

Spontaneous Cholecystocutaneous Fistula

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ABSTRACT

Spontaneous biliary fistulae are encountered, not very rarely, in one's surgical practice. These fistulae are of three types: Internal, external, and combined. Internal spontaneous biliary fistulae are the commonest. External fistulae could be spontaneous or because of therapeutic, iatrogenic, or traumatic reasons are extremely rare. Spontaneous cholecystocutaneous fistula (SCCF), secondary to calculous cholecystitis, is an extremely rare presentation in the present-day scenario. It used to be quite common before the year 1900, but is very rare now because of better management of cholecystitis and cholelithiasis. Usually, SCCF is a complication of neglected chronic cholelithiasis. This is seldom seen today because of the early diagnosis and better management made feasible by ultrasound as first-line investigation, broad spectrum antibiotics, and effective surgical management of biliary tract diseases. It is a very rare case of 35-year-old female patient presenting in the outpatient department, with the multiple stones carefully preserved, which she had been extruding through the fistulous opening in the umbilicus, for the last 1 year. She was investigated and was operated for the same condition. Though the entity is very rare, clinicians should keep this condition in mind while examining any case of chronic discharging sinus or fistula on the abdominal wall, particularly the wound extruding stones in which case the diagnosis is self-revealing. In the absence of positive history of expelling stones, the diagnosis can be confirmed by computerized tomogram fistulography. Though the early diagnosis and improvement in the management of gallbladder disease has improved tremendously, the possibility of this condition arising mostly from the neglected gallbladder disease should always be kept in mind as such cases are again being reported from all over the world.

Keywords: Cholecystocutaneous fistula, Cholecysto-umbilical fistula, Chronic calculous cholecystitis, Computerized tomogram fistulogram, Laparoscopic cholecystectomy.

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INTRODUCTION

Most of these internal fistulae communicate with duodenum (77%), colon (15%), and stomach (6%).¹ Rarely, it can communicate with the urinary system or bronchial

tree.² Spontaneous external biliary fistulae are very rare and were first described in 1670 by Thilesus.³ In the world literature, only 65 cases had been described, since the year 1900.^{2,4} Less than 20 cases prior to year 2006 have been reported in the last 50 years. Now, most biliary fistulae are postoperative complications of liver and biliary tract surgery or trauma. External biliary fistulae can be further subdivided into spontaneous, therapeutic, traumatic, and iatrogenic fistulae. External spontaneous cholecystocutaneous fistula (SCCF) is a very rare surgical complication of neglected calculous biliary disease that has become even increasingly rarer because of easy and early diagnosis and expedient surgical intervention for gallstone disease. External biliary fistulae sometimes occur spontaneously as a result of intrahepatic abscess (pyogenic or parasitic), necrosis or perforation of the gallbladder, or other inflammatory processes involving the biliary tree. Though the entity had almost vanished, recently, a few cases are being reported from all over the world. In spite of early diagnosis and better management of gallbladder disease, it is feared that this may not be a revisit by this, once not so uncommon entity, and a clinician should arouse a suspicion of SCCF in the patient having chronic discharging sinus or fistula on the abdominal wall whether the history of the gallbladder disease is forthcoming or not. A 35-year-old female patient presented to the outpatient department with a history of passing multiple stones, repeatedly, from her umbilicus for the last 1 year (Fig. 1). There was no history of episodes of fever, chills, rigors,



Fig. 1: Spontaneous external cholecysto-umbilical fistula, extruding gallstones from umbilicus

