Case study

A Rare Case of Post Cholecystectomy Cholecysto-cutaneous Fistula with Concomitant Common Bile Duct Stone

Abstract

Cholecysto-cutaneous fistula (CF) formation following cholecystectomy is a rare surgical condition. We report a case of post cholecystectomy CF in a 37-year-old male who presented with bile-like mucous discharge from the healed cholecystectomy scar. He was previously treated for gallbladder empyema presented with acute and severe cholecystitis by percutaneous cholecystostomy followed by open subtotal reconstituting cholecystectomy in another hospital. Intermittent pain on the surgical site and occasional mucous discharge from the surgical scar started two months after the surgery and was left untreated for almost a year. The patient presented in our emergency room (ER) with obstructive jaundice and cholangitis, along with an apparent bile fistula. Magnetic resonance imaging (MRI) confirmed the diagnosis of cholecysto-cutaneous fistula along with distal common bile duct (CBD) stone. We performed an open cholecystectomy, fistula excision, bile duct exploration, and a choledocho-duodenostomy bypass. Post cholecystectomy cholecysto-cutaneous fistula is a rare clinical entity that could happen after a complex cholecystectomy operation. MRI best describes the fistula with the possibility of concomitant stones. One-step surgery should minimize the possible morbidity and complications after multiple-step surgery in treating gallbladder empyema.

Keywords: gallbladder empyema, cholecysto-cutaneous fistula, Magnetic resonance imaging, cholecystectomy

Introduction

A cutaneous gallbladder fistula or a cholecysto-cutaneous fistula (CF) is an extremely rare surgical condition nowadays. Reports of spontaneous CF following an untreated biliary disease go back as early as the 17th century.[1] However, the occurrence of CF following cholecystectomy surgery has only been reported twice in the English language literature.[2,3] We report a case of cholecysto-cutaneous fistula, accompanied with common bile duct stone, following an open subtotal reconstituting cholecystectomy performed for acute cholecystitis.

Case Presentation

A 37 years old male presented with bile-like mucous discharge at the right upper abdominal wall from a healed open cholecystectomy scar. He had a history of open cholecystectomy surgery one year prior in another hospital. From his case history, we learned that approximately one year ago, he had visited the emergency room (ER) with a high fever, malaise and was delirious. He had excruciating right upper quadrant abdominal pain accompanied by nausea and vomitus, but no jaundice was reported. He had had a history of upper abdominal pain that comes and goes. His primary physician treated that as chronic gastritis. He had no history of jaundice before his initial admission and had only relented to go to the ER due to the high fever. Abdominal ultrasonography done in the ER revealed a severely inflamed gallbladder and the presence of gallbladder stones without bile duct obstruction.

After adequate resuscitation and antibiotic administration in the ER, the patient was taken to the operating room (OR) for a percutaneous cholecystostomy as source control and life-

saving procedure. Following the cholecystostomy, the patient's condition gradually improved. Open cholecystectomy surgery was performed two weeks after the procedure as the second stage surgery. The surgical report indicated a hostile abdomen with severe adhesion along the gallbladder fossa and a severely inflamed gallbladder. As a result, a reconstituting subtotal cholecystectomy was performed to avoid unwanted injury to the common bile duct (CBD).

Two months after the surgery, the patient started having intermittent pain on the surgical site accompanied by a small reddish mass that usually disappears on its own. At times, the mass would painfully enlarge and break, producing mucous discharge that will eventually dry up. The pain and mucous discharge would subside for a few weeks and recur again. The symptoms occurred around the COVID-19 pandemic, and the patient was reluctant to go to the hospital for a follow-up. He had attended to his wound himself. However, after almost eight months of having recurrent discharge from his wound, he decided to go back to his surgeon.

A fistula tract had formed between the remaining gallbladder and the front abdominal wall, as revealed by a magnetic resonance cholangiopancreatography. By injecting water-soluble contrast from the external wound, a fistulography was conducted, which revealed a clean route from the skin to the remaining gallbladder (**Figure 1**). When the patient was referred to our facility, we chose to perform surgery. The pandemic-related shortage of hospital and human resources, however, caused a little delay in the procedure.

