



Case report

Gallbladder duplication complicated by cholecystitis and unsuspected choledocholithiasis: A case report and review of literature

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ABSTRACT

Introduction: Gallbladder duplication is a rare congenital anomaly of the biliary tree. Although a double gallbladder by itself is not clinically significant, complications of gallstone disease increases the complexity of the management. Preoperative recognition decreases the risk of complications during laparoscopic cholecystectomy.

Presentation of case: Presented herein is the case of a 52 year old who presented with abdominal pain. A trans-abdominal ultrasound was suggestive of a gallbladder duplication with the larger gallbladder filled with cholelithiasis. Subsequent imaging studies, including an endoluminal ultrasound, confirmed the diagnosis and documented a choledocholithiasis. Endoscopic extraction of the biliary stone with subsequent laparoscopic cholecystectomy of both gallbladders was successfully performed.

Discussion: This case is being presented not only for the rarity of the condition but also of the challenges in management it poses. In gallbladder duplication, pathologic involvement of one gallbladder requires removal of both gallbladders. A high index of suspicion on initial scanning warrants further delineation of the important anatomic structures of the biliary tree to avoid perioperative complications.

Conclusion: Laparoscopic cholecystectomy may be safely performed in patients with gallbladder duplication. Preoperative recognition with appropriate imaging modalities, including ultrasound and MRCP may avoid surgical complications. In cases where the anomaly is detected intraoperatively during cholecystectomy, meticulous dissection and intraoperative cholangiography will avoid iatrogenic injuries and lead to successful outcomes.

1. Introduction

Gallbladder duplication is a rare congenital anomaly which occurs in only one in 4000 births. This anomaly is clinically significant particularly in cases where they are not detected prior to gallbladder surgery [1,2,3]. Anomalies of the gallbladder and the biliary tree may lead to misidentification of structures which increases the risk of complications during cholecystectomy, both laparoscopic and open. In this era of laparoscopic cholecystectomy, unrecognized variations in anatomy predispose patients to iatrogenic bile duct injuries and other complications [4]. Although very few are reported in the published literature, these anomalies may be associated with complications like calculous cholecystitis and choledocholithiasis. Preoperative documentation of these malformations is very important to avoid catastrophic intraoperative problems. Judicious use of diagnostic imaging modalities may provide an accurate diagnosis which will avoid iatrogenic injuries and ensure successful operative outcomes [5–10]. In cases diagnosed

intraoperatively, maneuvers such as an intraoperative cholangiography may avoid bile duct injuries. However, preoperative recognition of these anomalies, while difficult, is still preferred to avoid intraoperative problems. Laparoscopic cholecystectomy is still the treatment of choice when gallstone diseases arise [11–21].

This case is being reported not only for the rarity of the condition but also for the complexity in management it poses. This report is being submitted in line with the SCARE 2020 criteria [22].

2. Presentation of case

A 52 year old female patient was admitted for severe epigastric pain characterized as continuous, dull and non-radiating. It was aggravated by left decubitus position and food intake. The patient recalls no previous episodes of similar abdominal pain but claims occasional vague discomfort with intake of fatty foods. She has no identifiable co-morbidities but has had three previous Cesarean sections. She has a two and a

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half pack year smoking history and is an occasional alcoholic beverage drinker. She has no maintenance medications and no allergies. Family and psychosocial history were likewise unremarkable.

