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Abscess of ligamentum teres hepatis post-endoscopic retrograde cholangiopancreatography: A case report and a literature review

Zixiang Ji, Zhenyu Wang and Hao Li

Abstract

Abscess of the ligamentum teres hepatis has been described in the medical literature as an extremely rare clinical entity, which often presents a diagnostic dilemma. A 68-year-old man was hospitalized for upper abdominal pain and obstructive jaundice. The patient presented with low-grade intermittent fever. Laboratory investigations showed a white blood cell count of 32.38×10^9/L, a C-reactive protein level of 247.86 mg/L, abnormal liver enzyme and bilirubin levels, and elevated serum levels of amylase and lipase. He was first diagnosed with acute biliary pancreatitis. A computational tomography scan and magnetic resonance cholangiopancreatography revealed obstructive choledocholithiasis and cholecystolithiasis. The patient received preoperative antibiotics and symptomatic treatments for 5 days, followed by endoscopic retrograde cholangiopancreatography and a subsequent duodenal papilla incision to extract pigment and cholesterol gallstones. The patient recovered and was discharged on the fifth day after surgery. However, 10 days later, the patient was readmitted for the recurrence of acute calculous cholecystitis. Laboratory tests showed increases in total and direct bilirubin, γ-glutamyltransferase, and alkaline phosphatase, but not inflammatory parameters. After the patient's nutritional status improved on the 11th day after admission, a laparoscopic cholecystectomy was performed. Intraoperative exploration revealed extensive abdominal adhesions; a thickened edematous gallbladder wall; and an unexpected abscess of the ligamentum teres hepatis. Pus aspiration was performed laparoscopically after laparoscopic cholecystectomy, and to ensure elimination of the abscess, ultrasound-guided pus aspiration was also performed 1 week later. Fortunately, the patient made an uneventful recovery and was discharged with a drain tube on the 16th day after surgery. Doppler ultrasound indicated that the abscess had completely disappeared 2 weeks after discharge. This case highlights an unusual presentation of a ligamentum teres hepatis abscess caused by obstructive cholangitis but that appeared after the choledocholithiasis was resolved. However, the mechanism of abscess formation remained uncertain.

Keywords

Ligamentum teres hepatis, abscess, obstructive choledocholithiasis, calculous cholecystitis, acute biliary pancreatitis

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Introduction

An abscess of the ligamentum teres hepatis (LTH) is an infrequent clinical entity. Although the etiology remained unclear, the LTH abscess is supposedly induced by acute calculous cholecystitis, cholangitis, and pancreatitis in adults, as well as omphalitis and infected ventriculoperitoneal shunts in newborns/infants.[1–6] The most common symptoms of the disease are fever, abdominal pain, and vomiting, which make clinicians often confused with clinical manifestations that are primarily associated with pancreatitis, biliary tract infection, and other conditions, leading to missed or delayed diagnosis.

As for the treatment options, aspiration of the pus followed by surgical excision of the abscess is recommended to prevent septic shock with severe peritonitis.[7,8] However, conservative treatment with endoscopic biliary drainage and broad-spectrum antibiotics has also been effective in

Department of General Surgery, Affiliated Hospital of Yanbian University, Yanji, P.R. China

Corresponding Author:
Hao Li, Department of General Surgery, Affiliated Hospital of Yanbian University, 1327 Juzi Street, Yanji 133000, Jilin, P.R. China.
Email: lih@ybu.edu.cn

Table 1. Laboratory examination outcomes.

