

# Spontaneous cholecystocutaneous fistula draining from an abdominal scar from previous surgical drainage



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## Spontaneous cholecystocutaneous fistula draining from an abdominal scar from previous surgical drainage

*We present a rare case of cholecystocutaneous fistula draining from an old surgical scar in the right upper abdominal quadrant following chronic calculous cholecystitis. A 71 year old male presented to the emergency department with a persistent bilious drainage from an old surgical scare, from surgical drainage, of the right upper abdominal quadrant for about a week. Cultures from the draining fluid grew Staphylococcus hominis, Escherichia coli and Klebsilla pneumoniae and tigecycline 50mg twice a day was administrated intravenously to the patient according to sensitivity results. An abdominal US revealed the presence the gallbladder with calculi in a superficial position and the fistulogram revealed a cholecystocutaneous fistula arising from the fundus of the gallbladder. At laparotomy a fistula track was found connecting the gallbladder fundus to the skin, which was dissected and a cholecystectomy was performed. Spontaneous cholecystocutaneous fistula is rarely observed today, mostly as a complication of chronic calculous cholecystitis. Most often it arises from the gallbladder fundus and the clinical presentation is that of a painless draining sinus tract in the right upper quadrant. Diagnosis is aided by abdominal CT scan and ultrasound and treatment is with elective cholecystectomy and excision of the fistula.*

KEY WORDS: Cholecystitis, Fistula, Gallbladder, Liver

## Introduction

Spontaneous cholecystocutaneous fistula (CF) is defined as a rupture of the gallbladder through all layers of the abdominal wall with the formation of a fistulus tract that connects the gallbladder to the skin <sup>1</sup>. CF may be secondary to abdominal trauma, previous abdominal surgery, gallbladder stones and biliary malignancy <sup>2</sup>. Spontaneous CF is rarely observed today, mostly as a complication of chronic calculous cholecystitis <sup>3</sup>,

although there have been cases of spontaneous CF complicating acalculous cholecystitis and carcinoma of the gallbladder <sup>2,3</sup>. CF was first reported by Thilesus in 1670. Large series of CF have been reported in the 19<sup>th</sup> century. Courvoisier has reported 169 cases of CF out of a total of 499 gallbladder perforations while Naunyn has reported 184 cases <sup>4,5,6</sup>. In the past half century fewer than 25 cases have been reported <sup>3</sup>, because of the early diagnosis of gallbladder calculous diseases with ultrasound and the effective treatment with intravenous broad spectrum antibiotics and elective and emergency surgical management of biliary tract disease <sup>3,4</sup>.

## Case report

A 71 year old male presented to the emergency department with a persistent bilious drainage from an old surgical scare, from surgical drainage, of the right upper

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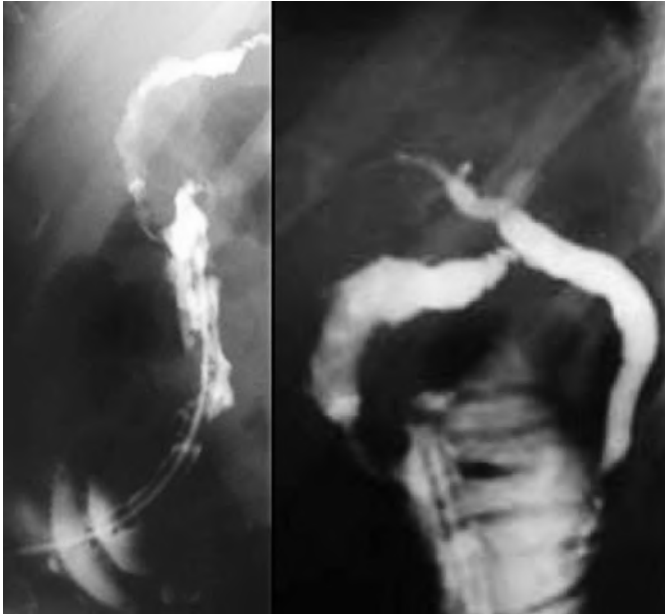


Fig. 1: Fistulogram through skin opening showing a cholecystocutaneous fistula arising from the fundus of the gallbladder by revealing entrance of dye into the biliary tree. Also no evidence of obstruction is observed and free flow of the dye into the duodenum is depicted.

abdominal quadrant for about a week. He had a history of right upper quadrant abdominal pain for about three weeks. He had undergone surgery 14 months ago through a middle epigastric incision for a penetrating gastric ulcer which was sutured and a drainage was placed in the right upper abdominal quadrant. Besides from the peptic ulcer his medical history revealed diabetes mellitus and arterial hypertension under treatment.

