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A case report of a patient with gallbladder agenesis resulting in a common bile duct injury

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ABSTRACT

INTRODUCTION: Congenital agenesis of the gallbladder is a rare embryological defect of the biliary system. While occurring equally in men and women, gallbladder agenesis is found clinically twice as often in women. Patients present with symptoms suggesting biliary colic. Abdominal ultrasound and cholecintigraphy or HIDA scan are usually inconclusive and, in some cases, may be read as positive for biliary colic. Patients can undergo surgery based on characteristics of pain.
PRESENTATION OF CASE: We report the case of a 60-year-old female presenting with symptoms of recurrent biliary colic and subsequently undergoing laparoscopic cholecystectomy after inconclusive workup.
DISCUSSION: We offer a review of past reported cases as well as a new approach to such patients during the intraoperative period. An intraoperative decision should be made whether to continue and search for a possible ectopic gallbladder or investigate further with imaging studies.
CONCLUSION: Gallbladder agenesis is a rare clinical presentation that the surgeon must be aware of. With inconclusive studies, the surgeon should consider congenital absence of the gallbladder and pursue further imaging if the gallbladder cannot be localized during the intraoperative period.

1. Introduction

Congenital agenesis of the gallbladder is a rare embryological defect of the biliary system. Its prevalence has been estimated to be about 9 per 10,000 [1]. The gallbladder develops from the hepatic diverticulum and hepatic primordium [2]. It begins as a hollow bud from the duodenum and grows into the septum transversum [2,3]. The bud divides, one part forming the gallbladder and cystic duct, and the other forming the glandular substance of the liver [2,3]. It is thought that either failure of this division or the failure of the solid bud to become hollow results in an absent gallbladder [2]. Congenital agenesis occurs equally in men and women, but it is found clinically twice as often in women [1]. Patients can present with symptoms of biliary colic. This work has been reported in line with the SCARE criteria [4].

2. Case description

A 60-year-old female with a BMI of 26.5 presented with a complaint of daily post-prandial epigastric pain and nausea lasting three to four hours for the prior six months. There were no alarm symptoms such as fevers or unintentional weight loss, and physical exam was normal. After an ultrasound demonstrated the presence of a gallstone, routine labs were ordered including complete blood count, hepatic function tests, lipase as well as a HIDA scan. All labs, including the direct and indirect bilirubin were normal. On HIDA the gallbladder was not visualized at 90 min (Fig. 1). A subsequent ultrasound demonstrated a wall echo shadow complex in the gallbladder fossa indicating a gallbladder filled with stones. A diagnosis of symptomatic cholelithiasis was made, and a laparoscopic cholecystectomy was recommended.

During the operation, a laparoscope was inserted, and the peritoneal cavity was explored. The gallbladder was not visualized on initial exploration. The gallbladder fossa appeared empty. An exhaustive dissection including skeletonizing the hepatoduodenal ligament failed to locate the organ and resulted in a CBD injury that required open exploration and roux-en-y hepaticojejunostomy reconstruction. No vestige of gallbladder or cystic duct was ever encountered. The patient did well and is asymptomatic with normal liver function one-year post-op.

3. Discussion

Although the clinical prevalence of gallbladder agenesis is 0.09%, the overall incidence is higher when taking into account autopsy reports [1]. The diagnosis of congenital absence of the gallbladder is rarely made preoperatively, except in rare familial cases when it might be suspected [6]. Although most patients are never diagnosed, a subset of patients, approximately 33–50%, demonstrate symptoms of biliary colic and consequently undergo an operation.