	First	Second	Normal range
White blood cell (WBC)	**32.38×10^9/L**	5.89×10^9/L	$4.0–10.0 \times 10^9$/L
Percentage of neutrophil granulocyte (%)	**96.20**	61.40	50–70
C-reactive protein (CRP)	**247.86 mg/L**	**10.2 mg/L**	0–5 mg/L
Total bilirubin (TBIL)	**84.0 μmol/L**	25.2 μmol/L	5.1–25.6 μmol/L
Direct bilirubin (DBIL)	**75.6 μmol/L**	**18.0 μmol/L**	1.7–6.8 μmol/L
Aspartate aminotransferase (AST)	**97 U/L**	25 U/L	0–40 U/L
Alanine aminotransferase (ALT)	**134 U/L**	37 U/L	0–40 U/L
Gamma-glutamyltransferase (GGT)	**370 U/L**	**154 U/L**	8–58 U/L
Alkaline phosphatase (ALP)	**200 U/L**	**175 U/L**	–42–140 U/L
Albumin	**27 g/L**	**31 g/L**	37.0–53.0 g/L
Carbohydrate antigen (CA)19-9	**>1962.0 U/mL**	29.5 U/mL	0–35 U/mL
Amylase	**1233 U/L**	83 U/L	0-220 U/L
Lipase	**4690 U/L**	152 U/L	73-393 U/L

Abnormal values are shown in bold.

improving condition.[4,7,9,10] The latter can be considered the primary choice when the patient's general conditions allow.

Case presentation

A 68-year-old man was admitted to treat abdominal pain and notable jaundice of the skin and sclerae. The pain had started 1 month earlier and was significant in the upper right quadrant without radiation to the back. It was associated with nausea and vomiting. He was treated with cephalosporins at a local clinic before admission to our hospital, but did not improve symptoms. On physical examination, his abdomen appeared flat and soft with palpable tenderness. No abdominal mass was palpable. Murphy's sign was negative. The patient presented with intermittent low fever (<38.5°C). Laboratory investigations showed a white blood cell count of 32.38×10^9/L, a C-reactive protein level of 247.86 mg/L, abnormal liver enzyme and bilirubin levels, and elevated serum amylase and lipase levels (Table 1). In addition, blood culture was tested to be negative for bacteria.

A computational tomography (CT) scan of the upper abdomen and magnetic resonance cholangiopancreatography (MRCP) revealed gallstones and choledocholithiasis with a 5.0-mm stone in the distal part of the common bile duct, leading to dilation of the intrahepatic and extrahepatic biliary duct (Figure 1(a) and (b)). Our first diagnosis was acute pancreatitis, obstructive choledocholithiasis, and cholecystolithiasis. The patient received anti-infection medications (meropenem, piperacillin, and levornidazole) for 5 days, followed by endoscopic retrograde cholangiopancreatography (ERCP) to remove gallstones with pigment and cholesterol. Introperative cholangiography (IOC) also confirmed dilation of the common bile duct and a filling defect in the distal part. The patient was discharged 5 days after surgery. He was scheduled for a laparoscopic cholecystectomy a month later.

However, within 10 days, the patient was readmitted with complaints of abdominal pain in the upper right quadrant, intermittent fever, nausea, and vomiting. The physical examination presented results similar to those of the first admission, except that skin and sclera jaundice were absent. Laboratory examinations showed that inflammatory parameters improved (Table 1). A laparoscopic cholecystectomy was performed after improving the patient's nutritional status after 11 days. Surprisingly, subsequent intraoperative exploration revealed an abscess of the LTH, initially considered an encapsulated effusion due to acute pancreatitis and mild peritonitis. A thorough retrospective examination on CT scans was conducted, although this LTH abscess was absent in the imaging at the first admission (Figure 1(c)).

We then reevaluated the MRCP results before surgery. The lesion appeared to show mixed signals on the T2-weighted imaging (T2WI) that extended from the median fissure of the liver to the lower abdomen (Figure 1(d) and (e)). The combined diffusion-weighted imaging (DWI) sequence showed high signal intensity and restricted diffusion (Figure 1(f) and (g)), representing the LTH abscess.

After cholecystectomy, a 1-cm parallel incision was made in the LTH and 40 mL of thick gray-yellow pus containing necrotic tissue was aspirated. The pus and blood culture were tested twice to be negative for bacteria. Twenty-two French drainage tubes were placed individually in the abscess incision and below the liver. The volumes of pus draining gradually decreased in a week.

