CASE REPORT

Rare case of Mirizzi syndrome associated with cholecystogastric fistula

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SUMMARY

A woman in her mid-30s presented with upper right quadrant abdominal pain. On examination, there was mild upper right quadrant tenderness and negative Murphy's sign. Basic laboratory investigations revealed normal results except for elevated alkaline phosphatase. Subsequent imaging revealed chronic calculous cholecystitis and the dilated common bile duct harbouring stones, with pneumobilia. The patient underwent open cholecystectomy with common bile duct exploration. Intraoperatively, the gallbladder was found contracted, and densely adherent to and fistulising with the common bile duct and stomach.

BACKGROUND

This case shows a rare presentation of Mirizzi syndrome (MS) with cholecystogastric fistula.

CASE PRESENTATION

A woman in her mid-30s presented with a 1-month history of upper right quadrant abdominal pain. A few days prior to her presentation, the pain had increased in severity. She had no fever, no gastrointestinal disturbances and no jaundice. She had no history of any medical illnesses. On examination, her abdomen was soft and non-distended, with mild upper right quadrant abdominal tenderness and negative Murphy's sign. General examination was otherwise unremarkable.

INVESTIGATIONS

Haematological and biochemical investigations revealed normal results apart from elevated alkaline phosphatase of 453 IU/L (upper limit of normal is 104 IU/L).

Ultrasonography of the abdomen revealed chronic calculous cholecystitis with dilated common bile duct (CBD) harbouring stones, with pneumobilia (figure 1).

Endoscopic retrograde cholangiopancreatography (ERCP) was performed but failed to extract the CBD stones. The contrast was going to the stomach, rather than filling the CBD (figure 2).

MR cholangiopancreatography (MRCP) revealed a 2.5 cm CBD with multiple stones, the largest measuring 2.4×1.7 cm, with moderate intrahepatic biliary dilatation (figure 3).

TREATMENT

An open exploration of the CBD was performed. Hence, an intraoperative diagnosis of MS was concluded. A small contracted gallbladder fistulising to

Figure 1 Ultrasonography of chronic calculous cholecystitis with dilated common bile duct harbouring stones, with pneumobilia.

the CBD and to the stomach was found (figures 4 and 5). Cholecystectomy with excision of distal CBD and division of cholecystogastric fistula, and Roux-En-Y hepaticojejunostomy was performed.

OUTCOME AND FOLLOW-UP

The patient did well postoperatively, and a 2-year follow-up showed no complications.

DISCUSSION

MS, first described by Mirizzi,¹ is a form of obstructive jaundice that is caused by a stone(s)

Figure 2 Endoscopic retrograde cholangiopancreatography showing common bile duct filling defect.

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Figure 3 MR cholangiopancreatography revealed common bile duct stones and moderate intrahepatic biliary dilation.

impacted in the neck of the gallbladder or the cystic duct, such that the common hepatic duct is narrowed.

Reported incidence of MS is approximately 1%,^{2,3} although it has been reported to be up to 5.7%,^{4,5} in various studies.

The syndrome arises from the sequence of events that initiate from the impaction of a gallstone to the erosion through the gallbladder and the CBD wall, forming a cholecystobiliary fistula.^{6,7} The same pathophysiological process explains other biliary fistulas, such as cholecystoduodenal, cholecystogastric and cholecystocolonic fistulas.⁸

Two pathomechanisms for the formation of an internal fistula are discussed. First, an adherence of the stone-containing gallbladder wall to the surrounding neighbouring structures, necrosis and ulceration of the wall may cause the stone to perforate through the adherent area into the intestine.⁹ Second, in cases of scleroatrophic stone cholecystitis,¹⁰ or obstruction of outflow in the CBD due to an increase in pressure in the gallbladder, a concrement is pressed against the wall, producing mucosal decubitus. Pericholecystitic adhesions are formed by local inflammation, and the stone transmigrates through the gallbladder wall and perforates into a hollow viscus, forming a fistula.¹¹

McSherry *et al.*⁶ proposed a classification that divides MS into two types based on ERCP findings: type I involves the

Figure 4 Cholecystogastric fistula (S, stomach, B, gallbladder merged with distal biliary tree, D, duodenum).

Figure 5 Cholecystogastric fistula (S, stomach, B, gallbladder merged with distal biliary tree, D, duodenum).

external compression of the common hepatic duct by calculus in the cystic duct or Hartmann's pouch. Type II includes a cholecystocholedochal fistula that is caused by calculus eroding the bile duct.

Csendes *et al.*⁷ modified McSherry's classification in 1989, and classified MS into four types: type I is the extrinsic compression of the CBD by an impacted gallstone. Type II consists of a cholecystobiliary fistula, secondary to an eroded gallstone, involving one-third of the circumference of the CBD. In Mirizzi type III, the cholecystobiliary fistula involves two-thirds of the circumference of the CBD. While in type IV, the cholecystobiliary fistula comprises the whole circumference of the CBD.

Since the natural history of MS may not just end with a simple cholecystobiliary fistula, and the continuous inflammation in the triangle of Calot area may result in a complex fistula involving not only the biliary tract but also the adjacent viscera, Csendes, in 2007, added a new type (type V) to his classification of MS, and it was validated by Beltran *et al.*⁵ this type corresponds to any type of MS associated with cholecystoenteric fistula with (V_b) or without (V_a) gallstone ileus.

MS carries a challenge for the surgeon in terms of management. Methods of operation depend on the type. Type I is adequately resolved by partial or subtotal cholecystectomy. While in type II, choledochoplasty with gallbladder flap and bilioenteric anastomosis, and Roux-En-Y hepaticojejunostomy is the preferred surgical approach. Partial cholecystectomy with or without T-tube is performed in type III along with Roux-En-Y hepaticojejunostomy. In type IV, hepaticojejunostomy is preferred. In type V, cholecystectomy, hepaticojejunostomy and division of the fistula are performed.

Although the open approach remains the standard of care,¹² in managing MS, laparoscopy can be used in certain conditions with a low threshold for conversion to open cholecystectomy in distorted anatomy.

Learning points

- ▶ The distorted anatomy in Mirizzi syndrome increases the risk of biliary injury intraoperatively.
- ▶ Biliary drainage into a fistula causes the patient to present with no obstructive jaundice.
- ▶ A laparoscopic approach in types I and II is possible, but a surgeon must lower the threshold for the conversion to open cholecystectomy.

Exploration of CBD should be undertaken due to the occurrence of choledocholithiasis in MS.^{3 13}

Performing a frozen section biopsy from the removed gallbladder is advisable, since there is an association between MS and gallbladder cancer.¹⁴

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Competing interests None declared.

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