



Duplicate Gallbladder: Overview and Management Challenges

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Abstract

Gallbladder (GB) anomalies are rare clinical diagnosis of biliary tract diseases. We present a case of a 42 years old female who presented with a 6 years history of intermittent right upper quadrant abdominal pain with occasional nausea and vomiting, diagnosed to have a duplicate gallbladder. Her basic blood investigations including liver function tests were within normal. Preoperative abdominal ultrasound (US) and computed tomography (CT) showed a non-enhancing lobulated cystic lesion in segment V with extension reaching the gallbladder. Thin sliced magnetic resonant cholangiopancreatography (MRCP) was done and showed multilobular cystic lesion communicating with the biliary tree most likely representing a duplicate gallbladder. The nature of the condition was explained to the patient and informed consent was obtained. She 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. In summary, gallbladder anomalies should be anticipated in the presence of cystic lesions reported around the gallbladder. The laparoscopic cholecystectomy remains the gold standard approach for intervention and these cases should be done by an experienced laparoscopic surgeon.

Keywords: Accessory gallbladder; Bilobed gallbladder; Gallbladder duplication; Laparoscopic cholecystectomy

Introduction

Gallbladder duplication is a rare congenital abnormality. More than 200 cases of duplicate gallbladder have been reported in the literatures [1,2]. Anticipation and recognition of this anomaly and its various types is important for the surgeons to be familiar with since it can complicate a simple hepatobiliary surgical procedure. Preoperative diagnosis is important to plan surgery and to further prevent possible surgical complications or re-operation if accessory gallbladder has been overlooked during initial operation. We report unusual case of a bilobed gallbladder, managed successfully by laparoscopic cholecystectomy. We reviewed current literature and proposed a diagnosis approach for surgeons to follow when evaluating patients presenting with right upper quadrant pain suspected to have gallbladder anomaly.

Case Presentation

A 42 year old lady was referred to Hepatobiliary Surgery outpatient clinic with six years history of intermittent right upper quadrant pain associated with occasional nausea and vomiting. She had no history of jaundice or fever. She has past medical history of Iron deficiency anemia secondary to menorrhagia, on iron supplement and oral contraceptive pill. Physical examination revealed soft abdomen, no tenderness or palpable mass. Her blood investigations were normal including complete blood count, liver function test, bilirubin and tumor markers. Abdominal US showed a multiseptated an echoic cystic lesion in the right liver adjacent to segment V and gallbladder. Abdominal CT also showed a non-enhancing tabulated cystic lesion in segment V with extension reaching the gallbladder (Figure 1). She was further investigated with MRCP (thin sliced) which revealed multilocular cystic lesion communicating with biliary tree most likely representing duplicated gallbladder (Figure 2). Patient was admitted for elective laparoscopic cholecystectomy. The nature of the condition and the complications of the surgery were explained to the patient. The anesthetist was alerted to maintain a low central venous pressure throughout the operation. During surgery, the liver appeared normal. Gallbladder was found to be covered by extensive omental adhesions. After release of those adhesions and partial exposure of the gallbladder, careful

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dissection further revealed significant thick fibrous adhesions and bands while dissecting Calot's triangle. The cystic duct was normal in diameter and slightly long, but was inserted in a normal anatomical position. The cystic artery was not identified probably thrombosed and cauterized along the way during dissection of the fibrous tissue. Strasburg critical view of safety was clearly identified before dividing the cystic duct. Combination of antegrade & retrograde approaches used to dissect the gallbladder from its bed. A thick fibrous structure was found adherent to its posterior surface at the infundibulum and going deep into the liver bed. Careful dissection of this fibrous band revealed it is communicating 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 it was thick walled surrounded by fibrous adhesions and deep into the liver with close proximity to the right portal vein seen on CT scan and extending partially into segment VIII. We stayed close to the perifibrous plane maintaining good retraction but removing part of the liver parenchyma surrounding the specimen. While separating the intrahepatic gallbladder from the wall of the right portal vein and its branches, bleeding occurred from a small branch that was managed by temporary pressure gauze and lowering intra abdominal pressure. Also there was minimal arterial bleed observed from different areas in the liver parenchyma that was also managed with pressure and careful cauterization. The dissection continued until the duplicate gallbladder was completely removed en bloc. After retrieval, the gallbladder was examined on the back table and confirmed to be a bilobed gallbladder joining at the infundibulum forming a single cystic duct, the intrahepatic gallbladder wall was thick with mucous content. (Figure 3). Postoperatively, while the patient in the recovery room, she was noted to be complaining of moderate central chest discomfort with normal oxygenation when lying supine. The discomfort was almost completely relieved few minutes after turning the patient on to her left side and aggravated by lying supine again. Pulmonary air embolism was suspected and this was probably caused when the bleeding from the wall of the right portal vein has happened. Intravenous fluid rate maintained, high flow F102 given and she was maintained on left decubitus position for 3-4 hours till the chest discomfort has completely improved. Central line insertion was considered but not attempted for suspected gas bubble aspiration as the patient showed progressive improvement. She was discharged home on post operative day 2. Microscopic examination of primary gallbladder 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.

