

# Retrospective Cohort Study of Short- and Long-term Outcomes in Surgical Management of Adult Choledochal Cysts: Ten-year Experience of a Tertiary Care Centre in Karnataka, India

SHILPA MARIAPPA CASABA<sup>1</sup>, GK ADITHYA<sup>2</sup>, BN VINAY<sup>3</sup>, HG VENUGOPAL<sup>4</sup>

## ABSTRACT

**Introduction:** Choledochal cysts in adults are rare, as most are diagnosed in childhood. The concern for malignancy in adults adds to the expenses associated with evaluation and management. A aetiology can be congenital or acquired. Acquired biliary wall thinning, caused by the reflux of pancreatic juice, can be a contributing factor in adults. Long-term follow-up after surgery is essential to optimise the treatment. However, not many studies have documented the long-term outcomes of surgery.

**Aim:** To study the short- and long-term outcomes of patients undergoing surgery for adult choledochal cysts {choledochal cyst excision and Hepaticojejunostomy (HJ)}.

**Materials and Methods:** This was a retrospective cohort study of patients aged 18 years and older with choledochal cysts who were diagnosed and operated on between 2012 and 2022 in the Department of Surgical Gastroenterology, Bangalore Medical College and Research Institute (BMCRI), Bengaluru, Karnataka, India. Patients were diagnosed using imaging and subjected to open choledochal cyst excision and HJ. Outcomes like morbidity and mortality associated with the procedure, both in the short-term and long-term, were studied. Continuous variables like

age were represented as mean±standard deviation, while categorical variables (such as CBD diameter) were represented as percentages/proportions. Statistical Package for the Social Sciences (SPSS) (version 25.0) was used for statistical analyses.

**Results:** A total of 128 patients were included in the study, with a mean age of 48.92±16.20 years. Type I cysts were seen in 61.72% of the cases, while Type IV A constituted 38.28%. The mean diameter of the Common Bile Duct (CBD) in the study sample was 23.13±14.88 mm, with fusiform dilatation. Endoscopic Retrograde Cholangiopancreatography (ERCP) with stenting/stone clearance was performed in 41 patients. Short-term complications occurred in 48 (37.5%) patients, with Surgical Site Infection (SSI) being the most common early complication. Early postoperative mortality was observed in one patient (0.78%), On long-term follow-up of 83 patients, mortality occurred in six patients. A total of 18 patients complained of abdominal pain during long-term follow-up.

**Conclusion:** Choledochal cyst still remains a significant surgical disease in the adult population, especially in a tertiary care centre. Timely surgical intervention is essential to prevent the short- and long-term complications associated with this condition.

**Keywords:** Bile leak, Biliary stricture long-term outcome, Hepaticojejunostomy, Surgical intervention

## INTRODUCTION

Cystic dilatation of the bile ducts is common in children, with an incidence of 1 in 100,000 to 150,000 live births in the western population. However, this incidence could be as high as 1 in 13,500 live births in the United States [1]. The incidence is even higher in the Asian population, with an estimated incidence of 1 in 1,000, mostly reported from Japan [2]. When the same dilatation occurs in adult bile ducts, it warrants attention, as multiple associated conditions complicate the scenario [3].

Todani T et al., modified the original Alonso-Lej classification (which consisted of three types) to include five types of congenital choledochal cysts [4]. Every imaging modality-whether it be Ultrasonography (USG), Computed Tomography (CT), or Magnetic Resonance Cholangiopancreatography (MRCP)-fairly detects the finding of cystic dilatation of the CBD or any other intra- or extrahepatic bile ducts. The sensitivity of these modalities ranges from 71% to 97% for USG, 90% for CT and 96% to 100% for MRCP in the detection of bile duct cysts [5,6]. The specific diameter of the CBD at which it should be classified as a choledochal cyst is not definitively known; it is the fusiform or saccular dilatation of the CBD that makes it a choledochal cyst [3].