The patient presented to the emergency room with jaundice while awaiting the scheduled elective operation as well as fever and right upper quadrant abdominal pain. Up to 250 cc of bile-colored liquid was now consistently oozing from his wound, which had by this point become a regular discharge. A second MRCP was conducted, which revealed extra and

intrahepatic bile duct dilatation as well as CBD obstruction caused by a CBD stone (**Figure 2**). While the patient was getting ready for elective surgery the following day, rehydration, antibiotics, and symptom medication were given.

On the operating table, after skin preparation, a small probe was inserted through the external fistula opening before the laparotomy incision. Upon entering the abdomen, there was moderate adhesion around the gallbladder fossa that was taken down carefully. The fistula tract was excised by following the probe, and cholecystectomy was performed. The bile duct was incised and explored, revealing a 2x1x1 cm stone near the ampulla. A choledochoduodenostomy bypass was performed, and a single drain was placed at the subhepatic region.

The postoperative period was unremarkable. The patient was started on clear fluid as soon as he was awake, mobilization by postoperative day (POD) 1, a nasogastric tube (NGT) was removed by POD 3, abdominal drain POD 4, and the patient was discharged POD 5. Upon follow-up, the patient was pain-free, no recurrent jaundice, and the surgical wound healed without any complications.

Discussion

A fistula is an abnormal communication between two epithelial surfaces that can be caused by a complication of a disease or surgical intervention. Fistulas are named after the two surfaces or lumens it connects; thus, a cholecysto-cutaneous fistula is an abnormal connection between the gallbladder and the skin. Since the first reported case by Thilesus in 1670, CF has become a clinical rarity in this modern era of diagnostics that leads to the early management of biliary diseases.[1,4]

Untreated gallbladder diseases, such as gallbladder stones with cholecystitis, and tumors, are the main risk factors of CF. In addition, CF can occur following the removal of percutaneous cholecystostomy drain removal.[1,4] CF following cholecystectomy procedure is rare, and we have only found two reported cases in the English literature. In both cases, the fistula was formed between the residual gallbladder to the skin.[2,3] While in one of the reports, Maynard et al. had reported since the beginning that the CF was formed in a patient following a subtotal cholecystectomy,[2] in the other report, Ping et al.[3] reported that the initial surgery was understood as a total cholecystectomy. However, upon exploration, Ping et al. soon discovered a remnant gallbladder, to which the fistula was connected, and an enlarged cystic duct.

Complications following cholecystectomies in which the gallbladders are removed entirely are infrequent. However, in some difficult cases in which the biliary anatomy is uncertain, subtotal cholecystectomy has been recommended as a safe and viable option.[2,5] Subtotal cholecystectomies are performed as 'subtotal reconstituting cholecystectomy' or as a 'subtotal fenestrating cholecystectomy'. An external bile fistula is more likely to occur with subtotal fenestrating cholecystectomy, in which one wall of the gallbladder is left in situ, and the cystic duct remained open to the peritoneal cavity. Meanwhile, a subtotal reconstituting cholecystectomy, in which the Hartman's pouch is left in situ, leaving a remnant gallbladder, is more likely to develop recurrent cholelithiasis and cholecystitis. Therefore, when a subtotal cholecystectomy is necessary, the operative record should include a complete and accurate description of the procedure, as this may influence the possibility of certain postoperative complications.[2]

The pathophysiology commonly associated with gallbladder fistula is the increasing pressure in the gallbladder, or gallbladder remnant, due to an obstruction in the cystic duct or the

common bile duct leading to cholecystitis and/or gallbladder empyema and compromised blood supply, which eventually leads to perforation. The leaked infected bile tracts towards the abdominal wall, eventually forming a visible abscess that drains out continuously as a biliary fistula.[1,2,4]

Although, as mentioned above, the formation of bile fistula is more likely after subtotal fenestrating cholecystectomy, cholecysto-cutaneous fistula after a subtotal reconstituting cholecystectomy may occur due to an obstruction in the cystic duct causing the sequence in which the gallbladder perforates, and the infected bile drains to the anterior abdominal wall, as seen in this case. In addition, the patient had had percutaneous gallbladder drainage before the subtotal reconstituting cholecystectomy, leaving a tract of inflamed tissue around the rubber drain, which may have aided in the pathological formation of the fistula tract through which the bile from the perforated gallbladder flowed to the anterior abdominal wall.

