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Congenital Cystic Dilatation of Main Bile Ducts: A Case Report

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ABSTRACT
Cystic dilatation of the bile ducts is a rare congenital malformation. It's due to an abnormality of the biliopancreatic junction involving the extra and intrahepatic bile ducts. It's the second leading cause of congenital disabilities of the bile ducts after atresia. According to Todani, several types depending on the site, shape, and distribution of the malformation. It's evoked in front of the inconstant clinical triad: pain, jaundice, and mass. Ultrasound and better sectional imaging confirm the diagnosis. Surgical excision is the treatment of choice to prevent malignant degeneration of the cyst wall and bile ducts.

Introduction

Cystic malformations of the bile ducts are rare congenital conditions. They are classified, according to Todani, according to the location, extent, and type of dilatation of the bile ducts. The anomaly of the biliary-pancreatic junction plays an essential role in the genesis of this malformation and the degeneration of the bile ducts. Surgical resection is the standard treatment [1].

Case report

We report the case of a 60 years old patient, operated on ten years ago for cholecystectomy, who presents since three months intermittent pains with a feeling of heaviness of the right hypochondrium without irradiation.

The clinical examination showed localized tenderness to palpation, without fever, jaundice, or other associated signs. The biological workup did not reveal any inflammatory syndrome or cholestasis. Ultrasound showed a significant cystic dilatation of the main bile duct, with no image of echo-detectable obstruction downstream. Then performed an abdominal CT scan showing a large fusiform cystic dilatation of the main bile duct, measuring 71x49mm, classified as type I according to the Todani classification, with slight dilatation upstream of the intrahepatic bile ducts (Figure 1). The MRI better appreciated this cystic dilatation of the main bile duct and moderately of the proximal intrahepatic bile ducts (figure 2) and excluded possible lithiasis of the lower bile duct or mass syndrome. The patient underwent surgical resection of the main bile duct with hepatico jejunal anastomosis. The postoperative course was simple.

Discussion

Congenital cystic dilatation of the main bile duct is a rare anomaly (1 in 100,000 and 150,000), but the most frequent malformation cause of extrahepatic cholestasis in infants, with a clear female predominance (80% of cases) [2]. It is an abnormality of the junction of the bile duct and the pancreatic duct with an abnormally long common biliopancreatic duct. This junction induces the reflux of pancreatic juice into the bile duct. [3]. Several classifications depend on the type of bile duct dilatation and whether or not it is associated with cystic dilatation of the intrahepatic bile ducts. The best known is Alonso-Lej, revised by Todani: Type I (80%)) Cystic dilatation of the main bile duct. Type II (10%): Supraduodenal diverticulum of the bile duct. Type III (4%): choledochocele. Type IV (11%). a: Dilatation of the intra- and extrahepatic bile ducts. b: Multisegmental dilatation of the main bile duct. Type V (< 1%): Isolated dilatation of the intrahepatic bile ducts [2]. The fusiform form is by far the most frequent, the diverticular form being exceptional.

The classic clinical triad of mass, pain, and jaundice is present in only 10% of cases. Most often, the diagnosis is made by ultrasound in the presence of abdominal pain and disturbances of liver balance in children under ten years of age. However, some cases are diagnosed prenatally and others in adulthood (20% of cases) [4].

Acute pancreatitis is often revealing in children. Other complications (lower bile duct lithiasis, cholangitis flare-up, perforation, biliary cirrhosis) can sometimes be showing with a risk of secondary degeneration of the cyst wall that increases with age [5]. The diagnosis of a bile duct cyst can be easily evoked on ultrasound in the presence of cystic dilatation of the main bile duct, located in the hepatic pedicle in continuity with the dilated BSIV and the gallbladder. This cyst can sometimes be the site of some small stones or echogenic debris gallbladder often has a thickened wall and may also contain sludge or stones. The pancreatic duct may be dilated, sometimes with visibility of its junction with the common bile duct in the head of the pancreas [6]. The differential diagnosis in the neonatal period is biliary atresia in its cystic form. The main differential diagnosis is a dilatation of the bile ducts secondary to primary lithiasis of the lower bile duct. The CT scan, and better still the MRI scan, makes the diagnosis of a biliary-pancreatic junction anomaly with an abnormally long common duct, exceeding 5 mm in children and 15 mm in adults [7]. In case of doubt,

