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What an Acute Abdomen can conceal in Children?

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ABSTRACT
Cystic malformations of the biliary tract are rare congenital conditions estimated at about 1/2,000,000 births. It is a condition that can lead to serious complications such as angiocholitis, chronic pancreatitis, progressive biliary cirrhosis, portal hypertension or biliary lithiasis. Its spontaneous perforation is one of the few complications, first described by Weber in 1934. Reported to us the case of an 18-month-old boy admitted for sub-occlusive syndrome with biliary peritonitis. An ultrasound scan was performed showing abdominal effusion with communicating cystic formation of the biliary tract associated with a sub-capsular effusion of the liver confirmed by a CT scan. The procedure consisted of a peritoneal toilet with a redon drain at the perforation level and a subhepatic drain without excision of the cyst. The patient was readmitted 6 months after this incident for final treatment.

1.0 Introduction

Cystic malformations of the biliary tract are rare congenital conditions[1]. The most commonly accepted etiology is the anomaly of the bilio-pancreatic junction, which is also incriminated in the degeneration of the biliary tract. The age of onset of this malformation is less than 10 years with feminine predominance[2]. The triad of pain, jaundice, mass, usually evokes a cystic dilatation of the biliary tract, but it is found in only 13 to 25% of cases [3]. Exceptionally, this condition can be seriously complicated. The resection of the cystic dilatation is the reference treatment. Kystodigestive anastomosis is currently discontinued as cystic malformation of the biliary tract is considered a precancerous condition [2].

2- Observation

An 18-month-old boy with no specific pathological history who has been admitted to pediatric emergency departments for food vomiting for the past 7 days, becoming bilious recently, screaming, and fever. The clinical examination at admission showed a temperature of 38.5°C, a bloated abdomen with diffuse abdominal tenderness. The general condition was kept out of asthenia, the hemodynamic condition was stable, without signs of shock. An abdomen without preparation was performed without abnormalities. An ultrasound scan showed abdominal effusion with communicating cystic formation of the biliary tract associated with subhepatic effusion.

An echoguided liver puncture was performed, affirming the bilious origin of the subhepatic fluid. The CT complement confirmed the ultrasound data (Figure 1). Biological markers were in favour of moderate inflammatory syndrome and liver enzymes were normal. A laparotomy under right costal was performed for suspicion of rupture of the bile duct cyst. Surgical exploration confirmed the presence of nonpurulent biliary peritonitis associated with perforation of a bile duct cyst. The initial surgical intervention consisted of a peritoneal toilet with a redon drain at the perforation level and a subhepatic drain without excision of the cyst.

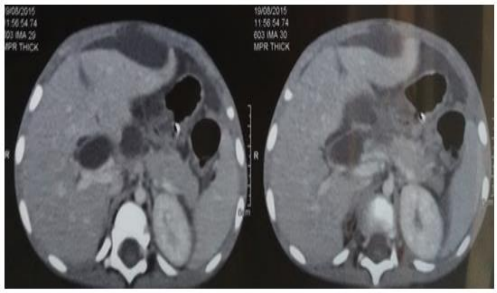

Figure 1. Aspect of bile duct cyst rupture on a CT scan.

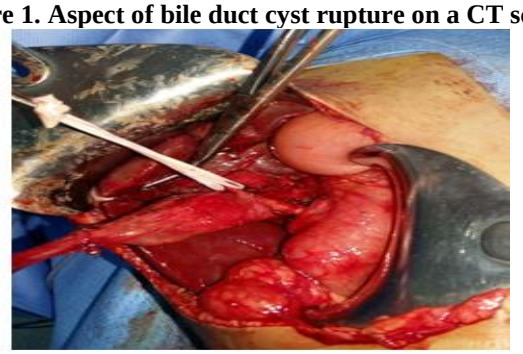

Figure 2. Appearance of intraoperative cystic dilatation.

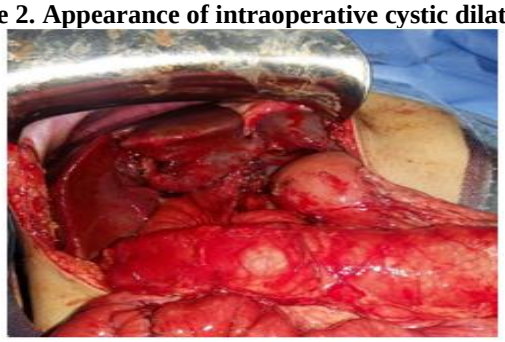

Figure 3. Choledochojejunal Y-anastomosis.

