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Case Report

Spontaneous Rupture of Common Hepatic Duct in an Infant, a Rare Entity

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Abstract

Pediatric spontaneous biliary duct perforation (SBDP) represents a rare clinical condition. The vast majority of pediatric cases typically manifest around 6 months of age; however, the initial onset of this condition has been documented to occur as early as 25 weeks of gestation and, conversely, as late as 7 years postnatally. Despite the encouraging fact that the condition is treatable with appropriate intervention, the often non-specific nature of its associated symptoms and signs can unfortunately result in a significantly delayed diagnosis. This delay, in turn, carries the potential for the development of severe, life-threatening conditions such as biliary peritonitis and sepsis. Therefore, it is of paramount importance for clinicians to establish an early suspicion and achieve a prompt diagnosis to mitigate these risks. An exceptionally uncommon variant within the spectrum of spontaneous biliary duct perforation is the spontaneous rupture of the common hepatic duct. This report presents an illustrative case of a 5-month-old male infant who experienced a spontaneous perforation of the common hepatic duct, leading to the formation of a biloma. This diagnosis was successfully established pre-operatively through the utilization of a contrasted abdominal computed topography (CT) scan. Subsequently, the patient underwent a primary surgical repair of the identified perforation site, in addition to percutaneous drainage facilitated by a biliary T-tube. The postoperative recovery period was remarkably uneventful, and the T-tube was successfully removed on the 12th postoperative day. Six weeks following discharge from the hospital, the patient exhibited entirely satisfactory progress, with a complete absence of any discernible complications.

Keywords

Spontaneous Perforation of Common Hepatic Duct, Biloma, Pediatric Surgery

1. Introduction

Bile duct perforation in the pediatric population is relatively rare, with only a limited number of cases of spontaneous bile duct perforation due to common hepatic duct perforation reported in the literature.

The majority of pediatric cases arise within the first year of life, although onset may occur as early as 25 weeks of gestation and can extend up to 15 years post-birth. The median age at presentation is 4 months, with a male-to-female ratio of 2:1 [1].

The reasons for the occurrence of SBDP in children remain poorly understood. Various theories may contribute, including congenital weakness of the bile duct, bile duct obstruction, and congenital anomalies of the pancreaticobiliary duct [2].

Perforation predominantly occurs at the junction of the

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cystic and common hepatic ducts, with only a limited number of cases reported at other locations within the biliary tree [1, 2].

Abdominal distension, ascites, and jaundice represent the most common presentations; however, additional symptoms can include pyrexia, localized or generalized peritonitis, and septic shock, with or without signs of biliary tract disease [2-10].

The management of SBDP varies from conservative strategies involving broad-spectrum antibiotics to interventions such as Endoscopic Retrograde Cholangiopancreatography (ERCP), percutaneous drainage, or more complex invasive surgical procedures [11-13].

2. Case Presentation

A 5-month-old male patient presented with low-grade fever and watery diarrhea for a duration of 2 weeks. The mother reported that he had chickenpox three weeks before admission, subsequently accompanied by recurrent non-bilious vomiting episodes. It was associated with hypoactivity, poor oral intake, and persistent crying. The vomiting ceased after one week. However, the child developed a fever that initially responded to antipyretics, along with watery non-bloody diarrhea and progressive abdominal distention, accompanied by facial pallor and scleral jaundice. The mother reported a weight loss of approximately 1 kg during this period. The infant has no previous surgical interventions or history of trauma. A history of asymptomatic atrial septal defect and ventricular septal defect was noted during a prior child's wellness visit.

On physical examination, the patient appeared unwell, exhibited pallor, mild jaundice, and displayed irritability. Vital signs showed a blood pressure of 102/56 mmHg, a heart rate of 140 bpm, a respiratory rate of 50 breaths per minute, and a temperature of 38.5 ℃. The abdomen revealed tenderness upon palpation, mild distension, slightly tense, a protruded but reducible umbilicus, and no signs of organomegaly. The right hip exhibited persistent flexion and tenderness during passive motion. Blood samples were collected and sent to the laboratory for analysis. Intravenous fluid replacement therapy was started.

