Cirujano General
January-March 2021
Vol. 43, no. 1 / p. 40-46

Experience in the management of common bile duct cyst in a general surgery service. Report of four cases

Experiencia en el manejo del quiste de colédoco en un servicio de cirugía general. Reporte de cuatro casos

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Keywords:
Cyst, common bile duct, adult.

Palabras clave:
Quiste, colédoco, adulto.

ABSTRACT

Choledochal cysts are dilatations of the biliary tract that occur mainly in females during childhood. In adulthood a high index of suspicion is required, especially in rural hospitals where there is no technology to diagnose them. Management can be by endoscopic or surgical approach, depending on the classification of the cyst. It is well determined that the older the patient is at the time of diagnosis, the greater the chances of malignant transformation, with up to 30% reported. In this report four female patients between 14 and 43 years of age were included. Three of them were not diagnosed as choledochal cyst of first intention; instead, they were diagnosed as hepatic cyst, acute cholecystitis, and biliary tract stenosis. The main symptom was abdominal pain, and only one of the patients showed jaundice and a palpable abdominal mass. Three of the patients presented a type I cyst and the other a type IV A cyst. All of them underwent open surgery with resection of the cyst and Roux-en-Y hepatic-jejunal-anastomosis. There were no postoperative complications. The histopathological study was negative for malignancy in all cases.

RESUMEN

Los quistes de colédoco son dilataciones de la vía biliar que se presentan principalmente en el sexo femenino durante la infancia, en la etapa adulta se requiere un alto índice de sospecha, sobre todo en hospitales rurales donde no existe la tecnología para diagnosticarlos. El manejo puede ser por abordaje endoscópico o quirúrgico, dependiendo de la clasificación del quiste. Está bien determinado que, a mayor edad durante el diagnóstico, las posibilidades de transformación maligna también son mayores, reportándose hasta en 30%. Se incluyeron cuatro pacientes mujeres entre 14 y 43 años. A tres no se les realizó el diagnóstico de quiste de colédoco de primera intención, siendo diagnosticadas como quiste hepático, colecistitis aguda y estenosis de la vía biliar. El síntoma principal fue dolor abdominal, sólo una de ellas mostró ictericia y masa palpable. Tres presentaron quiste tipo I y la otra tipo IV A. A todas se les realizó cirugía abierta con resección del quiste y hepatoyeyunoanastomosis en Y de Roux. No hubo complicaciones postoperatorias. El estudio histopatológico fue negativo a malignidad en todos los casos.

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Received: 03/26/2019
Accepted: 10/10/2019

INTRODUCTION

Choledochal cysts (CC) are congenital cystic dilatations of the biliary tree. The incidence is 1:100,000 to 1:150,000 in populations of Western countries. They mainly affect women and although sometimes they are diagnosed in childhood, up to 20% of patients are adults when the diagnosis is done.

The risk of developing cholangiocarcinoma in choledochal cysts increases with age, and they occur more frequently in types I (68%) and IV (21%),[1,2] which supports their complete removal when diagnosed. Several theories have been proposed to explain the pathogenesis of common bile duct cysts. The current and most accepted is the anomalous union of the pancreatic and biliary ducts outside the

How to cite: Reyes-García MA, Martínez-Bello A. Experience in the management of common bile duct cyst in a general surgery service. Report of four cases. Cir Gen. 2021; 43(1): 40-46.

duodenal wall, forming a single channel, which can reach a length between 10 and 45 mm. This anomaly causes reflux of pancreatic secretions into the biliary tree and because the pressure of the pancreatic duct is greater than the pressure of the biliary duct, it results in activation of pancreatic enzymes, alteration of the biliary composition, local inflammation, and damage to the biliary epithelium, causing weakness and dilatation of the biliary duct wall.[3,4]

The most used classification is that made by Todani and collaborators, who classify the common bile duct cyst in five types:[3]

Type I: saccular or cystic dilatation of the extrahepatic bile duct.

Type II: common bile duct diverticulum.

Type III: dilatation of the intraduodenal portion of the common bile duct (choledococele).

Type IV A: multiple intrahepatic and extrahepatic cysts.

Type IV B: multiple extrahepatic cysts.

