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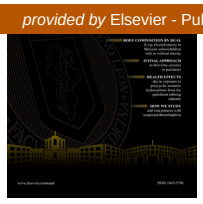

SCIENTIFIC LETTERS

Neonatal spontaneous biliary perforation: Case report

F. Reyna-Sepúlveda, A. Esparza-González, V. Govea-Ortiz, G. Martínez-Flores*, F. Montes-Tapia, U. Garza-Luna, G. Muñoz-Maldonado

Autonomous University of Nuevo León, General Surgery Department at the ''Dr. José Eleuterio González'' University Hospital, Monterrey, Mexico

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KEYWORDS
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Abstract
Objective: Spontaneous biliary perforation in neonates is rare. The etiology of this pathology is idiopathic and various management strategies ranging from non-operative treatment to complex operations, such as biliary-enteric reconstruction, are performed, with few reported outcomes.
Case report: A 3-week-old female, born at term, presented fever, abdominal distension, and acholic stool. An ultrasound was performed, which revealed generalized ascites and a poorly-defined collection. An emergency laparotomy confirmed perforation in the distal common bile duct and a biliary-enteric-anastomosis was performed.
Discussion: Wide drainage has been reported as the best initial management strategy for spontaneous biliary perforation, although it depends on the patient's clinical status and intraoperative findings.
Conclusions: Spontaneous infantile biliary perforation is rare. Main management is wide drainage with, most perforations being resolved in 2 weeks.

Introduction

Spontaneous biliary perforation in neonates is rare, even more so in the absence of a choledochal cyst.[1] No more than 150 cases have been reported, with an incidence of 1.5 out of every 1,000,000 live births.[2] The etiology of this pathology is idiopathic; however, some cases are associated with pancreatic malunion[3] or distal obstruction by stones or atresia.[4] A prompt diagnosis should be made, as it is the most common cause of surgical jaundice in infants[5] and severe sepsis can develop in a few days or even hours. Management includes non-operative treatment with antibiotics, endoscopic retrograde cholangiopancreatography (ERCP), percutaneous drainage[6] and biliary intestinal reconstruction. Biliary fistula and bile duct stenosis are the most common complications. We report a 3-week-old female

* Corresponding author at: Servicio de Cirugía General del Hospital Universitario ''Dr. José Eleuterio González'' de la Universidad Autónoma de Nuevo León, Av. Francisco I. Madero pte.y Av. Gonzalitos s/n col. Mitras Centro, Monterrey, Nuevo León, Mexico.
E-mail address: joseguillermo2506@hotmail.com (G. Martínez-Flores).

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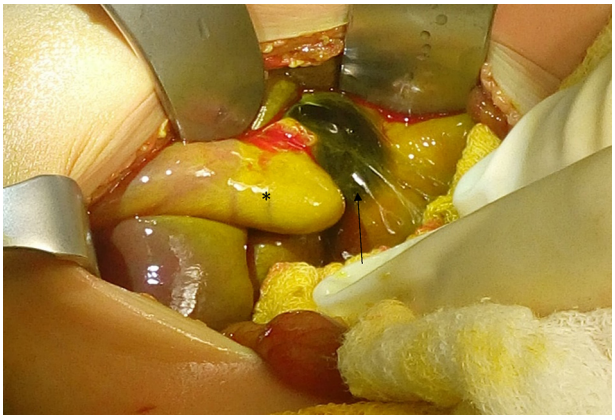

Figure 1 Contained bile perforation (black arrow) and gallbladder (asterisk).

neonate with spontaneous perforation and biliary reconstruction.

Case report

History and presenting condition

A 3-week-old female, born at term with no previous important obstetric history, presented with fever, abdominal distension, acholic stool and a history of diarrhea of 1-week duration. The abdomen was tender with no signs of sepsis. Laboratory studies showed a total bilirubin of 4.5 mg/dL with direct bilirubin of 2.1 mg/dL, transaminases were slightly increased, alkaline phosphatase was 177U/L with a normal blood count. Ultrasound revealed generalized ascites and a poorly-defined collection of 7.9 mL in a sub-hepatic location, with no evidence of dilation of the bile duct or presence of a choledocal cyst. Transhepatic cholangiography showed a leak of bile at the choledocal duct without observing material passing into the duodenum.

Intervention

An emergency laparotomy was performed, which revealed bilious ascites with inflammatory adhesions and an infrahepatic collection adjacent to the hepatoduodenal ligament (Fig. 1). An intraoperative cholangiogram via the gallbladder confirmed the leak with distal obstruction of the common bile duct and a perforation of 1 mm in the distal choledocal duct. Biliary reconstruction with biliary-enteric-anastomosis was performed.

Follow-up and outcome

Postoperatively, the patient had a positive evolution with nasogastric feeding. The drain was removed on day 7 and follow-up was continued by the neonatal intensive care unit.

Discussion

As mentioned before, this is an uncommon clinical complication. Jeanty et al.[2] reported 94 cases of biliary perforation, of which a bilioma was found in 24%, which was commonly mistaken for enteric duplication, a pancreatic pseudocyst or a choledocal cyst. The most common location for the perforation was at the junction of the cystic duct and common hepatic duct. Regarding management, they reported that 6% used non-operative treatment, 56% used surgical drainage and 33% used biliary reconstruction. Non-operative treatment included the use of broad-spectrum antibiotics only. From the surgical drainage group, 15% subsequently required delayed biliary-enteric anastomosis. Biliary fistula was the more common complication (10%); common bile duct obstruction due to stones[7] was more common than atresia. Importantly, wide drainage has been reported as the best initial treatment for spontaneous biliary perforation in order to control the leak without biliary reconstruction in 85% of patients, avoiding complex and difficult dissection in an inflamed operative field and minimizing additional injury. Diagnosis is a challenge, because standard studies such as ultrasound are not conclusive. We had to use interventional radiology resources. In this case, the surgeon's experience provided a permanent solution for the perforation without the need for drainage, which reduced the recovery time, minimized the risk of sepsis and avoided delayed reconstructive surgery. The limit of this case is the need to perform more prospective studies to accurately compare non-operative versus operative management.

Conclusions

Spontaneous infantile biliary perforation is rare. Main management is wide drainage, with most perforations being resolved in 2 weeks. Simple drainage or resection should depend on the patient's clinical status and intraoperative findings. Distal common bile duct obstruction must be evaluated.

Conflict of interest

The authors have no conflicts of interest to declare.

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