Clinical examination revealed a draining sinus and light abdominal tenderness of the right upper quadrant. The patient was afebrile with stable vital signs.

Haematological examinations showed a normal white cell count of  $5100/\text{mm}^3$  while liver function studies and other laboratory tests were within the normal limits. Cultures from the draining fluid grew *Staphylococcus hominis*, *Escherichia coli* and *Klebsilla pneumoniae* and tigeccycline 50mg twice a day was administrated intravenously to the patient according to sensitivity results. Biochemical examination of the fluid revealed aspartate transaminase 665 IU/L, Gamma-glutamyltransferase 1539 IU/L, Lactate dehydrogenase 16410 IU/L, alkaline phosphatase 1712 IU/L and bilirubin 3.12 mg/dL.

An abdominal US was performed which revealed the presence the gallbladder in a superficial position. The gallbladder was contracted and wizened, with calculi and thickening of the gallbladder wall while the intrahepatic and extrahepatic bile ducts and the common bile duct had normal size. A possible diagnosis of an enterocutaneous or cholecystocutaneous fistula was made. The fistulogram revealed a cholecystocutaneous fistula arising from the fundus of the gallbladder (Fig. 1).

At laparotomy a fistula track was found connecting the gallbladder fundus to the skin. The fistula was dissected and a cholecystectomy was performed.

Histopathologic examination of the gallbladder revealed chronic cholecystitis and cholesterolysis of the mucous. The postoperative course was unremarkable and the patient was discharged 7 days after surgery.

## Discussion

Spontaneous perforation of the gallbladder occurs in 10% of patients with acute calculous cholecystitis. It usually perforates towards the duodenum or colon and rarely to the bronchial tree, stomach, urinary tract or skin<sup>3,4</sup>. However, cases of cholecystoduodenal fistula caused by duodenal ulcer have also been reported<sup>7,8</sup>. While CF has even been reported to patients of young age, it usually presents in elderly females over 60 years old<sup>3,4</sup>. The typical presentation is that of a painless draining sinus tract in the right upper quadrant, but it may also appear in the left upper quadrant, the umbilicus, the right iliac fossa, the right groin, the right gluteal region, anterior chest wall and back<sup>3,4,9</sup>. Sometimes, the CF may present as an enlarging mass before spontaneous rupture<sup>4</sup>. In most cases, the patient's history is suggestive of biliary disease but fail to report an episode of acute cholecystitis<sup>4</sup>. In the present case the fistula was draining to the right upper quadrant at the site of an abdominal scar from previous surgical drainage, which has been reported only once before<sup>10</sup>. Differential diagnosis includes infected epidermal inclusion cyst, pyogenic granuloma and metastatic carcinoma<sup>2,4</sup>.

In almost all cases, there is obstruction of the cystic duct or the gallbladder usually because of calculi and rarely carcinoma which causes increased intraluminal pressure of the gallbladder<sup>1,3,4</sup>. This leads to impaired vascular supply and lymphatic drainage which can cause mural necrosis and perforation<sup>1,3,4</sup>. Perforation may be acute, resulting in peritonitis, subacute, resulting in abscess formation and chronic, resulting in fistula formation, internal or external<sup>1,3,4</sup>. Chronic perforation, most often arises from the gallbladder fundus. In these cases chronic inflammation may cause the gallbladder fundus to adhere to the abdominal wall and form a sinus tract, thus creating a fistula<sup>3,4</sup>.

Diagnosis is aided by abdominal CT scan and ultrasound. Ultrasound may demonstrate an abscess as a sonolucent mass with echogenic material adjacent to the anterior abdominal wall, while CT can delineate the abscess and show the fistula<sup>9</sup>. The diagnosis is confirmed by fistulogram which allows visualization of the origin and course<sup>4</sup>.

