Case study

Two abnormalities in one: A rare case of partial intrahepatic gallbladder with Fitz Hugh Curtis Syndrome.

Abstract:

Aim: Present the case of a female patient with chronic upper right quadrant pain that underwent laparoscopy with the findings of Fitz-Hugh-Curtis syndrome with a partial intrahepatic gallbladder and report the clinical importance of the case and the lessons learned, for optimal future treatment.

Presentation of case: A 55-year-old female presented on the general surgery outpatient clinic, with a history of 1-year-old chronic right upper quadrant pain. It was decided to perform abdominal ultrasound reporting gallstones. She underwent a laparoscopy, findings included perihepatic adhesions between the liver capsule and anterior abdominal wall. The gallbladder had a 70% intrahepatic surface which occasioned a technical difficulty because traction of the gallbladder to improve vision resulted in tearing of the hepatic capsule and, therefore, bleeding of the hepatic bed.

Discussion: It is important to acknowledge the technical difficulties in the cholecystectomy when a Fitz-Hugh-Curtis syndrome is present, this because the violin strings don't permit the gallbladder retraction, and therefore they must be lysed on both hepatic lobes.

Conclusion: We present a rare case of Fitz-Hugh-Curtis syndrome in a patient with a partial intrahepatic gallbladder. It highlights the need for a better diagnosis approach and imaging methods in order to achieve a better planning and decrease the risk of complications.

Key Words: Intrahepatic gallbladder, Fitz-Hugh-Curtis syndrome, perihepatitis, cholecystitis

Introduction:

Fitz-Hugh-Curtis Syndrome is an inflammation of the liver capsule secondary to a complicated pelvic inflammatory disease, where the most common etiologic agent is C. trachomatis.[1,2] This acute perihepatitis is presents with a typical “violin-string” adhesions between the liver and the anterior abdominal wall or the diaphragm.[3]

The gallbladder can be situated in a variety of anomalous positions. An partial intrahepatic gallbladder is one that is partially embedded within the liver parenchyma. First described in medical literature en 1935, intrahepatic gallbladders predominantly result from a developmental anomaly, but in some instances have been reported to be secondary to chronic inflammation.[4] Cholelithiasis rates can be as high as 60% in this cases,[5] and may be a technique challenge for the general surgeons during laparoscopic cholecystectomy, especially if its associated with another anomaly. We present a case of Fitz-Hugh-Curtis Syndrome associated with partial intrahepatic gallbladdler, presented with chronic cholelithiasis, and its laparoscopic approach.

Case presentation:

A 55-year-old multiparous female presented on the general surgery outpatient clinic, whose personal history highlights diabetes mellitus, no other diseases. She has a history of 1-year-old chronic right upper quadrant pain, and decreased appetite. She also described the onset of pain coincided with ingestion of cholecistoquinetics and sometimes was accompanied with nausea but never vomiting. She had never developed fever, altered bowel habit or other symptoms. On examination, she was hemodynamically stable, afebrile. Normal pulmonary and cardiac auscultation. Abdomen not distended, preserved peristalsis, soft, depressible, mildly painful on superficial palpation and defense in right quadrant with negative Murphy maneuver.

Blood tests showed Hb: 15.5 g/dl, WBC: 5,410, neutrophils 50%, platelets 238,000, creatinine 0.82 mg/dL, total bilirubin 0,5 mg/dL, Sodium 140, Potassium 4.1, AST 25, ALT 22, amylase 21. Urine sediment was normal. The chest radiograph was within normal limits. Due to suspicion of hepatobiliary disease, it was decided to perform abdominal ultrasound reporting gallbladder with

acoustic shadow in all its length with no perivesicular fluid, with the diagnostic impression of gallstones. The diagnosis of cholelithiasis was made. She underwent a laparoscopy, findings included perihepatic adhesions between the liver capsule and anterior abdominal wall (Figures 1 and 2). With cautery, these adhesions were lysed on the right hepatic lobe. The gallbladder was then retracted over the liver where it was seen that more of the 70% of its surface was intrahepatic. The critical view of safety was achieved with difficulty because traction of the gallbladder to improve vision resulted in tearing of the hepatic capsule and, therefore, bleeding of the hepatic bed. Cystic artery and duct where clipped and cut. Following, retrograde dissection of the gallbladder from the liver bed was performed with a L-hook monopolar energy device. There was no peritoneal plane between the gallbladder and liver bed, which result in bleeding (Figure 3). After complete disassociation of the gallbladder from its bed, hemostasis was made with monopolar energy. The patient was discharged asymptomatic the same day. At her 2 week and 1 month follow-up examination, she remained asymptomatic.