The complete blood count (CBC) indicated a normal age-correlated white blood cell (WBC) count of 14,000 cells/mm ^{3} Hemoglobin level measured 7.4 g/dl, with a mean corpuscular volume (MCV) of 49.5 fl. Hemoglobin Electrophoresis confirmed the diagnosis of beta thalassemia minor. The acute phase reactants came out as follows: The platelet count measured 786 x 10^9/L, C-reactive protein was 15.93 mg/dl (normal range up to 0.5 mg/dl), the erythrocyte sedimentation rate (ESR) was 45 mm/hr (normal range less than 13 mm/hr), and the ferritin level was 285.5 ng/ml (elevated). Liver function tests were normal, with the exception of direct hyperbilirubinemia (total bilirubin at 68.7 umol/L and direct bilirubin at 52.4 umol/L) (normal total bilirubin is less than 17.1 umol/L and direct bilirubin is less than 5.1 umol/L). Serial lactate dehydrogenase (LDH) levels showed a consistent increase, ranging from 332 to 812 U/L (normal levels up to 300 U/L). Lipase, amylase, and alpha-fetoprotein levels, along with kidney and thyroid function tests, were all within normal ranges. Both blood and urine cultures yielded negative results. The parents declined the lumbar puncture; therefore, we administered empirical antibiotic treatment.

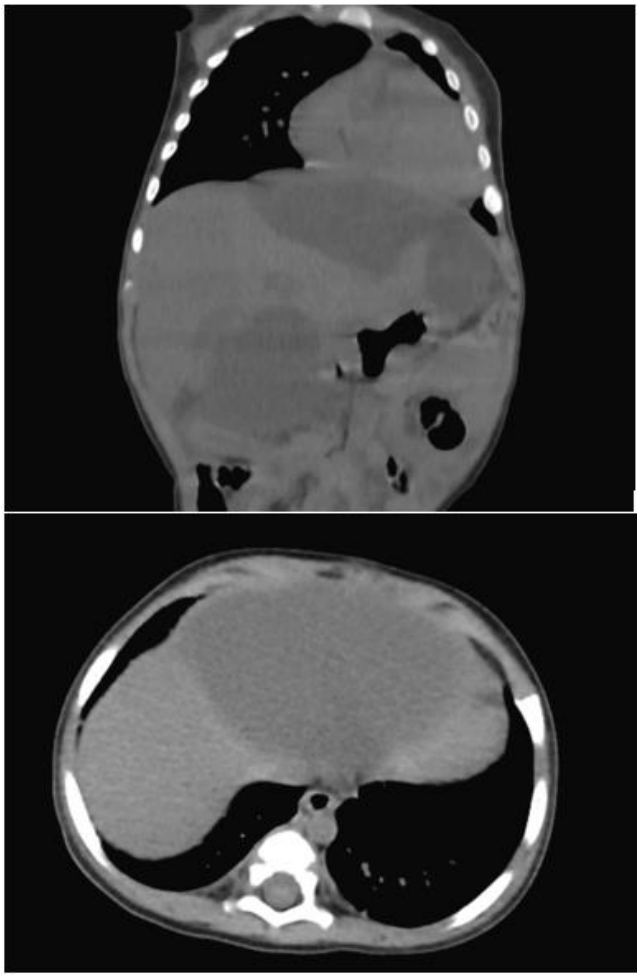

Figure 1. *A large subhepatic encysted fluid collection and a multiseptated collection in the gallbladder region.*

The abdominal ultrasound revealed mild hepatomegaly accompanied by a large cystic structure with septations in the left lobe of the liver. The gallbladder appeared contracted, with no evidence of pericholecystic edema. Both the common bile duct and intrahepatic biliary duct were not dilated, and the hepatic veins and arteries exhibited normal flows and velocities. However, the exam was suboptimal because of the patient's irritable condition. An abdominal computed tomography (CT) scan revealed a large left subcapsular septated hepatic cyst measuring 10.8 x 4.2 x 5.3 cm, exerting significant mass effect on the left lobe of the liver. Additionally, there was a large subhepatic encysted fluid collection measuring 5.5 cm in diameter and a multiseptated collection in the

gallbladder region measuring 4.3 cm in diameter (Figure 1). The pancreas, spleen, and kidneys exhibited normal findings, and there was an absence of intrahepatic or extrahepatic biliary dilatation.

The echocardiogram revealed a 4 mm subpulmonic ventricular septal defect and a 2 mm atrial septal defect. An ultrasound of the right hip and knee showed no abnormalities.

