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Successful laparoscopic management of duplicate gallbladder: A case report and review of literature

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ABSTRACT

INTRODUCTION: Gallbladder duplication is a rare congenital anomaly. Recognition of this anomaly and its various types is important since it can complicate a simple hepatobiliary surgical procedure.
PRESENTATION OF CASE: We report a case of a 42 year old female who presented a 6 year history of intermittent right upper quadrant abdominal pain. Her basic blood investigations including liver function tests were normal. Pre-operative imaging revealed a cystic lesion communicating with biliary tree representing duplicated gallbladder. She subsequently underwent successful laparoscopic cholecystectomy. The operative challenges were more than those anticipated at the usual laparoscopic gallbladder procedures. After six months follow up the patient remained asymptomatic.
DISCUSSION: Preoperative diagnosis plays a crucial role in planning surgery, and preventing possible biliary injuries or re-operation if accessory gallbladder has been overlooked during initial surgery. Magnetic resonance cholangiopancreatography (MRCP) is the imaging modality of choice for suspected duplicate gallbladder. Laparoscopic cholecystectomy for duplicate gallbladder is a challenging operation and should be performed with meticulous dissection of the cysto-hepatic triangle.
CONCLUSION: Gallbladder anomalies should be anticipated in the presence of a cystic lesion reported around the gallbladder. The laparoscopic cholecystectomy remains feasible for intervention and should be done by an experienced laparoscopic surgeon.

1. Introduction

Gallbladder duplication is a rare congenital anomaly. Anticipation and recognition of this anomaly and its various types are important to avoid surprises. Preoperative diagnosis plays a crucial role in planning surgery and preventing possible surgical complications or re-operation if accessory gallbladder has been overlooked during initial surgery. We present a case report in accordance with the case report (CARE) guidelines [35] of an unusual case of bilobed gallbladder managed successfully by laparoscopic cholecystectomy. Our review sought to determine the challenges in the diagnosis and management of this rare anomaly.

2. Case report

A 42 year old lady was presented to hepatopancreatobiliary surgery outpatient clinic with a six year history of intermittent right upper quadrant (RUQ) pain associated with occasional nausea and vomiting. She had no history of jaundice or fever. She had been on iron supplement and oral contraceptive pill for iron deficiency anemia secondary to menorrhagia. Physical examination revealed soft abdomen with no tenderness or palpable mass. Her blood investigations were normal including complete blood count, liver function test, bilirubin and tumor markers. Abdominal ultrasound (US) showed a multi-septated echoic cystic lesion in the right liver adjacent to segment V and gallbladder. Abdominal computed tomography (CT) showed a non-enhancing lobulated cystic lesion in segment V with extension reaching the gallbladder (Fig. 1). She was further investigated with magnetic resonance cholangiopancreatography (MRCP), which demonstrated a multilocular cystic lesion communicating with biliary tree most likely representing duplicated gallbladder (Fig. 2). Patient was admitted for elective laparoscopic cholecystectomy. Informed consent was obtained after explaining the surgical procedure and possible complications. During surgery, while dissecting the gallbladder from the liver bed, a thick fibrous structure was found adherent to gallbladder's posterior surface at the infundibulum. Careful dissection of this fibrous band revealed its communication with the intrahepatic cystic lesion that was anticipated and confirmed later to be the duplicate gallbladder.

The dissection of the intrahepatic gallbladder was challenging because of its close proximity to the right portal vein and middle

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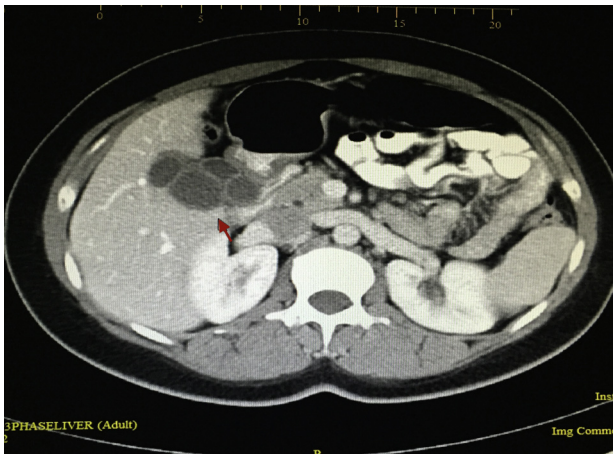

Fig. 1. A lobulated cystic lesion seen in segment V on CT scan.