Type V: intrahepatic cysts (Caroli's disease).

Less than half of patients present with the typical clinical triad of abdominal pain, jaundice, and an abdominal mass. In more than half of the cases the symptoms are nonspecific.[3,4] Since incomplete excision of the cyst can lead to recurrence, symptoms and malignant transformation within the remnant tissue, current surgery involves removal of the entire cyst (including the gallbladder) and restoration of biliary-enteric continuity.[5]

PRESENTATION OF CLINICAL CASES

Case report 1: a 14-year-old female with four-year evolution of colicky pain in the right hypochondrium. Four months prior to her admission she presented more frequent episodes, adding jaundice, coluria, acholia, nausea, and vomiting. She was admitted with jaundice, pain on palpation in the right hypochondrium and epigastrium, and an abdominal mass measuring approximately 15 × 15 cm. Liver function tests showed mixed hyperbilirubinemia (total bilirubin: 7.1 mg/dl, direct bilirubin: 3.7 mg/dl, increased alkaline phosphatase: 651 U/l). The ultrasonography scan revealed calculous cholecystitis and choledocholithiasis versus common bile duct cyst. The computed tomography (CT) scan showed a calculous cholecystitis and cystic dilatation of the bile duct *(Figure 1)*. The cholangial-resonance imaging showed a 72 mm choledochal cyst with intrahepatic bile duct dilatation *(Figure 2)*. Elective surgery was performed where a gallbladder with lithiasis and a type I choledochal cyst according to Todani's classification attached to duodenum measuring 17 × 10 × 10 cm was found *(Figure 3)*.

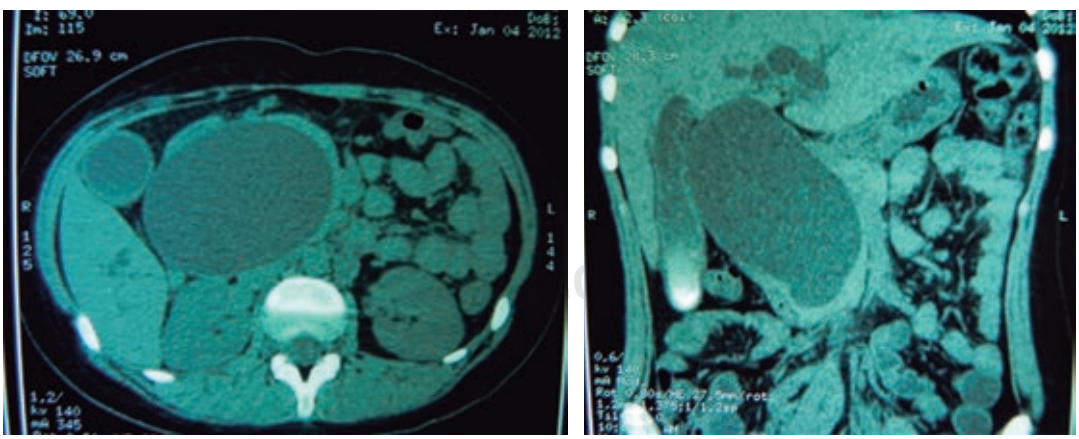

Figure 1: *Axial tomography scan showing a cystic image of the common bile duct, with thickening of the gallbladder wall and presence of stones in the gallbladder.*

