A case of late presentation of recurrent primary choledocholithiasis 30 years post-cholecystectomy: Presentation and management

Devin Bird BS, Haylee Flournoy MD, Robyn Tapp MD, Anna Rossini BS, Gamal Amir MD, FACS, Basem Soliman MD

ABSTRACT

Acute cholangitis typically occurs secondary to biliary obstruction and bile stasis. While the most common cause is secondary choledocholithiasis (i.e., stones form in the gallbladder and are expelled into the common bile duct), there is little information on primary choledocholithiasis as the principal source of this obstruction. Furthermore, it is particularly rare to see symptomatic choledocholithiasis years to decades later in patients who previously underwent cholecystectomy. We report a complex case of a 75-year-old woman who presented to the emergency department with a 3 to 4-day history of abdominal pain, nausea, generalized weakness, fever, and shortness of breath. She had septic cholangitis due to primary choledocholithiasis 30 years post-cholecystectomy and numerous other comorbidities that increased the complexity of her case. Endoscopic retrograde cholangiopancreatography (ERCP) was attempted to remove the stone but was unsuccessful due to duodenal diverticula. Eventually, this patient underwent common bile duct exploration using a robot-assisted approach. A 2 cm stone at the distal common bile duct was removed, and her clinical status dramatically improved. The efficiency and increased fine control of a robot-assisted approach introduces our idea that this approach should be an alternative management option for minimally invasive common bile duct exploration in patients who have high-risk comorbidities and failed ERCP.

Keywords: Acute cholangitis, primary choledocholithiasis, recurrent choledocholithiasis, post-cholecystectomy, robotic-assisted common bile duct exploration

INTRODUCTION

Acute cholangitis, also known as ascending cholangitis, refers to an infection of the biliary tree and can present with right upper quadrant abdominal pain, jaundice, and fever (Charcot's triad). In addition to Charcot's triad, patients with severe cases of acute cholangitis may also develop septic shock and altered mental status (known as Reynold's pentad).[1] While these are typically considered classic features of the disease, it is important to recognize that not all patients present with these symptoms and that Charcot's triad or Reynold's pentad alone have low diagnostic sensitivity for acute cholangitis.[2] Rather, diagnostic criteria should follow the 2018 Tokyo Guidelines which evaluates for signs of systemic inflammation (e.g., fever, elevated CRP, abnormal WBC, etc.) and cholestasis (e.g., jaundice, abnormal liver function tests, etc.) in the patient's clinical presentation, lab work, and imaging.[3] If left untreated, acute cholangitis can be life-threatening. Thus, identifying its etiology is critical to guiding disease management.

The pathophysiology behind acute cholangitis is biliary duct obstruction and bile stasis which allow bacteria to infect the biliary tract. Some causes of obstruction

Corresponding author: **Devin Bird**
Contact Information: Devin.Bird@ttuhsc.edu
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include gallstones, malignancy (e.g., pancreatic cancer, cholangiocarcinoma), and biliary strictures (e.g., primary sclerosing cholangitis, post-endoscopic retrograde cholangiopancreatography [ERCP] strictures).[1] The most common cause of obstruction are gallstones lodged within the common bile duct (CBD), which is known as choledocholithiasis. Most often, these stones first form in the gallbladder and are then displaced into the common bile duct (secondary choledocholithiasis). However, on rare occasions these stones may form de novo within the common bile duct (primary choledocholithiasis) and cause symptoms.

Primary choledocholithiasis is much less common than secondary choledocholithiasis, and there is little literature on its exact etiology and prevalence. Proposed risk factors for stone formation within the common bile duct include biliary infection, peripapillary diverticulum, irregular biliary structure/anatomy, and abnormal metabolism (e.g., hypothyroidism). Moreover, current research reports that despite treatment, some patients experience recurrence of primary choledocholithiasis, suggesting that long-term management may be difficult.[4–5] Despite its uncertain etiology and possible risk of recurrence, stone removal is always necessary, particularly in the setting of acute cholangitis.

Management of acute cholangitis consists of providing hemodynamic support, treating the infection with empiric antibiotics, and removing the biliary duct obstruction. In the case of choledocholithiasis, ERCP is considered first-line treatment for stone removal. In circumstances where ERCP fails to remove the stone or is unfeasible, surgical bile duct exploration (robot-assisted, laparoscopic, or open) is indicated. In patients with acute cholangitis secondary to choledocholithiasis, elective cholecystectomy remains standard of care to help prevent recurrence.[6]

We present a rare case of septic cholangitis secondary to recurrent primary choledocholithiasis in a patient 30 years post-cholecystectomy. This patient's case was further complicated by respiratory failure, an incidental 6.0 cm abdominal aortic aneurysm (AAA), nonocclusive deep venous thrombosis (DVT) within the left deep femoral vein, and failed ERCP. Given this patient's high-risk comorbidities and failed ERCP, robot-assisted bile duct exploration provided an effective treatment option for septic cholangitis secondary to primary choledocholithiasis.