On physical examination, her abdomen was soft but with tenderness at the epigastric and right upper quadrant area. There was no detectable jaundice. Liver function tests revealed ALT and AST at 859 μ /L and 195 μ /L, both elevated. Conjugated bilirubin was 21.38 mg/dL with unconjugated bilirubin at 1.38 mg/dL, both elevated. Alkaline phosphatase was likewise elevated at 337 μ /L. Lipase was normal. Protime and partial thromboplastin time were normal. Ultrasound showed a large gallbladder with multiple sub-centimeter stones with a slightly thickened wall and a smaller saccular structure beside it, suspicious of a gallbladder duplication. The intrahepatic and extrahepatic bile ducts were not dilated. The diagnosis of a double gallbladder was confirmed by an endoluminal ultrasound (EUS) which also detected a small, 0.4 cm common duct stone which was promptly removed by endoscopic retrograde cholangiopancreatography (ERCP) with balloon sweeping. The second gallbladder was not evident on the ERCP (Fig. 1). To prepare the patient for subsequent laparoscopic cholecystectomy and map out the biliary tree, a magnetic resonance cholangiopancreatography (MRCP) was requested. It clearly demonstrated the 2 gallbladders, one filled with stones and the other without filling defects (Figs. 2, 3, 4). Both gallbladders were drained by distinct cystic ducts separately draining into the bile duct. Diagnosis thus was Gallbladder duplication with cholecystitis and choledocholithiasis. The patient underwent laparoscopic cholecystectomy during the same admission. The procedure was performed by a senior attending surgeon, assisted by a fellow and a surgical resident. During the surgery, there were 2 gallbladders, the bigger one filled with stones and with a cystic duct inserting into the bile duct closer to the distal part of the common bile duct. The superiorly located 2nd gallbladder lay on top of the pathologic gallbladder and was grossly normal. The smaller cystic duct inserted into the bile duct one centimeter proximal to the other cystic duct. Both the cystic ducts were clearly identified, so an intraoperative cholangiography was deemed not necessary by the surgical team. The postoperative course was unremarkable and the patient was sent home recovered on the second postoperative day. Discharge instructions were provided for both interventions, the ERCP and the surgery. The macroscopic appearance is

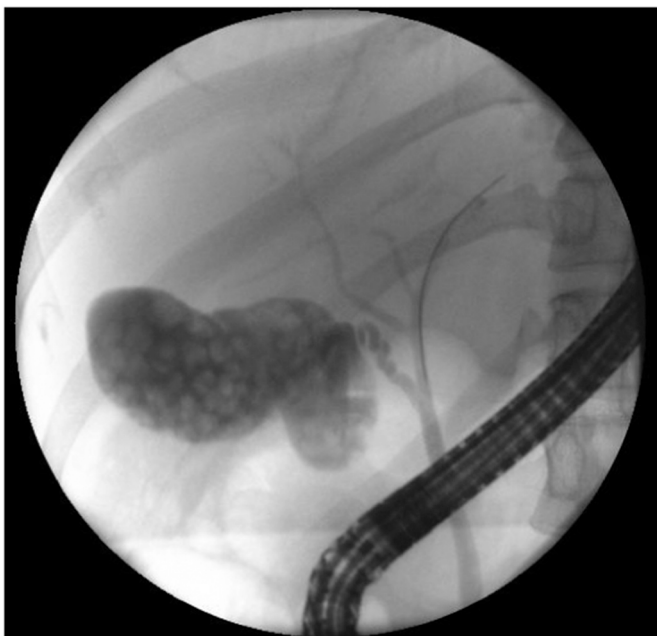


Fig. 1. Endoscopic retrograde cholangiopancreatography demonstrating one gallbladder with multiple filling defects. No visualization of the 2nd gallbladder. Film taken after duct stone extraction.

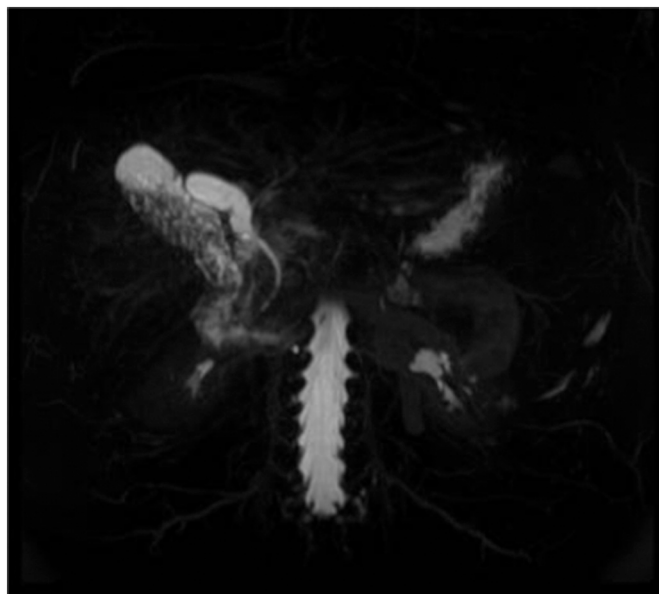


Fig. 2. MRI coronal view demonstrating 2 gallbladders, one with multiple stones. Both gallbladders are demonstrated to have separate cystic ducts.

