A 77-Year-Old Woman Presenting with Abdominal Pain, Distension, and Rigors due to Extrahepatic Biliary Leak, 9 Years Following Elective Laparoscopic Cholecystectomy

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Patient:
Female, 77-year-old

Final Diagnosis:
Biliary leakage

Symptoms:
Abdominal distension • abdominal pain • nausea

Clinical Procedure:
ERCP • laparoscopic washout

Specialty:
Gastroenterology and Hepatology

Objective:
Rare disease

Background:
Biliary leaks are an important cause of the acute abdomen condition, carrying significant levels of morbidity and mortality. They are most commonly the result of acute iatrogenic or blunt trauma, with an identifiable cause. In comparison, spontaneous and delayed biliary leaks are rare phenomena occurring in the absence of recent surgery, endoscopic intervention, or trauma. Here we report the case of a 77-year-old woman presenting with acute abdominal pain, distension, and rigors caused by an extrahepatic biliary leakage 9 years after laparoscopic cholecystectomy.

Case Report:
Laparoscopic hepatobiliary intervention, rather than open surgery, is associated with increased risk of biliary tree injury, including biliary leaks, which typically arise in the immediate postoperative period. This report concerns a 77-year-old woman presenting with acute abdominal pain, distension, and rigors due to extrahepatic biliary leakage 9 years following elective laparoscopic cholecystectomy for cholelithiasis. Computed tomography (CT) showed large-volume abdominopelvic ascites with no obvious source. Intra-abdominal exploration revealed a large biloma with 2 lacerations in the gallbladder fossa. Subsequent endoscopic retrograde cholangiopancreatography (ERCP) found only a vague leak in the extrahepatic biliary tree, with no focal defect or retained biliary calculi. Intra-abdominal drainage and common bile duct (CBD) stenting were performed. Repeat ERCP undertaken 3 months later, however, identified a calculus within the CBD.

Conclusions:
The patient’s rare presentation of biliary leakage 9 years after cholecystectomy raises the question of whether the condition was delayed or spontaneous in nature. The biliary calculus identified 3 months following the leakage raises the possibility of retained biliary calculi.

Keywords:
Bile Ducts • Cholangiopancreatography, Endoscopic Retrograde • Rupture

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Background

Biliary leakage is an uncommon but serious cause of the acute abdomen condition, which is associated with a high degree of morbidity and mortality [1]. Biliary leaks are typically attributed to bile duct injury or biliary stricture, which can occur within the intrahepatic or extrahepatic biliary tree, with the latter being the most common site of leakage [2]. Extrahepatic biliary leakage is predominantly caused by iatrogenic trauma, from procedures such as laparoscopic cholecystectomy, percutaneous transhepatic cholangiography, and endoscopic retrograde cholangiopancreatography (ERCP) [3,4]. Intrahepatic biliary leakage, on the other hand, can be caused by hepatic surgery, such as hepatic resection and abscess drainage, in addition to blunt or penetrating abdominal trauma [2].

In comparison with trauma-induced biliary leakage, spontaneous biliary leakage (SBL) is defined as a biliary leak with no identifiable cause, and is usually a diagnosis of exclusion [4]. The first case of SBL was described by Freeland in 1882 [5], in which a 65-year-old woman sustained spontaneous rupture of an intrahepatic duct, diagnosed on autopsy. SBLs are rare amongst adults, with diagnosis most often seen in infant and pediatric populations, usually as a result of congenital defects and anomalies of the hepatobiliary system [1,6]. To date, there have been over 70 cases of SBL reported, with most occurring within the extrahepatic biliary tree [7-11].

A number of potential etiologies have been suggested for why SBLs may occur [1,7,9]. These include the presence of biliary calculi eroding the duct wall, increased intraductal pressure due to distal bile duct obstruction (from calculi, tumors, strictures, or sphincter of Oddi dysfunction), thrombosis of a vessel supplying the bile duct wall, intramural duct infection from cholangitis, regurgitation of pancreatic secretions into the bile duct, bile duct diverticula, and acute pancreatitis [1,7,9,12-17].

This report concerns a 77-year-old woman presenting with abdominal pain, distension, and rigors due to an extrahepatic biliary leak 9 years following elective laparoscopic cholecystectomy.