Case

A 75-year-old woman presented to the emergency department with complaints of shortness of breath, cough, abdominal pain, nausea, diarrhea, generalized weakness, and fever that started 3 to 4 days prior while traveling from out-of-state. The patient's complex medical history included chronic obstructive pulmonary disease, hypothyroidism, hypertension, hyperlipidemia, active tobacco use of 1 pack per day, and obesity with a body mass index of 30.5 kg/m^2. Her surgical history was also significant for open cholecystectomy (approximately 30 years prior), ERCP with sphincterotomy (10 years prior), and tonsillectomy.

On initial presentation, the patient's vital signs showed tachycardia but were otherwise unremarkable. Generally, she looked ill and intermittently confused. Physical examination was significant for epigastric and right upper quadrant (RUQ) tenderness to palpation without guarding or rebound tenderness. Laboratory results showed leukocytosis (17.6 K/mcL) with neutrophil predominance (89.9%), a total bilirubin of 1.97 mg/dL, a lactic acid of 2.9 mmol/L, and elevated liver enzymes (ALT 115 units/L, AST 157 units/L, ALP 167 units/L).

Computed tomography (CT) imaging of the chest showed pulmonary emphysema and multiple tree-in-bud nodules in the right middle lobe, lingual, and bilateral lower lobes near the lung bases consistent with multifocal pneumonia. Computed tomography scans of the abdomen and pelvis showed obstruction of the distal CBD by a 2.0 cm gallstone and severe intrahepatic and extrahepatic biliary duct dilatation. A 6.0 fusiform infrarenal abdominal aortic aneurysm (AAA) was also identified (Figure 1).

The patient was found to have acute cholangitis and sepsis protocol was initiated. She was subsequently given fluids, breathing treatments, oxygen, broad spectrum empiric antibiotics, and admitted to the hospital. Gastroenterology and vascular surgery

