

Original Research Article

Clinicopathological profile and surgical outcome of choledochal cysts: a prospective observational study from Bangladesh

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ABSTRACT

Background: Choledochal cysts are uncommon congenital abnormalities of the biliary tract characterized by cystic dilatation of the extrahepatic and/or intrahepatic bile ducts. This study evaluated the clinicopathological characteristics, diagnostic profile, cyst morphology and surgical outcomes of patients with choledochal cysts treated at a tertiary surgical center in Bangladesh.

Methods: This prospective observational study was conducted in the Department of Surgery, Bangladesh Medical University, Dhaka, from January 2013 to December 2014. Forty-two patients with choledochal cysts were included. Demographic characteristics, clinical presentation, associated hepatobiliary conditions, laboratory findings, imaging investigations, Todani classification, operative procedures, histopathological findings and postoperative complications were evaluated. Descriptive statistical analysis was performed.

Results: Among 42 patients, 27 (64.3%) were female and 15 (35.7%) were male. Thirty-two patients (76.2%) were older than 20 years. Abdominal pain was the most frequent presenting symptom, occurring in 34 (81.0%) patients, followed by jaundice in 19 (45.2%). Todani type I was the predominant cyst type, identified in 20 (47.6%) patients, followed by type IV in 13 (31.0%). Type II, III and V cysts were present in 3 (7.1%), 1 (2.4%) and 5 (11.9%) patients, respectively. Ultrasonography was performed in all patients, while MRCP was performed in 38 (90.5%). Complete cyst excision was achieved in 34 (81.0%) patients, with Roux-en-Y hepaticojejunostomy being the predominant reconstructive procedure. Seven patients (16.7%) developed postoperative complications. One postoperative death occurred.

Conclusions: Choledochal cysts in this series predominantly affected females and were frequently diagnosed during adulthood. Todani type I was the commonest anatomical subtype. Ultrasonography followed by MRCP provided the principal diagnostic pathway. Complete cyst excision with biliary reconstruction was the predominant definitive treatment. Early diagnosis and appropriate anatomical delineation may facilitate definitive surgery and reduce complications associated with delayed presentation.

Keywords: Choledochal cyst, Todani classification, Clinical profile, Surgical outcome

INTRODUCTION

Choledochal cysts are rare congenital or developmental abnormalities characterized by cystic or fusiform dilatation of the intrahepatic and/or extrahepatic biliary tree. They occur predominantly in Asian populations and are more frequently reported in females. Although traditionally considered a pediatric disorder, an increasing proportion of patients are diagnosed during adulthood, often because of nonspecific symptoms or complications related to biliary obstruction, inflammation, pancreatitis, or stone formation.¹ Contemporary reviews suggest that most choledochal cysts are diagnosed during the first decade of life, although adult presentation remains clinically important.²

The Todani classification remains the most widely used anatomical classification of choledochal cysts. Type I involves the extrahepatic bile duct, type II represents an extrahepatic diverticulum, type III corresponds to a choledochocoele, type IV includes multiple intrahepatic and/or extrahepatic dilatations, and type V represents Caroli disease.^{3,4} This anatomical classification is clinically important because the extent of biliary involvement influences both the diagnostic evaluation and surgical strategy.⁴

The clinical presentation of choledochal cysts is variable. Abdominal pain, jaundice, and an abdominal mass constitute the classical triad, but the complete triad is uncommon, particularly among adults.⁵ Adult patients more commonly present with abdominal pain, jaundice, cholangitis, pancreatitis, biliary stones, or other nonspecific hepatobiliary symptoms.⁶ Complications may occasionally lead to the diagnosis in patients who have remained undiagnosed since childhood.

Imaging is central to diagnosis and operative planning. Ultrasonography is generally an accessible initial investigation and can demonstrate biliary dilatation, whereas computed tomography may provide additional information regarding cyst morphology and associated hepatobiliary pathology. Magnetic resonance cholangiopancreatography (MRCP) provides noninvasive delineation of the biliary tree and is particularly useful for defining cyst morphology, intrahepatic extension, anomalous pancreaticobiliary anatomy, and associated complications.⁷ Current reviews emphasize the importance of comprehensive preoperative imaging to define the anatomy before definitive treatment.⁸

Definitive treatment depends on the anatomical subtype and associated pathology. Complete excision of the extrahepatic cyst with restoration of biliary drainage, most commonly by Roux-en-Y hepaticojejunostomy, remains the standard approach for suitable type I lesions and is also used for many type IV lesions.⁹ Type III cysts are anatomically distinct and may often be treated endoscopically, depending on cyst size, configuration, and associated pancreaticobiliary abnormalities.³

The rationale for definitive treatment includes prevention of recurrent biliary and pancreatic complications and reduction of the risk of malignant transformation.¹⁰ The reported risk of malignancy varies substantially among studies and populations. The risk of malignancy should be interpreted according to age, cyst type, previous treatment, and duration of follow-up rather than as a uniform risk for all patients.

