

# Spontaneous cholecystocutaneous fistula: a rare complication of gallbladder disease

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### Introduction

Fistula is an abnormal condition, which results from abnormal connection between two epithelialized surfaces. Biliary fistulas are rare complications of gallstones, linkage between the biliary tract and various organs. There are two main groups of biliary fistulas: internal and external [1]. An internal biliary fistula forms a connection between the gallbladder and the gastrointestinal tract, typically resulting from chronic cholecystitis [2]. On the other hand, an external biliary fistula establishes a link between the gallbladder and the abdominal wall, with potential causes including spontaneous occurrence, postoperative complications, post-traumatic events, or iatrogenic injuries to the biliary tract [1,3].

Cholecystocutaneous fistula (CCF) belongs to the category of external biliary fistulas, creating a connection between the gallbladder and the skin. There are less than 100 cases of cholecystocutaneous fistula reported. The first reported case of CCF was in 1670 by Thilesus, who described this phenomenon for the first time. Nevertheless, during that

period, fistulas were a frequent complication arising from untreated cholecystitis [5]. As per a study conducted in 2005, a total of 226 cases have been documented, with fewer than 25 reported in the past five decades [4]. We don't see much of gallbladder fistula case in current practice due to rapid advancement in imaging, increased understanding of disease process and timely intervention either interventional radiologically or surgically. Mostly gallbladder calculus disease associated with fistula but at times we do see gallbladder carcinoma present with cholecystocutaneous fistula.

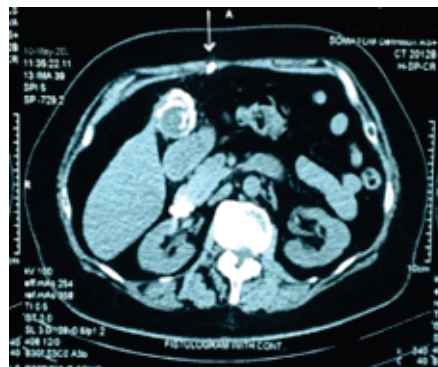
Commonest place of fistula opening is right hypochondrium which is self-explanatory due to underlying gallbladder but other known places are buttock, umbilicus and right inguinal region [4]. This entity is commonly seen in debilitated elderly over 60 years of age. However, cases have been reported in patients aged as young as 24 years. We report a case of a cholecystocutaneous fistula in a patient with previously undiagnosed gallstone disease.

### Case Summary

A 59-year-old woman with the history of hypertension and dyslipidemia was referred to our institution from a private sector with abdominal pain and discharging sinus in the epigastrium for 2 years. She has had excision biopsy of epigastric sinus 1 year back at a different hospital and histology revealed active chronic inflammation.




**Figure 1.** Fistulous opening in the epigastrium



**Figure 2.** Arrow showing the fistula

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Ultrasound of the abdomen showed 2cm tract in anterior abdominal wall in the epigastrium appears to communicate with peritoneal cavity, in favour of fistula formation with background chronic cholecystitis and gallbladder calculi. Subsequently CT fistulogram was preformed and findings were diagnostic of cholecystocutaneous fistula.

Liver function tests were normal, and there were no contraindications for surgery under general anesthesia. We performed laparoscopy and decided to convert to open surgery due to the density of the adhesions specially at the Calot's triangle. At laparotomy, the fistulous tract was demonstrated and found to enter the fundus of the gallbladder. Gallbladder was thick, fibrotic, and hard in consistency and was seen to adhere to the liver, with surrounding hard induration of the liver, and also the cystic duct - common hepatic duct junctional area, raising the suspicion of carcinoma of the gallbladder. There were several large and hard stones within the gallbladder. We proceeded with en bloc excision of aponeurotic muscle, skin and fistulous tract together with the gallbladder, and a 5cm cuff of the liver, and cystic duct - common hepatic duct junction, and the common bile duct down to the level of the superior border of the duodenum. A jejunal Roux-en-Y loop was raised, and end to side hepaticojejunostomy performed with 5/0 Polydioxanone interrupted sutures. A subhepatic drain was left in situ and patient received broad-spectrum antibiotics during and after surgery. The patient made a slow but uncomplicated recovery and was discharged home well on post-op D6. Superficial wound infection was noted on D9, which was managed with oral antibiotics and by D20 wound was completely healed. Histology of the specimen confirmed acute on chronic cholecystitis with a fistula between the skin and the gall bladder lumen, without evidence of malignancy or tuberculosis.

