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Management of Choledochal Cyst in Adults: A Four-Year Surgical Experience at a Cambodian Tertiary Center

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ABSTRACT

Introduction

Choledochal cyst is a congenital or acquired dilation of the biliary tree, affecting the extrahepatic and/or intrahepatic bile ducts. It is more prevalent in females and Asian populations. Early diagnosis and appropriate surgical management are essential to prevent complications and ensure optimal outcomes. This paper aimed to describe the clinical presentation, diagnostic approaches, surgical management, and postoperative outcomes of adult patients with choledochal cysts at a Cambodian tertiary center.

Methods

We conducted a retrospective case series of seven adult patients with choledochal cysts who underwent surgical treatment between January 2020 and December 2023 by checking medical records. We collected data on symptoms, imaging, Todani type classification, surgical procedure, and postoperative outcomes. Data were shown as mean, median, frequency and percentage

Results

Seven patients (five females, two males) were included, with a mean age of 33.7 years. Common symptoms include abdominal pain and jaundice. According to Todani's classification, these included 1 case of type Ia, 3 cases of type Ic, and 3 cases of type IVa. All patients underwent complete cyst excision with Roux-en-Y hepaticojejunostomy and cholecystectomy. No malignancy was found on histology in any case. The median follow-up duration was 24 months.

Conclusion

Choledochal cysts should be considered in the differential diagnosis of adult patients presenting with epigastric or right upper quadrant pain, jaundice, or signs of cholangitis. Thorough preoperative imaging and classification are essential for planning surgical intervention. Complete cyst excision with Roux-en-Y hepaticojejunostomy remains the standard treatment for extrahepatic types. Long-term follow-up is necessary for monitoring potential complications and early detection of malignant transformation.

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Keywords: Choledochal cyst, Anomalous pancreaticobiliary duct union, Roux-en-Y hepaticojejunostomy.

Introduction

A choledochal cyst is the congenital dilatation of the bile ducts. This condition is relatively rare, with an estimated incidence of 1 in 13,000–15,000 in Western populations. However, this condition is far more common in the East, with an incidence of 1 per 1000 in Japan (1, 2).

The precise etiology of choledochal cysts is not well understood, although an anomalous pancreaticobiliary duct union (APBDU) is a generally accepted hypothesis (3). A long common channel (>10 mm) proximal to the ampullary sphincter is thought to predispose to pancreatic enzyme reflux into the biliary tree with consequent biliary duct inflammation and pressure, leading to duct dilation (4–6). The theory is substantiated by the increased occurrence of APBDU among patients with choledochal cyst (approximately 30–96%) compared with 2% in the general population (3, 7). Experimental studies have corroborated the association between APBDU and choledochal cysts; however, this anomaly cannot be solely responsible, given the variable incidence in patients with choledochal cysts (3, 5).

In Cambodia, choledochal cysts in adults are infrequently encountered. A prior report from the Khmer-Soviet Friendship Hospital, documented in a thesis by Em Sokhom, identified only five adult cases over a three-year period (2008–2010). At Calmette Hospital, a national tertiary referral institution, seven adult cases were managed over a four-year span. These figures underscore the scarcity of local data regarding the presentation and management of choledochal cysts in the adult population. Accordingly, this study aims to describe the clinical characteristics, diagnostic modalities, surgical management, and postoperative outcomes of adult patients treated for choledochal cysts at our center.

Methods

Definition and classification

Choledochal cysts (CCs) are congenital or acquired dilations of the bile ducts, commonly associated with an anomalous pancreaticobiliary duct union. Todani classification was used to categorize CCs into types I–V based on the location and pattern of dilation (8–10). Type I is the most common (50–80%) one, in which the cystic or fusiform dilation of the extrahepatic bile duct, is subdivided into Ia (diffuse cystic), Ib (segmental), and Ic (fusiform with possible intrahepatic extension). Type II accounted for about 2%, in which there is a diverticulum of the extrahepatic bile duct. Type III accounted for less than 5% in which there is an intraduodenal dilation of the distal common bile duct. Type IV (15–35%) consists of multiple dilations with IVa involves both intra- and extrahepatic ducts; IVb is limited to extrahepatic ducts. Finally, type V, or is called Caroli's disease which accounted for about 20% in which there are intrahepatic ductal dilations, often associated with congenital hepatic fibrosis (**Figure 1**).