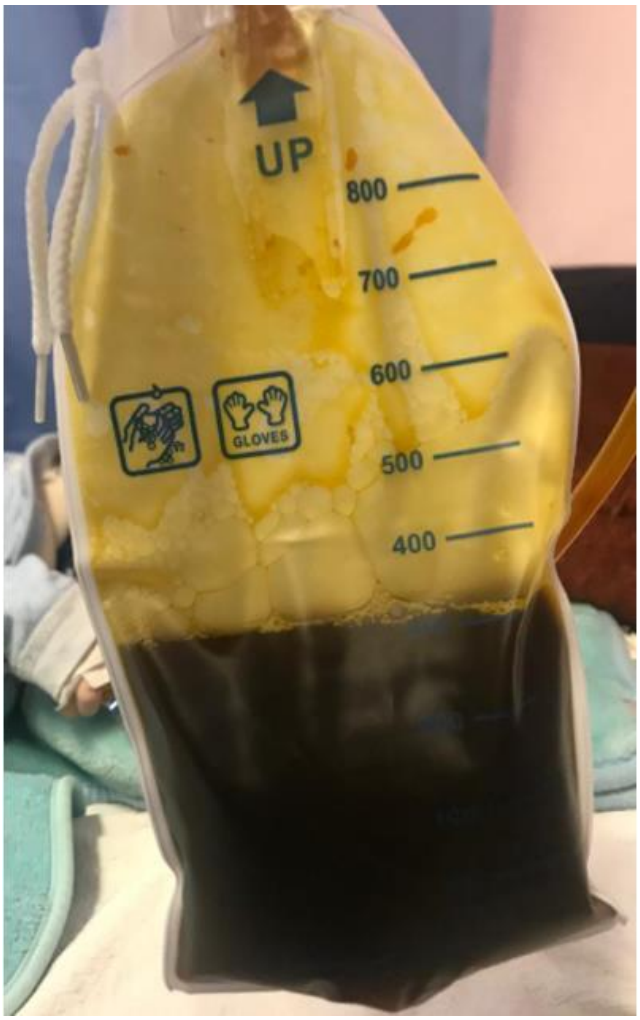

Figure 2. *Biliary drainage from the percutaneous drain.*

A diagnostic ultrasound-guided aspiration was performed on the largest collection, yielding approximately 100 cc of bilious fluid, which was subsequently sent for further evaluation. The bilirubin concentration in ascitic fluid was measured at 279.1 mg/dl, significantly exceeding the normal range of 0.7 - 0.8 mg/dl, thereby confirming the diagnosis of a biloma. The cytological analysis revealed a combination of inflammatory cells, including plasma cells, lymphocytes, and a few macrophages, while the cultures showed no bacterial growth. The patient underwent placement of a percutaneous drain and had been kept on broad-spectrum antibiotics. However, due to persistent symptoms and ongoing fluid drainage (Figure 2), surgical exploration was scheduled.

