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Case Report: Ectopic Liver (Choristoma) Discovered During Laparoscopic Cholecystectomy

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Patient: Male, 47-year-old **Final Diagnosis: Ectopic liver**

> **Symptoms:** Right upper quadrant pain

Medication: Clinical Procedure:

Specialty: Surgery

Objective: Congenital defects/diseases

Background: Ectopic liver (EL) is liver tissue that is not attached to the mother liver, and is also known as choristoma and hepar succenturiatum. According to Watanabe's 1060-patient series, it has an incidence rate of less than 1% (0.47%). The absence of an anatomical attachment to the hepatic proper is a rare congenital departure from the norm, known as EL. EL is thought to be caused by abnormal hepatic tissue growth from the foregut diver-

ticulum, particularly at cystic structures.

Case Report: We report the successful management of a 47-year-old male patient, not known to have any medical illness,

who was referred to the surgical team with a clinical picture of biliary colic. An ultrasound (US) abdomen was performed, and cholecystitis was confirmed. During a laparoscopic cholecystectomy, ET was identified by chance. He tolerated the operation well. Gallbladder histological examination revealed significant mucosal and wall ulcers, chronic inflammation, many black stones, no dysplasia or malignancy, and connected liver tissue measuring 12×5 mm. As a result, chronic calcular cholecystitis was confirmed, as was normal EL architecture and no

evidence of malignancy.

Conclusions: We conclude that EL is a rare condition, but there have been cases reported in the literature. Imaging modal-

ities such as US and computed tomography scans are recommended to rule out other underlying diagnoses and should be tailored to each individual when necessary. Because the presence of EL attached to the vesicle is a rare occurrence in the literature, a histological examination is required due to the elevated risk of hepatocellular carcinoma. Keeping such an abnormality in mind can help surgeons demarcate the embryological

plane of dissection during cholecystitis to avoid tumor cell spillages if present.

Keywords: Cholecystectomy, Laparoscopic • Cholecystitis • Choristoma

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Background

Ectopic liver (EL) is liver tissue that is not attached to the mother liver, and is also known as choristoma and hepar succenturiatum [1]. According to Watanabe's 1060-patient series, it has an incidence rate of less than 1% (0.47%) [1].

A literature review concluded that there have been 91 reported patients with gallbladder and EL connected to it [2]. More than two-thirds of EL cases (69.2%) were detected by chance during cholecystectomy.

EL is a rare congenital anomaly lacking an anatomical connection to the liver proper [3]. EL is thought to be caused by an abnormal migration of hepatic tissue from the foregut diverticulum at cystic formations. The attachment to the gallbladder surface is the most common location of EL, but it can occur elsewhere, including the heart, lungs, chest wall, diaphragm, umbilicus, spleen, pancreas, esophagus, stomach, jejunum, adrenals, kidneys, testes, retroperitoneum, hepatic ligament, and inferior vena cava [4-6].

Preoperative EL diagnosis was reported in only a few papers. EL does not usually cause symptoms and, due to its small size, can be overlooked on imaging [7]. The gallbladder must be removed, preferably with the assistance of an "endobag." Because malignant transformation to hepatocellular carcinoma of the choristoma has been observed in less than 5% of gallbladders associated with EL [7], there is a greater risk of neoplastic cell implantation in the portals of access to the abdominal cavity. We report our successful management of a case of completely excised EL on the gallbladder.

Case Report

Our patient was a 47-year-old Saudi man with no history of medical illness or surgery. He presented to the gastroenterology clinic in December 2020 with right upper-quadrant pain that had been present for 2 months, no nausea or vomiting, and no changes in urine or stool color. He had a history of Helicobacter pylori infection, was on Helicobacter pylori treatment, and reported no improvement. Examination was unremarkable, other than tenderness in the right upper quadrant. Laboratory workup revealed C-reactive protein (CRP) of 1.03 mg/L (normal range 0-3.0 mg/L), alkaline phosphatase (ALP) of 79 U/L (normal range 40-129 U/L), direct bilirubin of 0.231 mg/dL (normal range 0-0.2 mg/dL), total bilirubin of 0.729 mg/dL (normal range 0.146-1.4 mg/dL), aspartate aminotransferase (AST) of 64.8 U/L (normal range 0-40 U/L), and alanine aminotransferase (ALT) of 45 U/L (normal range 0-40 U/L). Complete blood count, creatinine, and serum electrolytes were all within normal limits. Serology results of hepatitis B, C, and HIV were negative.

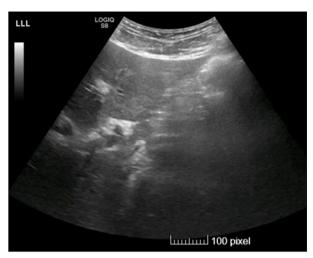


Figure 1. US abdomen showing gallbladder thickened wall (5 mm) with multiple stones.

The US abdomen (Figure 1) revealed a calcular gallbladder with a slightly thickened wall (5 mm), no evidence of focal lesion or intrahepatic biliary dilatation, and CBD, and the portal vein was within normal range. After completing *Helicobacter pylori* treatment, the patient was seen again in the gastroenterology clinic 1 week later. He still had pain, bloating, and decreased appetite. The examination was unremarkable. The results of the tests revealed that there was no *Helicobacter* antigen in the stool and no occult blood. The patient was referred to a surgery clinic for surgical management after being diagnosed with acute cholecystitis on top of chronic cholecystitis.