Discussion:

The gallbladder is typically located under the right lobe of the liver, in the plane of the interlobar fissure. The 4 most common ectopic locations of the gallbladder include under the left liver lobe, intrahepatic, transverse, and retroplaced.[4] The incidence of an ectopic gallbladder is reported to be 0.1 - 0.7%.[6] Complete or partial Intrahepatic gallbladders results from failure of the gallbladder to move from its intrahepatic position in the first trimester of gestation. This abnormality has more risk of cholelithiasis because they do not completely empty, leading to impaired function, contributing to stasis and formation of gallstones.[7]

Fitz-Hugh-Curtis syndrome is characterized by perihepatic inflammation concomitantly with pelvic inflammatory disease.[8] The mechanism by which this inflammation occurs in the liver capsule is due to the spread of these germs that ascend from the pelvis to the surface of the liver as a result of the peritoneal fluid movement through the paracolic gutter into subphrenic space.[9] This syndrome

provides a diagnostic challenge as it can mimic many other diseases (most often acute cholecystitis). To our knowledge, this is the first report in the literature that describes the incidence of partial intrahepatic gallbladder with a Fitz-Hugh-Curtis syndrome. In the case presented here, surgery remains the mainstay treatment to avoid future complications. It is important to acknowledge the technical difficulties in the cholecystectomy when a Fitz-Hugh-Curtis syndrome is present, this because the violin strings don't permit the gallbladder retraction, and therefore they must be lysed on both hepatic lobes. In this case the manipulation and dissection of the intrahepatic gallbladder was even more difficult, because in the beginning we only lysed the violin strings on the right hepatic lobe which contributed to a more significant bleeding, and forced us to lyse the left hepatic lobe adhesions in order to gain mobility of the structures

Conclusion:

Fitz-Hugh-Curtis syndrome associated with a partial intrahepatic gallbladder is a rare anomaly. It is challenging for both radiologists and surgeons. Both should be aware of this condition in surgical planning for a better patient management. Laparoscopy offers in both cases, a definitive diagnosis and immediate relief of the distressing symptoms when performed by a expert hepatobiliary surgeon.

Lessons Learned:

- When finding an anatomic abnormality, never overlook the possibility of having more abnormalities.
- When performing cholecystectomy in Fitz-Hugh-Curtis syndrome, always start with lysing the perihepatic adhesions on both hepatic lobes to gain more mobility.
- The most frequent complication in the dissection of a intrahepatic gallbladder, is bleeding.

Competing interests / Disclosure:

Nothing to disclose

Author's contributions:

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Figures:

