • high fever
Medication: —
Clinical Procedure: Emergent operation
Specialty: Gastroenterology and Hepatology

Objective: Rare disease
Background: Spontaneous perforation of the bile duct in adults is very rare, particularly in cases accompanied by retroperitoneal biloma. We report a patient with retroperitoneal biloma due to a spontaneous perforation of the left hepatic duct.

Case Report: An 82-year-old man was admitted to our institution with abdominal pain and a high fever. He had tenderness at the epi-mesogastrium. Computed tomography showed several stones in the gall bladder and common bile duct (CBD) and a few ascites. A substantial amount of fluid had collected from the dorsal stratum of the duodenum and pancreas head to the right paracolic gutter and anterior side of the right iliopsoas. Laboratory examination revealed a high inflammation score. He underwent emergent laparotomy. Biliary fluid was revealed after the mobilization of the pancreas head, duodenum, and right side of the colon. Bile duct perforation was suspected. Therefore, we exfoliated the dorsal side of the CBD to the cranial side, and intraoperative cholangiography was performed. However, the perforation site could not be detected. Cholecystectomy and choledocholithotomy were performed. A retrograde transhepatic biliary drainage tube was inserted, and primary closure of the CBD incision site was achieved. Postoperative cholangiography revealed leakage from the left hepatic duct near the caudate branch.

Conclusions: There are a few reports of spontaneous bile duct perforation cases in the literature, particularly on infants or children with congenital anomalies, but it is rare in adults. It usually causes bile peritonitis, although bile duct perforation should be considered in the differential diagnosis of spontaneous retroperitoneal fluid collection in adults.

MeSH Keywords: Bile Ducts, Intrahepatic • Retroperitoneal Space • Rupture, Spontaneous

Full-text PDF: <http://www.amjcaserep.com/abstract/index/idArt/897612>

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Background

Spontaneous perforation of the bile duct is very rare in adults. Most of the bile duct ruptures usually occur secondly to blunt or penetrating abdominal traumas, hepatobiliary operations, and instrumentation, and is more often seen in infants and children [1–3]. Patients with bile duct rupture mostly present with acute symptoms, fulminant bile peritonitis with pain, vomiting, fever, and abdominal distention. However, retroperitoneal biloma due to spontaneous perforation of the bile duct is very rare. Spontaneous perforation of the bile duct is difficult to diagnose preoperatively. Most patients are preoperatively diagnosed as having cholecystitis or choledochocystitis. These prognoses are inaccurate due to the prolonged duration of the symptoms [4].

Here, we report a patient with retroperitoneal biloma as a result of perforation of the left hepatic duct that was successfully treated with surgical intervention.

Case Report

An 82-year-old Japanese man was admitted to our institution due to abdominal pain lasting for 3 days and a high fever. He had a past medical history of emphysema, asthma, unstable angina, and gastric ulcer perforation for which he underwent an operation. He was an ex-smoker and daily drinker (2 cans of beer a day). He had a temperature of 38.7°C. An abdominal examination revealed distention and tenderness from the right hypochondrium to the midline. On laboratory examination, WBC was 14 700/mm³, CRP was 29.4 mg/dl, total bilirubin was 2.2 mg/dl, AST and ALT were normal, γ GTP was 220 IU/l, Cre was 1.44 mg/dl, and the myocardial marker level was normal. Although we performed an abdominal ultrasound examination, the upper right quadrant was nearly impossible to clearly visualize because of the presence of intestinal gas.

A computed tomography (CT) scan revealed several stones in the gallbladder (GB) and common bile duct (CBD) and retroperitoneal fluid collection extending from the dorsal stratum of the duodenum and pancreas head to the right paracolic space and anterior side of the right iliopsoas. There was no dilatation of the intrahepatic and extrahepatic bile duct (Figure 1).

Hemorrhage to retroperitoneal stratum, damage of the duodenum, and spread of colitis or appendicitis were considered in the differential diagnosis of the lesion. Our patient was elderly and had severe inflammation; therefore, we elected to perform a laparotomy.

We performed a median incisional laparotomy. There were a few ascites. The GB had a thick wall but was not perforated. After the mobilization of the pancreas head, duodenum, and right side of the colon, a substantial amount of bile was found. We suspected that perforation of the bile duct had occurred. Therefore, we exfoliated the dorsal side of CBD to the cranial side, and intraoperative cholangiography was performed. However, the site of perforation could not be detected. Additionally, we performed an intraoperative choledochoscopy, although the perforated site could not be found. Cholecystectomy and choledocholithotomy were performed, a retrograde transhepatic biliary drainage tube (RTBD tube) was inserted, and primary closure of the incision site of the CBD was achieved. After intraperitoneal irrigation with 5 L of saline, we inserted drains to the right paracolic gutter and Winslow's foramen.

