

Cholecystitis and Choledocholithiasis in Infant: Case Report of Surgical Innovation in Paediatrics

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Abstract

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The incidence of cholelithiasis in paediatric age is increasing. However, the guidelines concerning the diagnostic approach and treatment are not well established and are controversial. The authors report a rare diagnosis of calculous cholecystitis and choledocholithiasis in a premature infant that attended the emergency room for persistent vomiting, diarrhoea and abdominal swelling. At this age, the clinical presentation is unspecific, the diagnosis is not obvious and the treatment is not well documented. A detailed anamnesis and the combination of several predisposing risk factors must alert the physician to the diagnosis. After an unsuccessful attempt with conservative treatment, the decision was made to try a complex surgical procedure, with diagnostic and therapeutic purposes. So far, this procedure, at this age group, has not yet been reported in the literature. Accordingly, this makes this case unique and strengthens its use as an innovative approach that achieved therapeutic success.

Keywords: Cholecystitis, Acute; Cholelithiasis/diagnosis; Cholelithiasis/aetiology; Cholelithiasis/surgery; Cholecystectomy, Laparoscopic; Infant; Premature, Diseases; Parenteral Nutrition/adverse effects

Introduction

The literature concerning cholelithiasis in the paediatric age is scarce. However, the number of cases is increasing, not only for a higher prevalence of the disease, but also for a greater accuracy in diagnosis.¹

The incidence of cholelithiasis in paediatric age shows a biphasic distribution. The first peak of incidence occurs during the first year of life, with no differences between genders.¹ This finding is associated with a higher number of risk factors (parenteral nutrition,² sepsis, bronchopul-

monary dysplasia, haemolytic diseases, ileal diseases, dehydration, immaturity of liver glucuronyl transferase, antibiotic therapy with ceftriaxone), and an increased bile lithogenesis in this period.³ The second peak occurs in adolescence, with a higher prevalence among the female gender, related to oestrogen effects.

At the paediatric age, clinical presentation is unspecific,³ diagnosis is not obvious and treatment is not well documented. Therefore, the guidelines concerning the diagnostic approach and treatment of this entity are not well established and are controversial.⁴ The diagnostic test of choice is abdominal ultrasonography, which has 90%-95% sensitivity.^{1,5} The treatment of acute cholecystitis should include a laparoscopic cholecystectomy, which is more likely to be successful when performed within three days of the onset of symptoms.^{3,6,7} It is important to remember that gallstones can lead to a variety of other complications including choledocholithiasis, gallstone ileus and acute gallstone pancreatitis.⁸ If a complication is presumed, noninvasive – ultrasonography, computed tomographic cholangiography, magnetic resonance cholangiopancreatography (MRCP) – and invasive – endoscopic retrograde cholangiopancreatography and percutaneous trans-hepatic cholangiography – imaging modalities should be considered.^{9,10}

This case report stresses the side effects of parenteral nutrition and the importance of a high index of suspicion,² and presents an innovative surgical approach to cholelithiasis not previously reported in this age group.

Case Report

A 12 month-old Caucasian male, born in Portugal at 28 weeks of gestation, was admitted to the emergency room for persistent vomiting and diarrhoea in the last 7 days, associated with abdominal swelling, with no history of fever or presumptive food intoxication. The patient followed a typical Mediterranean diet for his

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age. He was fed exclusively with formula milk until 6 months of life, at which point he began food diversification with fruit and vegetable soup. Gluten, meat and fish were introduced between 6 and 7 months of age and egg yolks and whites at 9 and 11 months, respectively. From 2 weeks old until admission, he was daily supplemented with vitamin D.

The physical examination revealed a distended abdomen with a visible mass at the right upper abdominal quadrant that was palpable 3 cm below the right costal margin, with tenderness at superficial palpation. Anicteric, slightly sunken eyes and moderate nasal obstruction conditioning some snoring.

The abdominal ultrasound showed a distended gallbladder with thickened walls, biliary sludge, several gallstones in the infundibulum and dilatation of the common bile duct (Fig. 1), confirming the diagnosis of lithiasic cholecystitis. The liver was normal, without structural or morphologic changes and no dilatation of intra bile ducts. The pancreas and spleen were normal and there was no evidence of ascites.

