

CASE REPORTS

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# Spontaneous perforation of common hepatic duct: an intraoperative surprise

Y. Kerkeni\*, F. Thamri, A. Zouaoui, B. Aziza and R. Jouini

## Abstract

**Background:** Pediatric spontaneous bile duct perforation is one of the rare causes of acute abdomen in infants. With a highly variable presentation, diagnosis and treatment can prove challenging. We report a case of spontaneous common hepatic duct perforation mistaken for appendicular peritonitis.

**Case presentation:** An otherwise healthy 3-year-old boy presented to the emergency department with complaints of abdominal pain, distention, bilious vomiting, and fever evolving for 4 days. Preoperative ultrasound was suggestive of appendicular peritonitis. Laparotomy revealed abundant biliary fluid, a distended gallbladder with thickened and inflammatory wall, and a macroscopically normal appendix. Intraoperative cholangiography showed a leak of contrast from the anterior wall of the common hepatic duct, 2–3 mm below the upper biliary confluence and 2 cm above the junction of the cystic duct to the common hepatic duct. A cholecystostomy was performed, and two external intraabdominal drains were placed. A retrograde cholangiogram was performed on postoperative day 21 showing no extravasation of contrast product. The patient was discharged on postoperative day 25.

**Conclusion:** Spontaneous perforation of the common hepatic duct is a rare phenomenon. It should be considered as a differential diagnosis in pediatric patients that present with unexplained peritonitis. The optimal method of diagnosis and management remains controversial.

**Keywords:** Biliary tract, Common hepatic duct, Spontaneous perforation

## Background

Spontaneous common bile duct perforation is an unusual cause of acute abdomen in children, most often diagnosed at the stage of biliary peritonitis [1]. It is potentially fatal, and preoperative diagnosis is very difficult since the clinical presentation is non-specific [1]. The most common presentation is abdominal distension and jaundice. Diagnosis is often made during surgery. Although the actual etiopathogenesis of this uncommon disease has yet to be determined, several hypotheses have been suggested. Mortality decreased from 40 to 12% as a result of improved care. However, in the lack of a clear consensus, surgical management remains variable [2, 3].

Herein, we report a rare case of spontaneous common bile duct perforation in a 3-year-old boy mistaken for appendicular peritonitis. Through our case, we detailed our surgical management and we reviewed the literature on the surgical options for this rare condition.

## Case presentation

An otherwise healthy 3-year-old boy presented to the emergency department with complaints of diffuse abdominal pain, distention, bilious vomiting, and fever evolving for 4 days. On examination, the patient was febrile, dehydrated, and hemodynamically stable. Abdominal examination revealed abdominal distension with mild tenderness. Abdominal ultrasound was suggestive of appendicular peritonitis. It showed peri-appendiceal fat stranding, cecal apical wall thickening, and echogenic intraperitoneal effusion of great abundance without visualization of the appendix. A midline

\*Correspondence: yosrakerkeni@yahoo.fr  
Department of Pediatric Surgery "A", Children's Hospital, Tunis El Manar University, Boulevard du 9-Avril 1938, 1006 Tunis, Tunisia

laparotomy was performed revealing an abundant bilious effusion, a distended gallbladder without any calculus and perforations, and a macroscopically normal appendix. Intraoperative cholangiography through the gallbladder showed a leak of contrast from the anterior wall of the common hepatic duct, 2–3 mm below the upper biliary confluence and 2 cm above the insertion of the cystic duct (Fig. 1).

Biliary diversion with a cholecystostomy tube was performed. A drain was placed under the liver and a wavy blade at the level of the right iliac fossa. The postoperative course was uneventful. A retrograde cholangiogram was performed on postoperative day 21 showing no extravasation of contrast product (Fig. 2). Enteral feeding was introduced on day 15 post-operatively and was well tolerated. The cholecystostomy tube was removed after the cholangiography. The patient was discharged on postoperative day 25. He is doing well on follow-up.

### Discussion

Spontaneous or idiopathic bile duct perforation is a rare entity, mostly seen in children less than 4 years of age [1]. The actual etiopathogenesis is unknown and remains obscure. Congenital weakness of the common hepatic duct, trauma, choledochal cyst, viral infection,



**Fig. 1** Intraoperative cholangiography showing the leak of contrast from the anterior wall of common hepatic duct



**Fig. 2** Postoperative retrograde cholangiogram showing a permeable normal-sized bile duct with no extravasation of contrast product

intramural thrombosis, necrotizing enterocolitis, pancreatitis, diverticulum in the common hepatic duct, stenosis of the common hepatic duct, and anomalous union of the pancreatico-biliary ductal system are all believed to be involved [2]. Usually, the site of weakness is at the junction of the cystic duct and the common hepatic duct. The site of the perforation at the common hepatic duct is exceptional and has been reported in few adult cases [3]. To our knowledge, this is the second pediatric case which reported such location [3].