In the postoperative course, we performed cholangiography, which revealed a leakage from the left hepatic duct near the caudate branch (Figure 2). On postoperative day 5, the patient had a fever that reached 38.6°C and a high inflammation score upon laboratory examination. A CT scan revealed an abscess formation that was remaining at the ventral side of the inferior vena cava and right iliopsoas (Figure 3). We adjusted the drain in Winslow's foramen further inside to improve drainage

Figure 1. (A–C) An abdominal computed tomography scan. These figures show several stones in the gall bladder and common bile duct, as well as retroperitoneal fluid collection extending from the dorsal stratum of the duodenum and pancreas head to the right paracolic space and anterior side of the right iliopsoas.

Figure 2. Postoperative cholangiography showing leakage from the left hepatic duct near the caudate branch.

of bile spillage from the left hepatic duct and the drain at the right paracolic gutter further caudally with a use of a guide wire, and the patient gradually recovered. On postoperative day 30, the RTBD tube was withdrawn following a cholangiography that revealed no leakage from the biliary duct. At a 6-month follow-up examination, the patient was in good health, and the laboratory examination revealed normal liver function.

Discussion

Spontaneous perforation of the bile duct in adults unrelated to iatrogenic injury or severe trauma is extremely rare and is

more often seen in infants and children [1,2,5]. Freeland reported the first case of spontaneous perforation of the hepatic duct in 1882, diagnosed during an autopsy [6]. The most common perforation site of the biliary tract is the GB and the cause is typically cholecystitis and cholelithiasis [7]. Other perforation sites are the intrahepatic bile duct and extrahepatic bile duct. To date, 23 cases of intrahepatic bile duct perforations and 54 cases of extrahepatic bile duct perforation in adults have been reported [8–15]. Several causes of spontaneous perforation of bile duct have been proposed: (1) erosion by biliary stones that injured the duct wall; (2) increased intraductal pressure due to an obstruction of the distal bile duct (by stones, carcinomas, or a reflux spasm of the sphincter of Oddi); (3) thrombosis of a vessel supplying the bile duct wall; (4) intramural infection of the duct as a result of cholangitis; (5) regurgitation of pancreatic secretions into the bile duct; (6) diverticulitis of the bile duct; and (6) acute pancreatitis [4,15–17].

The most common perforation site is the CBD. Bile duct perforation presents as either a localized fluid collection or generalized biliary peritonitis. Therefore, only a few cases that have described the presence of fluid collection in retroperitoneum space in spontaneous perforation of the bile duct have been published. To date, only 6 cases of spontaneous perforation of the bile duct with retroperitoneal fluid collection have been reported [8,18–22]. It is very difficult to diagnose a spontaneous perforation of the bile duct presenting with retroperitoneal fluid collection preoperatively. In this case, the preoperative diagnosis could not be obtained, and an emergency laparotomy was performed. If the preoperative diagnosis had been obtained, an endoscopic retrograde cholangiogram could have been performed.

Figure 3. (A, B) A postoperative computed tomography scan showing abscess formation at the ventral side of the inferior vena cava and right iliopectas.

According to the surgical treatment, the repair of the perforation site and T-tube insertion was most frequently reported [4,10–16,23]. In our case, an accurate site of the perforation could not be detected. Therefore, we performed the insertion of a retrograde transhepatic biliary drainage tube. If there is a large defect, a more invasive operative method should be considered, such as a hepaticojejunostomy, hepatoduodenostomy, or flap patch [11,23]. Appropriate and careful drainage via the insertion of drains is very important, particularly in cases with retroperitoneal fluid collection, because it is difficult due to the narrow space. In this case, the position of the drain, which was located on the dorsum of the duodenum and pancreas head, was slightly shallow; therefore, we had to reposition it.

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Conclusions

Spontaneous perforation of the bile duct presenting retroperitoneal fluid collection is an extremely rare and potentially fatal condition. It is very important to consider it in the differential diagnosis whenever retroperitoneal fluid collection is present without an air bubble.

Declaration of interests

Kenjiro Ishii and other co-authors have no conflict of interest to declare.