However, a subsequent CT scan detected the persistent abscess, which appeared to be a draining tube attached to the wall, suggesting that the draining of the pus was insufficient (Figure 1(h)). Ultrasound-guided pus aspiration was followed immediately (Figure 1(i)). A total of 100 mL of pus was initially obtained, followed by a steadily reduced volume each day, and the color of the pus gradually turned light yellow. The tube was constantly flushed. The patient was discharged 16 days after surgery. Doppler ultrasound showed that the abscess cavity had completely disappeared 2 weeks after discharge (Figure 1(g)). The patient remained

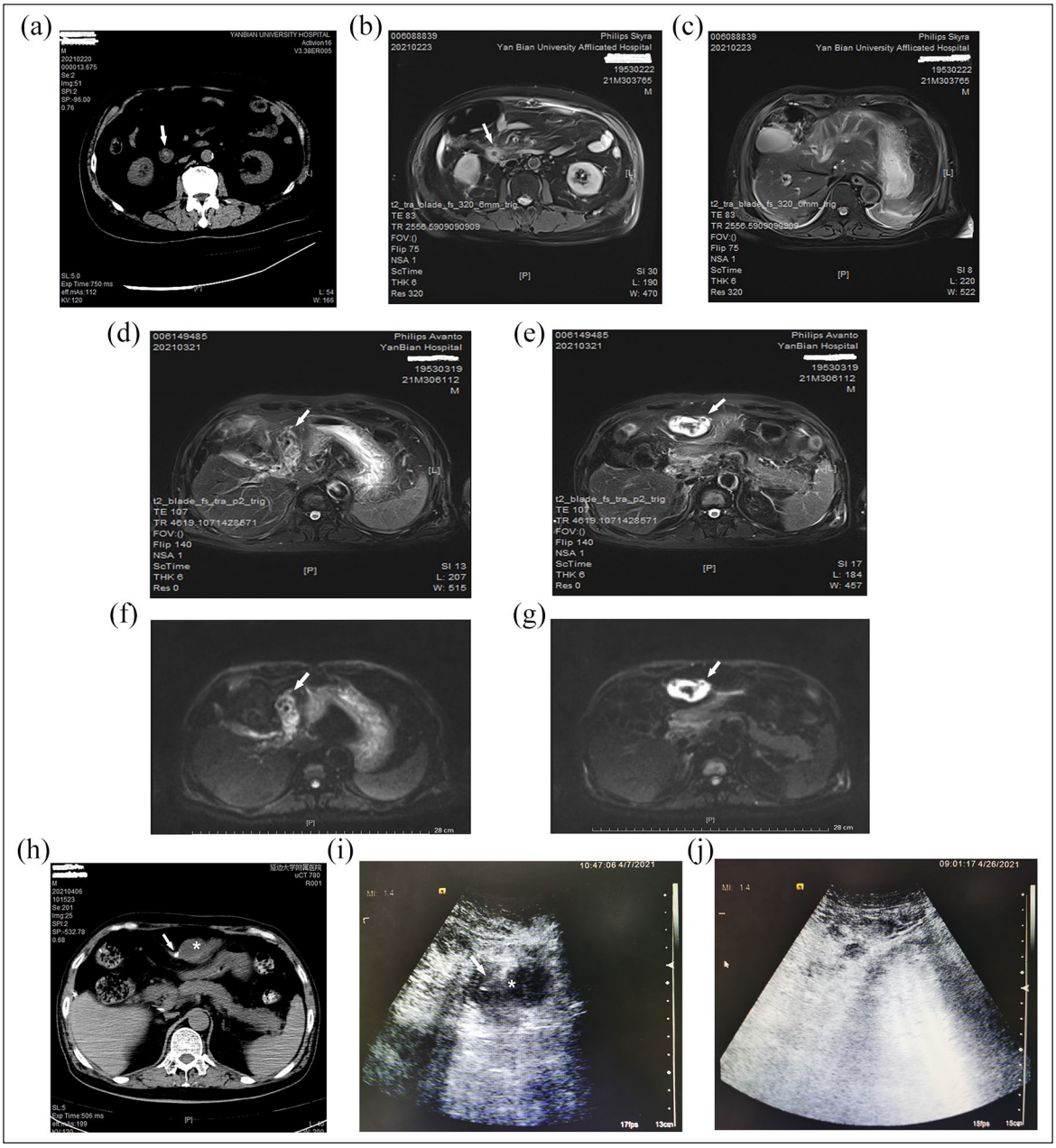

Figure 1. Images of CT, MRCP, and ultrasound at presentation. (a, b) CT and MRCP revealed choledocholithiasis with a gallstone in the distal part of the common bile duct. (c) Abscess of the LTH was not detected at the first visit. (d, e) T2WI showed a lesion with a mixed-signal intensity that extended from the median fissure of the liver to the lower abdomen. (f, g) Combined diffusion-weighted imaging (DWI) sequences showed increased signal intensity and restricted diffusion. The white arrows indicate the lesion in D–G. (h) Postoperative abdominal CT revealed an irregularly shaped low-density lesion in the upper/middle abdomen, confirming the persistent abscess. The white arrow indicates the drain tube attached to the abscess wall, suggesting insufficient pus drainage. (i) Ultrasound-guided abscess drainage was followed. The white arrow indicates the puncture needle. The white asterisk indicates the abscess cavity in H and I. (j) Doppler ultrasound showed that the abscess cavity had completely disappeared 2 weeks after discharge.

asymptomatic with normal laboratory biochemistry and the abscess had not recurred at the 1-year follow-up.

Discussion

The LTH, also known as the "round ligament of the liver," is a structure that makes up the falciform ligament (FL). Due to the rarity of LTH abscess, limited literature is available. We extensively reviewed the English literature (with abstract available) using PubMed, Google Scholar, and manual cross-referencing that covers abscess of LTH or FL up to 2021. To better compare with our case, only those reported in adults were included. A total of 18 reports containing the present one are listed (Table 2). As is known, the leading

Table 2. List of Reports.

Reference	Age	Sex	Main presentation	Concomitant disease	Microbiology	Treatment
Charuzi and Freund[11]	75	F	Abdominal pain, high fever	Not significant	(−)	Surgical resection
Sones et al.[12]	71	M	Abdominal pain and distension, palpable gastric mass	Cholecystitis, rupture of gangrenous gallbladder	Not described	Surgical resection
