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Case and Review

Two Anomalies in One: A Rare Case of an Intrahepatic Gallbladder with a Cholecystogastric Fistula

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Abstract
The gallbladder can be situated in a variety of anomalous positions. An intrahepatic gallbladder – the second most common ectopic location of the gallbladder – is one that is completely embedded within the liver parenchyma. Described in the literature as early as 1935, intrahepatic gallbladders predominantly result from a developmental anomaly but in some instances have been reported to be secondary to chronic inflammation. The significance of an intrahepatic gallbladder lies in the fact that 60% of the cases are associated with gallstones and may present a challenge for the general surgeon during cholecystectomy and other biliary operations in addition to causing misdiagnosis on imaging. Intrahepatic gallbladders are unusual, but the incidence of an intrahepatic gallbladder with a cholecystogastric fistula is rare. Cholecystogastric fistulas commonly are a complication of long-term cholelithiasis or chronic cholecystitis with subsequent gallstone ileus. Herein, we present the case of an 80-year-old man who presented with 2 months of progressive weakness, fatigue, decreased appetite, and intermittent right-sided abdominal pain, and was found to have a markedly distended and irregular intrahepatic gallbladder measuring 12.2 × 11.5 × 13.4 cm on CT, as well as a cholecystogastric fistula on esophagogastroduodenoscopy. During esophagogastroduodenoscopy, the gallbladder was entered directly via the fistulous tract. The patient was on i.v. antibiotics with tube feeds via a nasojejunal tube initially, followed by p.o. which he tolerated. He was eventually discharged with referral for surgical evaluation. Given the poten-

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tial for cholelithiasis and fistulation, physicians should have a high index of suspicion and recommend timely endoscopic and/or surgical management to avoid future complications.

Introduction

An intrahepatic gallbladder is one that is partially or completely embedded within the liver parenchyma [1, Couinaud C, cited in 2]. A developmental anomaly or failure of the gallbladder to move from its intrahepatic position to its normal superficial location during the second month of gestation may result in this ectopic location [3]. The condition should be suspected if a cholecystogram or ultrasonography shows a gallbladder in an unusually high location. Cholelithiasis rates are as high as 60% in patients with intrahepatic gallbladders [4] and may present a challenge for the general surgeon during cholecystectomy and other biliary operations in addition to causing misdiagnosis on imaging.

Cholecystogastric fistulas, or abnormal connections between the gallbladder and stomach, commonly are a complication of long-term cholelithiasis or chronic cholecystitis with subsequent gallstone ileus. The fistulous tract forms from the gradual erosion of the chronically inflamed and closely adherent wall of the gall bladder and stomach.

We present the case of an 80-year-old man who presented with 2 months of progressive weakness, fatigue, decreased appetite, and intermittent right-sided abdominal pain, with laboratory tests significant for leukocytosis, hyperbilirubinemia, and elevated alkaline phosphatase. He was found to have a markedly distended and irregular intrahepatic gallbladder with a small speck of gas in the fundus suggestive of fistulization to the bowel on CT. A cholecystogastric fistula was confirmed on esophagogastroduodenoscopy. The gallbladder that was in communication with the stomach was entered directly via the fistulous tract during the procedure.

The incidence of an intrahepatic gallbladder with a cholecystogastric fistula, as in our case, is rare. Since cholelithiasis rates are high in this patient population, they may be prone to fistulation between the gallbladder and the duodenum and/or stomach. Hence, physicians should have a high index of suspicion and recommend timely endoscopic and/or surgical intervention to avoid future complications.

Case Presentation

An 80-year-old man presented with 2 months of progressive weakness, fatigue, and decreased appetite. He also described a sharp, intermittent, nonradiating right-sided abdominal pain that had developed. He denied nausea, vomiting, fevers, chills, diarrhea, chest pain, shortness of breath, weight loss, acholic stools, or dark urine. Initial labs were significant for leukocytosis (19.0 K/μL), elevated total bilirubin (2.3 mg/dL), direct bilirubin (1.3 mg/dL), and alkaline phosphatase (168 IU/L).

CT of the abdomen and pelvis with i.v. contrast revealed a markedly distended and irregular gallbladder measuring 12.2 × 11.5 × 13.4 cm, closely abutting and causing mass effect upon the posterior inferior right hepatic lobe and inferior aspect of the medial left hepatic lobe with adjacent inflammation. The wall of the gallbladder appeared heterogeneous and thickened with cystic trabeculation. It was difficult to distinguish where the gallbladder wall terminated and the hepatic parenchyma began, i.e., intrahepatic gallbladder (Fig. 1).

There was trace intrahepatic biliary ductal dilatation. There was extension into the right lateral wall of the distal gastric antrum/proximal duodenum measuring 4.4 × 3 × 4.3 cm in anterior to posterior, transverse, and cranial caudal dimensions. No radiopaque gallstones were seen. Magnetic resonance cholangiopancreatography confirmed a large hypo-enhancing gallbladder with multiple enhancing septations and thickened irregular wall, with invasion into the adjacent hepatic parenchyma as well as possible fistulization into the first portion of the duodenum/pylorus (Fig. 2).

Esophagogastroduodenoscopy demonstrated a cholecystogastric fistula through which the gallbladder was directly entered (Fig. 3). A white exudate on the gallbladder wall and moderate purulence were noticed (Fig. 4). There was no obvious necrosis or extensive debris. Cytology of gall bladder aspirate fluid showed acute inflammatory exudate, macrophages, and proteinaceous debris. Pathology of gallbladder wall biopsy exhibited fragments of necrotic tissue with acute inflammation.