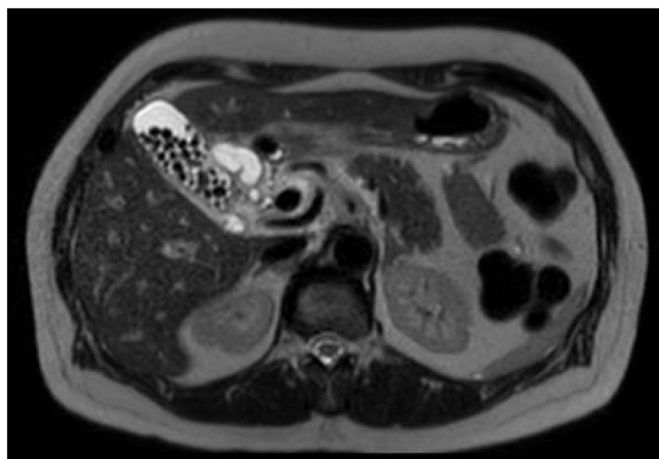


Fig. 3. MRI axial view demonstrating the 2 gallbladders.

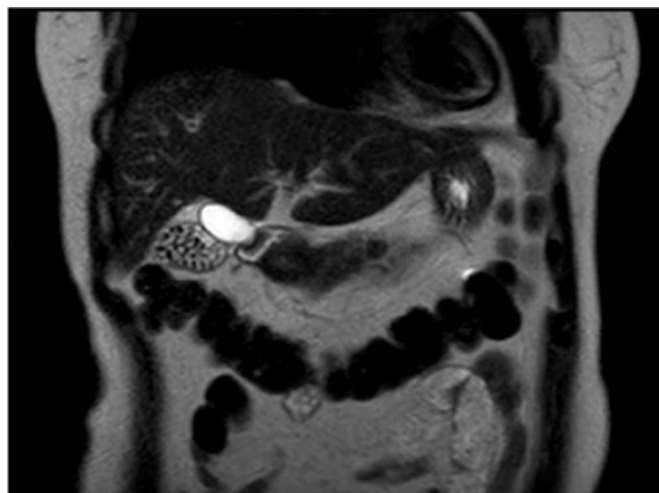


Fig. 4. MRI sagittal view demonstrating the 2 gallbladders.

consistent with a double gallbladder (Fig. 5). The pathology report revealed gallbladder duplication, with the lower gallbladder filled with stones. Microscopic evaluation revealed chronic cholecystitis. No malignancy was seen on histopathologic examination. The patient followed up 1 week and 1 month after discharge after the surgery with no post-operative complications.

3. Discussion

Gallbladder duplication is a rare congenital anomaly of the hepatobiliary tree, occurring in only about one in 4000 births. This may be an underestimation since the few cases that are diagnosed with certainty having this anomaly are only those who are symptomatic and those encountered incidentally during surgery, imaging studies or autopsy. Although gallbladder duplication by itself is not an indication for intervention, it is important to detect the condition when gallbladder diseases arise. Gallbladder duplications by themselves are clinically silent. Symptoms present when the condition is complicated by pathologies similar to those encountered in the single gallbladder. These include acute or chronic cholecystitis, cholelithiasis, empyema, torsion, biliary obstruction carcinoma and other gallbladder diseases. Any of these conditions affecting either of the double gallbladder warrants removal not only of the pathologic gallbladder but the other one as well. This is to avoid possible cholecystitis or symptomatic gallstones in the remaining organ. Reoperating for a subsequent pathology in the remaining gallbladder will technically be more difficult and may put the patient at risk for operative complications in the future [23]. Several diagnostic modalities that are used to evaluate the hepatobiliary tree may be used to detect gallbladder duplication and possible disease.