Blood tests revealed:

- Normal blood count without anaemia or leucocytosis: haemoglobin 11.7 mg/dL, leukocytes $9650 \times 10^9/L$;
- Thrombocytosis: $510 \times 10^9/L$;
- Elevated liver enzymes: aspartate aminotransferase (AST) 1358 UI/L, alanine aminotransferase (ALT) 1394 UI/L, gamma glutamyl transpeptidase (GGT) 333 UI/L, alkaline phosphatase 946 UI/L, lactate dehydrogenase (LDH) 874 UI/L;
- Unchanged coagulation tests;
- Direct bilirubin (DB) and total bilirubin (TB) slightly elevated with 1.13 mg/dL and 1.55 mg/dL, respectively;
- Albumin 3.8 g/dL, total proteins 6.1 g/dL;
- Amylase 13 UI/L;
- Normoglycaemia;
- Normal C-reactive protein (CRP).

The patient was transferred to paediatric surgery depart-

ment in order to complete seven days of intravenous antibiotic and *nil per os*, and schedule elective surgery out of the acute setting.

At the seventh day of admission, there was a sudden onset of fever, intermittent episodes of jaundice and acholia, along with no evidence of clinical improvement, and the patient was submitted to a MRCP that revealed cholelithiasis showing one 6 mm infundibular stone, some other millimetric stones, and biliary sludge at the common bile duct and principal biliary duct, which were both dilated (Fig. 2). The blood test control revealed improvement in liver enzymes (AST 112 UI/L, ALT 203 UI/L), but a rise of DB (4.69 mg/dL), TB (5.49 mg/dL) and CRP (9.12 mg/dL).

Consequently, after nine days of intravenous treatment with amoxicillin and clavulanic acid, a course of ceftriaxone (85 mg/kg/day) was initiated, along with intravenous fluid therapy and analgesia for 11 days.

At the 22nd day of admission, a laparoscopic procedure was performed. It started with the placement of four trocars (5 mm in the navel, 5 mm in the right flank, 3 mm in the epigastrium and 3 mm left flank), followed by the exposure of the Calot's triangle, with dissection and isolation of the cystic duct and artery. Then, a link with Hem-o-lok upstream of the cystic duct and a partial section downstream were made in order to perform an intraoperative cholangiography (Fig. 3). This allowed the confirmation of the choledocholithiasis diagnosis and a choledochoscopy with a 3 mm flexible uretero-roscope¹¹ was performed with extraction of two stones from the common bile duct, with a Dormia stone basket catheter. Post-choledochoscopy cholangiography revealed the remaining lithiasic debris in the common hepatic duct (Fig. 3). Consequently, saline irrigation of the biliary tree was performed and residual lithiasis was eliminated. The radioscopic confirmation of absence of lithiasis was performed and the cholecystectomy procedure was ended with the extraction of the vesicle through the umbilical trocar.

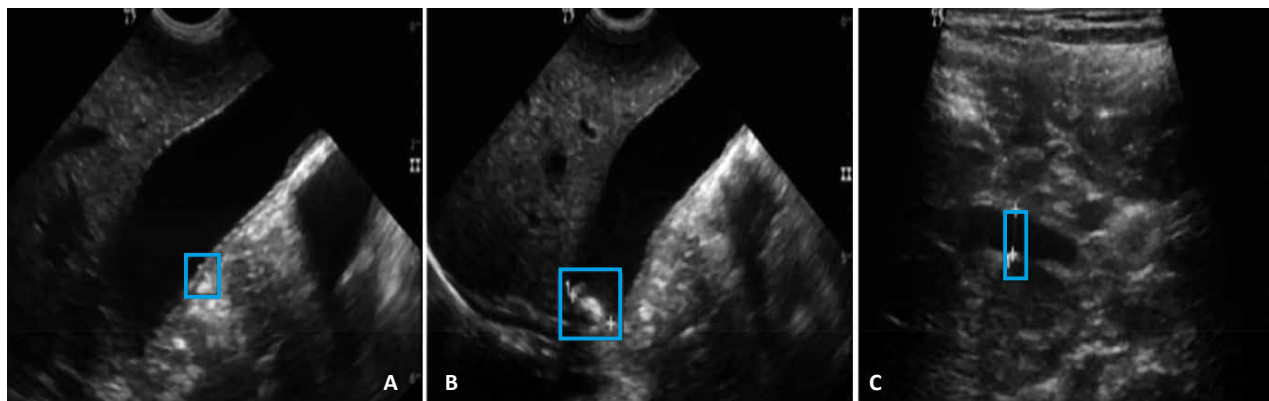


Figure 1. Abdominal ultrasound. Distended gallbladder with thickened wall (A), gallstones in the infundibulum (B), and dilatation of the common bile duct (C).

