

Spontaneous cholecysto-antral-cutaneous fistula: a consequence of neglected calculus cholecystitis

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ABSTRACT A biliary fistula from the gallbladder can develop as a result of the erosion of stones into the surrounding viscera. An 82-year-old woman with multiple comorbid diseases presented with a persisting discharge from what was thought to be an abscess in the right hypochondrium, which had previously been drained. Sinogram confirmed fistulous communication with the gallbladder and gastric antrum. The fistulous tract was excised together with the gallbladder, and the gastric defect was repaired. The patient made an uneventful recovery.

Keywords: biliary, calculus cholecystitis, complication, fistula, gastric
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INTRODUCTION

The first description of a spontaneous external biliary fistula was given in 1670 by Thelius.⁽¹⁾ In the 19th century, Courvoisier reported 169 cases of biliary fistula.^(1,2) Since then, less than 100 cases of cholecystocutaneous fistula have been reported in the literature.⁽¹⁻³⁾ The natural history of the disease has changed from suppurative cholecystitis with spontaneous rupture to operative external drainage of an abscess.⁽¹⁻⁴⁾ With the improvement in imaging techniques, antibiotics, operative techniques and perioperative care, such situations are rarely encountered. Early convalescence and low morbidity associated with laparoscopic cholecystectomy has brought about a revolution in the management of patients with biliary lithiasis.⁽⁵⁾ Occasionally, in a neglected, compromised and debilitated patient with a long-standing disease, such a catastrophe can occur.^(1-4,6) A debilitated woman who developed cholecystocutaneous fistula forms the basis of this present report.

CASE REPORT

An 82-year-old Indian woman who was suffering from diabetes mellitus, rheumatoid arthritis, seizure disorder and chronic obstructive airway disease presented with symptomatic biliary lithiasis for the past ten years. She developed redness, induration and swelling in the right hypochondrium three months prior to presentation. The symptoms resolved following mucopurulent discharge from the indurated area. She consulted a family physician who performed incision and drainage with a presumptive diagnosis of incompletely drained abscess. The patient subsequently noticed bile discharge from the drainage site.

Physical examination was unremarkable except for a bile-discharging sinus in the right hypochondrium. The patient's haematological, biochemical and coagulation parameters were normal. Sinogram revealed communication with the gallbladder, with multiple filling defects. The contrast was seen to reach the

gastric antrum through an abnormal communication (Figs. 1 & 2). The rest of the biliary system was not visualised. Magnetic resonance cholangiopancreatography (MRCP) revealed a normal biliary anatomy with evidence of cholelithiasis.

Cholecystectomy with excision of the fistulous tract and repair of the gastric antrum was performed through a subcostal incision. The gallbladder was found to be small, contracted and densely adherent to the parietal wall at the fistula site. The anatomy of Calot's triangle was normal. Communication with the gastric antrum was observed in the Hartmann's pouch. The gallbladder was carefully dissected off the gastric antrum. The defect in the gastric antrum was sutured with interrupted absorbable sutures. The gallbladder was thick-walled, with normal mucosa, and its lumen contained two stones, each measuring approximately 1 cm in diameter. Histopathology of the gallbladder and the excised fistulous tract, including the gastric antrum, revealed chronic inflammation. The patient had an uneventful recovery and was well until three years into follow-up when she died of an unrelated cause.

DISCUSSION

Spontaneous internal biliary fistulae are relatively common and well-described in the literature. More than 90% of them communicate with the duodenum or the colon.⁽⁵⁾ The remaining 10% communicate with the stomach or jejunum, or may have multiple communications.⁽⁵⁾ Postoperative external biliary fistulae are common,⁽⁵⁾ but those arising as a result of complicated biliary disease are a rarity.⁽¹⁻⁴⁾ This is evident from the fact that only 20 such cases have been reported in the last 50 years.⁽⁴⁾ This low incidence can be attributed to improved antibiotics and perioperative care.

The external opening of the cholecystocutaneous fistula is invariably located at the right hypochondrium.⁽¹⁻⁴⁾ However,

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Fig. 1 Sinogram (in left anterior oblique view) image shows opacification of the gallbladder and stomach by the non-ionic contrast injected through the external opening in the right hypochondrium. Both cholecystocutaneous (arrow) and cholecysto-antral (arrowhead) fistulae are outlined by the contrast.

other sites such as the umbilicus, left hypochondrium, lumbar,⁽⁶⁾ gluteal regions⁽⁷⁾ and the chest wall⁽⁸⁾ have also been described. In neglected cases, the condition may go on to cause portal vein thrombosis.⁽⁹⁾ This condition is usually seen in association with the presence of gall stones,^(1,3,4,6-9) although spontaneous occurrence without any biliary lithiasis has also been described.⁽²⁾

The simultaneous occurrence of an internal and external fistula is a rare event.^(8,10,11) To the best of our knowledge, only four such cases describing communication with the duodenum have been reported.⁽⁸⁻¹¹⁾ The occurrence of internal communication with the gastric antrum is a unique feature of the present case. This condition is usually seen in the elderly with comorbid conditions who have decreased host defense mechanisms and, probably, compromised blood supply to the gallbladder.^(1,3,4,8,9) This disease is usually long-standing.⁽¹⁻⁴⁾ Repeated episodes of inflammation results in adhesion formation with the parietal wall and adjacent viscera.⁽⁵⁾ Perforation of the gallbladder in such a situation is usually localised and can lead to fistula formation.^(1-4,6,7,9) The presence of large stone(s) further facilitates the process of fistulisation by pressure necrosis.^(10,11)

Usually, the efflux of the external fistula is mucopurulent, as an impacted stone or a tumour blocks the neck of the gallbladder.^(1,3,8,9) In the present case, the presence of bile-stained discharge may indicate the presence of an internal fistula and the release of the blockage. However, copious discharge of bile should raise the suspicion of a distal obstruction.⁽⁵⁾ This condition is best detected by a sinogram using a water-soluble contrast, which can delineate the anatomy of the fistula.^(1-4,6,7) However, more often than not, it fails to demonstrate the biliary anatomy.⁽¹⁻⁴⁾ Although MRCP may not be able to visualise fistulae, it is often required for the evaluation of the biliary system.^(1,5)

The primary treatment is drainage of abscess and control of sepsis. Cholecystectomy and excision of the fistula tract are



Fig. 2 Delayed sinogram image (taken ten minutes later in anterior view) shows filling defects in the contrast-opacified gallbladder, suggestive of gallstones. The gallbladder is seen in close proximity to the gastric antrum. Note the contrast visualised in the small bowel.

curative.^(1-4,6-13) Abscess drainage and cholecystectomy can be performed as a single-^(1,12) or two-stage procedure.⁽¹³⁾ Both conventional^(1,3,4) and laparoscopic approaches^(12,13) to tackle this problem have been described. Occasionally, nonoperative management via the placing of internal and external stents in a moribund patient has also been described.⁽¹¹⁾

In conclusion, cholecysto-antral-cutaneous fistula is a rare entity that is usually seen in elderly and debilitated patients. Contrast sinogram can delineate the anatomy of the fistula, and cholecystectomy is curative.

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