Although cyst excision with Hepaticojejunostomy (HJ) is the standard procedure of choice [7], morbidity and mortality associated with the procedure are not uncommon. Literature cites morbidity incidence rates ranging from 14% to as high as 64% [8]. Morbidity may include bile leaks, infections, anastomotic strictures (3.9%) and malignancy (0.7%) in the long-term [9,10]. In the field of surgery, every effort is being made to minimise these complications.

Long-term follow-up of these patients will undoubtedly enhance the understanding of the disease and treatment modalities, as they are continually evolving. The present study was carried out to highlight the short- and long-term outcomes of these patients and to optimise the therapy to the fullest.

## MATERIALS AND METHODS

This was a retrospective cohort study conducted in the Department of Surgical Gastroenterology at BMCRI, Bengaluru, Karnataka, India, where data was collected, compiled and analysed over a period of three months from January 2024 to March 2024, after obtaining ethical clearance (vide letter no. BMCRI/EC/03/24).

All cases involving patients over 18 years of age diagnosed with choledochal cysts, who underwent choledochal cyst excision in the study Institute from February 2012 to February 2022 were included in the study. Data was retrieved from the medical records.

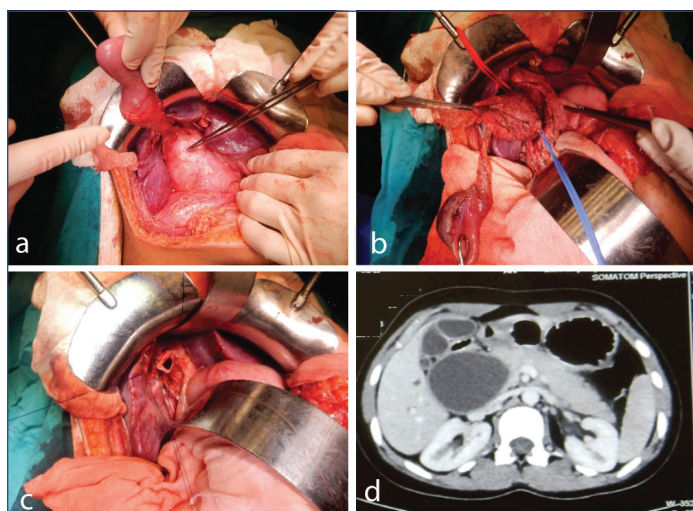
**Inclusion criteria:** Patients over 18 years of age diagnosed with choledochal cysts, patients with cystic dilatation of CBD or/and Common Hepatic Duct (CHD) and/or Intrahepatic Biliary Radicals (IHBR), significant CBD dilatation (in fusiform or saccular form) with or without CBD stones, who underwent choledochal cyst excision at the institute.

**Exclusion criteria:** Patients with bile duct malignant aetiology for CBD dilatation and those deemed high-risk for surgery were excluded.

## Study Procedure

All case sheets of patients in the study were reviewed in detail. The pattern of presentation, mode of diagnosis, type of intervention, intraoperative and immediate postoperative complications, management of complications, status at discharge, follow-up visits, long-term complications and their management were documented.

Patients presenting with acute cholangitis underwent ERCP and CBD stenting. The definitive procedure was deferred for 4 to 6 weeks in these cases. All patients underwent surgery in the form of open choledochal cyst excision and Roux-en-Y HJ [Table/Fig-1]. An abdominal drain was placed in all cases.



**[Table/Fig-1]:** Type IV A choledochal cyst: a) Large choledochal cyst in-situ; b) After the dissection of the cyst; c) Hepatic duct ready for Hepaticojejunostomy (HJ); d) CECT abdomen of the patient showing a large choledochal cyst extending to hepatic ducts making it a Type IV A choledochal cyst.

