

## Spontaneous Cholecystocutaneous Fistula: Still a Complication of Gallstones Disease

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### Abstract

A cholecystocutaneous fistula is a rare complication of gallstone disease. The first report was written in the seventeenth century by Thileus. During the last 50 years, 25 cases have been reported. This presentation was a case of cholecystocutaneous fistula successfully treated with excision and cholecystectomy. A 60-year-old lady presented with a persisting discharge from what was thought to be an 'abscess' in the right hypochondrium, which was spontaneously ruptured and afterwards it had been excised and drained. Investigation with ultrasound scanning and sinography, demonstrated a cholecystocutaneous fistula and calculous cholecystitis. The fistulous tract was excised together with the gallbladder. This condition was rarely seen nowadays due to the greater availability of antibiotic therapy and biliary surgery. Cholecystectomy was the preferred treatment for this case, the laparoscopic approach was an option reserved for the expert surgeon. Although in a few patients, the fistula may close spontaneously.

**Keywords:** Cholecystocutaneous fistula, Gallstones, Sinogram, Cholecystectomy.

### Introduction

Gallstones can present in different ways. It may present with biliary colic, acute cholecystitis, chronic cholecystitis, obstructive jaundice & even gallstone ileus.<sup>1</sup> Spontaneous cholecysto-cutaneous fistula defined as the link between gallbladder and exterior by means of rupture of abdominal wall layers, all of which strengthens the fistula tract.<sup>2</sup> Thileus first described an external biliary fistula in 1670. Courvoisier reported 169 cases in the nineteenth century.<sup>3</sup> However, since 1900 just 65 cases have been reported<sup>4</sup>, and only 15 in the last 50 years.<sup>5</sup> The condition has changed from being a complication of suppurative cholecystitis to being caused by trauma, which is usually operative.<sup>6</sup> The rarity of external biliary fistula stems from improved diagnostic investigations such as ultrasound scans and the greater availability of treatment

in the form of antibiotics and biliary surgery. This report was a case of a cholecystocutaneous fistula in a patient with previously undiagnosed gallstone disease.

### Clinical Presentation

An elderly lady, Mrs. Jahanara Begum (60) attained outpatient department (OPD) of North East Medical College Hospital (NEMCH), Sylhet on 18<sup>th</sup> August 2014 with complains of chronic discharging sinus at the upper right side of abdomen for 6 years (Fig 1). She was admitted into Female Surgical Ward (FSW) in this hospital as an academic case. The patient gave the past history of insidious onset of a swelling in right upper abdomen which was spontaneously ruptured 6 years back then she admitted into Sylhet MAG Osmani Medical College Hospital (SOMCH) in surgery ward on 09.02.2008 as chronic discharging sinus and was operated (Excision of sinus tract) under local anaesthesia, followed by daily dressing. Histological report of sinus tract was granulation tissue. Her condition improved some extends but not cured and she visit to OPD of SOMCH on 24.04.2008 and was advised for daily dressing with sinogram. Patient did not perform the sinogram.

After got admission into NEMCH she was advised for Ultrasonography of whole abdomen, sinogram, haematological investigations. USG show distorted anechoic area in gallbladder fossa, part of Common Bile Duct (CBD) & intrahepatic channels were mildly dilated, no intra-abdominal fluid or subcutaneous collection noted in sinus area (Fig 2). Sinogram show fistula tract between exterior and gallbladder and contrast passed into cystic duct and CBD (Fig 3). So the working diagnosis cholecystocutaneous fistula made and the patient was counseled about the disease condition and for laparotomy and cholecystectomy with excision of fistula tract.

Laparotomy through right Kocher's incision under general anaesthesia, gross intra-abdominal adhesion of omentum with gall bladder. Fistula tract from skin communicate with gall bladder and stone impacted within fundus at

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the internal end of fistula tract (Fig 4). Cholecystectomy and excision of fistula tract done (Fig 5) and sent for histopathological examination. Abdomen closed in layers after keeping a drain in situ. The recovery of the patient was uneventful and the patient keeps on oral feeding after 2<sup>nd</sup> post operative day. Histology report of the tract was chronic inflammatory tissue, no malignant cells or tubercular lesion found. Her wound stitches were off after 2 weeks. Wound found healthy and dry (Fig 6).