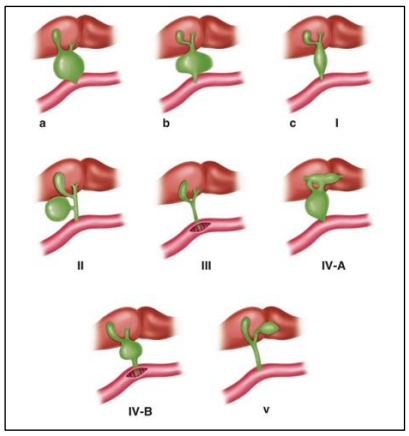

Figure 1: Todani classification of choledochal cyst (7)

Setting and data collection

We conducted a retrospective case series at the Department of Surgery, Calmette Hospital, Phnom Penh, Cambodia. The study included adult patients aged ≥18 years diagnosed with CCs and treated surgically between January 1st, 2020, and December 31st, 2023. Eligible patients were identified through operative logs, imaging records, and admission files. The following variables were extracted: from the medical records: Demographics: Age, sex, clinical presentation such as onset and duration of symptoms, imaging findings (US, MRCP, CT, ERCP), cyst types, surgical details (type of procedure, intraoperative findings, and postoperative course (complications, length of hospital stays, follow-up data). Also, patients were followed up every three months for the first two years postoperatively and annually thereafter. At each visit, a clinical examination, liver function test, and abdominal ultrasound were performed. Follow-up information was obtained from hospital records or by direct phone contact with patients. Data were shown as mean, median, frequency and percentage.

Results

Demographic characteristics

A total of seven adult patients diagnosed with choledochal cysts underwent surgical management during the study period. There were five females (71.4%) and two males (28.6%), with a mean age of 33.7 years (range: 25–54 years). Four patients were from different provinces, and 3 others were from Phnom Penh. Occupations included 2 office workers, 2 garment workers, 1 electrician, 1 blacksmith, and a farmer. None had prior hepatobiliary surgery. One patient had a history of recurrent biliary colic, and another had previously documented pancreatitis (**Table 1**).

Table 1: Demographic characteristics of the 7 patients with CCs

Variables	Freq.	%
Ages, mean in years (SD)	33.7 (9.7)	
Range, years	25-54	
Sex		
Male	2	28.6
Female	5	**71.4**
Occupation		
Garment worker	2	28.6
Office worker	2	28.6
Blacksmith	1	14.3
Electrician	1	14.3
Farmer	1	14.3
Province		
Phnom Penh	3	**42.9**
Kandal	1	14.3
Kampot	1	14.3
Kep	1	14.3
Sihanouk Ville	1	14.3
Mean hospital stay in days (SD)	7.0 (0.58)	
Range, days	6-8	

Clinical presentation and imaging and surgical procedures

According to **Table 2**, the most common symptoms were abdominal pain (7/7, 100%) and jaundice (4/7, 57.1%). One patient presented with symptoms of acute cholangitis and another with acute pancreatitis. A palpable mass was not reported in any case. All patients underwent transabdominal ultrasound as the initial investigation. MRCP was used in 4 patients (57.1%). CT scan was used in 4 patients (57.1%). Both CT and MRCP were used in combination in 1 patient (14.3%). One patient underwent ERCP for diagnostic and therapeutic purposes. Imaging confirmed that choledochal cysts were present in all cases, with no evidence of malignancy. Four patients were classified into type I (3 patients as type Ic, one patient as type Ia), and 3 patients were identified as type IVa (**Figure 2A & B**).

Table 2: Clinical and imaging presentation and surgical procedures of CCs

Variables	Freq.	%
Symptoms		
Abdominal pain	3	42.9
Pain, jaundice	3	42.9
Pain, jaundice, pancreatitis	1	14.3
Types of choledochal cysts		
Ia	1	14.3
Ic	3	42.9
IVa	3	42.9
History		
None	5	71.4
Biliary colic	1	14.3
Pancreatitis	1	14.3
Imaging*		
US/CT	3	**42.9**
US/MRCP	2	28.6
US/CT/MRCP	1	14.3
US/ERCP/MRCP	1	14.3
Surgery performed		
CCE, CEx, R-YHJ	7	100.0

** US - ultrasound, MRCP - magnetic resonance cholangiography, ERCP - endoscopic retrograde cholangiopancreatography, CCE - cholecystectomy, CEx - complete excision of cyst, R-YHJ - Roux-en-Y hepaticojejunostomy*