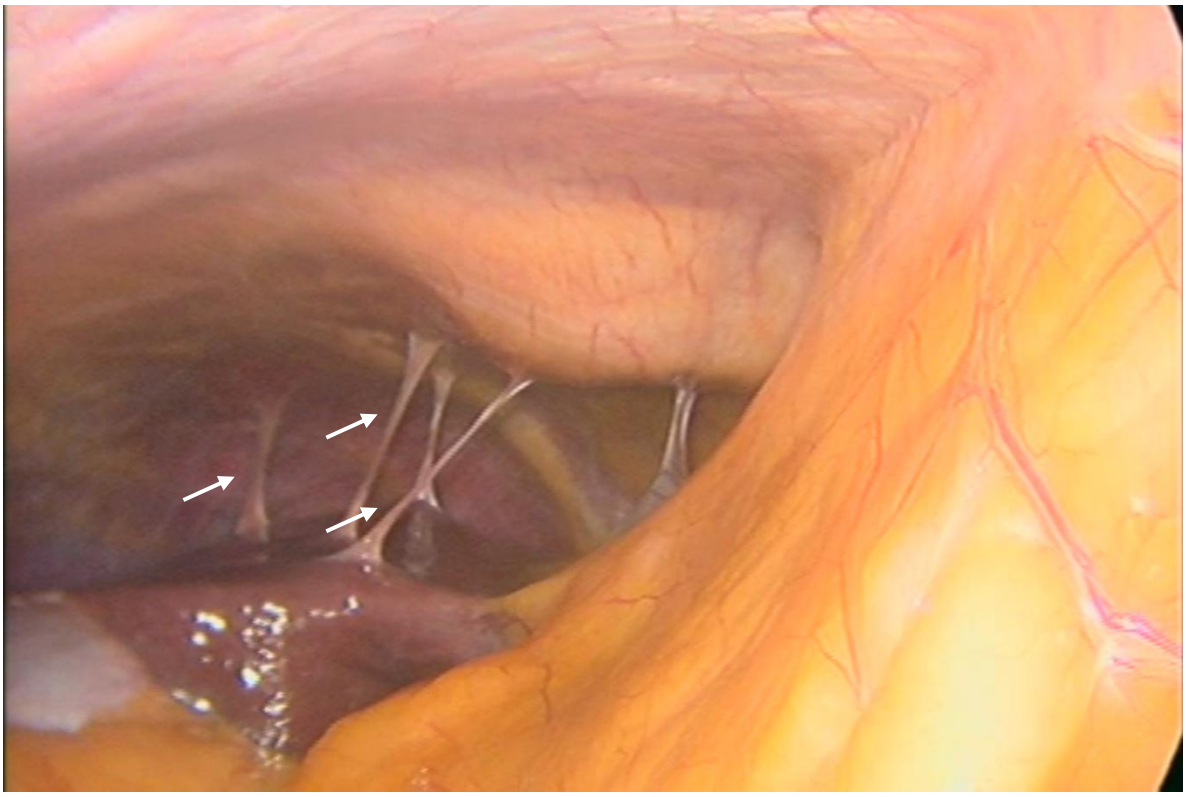

Figure 1: Laparoscopic view of the right hepatic lobe with visualization of violin-string adhesions between anterior surface of liver and anterior abdominal wall (arrows).