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and anorexia. On examination, thinly built, asthenic patient had an umbilical fistula, having serous discharge from the umbilicus. The patient presented the carefully preserved stones which she used to frequently expel from her umbilicus. The fistula was chronic and painless. Clear history of passing large, faceted, multiple stones repeatedly through the umbilicus was the mainstay of the clinical diagnosis, which was confirmed by the computerized tomogram fistulogram, as it delineated the tract and demonstrated the multiple stones present in the fistulous tract leading to the umbilicus. There was no history of abdominal trauma or previous surgery in this patient. Cholecystectomy of the chronically inflamed and shrunken gallbladder was done along with excision of the fistulous tract. The fistulous tract containing multiple stones, extending from gallbladder to umbilicus and traversing anterior abdominal wall was dissected and excised from anterior abdominal wall. The tract was identified emerging from the gallbladder, which was shrunken and fibrosed. There were no stones in the gallbladder. Postoperative period remained uneventful. Spontaneous external biliary fistula discharging into the skin surface, as in the present case, is rare. It is defined as a rupture of the gallbladder through all layers of the abdominal wall, with the creation of a fistulous tract to the skin, not preceded by any biliary surgery or trauma. The process of fistula formation is precipitated by obstruction of the cystic duct, which raises the pressure in the gallbladder, impairing the vascular supply and resulting in focal necrosis. This inflammatory process is typically insidious and recurrent. The fistula usually forms through the fundus of the gallbladder, since this part of the gallbladder is farthest from the cystic artery and thus most likely to be affected by ischemia. Chronic inflammation of the gallbladder can cause the gallbladder fundus to adhere to the abdominal parities triggering the

formation of fistulous tract. Underlying pathophysiology is the perforation of gallbladder which may be acute, sub acute, or chronic; it is the chronicity of the diseased gallbladder which is responsible for SCCF. Retained gallstones following laparoscopic cholecystectomy may cause biliary fistula or abdominal wall sinuses.⁵ This occurs because gallstones can harbor bacteria, which may form localized abscess with localized sinus, in an attempt to discharge the foreign body. *Salmonella typhi*, which has a predilection for the gallbladder, can cause chronic cholecystitis and may predispose to spontaneous SCCF.⁶ Communication to the umbilicus may be through the falciform ligament.

Sometimes the only manifestation may be the passing of the stones and discharge from the fistula, which is the case in the present patient also. The fistula itself may be painless as in the present case. Passing of the stones through the fistula with the discharge confirms the diagnosis clinically as in the present case. Various conditions which can be considered for differential diagnosis are metastatic carcinoma, tubercular sinus, pyogenic granuloma, chronic osteomyelitis of ribs with sequestrum, and infected epidermal inclusion cyst. Possibility of the SCCF should always be considered in any patient who has a chronic discharging sinus in abdominal wall or umbilicus. Moreover, the typical history of the patient is self-revealing and diagnostic.

Computerized tomogram fistulogram can demonstrate the fistulous tract and its contents, making a definitive diagnosis as was done in the present case (Figs 2, 3). The contrast will demonstrate the tract and gallbladder. Fistulography also may demonstrate the common bile duct, permitting evaluation of the biliary anatomy. It may also demonstrate multiple fistulous tracts or communications in some rare cases. Clinical presentation and radiological imaging provide valuable



Fig. 2: Computerized tomogram fistulogram demonstrating cholecysto-umbilical fistulous tract containing multiple gallstones



Fig. 3: Computerized tomogram fistulogram demonstrating cholecysto-gastric fistulous tract containing multiple gallstones

information in making the diagnosis of this rarely seen condition. Surgery is required most of the time and includes cholecystectomy with the excision of the tract, as both the gallbladder and the fistulous tract need to be excised to achieve a cure; in the present case open cholecystectomy with the excision of the tract was done and a subhepatic drain was placed which was removed on the third day.

The gallbladder was shrunken and fibrosed, whereas the fistulous tract contained multiple gall stones and the tract was adherent with the anterior abdominal wall as well with the surrounding tissue. The tract was opening into the umbilicus externally. The diagnosis of this rare entity often proves challenging if the clear history of passing the calculi per fistula is not there as a significant proportion of these patients present with nonspecific symptoms. Ideally, the treatment should include broad spectrum antibiotics, drainage of the abscess, and elective cholecystectomy with excision of the fistula.⁷

The possibility of external SCCF though very rare in present-day scenario should be kept in mind, in a patient having discharging sinus over abdomen or lower chest wall. In the patient passing stones through these fistulae, the diagnosis is obvious. Though with the advent of newer and efficient investigative and operative

modalities, the diagnosis and management of gallbladder disease has been made easier, yet the entity may be the result of neglected gallbladder disease as quite a few of these cases have been recently observed, as the modern-day advances in the treatment of gallstone disease are still not available in some pockets of the poorer population who prefer to bear or ignore the disease for the socioeconomic reasons, or is it a revisit by this entity?

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