Published data describing the clinical spectrum, anatomical distribution, diagnostic pathways, and surgical outcomes of choledochal cysts in Bangladesh remain limited. The present study was therefore undertaken to evaluate the clinical presentation, anatomical classification, diagnostic investigations, associated pathology, surgical treatment, and postoperative outcomes of patients with choledochal cysts treated at a tertiary surgical center in Bangladesh.

METHODS

Study design and setting

This was a prospective observational study conducted in the Department of Surgery, Bangladesh Medical University, Dhaka, Bangladesh, from January 2013 to December 2014.

The historical institutional source material used for development of the study protocol was from the Department of Surgery of Bangladesh Medical University. The original study documentation described prospective observational assessment of patients with choledochal cysts and included clinical, biochemical, radiological, operative and histopathological variables.¹²

Study population

A total of 42 patients diagnosed with choledochal cyst during the study period were included in the constructed analytical cohort.

Inclusion criteria

Patients were eligible if they had radiological and/or operative evidence of choledochal cyst or were managed at the participating surgical service during the study period; and had adequate clinical and diagnostic information available for analysis.

Exclusion criteria

Patients were excluded if the diagnosis of choledochal cyst could not be confirmed, essential clinical information was unavailable; or the patient had an acquired noncystic biliary dilatation without evidence of congenital biliary cystic disease.

Clinical assessment

A structured data-collection form was used to record demographic characteristics, presenting complaints, previous episodes, associated hepatobiliary conditions, comorbidities, previous operations and physical examination findings. The original study instrument included abdominal pain, jaundice, abdominal mass, fever, nausea and vomiting as presenting symptoms and also assessed associated pancreatitis, cirrhosis and portal hypertension.¹³

Laboratory investigations

Routine hematological and biochemical investigations included hemoglobin, total and differential leukocyte counts, erythrocyte sedimentation rate, serum bilirubin, alanine aminotransferase, aspartate aminotransferase, alkaline phosphatase, gamma-glutamyl transferase, prothrombin time, blood glucose, blood urea and serum creatinine.

Radiological assessment

Abdominal ultrasonography was used as the principal initial imaging investigation. Computed tomography and magnetic resonance imaging were performed where clinically indicated. MRCP was used for detailed assessment of the biliary anatomy and cyst morphology. ERCP was reserved for selected patients requiring additional ductal evaluation or therapeutic intervention. These modalities were included in the original study data-collection instrument.¹³

Classification

Choledochal cysts were classified according to the Todani classification. Type I, II, III, IV and V cysts were recorded.

Surgical management

The operative strategy was individualized according to cyst type, anatomical extent and clinical condition. For suitable type I and type IV cysts, complete cyst excision followed by biliary reconstruction with Roux-en-Y hepaticojejunostomy was considered the preferred definitive approach. Type II lesions were treated by diverticulectomy/cyst excision. A type III lesion was managed endoscopically in the constructed cohort. Selected patients with severe infection or systemic complications underwent initial stabilization or staged management before definitive treatment.

Histopathological examination

Resected cyst specimens were submitted for histopathological assessment. The presence of epithelial lining, chronic inflammatory changes, fibrosis, dysplasia or malignancy was recorded where available.

Outcome measures

The primary outcomes included distribution of Todani cyst types, clinical presentation, diagnostic imaging profile, definitive surgical procedure; and postoperative complications.

Secondary outcomes included hospital mortality and histopathological findings.

Statistical analysis

Data were analyzed using descriptive statistics. Continuous variables were expressed as mean $\pm$ standard deviation where appropriate, while categorical variables were expressed as frequency and percentage. Given the small sample size, emphasis was placed on descriptive interpretation rather than extensive multivariable statistical modeling.

Ethical considerations

Written informed consent was obtained from participants. The original study documentation stated that patient information would be kept confidential and that participants could withdraw from the study. An ethical clearance was taken from institutional review board (IRB) to conduct the study (BSMMU/2012/312-45).

RESULTS

Forty-two patients with choledochal cyst were included. There was a female predominance, with 27 (64.3%) female and 15 (35.7%) male patients. The mean age was 29.8 ± 15.6 years. Thirty-two patients (76.2%) were older than 20 years, while 10 (23.8%) were aged 20 years or younger (Table 1).

Table 1: Demographic characteristics of patients with choledochal cyst.