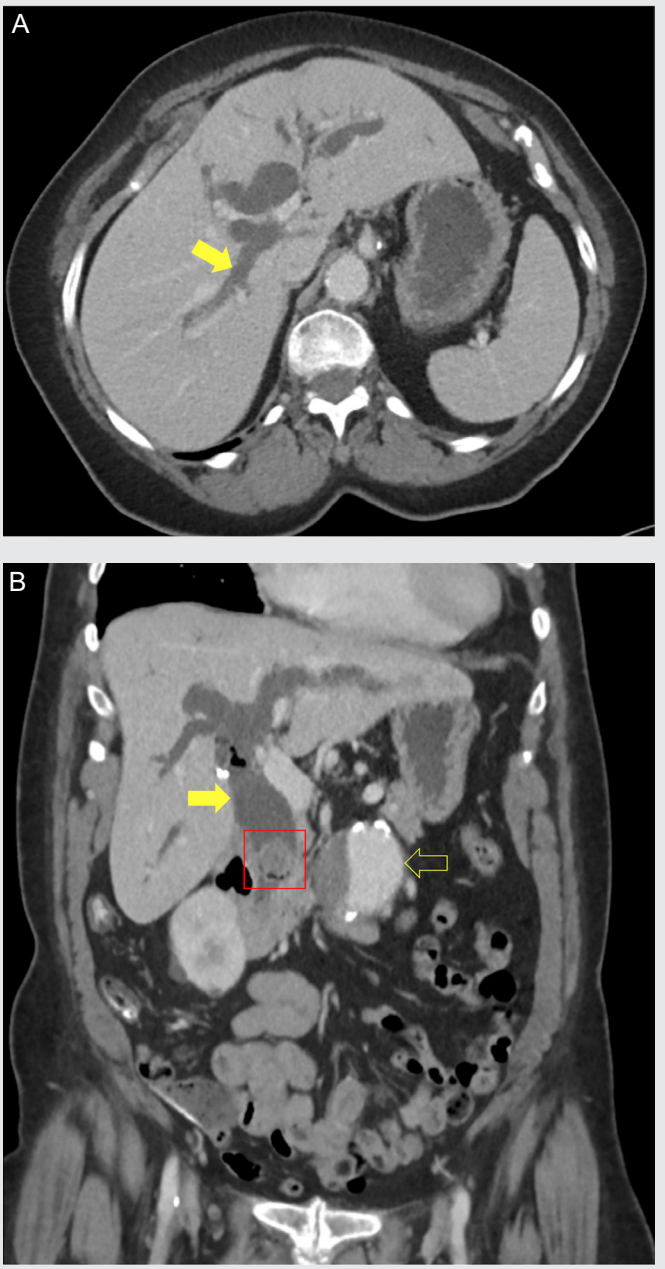

Figure 1. CT abdomen and pelvis with contrast, coronal (**A**) and axial (**B**) views. Shown is a gallstone obstructing the distal common bile duct (red box), intrahepatic and extrahepatic biliary ductal dilatation (filled yellow arrows), and an abdominal aortic aneurysm (unfilled yellow arrow).

services were consulted for CBD stone removal and AAA repair respectively. Prior to completion of these procedures, the patient experienced additional complications throughout her hospital stay.

On hospital day 2, her respiratory condition deteriorated further with an oxygen saturation as low as 74% and she became increasingly tachycardic and diaphoretic, which warranted elective intubation and transfer to the intensive care unit. Sputum cultures grew *Streptococcus pneumoniae*, and antibiotics were adjusted accordingly. The patient was also started on corticosteroids and found to have a white blood cell count of 69.9 K/mcL.

On hospital day 3, a duplex compression ultrasound was performed due to a swollen left extremity. A nonocclusive DVT in the left deep femoral vein, possibly chronic, was discovered even though she had been on enoxaparin for DVT prophylaxis. Eventually, the patient underwent ERCP for stone removal. During ERCP, two large diverticula were found in the second portion of the duodenum. Despite multiple attempts, locating the ampulla of Vater within the diverticulum was unsuccessful, resulting in the procedure's termination without stone removal.

On hospital day 4, interventional radiology was consulted for guided percutaneous transhepatic cholangiography and drain placement. Obtained cultures revealed the presence of *Escherichia coli*, *Enterococcus faecalis*, *Clostridium perfringens*, *Citrobacter freundii*, and *Candida albicans*. The patient's antibiotic regimen was adjusted based on the sensitivity results to include meropenem, ampicillin, and micafungin. This treatment approach successfully resolved the cholangitis and the patient's condition stabilized.

On hospital day 6, the patient was extubated and weaned off the vasopressors. The patient was advised against traveling home for further treatment due to the risk of AAA rupture and drain malfunction.

On hospital day 9, a robotic CBD exploration procedure was conducted. On visualization of the target anatomy, thick adhesions obscuring the subhepatic space were discovered that required 40 minutes of adhesiolysis. Absence of the gallbladder was confirmed, the

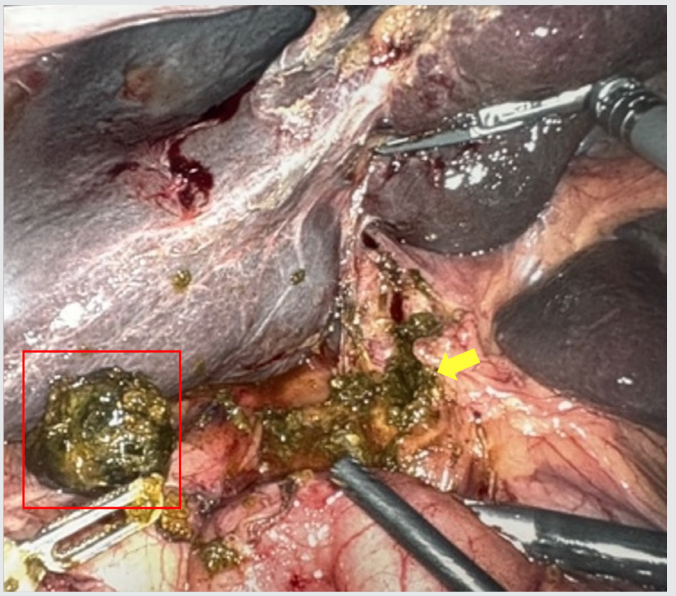

Figure 2. Intraoperative common bile duct exploration. Choledochotomy (yellow arrow). Stone removed from the common bile duct (red box).