In addition to the symptoms of abdominal wall abscess, fever, excretion of bile from the skin, and sometimes obstructive jaundice, radiological studies are needed in confirming the diagnosis of CF. While ultrasonography (US) is helpful in providing abnormal findings, such as abscess formation, gallbladder stones, edema, thickened gallbladder wall, and dilated bile ducts, it often fails to establish the CF tract. An X-ray fistulogram can show the CF tract, which confirms the diagnosis, but is not able to evaluate the gallbladder and bile ducts. Computed tomography (CT) shows abnormal findings, which point towards a CF diagnosis but fails to identify the tract in some cases. In those cases, a CT fistulogram may aid in showing the fistula tract. Magnetic resonance imaging (MRI), along with magnetic resonance cholangiopancreatography (MRCP), provides a more accurate picture of the fistula and surrounding structures when CT detects no abnormalities.[4]

In our case, the fistula formed two months after cholecystectomy surgery and drained through the incision site. The symptoms were intermittent for a year before the patient developed obstructive jaundice with cholangitis. Our patient's MRCP clearly defined the fistula tract between the severely inflamed remnant gallbladder and the external cutaneous opening along with dilated intra and extrahepatic bile duct caused by a distal CBD stone.

Gallstones-related complications may occur at any time following all types of subtotal cholecystectomy, causing symptoms such as recurrent right upper quadrant pain, gallstone pancreatitis, and obstructive jaundice.[2,5] There are three alternate conditions that may cause the recurrence of stones in a gallbladder remnant: 1. Inadvertent incomplete gallbladder removal, 2. Incorrectly performed subtotal intentional cholecystectomy (fundectomy alone), or 3. Existence of a duplicated or even triplicated gallbladder was inadvertently missed at the procedure[5]. Examining the remaining gallbladder intraoperatively in our case, we believe a fundectomy alone rather than a true subtotal cholecystectomy was out.

Management of CF varies according to disease severity, age, and patient's preference. Conservative management of antibiotics, fluids, and endoscopic retrograde cholangiopancreatography (ERCP) removal of calculi and sphincterotomy, along with abscess drainage, can be an option, especially in elderly patients who are unable to tolerate surgery. Open cholecystectomy with excision of the fistula tract is curative in most cases and considered as a standard management option. However, in the hands of advanced experienced laparoscopic surgeons, laparoscopic cholecystectomy with tract excision can be an acceptable option.[4] We elected the open approach as the patient had had an open approach previously, and we suspected a hostile abdomen with severe adhesion along the gallbladder fossa. A probe was inserted through the external fistula opening that served as a guide in the fistula tract excision. The adhesions around the cholecyst were meticulously removed before

the operation. We assume that an incomplete subtotal cholecystectomy (fundectomy) was previously conducted based on the size of the remaining gallbladder. To clear the bile duct obstruction, we underwent a choledochoduodenostomy bypass and a CBD exploration. The patient made a full recovery with no complications.

Conclusion

A difficult cholecystectomy procedure, whether performed openly or laparoscopically, may result in an uncommon clinical phenomenon known as a Post Cholecystectomy Cholecysto-cutaneous Fistula. The patient, the patient's family, and the surgeon should all be aware of this possibility. It is important to perform a thorough examination to determine the extent of the issues; an MRI is the greatest tool for detecting fistulas and the potential presence of internal stones. To reduce the likelihood of morbidity and problems following multiple-step surgery to treat gallbladder empyema, one-step surgery should be performed while the patient is in their best physical and nutritional condition.

