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Case Report / Приказ болесника

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Hepatolithiasis followed by recurrent cholangitis as a consequence of inadequate hepaticojejunostomy for common bile duct injury

Хепатолитијаза праћена рекурентним холангитисима као последица неадекватне хепатико-јејуностомије услед повреде заједничког жучног канала

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SUMMARY

Introduction Hepatolithiasis (HL) is defined as gallstones present in bile ducts above the common bile duct confluence, regardless of the coexistence of gallstones in other parts of the biliary tract. HL is common among patients with recurrent pyogenic cholangitis. Chronic infection can lead to the development of malignancy.

Case outline A 65-year-old woman presented with intermittent fever, jaundice, abdominal pain, and nausea. Eighteen years ago, patient had an open cholecystectomy due to acute cholecystitis. During the early post-operative days patient developed icterus. Intrahepatic biliary ductal dilatation was confirmed by abdominal ultrasound. Due to suspicion of iatrogenic common bile duct injury patient underwent second operation during which said injury was confirmed. "Non-Roux-en-Y" hepaticojejunostomy (HJ) was performed as a problem resolving procedure. Despite performed biliary bypass, patient continued to have episodes of recurrent cholangitis over the eighteen years. Given the patient's recurrent symptoms and results of MRI consistent with HL, surgical treatment was indicated. A left hepatectomy was performed with Roux-en-Y HJ biliary reconstruction. Post-operative course went without complications and since then patient is symptoms free.

Conclusion The main purpose of treating HL is to eliminate infection which leads to recurrent cholangitis and subsequent hepatic fibrosis. Adequate solution of HL decreases the need for repeated interventions and prevent progression of the disease to cholangiocarcinoma.

Keywords: hepatolithiasis; cholangitis; bile duct stones; hepatectomy; Roux-en-Y hepaticojejunostomy; bile duct injury

САЖЕТАК

Увод Хепатолитијаза (ХЛ) се дефинише као присуство каменаца у жучним водовима рачве заједничког жучног вода, без обзира на присуство каменаца у другим деловима жучног тракта. ХЛ је често присутна код болесника са рекурентним холангитисом. Присуство хроничне инфекције може довести до развоја малигнитета.

Презентација болесника Приказана је 65-годишња болесница са тегобама у виду повремене температуре, жутице, болова у трбуху и мучнине. Болесници је пре осамнаест година начињена отворена холецистектомија због акутног холециститиса. У раном постоперативном току болесница развија иктерус. Ултразвук абдомена потврђена је дилатацију интрахепатичних жучних водова. Индикована је поновна операција услед сумње на јатрогену повреду заједничког жучног вода, која је интраоперативно потврђена. У циљу решавања повреде начињена је хепатикојејуностомија по типу "не-Roux-en-Y" анастомозе. Упркос начињеном билијарном бајпасу, болесница је наредних осамнаест година имала епизоде рекурентних холангитиса. На основу тегоба болеснице те налаза магнетне резонанце који су указивали на хепатолитијазу индиковано је оперативно лечење. Начињена је лева хепатектомија са Roux-en-Y хепатикојејуностомијом. Постоперативни ток је протекао уредно и од тада болесница је без тегоба.

Закључак Примарни циљ лечења хепатолитијазе је ерадикација постојеће инфекције која доводи до рекурентног холангитиса и подледичне хепатичне фиброзе. Адекватан третман ХЛ доводи до смањења потребе за понављаним лечењем и превенира настанак холангиокарцинома.

Кључне речи: хепатолитијаза; холангитис; каменци жучних водова; Roux-en-Y хепатикојејуностомија; повреде жучних водова

INTRODUCTION

Hepato­lithiasis (HL) is defined as gallstones present in bile ducts above the common bile duct confluence, regardless of the coexistence of gallstones in other parts of the biliary tract. Hepato­lithiasis occurs most often in East Asia while it is rare in Western countries [1].

HL was the third most common cause of emergency abdominal surgery at a university hospital in Hong Kong during the 1960s. A downward trend has been observed over the years

possibly due to improved standards of living and Westernized diet. Increasing incidence of HL has been noted in Western countries with increasing immigration from East Asia to the West [2, 3, 4].

