

Spontaneous perforation of the common bile duct in a pediatric patient. Case report and short review of the literature

Perforación espontánea de colédoco en un paciente pediátrico. Presentación de caso y revisión breve de la literatura

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Abstract

Introduction: Spontaneous perforation of the common bile duct is rare. The cause is usually unknown, although it is sometimes related to the malformation of the bile duct. **Clinical case:** Female of 1 year, with abdominal distention, hyporexia and fever, tomography with ascites, surgical findings included retroperitoneal bilioma, peritonitis and perforation of common bile duct. Block resection and primary hepato-duodenal anastomosis were performed. **Discussion:** Spontaneous perforation of the common bile duct tend to evolve insidiously and delay in diagnosis is frequent. Surgical management is decisive, and there are different techniques described. **Conclusion:** A high index of suspicion is important, treatment must be individualized according to each patient.

Key words: Bilioma. Spontaneous perforation of the biliary tract. Biliary peritonitis.

Resumen

Introducción: La perforación espontánea del colédoco es rara. La causa generalmente se desconoce, aunque en ocasiones guarda relación con una malformación de la vía biliar. **Caso clínico:** Niña de 1 año, con distensión abdominal, hiporexia y fiebre, tomografía con ascitis, se interviene y se encuentra bilioma retroperitoneal, peritonitis y perforación de colédoco. Se realiza resección en bloque y anastomosis hepatoduodenal primaria. **Discusión:** La perforación espontánea del colédoco tiende a evolucionar insidiosamente y el retraso en el diagnóstico es frecuente. El manejo quirúrgico es resolutorio; existen distintas técnicas descritas. **Conclusión:** Es importante un alto índice de sospecha. El tratamiento se debe individualizar en cada paciente.

Palabras clave: Bilioma. Perforación espontánea de la vía biliar. Peritonitis biliar

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Introduction

Spontaneous perforation of the bile duct is a rare event in pediatrics, with few cases reported in the international literature. The cause is not entirely known, with the most accepted theory involving pancreaticobiliary junction malformation and consequences thereof¹. Usually, it occurs as abdominal distention associated with jaundice and hyporexia, and on rare occasions it can occur as acute abdomen. Diagnostic studies are poorly sensitive or specific, and treatment in all cases is surgical. The procedure will depend on patient condition upon entering the operating room and the degree of inflammation of surrounding structures².

Case presentation

One-year-old girl, with no relevant medical history, who presented with abdominal distention accompanied by nausea with vomiting and decreased consistency stools, which was managed as a gastrointestinal infection. Of insidious evolution, on seventh day fever, dehydration and peritoneal irritation were added. Laboratory tests showed mild leukocytosis without neutrophilia. Computed tomography revealed free fluid in the cavity and a right subdiaphragmatic collection with thin septa (Fig. 1). Laparotomy reported 250 ml of free icteric fluid in the cavity, lax subhepatic adhesions, intense peritonitis and biliary impression in the retroperitoneum near the common bile duct (Fig. 2). Intraoperative cholangiography identified bile leakage in the retroduodenal portion of the common bile duct (Fig. 3), with a 0.5-cm punctiform perforation being therefore dissected and identified on the posterior surface, at 1 cm from its insertion with the pancreas. En bloc resection of the common bile duct and gallbladder was decided, and hepatoduodenal anastomosis was performed. Postoperative clinical evolution was satisfactory, and discharge was possible 7 days after the procedure. With a 6-month postoperative follow-up, she is currently asymptomatic.

Discussion

Spontaneous perforation of the bile duct is a rare pathological event in pediatrics. It is more commonly observed in patients younger than 20 weeks of life, with a presentation ratio by gender of 1:1³ and an approximate incidence of 1.5 per 1,000,000 live



Figure 1. Abdominal computed tomography scan, where free fluid is appreciated in the abdominal cavity.