Case Report

A 77-year-old woman with a prior history of laparoscopic cholecystectomy 9 years previously for cholelithiasis and a previous transient ischaemic attack presented to our hospital with a 3-day history of generalized abdominal pain which had progressively worsened in severity, with no apparent trigger. The pain was accompanied by increasing abdominal distension, nausea, reduced oral intake, and rigors. She reported adequate bowel movements with no lower urinary tract symptoms. There was no history of recent abdominal or pelvic surgery, endoscopic intervention, chronic liver disease, or abdominal trauma.

On examination she was febrile with a temperature of 38.0°C, blood pressure of 140/77 mmHg, and heart rate of 106 beats per minute. There was no clinical evidence of significant jaundice. The abdomen was grossly distended, with guarding and rebound tenderness evident in all 4 quadrants. The patient was admitted under the surgical team for abdominal sepsis, suspected to be due to biliary sepsis or abdominal viscus perforation.

Initial blood tests revealed a white blood cell count of 20,10^9/L (normal reference range: 4-11,10^9/L) and a C-reactive protein of 326 mg/L (normal reference range: 0-4.9 mg/L). Liver function tests highlighted an alanine aminotransferase of 237 units/L (normal reference range: 3-40 units/L), bilirubin of 57 µmol/L (normal reference range: 3-17 µmol/L), and alkaline phosphatase of 247 µmol/L (normal reference range: 30-100 µmol/L). Serum creatinine was 212 µmol/L (normal reference range: 44-80 µmol/L) and urea was 11.8 mmol/L (normal reference range: 2.5-7.8 mmol/L). Serum lipase was within normal limits.

The patient was resuscitated with intravenous (IV) antibiotics in the form of piperacillin-tazobactam and IV fluids. An urgent computed tomography (CT) scan of the abdomen and pelvis displayed a cystic remnant in the porta hepatis consistent with a prior history of cholecystectomy (Figure 1A), and a small volume of free fluid surrounding the liver, from no clear source (Figure 1B).Appearances of the small bowel and intra- and extra-hepatic biliary tree were otherwise unremarkable.

Following CT imaging, a magnetic resonance cholangiopancreatography (MRCP) scan was undertaken, which showed only prominent extrahepatic channels with mild ascites (Figure 2). The common bile duct (CBD) measured 9.5 mm with no evidence of intraductal filling defect or calculus, and there was no evidence of intrahepatic biliary dilatation.

The patient received optimized medical therapy for the following 10 days; however no response to treatment was observed, and her biochemical markers continued to worsen. A repeat CT scan of the abdomen and pelvis demonstrated progressive large-volume abdominopelvic ascites with no obvious source (Figure 3). Following this, the decision was made to proceed to exploratory laparoscopy. Intraoperatively, the abdominal cavity was found to be grossly soiled with bile, with 2 lacerations to the liver at the site of the previous gallbladder fossa. The CBD was grossly distended yet no retained biliary calculi were identified. A total of 4300 ml of bile was removed, and 4 intra-abdominal drains were inserted. Intra-abdominal bile specimens later grew Enterococcus faeci um anaerobically; hence, antibiotic treatment was changed to teicoplanin and meropenem based on sensitivity of this.
species to these antibiotics. Cytological analysis was negative for malignancy.

Following diagnostic laparoscopy and drainage, ERCP was undertaken to localize and treat the source of the biliary leak. ERCP revealed only a vague biliary leak within the extrahepatic biliary tree, with no focal site of leakage. No retained biliary calculi or cause of the biliary leak were found (Figure 4A, 4B). A straight plastic stent was successfully inserted into the CBD (Figure 4C).

Postoperative output from the intra-abdominal drains gradually decreased, and all 4 drains were removed uneventfully within 10 days. The patient’s clinical symptoms and inflammatory markers gradually improved during this period, and she was discharged 1 week later. A repeat ERCP performed 3 months following discharge found a CBD stone which was

Figure 1. A computed tomography (CT) scan of the abdomen and pelvis showing a cystic remnant in the porta hepatis following cholecystectomy (A) and a small volume of free fluid in the upper abdomen surrounding the liver (B).