Recurrent pyogenic cholangitis is frequently followed by hepato­lithiasis. Common presence of bacteria in bile and gallstones indicates the possibility of pattern connection

between bacterial infection and the formation of brown pigment stones. *Escherichia coli*, *Clostridium* and *Bacteroides* show beta-glucuronidase activity and are most common bacterial species isolated from the bile of patients with hepato­lithiasis. Clinically HL may present as acute obstruction and recurrent bacterial cholangitis with all its possible complications, such as liver abscess and septicemic shock, or with chronic complications which refers to cholangiocarcinoma [4–8].

The main purpose of treating HL is to eliminate infection which leads to recurrent cholangitis and subsequent hepatic fibrosis. Adequate solution of HL decreases the need for repeated interventions and prevent progression of the disease to cholangiocarcinoma.

The surgical treatment of hepato­lithiasis implies removal of the affected hepatic segment(s). Complete removal of the diseased hepatic segment or lobe is crucial to preventing disease recurrence and further chronic consequences [9–13]. Best surgical approach for treating hepato­lithiasis is based on high-volume experience from a single center in China that proposed system of classification known as “Dong's Classification” (Table 1) [9].

Here in we present a case of hepato­lithiasis followed by recurrent cholangitis as a consequence of inadequate hepaticojejunostomy for common bile duct injury.

CASE REPORT

A 65-year-old woman presented with intermittent fever, jaundice, abdominal pain, and nausea over the eighteen years. During this period patient was admitted to hospital numerous times due to recurrent cholangitis.

Eighteen-years ago patient underwent open cholecystectomy for acute cholecystitis. During the early post-operative days patient developed icterus. Intrahepatic biliary ductal dilatation was confirmed by right upper quadrant abdominal ultrasound. Due to suspicion of iatrogenic common bile duct injury patient underwent re-operation during which said injury was confirmed. “Non-Roux-en-Y” hepaticojejunostomy (HJ) was performed as a problem resolving procedure for the said injury. Despite performed biliary bypass, patient continued to have episodes of recurrent cholangitis over 18 years.

At the time of the last hospitalization the following blood test results were performed: Hemoglobine (Hgb) 130 g/l, erythrocytes $4.82 \times 10^{12}/L$, leukocytes $5.2 \times 10^9/L$, platelets $194 \times 10^9/L$, total bilirubin 9.4 $\mu\text{mol}/L$, aspartate aminotransverase (ALT) 14 U/L, alanine aminotrasferase (ALT) 14 U/L, Gamma-glutamyltransferase (GGT) 54 U/L, and alkaline phosphatase (ALP)132 U/L. Serology for Hepatitis B and C virus was negative. Signs of HL were present on pre-operative performed abdominal magnetic resonance imaging (MRI) and magnetic resonance cholangiopancreatography (MRCP) (Figure 1).

Surgical treatment was indicated considering the patient's complaints as well as the abdominal MRI findings that suggested HL.

The presence of intrahepatic biliary calculi within the left hepatic lobe were confirmed by intra-operative ultrasound. Referring to Dong’s Classification operation of choice was left hepatectomy with Roux-en-Y hepaticojejunostomy biliary reconstruction (Figures 2 and 3). Tested intra-operative bile cultures came positive for *E. coli* and *Pseudomonas* sp.

Post-operative course was uneventful, and the patient is symptoms free since then. Histopathology showed findings consistent with chronic hepatolithiasis, chronic cholangitis, and secondary biliary cirrhosis (Figure 4).

This case report was written and submitted after obtaining consent of patient.

DISCUSSION

Hepatoolithiasis (HL) is defined as gallstones present in bile ducts above the common bile duct confluence, regardless of the coexistence of gallstones in other parts of the biliary tract. And regardless of whether the confluence is located intra- or extra-hepatically [1–4].

Hepatoolithiasis is most common in East Asia (Singapore 1.7%, Japan 2.2%, Hong Kong 3.1% and Taiwan 50%). Once rare in Western countries, the rate of HL has been rising due to increased immigration from East to West (Western country prevalence <1%). The highest incidence of hepatoolithiasis occurs in the 50th to 60th decades of life and has been reported typically between the age 30 to 70. The combination of intra- and extra-hepatic HL is more frequent in the older groups while intrahepatic form of the disease occurs in the younger age groups [1–10].