Short-term measures included 30-day morbidity and mortality, the requirement for readmission and resolution of symptoms. Long-term outcomes included long-term symptom relief (minimum of 6 months), HJ stricture rate, liver fibrosis and operative site morbidities.

## STATISTICAL ANALYSIS

Continuous variables, like age, were represented as mean±standard deviation. Categorical variables, such as CBD diameter, were represented as percentages or proportions. Statistical analyses were performed using SPSS (version 25.0).

## RESULTS

A total of 128 patients who underwent choledochal cyst excision at the institute were included in the study. Among them, 85 (66.4%) were female and 43 (33.6%) were male. The patients belonged to an age group of 19 to 75 years, with a mean age of 48.92±16.20 years [Table/Fig-2]. Abdominal pain was the main complaint, followed by jaundice, fever, pruritus and weakness. A total of 46 patients had either a history of or current clinical signs of acute cholangitis. ERCP was performed with sphincterotomy and biliary stenting in

Patient characteristics	n (%)
Total patients (N)	128
Age (years) (mean±SD)	48.92±16.20 (mean)
Females	85 (66.4%)
Co-morbidities	42 (32.8%)
<b>Presenting symptoms (n)</b>	
Abdominal pain	120
Fever	46
Jaundice	22
Vomiting	18
Pruritus	10
Cholangitis	46 (35.9%)
ERCP and intervention	41 (32%)
Multiple ERCPS	3 (2.3%)
Post ERCP pancreatitis (PEP)	2 (1.5%)
<b>Types of choledochal cyst</b>	
Type I	79 (61.7%)
Type IV A	49 (38.28%)
Hyperbilirubinaemia	40 (31%)
Elevated levels of serum transaminases	31 (24.2%)
Elevated levels of serum alkaline phosphatase	55 (42.9%)
Low serum albumin level	28 (21.88%)
Previous surgeries	26 (20.3%)

**[Table/Fig-2]:** Table showing patients' demographic and clinical characteristics.

these patients. In five cases, it was managed conservatively with antibiotics and they were subsequently taken up for surgery. One patient underwent percutaneous transhepatic biliary drainage. Three patients required multiple ERCPS with repeated stenting, which resulted in post-ERCP pancreatitis (PEP) in two of them.

Past surgeries included laparoscopic cholecystectomy in 10 patients, truncal vagotomy with gastrojejunostomy in five, appendectomy in five, CBD exploration in three, one lateral pancreatico-jejunostomy, one surgery for intestinal obstruction and one for gallbladder perforation with abdominal wall abscess.

Type I choledochal cysts were the most common, observed in 79 patients (61.7%), followed by Type IV A in 49 patients (38.3%). Other forms were not seen in the present data. Hyperbilirubinaemia was seen in 40 patients (31%), with 7 patients (5.4%) having total bilirubin levels exceeding 5 mg%. Elevated levels of transaminases and alkaline phosphatase were observed in 31 patients (24.2%) and 55 patients (42.9%), respectively. Serum albumin levels were less than 3.4 g/dL in 28 patients (21.9%), while the remainder had levels exceeding 3.4 g/dL [Table/Fig-1].

The majority of cases were operated on between the years 2015 and 2018. The different CBD diameters in choledochal cyst patients (N=128) shown in [Table/Fig-3].

CBD diameter	n (%)
Less than 12 mm	7 (5.46)
12-15 mm	43 (33.59)
16-18 mm	18 (14)
19-20 mm	24 (18.75)
21-40 mm	23 (17.96)
More than 40 mm	13 (10.15)

**[Table/Fig-3]:** Demonstrating the different diameters of CBD for those who underwent surgery.

The USG was the primary investigation of choice, followed by MRCP for the evaluation of the cysts. In the present study institution, a CT scan is mostly accompanied by complementary MRCP. MRCP was performed on all patients as a basic investigation for evaluating the

biliary tree. Fusiform dilatation of the CBD was the most common finding, with or without concomitant CHD and IHBR dilatation. CBD stones were the most common additional feature, observed in 64.06% of the cases. The other MRCP findings in these patients are shown in [Table/Fig-4].