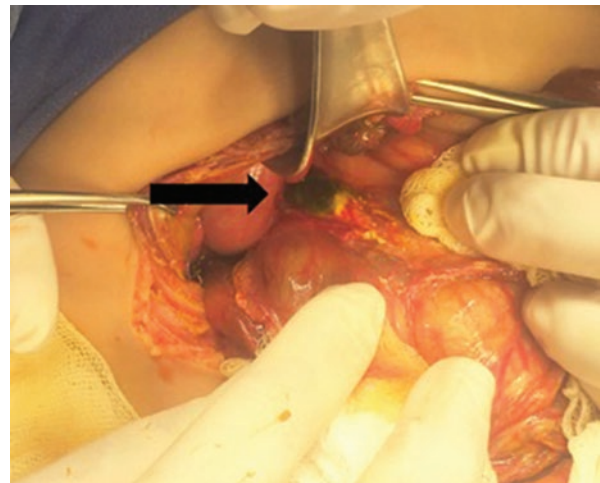


Figure 2. Image of the retroperitoneal biloma.



Figure 3. Transoperative cholangiography, where contrast material leakage is observed.

newborns. During the last 15 years, only 90 cases have been reported⁴. Most affected patients are usually previously healthy and generally do not have a significant prenatal or postnatal history⁵.

From the etiopathological point of view, the most common cause is idiopathic⁶, which is believed to be associated with a malformation of the common bile duct on its extreme at the duodenal level, which causes pancreatic enzymes reflux, wall weakness and predisposition to perforation⁷. Gallstones, trauma, previous surgical intervention, and congenital disorders, such as common bile duct cyst, are present to a lesser extent⁸.

The usual perforation site is the anterior surface of the common bile duct, near the junction with the cystic duct, which tends to generate intra-abdominal collection and biliary peritonitis⁹. Those that occur in the posterior aspect of the duct are considered rare, determine retroperitoneal collection and in turn delayed diagnosis, which in most cases is made at the time of surgical exploration¹⁰.

Clinical presentation is usually divided in two: an insidious form, which accounts for 80% of cases and is characterized by progressive abdominal distension, hyporexia, jaundice, acholia, and choluria; or a form with acute symptoms, which accounts for the remaining 20% and is characterized by marked irritability, abdominal distention, vomiting, fever and peritoneal irritation.

Blood cell count is usually nonspecific, although at advanced stages of the disease, leukocytosis and neutrophilia tend to appear¹¹. In these cases, abdominal ultrasound has been documented to show normal intrahepatic and extrahepatic bile ducts, with presence of free fluid in the abdominal cavity that is sometimes associated with loculi. Conversely, tomography does reveal wall defects, although it is limited due to the need for intravenous contrast to be administered¹². The most sensitive and specific study is liver and bile duct scintigraphy. Another modality is diagnostic paracentesis, which reveals the presence of free bile in the abdominal cavity¹³.

Several therapeutic modalities have been proposed, which range from observation to biliodigestive diversion. Simple peritoneal or bile duct drainage with T-tube placement is reported to be the treatment of choice in uncomplicated cases, with good results, as described in current literature¹⁴, and the most common cause of failure in this modality is distal common bile duct obstruction by bile sludge and inflammation, which causes an increase in bile leakage and difficulty

for closure. Another more complex technique is swollen tissue resection and biliodigestive diversion to the duodenum or jejunum¹⁵. The decision will depend on patient clinical characteristics at diagnosis and the degree of inflammation of the periportal region¹⁶. One of the complications of drainage as single treatment is acquired atresia or bile duct stenosis¹⁷. Other less frequent associated complications include cholangitis, portal vein thrombosis and biloma¹⁸. It is important for diagnosis and surgical resolution to be made early in order to reduce the risk of complications for the patient¹⁹. The prognosis depends on the severity of the clinical presentation, the inflammation it causes and the time elapsed until definitive surgical resolution, with variable degrees of morbidity and mortality that are directly associated with deferred surgical management²⁰.

Conclusion

Spontaneous perforation of the common bile duct is rare in pediatrics. It is important to have a high index of suspicion in patients with biliary peritonitis. The tomographic study helps for diagnosis to be established. Several surgical techniques have been described, which range from simple external drainage to resection and biliodigestive diversion; the decision will be made taking into account patient general conditions and the degree of local inflammation in the periportal region. In our review of cases, we found good to excellent results for life and function of these patients regardless of the surgical option chosen and patient characteristics.

Conflict of interests

The authors declare that there are no conflicts of interest.

Ethical disclosure

Protection of people and animals. The authors declare that no experiments were performed on humans or animals for this research.

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

Right to privacy and informed consent. The authors have obtained informed consent from the patients and/or subjects referred to in the article. This

document is in the possession of the corresponding author.

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