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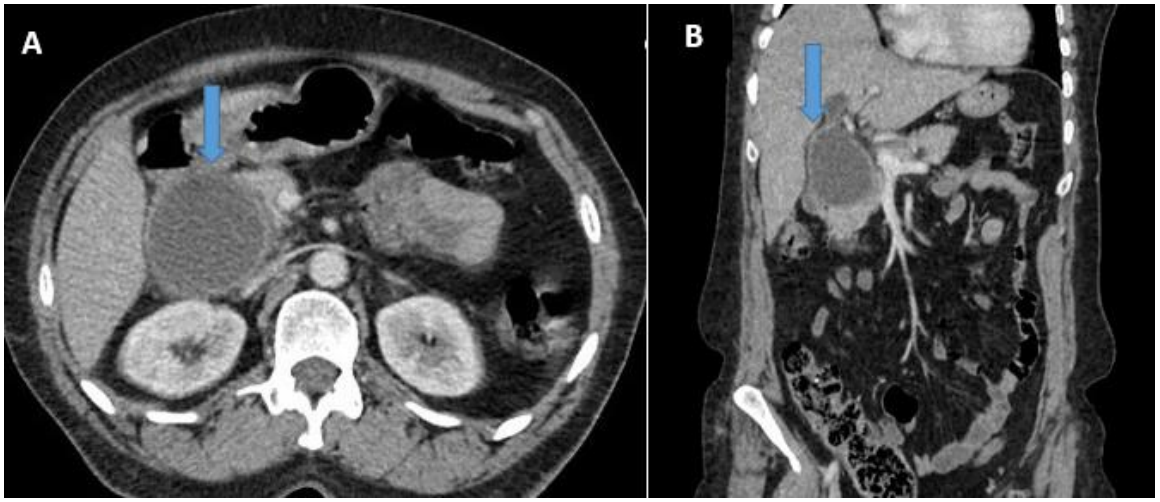

Figure 1. Abdominal CT scan with iodinated contrast injection (A: axial slice B: coronal slice) showing fusiform cystic dilatation of the main bile duct ( ) , extending superiorly to the hepatic hilum with mild dilatation of the common hepatic ducts and intrahepatic bile ducts, classified as TODANI type I, exerting a soft mass effect on the duodenal frame and pancreatic parenchyma.

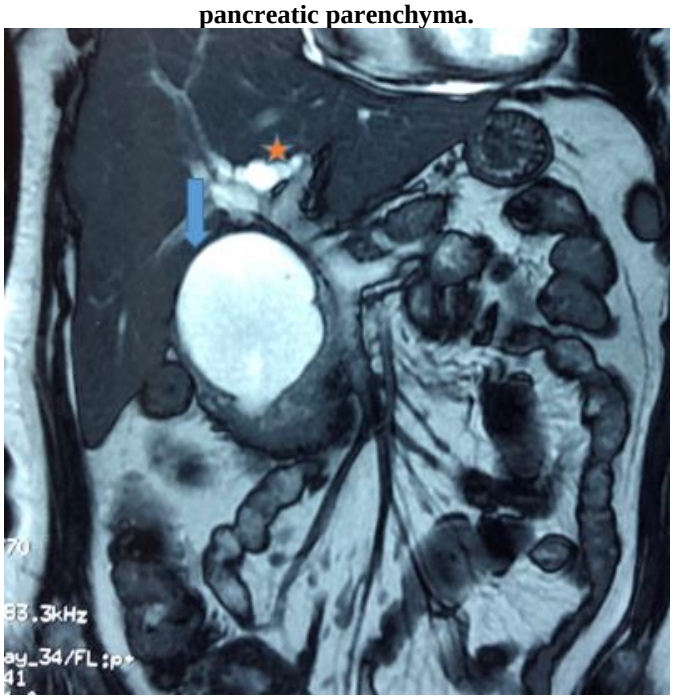

Figure 2 . Abdominal MRI T2-weighted coronal slice showing a spindle-shaped cystic dilatation of the main bile duct ( ) with thin wall and fluid content of the homogeneous signal and identical to the bile with slight dilatation upstream of the intrahepatic bile ducts ( ).

preoperative or intraoperative biliary tract opacification and elevated pancreatic enzyme levels in the bile can confirm the diagnosis.

Complete surgical resection of the cyst and gallbladder with hepatico jejunal anastomosis is the standard gold treatment due to the increased risk of malignant degeneration [1].

Conclusion

Cystic bile duct dilatation is a rare congenital malformation of the main bile duct. Imaging with ultrasound, MRI, and especially BILI-MRI plays an important role in their diagnosis. The treatment must be adapted because of serious and multiple complications to each type of malformation, where the complete resection possible is the intervention of choice.

Conflict of Interest

The authors declare that they have no conflicts with this manuscript.

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