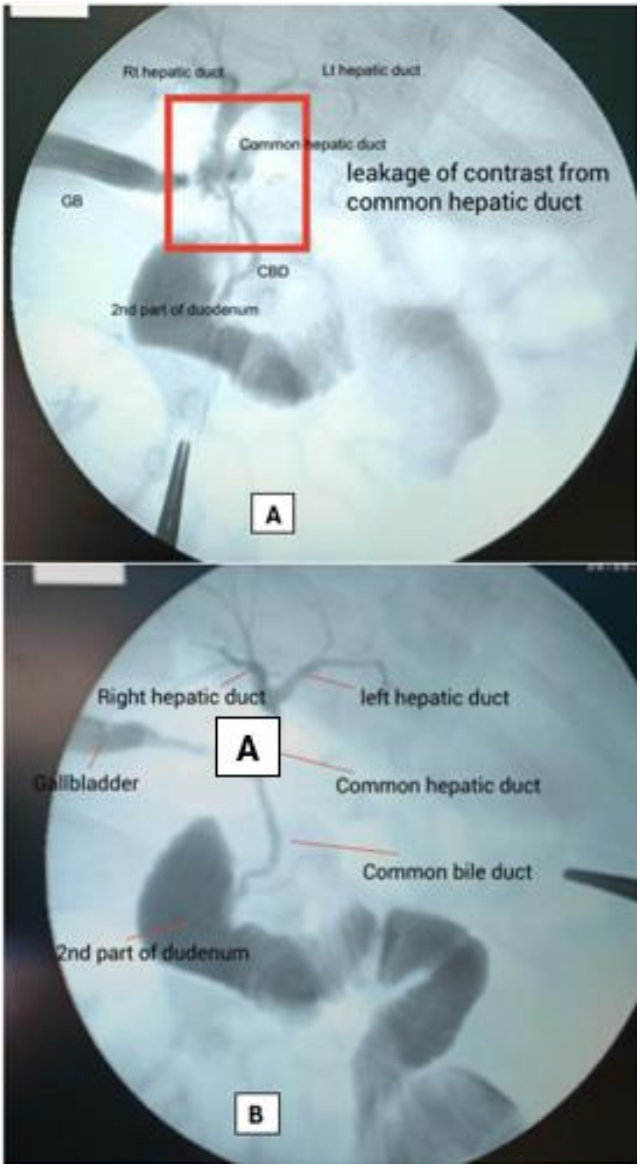

Figure 3. *Intraoperative cholangiogram. A: Contrast leakage from the common hepatic duct. B: Repeat intraoperative cholangiogram showed no contrast leakage from the previous site.*

An exploratory laparotomy was performed via an upper transverse incision at the peritoneal cavity's entrance near the percutaneous drain, located beneath the liver's inferior surface and extending to the porta hepatis. The adjacent bowel exhibited bile staining. The percutaneous drain was removed, and an intraoperative cholangiogram was performed (Figure 3A), revealing contrast leakage from the common hepatic duct. Examination of the common bile duct, common hepatic duct, right hepatic duct, and left hepatic duct identified a 5mm perforation on the posterior wall of the common hepatic duct. The perforation site was repaired, and a repeat intraoperative cholangiogram showed no contrast leakage from the previous site (Figure 3B). Subsequently, a cholecystectomy was performed via the cystic duct stump, and a T tube was inserted.

A subhepatic drain was inserted. The postoperative period was without complications, and the patient was permitted oral

intake on the second day following surgery. The subhepatic drain was removed on day 5. A cholangiogram conducted on day 11 indicated no leakage of contrast at the primary repair site, leading to the removal of the T tube. The patient was subsequently discharged the following day. The patient was doing well during the follow-up assessment conducted two months post-discharge.

3. Discussion

The first reported case of bile duct perforation was in 1882 by Freeland in adults [14], while the first case in infants was reported by Dijkstra in 1932 [15]. Spontaneous perforation of the biliary system in pediatric patients is rare, with the most frequent location occurring at the junction of the cystic and common hepatic duct. Isolated perforations in the common hepatic duct, as observed in our case, are relatively rare, with only a limited number of pediatric cases documented in the literature. Sunil reported three cases, while Yan reported one case involving children [2, 8].

Our case presented with jaundice, progressive abdominal distension, and vomiting, consistent with symptoms documented in the literature [2, 3]. Perforations can lead to peritonitis [9] and may advance to septic shock, requiring the administration of empiric antibiotics. SBDP exhibits a higher prevalence in males, as evidenced in our case, with a median age of 4 months, which aligns with the 5 months observed in our case [1].

Perforations in pediatric patients may occur as a result of stones, trauma, or congenital abnormalities. The precise pathophysiology of idiopathic perforations remains incompletely elucidated [2]. No definitive secondary cause was identified in our case.

The main goal of SBDP treatment is to drain the biliary ascites, prevent complications and restore biliary tract patency [2]. SBDP may be managed either operatively or conservatively. Conservative management includes broad-spectrum antibiotics and drainage, which can be performed percutaneously or via ERCP [11-13]. ERCP is less invasive than surgery and serves both diagnostic and therapeutic purposes; endoscopic management of the leak may involve the placement of biliary stents [11], in our case, ERCP was unavailable, and the patient was referred to for surgical intervention. Surgical intervention can be through laparotomy, laparoscopy, or robotics [2, 11-13], in our case the patient underwent laparotomy during which the perforation site was identified and repaired.

Early detection and management typically yield a favorable prognosis [12]. The patient remained asymptomatic upon follow-up two months later.

4. Conclusion

SBDP is a rare yet significant differential diagnosis in children and infants presenting with biliary ascites. The symptoms are typically nonspecific; however, if not identified promptly, they may advance to severe peritonitis, underscoring the importance of early recognition. The prognosis is favorable with either conservative or surgical management approaches.

Abbreviations

SBDP	Spontaneous Biliary Duct Perforation
CT	Computed Topography
ERCP	Endoscopic Retrograde Cholangiopancreatography
Kg	Kilogram
CBC	Complete Blood Count
WBC	White Blood Cells
LDH	Lactate Dehydrogenase
MCV	Mean Corpuscular Volume
ESR	Erythrocyte Sedimentation rate

Conflicts of Interest

The authors declare no conflicts of interest.

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