Watson et al.[13]	84	F	Abdominal pain, vomiting	Cholecystolithiasis	Not described	Surgical resection
Losanoff and Kjossev[8]	18	M	Abdominal pain, nausea, vomiting	Not significant	Blood (−), peritoneal pus (+*Escherichia coli*)	Surgical resection
De Melo et al.[14]	65	M	Abdominal pain, fever	Calculous cholecystitis	Not described	Laparosopic abscess drainage (recur), cholecystectomy
Martin[15]	52	F	Light abdominal pain, epigastric burning pain, nausea, vomiting	Cholecystolithiasis, cholecystitis	Not described	Surgical resection
Tsukuda et al.[16]	70	F	Abdominal pain, high fever	Choledochitis and pancreatitis due to choledocholithasis	Pus (+*Staphylococcus epidermidis*)	Antibiotics, ERCP, cholecystectomy, surgical resection
Arakura et al.[7]	63	M	Epigastralgia, high fever	Cholecystolithiasis, portal thrombosis	Bile and blood (+*Streptococcus anginosus*)	Antibiotics, thrombolytic therapy
Czymek et al.[17]	44	F	Epigastralgia	Cholecystolithiasis	Resected specimen (+*Staphylococcus epidermis*)	Surgical resection
Warren et al.[18]	73	M	Extreme tenderness in upper abdomen, jaundice, anorexia, nausea	Cholangitis, obstructive ampullary carcinoma, portal pyemia	Not described	Antibiotics, ERCP, pancreaticoduodenectomy, surgical resection
Atif and Khaliq[19]	40	M	Epigastralgia, anorexia, vomiting, a noticeable mass in epigastrium	Pancreatitis	Not described	Excision of falciform ligament and para-duodenal abscess
Sen et al.[9]	40	M	Acute abdomen, palpable sausage-shaped, supraumbilical mass	Obstructive choledocholithiasis, cholangitis, portal thrombosis	Pus (+*Escherichia coli*)	Pus aspiration, ERCP, cholesytectomy, surgical resection
Jain et al.[20]	65	F	Epigastralgia, vomiting	Cholecystitis, cholelithiasis	Not described	Pus aspiration, antibiotics, cholecystectomy
Fujikawa and Araki[10]	86	F	Hypochondralgia, fever	None	Pus (+*Serratia marcescen*)	Pus aspiration, antibiotics
Bhattacharya et al.[21]	69	F	Abdominal pain, palpable epigastric lump	Calculus cholecystitis	Not described	Antibiotics, upcoming cholecystectomy
Zorgdrager et al.[22]	66	M	Periumbilical painful swelling, loss of appetite, fever	Abdominal wall abscess, partial portal thrombosis, liver atrophy	Pus (+*Enterobacter aerogenes, Enterobacter cloacae, Streptococcus milleri*)	Surgical resection
Fang and Huang[23]	33	M	Epigastralgia	Calculous cholecystitis	Not described	Surgical resection of the gallbladder and round ligament
Current case	**68**	**M**	**Abdominal pain, jaundice, nausea, vomiting, low-grade fever**	**Pancreatitis, obstructive choledocholithiasis, cholecystolithiasis, calculous cholecystitis**	**Blood (−), pus (−)**	**Antibiotics, ERCP, cholecystectomy, laparoscopic pus drainage/US-guided aspiration**