Hepatoolithiasis and recurrent pyogenic cholangitis are in thin connection since most patients with HL experienced symptoms of pyogenic cholangitis at least once during the period of the disease. The high incidence of bacteria infested bile and gallstones indicate that there is a close association between bacterial infection and the formation of intrahepatic stones. There are several scenarios how bacteria find route into the biliary tract. One of them is ascending infection through the Sphincter of Oddi, then bacteribilia via the portal venous system. Also, transient infection due to biliary stasis is possible. The most common isolated bacterial species from the bile of patients with hepatoolithiasis are *E. coli*, *Clostridium* and *Bacteroides* spp. This literature data matches with the bacteriology results in our case [6–9].

The main purpose of treating HL is to eliminate infection which leads to recurrent cholangitis and subsequent hepatic fibrosis. Adequate solution of HL decreases the need for repeated interventions and prevent progression of the disease to cholangiocarcinoma.

Choice of the treatment strategy for hepatoolithiasis needs to be based on: 1) the structure of the calculi (cholesterol or calcium bilirubinate); 2) the location of the calculi in bile duct and the most feasible approach to them; 3) well planned treatment that includes resolving of bile duct stenosis; 4) evaluation of liver function, the extent of liver resection, and residual liver volume; and 5) investigation of the presence of intrahepatic bile duct malignancy. Therapeutic strategy must be planned by taking into consideration the history, nature and extent of biliary tract surgery [11, 12, 13]. According to the Clinical Guidelines for Cholelithiasis written by the

Japanese Society of Gastroenterology, treatment selection should be based on the presence or absence of prior biliary tract surgery [10].

Untreated hepatolithiasis can lead to serious consequences such as biliary cirrhosis and even cholangiocarcinoma. Resection of the affected hepatic lobe that contains strictures, atrophy, and multi-segmental distribution of intrabiliary calculi has been effective in reducing disease recurrence and progression of liver disease [11]. Uchiyama [12] performed a retrospective study which compared invasive and non-invasive treatments and procedures in HL treatment to analyze the rate of residual stones and complications, as well as the long-term outcome. Out of 105,062 patients with cholelithiasis treated between 1989 and 1992, among all patients 2,353 (2.24%) were diagnosed with hepatolithiasis. The authors concluded that the most effective therapy was surgery [12]. According to a report of Japanese multi-center-based surveys, there has been a progressive increase in treatment of patients with hepatolithiasis who had previously undergone biliary surgery [14].

We present a patient with an eighteen-year long history of recurrent cholangitis after cholecystectomy during which bile duct injury was made. Patient underwent early reoperation and non-Roux-en-Y hepaticojejunostomy. As it shows in early postoperative days, this form of biliary reconstruction was inadequate, given the absence of dysfunctional jejunum loop (Roux-en-Y) [15].

Safar et al. [16] investigated 12 patients to compare CT, MRI and MRCP findings of hepatolithiasis. Although cross-sectional imaging with CT scan is a useful technique for screening for intrahepatic stones with a sensitivity of 80–90%, CT is less useful than MRCP for precise topographic localization of stones proximal to the biliary confluence [17]. MR cholangiography is a non-invasive test providing high quality multi projection images. It not only detects the stones, but also provides detailed information of the biliary anatomy, which is useful for surgical planning [16, 17].

Pre-operative abdominal MRI in case we presented showed that the gallbladder was surgically removed, while the common bile duct was accidentally resected, with stenosis of the non-Roux-en-Y hepaticojejunal biliary reconstruction. The intrahepatic biliary ducts of both hepatic lobes were dilated, with moderate dilatation noted on the extrahepatic biliary ducts (diameter of the left and right hepatic duct up to 7 mm). Intra and extra hepatic biliary ducts showed intense contrast enhancement signal primarily due to inflammation.

The proposed system for classification of hepatolithiasis, "Dong's Classification," is utilized to determine the best surgical approaches to resolve this disease (Table 1) [9]. Considering hepatectomy, the best candidates are patients with Type I and Type IIb hepatolithiasis. Patients with type II hepatolithiasis have a high risk of stone recurrence, thus the best solution is biliary stone extraction along with Roux-en-Y hepaticojejunostomy or hepaticocutaneous jejunostomy. For the treatment of type IIb hepatolithiasis with segmental atrophy and/or strictures of the intrahepatic bile ducts hepatectomy is considered the optimal approach. Complete removal of the affected lobe or segment is mandatory to preventing recurrence of bile stone and progressive liver disease including fibrosis and malignancy [18, 19].

