

Case Series

A case series on laparoscopic management of choledochal cyst in adults at a tertiary care centre

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ABSTRACT

Choledochal cysts represent a rare congenital anomaly characterized by the cystic dilation of the biliary tract. Although a vast majority of the cases present in early childhood, sometimes these cases can present in adulthood as well. The preferred treatment involves laparoscopic complete excision of the choledochal cyst with subsequent reconstruction of the biliary tract. While laparoscopic hepaticoduodenostomy is a documented procedure in the literature, laparoscopic Roux-en-Y hepaticojejunostomy has gained widespread acceptance. This study details a series of fifteen cases illustrating our experiences with these procedures. Between August 2022 and February 2024, a total of eighteen patients, including fifteen females and three males, were diagnosed with choledochal cysts. Fifteen of the patients underwent laparoscopic cyst excision with Roux-en-Y hepaticojejunostomy. Evaluation parameters included age, sex, clinical symptoms, Todani classification, operative time, complications, and hospital stay. Assessment utilized clinical history, ultrasonography, and magnetic resonance cholangiopancreatography. Out of the total eighteen cases, fifteen patients were operated, two patient developed right-sided pleural effusion which was managed with chest physiotherapy and incentive spirometry. Anastomotic leak was observed in the case of two patients which was managed conservatively. Of these patients, three developed fevers, which were managed using intravenous antipyretic drugs. In a further 3 months follow-up, no new complications were observed in any of the cases. Laparoscopic management of choledochal cysts is increasingly favoured, providing patients with the benefits of minimally invasive surgery. However, such procedures must be performed by highly experienced laparoscopic surgeons.

Keywords: Choledochal cyst, Biliary malformations, Hepaticojejunostomy

INTRODUCTION

Choledochal cysts are uncommon congenital cystic dilation affecting the biliary tract. A majority of cases are observed in the pediatric age group, usually less than 10 years, it can also manifest in adults.¹ Because of recent advancements in understanding epithelial markers and recognizing varied pathophysiology among different etiologic subtypes, the term 'biliary malformations' is now used instead of cysts.²

These are uncommon, with an incidence ranging from 1:100,000 to 1:150,000 in Western populations, but are

more prevalent in Eastern populations. Choledochal cysts predominantly impact females, occurring three to eight times more frequently than in males. While often identified in infancy or childhood, up to half of the patients receive a diagnosis in adulthood.^{3,4}

Choledochal cysts are classified based on the Todani modification of the Alonso-Lej classification. Type I, characterized by fusiform or cystic dilations of the extrahepatic biliary tree, is the most prevalent, constituting more than half of all choledochal cysts. Type II represents a saccular diverticulum of an extrahepatic bile duct and is rare, accounting for less than 5% of cases.

Type III, involving bile duct dilatation within the duodenal wall (choledochoce), comprises approximately 5% of choledochal cysts. Types IVa and IVb, characterized by multiple cysts, collectively make up 5% to 10% of choledochal cysts. Type IVa affects both extrahepatic and intrahepatic bile ducts, while type IVb cysts impact the extrahepatic bile ducts exclusively. Lastly, Type V, featuring intrahepatic biliary cysts, is exceedingly rare, constituting only 1% of choledochal cyst cases.²⁻⁵

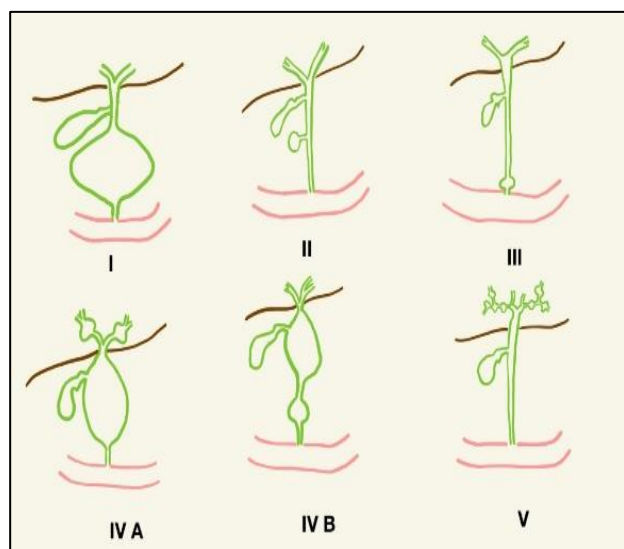


Figure 1: Classification of choledochal cysts.

As described by Todani et al the choledochal cysts can be classified into type I: fusiform or cystic dilations of the extrahepatic biliary tree. Type II: saccular diverticulum of an extrahepatic bile duct. Type III: involving bile duct dilatation within the duodenal wall (choledochoce). Type IVa: affects both extrahepatic and intrahepatic bile ducts, while type IVb cysts impact the extrahepatic bile ducts exclusively. Lastly, type V, features intrahepatic biliary cysts. Green colour indicates the biliary tract, brown represents the liver while pink represents the drainage of the biliary system into the second part of the duodenum.