Findings	n (%)
Cholelithiasis	48 (37.5)
CBD stones	82 (64.06)
Hepatolithiasis	11 (8.59)
CBD Stent in-situ	25 (19.53)
Chronic pancreatitis	3 (2.34)
Liver cysts	2 (1.56)
Enterobiliary fistula	6 (4.68)
Remnant GB	1 (0.78)
Cholangitis abscess	3 (2.34)
Carcinoma gallbladder	2 (1.56)
Biliary cystadenoma	1 (0.78)
Anomalous Pancreaticobiliary Junction (APBJ)	2 (1.56)
Haemangioma of liver	2 (1.56)
Pancreatic divisum	1 (0.78)
Gallbladder polyp	1 (0.78)
Portal vein thrombosis	4 (3.13)

**[Table/Fig-4]:** Other MRCP findings of studied patients.  
 CBD: Common bile duct; GB: Gallbladder

All patients underwent standard open choledochal cyst excision with Roux-en-Y HJ. Only one patient had the posterior wall of the cyst left behind due to its dense adhesion to the main portal vein. The CBD diameter and architecture matched with MRCP findings. Few patients, 21 (16.4%), had other intraoperative findings [Table/Fig-5].

Other intraoperative findings	No. of patients
Cholecystoduodenal fistula	3
Choledochoduodenal fistula	1
Cholecystocolic fistula	1
Carcinoma gall bladder	2
Gastrojejunostomy	5
Type I Mirizzi's syndrome	1
Type II Mirizzi's syndrome	1
Cholestatic liver	2
Atrophy of right lobe of liver	1
Jejunal diverticula	1
Empyema gall bladder	1
Abscess in lesser sac	1
Low insertion of cystic duct into intraduodenal part of CBD	1
Total	21

**[Table/Fig-5]:** Other intraoperative findings noted during the surgery (n=21).  
 Additional surgeries done in patients

Some patients, 13 (10.15%), required additional surgeries along with choledochal cyst excision [Table/Fig-6].

**Short- and Long-term Outcomes:** Postoperative complications occurred in 48 patients (37.5%) during the short-term follow-up period (30 days). Postoperative mortality was observed in one patient (0.78%). The types of postoperative complications is depicted in [Table/Fig-7]. All these patients were managed conservatively, except for three who required laparotomy for one case each of strangulated umbilical hernia, acute intestinal obstruction and HJ leak with biliary sepsis. The last patient later succumbed to sepsis. Patients were able to tolerate an oral diet at discharge, although three had an abdominal drain in-situ (for bile leak), which was removed during follow-up.

Other procedures	Number of patients
Enterobiliary fistula take down	5
Radical cholecystectomy	2
Appendectomy	1
Left lateral sectionectomy	1
Frey's procedure	1
Right posterior duct cholangiojejunostomy	1
Umbilical hernia repair	1
Jejunal diverticular excision	1
Total	13

**[Table/Fig-6]:** Other additional procedures performed in patients (n=13).

Immediate postoperative complications	n (%)
Superficial Surgical Site Infections (SSI)	25 (52%)
Bile leak	9 (18.75%)
Postoperative bleeding	4 (8.3%)
Ileus	3 (6.25%)
Pancreatic leak	2 (4.1%)
Urinary retention	1 (2%)
Severe acute respiratory illness	1 (2%)
Subcostal incision dehiscence	1 (2%)
Acute intestinal obstruction	1 (2%)
Strangulated umbilical hernia	1 (2%)

**[Table/Fig-7]:** Immediate postoperative complications (n=48).  
 \*Death (one of bile leak patient)

A total of 83 patients (83/127) (65.35%) were available for long-term follow-up to measure long-term outcomes [Table/Fig-8]. The mean follow-up period was 44.58±27.86 months. One patient died within six months of follow-up due to exacerbation of bronchial asthma. Two patients with carcinoma of the gallbladder succumbed within one year of surgery. Another patient died of jaundice, with details unavailable (potentially due to biliary sepsis) and two patients died of unknown causes.