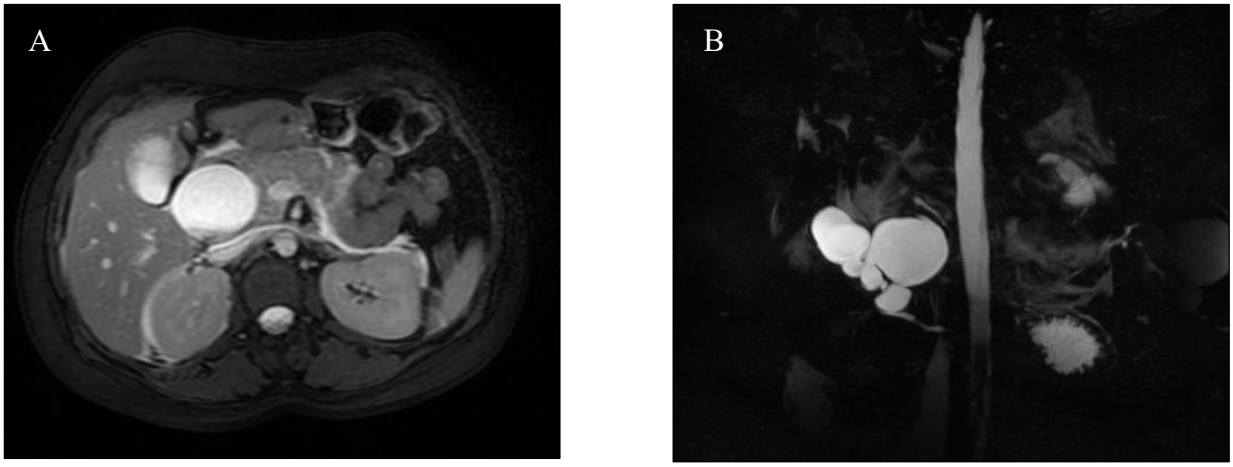

Figure 2: (A). MRCP showing type I-A saccular choledochal cyst (cross section), (**B**). MRCP of Type I-A CCs with APBDU

All patients underwent open complete cyst excision, along with cholecystectomy and Roux-en-Y hepaticojejunostomy**.** No intraoperative complications were reported.
The median hospital stay was 7 days (range: 6–8 days). Postoperative recovery was uneventful in all patients. Median duration of the follow up was 24 months (range: 12–36 months). No patient developed anastomotic stricture, cholangitis, or malignant transformation. Moreover, liver function tests remained within normal range, and all patients reported no recurrence of symptoms **(Figure 3A & B)**.

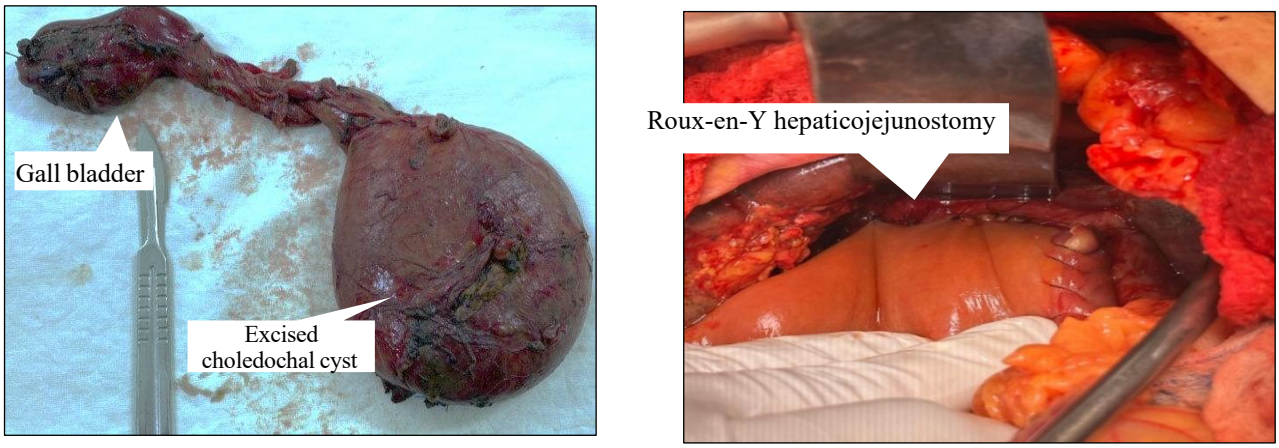

Figure 3: (A). Completely excised choledochal cyst with the gall bladder, **(B)**. Choledochal cyst excision the completion of Roux-en-Y hepaticojejunostomy

Discussion

Choledochal cysts are rare congenital biliary anomalies, with most cases diagnosed in childhood. While the classical triad of right upper quadrant pain, palpable mass, and jaundice is seen in up to 85% of pediatric cases, it appears in only about 25% of adults. In our series, abdominal pain and jaundice were the most frequent presenting symptoms, consistent with findings from international case series and studies by Erraoui G (12), and Honnavara Srinivasan P, et al. (13), reported comparable patterns of symptom presentation in adult patients with CCs. Type I and type IVa cysts were the most common types observed, which is consistent with the global literature. Studies in India (13) and in the U.S (14) also reported a predominance of type I and type IVa cysts in adult cases. Moreover, the female predominance in our case series

is also widely observed in previous research, suggesting a possible hormonal or genetic component, though the exact cause remains unclear.