As for our patient, the preoperative diagnosis is extremely rare given the absence of specific signs. Most common clinical presentations are abdominal distension, jaundice, symptoms of peritonitis, septicemia, and symptoms of biliary tract disease [3]. Abdominal ultrasound visualizes the defect in 38.4% of cases. An abdominal CT scan helps to make the diagnosis in 69.2% of cases [4].

Controversy exists regarding the optimal surgical modality. Most of the authors use T-tube drainage

**Table 1** Literature review of surgical management of common bile duct perforations in children

Study	Management	Number of patients
Ozdemir et al. [6]	T-tube drainage of the bile duct	5
Howard et al. [16]	Cholecystenterostomy using a Roux-en-Y loop of the jejunum	2
Sai Prasad et al. [10]	Excision of the gall bladder and common bile duct + Roux-en-Y hepaticojejunostomy	1
	Cholecystostomy drainage	1
Haller et al. [7]	T-tube drainage of the common hepatic duct	3
	Cholecystostomy tube + T-tube drainage of the bile duct	1
	Surgical repair of the bile common duct	1
Sunil et al. [3]	Surgical repair of the common hepatic duct over T-tube	1
Fukuzawa et al. [11]	Cholecystostomy	13
	T-tube drainage of the bile duct	2
Lloyd and Mickel [17]	Choledocoduodenostomy	1
	Surgical repair of the bile common duct	1
Xanthakos et al. [12]	Cholecystectomy + intraabdominal drainage	1
Ford WD [18]	Roux-en-Y hepaticojejunostomy	1
Charu Sharma et al. [15]	Insertion of a metal fully covered stent with the tip beyond the leak	1
Chardot et al. [9]	Hepaticojejunostomy by means of a Roux-en-Y loop + cholecystostomy + external drainage of the subhepatic area	1
	Hepaticojejunostomy by means of a Roux-en-Y loop + cholecystectomy	2
	Hepaticojejunostomy by means of a Roux-en-Y loop + cholecystectomy + T-tube drainage of the bile duct	2
	Cholecystostomy + external drainage of the subhepatic area	2
	T-tube drainage of the bile duct + external drainage of the subhepatic area	1
Mirza et al. [13]	Surgical repair of the bile common duct + cholecystostomy tube + external drainage of the subhepatic area	3
Kohli et al. [14]	Cholecystectomy + Roux-en-Y hepaticojejunostomy	1
Upadhyaya et al. [8]	Lavage + subhepatic and pelvic drain	2
	Lavage + subhepatic and pelvic drain + T-tube insertion + definitive repair after 6 weeks type Roux-en-Y loop	3
	Cholecystostomy followed by definitive repair after 3 months type Roux-en-Y loop	1

placed through the bile duct and a subhepatic drain [3, 5–8]. T-tube drainage ablation is usually done 3 weeks post-operatively. Other techniques are described such as biliary diversion using a cholecystostomy tube with or without placing a Kehr drain [1, 3, 9–14], surgical repair of the common hepatic duct, and cholecystectomy with intraabdominal drainage [12]. Insertion of a metal fully covered stent with the tip beyond the leak [15] and hepaticojejunostomy by means of a Roux-en-Y loop were also described as surgical modalities [8–10, 14, 16–18]. Table 1 summarizes the different surgical approaches used in the treatment of common bile duct perforations described in the literature. The particularity of our patient is that we opted for a non-invasive treatment using a single biliary diversion with cholecystostomy tube and drainage of the peritoneal cavity. The cholecystostomy drains the bile, thus reducing the pressure at the level of the common hepatic duct, and facilitates its cicatrization to healing.

## Conclusion

Spontaneous perforation of the common hepatic duct is rare. The optimal method of diagnosis and management remains controversial. Intraoperative cholangiography is essential to locate the perforation. Several therapeutic methods are used. Biliary diversion via a cholecystostomy tube is simple and safe and proves to be an effective method of treatment.

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## Authors' contributions

KY drafted the manuscript. JR supervised the study. TF, ZA, and BA performed perioperative management of the patient. The authors read and approved the final manuscript.

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## Declarations

### Ethics approval and consent to participate

Not applicable.

### Consent for publication

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### Competing interests

The authors declare that they have no competing interests.

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