**Figure 1:** External fistulous opening



**Figure 2:** Per-operative fistula tract with Gallbladder



**Figure 3:** USG anechoic Gallbladder



**Figure 4:** Fistulogram showing the tract is continuous with the gallbladder



**Figure 5:** Post cholecystectomy specimen



**Figure 6:** Healthy & dry postoperative wound along with the fistulous tract

## Discussion

Biliary fistulas can be either internal or external. Internal fistulas 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 cholecystoduodenocolic fistula.<sup>7</sup> Spontaneous cholecystocutaneous fistula was a common complication of gallstones until the beginning of twentieth century and a total 226 cases have been reported till now.<sup>8</sup> It has become a rare complication of chronic calculus cholecystitis because currently gallstones are diagnose & treated at early stage.<sup>8</sup> They usually complicate gallstone disease, but can occur secondary to biliary injury during a surgical procedure<sup>6</sup>, cholangiocarcinoma<sup>9</sup> and other traumatic causes.<sup>10</sup> The external opening of a cholecystocutaneous fistula is generally in the right hypochondrium. However, other sites can be involved such as the left hypochondrium (45%), the umbilicus (27%), the right lumbar region, the right iliac fossa<sup>3</sup> and the gluteal region.<sup>11</sup>

A fistula such as this one is an end result of perforation of the gallbladder secondary to acute calculous cholecystitis. Perforation of the gallbladder can also occur, albeit rarely, in the absence of gallstones.<sup>4</sup> Chronic bile outflow obstruction increases intra-gallbladder pressure, decreases gallbladder perfusion proportionally and leads to necrosis and perforation of the gallbladder.<sup>12</sup> Once gallbladder becomes perforated, it may drain into the peritoneal cavity, adjacent viscera or adhere to the abdominal wall to form an external fistula. Most patients are females in fifth decade of life and an erythematous lesion may be the only presenting sign. It has been suggested to be associated with polyarteritis nodosa, trauma, typhoid and drug treatment.<sup>8</sup> Two cases of combined internal and external fistulas have been described, communicating in each case with the duodenum.<sup>13,14</sup>

Ultrasonography and fistulogram are presently the imaging modalities for diagnosing this complication. However, magnetic resonating cholangiopancreatogram may be helpful for diagnosis. If it presents early with an abscess, drainage with proper antibiotic is the initial treatment of choice. Subsequently, elective cholecystectomy with excision of the fistulas tract has been suggested as a definitive treatment modality. However, a percutaneous treatment may be considered in high-risk patients.<sup>14</sup> Malik et al. described a different approach that involves the laparoscopic removal of the gallbladder and dissection but without excision of the fistula from the abdominal wall.<sup>15</sup> However, the process unleashed by the pathology is related to the chronic and severe gallbladder inflammation. It also jeopardizes bile duct structures with minimal invasion.<sup>16-20</sup> This approach may provide an alternative option to open excision of the fistula for co-morbid elderly patients.

## Conclusion

There is currently low incidence of cholecystocutaneous fistula and it seems that the current trend is for it to become

an anecdotal condition. However, surgeons should keep in mind by as one of the complications of biliary related pathologies. Even though the efforts of health system in establishing a prompt diagnosis and treatment for gallbladder diseases that reduced theseverity of complications such as fistulation, in the lowest social strata of the population. There is still a small percentage that does not have access to such benefits and that is referred to a surgical service with pathologies as described in this text.

There must be preoperative comprehensive assessment that informs the fistula anatomy as a bile duct in order to establish an optimal surgical plan. As regards the approach, abdominal exploration by sub-costal means remain as the first option due to the etiological features of the condition, and since laparoscopic access is an alternative reserved for expert surgeons, as well as the patience needed to treat the pathology comprehensively via this access.

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