The liver looked normal without signs of cirrhosis. The postoperative follow-up was straightforward and the child was able to leave the ward on the 10th post-operative day. The patient was readmitted 6 months after this incident for

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permanent treatment of his malformation with a complete removal of the dilatation of the main biliary tract, with hepaticojejunal anastomosis on a Roux Y-shaped handle (Figure 2 and 3).

3- Discussion

The first description of root canal dilatation of the biliary tree dates from 1723 by Vater [4]. It is a rare malformation, which is readily seen in children (75% of cases are discovered before the age of ten years), with a clear feminine predominance (sex-ratio between 0.23 and 0.43) [2]. Its incidence is of the order of 1/2000,000 births [1]. It is most evident in Asian countries [5]. Although affecting the child preferentially, the bile duct cyst (BCCD) may not manifest itself until adulthood as a complication, mainly infectious [6]. The most commonly proposed etiological hypothesis is that the bile duct cyst is the result of an anomaly in the pancreaticobiliary junction [7]. Recently, abnormal functioning of the Oddi sphincter has been reported as predisposing to reflux of pancreatic juice into the biliary tract, and spasms of the Oddi sphincter have been noted in association with bile duct cysts [8].

Todani's classification is the most widely used and comprises five types [7], the most common type is Type I, corresponding to dilatation of the extrahepatic biliary tract, and divided into 3 subtypes: Ia = cystic dilatation, Ib = segmental dilatation and Ic = fusiform dilatation [7].

The diagnosis can be evoked in antenatal before the observation of fluid formation at the level of the hepatic hilum region [5,9]. In children, its discovery is fortuitous, and when it is symptomatic, it manifests itself in the classical triad: abdominal pain, jaundice, mass of the right hypochondrium[3]. This symptomatology is observed in only 13 to 25% of cases [3]. DKC is rarely found at the stage of complications [10], and are: lithiasis which can block bile ductal low, infection (angiocholitis, hepatic abscess or sepsis) [5], primary biliary cirrhosis, portal hypertension, cholangiocarcinoma and malignant degeneration with cholangiocarcinoma with a variable frequency in Spontaneous rupture is one of those rare complications. Its incidence is 1.8% to 2% [3]. In the majority of cases it occurs in children under 4 years of age [3] and was first described in 1934 by Weber [3]. The clinical picture in this case is usually progressive abdominal distension, vomiting, and shock with or without jaundice [3]. Traumatic rupture is even rarer, with only a few cases described [5,12].

In ultrasound, DKC is presented as a cystic mass appended or replacing the bile duct and extending upwards with the cystic duct and liver duct, and down with the terminal or Wirsung duct [3]. This examination may be sufficient for diagnosis and CT does not provide additional information [3,10]. Cholangio-TDM allows to visualize the accumulation of the contrast medium in the cyst. A mapping of the biliary tree and a precise delineation of the lesion are thus obtained [10]. Cholangio MRI or bili-IRM is a recent and non-invasive technique that is highly effective in the diagnosis of anomalies of the bilio-pancreatic junction [3].

As soon as the diagnosis of a cystic dilatation of the bile duct is diagnosed, the surgical cure should be performed at 6 months of age [13]. The most commonly accepted surgical operation consists of resecting the entire dilated portion of the biliary tract and performing bile duct anastomosis or hepaticojejéjunale or hepaticojejunale on a Roux Y-shaped handle [3,5,14]. The resection is wide considering the risk of secondary degeneration of the dysplastic biliary wall [3].

For intrahepatic cysts and intrahepatic dilatations (Todani type IV), other procedures may be necessary, such as segmentectomy, partial hepatectomy or intrahepatic kystoenterostomy [2,15]. Hepatic biopsy is recommended to detect early signs of hepatic cirrhosis [5].

4- Conclusion

The rupture of a bile duct cyst is a rare cause of acute abdominal pain in children and can develop on low noises for a few days. Diagnosis is evoked by ultrasound and CT scan and confirmed intraoperatively. The treatment consists of peritoneal cleansing and resection of the cyst with a Y-choledochojejéjunal anastomosis.

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