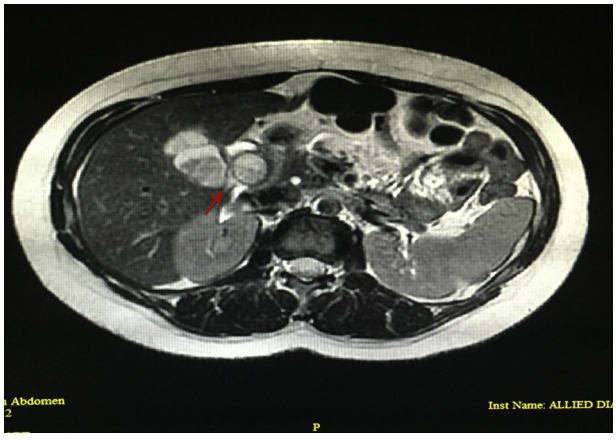

Fig. 2. Duplicate gallbladder on MRCP.

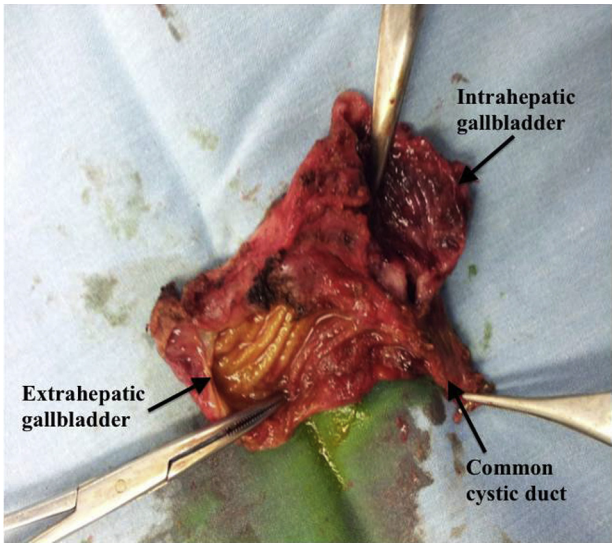

Fig. 3. Specimen of bilobed gallbladder.

hepatic vein, and extending partially into segment VIII. The dissection was continued carefully until the duplicate gallbladder was completely removed en bloc. On the back table the gallbladder was confirmed to be bilobed joining at the infundibulum with a single cystic duct. The intrahepatic gallbladder wall was thick with mucous content (Fig. 3).

In the recovery room, patient complained of mild chest tightness and shortness of breath. Air pulmonary embolism was suspected, which was resolved with oxygenation and changing patient's position to the left lateral decubitus. Patient was discharged home on postoperative day 2. Final histopathology revealed features of chronic cholecystitis. The adjacent pouch showed ulcerated epithelium with extensive hemorrhage in the wall, and proliferation of glands with gastric metaplasia. No evidence of dysplasia or malignancy was found. After six months follow up the patient remained asymptomatic.

3. Discussion

Gallbladder duplication is a rare congenital anomaly that occurs in 1 per 4000 individuals [1], occurring nearly twice in women than in men [2]. Duplication of gallbladder occurs during the 5th or early 6th embryonic week during which a single primordium bifurcates [1]. The time that bifurcation occurs determines the type of duplication that will occur i.e. the earlier the bifurcation; the more complete the degree of duplication [1]. A true accessory gallbladder arises from two separate primordia on the biliary tree and possesses a separate cystic duct. Histologically, gallbladder duplication is differentiated from a choledocal cyst by the presence of a muscular wall with an epithelial lining [3].