In March 2021, the patient was admitted voluntarily for lap-aroscopic cholecystectomy. Prophylactic antibiotics were given in the form of cefoxitin sodium 1000 mg. The patient was intubated and ventilated mechanically. He was placed in supine position. The abdomen was prepared and draped before 4 ports were inserted in a similar manner. The gallbladder was mildly inflamed, with attachment to the duodenum and EL on the gallbladder (Figure 2). Adhesolysis was performed first by exposing the Calot's triangle. A critical view of safety was maintained. The cystic artery and duct were clipped using a hemoclip. The gallbladder was removed using an endobag, and hemostasis was ensured (Figure 3). Fascia and skin were closed. The patient was extubated without complications and was transferred to the recovery room in stable condition.

Postoperatively, the patient was given omeprazole 40 mg i.v. once daily, as well as analgesia (ibuprofen 400 mg i.v. every 6 h) and cefoxitin sodium 1000 mg i.v. every 8 h. Six hours after surgery, he began a low-fat diet. He was hospitalized for 1 day. He had no postoperative symptoms, was able to eat, and had no nausea or vomiting. He was released from the hospital in good condition.

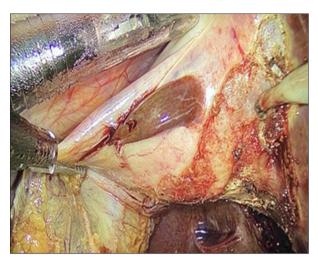


Figure 2. Ectopic liver tissue on the gallbladder during laparoscopic cholecystectomy.



Figure 3. Ectopic liver tissue on the gallbladder during retrieval.

The patient was seen and evaluated in the surgery clinic 2 weeks after the procedure as part of a routine follow-up. He had no symptoms. The examination was uneventful, and the surgical wounds were clean and dry.

Gall bladder histological examination revealed that the gall-bladder measured $8.6 \times 3 \times 1.3$ cm, the serosa was greenish brown, the mucosa was dark green and rough, the wall thickness ranged from 0.2 cm to 0.4 cm, and there were several black stones. The gallbladder was described microscopically as having extensive ulceration of the mucosa and wall, as well as chronic inflammation, with no evidence of malignancy, and liver tissue measuring 12×5 mm attached. As a result, the diagnosis of chronic calcular cholecystitis with normal EL architecture and no dysplasia or malignancy was reached.

Discussion

Ectopia is defined as the relocation or misalignment of an organ or a body part. Ectopic liver was discovered for the first time in 1922 [8]. EL can be found anywhere other than the mother liver, such as the thorax, umbilical cord, intra- or extra-pleural, retroperitoneal, jejunal, pancreas gland, adrenal glands, spleen, falciform ligament, pylorus, esophagus, or pericardiac cavity [4]. EL is most often noted on the gallbladder [4]. Similarly, in our case, an EL was discovered on the gallbladder.

Several mechanisms could explain the presence of EL in various locations [9]. Most researchers agree on 2 possible explanations: the first is the development of an EL, in addition to decay or reduction of the original attachment to the main liver, and the second is the displacement or migration of a portion of the pars hepatica to the rudiments of various organs [10,11].

Most cases of EL are asymptomatic, and imaging studies are rarely used to diagnose it before a surgical procedure [2]. Diagnosis may be missed due to its small size [7,12] and because radiologists may be unaware of it. There is an established correlation between EL and gallbladder diseases such as cholelithiasis and cholecystitis [6]. Nonetheless, it can present with recurrent abdominal pain due to hemorrhagic necrosis, torsion, or rupture, or with pressure symptoms due to mass formation due to malignant alteration [13], including esophageal obstruction and portal vein obstruction. Our patient presented with symptoms of cholecystitis, and ectopic liver tissue was discovered by chance during laparoscopic cholecystitis surgery.

EL has the same histopathology as the mother liver, including cirrhosis, hemosiderosis, hepatitis, and hepatocellular carcinoma (HCC) [3]. HCC was found in less than 5% of gallbladder cases associated with EL, and several cases had normal native liver tissue [7,14]. Although HCC is uncommon, it requires the same approach as carcinoma in the mother liver, according to National Comprehensive Cancer Network Guidelines [12]. Our histological examination of the gallbladder revealed no evidence of malignancy.

Ultrasound-guided (USG) percutaneous biopsies have been shown to be useful for preoperative diagnosis by revealing normal liver parenchyma [15]. In addition, computed tomography (CT) and hepatobiliary iminodiacetic acid (HIDA) scans can help with diagnosis. Furthermore, color Doppler USG or angiography could show a nourishing vessel [15]. However, our patient presented with cholecystitis symptoms and thus underwent abdominal ultrasound (US). Because the US abdomen revealed no ectopic tissue, it was not possible to order additional investigations, such as a CT, MRI, or HIDA scan in a case of cholecystitis. As a result, imaging modalities should be tailored to each patient.

Conclusions

We conclude that EL is a rare entity, but there have been reports of cases in the literature. Imaging modalities, such as US and CT scans, are suggested to eliminate other deferential diagnoses and should be tailored to each individual when necessary. Due to the increased risk of hepatocellular carcinoma, the presence of EL adhered to the vesicle is a possibility that is not commonly reported in the literature, so a histopathological study is mandatory. In addition, the EL must be removed in an endobag to avoid any possibility of port sealing of cancer cells when present. Being aware of this anomaly can help surgeons delineate the embryological plane of dissection during cholecystectomy and eliminate the risk of malignant transformation of EL.

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Conflict of Interest

None declared.

Declaration of Figures Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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