Type of long-term complication	n (%)
Pain abdomen on and off	9 (10.84%)
Incisional hernia	5 (6.02%)
Itching	3 (3.6%)
Subacute intestinal obstruction	1 (1.2%)
Death	6 (7.22%)
Lost to follow-up	44

**[Table/Fig-8]:** Long-term follow-up (n=83 available for follow-up).

Postoperative hospital stays ranged from four to 20 days, with a mean of eight days. The longer postoperative period was mainly due to complications, with two patients requiring re-laparotomy while the others were managed conservatively.

## DISCUSSION

Choledochal cysts are cystic dilations of the bile duct or ducts that occur congenitally in infants, although they can also present in children or, less commonly, in adults. When similar conditions are observed in adults, there are various potential causes. One of the most notably cited aetiology is the Abnormal Union of the Pancreatobiliary Duct Junction (AUPBD) [11]. A nationwide survey conducted by a Japanese research team, which included 645 patients, found a strong association of 90.2% between choledochal cysts and AUPBD [12]. However, this association is not as prevalent in the present study population, as the authors identified only two patients with this finding on MRCP. Other aetiological factors mentioned in the literature include bile duct obstruction due to sphincter of Oddi dysfunction, inadequate autonomic innervation of



the bile duct leading to hypokinesia, as well as atresia, stenosis, or fibrosis of the terminal CBD [13,14]. In adults, the most common pathology appears to be hypokinesia of the bile ducts leading to stasis and lithiasis.

The most common manifestation of the disease is abdominal pain. Secondary symptoms may include jaundice, fever, sepsis, or even severe sepsis. The presence of a palpable mass in the abdomen, as observed in children, is rare in adults, as noted in the present study. Cholangitis was seen in 46 patients in the series, which is not commonly reported in other studies. USG was the primary investigation of choice, followed by MRCP for the evaluation of cysts. In the present study institution, CT scan is mostly accompanied by complimentary MRCP, which provides a diagnosis in almost 100% of patients, as has been observed in other series as well [15]. Type I choledochal cysts were seen in 67.1% of cases, followed by Type IV A.

The presence of CBD stones was noted in 64% of patients, suggesting a possible acquired cause (hypokinesia) of choledochal cysts. This also accounts for the concomitant dilatation of the central intrahepatic bile ducts due to pressure. The prevalence of biliary calculi varies, ranging from 9.0% in children to 24.1% in adults, with gallstones found in 12.7% of cases, CBD stones in 65.8% and hepatolithiasis in 21.5% of cases [12]. The presence of such a significant number of stones also explains the incidence of cholangitis in these patients. One of the present study patients had a previous history of pancreatitis. However, perforation of the bile duct, as seen in children, was not observed in any of the patients.

Hepatobiliary malignancy is a well known complication of long-term choledochal cysts. In present series, the authors had two patients diagnosed with carcinoma of the gallbladder. Hepatobiliary malignancy has been reported to occur in 2.5% to 28% of patients with choledochal cysts [16,17]. The risk is age-related, with an incidence quoted at 14.5% in patients older than 20 years [18]. Chronic inflammation induced by bile stagnation, along with the possible development of carcinogens and the sparse distribution of protective mucin-secreting glands in the bile duct, may contribute to carcinogenesis [19]. Genetic analysis of choledochal cysts has revealed Tumour Protein53 (TP53), RNA Binding Motif Protein 10 (RBM10), Kirsten Rat Sarcoma viral oncogene homolog (KRAS) mutations and Fibroblast Growth Factor Receptor 2 (FGFR2) rearrangements, which have been shown to trigger cancer development to some extent and warrant further studies [20].