Symptoms vary between paediatric and adult age groups, with right upper quadrant pain being the most common, occasionally accompanied by jaundice and an abdominal mass. Despite its benign nature, choledochal cysts are linked to severe complications, including malignant transformation, cholangitis, pancreatitis, cholelithiasis, and cyst perforation. Due to the risk of malignant transformation, early surgical intervention is often recommended.^{6,7}

The sole method for confirming the diagnosis is diagnostic imaging. The first investigation that patients usually undergo is ultrasonography. Due to the widespread use of CT scans, the suspicion of a choledochal cyst often arises through this method, but its classification typically requires additional assessment using MRCP or ERCP. In cases where the distal bile duct proves challenging to assess via MRCP, ERCP becomes a

more valuable tool for defining the distal biliary tree and pancreaticobiliary junction. ERCP has disadvantages like invasiveness, radiation to exposure, and difficulty in performing the procedure in cases with inflammation or scarring. Because of these reasons, MRCP is considered to be the gold standard for diagnosing choledochal cysts.^{2,8}

The surgical approach to managing choledochal cysts involves the complete removal of the cyst followed by appropriate surgical reconstruction. For type I cysts, the treatment involves complete cyst excision, cholecystectomy, and Roux-en-Y hepaticojejunostomy. The resection should extend to the nondilated biliary tree, and anastomosis may be necessary with the left and right hepatic ducts. Type II cysts should undergo complete excision. Type III cysts, being rare, may be approached transduodenally, and if they don't involve APBJ, endoscopic drainage might be sufficient. In cases of duodenal or biliary obstruction, transduodenal excision or sphincteroplasty can be considered. The surgical approach for type IV cysts needs careful customization based on the affected anatomy. Those affecting only the extrahepatic bile ducts are managed similarly to type I cysts, involving excision and hepaticojejunostomy. Type IV cysts with intrahepatic extension in one lobe may require partial hepatectomy and reconstruction. Surgical options for Caroli disease vary from resection for unilobed cases to liver transplantation for diffuse disease.^{2,4}

CASE SERIES

This observational study including 18 patients admitted during the period from August 2022-February 2024 in the department of general surgery of a tertiary centre, was conducted after taking approval from the ethical committee of the institute.

A total of eighteen patients, of which three were men and fifteen women, with a mean age of 21.5 years were observed. All of the patients presented with complaints of Pain in the upper abdomen, of which six were associated with jaundice and pruritus, and five with complaints of a lump in the abdomen. Nausea and vomiting were noted in seven of the abovementioned patients, while 9 patients had associated anorexia. After noting the demographic details of the patients, the patients underwent a thorough history taking and physical examination to ascertain a probable diagnosis. Following this, blood samples were drawn to assess the serum levels of bilirubin and hepatic enzymes. Ultrasonography was used as the first radiological investigation which revealed dilated common bile Duct. Magnetic resonance cholangiopancreatography (MRCP) was conducted to confirm the diagnoses. Classification of the choledochal cysts was done based on the Todani modification of Alonso-Lej classification.

Three patients who were categorized as type 3 and type 4 A choledochal cyst were referred to respective departments for further management. Fifteen of the patients were operated on by using the minimal access technique. Aspects like the duration of the surgery, post-operative pain using the visual analogue scale, complications, first oral feed after surgery, and day of discharge were also noted.

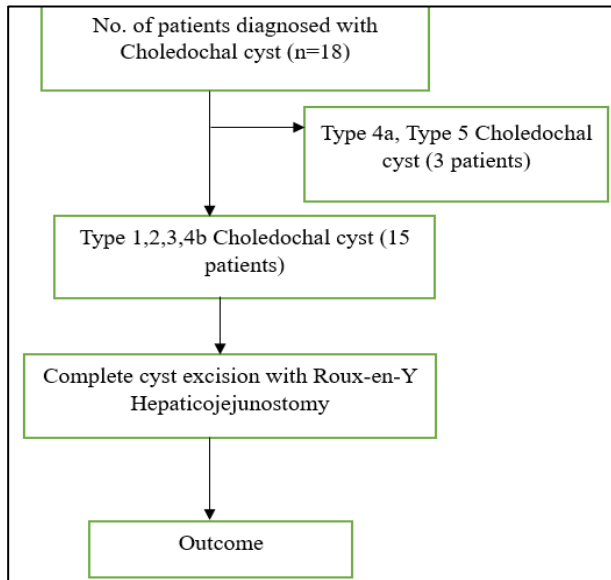


Figure 2: Flowchart depicting patient selection and measuring the outcome.

Pre-operative workup

Out of the various patients admitted through the outpatient department, eighteen patients underwent treatment for choledochal cysts in our institute. Fifteen of these patients were women and three were men. The age and sex proportions are also represented in Table 1 with mean age of presentation 21.5 years. Serological profiles of the patients revealed raised bilirubin in nine of the patients as indicated in Figure 3.

Table 1: Sex and age distribution (n=18).