Management of CF depends on the presence of acute inflammation. In cases of abscess formation broad spectrum antibiotics and incision and drainage of the abscess is mandatory, including adequate analgesia and resusci-

tation<sup>4,9,11</sup>. After the acute inflammation has subsided elective cholecystectomy and excision of the fistula is performed usually by an open approach although laparoscopic treatment is also feasible<sup>3,4,9</sup>.

### Riassunto

Si presenta un raro caso di fistola colecisto-cutanea drenante attraverso una vecchia cicatrice chirurgica nel quadrante superiore destro dell'addome quale evoluzione di una colecistite cronica litiasica. Un uomo di 72 anni si era rivolto al nostro dipartimento di emergenza per la persistenza da circa una settimana di una fuoriuscita di bile attraverso la vecchia cicatrice chirurgica di un drenaggio a livello del quadrante superiore destro dell'addome. L'esame colturale del fluido drenante aveva dimostrato lo sviluppo di *Staphylococcus hominis*, *Escherichia coli* e *Klebsilla pneumoniae*, e seguendo le prove di sensibilità vennero somministrati per via endovenosa 50 mg due volte al dì di tigecycline.

Con l'ecografia addominale si dimostrò la presenza di una colecisti in posizione superficiale e sede di calcoli, ad una fistolografia dimostrò una fistola colecisto-cutanea che si originava dal fondo della colecisti. Alla laparotomia venne isolato un tratto fistoloso in connessione tra il fondo della colecisti e la cute, e si provvide alla sua asportazione insieme alla colecisti.

La fistola spontanea colecisto-cutanea è attualmente di rara osservazione, per lo più come complicanza di una colecistite litiasica cronica. Più spesso essa trae origine dal fondo della colecisti e il quadro clinico che ne consegue è quello di un orificio drenante nel quadrante addominale superiore destro in assenza di dolore. La diagnosi è facilitata dalla TC addominale e dall'ecografia, ed il suo trattamento è la colecistectomia di elezione con escissione del tramite fistoloso.

### References

1. Carragher AM, Jackson PR, Panesar KJ: *Subcutaneous herniation of gall-bladder with spontaneous cholecystocutaneous fistula*. Clin Radiol, 1990; 42:283-84.
2. Chang SS, Lu CL, Pan CC et al.: *Spontaneous cholecystocutaneous fistula presenting with a cellulitis and portal vein thrombosis*. J Clin Gastroenterol, 2002; 34:99-100.
3. Ijaz S, Lidder S, Mohamid W et al.: *Cholecystocutaneous fistula secondary to chronic calculous cholecystitis*. Case Rep Gastroenterol, 2008; 2:71-75.
4. Vasanth A, Siddiqui A, O'Donnell K: *Spontaneous cholecystocutaneous fistula*. South Med J, 2004; 97:183-85.
5. Courvoisier L: *Pathologie and Chirurgie der Gallenwege*. Leipzig, 2. Germany: FCWVogel: 1890.
6. Naunyn B: *Ulcerative affections of the biliary passage and fistula formation*. In: *A Treatise on Cholelithiasis*. New Sydenham Society; (English version 1896). New Sydenham Society, 1892; 138-51.
7. Cavallaro A, Lauretta A, Lizzio A, Cavallaro M, Cavallaro V: *Duodeno-colecystic fistula, a rare complication of duodenal ulcer*. A case report (In Italian). Ann Ital Chir, 2005; 76:573-76.
8. La Greca G, Grasso E, Sofia M, Gagliardo S, Barbagallo F: *Complicated duodeno-biliary fistula in bleeding duodenal ulcer: Case report an literature review* (In Italian). Ann Ital Chir, 2008; 79:57-61.
9. Malik AH, Nadeem M, Ockrim J: *Complete laparoscopic management of cholecystocutaneous fistula*. Ulster Med J, 2007; 76:166-67.
10. Abril A, Ulfohn A: *Spontaneous cholecystocutaneous fistula*. South Med J, 1984; 77:1192-93.
11. Flora HS, Bhattacharya S: *Spontaneous cholecystocutaneous fistula*. HPB (Oxford), 2001; 3:279-80.