Figure 2. A magnetic resonance cholangiopancreatography (MRCP) scan demonstrating prominent extrahepatic biliary ducts only, with no clear cause or source of the biliary leak found.

Figure 3. A repeat computed tomography (CT) scan of the abdomen and pelvis displaying large-volume abdominopelvic ascites, of no obvious source.
extracted but no evidence of ongoing biliary leak, hence the biliary stent was removed.

Discussion

This is a case of a 77-year-old woman presenting with a biliary leak 9 years after elective laparoscopic cholecystectomy. The case highlights that biliary leaks can occur in the long-term phase numerous years following hepatobiliary intervention as well as in the acute postoperative period. The potential etiologies of biliary leaks presenting in the long-term phase include spontaneous leaks, such as from retained biliary calculi, as well as delayed leaks from encapsulated bile collections or from progressive hepatobiliary trauma.

The presentation of biliary leaks can be acute or insidious, with the latter being more frequent [18]. Acutely, fulminant biliary peritonitis is typically observed with generalized abdominal pain and distension, bilious or non-bilious vomiting, fevers, and ascites [11]. Insidious SBLs tend to be subacute or chronic in onset, presenting with painless and progressive abdominal distension accompanied by jaundice and pale-colored stools [2]. SBLs can also be asymptomatic with incidental diagnosis being made on imaging studies, depending on the anatomical location and size of the leak [3].

The clinical presentation of our patient’s case was more consistent with an acute biliary peritonitis picture. The initial small volume of ascites highlighted on CT and MRI was perhaps misleading, as this was thought to not be large enough to originate from a perforated viscus, and thus was assumed to have limited clinical significance. However, given the severity of the sepsis and derangement of liver function tests in the presence of relatively unremarkable hepatobiliary imaging, it could be postulated that exploratory laparoscopy would ultimately be required for both diagnosis and management. Nonetheless, the 10-day period of medical therapy and close clinical observation was crucial in indicating this need. The growing volume of biliary ascites during this period and its lack of response to

Figure 4. Endoscopic retrograde cholangiopancreatography (ERCP) highlighting a vague biliary leak in the extrahepatic tree (A, B), and biliary stent in situ in the common bile duct (C).
medical therapy was manifested by ongoing clinical evidence of peritonitis and rising inflammatory markers. This raised the suspicion of a spontaneous or delayed biliary leak, and thus the need for exploratory laparoscopy.

Hepatobiliary surgery is not without its complications [19]. Bile duct injury is among the most common complications arising from hepatobiliary intervention [19-21]. Bile duct injury can be incomplete, involving a partial circumference of the duct, or complete, resulting in the duct being divided [19]. Both incomplete and complete bile duct injuries can lead to biliary leaks, the development of a biloma, and life-threatening biliary peritonitis [4,7,20]. Biliary fistulae can also arise from biliary leaks, due to increased intraductal pressures as well as unsuccessful clipping of cystic duct stumps or thermal lesions of bile ducts [20].

The occurrence of biliary leaks increases mortality following hepatobiliary intervention [22]. A study by Vining et al [22] of outcomes after elective hepatectomy in over 15,000 patients found a significantly increased 30-day mortality in patients who sustained biliary leaks versus those who did not (2% vs 0.9%, P<0.001) [22].

There are several potential hypotheses as to the etiology of our patient’s biliary leak with respect to her laparoscopic cholecystectomy performed 9 years previously. Firstly, the cystic remnant in the porta hepatis visible on CT imaging may have in fact represented a chronic encapsulated collection of bile formed following laparoscopic cholecystectomy. In the latter years this collection of bile may have subsequently become infected, enlarged, and perforated, which would account for the lack of a specific site of leakage in the biliary tree.

Another potential explanation for the biliary leak is minor hepatobiliary trauma following surgery, which gradually progressed over the long term, eventually leading to bile duct perforation. Two case reports published by Kaffes et al [21] and Gundara et al [23] describe biliary leaks 4 and 6 years after open cholecystectomy and elective laparoscopic cholecystectomy, respectively. This raises the hypothesis that the biliary leak was delayed as opposed to being spontaneous. It is thus pertinent to be aware that iatrogenic biliary leaks can develop chronically, with biliary trauma or defects gradually increasing in size and clinical significance [21-24].