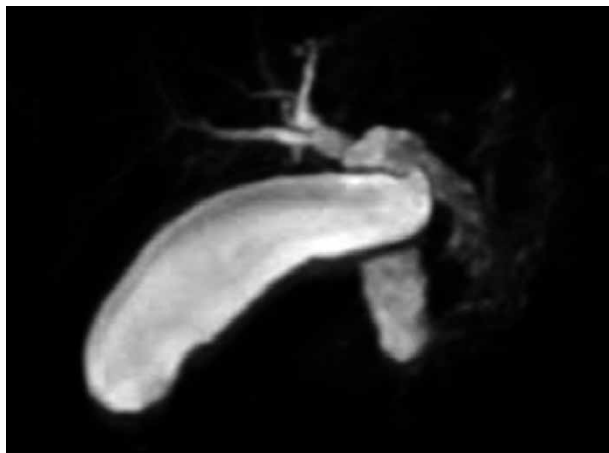


Figure 2. Magnetic resonance cholangiopancreatography. Cholelithiasis, showing one infundibular stone of 6 mm, some other millimetric stones nearby and also biliary sludge at the choledochal duct and principal biliary duct, which are both dilated.

The postoperative period progressed uneventfully and surgical wound healing was excellent. There was the need to optimise the analgesia with paracetamol, tramadol and ketorolac. Ultrasound and blood testing were performed three days after surgery, showing the normalisation of the results.

Presently, after two years of follow-up, the child remains well, without complaints or complications related to this diagnosis or to the surgical procedure.

Discussion

Cholelithiasis typically manifests as an acute abdominal pain, especially in the right upper quadrant or epigastrium, jaundice, nausea, vomiting, anorexia, fat intolerance, pancreatitis and fever. Contrary to this presentation, lithiasic cholecystitis in infants can go unnoticed,^{1,4} which may explain the diagnostic delay in this clinical case.

Nevertheless, we found a history of longstanding parenteral nutrition for the first 40 days of life, that should

have raised the suspicion of the diagnosis.² This method implies the absence of enteric transit leading to a lower secretion of intestinal hormones, reduced bile flow and stasis, which are very important mechanisms in the emergence of cholestasis, biliary sludge and cholelithiasis.¹ Moreover, after birth, the patient suffered several complications of prematurity including:

- Grade 3 hyaline membrane disease with exogenous surfactant administration;
 - Neonatal jaundice treated with phototherapy for two days;
 - Cholestasis with 9.69 mg/dL of TB, direct bilirubin 20% of TB and alkaline phosphatase of 1211 UI/L (34 days after initiating parenteral nutrition);
 - Hospital-acquired sepsis treated with antibiotics (ampicillin, gentamicin, vancomycin and amikacin);
 - Gastrointestinal bleeding treated with ranitidine;
 - Anaemia requiring a total of four red blood cell transfusions, with the lowest haemoglobin value of 5.8 g/dL.
- Therefore, not only one, but the combination of several risk factors contributed in triggering this outcome.^{1,4}

Thus, in patients with predisposing risk factors, a routine follow-up with a particular focus on the biliary system is essential.¹ This approach allows for timely prediction or diagnosis and may even prevent some of its complications.

Apart from the lithiasic cholecystitis, the diagnosis of choledocholithiasis at this age makes this case even more unusual. The treatment of choledocholithiasis is not well established in infants. The endoscopic retrograde cholangiopancreatography (ERCP) might be particularly challenging⁹ or even impossible. Therefore, the authors decided to perform a laparoscopic surgery that involved intraoperative cholangiography, choledochoscopy and cholecystectomy, during the same surgical intervention. To the best of our knowledge, this laparoscopic procedure has not yet been reported in this age group, which makes this case unique.