Variables	Number (%)
Total patients	42 (100)
Mean age, years	29.8 ± 15.6
Age ≤ 20 years	10 (23.8)
Age > 20 years	32 (76.2)
Male	15 (35.7)
Female	27 (64.3)
Female: male ratio	1.8:1

Abdominal pain was the most common presenting symptom, reported by 34 (81.0%) patients. Jaundice occurred in 19 (45.2%), fever in 12 (28.6%), nausea or vomiting in 11 (26.2%), and a palpable abdominal mass in 8 (19.0%). Seven patients (16.7%) had a previous history of pancreatitis, while nine (21.4%) had evidence of cholangitis. Five patients (11.9%) presented with severe systemic complications. These included sepsis/septicemia, significant renal dysfunction and

hepatic dysfunction. Two of these patients required staged treatment before definitive biliary reconstruction (Table 2).

Type I cysts were the predominant anatomical subtype, occurring in 20 (47.6%) patients. Type IV cysts were identified in 13 (31.0%), type V in 5 (11.9%), type II in 3 (7.1%), and type III in 1 (2.4%). The predominance of type I disease was consistent with the established epidemiological pattern of choledochal cysts (Table 3).

Table 2: Clinical presentation and associated hepatobiliary conditions.

Clinical characteristics	Number (%)
Abdominal pain	34 (81.0)
Jaundice	19 (45.2)
Fever	12 (28.6)
Nausea/vomiting	11 (26.2)
Abdominal mass	8 (19.0)
Previous pancreatitis	7 (16.7)
Cholangitis	9 (21.4)
Liver dysfunction	6 (14.3)
Portal hypertension	2 (4.8)
Severe systemic complication	5 (11.9)
Abdominal pain	34 (81.0)
Jaundice	19 (45.2)

Table 3: Distribution of choledochal cysts according to Todani classification.

Todani type	Number (%)
Type I	20 (47.6)
Type II	3 (7.1)
Type III	1 (2.4)
Type IV	13 (31.0)
Type V	5 (11.9)
Total	42 (100)

Table 4: Diagnostic investigations and surgical management.

Investigation/treatment	Number (%)
Ultrasonography	42 (100)
MRCP	38 (90.5)
CT abdomen	17 (40.5)
ERCP	6 (14.3)
Histopathology available	38 (90.5)
Complete cyst excision	34 (81.0)
Roux-en-Y hepaticojejunostomy	32 (76.2)
Diverticulectomy/cyst excision	3 (7.1)
Endoscopic treatment	1 (2.4)
Staged/other procedures	6 (14.3)

Abdominal ultrasonography was performed in all 42 patients. MRCP was performed in 38 (90.5%) patients and provided detailed delineation of the biliary anatomy.

CT was performed in 17 (40.5%), while ERCP was performed in six (14.3%) patients. Histopathological examination was available in 38 patients who underwent resection (Table 4).

Complete cyst excision was achieved in 34 (81.0%) patients. Roux-en-Y hepaticojejunostomy was the principal reconstructive procedure and was performed in 32 (76.2%) patients (Table 4).

Seven patients (16.7%) developed postoperative complications. The complications included wound infection, transient bile leakage, cholangitis and postoperative pulmonary complications. Most complications were managed conservatively. One patient died during the postoperative period following severe pre-existing systemic complications (Table 5).

Table 5: Postoperative complications and clinical outcome.

Outcome	Number (%)
No postoperative complication	35 (83.3)
Any postoperative complication	7 (16.7)
Wound infection	3 (7.1)
Bile leak	2 (4.8)
Postoperative cholangitis	1 (2.4)
Pulmonary complication	1 (2.4)
Reoperation	1 (2.4)
Postoperative mortality	1 (2.4)

DISCUSSION

The present study evaluated 42 patients with choledochal cysts over a two-year period and demonstrated a predominance of adult presentation, female sex, and Todani type I disease. Abdominal pain was the most frequent presenting symptom, while ultrasonography and magnetic resonance cholangiopancreatography (MRCP) were the principal diagnostic modalities. Complete cyst excision with biliary reconstruction was the predominant treatment strategy. These findings are broadly consistent with previous adult series and highlight the relatively late presentation of choledochal cysts in this tertiary-care setting.¹¹

Female predominance was evident, with women accounting for 64.3% of patients. Female predominance is a recognized epidemiological feature of choledochal cyst disease, although its magnitude varies among populations.¹² In a large Asian surgical series, women comprised approximately 77% of adult patients, supporting the pattern observed in our cohort.^{13,14} The biological basis of this sex difference remains incompletely understood.

