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Management of choledocholithiasis in an infant

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ABSTRACT

Choledocholithiasis is a rare diagnosis in the infant population with poorly defined optimal management. Endoscopic and operative interventions are both technically challenging. Endoscopic retrograde cholangiopancreatography (ERCP), common bile duct exploration and laparoscopic cholecystectomy are typically safe and successful when performed. However, choledocholithiasis has been reported to spontaneously resolve in up to 60% of cases in neonates and infants. We present five cases of choledocholithiasis in infants and a comprehensive review of the literature. Based on our review and experience, we make a recommendation that clinicians should proceed with a selective approach in the management of this disease based on severity of symptoms, co-morbidities, and the available surgical and endoscopic capabilities of each institution.

1. Introduction

While there is not yet a gold standard in the management of choledocholithiasis (CDL) in the pediatric population, the treatment approach has become clearer in recent years. In centers where pediatric ERCP is offered, the endoscopic approach for relieving biliary tract obstruction is safe and effective and is often employed as the first option [1–4]. Likewise, laparoscopic common bile duct (CBD) exploration has also been proven to be safe and effective and is an appropriate alternative, especially in locations where pediatric ERCP is not available [5–9]. As in adults, laparoscopic cholecystectomy for biliary stone disease in children is well accepted as the standard of care [8,10].

Given the rare occurrence of CDL in infants, its management is poorly defined and often perplexing. Data suggest that 0.15%–0.22% of children younger than 16 years of age have cholelithiasis [11]. Of these, approximately 10% will have CDL [12]. Based on these numbers, the rate of CDL in neonates and infants can be surmised to be far less than 1 in 5000. The small body habitus of infants makes standard surgical procedures, as well as ERCP, technically more challenging. Some authors have published CBD injury rates as high as 4.5% in children less than 5 years of age [13]. Many hospitals, including large academic referral centers, do not have an endoscopist capable of performing ERCP in infants. Infants who develop stone disease often have co-morbidities which can complicate the clinical scenario—surgery, sepsis, prematurity, total parenteral nutrition (TPN) dependence, bronchopulmonary dysplasia, hemolytic disease, and malabsorptive processes [14]. Finally, cholelithiasis and CDL have been reported to spontaneously resolve in 35–60% of cases with a low incidence of recurrence [15–18]. This has led to some degree of incongruity in the literature regarding appropriate management of CDL in infants. The purpose of this report is to describe our experience with infantile choledocholithiasis, as well as to review the body of literature regarding this disease entity.

2. Case series

2.1. Patient A

A full-term 3-month-old, 6.1 kg female presented to the emergency department (ED) with acholic stools and jaundice. Laboratory tests demonstrated a conjugated hyperbilirubinemia and mildly elevated transaminases. An abdominal ultrasound showed a dilated CBD (5 mm) with sludge in the gallbladder. MRCP the following day

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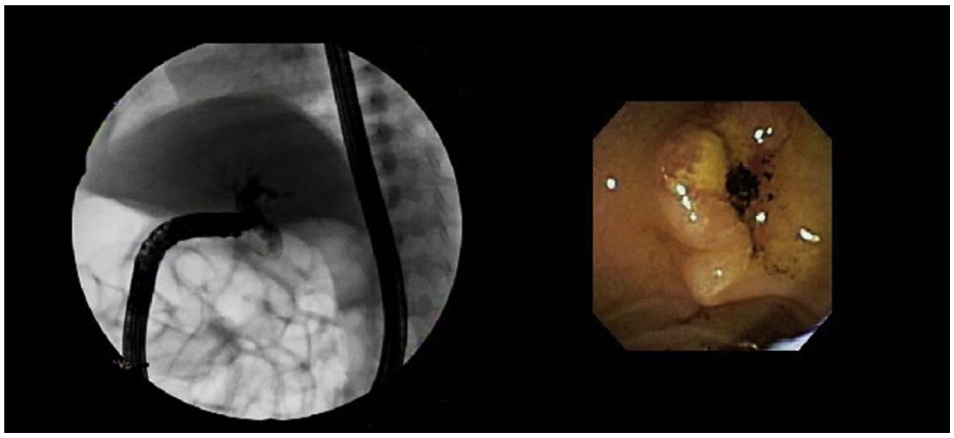

Fig. 1. Pre-operative ERCP for Patient A showing non-filling of the duodenum (left) and extraction of black pigmented stones (right).