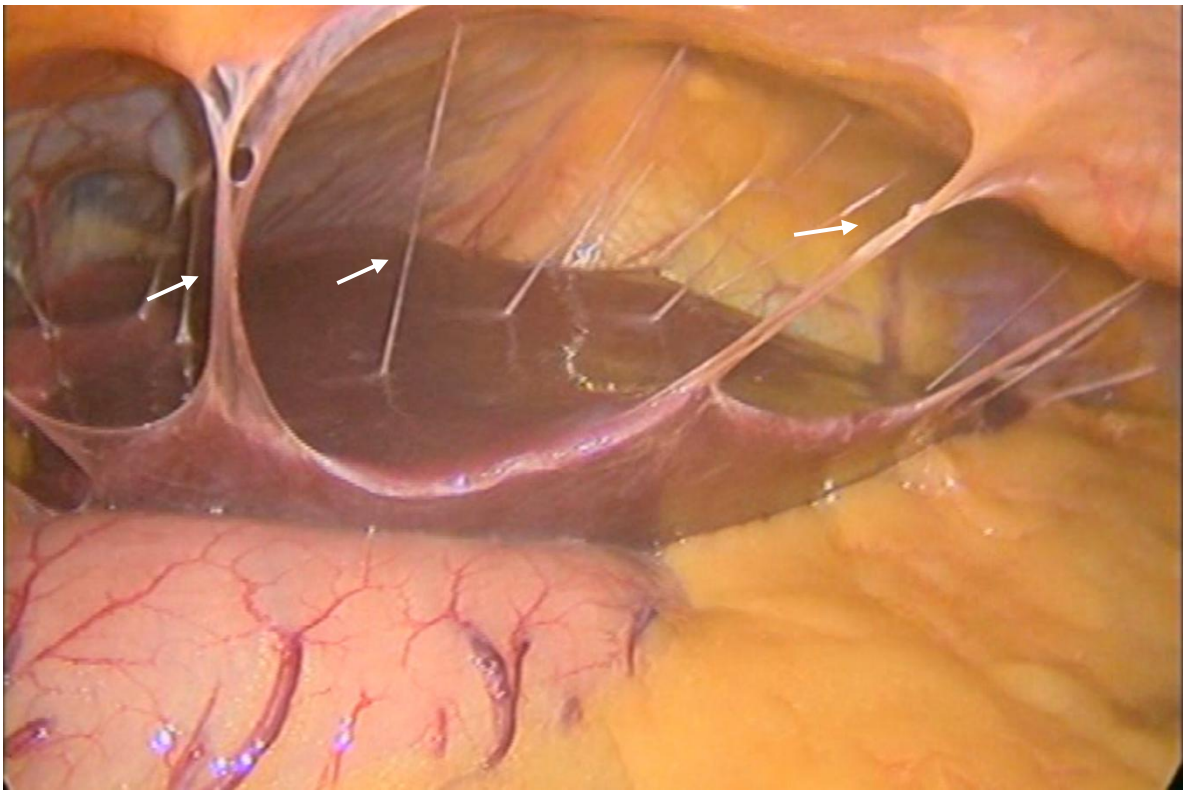

Figure 2: Laparoscopic view of the left hepatic lobe with visualization of violin-string adhesions between anterior surface of liver and anterior abdominal wall (arrows).

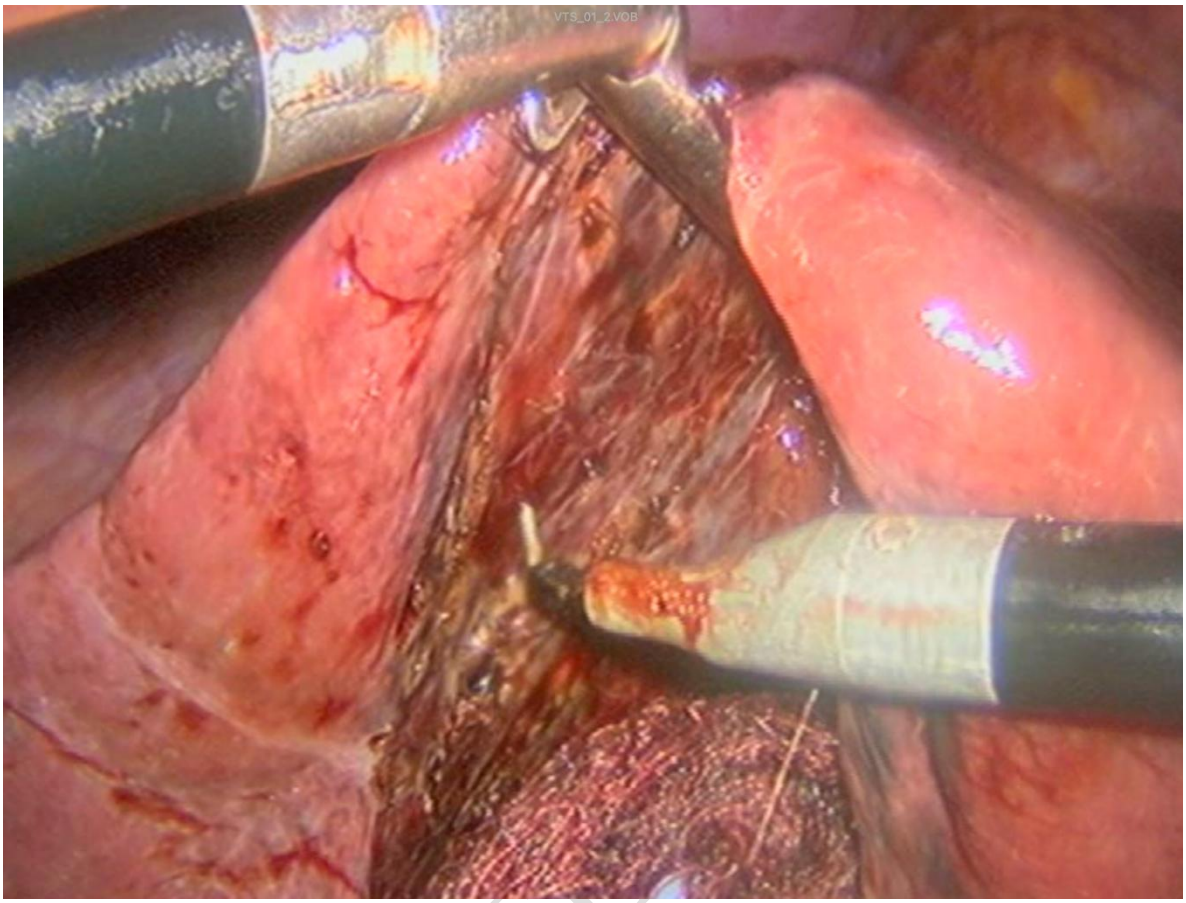

Figure 3: Bleeding of the liver bed after cholecystectomy of a partial intrahepatic gallbladder.

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