ERCP: endoscopic retrograde cholangiopancreatography.

presentation of LTH abscesses is abdominal pain (epigastralgia), fever, and vomiting. Of the 18 cases, 4 presented palpable/noticeable mass in the epigastrium.[9,12,19,21] The majority of cases are accompanied by at least one of these complications, pancreatitis, obstructive choledocholithiasis, cholangitis, and portal thrombosis. An LTH or FL abscess was first suspected in most cases by imaging and confirmed by intraoperative exploration followed by treatments. There are only two cases, the current case and the other reported by Tsukuda et al.,[16] that presented abscess in a delay after ERCP was conducted. Six patients were cured with conservative therapy,[7,10,14,20,21] while the rest underwent surgical resection, including some who received conventional treatment primarily but recurred or did not shrink the abscess.[8,9,11–13,15–19,22,23]

As such, scholars classify LTH abscess as a complication of biliary tract obstruction interpreted as retrograde infection of the portal venous system.[18,19,23] When bacteria invade the hepatic sinusoids and systemic circulation, they can cause abscesses in the round ligament of the liver. However, some cases develop an LTH abscess without a history of pancreatitis or biliary tract infection, particularly in children.[1–4] Others have proposed that the lymphatic spread of infection could be another cause of abscess formation.[14] Because the superficial lymphatics of the liver drain the lymph from the FL, it could spread infections and malignancies to the FL through blood or lymphatics. It could also collect infections or malignancies from surrounding structures and seed them in the FL.[18]

Our patient presented with severe infection and was initially treated with ERCP for obstructive cholangitis. However, when he was readmitted for laparoscopic cholecystectomy, an LTH abscess was incidentally identified during surgery. A detailed retrospective investigation in imaging revealed that this abscess appeared on the second visit but not on the first, raising a causal inference dilemma. Here, we discuss some potential causes that could link to this abscess formation.

