Whipple procedure in the treatment of Todani type IVa bile duct cyst. A case report

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Background: Introduction: Congenital bile duct dilation is a lesions that can be extrahepatic, intrahepatic, or both. There is a predominance in the female population. The Todani's classification has become the basic classification system, with type I being the most common, of all bile duct cysts, generally the most of cases are infants and young children. In bile duct cysts, resection is justified to prevent future malignant tumors and complications.

Case Report: We present the case of a male young of 16 years old who presented to the hospital with important increase in abdominal perimeter, intermittent pain and jaundice, and with help of studies of imaging is identified a giant atypical bile duct cyst and we decided to give surgical treatment

Conclusion: Due to the risk of infection or malignancy, the principle of treatment is bile duct resection with bilio-digestive reconstruction, but the age of this patient made this procedure more complex

Case Report

General Surgery

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Congenital bile duct dilation was first described in 1723 by Vater and Ezler (1). These lesions can be extrahepatic, intrahepatic, or both (1,2). The incidence in the Western population is 1 in 100,000 to 150,000 live births; in Asian populations, it is 1 in 1,000–13,000 (2,6), with Japan having two-thirds of all cases. There is a predominance in the female population, more frequent in the first decade.

Since 1977, Todani's classification has become the basic classification system, with type I being the most common, accounting for 80 to 90% of all bile duct cysts, while type IV accounts for 15% to 20% (6).

Generally, 80% of cases are infants and young children (4). The clinicopathological characteristics vary with age. Abdominal discomfort, jaundice, and an abdominal mass are the most common symptoms in children. Adults present with biliary or infectious problems and are more prone to progress to choledocholithiasis and malignant transformation (2).Both diagnosis and classification of subtypes can be made by abdominal ultrasound, computed tomography (CT), magnetic resonance cholangiopancreatography (MRCP), endoscopic retrograde cholangiopancreatography (ERCP) (7).

In bile duct cysts, resection is justified to prevent future malignant tumors and complications (5). The risk reduction for future malignancy development varies depending on the patient's age and type of lesion. Management includes total cyst excision and bilioenteric reconstruction (6). This case report describes a unique case of a giant atypical bile duct cyst and the complexity of its surgical treatment.

Case report

A 16-year-old male from Saltillo, Coahuila, was attended in the general surgery consultation at a third-level hospital in northern Mexico with complaints of moderate colicky epigastric pain relieved by oral analgesics for the past two years. The patient presented with generalized jaundice, pruritus, nausea, and vomiting on multiple occasions with biliary characteristics, as well as dark urine, prompting him to seek evaluation by our service. He denied fever and other constitutional symptoms. He reported no known comorbidities or previous surgeries. On physical examination, the patient was in a fair general state, with moist mucous membranes but with frank jaundice. Abdominal examination revealed a very distended abdomen and an intra-abdominal mass encompassing the entire abdomen, with present peristalsis, soft and non-tender to palpation, dull on percussion.

Laboratory tests revealed findings compatible with cholestatic jaundice: complete blood count without left shift, total bilirubin of 15.97 mg/dL at the expense of the direct fraction (13.3 mg/dL), elevated transaminases (ALT = 265 mg/dL and AST = 219 mg/dL), and elevated alkaline phosphatase (857 mg/dL).

A CT scan and subsequently an MRCP showed a dilated intrahepatic bile duct due to a large oval-shaped image of liquid density, with well-defined borders, located at the hepatic hilum extending caudally to the lower abdomen, measuring up to 17.7 x 21.4 x 28.3 cm, displacing intestinal loops and the

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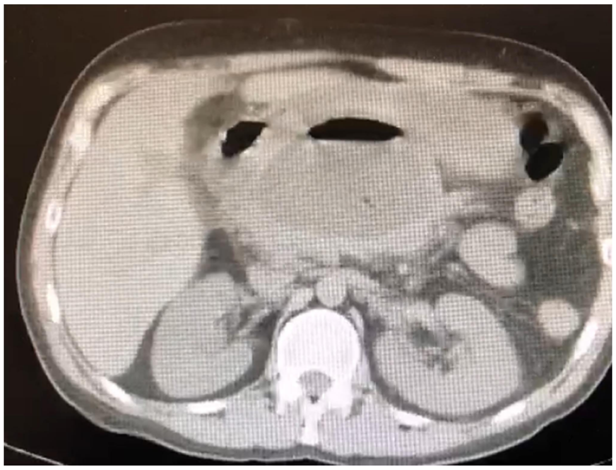

Figure 1. Simple CT bile duct cyst (2 months before surgery)

right kidney, corresponding to a type IV-A choledochal cyst.