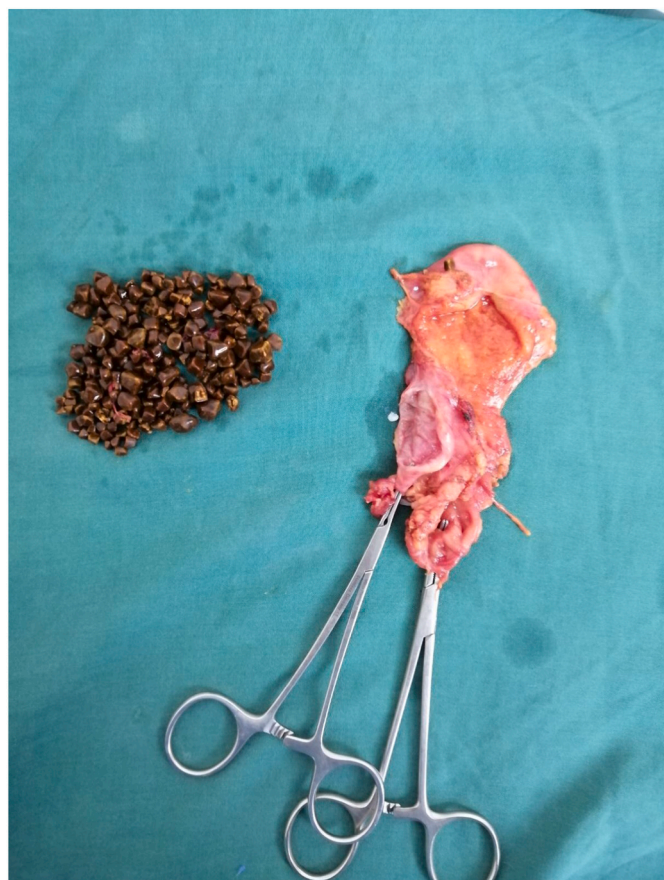


Fig. 5. Gross macroscopic appearance of the 2 gallbladders, with the instruments inserted into the independent cystic ducts. Stones were taken from the larger gallbladder.

These include ultrasound, computed tomography scanning (CT scan), magnetic resonance imaging (MRI), MRCP, ERCP and EUS. Ultrasound is most commonly used in patients suspected to have biliary disease. It thus most commonly detects a possible double gallbladder. Although ultrasound may suggest a double gallbladder, the cystic duct may be difficult to identify in most cases and thus difficult to distinguish from a bilobed gallbladder. For most cases, ultrasound will demonstrate the presence of 2 gallbladders. Although it is possible to demonstrate the draining cystic ducts, it may be more evident using the other modalities. MRI has proven to be a very useful imaging technique to evaluate the gallbladder and the biliary tree after an initial transabdominal ultrasound. Helical CT scan can also be helpful [23,24].

Duplication of the gallbladder has been detected by oral cholecystography, scintigraphy, and percutaneous transhepatic cholangiography but these examinations are not routinely used in patients with biliary disease [6–10]. In this case being presented, the initial transabdominal ultrasound raised the suspicion of a double gallbladder. The ultrasound findings revealed the presence of a second saccular structure superior to the stone-filled gallbladder. As narrated above, this was confirmed in the succeeding battery of tests requested. For an accurate surgical planning, gallbladder duplication must be classified into one of several types identified in Boyden's classification as shown in Fig. 6. Anatomic variants of gallbladder duplication are differentiated according to Boyden's classification as follows (1) Vesica fellea divisa (bilobed or bifid gallbladder, double gallbladder with a common neck), (2) Vesica fellea duplex (double gallbladder with two cystic ducts), (i) Y-shaped type (the two cystic ducts uniting before entering the common bile duct), (ii) H-shaped type (ductular type, the two cystic ducts entering separately into the biliary tree) [24]. Differential diagnosis includes gallbladder diverticula, gallbladder fold, Phrygian cap, choledochal cyst, pericholecystic fluid, focal adenomyomatosis, and intraperitoneal fibrous bands. In this case, the gallbladder consisted of 2 separate chambers, with adjacent walls. The chambers had distinct cystic ducts and which both drained into the common bile duct, classifying it as the H-type ductular type. In the course of the procedure they were noted to be supplied by separate cystic arteries. Double gallbladders, and those associated with other anomalies, present technical challenges to the surgeons which may cause perioperative difficulties and complications.