Most often, removal of the affected hepatic segment(s) is the best possible surgical approach. Stone extraction, resolving of strictures and consequent biliary stasis, which is responsible for stone formation, is achieved by liver resection [20]. Criteria for segmental/lobe liver resection in hepatolithiasis include: 1. Hepatolithiasis limited to one lobe, particularly left-sided; 2. Cholangitis followed by atrophy, fibrosis and multiple abscesses; 3. Suspected the existence of associated cholangiocarcinoma and 4. Multiple intrahepatic stones with biliary strictures that cannot be treated percutaneously or endoscopically. Complete removal of the affected liver segment/lobe is mandatory to prevent recurrence and progressive disease [21, 22].

In case we presented, inadequate biliary-enteric anastomosis (absence of dysfunctional jejunum loop) lead to reflux of digestive juice into the intrahepatic bile ducts, followed by intermittent bacterial infection and recurrent cholangitis. Chronic recurrent infection led to biliary strictures, formation of intrahepatic stones and the increased risk for neoplasia in the form of cholangiocarcinoma. Therefore, we performed left hepatectomy followed by Roux-en-Y hepaticojejunostomy.

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Conflict of interest: None declared.

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Paper accepted

Table 1. Dong's classification of hepatolithiasis [9]

Type	Definition or content
Type I	Localized stone disease: unilobar or bilobar.
Type II	Diffuse stone disease.
IIa	No atrophy of the hepatic parenchyma or stricture of the intrahepatic bile ducts.
IIb	Segmental atrophy or/and stricture of the intrahepatic bile ducts.
IIc	Biliary cirrhosis and portal hypertension.
Additional Type E	Extrahepatic stones.
Ea	Normal sphincter of Oddi.
Eb	Relaxation of the sphincter of Oddi.
Ec	Stricture of the sphincter of Oddi.