CBD was exposed, and indocyanine green fluorescent cholangiography was performed to confirm the CBD location. There was no visualization of a cystic duct remnant at that time. The CBD was incised longitudinally to perform a choledochotomy and a 2 cm stone was removed (Figure 2). The choledochotomy was closed and intraoperative cholangiogram (IOC) showed patency of the CBD with no filling defects and flow of contrast to the duodenum. The retrieval bag was removed, a Jackson-Pratt drain was placed, and the port sites were closed. A week later, the patient's leukocytosis had improved, and she underwent repair of the 6.0 cm AAA. Over the following days, the patient continued to improve and was discharged on hospital day 19 with oral antibiotics.

The case is unusual for several reasons. First, not only did the patient have septic cholangitis secondary to primary choledocholithiasis, but it was recurrent primary choledocholithiasis and it occurred 30 years post-cholecystectomy, all of which is very rare. Second, the patient was found to have multiple incidental comorbidities which complicated her case. And third, with regard to treatment of late primary choledocholithiasis in the setting of failed ERCP, there is little evidence on how it should best be managed. In this case, robot-assisted CBD exploration was an effective treatment option.

Discussion

Acute cholangitis, an infection of the bile and bile ducts, is a life-threatening condition if not properly treated. Biliary obstruction and subsequent infection by bacteria underlie the pathophysiology behind acute cholangitis, and the source of obstruction has many several etiologies. These causes of obstruction include biliary lithiasis, benign biliary stenoses, malignant biliary stenoses, biliary stents, surgical clips, duodenal diverticulum, and others.[7] Regardless of the etiology, the goal of care is the same, treatment of the infection with antibiotics and removal of the obstruction. The most common cause of obstruction is biliary lithiasis, with secondary choledocholithiasis being the most common source of stones and primary choledocholithiasis being less common.

Acute cholangitis secondary to primary choledocholithiasis is uncommon, and there is little literature on the subject. The available literature mentions that these stones are typically composed of calcium bilirubin and have been reported to reappear in patients who previously received treatment.[4–5] Even more uncommon than primary choledocholithiasis itself is choledocholithiasis that presents years to decades after cholecystectomy. Most primary bile stones recur within 3 years of cholecystectomy.[8,9] A few cases have been reported of primary bile stones decades after the gallbladder was removed.[10,11]

The diagnosis of choledocholithiasis involves assessment of a patient's clinical presentation, lab tests, and imaging. Depending on severity, patients with biliary stone obstruction may present with RUQ abdominal pain, jaundice, fever, hypotension, or altered mental status on physical examination. If clinical presentation is concerning for an obstruction, laboratory studies and imaging can help guide the diagnosis. Elevated gamma-glutamyl transpeptidase, total bilirubin, and alkaline phosphatase are frequently associated with

choledocholithiasis. RUQ abdominal ultrasound is an appropriate first choice for most patients. It is cheap, accessible, and can easily identify stones within the gallbladder and common bile duct. A CT scan of the abdomen is another imaging option and is reported to have a similar specificity and greater sensitivity than ultrasound for diagnosing choledocholithiasis. Computed tomography can be particularly useful when a cause other than secondary choledocholithiasis is suspected (e.g., malignancy). ERCP is an additional option that is both diagnostic and therapeutic for patients with a high likelihood of gallstone obstruction. Last, endoscopic ultrasound and magnetic resonance cholangiopancreatography are both less invasive than ERCP and are considered to have greater sensitivity than abdominal CT or ultrasound. However, these last two modalities are time consuming and not widely available.[12]

ERCP remains the gold standard for management of choledocholithiasis. However, when ERCP fails or is contraindicated, surgery is necessary. Options for surgical intervention of primary choledocholithiasis include open, laparoscopic, or robotic-assisted CBD exploration.[10] The first published report of a robotic-assisted CBD exploration was in 2003, and its use has increased since then. The benefits using a robotic-assisted approach include better range of motion for surgical intervention, increased accuracy in suturing, and better visualization of target anatomy when compared to a laparoscopic approach.[13] With regard to treatment of late primary choledocholithiasis in the setting of failed ERCP, there is little evidence on how it should best be managed. In this case report, robotic-assisted CBD exploration was an effective treatment option.

Conclusion

Literature on the management of late primary choledocholithiasis is limited, and clinicians need an update to its standard care. We propose that based on the ease and ability for fine control amidst high-risk comorbidities, robotic-assisted intervention is a preferred alternative management option for common bile duct exploration following unsuccessful or contraindicated ERCP.

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From: School of Medicine (DB, AR) and Department of Surgery (BS) Texas Tech University Health Sciences Center, Amarillo, Texas; Department of Pediatrics (HF), University of Texas Southwestern Medical School, Dallas, Texas; Department of Surgery (RT), University of Colorado School of Medicine, Denver, Colorado; National Cancer Institute (GA), Cairo University, Egypt
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