Formosan Journal of Surgery (2015) 48, 133–136

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CASE REPORT

Spontaneous biliary perforation in a 7-year-old child

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Received 10 February 2015; received in revised form 7 March 2015; accepted 26 March 2015
Available online 18 June 2015

KEYWORDS
bilious;
peritonitis;
spontaneous biliary perforation

Abstract Spontaneous biliary perforation is a rare cause of surgical jaundice in infants and children. It typically presents in infants aged 1–12 weeks. Fewer than 200 cases have been reported in the medical literature. The lack of knowledge regarding the pathogenesis of spontaneous biliary perforation and its extreme rarity makes preoperative diagnosis and its management difficult. We report a case of a 7-year-old boy with spontaneous biliary perforation, who presented with clinical features of acute peritonitis. Cholecystectomy with simple drainage by using a T-tube resulted in satisfactory recovery. We suggest that in a jaundiced child with symptoms of abdominal pain, distension, vomiting, acholic stools, and a previous anicteric period of good health, the diagnosis of spontaneous biliary perforation should be considered if there is a triad of: (1) signs of peritonitis; (2) absence of pneumoperitoneum on erect abdominal radiography; and (3) bilious abdominal paracentesis under ultrasonographic guidance. Routine abdominal paracentesis is not recommended for infants and children. Although there are several surgical options for the disease, cholecystectomy, flushing of the bile duct and external biliary drainage, constitute the most favorable treatment. Pediatricians and surgeons should consider this disease because early surgical intervention is the only option for ensuring a favorable prognosis.

Conflicts of interest: All contributing authors declare no conflicts of interest.

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http://dx.doi.org/10.1016/j.fjs.2015.03.001

1. Introduction

Spontaneous biliary perforation is a rare surgical condition in children.[1] It was first reported in 1932 by Dijkstra.[1] Based on a literature review, fewer than 200 cases have been reported in the medical literature.[1] This condition typically presents in infants aged 1—12 weeks and is extremely rare in late infancy and childhood.[1–3] The cause is idiopathic.[2] Trauma and Choledochal cyst are two differential diagnoses that are ruled out in a child presenting with spontaneous biliary perforation.[1–3] Preoperative diagnosis and management of the condition is difficult. Early and limited surgical intervention to achieve adequate external biliary drainage produces optimal results.[4] Herein, we report a rare case of a 7-year-old boy with spontaneous biliary perforation and discuss the dilemma in its management.

2. Case report

A 7-year-old boy presented to our emergency room with acute abdominal pain, abdominal distension, vomiting, and acholic stools for 2 days. He exhibited an absence of prodrome and was previously well with no relevant medical history suggestive of hepato-biliary disease. On examination he had a feeble pulse, pulse rate of 140 beats/min and a respiratory rate of 45 breaths/min with icterus. Abdominal signs of generalized peritonitis were present, with more tenderness in the right hypochondrium as compared to other quadrants of the abdomen. Laboratory values revealed hemoglobin of 11 gm/dL and total leukocyte count of 17,000/mm3 or 17,000/μL. Liver function tests revealed total bilirubin of 2.2 mg/dL (conjugated hyperbilirubinemia) and alkaline phosphatase at 249 IU/L (high). Serum transaminases were normal. There was no free gas under the domes of the diaphragm on erect abdominal radiography. Abdominal ultrasonography (USG) revealed fluid collection around the common bile duct (CBD), gallbladder, and in the rest of the peritoneal cavity. No calculus was demonstrated in the biliary tree. Abdominal paracentesis under USG guidance revealed bilious ascites (with high bilirubin in the ascitic fluid). Computed tomography (CT) was unavailable. The patient was resuscitated, preoperative optimization was performed, and he was subjected to laparotomy, because of the unavailability of a laparoscopic facility.

Laparotomy showed bilious ascites (~200 mL, mostly in the Morrison's pouch) with mucus flakes over the gallbladder and in the Calot's triangle (Fig. 1). A perforation was observed in the CBD, immediately inferior to its junction with the cystic duct (Fig. 1). The patient underwent cholecystectomy, thorough abdominal lavage, and external biliary drainage with a T-tube after flushing of the bile duct. The rest of the abdominal viscera were normal. The patient was administered third generation cephalosporin in the postoperative period.

On the 7th postoperative day, the patient developed central abdominal pain, distension and episodes of vomiting that were initially managed conservatively. There was a recurrence of obstructive symptoms on the 16th postoperative day, which necessitated a second exploratory laparotomy. Numerous interloop small bowel adhesions were revealed. Adhesiolysis was performed. The postoperative period was uneventful. The T-tube was clamped intermittently, beginning 3 weeks after the first surgery. T-tube cholangiogram was performed at 4 weeks and

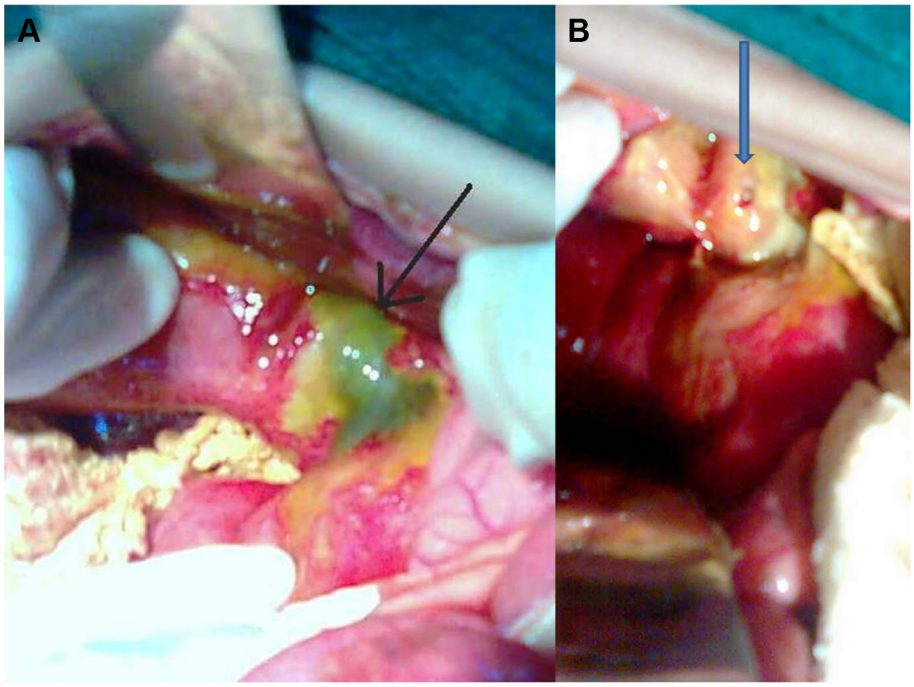

Figure 1 (A) Intraoperative photograph showing mucus flakes over the gallbladder and in the Calot's triangle, as indicated by the black arrow. (B) Intraoperative photograph showing perforation of the common bile duct, as indicated by the blue arrow (observed after removing the mucus flakes).