CASE REPORT

Spontaneous cholecystocutaneous fistula as a rare complication of gallstones

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Abstract: Spontaneous perforation of gallbladder as a complication of biliary stones may lead to a cholecystocutaneous abscess or fistula. The pathophysiology of this condition has been associated with increased pressure in the gallbladder, secondary to biliary obstruction. Ultrasonography and Computed Tomography (CT) can be used for diagnosis of cholecystocutaneous abscess or fistula. The treatment of fistula requires adequate drainage, antibiotics, followed by elective cholecystectomy with excision of the fistula. We report a case of spontaneous cholecystocutaneous fistula in an 89-year-old female patient who presented with obstructive jaundice and subcutaneous abscess in the right subcostal area. Abdominal CT scan showed gallstones and communication between the abscess and the gallbladder. First abdominal wall abscess was drained externally then cholecystectomy and exploration of common bile duct was performed (*Fig. 3, Ref. 8*). Full Text in PDF *www.elis.sk.* Key words: cholecystocutaneous fistula, gallstone, spontaneous, tomography, cholecystectomy.

Cholecystocutaneous fistula is very rare in surgical practice and usually is a result of hepatobiliary surgery. Spontaneous perforation of gallbladder as a complication of biliary stones may lead to cholecystocutaneous abscess or fistula. Cholecystocutaneous fistula was first described by Thilesus in 1670 (1) and was common till the half of the last century. Improved diagnostic investigations and modern biliary surgery have lead a decrease in the incidence of this complication. In this report, we present a case of spontaneous cholecystocutaneous fistula in an 89-year-old female patient who presented with obstructive jaundice and large subcutaneous abscess in the right subcostal area with surrounding cellulitis and crepitus.

Case report

An 89-year-old woman was admitted to our hospital complaining of a right upper abdominal pain and icterus that had lasted for 3 days (Fig. 1). Gastroenterologists evaluated the patient and planned endoscopic retrograde cholangiopancreatography (ERCP) for obstructive jaundice. In this period, the complaints and bilirubin levels improved, but a painless concomitant right subcostal mass with surrounding cellulitis and crepitus developed. The patient was readmitted to our service with this mass. There was no previous history of abdominal pain, local trauma, dyspepsia or diabetes. On readmission, the patient was in a good clinical condition, with a heart rate of 84 beats/min and systolic blood pressure of 110 mmHg and she was afebrile.

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Address for correspondence: Y. Ozdemir, MD, GATA Haydarpasa Egitim Hastanesi, Genel Cerrahi Servisi, Uskudar, 34668, Istanbul, Turkey. Phone: +90.216.5422020, Fax: +90.216.4494480 Abdominal examination revealed a 10-cm abdominal wall abscess in the right upper quadrant area with surrounding cellulitis, crepitus and tenderness. The white blood cell count was 7,700 x 10^{9} /L. The levels of some blood tests were as follows; alkaline phosphatase 305 U/L, ALT: 295 U/L, AST: 644 U/L, GGT: 207 U/L, direct bilirubin: 38.47 µmol/L, total bilirubin: 64.98 µmol/L, CRP: 48 mg/L. The other laboratory values, including hemoglobin, amylase and glucose levels were unremarkable. Abdominal ultrasound (US) and abdominal computed tomography (CT) scan clearly showed gallstones, common bile duct stone and communication between the abscess and the gallbladder.

A small incision was performed to the fluctuating area of the abscess. Initially, 15 cc of purulent material were drained from the incision and following two days the drainage continued. Initial treatment included local control of septic focus and broad-spectrum antibiotics administration. The bile leakage stopped after 7 days and the inflammation subsided within 12 days. All laboratory data was normal on the 21st day. Laparotomy was performed on the 24th day, by using a right subcostal incision including the fistula tract. During laparotomy, the fistula formation between the abdominal wall and the fundus of the gallbladder was also exposed (Fig. 2). Cholecystectomy and excision of the fistula tract were performed. After this procedure the common bile duct was explored and the stone was extracted. Common bile duct was closed primary and a silicon drain was inserted to the Winslow space. Postoperative recovery of the patient was uneventful. Histopathological analysis revealed chronic cholecystitis with no evidence of malignancy.