The size of the CBD on imaging helps in diagnosing the disease, particularly if it is fusiform; however, the specific diameter at which it should be considered a choledochal cyst remains unclear, as the literature does not provide guidance on this matter. The excision of the cyst and Roux-en-Y HJ is the treatment of choice. This provides adequate clearance of the disease and optimal drainage. At the institution, the authors routinely extend the HJ to the left duct for a wider anastomosis. In some cases, intraoperative endoscopy was utilised to ensure complete clearance of ductal calculi.

In the present study, authors observed that the size of the duct did not increase the incidence of complications. Previous cholangitis and ERCP interventions did not increase the complexity of surgery or lead to increased postoperative complications.

Complications following choledochal cyst excision and HJ are common. The literature cites an incidence of complications ranging from 14% to as high as 64% [8]. Srinivasan PH et al., reported an early bile leak rate of 11% and a late anastomotic stricture rate of 2.7% [21]. In the present study, the bile leak rate was 7% and there were no long-term strictures, which is comparable. The long-term rate of anastomotic stricture reported by Singham J et al., and Lipsett PA et al., was 12% and 2.3%, respectively [22,23]. They emphasised factors like tension-free, adequately sized anastomosis to prevent the formation of anastomotic strictures in the long-term. A study conducted by Kumar S et al., identified one case of anastomotic

stricture in the long-term [24]. Machado NO et al., followed-up with 10 cases for six years and did not find any instances of anastomotic stricture; however, two patients experienced recurrent cholangitis, which was managed conservatively [25]. Cho MJ et al., reported a 14-year surgical experience with adult Choledochal Cyst Disease (CCD), focusing on clinical outcomes after surgical treatment. They noted that out of 204 patients, extrahepatic cyst excision with HJ was performed in 185 patients (90.7%) [26]. Major perioperative complications occurred in 5 patients (2.5%), resulting in no mortality. Late complications were observed in 48 patients (23.6%) and concurrent cancer was diagnosed in 20 patients (9.8%) [26]. The present study had two cases of gallbladder cancer.

An ectatic bile duct in adults is a challenging condition for any Hepatopancreatobiliary (HPB) surgeon, as it always requires additional evaluation to rule out malignancy. Once malignancy is ruled out, a decision must be made regarding whether to drain the bile duct or excise it and perform HJ. The institutional protocol is to excise the bile duct when it is more than double the size expected for that age with fusiform dilatation and to perform wide HJ. This approach provides long-term symptom relief for patients. Long-term complications, such as stricture, biliary cirrhosis and malignancies in the remnant duct, are known to occur, which necessitates regular follow-up for these patients.

### Limitation(s)

The study has a few limitations. The retrospective data from older patients regarding surgical details and short-term outcomes may introduce bias. There are no standard criteria for diagnosing the disease, which could lead to overdiagnosis or underdiagnosis in some patients. Additionally, almost one-third of the patients are not available for long-term follow-up.

### CONCLUSION(S)

Adult choledochal cysts remain a concerning issue in tertiary care referral centre. Timely diagnosis and intervention provide long-term symptom relief and help prevent complications.

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**PARTICULARS OF CONTRIBUTORS:**

1. Associate Professor, Department of Surgical Gastroenterology, BMCRI, Bengaluru, Karnataka, India.
2. Assistant Professor, Department of Surgical Gastroenterology, BMCRI, Bengaluru, Karnataka, India.
3. Professor, Department of Surgical Gastroenterology, BMCRI, Bengaluru, Karnataka, India.
4. Professor, Department of Surgical Gastroenterology, BMCRI, Bengaluru, Karnataka, India.

**NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:**

GK Adithya,  
543, Sobha Dew Flower Apartment Sarakki Main Road,  
Bengaluru-560078, Karnataka, India.  
E-mail: adi3anny@gmail.com

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