Discussion

Gallbladder duplication is a rare congenital anomaly that is reported to occur in 1 per 4000 individuals, occurring nearly twice in women than in men [1]. Duplication of gallbladder occurs during the 5th or early 6th embryonic week during which a single primordium bifurcates. 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'. 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. In 1929 Boyden reported 20 cases of double gallbladder he found in the literature from 1674 to 1929 [3-8]. He described a system to classify gallbladder duplications including

"vesica fellea divisa" (billed 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-F6.

In 1977, Harlaftis et al. [1] further modified the classification by describing two main types based on morphology and embryogenesis' (Table 1). His classification is the most universally accepted

A recently published data have described a modified Harlaftis classification that added a left trabecular variant to type 2 classification'. Hassan et al. [1] reported an accessory gallbladder branching from both the left and right hepatic ducts. Causey et al. [9] reported a new variant in which a septated type 1 gallbladder has 2 cystic ducts. 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. The same spectrum of disease seen in a single gallbladder can affect a duplicated gallbladder including cholelithiasis, acute/chronic cholecystitis, empyema, fistula, torsion, papilloma and carcinoma as described in case reports [2,5,10-15]. Among these, the most common complication is stone formations. However, the risk of stone formation in the duplicate gallbladder is the same as with a single gallbladders [16]. Therefore there is no indication to remove a duplicated gallbladder if found incidentally. Surgery should be the treatment of choice only in symptomatic gallbladder duplication. It is recommended to remove both gallbladders at one setting to prevent subsequent disease in the remnant gallbladder at a later date.

Preoperative diagnosis of duplicate gallbladder is very important because diagnosis of a second gallbladder may be overlooked during the surgery. Factors that can lead to overlooking of the diagnosis include lack of specific signs and symptoms, lack of awareness of the surgeon of the anatomic variations and inadequacy of imaging techniques [17,18]. This may result in recurrence of symptoms or biliary complications. Therefore, preoperative imaging is very important in diagnosing duplicate gallbladder. Although successful preoperative diagnosis is reported in only half of all cases', the imaging methods for viewing anatomic structures of the biliary tree and diagnosing the disease have progressed recently [19].

Abdominal US can be helpful for preoperative diagnosis of gallbladder disease. It can recognize duplicate gallbladder in the presence of two cystic structures occupying the gallbladder fossa[5]. However, US does not reliably delineate the anatomic detail of the cystic ducts neither their relations to the biliary tree [20,21]. Therefore, further investigations must be performed to determine the type of the anomaly. From the 17 case reports we reviewed, there were 3 cases in which duplicate gallbladders could be recognized by ultrasound. The rest reported a cystic lesion seen in the gallbladder fossa which raised the suspicion of duplicate gallbladder and required further investigations to confirm the diagnosis.

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. However, recently 3-dimensional intravenous infusion cholangiography- spiral CT (IVC-SCT) was reported to be useful in the diagnosis of duplicate gallbladder [22].

Given the limitations of US and CT, MRCP can correctly identify the specific type of duplication as it has better diagnostic capability than US [19]. MRCP images and the 3D maximum intensity projection (MIP) images can further delineate the anatomy of the biliary tree. MRCP has the advantage of being a non-invasive tool and proved to be a valid method for the evaluation of patients with suspected gallbladder anomalies after initial scanning with US [19].

Endoscopic retrograde cholangiopancreatography (ERCP) can accurately delineate the biliary tract anatomy in gallbladder duplications [5]. It can be used as 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 to help identify additional anomalous structures, especially if MRCP has not been carried out and an anomaly is encountered during laparoscopic cholecystectomy [23]. This is highly recommended by several authors to avoid inadvertent damage to the biliary ductal system and overlooking of second gallbladder [23].

In our case, IOC was not performed for three reasons. First, diagnosis of duplicate gallbladder and normal appearing bile ducts on 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.

Although some authors advise an open surgical approach to prevent missed diagnosis, several authors have reported cases of duplicate gallbladder successfully treated by laparoscopic cholecystectomy [10,24-26]. 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 [27]. Furthermore, laparoscopy is associated with less post operative pain, shorter hospital stay and faster return to activities of daily living. Shirahane reported utilizing Endoscopic Nasobiliary Tube (ENB) in removing duplicated gallbladder by laparoscopy successfully [28].

Comparison between different articles written on individual cases of duplicate gallbladder (Table 2) revealed that, the most common symptom is the epigastric or right upper quadrant abdominal pain. Abdominal US was able to demonstrate the presence of a separate cystic lesions near the gallbladder with or without stones and whether it is complicated with acute cholecystitis or intra or extrahepatic biliary dilatation. However, the CT scan did not add much 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 apart from few cases were converted for uncontrolled bleeding or done open for missed duplicate gallbladder and bowel obstruction.

Laparoscopic cholecystectomy for duplicate gallbladder is a challenging operation. Although the frequency of complications has not been well studied probably because of the small 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. The risk of conversion rate might be slightly higher due to risk of bleeding

associated with intrahepatic dissection. In addition, gallbladder anomalies are not associated with increased risk of other biliary anomalies.

We propose the following approach for evaluating patients presenting with upper abdominal pain and suspected to have duplicate gallbladder anomalies (Figure 4).

Conclusion

Duplication of gallbladder is a rare congenital anomaly that requires special attention. Preoperative diagnosis can be challenging to the laparoscopic surgeon who should be aware of the anatomic variations of the gallbladder & 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, and to plan surgery. Abdominal CT exposes patients to radiation and might not be able to provide detailed anatomy of the gallbladder anomalies compared to MRCP which 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.

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