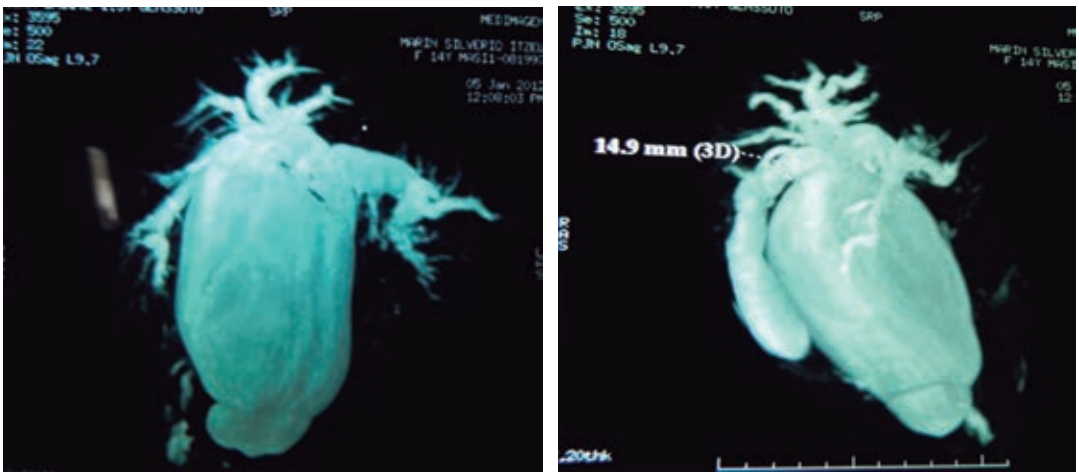

Figure 2: *MRI cholangiopancreatography showing a giant common bile duct cyst type I.*

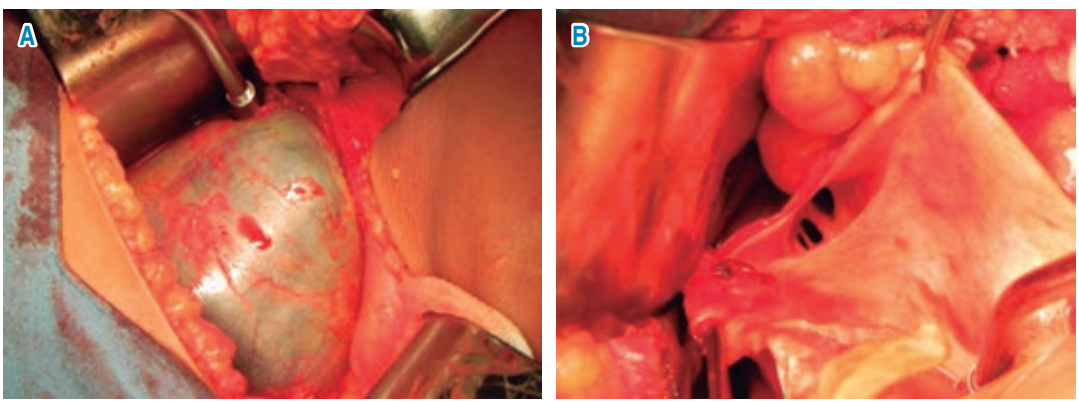

Figure 3: A) *Image of a common bile duct cyst once the adhesions were released.* ***B)*** *After the cyst is opened, the hepatic ducts and the cystic orifice are identified.*

Case report 2: a 43-year-old woman presented with colicky pain in the right hypochondrium, without accompanying symptoms; she attended a rural hospital, where she underwent open cholecystectomy during which an increase in the diameter of the common bile duct was observed. It was decided to perform a choledochotomy with T-tube placement. In the outpatient clinic, a T-tube cholangiography was requested, which showed a common bile duct cyst, so she was referred to our hospital. She was admitted without jaundice and an open Kerr type tube with biliary output. Liver function tests were within normal parameters and only shoed hypoalbuminemia. Surgery was performed with findings of a common bile duct cyst measuring 15 × 8 × 7 cm attached to the duodenum, stomach, pancreas, portal vein and vena cava. A right and left hepatic duct with a diameter of 1 cm was found.

Clinical case 3: a 15-year-old female, with a history of right hypochondrium pain and fever. The ultrasonography scan revealed a hepatic abscess. She underwent exploratory laparotomy where a giant tension common bile duct cyst with necrosis and adhesions to the gallbladder was found. Choledochotomy, aspiration and primary closure with placement of a Penrose drainage was performed. On the second postoperative day, 400 cm^3 of bile came out through the drainage and, by ultrasound,

free liquid in the cavity was seen, for which reason she underwent a new laparotomy. In this second surgery drainage, cholecystectomy, and placement of a Kerr probe in the cyst bag were performed. She was referred to our hospital, where she was maintained with medical treatment and later sent to outpatient care. A T-tube cholangiography revealed a cystic pocket compatible with a common bile duct cyst. A scheduled surgery was performed, and a common bile duct cyst of approximately 8 × 5 cm with a T-tube inside and multiple adhesions was detected *(Figure 4)*.