Given the high risk of bile duct malignancy, after preoperative exams, the patient underwent a complete resection of the extrahepatic bile duct with a Whipple procedure (duodenopancreatectomy, reconstruction with Hepp-Couinaud hepaticojejunostomy, and cholecystectomy). Findings included a choledochal cyst approximately 30x25 cm with 8 liters of biliary content extending from the hepatic border to the iliac arteries. The surgery involved approximately 4000 milliliters of blood loss and a surgical time of 840 minutes.

Postoperative Evolution: The immediate postoperative evolution was torpid within the first 24 hours due to hemodynamic instability and suspected postoperative bleeding, necessitating an exploratory laparotomy that revealed 2000 cc of hemoperitoneum and layer bleeding near the pancreatojejunostomy site. Subsequently, the patient continued to evolve adequately and was discharged on the 19th postoperative day. The surgical specimen's histopathological analysis revealed acute and chronic inflammatory processes, fibrosis, vascular ectasia, and hemorrhagic foci in the pancreatic parenchyma and the choledochal cyst wall.

Discussion

In this case report, the patient presented with right upper quadrant pain, jaundice, an abdominal mass, and cholestatic alterations. Initially, based on our diagnostic suspicions, the first step should have been to request a liver and bile duct ultrasound as it is the initial evaluation method, distinguishing biliary calculi, hepatic cystic disease, and biliary tract cystic disease or agenesis. In our case, a CT scan was chosen to expedite surgical planning.

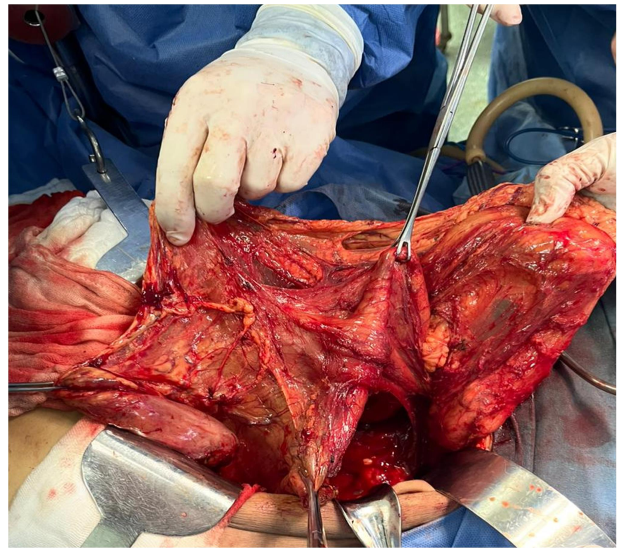

Figure 2. Wall of bile duct cyst after drainage of 8 liters of liquid content

If bile duct dilation with an evident obstructive factor is detected, the next test should be MRCP, which has good sensitivity (80-100%) (8).
Treatment for types I and IV choledochal cysts involves complete excision of the extrahepatic bile duct cyst, up to the level of communication with the pancreatic duct, cholecystectomy, and restoration of bilioenteric continuity (5,6).

In cases of severe chronic pancreatitis associated with a choledochal cyst and atrophic pancreatic head due to an anomalous junction of the pancreatic and bile ducts, a duodenopancreatectomy may be indicated (9,10). The patient in this report had a symptomatic type IV-A choledochal cyst according to Todani's classification; hence, a high-complexity surgical procedure (Whipple) was opted for to avoid residual neoplasia risks due to incomplete cyst resection (6, 11).

Conclusion

Due to the risk of infection or malignancy, the principle of treatment is bile duct resection with bilio-digestive reconstruction. In this patient, the cyst measured approximately 30 centimeters, involving vascular structures with a point of no return in dissection, leading to the decision to perform a Whipple procedure, with favorable evolution. The management of this type of case requires a careful and detailed evaluation prior to choosing the surgical procedure to be used, to avoid complications.

Conflicts of interests

The authors declare no conflicts of interests.

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