CASE REPORT

Complicated triple gallbladder: clinical presentation and surgical approach

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Accepted 3 July 2016

SUMMARY

We report a patient with previous biliary symptoms and endoscopic interventions who presented with clinical features suggestive of choledocholithiasis. Open surgical exploration disclosed three gallbladders with copious stones and varying degrees of acute and chronic inflammation. Literature review revealed only 16 previously reported cases. We review the aetiology of triple gallbladder as being due to failure of rudimentary bile ducts to regress during embryological development, as well as the classification scheme of triple gallbladder based on size, location and number. We also discuss the clinical evaluation and appropriate surgical management of this entity, as triple gallbladders can be associated with cholecystitis or carcinoma.

BACKGROUND

Congenital anomalous biliary multiplications occur in ~1 in 4000 patients, and can be classified based on the size, location and number of accessory gallbladders.^{1–16} The clinical presentation is variable and accurate preoperative diagnosis may require use of several imaging modalities. Despite thorough preoperative workup, however, the diagnosis can be confused with cholangitis, choledocholithiasis or biloma. The clinician must be aware of the possibility of triple gallbladder, as complete removal of all gallbladders via an open or laparoscopic approach is essential for adequate treatment.

CASE PRESENTATION

A 30-year-old man presented with worsening abdominal pain, jaundice and fever. He had a history of cholelithiasis, choledocholithiasis, two endoscopic retrograde cholangiopancreatographies (ERCP) and biliary stent placement. The symptoms of the recent illness began 5 months earlier, 2 months after he had returned from India. A search for an infectious aetiology was unrevealing. On examination, the abdomen was soft but tender in the right upper quadrant; he was jaundiced and febrile to 101.1°F.

INVESTIGATIONS

Total serum bilirubin was 12.2 mg/dL (direct bilirubin 7.9 mg/dL), alkaline phosphatase 218 U/L, alanine aminotransferase 105 U/L, aspartate aminotransferase 38 U/L. Right upper quadrant ultrasound (figure 1) confirmed cholelithiasis, gallbladder sludge and wall thickening; the common bile duct (CBD) measured 7 mm in diameter. Abdominal CT scan (figure 2) revealed the biliary stent in good position; there was a 5.3×11 cm mass in the gallbladder

Figure 1 Preoperative right upper quadrant ultrasound scan with dilated gallbladder suspicious for acute cholecystitis. SAG, sagittal.

Figure 2 Preoperative CT scan abdomen/pelvis with intravenous and oral contrast concerning for postprocedure haematoma in gallbladder fossa.

fossa. Given his abdominal pain coupled with the large collection seen on CT scan, as well as his persistent fevers and leucocytosis, the decision was made to proceed directly to the operating room, instead of first performing MR cholangiopancreatography (MRCP).

DIFFERENTIAL DIAGNOSIS

The differential diagnosis for this patient included cholecystitis, cholangitis, choledocholithiasis, bile leak, biloma or biliary haematoma. Given his elevated bilirubin level despite appropriate positioning

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To cite: Copeland-Halperin LR, Kapoor K, Piper JB. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2016-215901

of the stent on imaging, we were particularly concerned about a large bile leak due to bile duct perforation.

TREATMENT

Following appropriate informed consent, the patient was taken for open surgical exploration with CBD exploration. The gallbladder was necrotic, haemorrhagic and ruptured (figure 3, gallbladder A), and there was evidence of bile peritonitis. Using a fundus-first approach, the gallbladder was dissected free from the cystic duct, which was draining into the CBD. Two fluctuant liver masses were palpated that were initially believed to be abscesses, but on further dissection were found to be lined with gallbladder mucosa (figure 3, gallbladders B and C) and contained clear mucus consistent with a gallbladder hydrops. Their cystic ducts drained individually into the CBD (figure 4) and appeared chronically obstructed. Exploration of the CBD revealed hundreds of small stones that could not be completely cleared, so the biliary stent was left in place to allow for ongoing drainage. ERCP performed 8 weeks postoperatively showed a 15 mm stricture of the lower third of the main bile duct and so his stent was replaced at that time.

OUTCOME AND FOLLOW-UP

The patient tolerated the procedure well and was discharged on postoperative day 2. Pathological analysis confirmed triplication of the gallbladder with severe, acute and chronic haemorrhagic cholecystitis (gallbladder A), cholesterosis with less inflammation (gallbladder B) and nearly denuded mucosa with an extensive haemorrhagic granulation tissue and haematoma (gallbladder C).

At follow-up, the patient has been asymptomatic.

DISCUSSION

Triple gallbladder (vesica fellae triplex)¹⁷ is a rare, congenital anomaly that occurs when rudimentary bile ducts fail to regress during embryological development and form accessory gallbladders.^{1 16} The first case was reported in a human cadaver in 1752.² Three types have been described, based on the anatomy of the cystic duct. Type 1 consists of three gallbladders draining into the CBD through separate cystic ducts, the variant

Figure 3 Intraoperative photograph of three gallbladders (labelled A, B and C).

Figure 4 Intraoperative photograph of three cystic ducts (white arrows) draining into the common bile duct (black arrow).

Figure 5 Three types of triple gallbladders. (A) Three cystic ducts combine to form a single cystic duct prior to joining the common bile duct (CBD). (B) Two cystic ducts form a single cystic duct and enter the CBD separate from the third cystic duct. (C) Three cystic ducts enter the CBD independently.

encountered in this patient. In type 2, two gallbladders connect to a common cystic duct and the third gallbladder drains into a separate duct. In type 3, three gallbladders share a common cystic duct (figure 5).¹⁷ The relationship between the gallbladders and cystic duct depends on the location of the embryological outpouchings. When these arise from the hepatic duct or CBD, they have separate cystic ducts, when they originate from the cystic duct, the gallbladders typically share a common cystic duct that drains into a morphologically normal CBD.^{9 10 15}

Multiple gallbladders are often associated with pathological conditions such as cholecystitis or carcinoma,¹⁸ hence a histological examination is essential. Preoperative diagnosis can be challenging. In the majority of cases, preoperative imaging fails

Learning points

- Congenital anomalies of the gallbladder can be classified based on size, location and number.
- Triple gallbladder occurs when rudimentary bile ducts fail to regress during embryological development and form accessory gallbladders.
- Multiple gallbladders are often associated with pathological conditions such as cholecystitis or carcinoma.
- The presence of multiple gallbladders is rare, but clinicians must be aware of this anomaly, as all gallbladders should be removed, either via open or laparoscopic surgery.

to identify all three gallbladders unless multiple modalities, including ultrasonography, endoscopic ultrasonography and MR ultrasonography are employed.¹¹ When any of the gallbladders is symptomatic, complete removal is recommended to avoid recurrent symptoms and the need for reoperation.^{9 19} Several approaches to removal of triple gallbladder have been described. Some authors favour open cholecystectomy because of the variable anatomy.²⁰ If a laparoscopic approach is undertaken, intraoperative cholangiography is recommended.^{12 13 5}

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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