In 1929 Boyden reported 20 cases of double gallbladder he found in the literature from 1674 to 1929 [1]. He described a system to classify gallbladder duplications including "vesica fellea divisa" (bilobed gallbladder that has one cystic duct) and "vesica fellea duplex" (true gallbladder duplication). The latter is subclassified into "Y-shaped type" (two cystic ducts uniting before entering the common bile duct), and "H-shaped or ductular type" (two cystic ducts enter separately into the common bile duct).

In 1936, Gross described congenital abnormalities of gallbladder and classified them into six types labeled A–F [4]. In 1977, Harlaftis et al. further modified the classification by describing two main types based on morphology and embryogenesis [2] (Table 1). Although his classification is the most universally accepted, a modified Harlaftis classification has been reported in the literature by describing a left trabecular variant to type 2 classification [5]. Hassan et al. reported an accessory gallbladder branching from both the left and right hepatic ducts [6]. Causey et al. reported a new variant in which a septated type 1 gallbladder has 2 cystic ducts [7]. Our case represents Boyden type I, Gross type C, and Harlaftis type I septated gallbladder.

There are no specific symptoms or signs associated with duplicate gallbladders. Cholelithiasis, acute/chronic cholecystitis, empyema, fistula, torsion, papilloma and carcinoma that are seen in a single gallbladder can affect a duplicated gallbladder [8–13]. However, the most common complication is stone formation [14], and the risk is similar to that of a single gallbladder [3,15]. Surgery should be the treatment of choice only in symptomatic gallbladder duplication. It is recommended to remove both gallbladders at one stage to prevent subsequent disease in the remnant gallbladder at a later date.

Preoperative diagnosis of duplicate gallbladder is important because diagnosis of a second gallbladder may be overlooked during surgery. Factors that can lead to overlooking of the diagnosis include non-specific signs and symptoms, lack of awareness of the surgeon of the anatomic variations and inadequacy of the imaging techniques [16]. This may result in recurrence of symptoms or biliary complications. Although successful preoperative diagnosis is reported in only half of all cases [5], the imaging methods for viewing anatomic structures of the biliary tree and diagnosing the disease have progressed recently [17,18].

Table 1
Harlaftis classification of duplicate gallbladder [2].

Harlaftis classification of duplicate gallbladder
Typ1. The split primordium
• V-shaped (2 separate gallbladders at the fundus but join at the neck) • Y-shaped (2 separate gallbladder each with a cystic duct combine to form one cystic duct before entering the CBD) • Septate or bilobed (there is a single a septum that divides the two gallbladder).
Typ2. The accessory gallbladder
• Ductular type (2 gallbladders each with a cystic duct entering separately into the CBD) • Trabecular type (2 separate gallbladders, the superior cystic duct enters the right hepatic duct)

Abdominal US can be helpful for preoperative diagnosis. It can recognize duplicate gallbladder in the presence of two cystic structures occupying the gallbladder fossa [3]. However, US does not reliably delineate the anatomic detail of the cystic ducts neither their relations to the biliary tree [19,20]. Therefore, further investigations must be performed to determine the type of the anomaly. Abdominal CT cannot differentiate the gallbladder anomalies and its relation to the biliary tree in most of the cases. The differential diagnosis given by CT in our case was biliary cyst adenoma or hydatid cyst. A 3-dimensional intravenous infusion cholangiography- spiral CT (IVC-SCT) may be useful in the diagnosis of duplicate gallbladder [21].

Given the limitations of US and CT scan, MRCP can correctly identify the specific type of duplication. MRCP images and the 3D maximum intensity projection (MIP) images can further delineate the anatomy of the biliary tree [18]. MRCP has the advantage of being a non-invasive tool and a valid method for the evaluation of patients with suspected gallbladder anomalies after initial scanning with US [18]. Endoscopic retrograde cholangiopancreaticography (ERCP) can accurately delineate the biliary tract anatomy in gallbladder duplications [3]. It can be used as a helpful adjunct method but not as routine because of its invasive nature. Similarly intraoperative cholangiogram (IOC) can be used to define the biliary tract anatomy and help identify additional anomalous structures, especially if MRCP has not been carried out and an anomaly is encountered during laparoscopic cholecystectomy [22]. In our case, IOC was not performed for three reasons. First, diagnosis of duplicate gallbladder with normal appearing bile ducts from MRCP did not necessitate further intraoperative diagnostic evaluation. Second, no significant abnormal biliary anatomy was encountered intraoperatively prior to dividing the cystic duct. Third, dissection of gallbladder was carried out without any concern of biliary injury.