Case report 4: a 43-year-old woman presented with two-year evolution of colicky pain in the right hypochondrium. Two months prior to her referral to our hospital she presented with increasing pain intensity, accompanied by nausea, vomiting and fever. An endoscopic retrograde cholangiopancreatography (ERCP) was performed, which reported common bile duct dilatation measuring 25 mm of the intrahepatic and extrahepatic bile duct. A sphincterotomy and stent placement were performed. The cytology brushing was negative for malignancy. She was sent to our hospital, where a cholangial-resonance imaging was performed, which showed fusiform dilatation of the main biliary tract, and two more smaller dilatations in the right and left hepatic duct. A diagnosis of type IV A choledochal cyst was made. Liver function tests showed a serum alkaline phosphatase level of 595 U/l, an aspartate aminotransferase (TGO) level of 139 U/l, and alanine aminotransferase (TGP) level of 181 U/l and a total bilirubin level of 1.2 mg/dl.

All patients underwent cyst resection and Roux-en-Y choledochal-duodenal-anastomosis, and two of them also underwent cholecystectomy. They were discharged without complications *(Table 1)*.

DISCUSSION

The choledochal cyst (CC), although rare, is a well-described clinical entity, affecting women more frequently. Even though choledochal cysts are diagnosed in infancy or childhood, up to 20% of patients are adults when the diagnosis is made. The risk of a choledochal cyst transforming into a cholangiocarcinoma increases with age and occurs more frequently in types I (68%) and IV (21%),[1,2] which justifies its complete removal when diagnosed. The most accepted theory is the anomalous union of the pancreatic and biliary ducts outside the duodenal wall, forming a single duct, which can reach a length between 10-45 mm.[6] The most frequent symptoms are right upper abdominal pain and jaundice. Fever may also

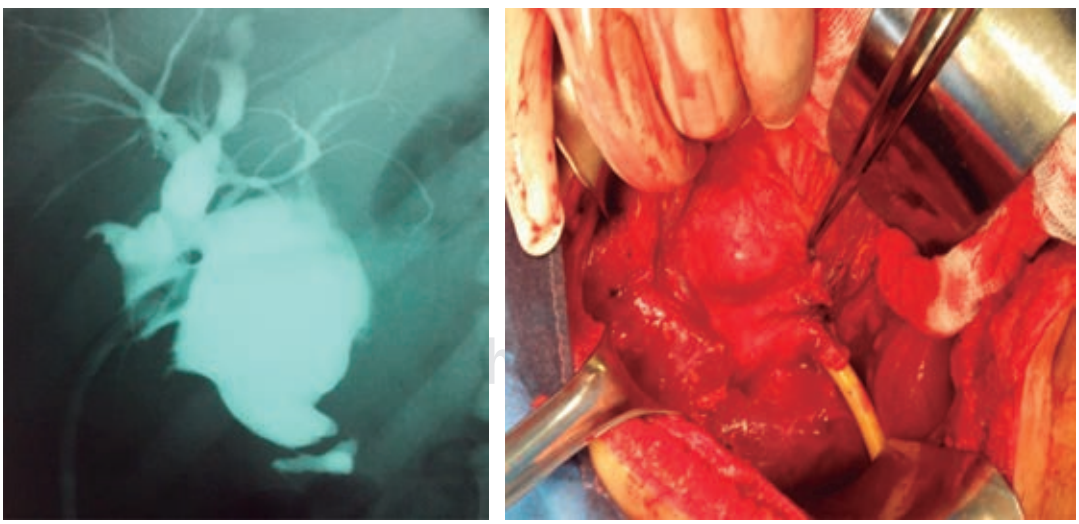

Figure 4: *Cholangiography with a Kerr probe demonstrating the presence of a common bile duct cyst; intraoperative findings exhibiting abundant adhesions and the probe being inserted into the cyst cavity.*

Table 1: Clinical and paraclinical features and surgical findings of patients with common bile duct cyst.