## Discussion

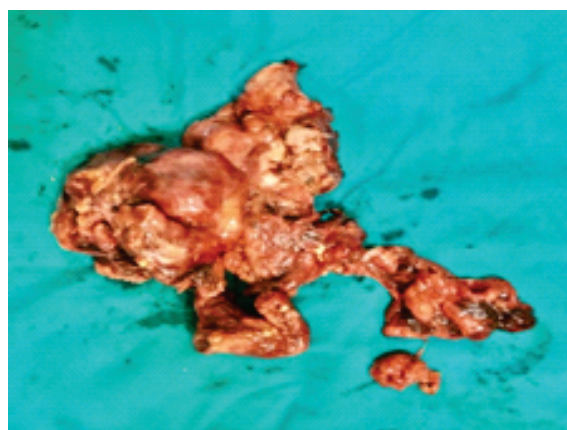
The better understanding of pathology and evolution of sophisticated investigations lead to a rare occurrence of spontaneous cholecystocutaneous fistula. Over the past 50 years, less than 20 cases of spontaneous CCFs have been reported [4]. A neglected biliary tract disease is being the culprit of CCF. They are painless and right upper hypochondrium being the commonest location. However, they have been reported at the umbilicus, left costal margin, right iliac fossa, right groin and the back [5]. A pyogenic granuloma, infected epidermal inclusion cyst, chronic osteomyelitis of ribs, enterocutaneous fistula, discharging tuberculosis and metastatic carcinoma should be considered as mimics of CCF by external appearance [5]. The patency of the cystic duct determines the nature of the discharge which can be purulent, mucoid or bile.

In a recent analysis by Kaminsky concerning the prevalence of biliary fistulas to the gastrointestinal tract, the majority were observed to be connected to the duodenum (60%), followed by the colon (24%), stomach (6%), and choledochal duct (5%). Out of all, only 2% cholecystocutaneous abscesses or fistulas are accounted. Key factors increasing the risk of spontaneous cholecystocutaneous fistulas (CCFs) encompass older women (> 50 years of age), steroid therapy, a history of typhoid, bacterial spread, trauma, immunocompromised conditions, etc.

The pathophysiology of CCF can be studied step by step in following manners: cholecystocutaneous fistula is a sequelae of cystic duct blockage leading to increased gallbladder pressure, either caused by stone or malignancy. As a result of increased intraluminal pressure, blood and lymphatic flow is compromised to gallbladder, result in mural necrosis and perforation.



**Figure 3.** 3D Constructed fistulogram



**Figure 4.** Resected gallbladder with fistulous tract

In literature, perforation classically described as acute, subacute and chronic. These fistulas, as presented in our case, frequently arise from the fundus of the gallbladder. Famous surgeon Nayman coined the term “empyema necessitatis” – A state prior to spontaneous rupture, also known as “burrowing abscess”.

Based on the underlying etiology, the external biliary fistula management differs. The septic presentation requires adequate antibiotics, analgesics and fluid resuscitation. Not all external biliary fistula warrants surgical intervention because a proportion of patients exhibits spontaneous healing. So, in elderly or debilitated patients major interventions can be avoided. Cholecystostomy and cholecystectomy are the possible surgical options. As cholecystostomy carries the possibility of further stone formation in the gallbladder, cholecystectomy is usually the treatment of choice.

In conclusion, gallstones-disease related complications can be prevented by early laparoscopic cholecystectomy. In patients with anterior abdominal wall discharging sinus should warrant early referrals. Rare possibility of malignancy should be kept in mind while dealing with spontaneous CCF. In these cases, proper preoperative planning with imaging like CT scan is pivotal. At last, patient's clinical status, local expertise and best post-operative outcome decide whether

laparoscopic versus open and one-stage versus two-stage approach.

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## Learning Points:

- Early laparoscopic cholecystectomy for gallstones may prevent chronic cholelithiasis complications.
- High level of suspicion needs to be maintained in patients with discharging sinus located in the anterior abdominal wall.
- Rare possibility of malignancy should be kept in mind while dealing with spontaneous CCF.
- Judicious use of CT as imaging modality should be considered to rule out the diagnosis and for proper preoperative planning.