We reviewed 17 case reports summarized in Table 2. Our review demonstrated that the most common symptom of duplicate gallbladder is epigastric or RUQ abdominal pain. Ultrasound confirmed duplicate gallbladder in only 3 cases. While in other studies, presence of a cystic lesion in the gallbladder fossa on ultrasound raised the suspicion of duplicate gallbladder and necessitated further investigations to confirm the diagnosis. CT scan did not add to the abdominal US compared to MRCP, which was able to delineate the anatomy of the biliary tree and describe the gallbladder anomalies in the majority of cases. Almost all cases were managed with laparoscopic cholecystectomy. Few cases were converted to open for uncontrolled bleeding or were done open for missed duplicate gallbladder and/or bowel obstruction.

Although some authors advised an open surgical approach to prevent missed diagnosis, several authors have reported cases of duplicate gallbladder successfully treated by laparoscopic cholecystectomy [9,23–25]. In view of its advantages, laparoscopy has become the procedure of choice. It allows visualization of the hepatic hilum, gallbladder bed and local adjacent structures more easily and efficiently than open surgery [26]. Furthermore, laparoscopy is associated with less postoperative pain, shorter hospital stay and faster return to activities of daily living.

Although the frequency of complications associated with the laparoscopic approach for duplicate gallbladder has not been well studied, probably because of the small number of reported cases, but one would expect the risks to be slightly higher than the standard laparoscopic cholecystectomy. Unfortunately, the rarity of this condition, does not allow conducting randomized controlled studies to prove or disprove that. However, the available data showed no increased risk of biliary leak or gallbladder cancer. In addition, gallbladder anomalies are not associated with increased risk of other biliary anomalies. The risk of conversion rate might be slightly higher due to risk of bleeding associated with intrahepatic dissection.

4. Conclusion

Duplication of gallbladder is a rare congenital anomaly that requires special attention. Preoperative diagnosis can be challenging to the surgeon who should be aware of the anatomic variations of the gallbladder and biliary system. Presence of cystic lesions adjacent to the gallbladder on imaging should raise the suspicion of gallbladder anomaly. Further diagnostic preoperative imaging is important to avoid surprises, complications and overlooking of a second gallbladder. MRCP should be the imaging modality of choice for suspected duplicate gallbladder. Overall, we think the risks associated with laparoscopic cholecystectomy for duplicate gallbladders are comparable to those with non-duplicate gallbladder. However, these cases probably do better in the hand of an experienced laparoscopic surgeon or a hepatobiliary surgeon.

Conflict of interest

All authors certify that no actual or potential conflict of interest in relation to this article exists.

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Ethical approval

No Ethics Review was required for this case report.

Author contribution

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Analysis and interpretation of data: Aziza Al Rawahi, Yahya Al Azri, Salah Al Jabri, Abdulrazaq Al Fadhli, Suad Al Aghbari.

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Table 2
Summary review of different case report articles on duplicate gallbladder.

