

Spontaneous Cholecystocutaneous Fistula: A Consequence of Neglected Calculus Chronic Cholecystitis

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ABSTRACT

Spontaneous cholecystocutaneous fistulae are extremely rare complications of neglected gall stone disease and it has become even rare during the last decades over the past 50 years few cases of spontaneous cholecystocutaneous fistulae have been described in the literature. We have here reported the first case of its own kind where cholecystocutaneous fistula presented with multiple gallstones over anterior abdominal wall in right hypochondrium. After through investigations, open cholecystectomy was performed along with removal of fistula tract. This case report demonstrates neglected calculus chronic cholecystitis where patient is hardly aware of this deadly complication. So a high degree of suspicion of this rare entity must be kept in mind for achieving correct preoperative diagnosis. We suggest that proper pre-operative investigations must be performed in all cases of unexplained abdominal wall suppuration to avoid these kinds of complications.

Key Words: Cholecystocutaneous fistula, Abscess, Gallstones, CECT, MRCP, open Cholecystectomy.

INTRODUCTION

Spontaneous cholecystocutaneous abscess or fistula is an extremely uncommon complication of gallbladder disease. The first description of this condition was made by Thilesus in 1670 and at that time fistulae were common complications of chronic and untreated cholecystitis. [1] 169 cases had been described by Courvoisier, with Henry and Orr had added another 36 cases, of external biliary fistulae. [2] According to a 2005 study, 226 cases have been reported in total, with fewer than 25 in the last 50 years. [2] We are here presenting a very unique case of gallbladder fistula with stone spontaneously spilling out over anterior abdomen wall.

CASE REPORT

A 72 year old female presented in our outdoor with swelling in right upper abdomen for three months. There was history of intermittent fever with rigors and chills. The swelling was progressively increasing in size and reached up to 4×3 cm. Again on follow up she was giving history of serous discharge from the swelling in right hypochondrium and on examination there was a small sinus present in the centre of swelling. The swelling appears slightly red in color and local temperature was not raised. There were no signs of generalized or local peritonitis on abdomen examination except tenderness in and around swelling. There was no previous history of local

trauma, dyspepsia or diabetes. On presentation the patient was in a good clinical condition, so she was investigated as outdoor patient. Her all biochemical investigations were normal. Ultrasound examination of abdomen shows small localized collection in subcutaneous plane and rest of abdomen examination was normal they advised CECT abdomen for further evaluation. The CECT reported as chronic cholecystitis with cholelithiasis. They also reported a suspicious rent in fundus of gallbladder with small ill defined collection of size 25×6 mm in muscle plane of anterior abdomen wall in right hypochondrium with overlying small discharging sinus. Sealed gallbladder perforation was made on CECT. Radiologist further advised MRCP for further evaluation and later on cholecystocutaneous fistula was confirmed along with gallbladder stones. She came for follow up with reports. The wound was opened for dressing. We were surprised to see the gall stones over abdomen wall coming through the fistula tract. [Figure 1] We have never seen such a patient presenting with gallbladder fistula and multiple stones on anterior abdominal wall. She was kept for elective open cholecystectomy on priority basis. [Figure 2&3]



Figure 1 Showing gallbladder fistula with stone spontaneously coming out over abdomen wall.

She was operated with en block excision of fistula tract with anti-grade cholecystectomy. There were dense adhesions present around gallbladder and Calot's triangle anatomy was not clear.

Cystic duct and CBD was dilated but no stone was found intra-operatively as well as on MRCP. The immediate postoperative course was uneventful. She was kept on antibiotics and on third day she was discharged in stable condition with advised to follow up in our outdoor. Histopathological analysis revealed chronic cholecystitis with no evidence of malignancy.

DISCUSSION

Cholecystocutaneous fistulas are often the result of neglected biliary tract disease. Patients with this complication usually do not report a distinct episode of acute cholecystitis in their history, since this would have brought such a patient to seek medical attention sooner. The patients are usually women over the age of 60. [3] Biliary fistulae can be either internal or external. Internal fistulae are very much commoner, 75% of them connecting to the duodenum and 15% to the colon. The remaining 10% of internal fistulas connect with the stomach or jejunum, or have multiple communications such as cholecysto-duodenocolic fistula. [4] External biliary fistulae are very rare. They are usually the complications of gallstone disease, but can be secondary to biliary injury during a surgical procedure. [5] Niemeier classified perforation of the gallbladder into acute, sub acute and chronic varieties, only the latter being characterized by fistula formation. [6] External biliary fistula formation is chiefly a disease of females in the 5th-7th decades. [7] Our patient is also 72 years old female.



Figure 2: Cholecystocutaneous fistula: just before open cholecystectomy.

