Spontaneous Cholecystocutaneous Fistula: A Rare Presentation of Gallstones

Authors: Leela Sayed (1), Sam Sangal (2) and Guy Finch (3)
Location: (1) University of Leicester Medical School, Leicester (2) Leicester General Hospital, Leicester (3) Northampton General Hospital, Northampton, UK
Citation: Sayed L, Sangal S, Finch G. Spontaneous Cholecystocutaneous Fistula: A Rare Presentation of Gallstones. JSCR. 2010 5:5

ABSTRACT

Spontaneous cholecystocutaneous fistula, one of the rarest complications of acute cholecystitis, has been reported in fewer than 25 cases over the past 50 years. Not only is this case rare but interestingly the patient experienced no pain or symptoms consistent with gallbladder pathology leading up to her hospitalisation. Furthermore, laboratory studies, microbiology and computed tomography scanning did not establish a diagnosis until the fistula passed calculi.

An 85-year-old lady with multiple co-morbidities presented to the Emergency Department with an erythematous soft and non-tender mass in her right flank. The mass had spontaneously ruptured and was discharging a serous-like material. Prior to further investigation a working diagnosis of an eroding/fungating caecal tumour was made. The lesion continued to discharge over a 3 month period which heralded the passage of 11 small, brown calculi thought to be gallstones. At this point spontaneous cholecystocutaneous fistula was diagnosed and was later confirmed by magnetic resonance imaging cholangiopancreatography.

INTRODUCTION

Reports of spontaneous cholecystocutaneous fistulae have been found in medical literature dating back to the 17th century. Fortunately, spontaneous cholecystocutaneous fistulae are a rare complication of cholelithiasis nowadays as prompt medical and surgical treatments address the underlying pathology when symptomatic. Diagnosis of this rare entity often proves challenging as a significant proportion of patients with this complication present with non-specific symptoms.

CASE REPORT

An 85-year-old lady with diagnosed gallstones (1996), venous leg ulcers, congestive heart failure, hypertension, atrial fibrillation, anaemia and hypothyroidism presented to her GP with a 10 day history of a swelling in her right flank. Three days later the mass spontaneously ruptured and she arrived at the Emergency Department with a 3cm x 2cm erythematous, soft and non-tender mass which appeared to be discharging a serous-like material. When questioned she denied any previous history of pain, sudden weight loss, anorexia, jaundice or
upper or lower gastrointestinal disturbances. Physical examination was essentially unremarkable aside from a heart rate of 128 beats per minute.

The patient was admitted for further investigations. Laboratory investigations revealed that she was anaemic (haemoglobin; 10.3g/dL), hyperkalaemic (5.3 mmol/L) and her WBC count was within the lower limits of normal (4.8 x 10^9/L). At this point a working diagnosis of an eroding/fungating caecal tumor was made. A computed tomography scan showed a low density mass measuring 8.1 x 2.6 x 5.1cm in the right lateral wall of the abdomen (Figure 1A). There was no evidence of a bowel related mass; however there was a track of abnormal density that appeared to extend either from the biliary tree or the second part of the duodenum to the abdominal wall (Figure 1B). The gallbladder was unidentifiable and the common bile duct measured 1cm in diameter; the upper limit of normal. A wedge biopsy reported purulent granulomatous tissue formation consistent with abscess formation and stated that there was no evidence of atypia or malignancy. The patient was discharged back under the care of her GP with the view of managing what was thought to be an abscess.

Over a three month period the lesion continued to discharge and required thrice weekly dressings by a district nurse. It was only when the district nurse reported 11 small, brown, hard calculi present in the dressings it became clear that the abscess was in fact a tract that directly communicated with the gallbladder/biliary tree; a spontaneous cholecystocutaneous fistula. The diagnosis was later confirmed by magnetic resonance imaging cholangiopancreatography (MRCP) which reported a track of fluid that ran from the superior aspect of the common bile duct and passed to the lateral abdominal wall (Figure 2A). Multiple calculi were still present in the common bile duct (Figure 2B).

It was thought that removal of any remaining calculi may aid spontaneous closure of the
fistula. Given the patient’s age and co-morbid state it was decided that a conservative approach was appropriate and removal of the calculi was performed by endoscopic retrograde cholangiopancreatography (ERCP) balloon trawl and sphincterotomy. Figures 3 and 4 show the fistula 6 months after initial presentation and following endoscopic retrograde cholangiopancreatography and balloon trawl.

**DISCUSSION**

Spontaneous cholecystocutaneous fistulae generally form as a result of neglected cholelithiasis, although a handful of cases have reported acalculous cholecystitis and carcinoma of the gallbladder as causative factors (1,2). Biliary outflow obstruction increases intramural pressure, restricts gallbladder perfusion and precipitates necrosis and perforation of the gallbladder. Once perforated, it may drain into the peritoneal cavity, adjacent viscera or less commonly adhere to the abdominal wall to form an external fistula. Most frequently, external biliary fistulae drain via a sinus in the right upper quadrant, however alternative locations such as the umbilicus, right groin, anterior chest wall and the gluteal region have also been reported (3,4).

Treatment should include broad-spectrum antibiotics, drainage of the abscess and elective cholecystectomy with excision of the fistula. Malik et al describe an approach that involves the laparoscopic removal of the gallbladder and dissection but not excision of the fistula from the abdominal wall (3). This approach may provide an alternative option to open excision of the fistula for co-morbid elderly patients.

This case elucidates the importance of maintaining a high level of suspicion and a low threshold for referral if a similar picture presents in primary care, particularly if the patient is
elderly. A significant proportion of elderly patients do not have the classic symptoms normally associated with cholecystitis (5). Their symptoms tend to be more non-specific and often complicated by co-existent disease. Delays in presentation due to psychosocial obstacles consequently mean that their disease may be more advanced or associated with more complications at initial presentation.

REFERENCES

1. Birch BR, Cox SJ. Spontaneous biliary fistula uncomplicated by gallstones. Postgrad Med J. 1991;67:391-2
2. Vasanth A, Siddiqui A, O'Donnell K. Spontaneous cholecystocutaneous fistula. South Med J. 2004;97:183-5
3. Malik AH, Nadeem M, Ockrim J. Complete laparoscopic management of cholecystocutaneous fistula. Ulster Med J. 2007;76:166-167
4. Nicholson T, Born MW, Garber E. Spontaneous cholecystocutaneous fistula presenting in the gluteal region. J Clin Gastroenterol. 1999;28:276-7
5. Lyon C, Clarke DC. Diagnosis of acute abdominal pain in older patients. Am Fam Physician. 2006;74:1537-1544