First, we suspect that due to obstructive choledocholithiasis, the retrograde spread of the infection from the biliary tree to the liver sinusoids could have led to an abscess of the LTH.[18] However, since the patient had a low intermittent fever and aspirated pus and blood cultures were negative for bacteria, the antibiotics were assumed to be effective. Therefore, the hematogenous spread of the infection does not appear to have directly triggered this abscess. Second, could this LTH abscess be an ERCP complication? Tsukuda et al.[16] described a case of the round ligament abscess that occurred after an endoscopic papillotomy for choledocholithiasis, similar to our case, although the mechanism was unclear. In general, injection of contrast dye and biliary flush during ERCP could temporarily increase pressure in the biliary duct, leading to the retrograde spread of bacteria through the biliary tract to the hepatic sinusoids and blood circulation. It can also cause liver damage and LTH abscess.

Nevertheless, this route of infection is consistent with the first. Given the clinical manifestations and treatments provided, it is difficult to infer that sinusoidal infection or hematogenous spread of infection led to this abscess of LTH. Besides, the patient received antibiotics before and after the operation and endoscopic nasobiliary drainage (ENBD) was also performed. Thus, it is unlikely that ERCP directly resulted in an abscess of the LTH.

Next, the round ligament of the liver is a degenerative cord of tissue that extends from the umbilicus to the transverse fissure of the liver. Increased pressure in the portal vein could recanalize the FL to allow blood flow. Yet, no evidence from laboratory tests or associated examinations suggested that the patient had cirrhosis or portal hypertension.

Then, would the lymphatic spread of the infection be the route? Due to its close association with the liver, when there is severe inflammation in the bile duct, it is likely that the LTH is infected by the retrograde lymphatic route to form an abscess. Finally, given that extensive adhesion was observed in the abdomen, direct infection by peritonitis cannot be ruled out as the source of this abscess formation.

Here, we elaborate on the "likely" and "unlikely" routes to spread the infection in this case, but the mechanism of LTH abscess formation remains controversial. It is also interesting to note that no abscess was detected at the initial admission but found at the second. We assume that our patient was on antibiotics before (cephalosporin) and after entry (meropenem, piperacillin, and levornidazole), which slowed but could not stop the development of the abscess.

Regarding the treatment option, surgical excision is recommended to completely resolve the abscess.[5] However, quite a few show that conservative treatment with endoscopic biliary drainage and broad-spectrum antibiotics has improved the condition. We assume that the primary cause of LTH abscess was biliary tract infection due to obstructive cholangitis, which was managed by ERCP and antibiotics. An isolated LTH abscess was discovered coincidently during laparoscopic cholecystectomy (LC); however, his general condition was not sustainable for surgical excision of the LTH immediately after LC because it could have spread the infection and cause diffuse peritonitis or even septic shock. In addition, intraoperative examination revealed that the patient had extensive intra-abdominal adhesions.

Therefore, considering that the patient's inflammatory parameters improved significantly, we believed that the conservative approach using laparoscopic pus aspiration and drainage was the best option for the patient at the time the abscess was confirmed after LC. An ultrasound-guided pus aspiration was also performed to ensure thorough removal of the abscess. Fortunately, drainage of the pus and antibiotic treatment diminished the abscess. Our patient showed improved clinical symptoms and remained asymptomatic with normal biochemical parameters at follow-up.

Conclusion

Imaging techniques are an essential means of inspecting round ligament abscesses. However, exploratory laparotomy or intraoperative diagnosis is often used for confirmation. In the present case, the mechanism of the formation of LTH abscesses is not fully understood. However, in patients with severe infection due to obstructive cholangitis and cholecystitis, although it is not necessarily related to the ERCP procedure, an abscess of LTH can occur as a rare complication afterward. Furthermore, when the infectious states of the patients allow, it is feasible to simultaneously address predisposing conditions, such as laparoscopic cholecystectomy, intraoperative diagnosis, and subsequent conservative treatment with endoscopic biliary drainage and antibiotics.

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Ethical approval

The study is approved by the Ethics Committee of Yanbian University Hospital (No. 2021165).

Informed consent

Written informed consent was obtained from the patient for his anonymized information to be published in this article.

ORCID iDs

Zixiang Ji https://orcid.org/0000-0002-3737-4482

Hao Li https://orcid.org/0000-0001-9441-6776

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