	Case 1	Case 2	Case 3	Case 4
Sex	Female	Female	Female	Female
Age (years)	14	43	15	43
Pain	Yes	Yes	Yes	Yes
Jaundice	Yes	No	No	No
Abdominal palpable mass	Yes	No	No	No
Classic clinical triad	Yes	No	No	No
Fever	No	No	Yes	Yes
LFT abnormalities	Yes	No	No	No
Ultrasound	Vesicular lithiasis Probable common bile duct cyst	Lithiasis cholecystitis	Hepatic abscess	Bile duct dilatation
Diagnosis	Cholangial-resonance	Cholangiography with a Kerr probe	Cholangiography with a Kerr probe	Cholangial-resonance
Type of cyst	Todani I	Todani I	Todani I	Todani IV A
Surgery	ERYHJA	ERYHJA	ERYHJA	ERYHJA
Hospital stay (days)	6	6	6	6
Complications	No	No	No	No
Histopathological	Benign	Benign	Benign	Benign

LFT = liver function tests; ERYHJA = excision and Roux-en-Y hepato-jejunal-anastomosis.

be detected when associated with cholangitis. On physical examination, an abdominal mass may be palpated in the right upper quadrant.[6,7] In our patients, all of them started with pain and two of them (50%) had fever as a symptom of cholangitis. The classic triad of abdominal pain, jaundice and an abdominal mass is present in less than 15% of adult cases. We saw it in only one patient. A study of 20 patients revealed that 100% were female, 19 (95%) had a type I choledochal cyst, while one had a choledochal cyst type IV. The main symptom was abdominal pain in 15 women (75%) and the other five (25%) had jaundice and/or cholangitis, which coincides with the findings seen in our patients. The diagnostic suspicion was made by ultrasonography scan in 50% of the cases and 18 (90%) were diagnosed by magnetic resonance cholangiopancreatography (MRCP).[8] In our study we observed that with ultrasonography scan, being an operator-dependent study, only one patient had a choledochal cyst suspected, and the diagnosis was attained by means of a Kerr probe cholangiography and two of them by MRCP. Two patients were operated on under suspicion of another diagnosis. The Todani classification is widely adopted for the classification of choledochal cysts and the choice of surgical management. The concept of complete excision is the ideal treatment. In proximal cyst removal the procedure must be performed carefully to protect the portal vein and hepatic artery. In distal cysts, the cyst usually extends into the pancreas, so excision may be difficult due to surrounding adhesion to the pancreas. Sometimes complete excision is not possible due to postoperative risk, pancreatic leakage, bleeding, and peritoneal infection.[9,10] However, the unresected intrapancreatic choledochal cyst gives rise to the formation of a dead space within the pancreas. Reflux of intestinal secretions results in activation of pancreatic enzymes in the remaining cyst, which can lead to infection, stone formation, and increased risk of malignancy.[11] Type I choledochal cyst, which has no obvious

relation to the pancreatic duct, intrapancreatic cyst or apparently normal bile duct, shall be completely excised and the stump is sutured. In our study, three patients were carriers of type I choledochal cysts, so total resection of the cyst was performed, taking special care in the intrapancreatic portion and in the dissection of the portal adhesions. In type IV choledochal cysts according to Todani's classification with extrahepatic and intrahepatic cysts, the extrahepatic cyst must be completely excised. If the intrahepatic cyst is limited, a partial hepatectomy with biliary-enteric reconstruction is performed.[12] In Mexico, Martínez-Ordaz carried out a study that analyzed 23 patients in 17 years that were operated on. Of these, 70% were women, 87% presented with pain and only four of them had the classic clinical triad; 74% had a type I choledochal cyst. Patients with type I and IV A cysts underwent resection of the cyst and Roux-en-Y choledochal-jejunal-anastomosis by open surgery. Three patients were reoperated for biliary leakage. There were no deaths.[13]