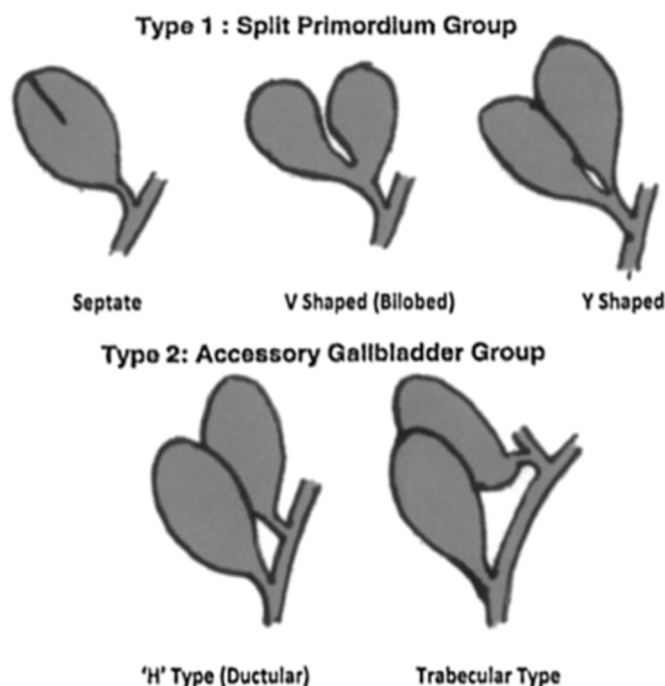


Fig. 6. Boyden's classification.

Performing biliary surgery on a patient with double gallbladder which is not detected preoperatively increases the risk of iatrogenic complications [4,25]. An accurate preoperative diagnosis will prepare the surgeon and may prevent misidentification of structures. It is very fortunate that the endoluminal ultrasound performed for this patient documented the presence of a choledocholithiasis, allowing endoscopic extraction. This further increases the rarity of this case, with only one published report documenting its occurrence in double gallbladders. A review of the studies published in English identified 13 laparoscopically-managed cases [11–20]. The majority of cases did not require conversion to an open cholecystectomy. For those cases successfully managed laparoscopically, an accurate interpretation of preoperative imaging data and meticulous dissection of the gallbladders are necessary prior to the ligation of tubular structures. In cases with the H-shaped subtype, the possibility of injury to the bile duct and hepatic artery is high [1,21]. In our case, the laparoscopic cholecystectomy was successfully completed safely without requiring intraoperative cholangiography. This supports the assertion that a carefully performed standard 4-port laparoscopic cholecystectomy technique may suffice.

The patient was discharged markedly improved. She verbalized her appreciation of the care she received from the surgical team.

4. Conclusion

Presented herein is a case of a double gallbladder complicated by calculous cholecystitis and choledocholithiasis managed by ERCP with stone extraction and laparoscopic cholecystectomy. A double gallbladder is a rare congenital anomaly that requires no active intervention in an asymptomatic patient. In cases complicated by gallstones or choledocholithiasis however, an unsuspected gallbladder duplication markedly increases the risk of iatrogenic injuries during surgery. While it is feasible to perform a laparoscopic cholecystectomy and removal of a double gallbladder, preoperative awareness of the condition through judiciously requested imaging studies will avoid intraoperative problems and ensure successful outcomes. In cases where the condition is diagnosed intraoperatively, a careful, meticulous dissection assisted by intraoperative cholangiography may prevent inadvertent injury to the biliary and vascular structures.

Ethical approval

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CRedit authorship contribution statement

Anthony R. Perez, MD: Study concept, writing the paper, documentation

Mary Ellen Perez, MD: Data collection, study design, manuscript editing

Michael de Jesus Magcase, MD: Data collection, review of literature, final draft.

Guarantor

Anthony R. Perez, MD, MHA

Provenance and peer review

Not commissioned, externally peer-reviewed.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Declaration of competing interest

There were no conflicts of interest.

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