

## Choleperitoneum due to Intrahepatic Bile Duct Rupture - Case Report

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### Rezumat

#### *Coleperitoneu prin ruptură de căi biliare intrahepatice - prezentare de caz*

Perforațiile netraumatice ale căilor biliare extrahepatice sunt entități rar întâlnite, cu atât mai mult cele ce afectează căile biliare intrahepatice, exteriorizându-și conținutul biliar în marea cavitate peritoneală. Prezentarea unui astfel de caz a determinat autorii să facă o atentă trecere în revistă a cazurilor prezente în literatură. Se conturează faptul că obstrucția căii biliare principale de cauză litiazică determină, prin creșterea presiunii, dilatarea arborelui biliar, stază și infecție urmate de tromboza vaselor intramurale ce duce la perforația canalelor biliare, totul pe fondul unor malformații necunoscute ale căilor biliare intrahepatice.

**Cuvinte cheie:** peritonită biliară, duct biliar intrahepatic, ruptură spontană.

### Abstract

Non-traumatic perforations of the bile ducts are unfrequently encountered entities, all the more when they affect the intrahepatic bile ducts, exteriorizing their biliary content in the

great peritoneal cavity. Reporting such a case has determined the authors to perform a careful overview of the cases present in the literature. An observation that can be made based on these is that the obstruction of the main bile duct due to lithiasis determines, by pressure increase, the dilation of the bile system branches, all on the background of an unknown malformation of the intrahepatic bile ducts.

**Key words:** biliary peritonitis, intrahepatic bile duct, spontaneous rupture.

### Introduction

Congenital dilations of the intra- and extrahepatic bile ducts present a great anatomical diversity and, therefore, their clinical expression is also very variable. Malformations are autosomal recessively transmitted and can affect, according to the classifications of Alonso-Lej and Todani: 1. the intrahepatic bile ducts exclusively - Caroli disease; 2. the main hepato-choledochal duct; 3. complex and various associations of the first two variants. Most cases occur in childhood or adolescence, but there are cases with severe manifestations (peritonitis) which are clinically expressed in adults. Biliary peritonitis through intrahepatic bile duct rupture is rare, very few cases having been reported in the literature (1,2). These rare forms of peritonitis are unfortunately accompanied by high mortality, up to 50%, despite the recommended surgical treatment and postoperative intensive reanimation (3).

Further on we will be presenting a clinical case exemplifying the need of a complete and detailed exploration of the biliary tract in case of choleperitoneum, as well as the

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adaptation of the surgical cure to the type of anatomical anomaly discovered intraoperatively.

## Case Report

A female patient, 77 years of age, is admitted to the clinic for scleral and tegumentary jaundice, fever and chills, as well as moderate pain at the level of the right hypochondrium irradiating into the epigastrium. The onset of the jaundice occurred 7 days prior, when the patient described moderate pain in the entire superior abdomen. It is to be mentioned that the patient presented dyspeptic biliary dysfunctions for approximately 10 years, for which she had received symptomatic treatment.

Of the paraclinical test results at admission the following are of significance: white count (WC) =  $8300/\text{mm}^3$ , total bilirubin (TB) = 17.43 mg%, direct bilirubin (DB) = 13.90 mg%, ESR = 28 mm/h, with specific hepatic tests within normal limits. Imaging exams (ultrasound) revealed a gallbladder moulded to the calculi, a main bile duct (MBD) of approximately 19 mm in diameter presenting a calculus of 14-15 mm at the interior and non-homogenous structure of the pancreas. An endoscopic retrograde cholangiopancreatography (ERCP) is performed, manoeuvre which led to a moderate quantity of bile fluid leaking through the duodenal papilla. After catheterization of the papilla and introduction of the contrast agent one could observe a MBD increased in volume, with a mobile calculus at the interior (Fig. 1), no agent intake at the level of the gallbladder and no dilatation of the intrahepatic bile ducts. Due to the size of the calculus and the impossibility of performing lithotripsy, endoscopic papillosphincterotomy was no longer performed as intended. Patient was further cared for by electrolyte and metabolic rebalancing and administration of antibiotic therapy.