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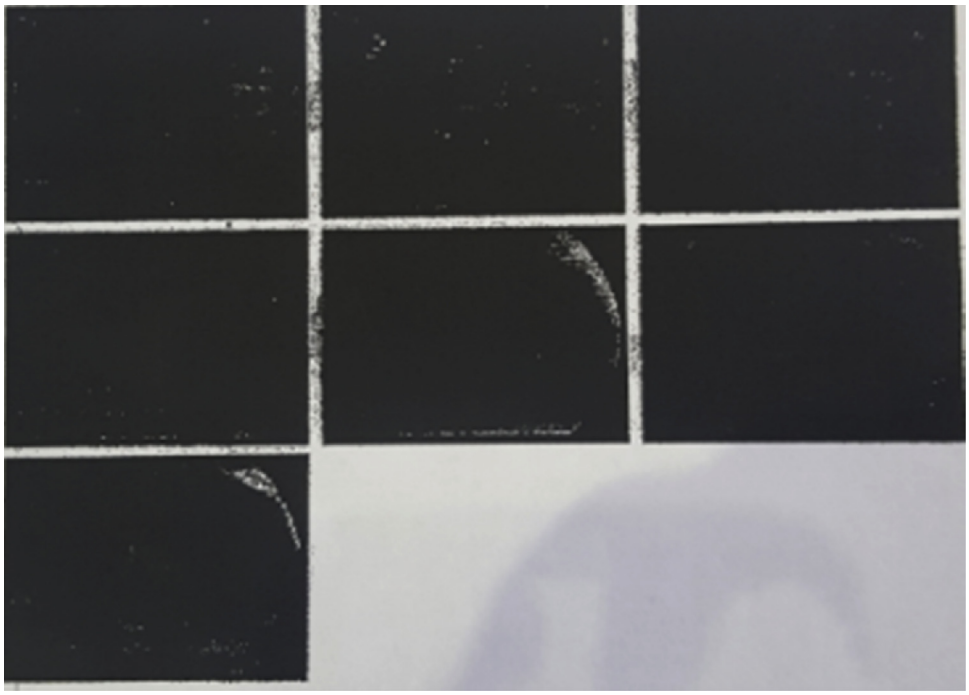

Fig. 1. HIDA scan showing uptake of the isotope by the common bile duct at 7 min and activity within the small bowel by 12 min. At 90 min, the counts in the liver were nearly exhausted, and no gallbladder activity was demonstrated.

In a study of 60 cases of congenital absence of the gallbladder reported by Dixon and Lichtman, 58% of the patients were symptomatic for cholecystic disease [5].

Otherwise known as symptomatic agenesis, this comprises a third to half of the patients with gallbladder agenesis [6,7]. Half of these symptomatic patients will possess a dilated common bile duct and/or stones in their common bile duct [7]. Other common symptoms include chronic right upper quadrant pain (90%), dyspepsia (30%), nausea and vomiting (66%), fatty food intolerance (37%), and jaundice (35%) [7]. In such cases, studies have demonstrated the use of ultrasound is limited [6,7].

Current recommendations for diagnostic imaging of the diseased gallbladder include MRCP, ERCP, and HIDA scan. While ERCP is the gold-standard for visualizing the biliary anatomy, it has complication rates up to 11.0%. Complications including pancreatitis, hemorrhage, and perforation have been reported. HIDA scanning, while not the primary means to diagnose biliary colic, is utilized when gallstones are not visualized on ultrasound. With a sensitivity of 90–97% and specificity of 70–91% for acute cholecystitis, it is often used in the diagnostic workup to facilitate a surgical decision [8]. However, in cases of gallbladder agenesis, failure to visualize the gallbladder on HIDA scan often leads to a false conclusion that the gallbladder is diseased [7]. These results ultimately lead to surgery.

In the past, congenital absence of the gallbladder is confirmed intraoperatively by dissecting the extrahepatic biliary tree [9]. In 1995 Peloponissios et al., recommended searching for an ectopic location of the gallbladder intraoperatively in such cases. Areas of interest include the retrohepatic space, on the left side, or within the lesser omentum or falciform ligament [10]. If maneuvers fail to localize the gallbladder, then peri-operative cholangiography is warranted to rule out an intrahepatic gallbladder. It is also useful to confirm the location of the common bile duct as well as look for stones that may be present which can cause symptoms that mimic biliary colic.

More recent studies suggest the use of endoscopic ultrasonography as a tool in the diagnosis of extrahepatic diseases [11]. Intraoperative exploration exposes the patient to an increased risk of poor outcomes such as injury to nearby structures like the small intestine or common bile duct [10]. Therefore, when faced with the decision to convert from laparoscopic operation to an open exploration to localize the gallbladder, it is now recommended to abort the operation and investigate with computed tomography and/ or magnetic resonance cholangiopancreatography (MRCP) [6]. These studies can also be utilized when the index of suspicion for gallbladder agenesis is high in order to avoid surgery [6]. Current consensus suggests MRCP is the test of choice to investigate whether the gallbladder is present or if it is in an ectopic position [1].