The laparoscopic approach is gradually replacing open surgical treatment of choledochal cysts in pediatric patients.[14] In contrast, laparoscopically choledochal cyst excision in adults is technically difficult and associated with a high rate of complications and conversion into open surgery; however, some authors have reported results like those obtained in children. In a study of 20 patients, Hirdaya found that laparoscopic excision could be completed in 16 cases (80%), while the other four (20%) required conversion to the open approach. The main reason for conversion was technical difficulty due to the initial learning curve, the presence of adhesions, and inflammation of the cyst and/or duct wall. There were no deaths.[8] In a study of 110 patients, where half were under 16 years of age, Palanisamy et. al. observed that type I cysts were the most common (71.82%) with an average size of 4.67 ± 1.59 cm. They were operated under laparoscopic approach. They compared the results obtained in the group of children versus adults and demonstrated that minimally invasive surgery had better results in pediatric patients with a lower conversion to open approach rate, a shorter hospital stay and a lower rate of complications. The reported hospital mortality after laparoscopic choledochal cyst excision is 1.8% and that of open choledochal cysts excision is 3 to 4%.[15] Altered anatomy, presence of adhesion and lack of palpatory sensation during the laparoscopic excision make it technically difficult and increase the risk of injury. In our hospital unit the patients had a good postoperative evolution, with no morbidity or mortality. In our population there is still no experience in the laparoscopic approach due to the low number of patients with choledochal cysts.

REFERENCES

1. Wiseman K, Buczkowski AK, Chung SW, Francoeur J, Schaeffer D, Scudamore CH. Epidemiology, presentation, diagnosis, and outcomes of choledochal cysts in adults in an urban environment. Am J Surg. 2005; 189: 527-531.
2. Singham J, Yoshida EM, Scudamore CH. Choledochal cysts: part 2 of 3: diagnosis. Can J Surg. 2009; 52: 506-511.
3. Piriz-Momblant A, Figueras-Torres B, Gómez-Claro M. Choledochal cyst. Review of the subject. Apropos of a patient. Rev Inf Sci. 2016; 95: 508-523.
4. Brunicardi FC. Schwartz principles of surgery. 10th ed. Mexico: McGraw Hill; 2015.
5. Ronnekleiv-Kelly SM, Soares KC, Ejaz A, Pawlik TM. Management of choledochal cysts. Curr Opin Gastroenterol. 2016; 32: 225-231.
6. Le Roy B, Gagniere J, Filaire L, Fontarensky M, Hordonneau C, Buc E. Pancreaticobiliary maljunction and choledochal cysts: from embryogenesis to therapeutics aspects. Surg Radiol Anat. 2016; 38: 1053-1060.
7. Robertson JF, Raine PA. Choledochal cyst: a 33-year review. Br J Surg. 1988; 75: 799-801.
8. Nag HH, Sisodia K, Sheetal P, Govind H, Chandra S. Laparoscopic excision of the choledochal cyst in adult patients: An experience. J Minim Access Surg. 2017; 13: 261-264.
9. Saluja SS, Nayeem M, Sharma BC, Bora G, Mishra PK. Management of choledochal cysts and their complications. Am Surg. 2012; 78: 284-290.
10. Cho MJ, Hwang S, Lee YJ, Kim KH, Ahn CS, Moon DB, et al. Surgical experience of 204 cases of adult choledochal cyst disease over 14 years. World J Surg. 2011; 35: 1094-1102.
11. Khandelwal C, Anand U, Kumar B, Priyadarshi RN. Diagnosis and management of choledochal cysts. Indian J Surg. 2012; 74: 29-34.
12. Liu Y, Sun J, Guo S, Liu Z, Zhu M, Zhang ZL. The distal classification and management of choledochal cyst in adults: based on the relation between cyst and pancreatic duct. Medicine (Baltimore). 2017; 96: e6350.

13. Martínez-Ordaz JL, Niño-Solís J. Adult common bile duct cysts. Cir Cir. 2010; 78: 61-66.
14. Liem NT, Pham HD, Dung le A, Son TN, Vu HM. Early and intermediate outcomes of laparoscopic surgery for choledochal cysts with 400 patients. J Laparoendosc Adv Surg Tech A. 2012; 22: 599-603.
15. Senthilnathan P, Patel ND, Nair AS, Nalankilli VP, Vijay A, Palanivelu C. Laparoscopic management of choledochal cyst-technical modifications and outcome analysis. World J Surg. 2015; 39: 2550-2556.

Ethical considerations and responsibility: The authors declare that they followed their center's protocols on the publication of patient data and safeguarded their right to privacy through the confidentiality of their data.

Funding: No financial support was received for this work.

Disclosure: The authors declare that there was no conflict of interest in carrying out this work.

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