Paper accepted

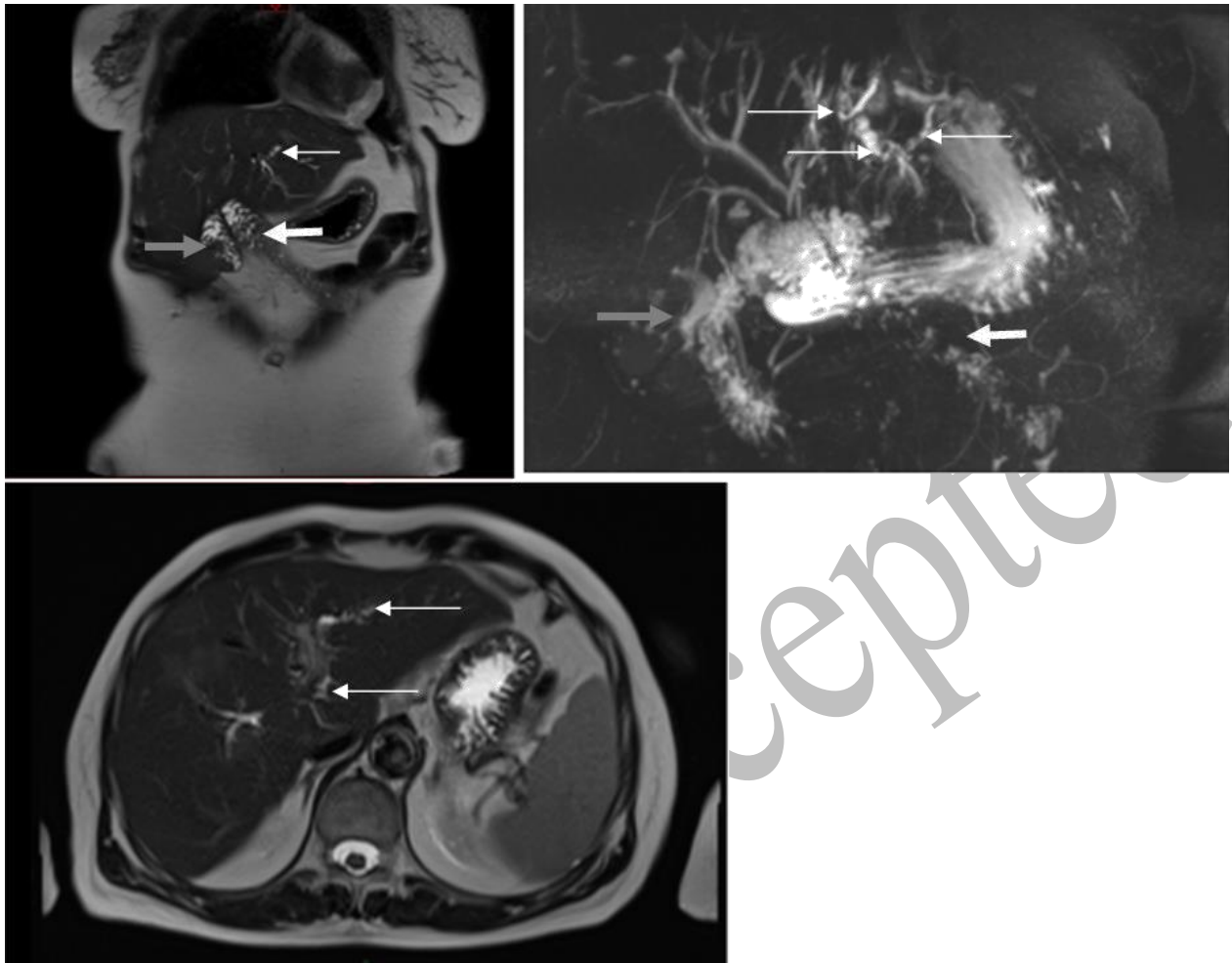


Figure 1. Abdominal magnetic resonance imaging and magnetic resonance cholangiopancreatography made before problem resolving operation: afferent jejunal limb of the non-Roux-en-Y hepaticojejunostomy (pointed by thick white arrow), efferent jejunal limb (pointed by gray arrow) and intrahepatic biliary calculi (pointed by thin white arrows)

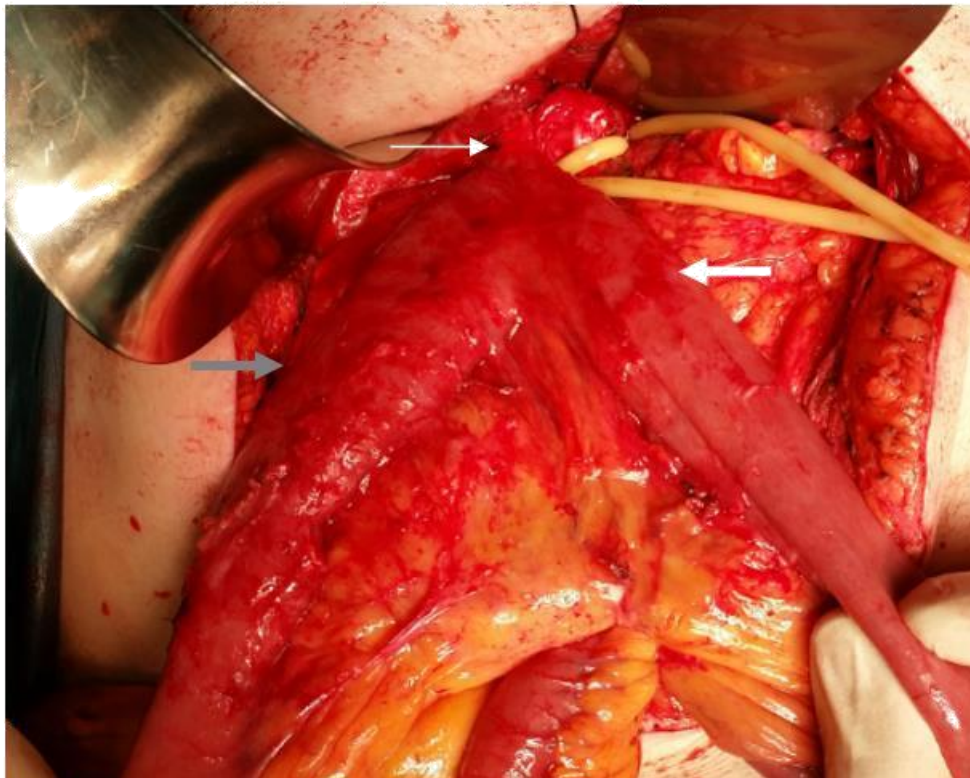


Figure 2. Cause of the disease: inadequate hepaticojejunostomy (non-Roux-en-Y) created at previous surgery (marked by thin white arrow); efferent jejunal limb (marked by gray arrow) and afferent jejunal limb (marked by white arrow)



Figure 3. Treatment decision: left hepatectomy with Roux-en-Y hepaticojejunostomy reconstruction

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Figure 4. Specimen photography made by pathologist: multiple intra-hepatic biliary stones

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