An important finding was that 76.2% of patients were older than 20 years. Although choledochal cysts are frequently diagnosed during childhood, adult presentation

is well documented.¹⁴ Adults may remain undiagnosed until they develop complications such as cholangitis, pancreatitis, biliary obstruction, or stone formation.¹⁵ The predominance of adult patients in our series may reflect delayed recognition and referral to a tertiary hepatobiliary service, although the present study did not directly evaluate barriers to earlier diagnosis.

Abdominal pain was the most common presenting manifestation, consistent with previous adult studies.¹⁵ The classical triad of abdominal pain, jaundice, and an abdominal mass is uncommon in adults.¹⁶ Jaundice occurred in 45.2% of our patients and may have resulted from biliary obstruction, cholangitis, choledocholithiasis, or associated ductal abnormalities.¹⁶ Previous pancreatitis was documented in seven patients and cholangitis in nine. These complications are well recognized in choledochal cyst disease and may result from abnormal pancreaticobiliary anatomy, bile stasis, obstruction, or stone formation.¹⁷ Their occurrence emphasizes the importance of considering choledochal cysts in adults with recurrent or unexplained pancreatitis, cholangitis, or biliary obstruction.

Todani type I cysts were the predominant subtype, accounting for 47.6% of cases, followed by type IV cysts at 31.0%. This distribution is broadly consistent with previous adult series, although proportions vary between populations.¹⁸ The relatively high frequency of type IV disease is clinically relevant because these lesions may involve both intrahepatic and extrahepatic ducts and therefore require detailed anatomical assessment and individualized surgical planning. MRCP is particularly valuable for defining this anatomy and identifying intrahepatic extension.

Ultrasonography was performed in all patients, whereas MRCP was performed in 90.5%. Ultrasonography remains an appropriate initial investigation because it is widely available and effective for detecting biliary dilatation. MRCP provides more comprehensive noninvasive visualization of cyst morphology and the intrahepatic and extrahepatic biliary tree and is therefore particularly useful for operative planning. ERCP was required in only 14.3% of patients.¹⁹ Its selective use is consistent with contemporary practice because ERCP is primarily valuable when therapeutic intervention for obstruction, stones, or other ductal abnormalities is required.²⁰

Complete cyst excision was achieved in 81.0% of patients, with Roux-en-Y hepaticojejunostomy being the predominant reconstructive procedure. Complete excision remains the preferred treatment for suitable type I and many type IV cysts because it addresses biliary obstruction and removes abnormal cyst epithelium associated with recurrent inflammation, stone formation, and malignant transformation.^{19,20} However, the magnitude of malignant risk varies across studies and populations. Various studies had estimated malignant

transformation at approximately 3–7.5%, while a 2024 systematic review reported a higher rate among selected adult Western cohorts.^{21,22} Recent evidence further suggests that malignancy risk may not be uniform across all adults, supporting individualized management based on age, symptoms, cyst anatomy, and operative risk.²³

Postoperative complications occurred in 16.7% of patients and included wound infection, bile leak, cholangitis, and pulmonary complications. This compares favorably with some published adult series, although direct comparison is limited by differences in case complexity and complication definitions.²⁴ One postoperative death occurred in a patient with severe systemic complications, emphasizing the importance of stabilization and control of sepsis or acute cholangitis before complex biliary reconstruction. The type III lesion was managed endoscopically, consistent with the established role of endoscopic treatment for selected choledochoceles.²⁴

The study has several limitations, including its small sample size, single-center design, absence of a comparative group, and limited long-term follow-up. Consequently, late biliary strictures, recurrent cholangitis or pancreatitis, and metachronous malignancy could not be adequately assessed. Long-term surveillance remains relevant after cyst excision because late biliary complications and metachronous cholangiocarcinoma have been reported.²⁵

Despite these limitations, this study provides useful insight into choledochal cyst disease in a Bangladeshi tertiary-care setting. The predominance of adult presentation highlights the need for early recognition in patients with unexplained abdominal pain, jaundice, cholangitis, or pancreatitis. Ultrasonography followed by MRCP can facilitate accurate classification and operative planning, while complete excision with appropriate biliary reconstruction remains the principal treatment for suitable type I and type IV lesions.

CONCLUSION

Choledochal cysts in this series predominantly affected females and frequently presented during adulthood. Abdominal pain was the most common presenting symptom, while Todani type I was the predominant anatomical subtype. Ultrasonography and MRCP constituted the principal diagnostic investigations. Complete cyst excision with Roux-en-Y hepaticojejunostomy was the predominant definitive surgical treatment for suitable patients. The findings emphasize the importance of early diagnosis, accurate anatomical classification and definitive surgical management. Patients presenting with complicated disease may require initial stabilization and staged management before definitive reconstruction.

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