Authors	Type of duplication	Symptoms	US	CT	MRCP	ERCP	Procedure	Challenge/recommendation
Yorganci et al. [27]	Accessory GB	RUQ pain	Cystic lesion	Cystic mass	–	–	Lap chole	Lap is safe and correct choice for management
Weibel et al. [28]	Accessory GB	Pain post lap chole	Accessory GB	–	–	Accessory GB arising from RHD with stones	2nd lap chole	Conversion to open due to hemorrhage
Mazziotti et al. [18]	Ductular type	RUQ pain	2 cystic structures in GB fossa one containing stones	–	2 vesica with 2 separate cystic ducts, stones in one GB	–	Open chole	MRCP is recommended to detect anatomical variant
Goel et al. [29]	Accessory GB	RUQ pain, dyspepsia	Double GB Acute cholecystitis	–	Confirmed findings of US and ERCP	Double GB Normal bile duct and biliary tree	Lap chole	Detailed preoperative investigations are essentials before considering lap chole
Ozmen et al. [25]	Bilobed GB	RUQ pain Nausea	Bilobed GB	–	Confirmed US finding	–	–	Laparoscopy is safe and effective. IOC is recommended to avoid complications
Shirahane et al. [30]	Accessory GB	RUQ pain	2 cystic structures in GB fossa, one containing stones	–	–	2 GB each has cystic duct draining into CBD separately	Lap chole with ENB to identify biliary tree anatomy	Successful use of ENB in removing duplicated GB by laparoscopy
RolDan-Valadez et al. [31]	Vesica fellea duplex	Annual evaluation	2 cystic structures in GB fossa.	–	Y-shaped duplication of GB	–	–	MRCP is recommended
Hishinuma et al. [32]	Accessory GB	Epigastric pain	Cystic structure next to GB	Cystic structure between GB and liver	Could not delineate GB	GB filled with stones. Adjacent cystic structure accessory cystic duct entering RHD	Lap chole	ERCP confirmed the diagnosis
Sasaki et al. [24]	Accessory GB	Epigastric pain	Multi-lobulated cystic structure adjacent to GB	GB branching from CBD	–	Accessory GB draining into duodenum, communicating with dilated pancreatic duct	Lap chole	3D IVC-SCT+/– ERCP recommended for diagnosis
Vijayaraghavan and Belagavi [16]	Ductular type	RUQ pain	Separate GB with stones	–	–	–	Lap chole + IOC	IOC is recommended to delineate anatomy
Singh et al. [33]	Trabecular type	Jaundice 1 year post lap chole	GB with dilated intra- and extra-hepatic biliary tree	CT confirmed presence of remnant GB	–	–	Laparotomy	Missed duplicate GB in 1st lap chole. Second operation findings; GB draining into RHD. Adenocarcinoma of CBD
Desolneux et al. [34]	Y shaped GB	RUQ pain, fever, nausea and vomiting	Wall thickening of GB and gallstones	–	Bilobed GB	–	Lap chole + IOC	IOC is recommended
Brady and Mitchell [8]	Accessory GB ductular type	Cerebral palsy with abnormal laboratory tests	Complex mass adjacent to GB with irregular wall containing debris.	–	Complex mass displacing CBD. Intrahepatic and extrahepatic duct dilation. Mass communicating with duodenum	–	Emergency laparotomy for bowel obstruction. 2 GBs with 2 cystic ducts to CHD and RHD. IOC confirmed anomaly	Difficult preoperative diagnosis. IOC is recommended
Causey et al. [7]	Bilobed GB	RUQ pain	Cholethiasis, retrospectively a septum dividing the 2 GBs	–	–	–	Lap chole	Proposed an unified classification of multiple GB
Smelt et al. [36]	Double GB	RUQ pain	Two GBs, one contained gallstones	–	2 separate cystic ducts with stones in posterior GB	–	Lap chole	Awareness of anatomic variations is important
Bulus et al. [15]	Accessory GB	RUQ and epigastric pain	Gallstones Cystic mass behind GB	Duplicate GB	Confirmed duplication of GB but could not show cystic duct	–	Lap chole	Preoperative diagnosis is important
Hassan et al. [6]	Accessory GB	Generalized abdominal pain	Lesion on GB wall. No stones	Small neoplastic lesion in the GB	Accessory GB containing stone and connecting to both right and left HD. Main GB joins CHD via its own cystic duct	–	Not operated due to age and multiple medical comorbidities	MRCP is recommended to detect anatomical variant

Abbreviations: GB = Gallbladder, RUQ = Right upper quadrant, CHD = common hepatic duct, RHD = Right hepatic duct, Lap chole = Laparoscopic Cholecystectomy, ENT = Endoscopic Nasobiliary Tube.

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