Discussion

Biliary fistula of the gallbladder is an abnormal connection between the gallbladder and abdominal viscera (internal) or skin



Fig. 1. Draining site of the abscess after incision in the right upper quadrant.

(external). Internal fistulas are much common than external fistulas and occur with connection to the duodenum (60 %), followed by the colon (24 %), stomach (6 %) and choledochal duct (5 %) (2). External biliary fistula (cholecystocutaneous fistula) is rarely observed today. Thilesus reported the first case of an external biliary fistula in 1670 (1). In 1890, Courvoisier described a series of 499 cases of gallbladder perforation, and 169 of these cases were cholecystocutaneous fistulas (1). Cholecystocutaneous fistula formation is a result of chronic perforation of the gallbladder. Obstruction of cystic duct or common hepatic duct, frequently with a calculus, plays an important role in the pathophysiology of the perforation. Because of the higher incidence of gallbladder stones in women, patients with cholecystocutaneous fistula are usually women. Most of the cases are over the age 60, but also patients as young as 24 years old has been reported (3). Our case was an 89-years-old woman.

Cholecystocutaneous fistulas more frequently drain to the right upper quadrant and right flank area, although other locations such as the epigastrium, umbilical area, right groin and even the gluteal region have also been described (2, 3, 4).

The increasing pressure in the gallbladder, secondary to cystic duct obstruction, caused by a calculus is a possible explanation for the pathophysiology of this condition. Due to the obstruction, intraluminal pressure increases, and as a result blood flow and lymph supply of the gallbladder impairs, mural necrosis and perforation occurs. Perforation of gallbladder is classified by Niemeier as acute, subacute and chronic (5). Acute perforation is a free perforation, and leads to peritonitis. Subacute perforation results in an



Fig. 2. Intraoperative exploration (a) and (b) fistula formation between the abdominal wall and the fundus of the gallbladder.



Fig. 3. Computed tomographic scan of the abdomen showing (a) enhancement of the gallbladder wall, and (b) prominence of subcutaneous tissues and muscle.

abscess formation around the gallbladder. Only chronic perforation is characterized by an internal or external biliary fistula formation. These fistulas, as presented in our case, frequently arise from the fundus of the gallbladder (3, 4). Furthermore there was stone at common bile duct in our case.

Ultrasonography and CT (Fig. 3) can be used for diagnosis of biliary tract pathologies and associated other pathologies (6). In the presence of a fistula opening, a fistulogram can confirm a communication with gallbladder or biliary tract. However, we did not need to perform a fistulogram in our patient, because the ultrasonography revealed gallbladder adherence to the abdominal wall and a tract between the gallbladder and the skin. We also observed characteristic bile drainage from the incision.

The treatment of fistula requires adequate drainage, antibiotics, followed by elective cholecystectomy with excision of the fistula. Because up to 20 % of cholecystocutaneous fistulas heal spontaneously, in the high-risk patients conservative treatment can be considered (7). Common bile duct obstruction, caused by a stone can be treated by endoscopical extraction of stones first, and then cholecystectomy with excision of the fistula. Patients with ampullary stone impaction, biliary pancreatitis and multiple comorbidities are considered good candidates for preoperative endoscopic therapy. If clearance is not possible because of multiple stones, intrahepatic stones, impacted stones, duedonal diverticula, or biliary stricture, this information is known before surgery. Endoscopic stone extraction is well tolerated in most patients. Mathonnet et al. suggested laparoscopic cholecystectomy as a feasible procedure, but it has a high conversion rate (8). The control of acute inflammation in the abdominal wall, followed by an elective open cholecystectomy

with the excision of the fistulous tract, is an adequate approach for efficient treatment.

The possibility of cholecystocutaneous fistula should be considered in all cases of unexplained abdominal wall suppuration or cellulitis. Suspicion of cholecystocutaneous fistula is helpful in achieving correct preoperative diagnosis and adequate treatment.

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