We have reported a rare case of spontaneous gallbladder perforation resulting in cholecystocutaneous abscess later on spontaneous burst with multiple gallstones spilled out. To our knowledge, there has not been any other case of cholecystocutaneous abscess following fistula presenting with multiple subcutaneous gallstones in the English-language medical literature since 1949 and also agree with this statement. [8] Cholecystocutaneous fistulas are most often seen in elderly women, with coexistent of disease and non-specific symptoms of cholecystitis interfering with diagnosis of this rare entity. [9] The natural history of the disease has changed from suppurative cholecystitis with spontaneous rupture to external drainage of an abscess. Early and effective medical and surgical management of biliary tract disease can prevent this rare condition. [10]

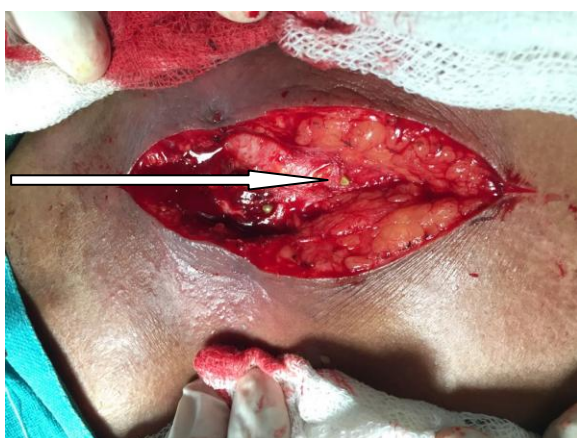


Figure 3 : intra-operative picture: fistula tract some stones are still in subcutaneous plane

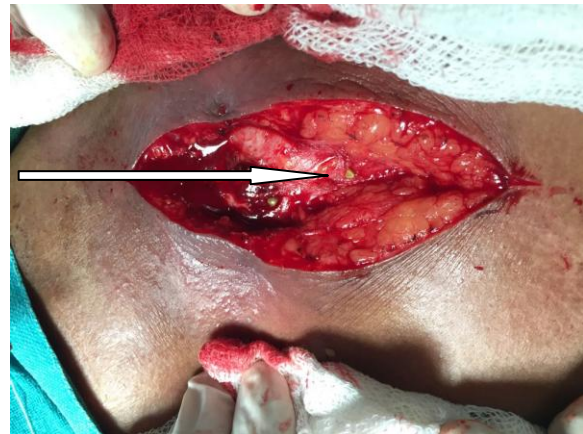


Figure 3 A: Intra-operative picture of fistula tract and some stones are still in subcutaneous plane.

However, over the past 50 years fewer than 20 cases of spontaneous cholecystocutaneous fistulas have been described. The marked decrease in the incidence of this complication is probably associated with the advent of contemporary diagnostic methods, broad-spectrum antibiotic therapy, and early and effective surgical management of biliary tract disease. [3] The pathophysiology of this condition has been associated with increased pressure in the gallbladder, secondary to cystic duct obstruction, either caused by a calculus or neoplasia. The increase in intra luminal pressure leads to impairment of the blood flow and lymph supply to the gallbladder, thus causing mural necrosis and perforation with the formation of an internal or external biliary fistula. [8] In our case there were no malignant changes on histopathological examination of gallbladder.

Traditional treatment for a spontaneous cholecystocutaneous fistula or abscess has consisted of abdominal wall incision and drainage followed by “staged” cholecystectomy. [11] The diagnostic process always begins with upper abdomen ultrasound and ends with hepatobiliary MRI to visualize the biliary tree and same was followed in our case too. CECT and MRCP confirmed the fistula before surgery. Cholecystocutaneous fistula has always been treated by two different strategies. The first includes a two-step approach: percutaneous drainage and antibiotic therapy, and subsequently cholecystectomy. The second directly involves laparotomy

cholecystectomy execution with en-block aponeurotic muscles, as well as skin and fistula orifice excision. The second strategy is the most commonly used since the two-step approach treatment is reserved for patients with sepsis and poor general condition. [3] The second approach was followed in our patient too because patient was in good clinical condition. Although laparoscopic approaches have option for treatment of cholecystocutaneous fistulae but still open technique is being used for most cases because of lower risks of post-surgical complications. [12]

CONCLUSION

Although complications like cholecystocutaneous spontaneous fistula is not common but if not quickly treated, it can rapidly evolve into a generalized septic state with severe impairment and poor prognosis. In our case, the patient was in good general health maybe because the fistula was draining the most of the abscess and stone outside the body and not in the peritoneum space. Surgical treatment is essential normal bile flow and addition of broad-spectrum antibiotic treatment is must to avoid the risk of post-operative complications.

Conflict of interest: All authors have no conflict of interest.

Contribution of Authors: All the authors have equally participated in preparation of this article.

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