36 hours after ERCP the patient described intense abdominal pain, initially in the left hypochondrium, altered general state and peritoneal irritation syndrome. Abdominal ultrasound described a moderate quantity of fluid free in the peritoneal cavity. Patient's condition did not allow for her transportation for an emergency CT exam. Under the suspicion of acute post-ERCP peritonitis an emergency surgical intervention was initiated immediately. After opening the abdominal cavity approximately 2 litres of bile fluid could be observed, which was afterwards evacuated. The peritoneal cavity was carefully explored, without identifying the source of the choleperitoneum. Cholecystectomy was decided for; a bilio-biliary cholecisto-choledochal fistula was observed, a choledocholithotomy with removal of a migrated choledochal calculus of approximately 15 mm in diameter performed, and an external Kehr type biliary drainage was placed. Control intraoperative cholangiography on the Kehr tube indicated a satisfying passage of contrast agent at Oddi level, a good intake of the MBD, but also presence of the agent in the abdominal cavity, in the left subdiaphragmatic area. After re-administering the contrast agent (in greater quantity) via the Kehr tube its presence in the peritoneal cavity is confirmed (hepato-cholangio-peritoneal fistula) (Fig. 2). Enlargement of the

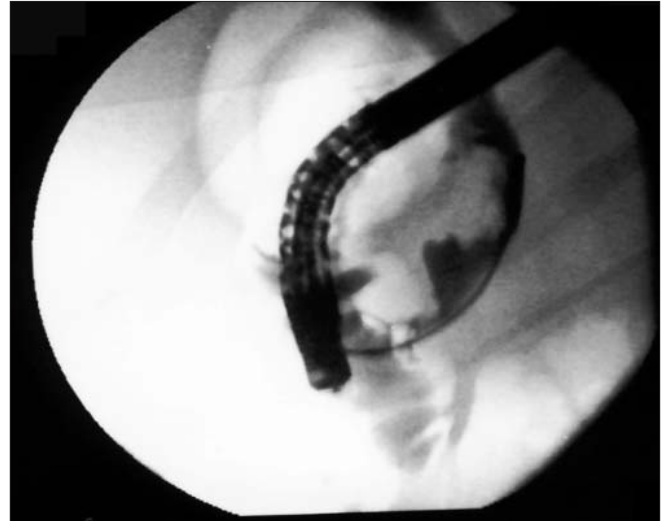


Figure 1. ERCP with image of the calculus in the MBD



Figure 2. Cholangiography via the Kehr tube. The arrow indicates the diffusion of the contrast agent in the left subphrenic space (hepato-cholangio-peritoneal fistula)

incision was decided for and at the level of the left hepatic lobe (segments 2-3), on the diaphragmatic aspect as well as at the level of the anterior margin and the visceral aspect of the liver, the presence of subglissonian bile lakes leaking in the great peritoneal cavity (multiple hepato-cholangio-peritoneal fistulae) was observed. The introduction of methylene blue via the Kehr tube served to better highlight these fistulae and the abundance of coloured fluid free in the peritoneal cavity (Fig. 3). Large calibre bile ducts (3-4 mm) were observed in these superficial areas, an anatomical anomaly, their presence being normal at a much more proximal level. In order to highlight their lumen, one such duct is catheterized with a no. 12 Benique (2 mm). Obturation of the ducts with non-absorbable threads passed in "U" or "X" was attempted, but unsuccessful. Under these circumstances, a "necessity" adjusted hepatectomy of segment 3 (Fig. 4), followed by



**Figure 3.** The arrow indicates subglissonian bile lakes leaking in the peritoneal cavity (hepato-cholangio-peritoneal fistulae). Introduction of methylene blue via the Kehr tube highlights their presence and the abundance of coloured fluid in the peritoneal cavity



**Figure 4.** Subglissonian bile lakes on the resection piece (segment 3 hepatectomy)

multiple drainage of the peritoneal cavity was decided for.

The enlarged bile ducts and the presence of subglissonian bile lakes at the level of the visceral aspect could be observed on the removed sample (Fig. 4). Histology exam showed: subcapsular microabscesses and thickening of the capsule, rich diffuse inflammatory infiltrate and also in the portal spaces, with predominance of polymorphonuclear neutrophils, bile stasis and hepatocytic lesions, dilated small calibre bile ducts lined with unistratified flattened epithelium and cystic dilation opening at the level of the hepatic capsule (Fig. 5).

The patient had a satisfying evolution for 72 hours post-operatively after which, despite the intensive treatment applied in order to attain hydroelectrolytic and metabolic balance, she developed severe hepato-renal failure and deceased.