The possibility of retained choledocholithiasis after laparoscopic cholecystectomy could also account for the patient’s biliary leak. Our patient had a history of cholelithiasis, which was the primary indication for laparoscopic cholecystectomy. Given this history, it is plausible that the patient had retained stone(s) within the biliary tree which eroded through the duct wall and later entered the abdominal cavity, thus going undetected and causing biliary peritonitis. This is supported by the repeat ERCP performed 3 months following discharge, which discovered a calculus within the CBD that may not have been visualized on the first ERCP.

Furthermore, chronic pancreatitis secondary to numerous pathologies including choledocholithiasis remains an alternative explanation for SBLs. Yasar et al [8] highlighted the case of an SBL in a patient 8 years following laparoscopic cholecystectomy for cholelithiasis. The patient was found to have pancreatic calcification and atrophy consistent with chronic pancreatitis on CT imaging; however, no calculi were identified radiologically or intraoperatively. Chronic pancreatitis is known to increase the risk of biliary strictures and bile reflux, both of which can increase intraductal pressure leading to SBLs [24]. Despite this, the CT and MRCP imaging for our patient failed to reveal definitive evidence of chronic pancreatitis or biliary strictures, making this a less likely etiology.

The diagnosis of SBL can be established preoperatively, intraoperatively, or postoperatively through several different investigation modalities [3,4]. For example, the use of hepatobiliary iminodiacetic acid imaging with a radioactive tracer has localized the site of SBL prior to surgery or ERCP [3,4,6]. In comparison, abdominal and pelvic CT imaging can confirm fluid collections, which may represent intra-abdominal bile. However, CT is typically unable to identify the focal site of SBL.

Several techniques can be utilized for intraoperative diagnosis of SBLs. Direct bile duct exploration, choledochoscopy, and intraoperative cholangiography can localize SBLs [1,8,9].

Despite the benefits of non-invasive imaging and intraoperative exploration, the diagnosis of SBLs is fundamentally dependent on ERCP [7,12,13]. Multiple cases of SBL have been diagnosed only after ERCP was performed, with CT imaging, MRCP, and even intraoperative bile duct exploration failing to identify the biliary leakage [7,12,19,20].

The management of choice in most cases of delayed or spontaneous biliary leak has been surgical drainage in the form of laparoscopy or laparotomy, followed by ERCP and temporary biliary stenting to close the site of the leak [1,6-9,11,12,17,19]. Surgical and/or endoscopic intervention is accompanied by broad-spectrum intravenous antibiotics, given that bacteremia caused by biliary leaks can consist of gram-positive, gram-negative, and/or anaerobic bacteria [25]. Penicillin, cephalosporin, aminoglycoside, lincosamide, fluoroquinolone, and carbapenem antibiotics have all been found to have adequate penetration for treating sepsis-related biliary leaks [25].

Although the management of choice mirrors our patient’s treatment, recent advances in endoscopic ultrasound (EUS)
have provided a highly successful and safer management option for the diagnosis and management of biliary leaks [26-29]. EUS-guided biliary drainage is particularly useful in complicated cases of biliary leak, in which ERCP has been unsuccessful, such as a failure to access the duodenal papilla due to duodenal stenosis or obstruction, or post-surgical changes in local anatomy [26,27]. EUS-guided biliary drainage can be performed using several methods. This includes EUS-ERCP rendezvous, a combined approach that entails EUS to puncture the culprit duct allowing for guidewire insertion through the defect to the duodenal papilla for subsequent rendezvous with ERCP [26]. Alternatively, the creation of a temporary or permanent fistula between the culprit duct and either the gastric or small bowel wall can be made, allowing biliary stent placement. Several studies have found EUS to have a sound success rate, ranging from 73 to 97% [27-29].

Conclusions

Biliary leaks are typically regarded as acute complications of iatrogenic or blunt trauma. Yet, they can present many years after hepatobiliary intervention. In this chronic phase, biliary leaks can be delayed or spontaneous in nature. In our patient’s case, the biliary leakage could be the result of retained biliary calculi 9 years following laparoscopic cholecystectomy.

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Declaration of Figures’ Authenticity

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