We suggest that during the intraoperative period, if the gallbladder cannot be visualized in an adequate time, and a dilated structure is present, to terminate the procedure and further investigate the location of the gallbladder using MRCP as shown in Fig. 2. Malde has suggested a workup of patients who present with symptoms suggestive of biliary disease with utilization of ultrasound followed by MRCP, CT, ERCP or endoscopic ultrasound if the gallbladder is not identified [12]. Once gallbladder agenesis is confirmed, current treatment recommendations include conservative management with smooth muscle relaxants. If conservative treatment fails, a sphincterotomy may be warranted [12].

Unfortunately, even radiological studies may lead to inadequate findings unable to confirm gallbladder agenesis [1]. A recent surge in laparoscopic cholecystectomy, largely attributed to obesity in the youth, may lead to more patients with gallbladder agenesis presenting with symptoms of biliary colic. While exceedingly rare, biliary surgeons need to bear in mind this possibility to avoid the type of complications illustrated by this case.

4. Conclusion

Gallbladder agenesis, is a rare clinical entity that may lead to symptoms and imaging resulting in the incorrect diagnosis of biliary colic or cholecystitis. Surgeons should be aware of gallbladder agenesis when imaging studies are inconclusive. In such patients, the gallbladder may not be visualized at the time of surgery.

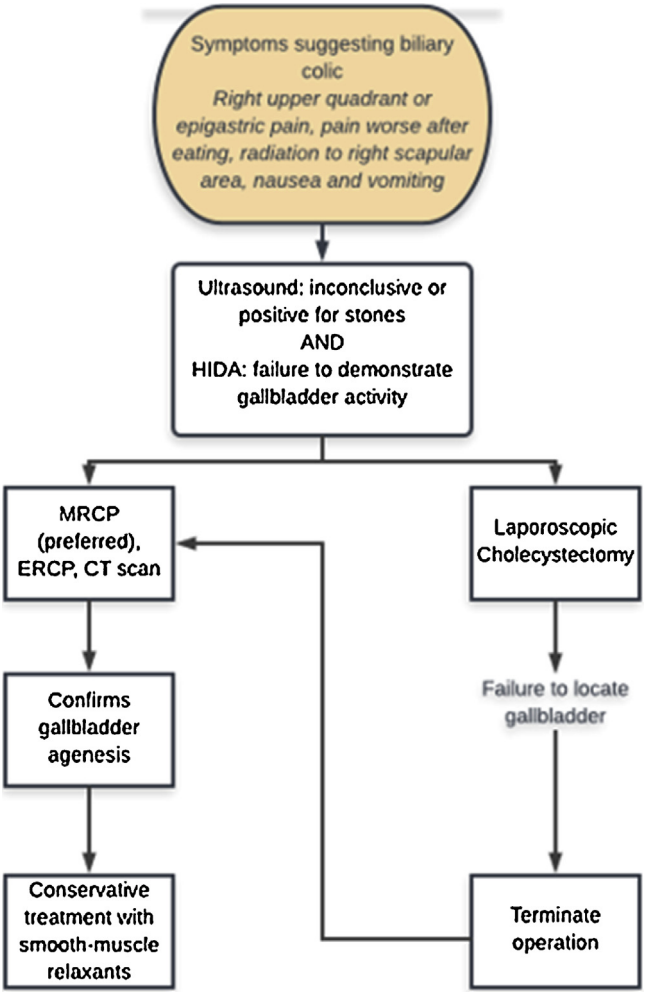

Fig. 2. HIDA: Hepatobiliary scintigraphy; MRCP: Magnetic resonance cholangiopancreatography; ERCP: Endoscopic retrograde cholangiopancreatography; CT: Computed tomography.

Rather than continue surgical exploration, the procedure should be aborted, and the patient should undergo further diagnostic testing. However, when there is a high clinical suspicion pre-operatively, diagnostic studies such as MRCP must be considered.

Conflicts of interest

Nothing to declare.

Funding

Nothing to declare.

Ethical approval

Ethical approval has been exempted by our institution, Medical College of Georgia, for this study.

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.

Authors contribution

Anoosh Bahraini- writing the paper.
Dr. John Odom- data analysis and interpretation.
Dr. Asif Talukder- data analysis and editing of the manuscript.

Registration of research studies

Nothing to declare.

Guarantor

Anoosh Bahraini.

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