The patient was on i.v. antibiotics with tube feeds via a nasojejunal tube initially, followed by p.o. which he tolerated. He was eventually discharged with referral for surgical evaluation.

Discussion

The gallbladder is typically located under the right lobe of the liver, in the plane of the interlobar fissure [5]. Embryologically, this organ arises from the caudal bud which itself originates from the hepatic diverticulum of the primitive midgut during the organogenesis of the liver, biliary tract, and ventral pancreas around the 4th week of gestation [6]. The 4 most common ectopic locations of the gallbladder include under the left liver lobe, intrahepatic, transverse, and retroplaced. The incidence of an ectopic location of the gallbladder is reported to be 0.1–0.7% [7]. Malpositioning of the gallbladder is of great clinical significance as it can confound presenting signs and symptoms of infection and/or inflammation and create technical problems during cholecystectomy and other biliary operations, in addition to causing misdiagnosis on imaging.

An intrahepatic gallbladder can be partially or completely surrounded by the liver parenchyma and is a product of a developmental glitch or failure of appropriate translocation during gestation. It poses a diagnostic challenge since scintigraphy findings can be interpreted as being consistent with a mass. Ultrasonography and angiography (with identification of the cystic artery) helps with identification [8]. Cholelithiasis rates are as high as 60% in patients with intrahepatic gallbladders, likely due to stasis. Our patient did not have any history of gallstone disease but had signs of inflammation of the gall bladder, e.g., thickened wall, on imaging.

Cholecystogastric fistulas commonly are a complication of long-term cholelithiasis or chronic cholecystitis with subsequent gallstone ileus. The fistulous tract forms from the gradual erosion of the chronically inflamed and closely adherent wall of the gall bladder and stomach. Cholecystogastric fistulas have been reported as far back as 1968 [9] and though once associated with high mortality, the advent of improved radiologic, endoscopic, and surgical modalities has led to successful management. CT appearances of cholecystogastric fistulas consist of a slight deficiency of the gall bladder wall [10] and attachment of the edematous wall of the gastric antrum and the thickened wall of the gall bladder fundus [11]. A small speck of gas in the fundus of the gallbladder was noted on our patient's CT suggestive of fistulization to the bowel.

Endoscopic treatment of cholecystogastric fistulas is safe and practical. However, the presence of large gallstones (>3 cm), gastrointestinal tract hemorrhage, stone impaction, and improper or partial stone manipulation are often mitigating factors for endoscopic failure necessitating surgical intervention [12]. Therapeutic interventions include 1-stage or 2-stage stone removal, fistula repair and cholecystectomy, laparoscopic cholecystoenteric fistula transection, laparoscopic intraperitoneal suturing of the fistulous tract, endoscopic treatment, or only stone removal without fistula repair and cholecystectomy [13]. Our patient was referred for surgical evaluation.

The incidence of an intrahepatic gallbladder with a cholecystogastric fistula, as in our case, is rare. Since cholelithiasis rates are high in this patient population, they may be prone to fistulation between the gallbladder and the duodenum and/or stomach. Surgical and/or endoscopic management should be recommended by physicians to avoid future complications.

Statement of Ethics

The authors have adhered to the ethical guidelines for authorship and publishing stated in the journal *Case Reports in Gastroenterology*. Consent was obtained from the patient for publication of this case report.

Disclosure Statement

The authors have no conflicts of interest to declare.

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Fig. 1. Coronal CT image showing a markedly distended and irregularly shaped intrahepatic gallbladder (red arrow) with surrounding edema.

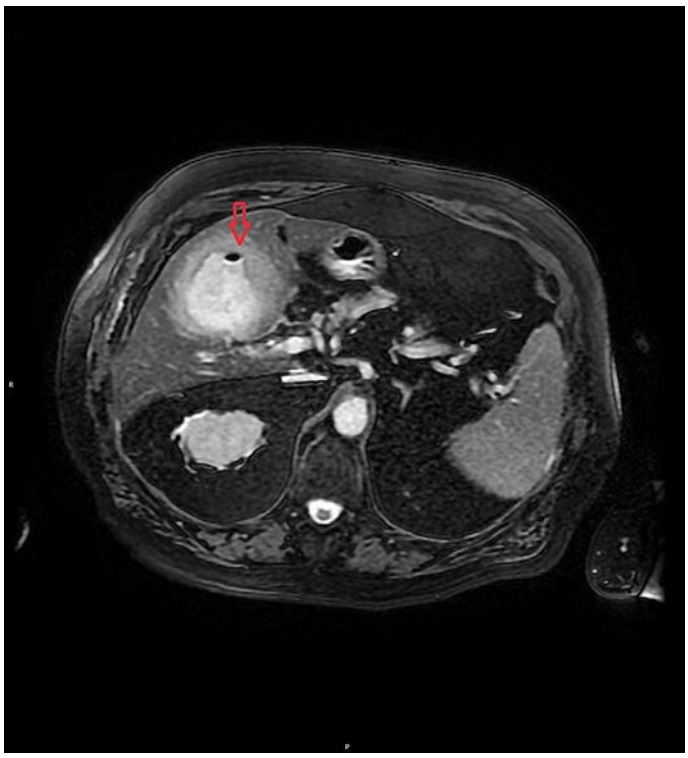

Fig. 2. MR image (T2-weighted, axial) demonstrating marked thickening of the gallbladder wall and debris in the gallbladder (red arrow).

Fig. 3. Opening of the cholecystogastric fistula with purulence (red arrow); gastric antrum in the background.

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Fig. 4. Cholecystogastric fistula (left) and lumen of the gallbladder with exudative covering of wall (right).