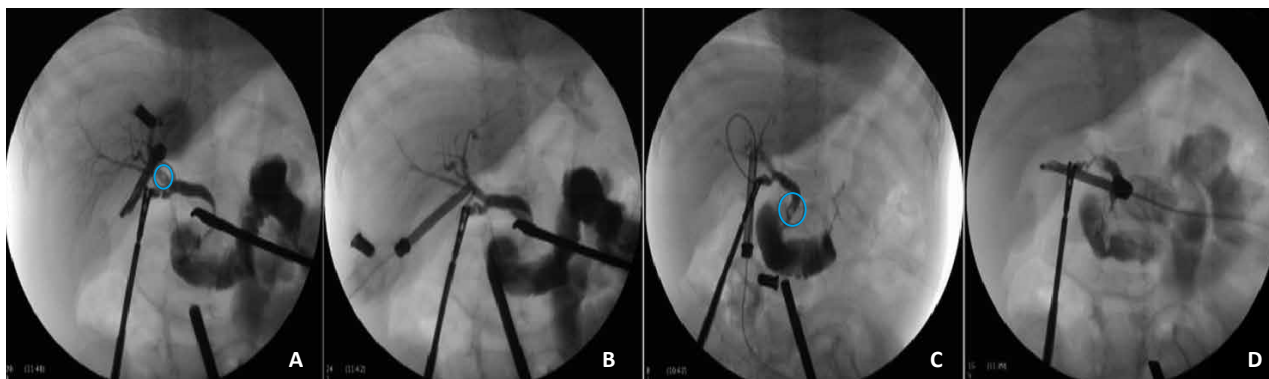


Figure 3. Intraoperative cholangiography showing remaining lithiasic debris observed in different perspectives (A to D).

WHAT THIS CASE REPORT ADDS

- The diagnosis of choledocholithiasis in the first year of life is rare, but possible.
- Contrary to the usual clinical course in adults, lithiasic cholecystitis may go undetected in small infants.
- Endoscopic retrograde cholangiopancreatography may be difficult or even impossible to perform in the first year of life.
- A unique example of therapeutic success in a small infant undergoing laparoscopic surgery with intraoperative cholangiography, choledochoscopy and cholecystectomy, during the same surgical intervention.
- In patients with risk factors for cholelithiasis, adequate follow-up allows for timely prediction or diagnosis and may even prevent some of its complications.

Conflicts of Interest

The authors declare that there were no conflicts of interest in conducting this work.

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Protection of human and animal subjects

The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).

Confidentiality of data

The authors declare that they have followed the protocols of their work centre on the publication of patient data.

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Colecistite e Coledocolitíase em Lactente: Caso Clínico de Inovação Cirúrgica em Pediatria**Resumo:**

A incidência de colelitíase em idade pediátrica está a aumentar. No entanto, não existem linhas de orientação para a sua correta abordagem diagnóstica e terapêutica, que constituem um tema controverso. Os autores relatam um caso raro de colecistite litíase e coledocolitíase num lactente prematuro. Recorreu ao serviço de urgência por quadro de vômitos persistentes, diarreia e distensão abdominal. Nesta faixa etária, a apresentação clínica é inespecífica, a hipótese diagnóstica nem sempre é equacionada e a abordagem terapêutica não está bem documentada. A anamnese detalhada e combinação dos fatores de risco predisponentes devem alertar-nos para este

diagnóstico. Após uma tentativa mal sucedida com tratamento conservador, a decisão foi tentar um procedimento cirúrgico complexo, para fins diagnósticos e terapêuticos. Não estão publicados relatos científicos do uso deste procedimento nesta idade. Este facto torna este caso único, realçando a escolha de uma abordagem inovadora, que decorreu sem complicações e garantiu sucesso terapêutico.

Palavras-Chave: Colecistectomia Laparoscópica; Colecistite Aguda; Colelitíase/cirurgia; Colelitíase/diagnóstico; Colelitíase/etiologia; Doenças do Prematuro; Lactente; Nutrição parentérica/efeitos adversos