Diagnostic imaging played a crucial role in our preoperative planning. While ultrasonography was effective as an initial tool, MRCP and CT scans were more definitive in classifying the cyst type and identifying associated complications. This is in line with studies by Lee et al. (1) and Park et al (3) who emphasized the superiority of MRCP in diagnosing choledochal cysts and detecting APBDU. However, in resource-limited settings like ours, access to advanced imaging modalities can be inconsistent, which may impact early and accurate diagnosis.

The management approach of CCs depends on the cyst type and the extent of hepatobiliary pathology. As a rule, all cysts should be resected; and bile flow should be restored. Early surgical excision of CCs is recommended, as Diao et al (15). The aims of preoperative management are a complete cholangiographic definition of the extent of the cystic process and associated ductal pathology and control of biliary infections.

Surgical excision remains the treatment of choice for extrahepatic CCs. All patients in our study underwent complete cyst excision with Roux-en-Y hepaticojejunostomy, which is widely accepted as the gold standard. The use of hepaticojejunostomy over hepatico-duodenostomy was guided by existing literature indicating lower risks of bile reflux and gastritis. Our outcomes were favorable, with no postoperative strictures, infections, or malignancy noted during follow-up, which suggests that complete resection with proper reconstruction provides good long-term results.

Despite these positive outcomes, our study has several limitations. The small sample size limits the generalizability of our results, and the case series nature of the study may introduce bias. Furthermore, follow-up duration was limited to a median of 24 months, which may not be sufficient to detect late complications such as malignancy or anastomotic strictures. Additionally, our institution lacks laparoscopic or robotic facilities, which are increasingly used in specialized centers and may offer better cosmetic and postoperative outcomes.

Another challenge is the lack of standardized national guidelines for managing adult CCs in Cambodia. This leads to variability in diagnostic and treatment approaches across institutions. Our findings highlight the need for increased awareness and training in hepatobiliary diseases, as well as the importance of multidisciplinary care in achieving optimal outcomes.

Our findings suggest that choledochal cysts in adults, although rare, can be managed effectively with early diagnosis and complete surgical excision. Reports from regional and international studies support our approach, but larger prospective studies are needed to strengthen the evidence and guide national protocols.

Conclusion

Choledochal cysts, although predominantly diagnosed in children, are increasingly recognized in adults and should be considered in any patient presenting with nonspecific biliary symptoms such as abdominal pain or jaundice. Their pathogenesis is multifactorial, with congenital anomalies such as an anomalous pancreaticobiliary duct union and embryologic maldevelopment being key contributors.

Prompt diagnosis and definitive surgical management are essential to reduce the risk of severe complications, including cholangitis, hepatolithiasis, biliary cirrhosis, spontaneous perforation, and malignant transformation. Complete cyst excision with Roux-en-Y hepaticojejunostomy remains the gold standard for treating extrahepatic CCs. Intrahepatic involvement may require segmental hepatic resection, while diffuse disease or advanced hepatic dysfunction may warrant liver transplantation.

Lifelong follow-up is recommended due to the persistent risk of cholangiocarcinoma, even after surgical resection. Increased awareness, timely referral, and comprehensive surgical care are vital for improving outcomes in adult patients with choledochal cysts, especially in low-resource settings where diagnosis is often delayed.

Abbreviation

APBDU: Anomalous pancreaticobiliary duct union, CCE: cholecystectomy, CEx: complete excision of cyst, Choledochal cysts: CCs, ERCP: endoscopic retrograde cholangiopancreatography, MRCP: magnetic resonance cholangiography, R-YHJ: Roux-en-Y hepaticojejunostomy, US: ultrasound,

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Author contributions

Conceptualization, data collection, intraoperative assistance, and manuscript drafting by Sim Pichdavong; supervision, and critical manuscript revision by H.E. Prof. Lem Dara. Main operator, surgical supervision, editing, and manuscript revision by Dr. Dara Vithiea. Dr. San Corine served as intraoperative assistance, surgical data contribution. Dr. Tha Oudom served as intraoperative assistance, Postoperative care and follow-up data interpretation. All authors have read and approved the final manuscript.

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Declaration

All data collection was conducted with prior approval from Calmette Hospital and the University of Health Sciences, Phnom Penh, Cambodia. Patient identities were protected, and informed consent was obtained from all individuals for the use of their anonymized clinical data and imaging in this publication.

Competing interest: The authors declare that they have no competing interests.

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