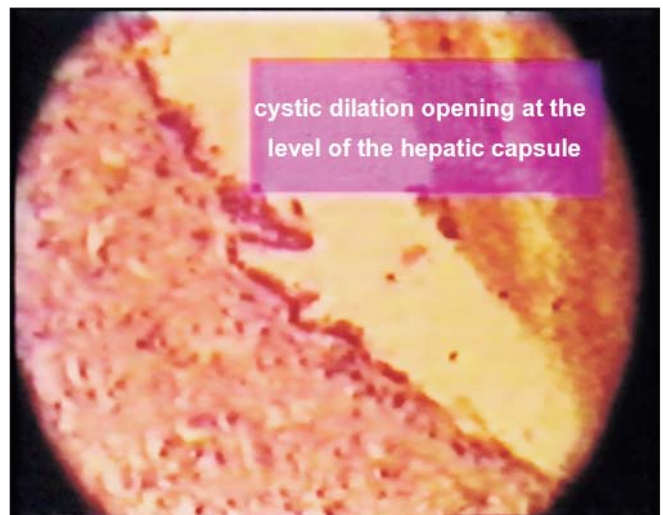
## Discussions

Classification type V (according to Todani) of congenital dilations of the bile ducts includes only cysts of intrahepatic bile ducts, which can be single or multiple. These segmentary multifocal cystic dilations are characteristic for Caroli disease and can be found in a single hepatic lobe (more frequently the left one), but can also affect the entire liver (1).

Spontaneous rupture of bile ducts in an adult is an extremely rare event, the first mention of such pertaining to Freeland in 1882, the entire British medical literature reporting only 22 cases. In 1912, McWilliams describes a single case of biliary peritonitis of hepatic origin out of 108 cases of choleperitoneum (3).

A series of mechanisms for spontaneous perforation of the bile ducts have been described in the literature, such as (3,4):

- increase in intraductal pressure through impaction of



**Figure 5.** Histopathology exam of the resection piece

calculi in the bile ducts or at the level of the Oddi sphincter, and blockage and/or spasming of the sphincter;

- subcapsular cholangitic microabscesses (confirmed by histology exam) which could reduce the ductal wall resistance to pressure increase;
- ductal intramural infection which could lead to thromboses of the parietal vessels followed by local necrosis and perforation at this level;
- autodigestion through pancreatic secretion reflux;
- congenital cystic dilations (Caroli disease), presenting a thin wall, lined by unistratified epithelium, which can predispose to perforation (5).

In the case we are reporting the most probable mechanism was pressure increase in the MBD due to the calculus in the biliary pathway, associated with the cholangitic process present at the level of the intrahepatic bile ducts, together leading to

perforation. Septic mechanism involvement in the perforation process is also supported by the patient's subsequent evolution, with a clinical picture suggestive of multiple organ failure, with renal and hepatic failure leading to exitus in the end.

Preoperative ultrasound allowed the detection of cholecysto-choledochal lithiasis, of MBD dilation, but did not reveal the intrahepatic cystic dilations. No additional preoperative imaging investigations were conducted, such as CT or MRI exam. Intraoperative cholangiography with contrast agent administered via the Kehr tube was relevant in diagnosing the peritonitis developed due to the perforation of the dilated intrahepatic bile ducts at the level of the left hepatic lobe, segment 3. Intraoperative confirmation was obtained through methylene blue cholangiography as well, in a manner similar to other authors, who however used indigo carmine as an agent instead (6).

Surgical treatment in our case consisted of cholecystectomy and choledocholitotomy, in performing a "necessity" adjusted hepatic resection of the left hepatic lobe, segment 3, other surgeons preferring an intermittent continuous suture with absorbable threads (such as Prolene, Vicryl, no. 3/0 or 4/0) to obliterate the perforations, followed by Kehr tube drainage of the MBD (3,7,8). When there is a form of Caroli disease located at the level of a single lobe (high frequency at the level of segment 3 of the left hepatic lobe) hepatectomy (left) can be recommended, and in diffuse forms hepatic transplant can be indicated (1).

Biliary peritonitis due to intrahepatic bile duct rupture is hard to diagnose preoperatively, rarely manifesting through jaundice, diffuse abdominal pain, more intense in the right hypochondrium, and signs of peritoneal irritation, patients sometimes being in shock; from a bioumoral point of view direct bilirubin predominates, presenting high values. It can be remarked that most intrahepatic biliary perforations occur in women, usually over the age of 60 and with various associated comorbidities, which determined an increased mortality rate of up to 30-50% (9,10), despite all measures taken for hydro-electrolytic and metabolic rebalancing (10,11).

## Conclusions

1. The patient presented cystic dilation in the sub-glissonian area of the left hepatic lobe (segment 3), associated with cholangio-biliary malformations.
2. These determined the spontaneous rupture of cystic areas with minimum resistance, followed by presence of bile in the peritoneal cavity.

3. This case presents the necessity of meticulous intra-operative evaluation of the bile ducts in cases of biliary peritonitis (particularly the left hepatic lobe), the diagnosis being established intraoperatively in most cases.
4. Surgical intervention can span from cystic dilation suture to radical interventions such as "necessity" partial hepatectomies, in localized forms of Caroli disease associated with perforation, or even liver transplant in diffuse forms.
5. Radical interventions are not always possible because of altered general state of the patients (biliary peritonitis).

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