References

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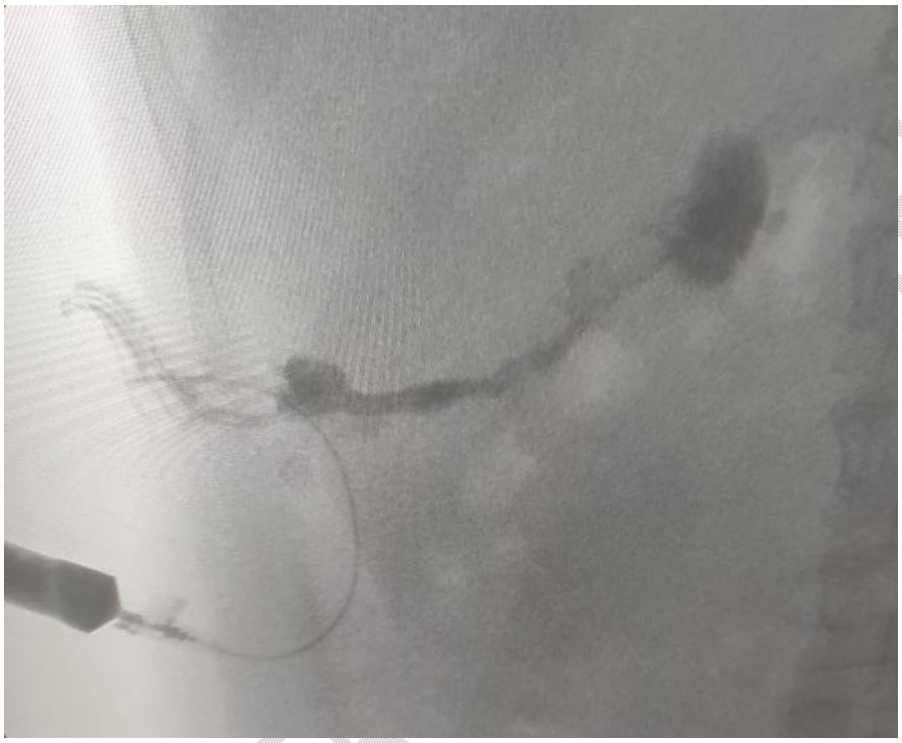

Figure 1. Fistulography revealed a clean route from the skin to the remaining gallbladder.

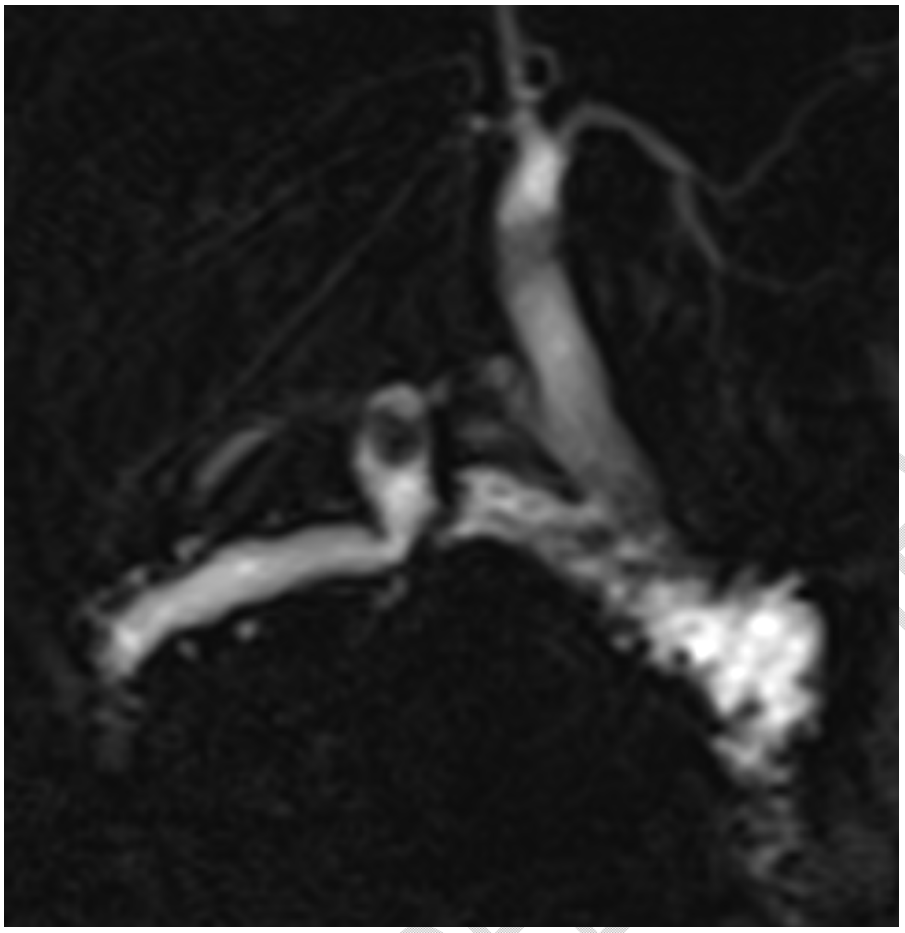

Figure 2. MRCP revealed extra and intrahepatic bile duct dilatation as well as CBD obstruction caused by a CBD stone.