revealed cholelithiasis, dilated intra- and extrahepatic bile ducts, and CDL of the distal CBD. The patient underwent ERCP, which was performed using an Olympus pediatric side-viewing scope. A cholangiogram was performed which showed multiple filling defects and a markedly dilated CBD. A biliary sphincterotomy was performed and multiple black pigmented stones were removed using a Cook Memory basket. Fig. 1 shows representative images of the ERCP (left) and black pigmented stones seen on endoscopy (right). The patient was taken to the operating room where she underwent laparoscopic cholecystectomy using a standard 4-port technique. She had an uncomplicated post-operative course with resolution of jaundice. The patient was discharged home on post-operative day one. Given the black pigmented stones, a workup for hemolytic anemia was undertaken but it was negative. The patient was seen in follow-up 5 weeks later and was found to be doing well with no further episodes of jaundice or acholic stools.

2.2. Patient B

A 6-month-old male with a history of heart failure, ventricular septal defect, and atrial septal defect was scheduled to undergo cardiac surgery. However, several days prior to his repair, he was admitted with a 2-day history of poor oral intake, irritability, and acholic stools. His weight at the time of admission was only 6.2 kg (2nd percentile). Physical exam showed jaundice and abdominal tenderness. Laboratory evaluation revealed conjugated hyperbilirubinemia. Ultrasound showed mild intra- and extrahepatic biliary dilation with sludge in the gallbladder and bile ducts. Computed tomography of the abdomen several small ($\leq$2 mm) stones in the distal CBD, in addition to the biliary dilatation.

He was not felt to be a candidate for ERCP because of his size. He was taken to the operating room for an open cholecystectomy with transcystic CBD exploration through a right subcostal incision. The cystic duct was found to be long and tortuous with an insertion into the CBD just above the duodenum. A cholangiogram was performed through the dilated cystic duct without distal passage of contrast into the duodenum (Fig. 2). A #2 Fogarty catheter (Edwards Lifesciences Corporation, Irvine, CA) was then passed through the cystic duct into the duodenum with successful removal of bilious sludge. Repeat cholangiogram revealed no further filling defects. It showed opacification of both right and left hepatic ducts, and contrast passage into the duodenum. Cholecystectomy was then performed and a closed suction drain was placed in the right upper quadrant. The child had an uneventful post-operative course. He tolerated a diet immediately after surgery and the drain was removed prior to discharge.

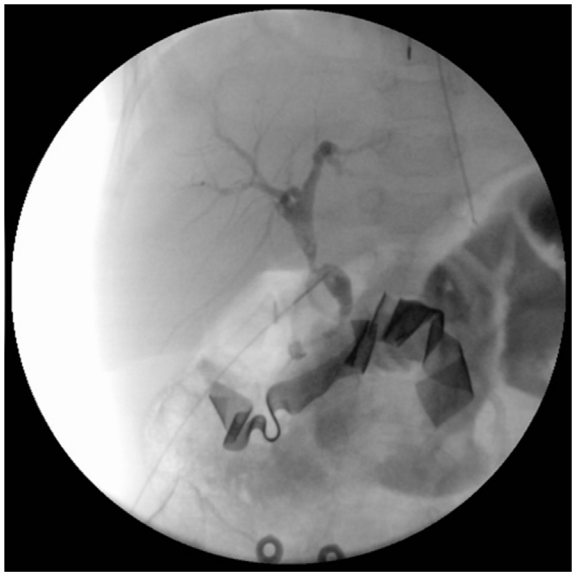

Fig. 2. Intraoperative cholangiogram for Patient B showing intra- and extra-hepatic biliary dilation as well as filling defects in the common bile duct.

2.3. Patient C

A 14-month-old, previously healthy male was admitted to the pediatric service with acholic stools, vomiting, and fever. His transaminases were mildly elevated with a normal total bilirubin. His mother was a known carrier for the alpha-1-antitrypsin phenotype, but the patient had no evidence of intrinsic liver disease or alpha-1-antitrypsin deficiency on work-up. Abdominal ultrasound demonstrated cholelithiasis without evidence of CDL and a normal common bile duct of 1.2 mm diameter. His stools returned to normal, he was tolerating an oral diet, and he was discharged home with plans for outpatient cholecystectomy.

One month later, he was found to be jaundiced with continued intermittent acholic stools and hyperbilirubinemia. Due to his size, he