Variables	N	Percentage (%)
Gender		
Male	3	20
Female	15	80
Age group (in years)		
16-20	6	33.3
21-25	8	44.4
26-30	3	16.7
31-35	1	5.5
>35	0	0

The Table 1 describes the gender and age distribution observed among the cases in the study, 80% of the cases observed were women and the rest were men. 33.3% of the cases belonged to the age group 16-20 years, 44.4%

of the cases were from the 21-25 years age group, 16.7% of the cases from 26-30 years of age and the remaining cases were from the 31-35 years age group. The overall data shows a female propensity in the prevalence of Choledochal cysts, presenting at a younger age.

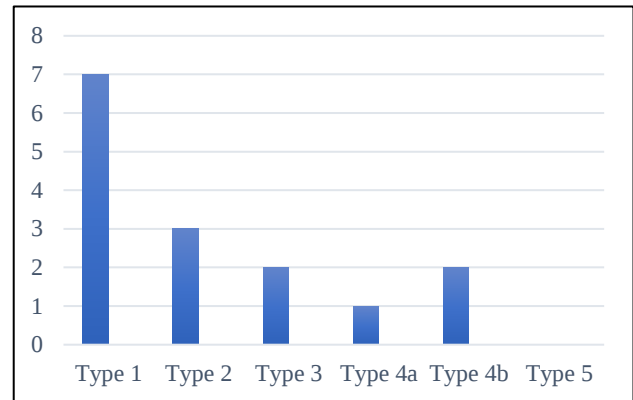


Figure 3: Distribution of cases among various types of choledochal cyst.

Seven of the patients were classified as type 1 (33.8%), three as type 2 (16.6%), two as type 3 (11.1%), one as type 4a (5.5%) and remaining two as type 4 B (11.1%) choledochal cyst. No type 5 choledochal cysts were recorded. These findings are also indicated in Figure 4. Patients diagnosed with type 3 and 4a cysts were referred to respective departments for further management. Fifteen patients underwent laparoscopic excision of the cyst, cholecystectomy with Roux-en-Y hepaticojejunostomy.

Seven out of a total of eighteen cases were found to be type 1 choledochal cyst, three of the cases were type 2 choledochal cyst, two cases were type 3 choledochal cyst, a single case of type 4a was observed, two of the cases were of type 4b choledochal cysts. None of the observed cases belonged to type 5 choledochal cysts.

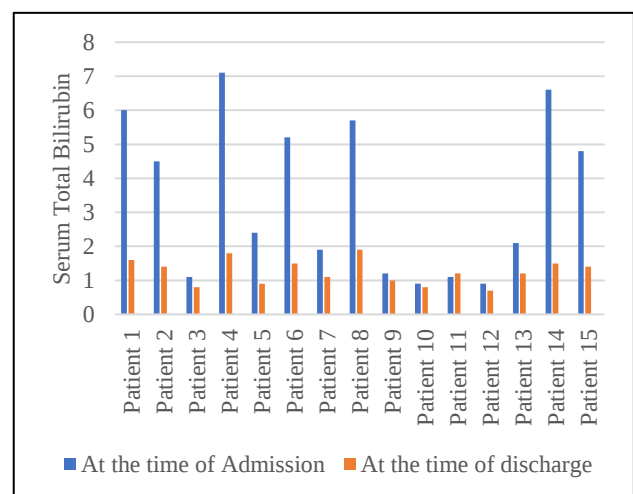


Figure 4: Serum total bilirubin levels (mg/dl) at the time of admission and discharge.

The chart depicts the comparative change in the levels of serum total bilirubin at the time of admission to the levels at the time of discharge in various patients. The blue column indicates the levels at time of admission, while orange columns indicate the levels at time of discharge.

Operative technique

The procedure and its risks are explained to the patient and written informed consent is taken. The patients are placed in the supine position, supraumbilical port is created using Hassan's open port technique. pneumoperitoneum is created and the rest of the ports are placed in the standard port position for laparoscopic cholecystectomy. A Nathanson's retractor is placed through an additional port to lift the liver cranially to ease the dissection. The energy source harmonic scalpel and monopolar electrocautery were used. To achieve adequate exposure of the working area cranial retraction of the gallbladder and caudal traction of the transverse colon and duodenum is done (Figure 5). Dissection was then extended cranially up to the level of the normal-sized hepatic ducts, and the cyst was excised in toto. This usually required clipping of the cystic duct junction. The distal bile duct was divided at the level where it was of normal calibre, and it was sutured with 3-0 Vicryl sutures (Figure 6 and 7). The duodenojejunal flexure was located, and the jejunum, specifically at the level of the first well-vascularized arcade, was separated using a linear cutter. A segment of at least 40 cm from the long limb was utilized for the anastomosis with the proximal hepatic duct. The jejunojejunostomy was established at the 30-cm level through an intracorporeal side-to-side anastomosis employing endostaplers. The creation of the Roux loop was entirely intracorporeal, and it was brought up for anastomosis in a retrocolic manner, ensuring that the long limb was closely aligned with the lateral abdominal wall. Hepaticojejunostomy was conducted in an end-to-side fashion, utilizing intracorporeal suturing with polyglactin 3-0 sutures. A subhepatic drain was placed near hepatojejunal anastomosis before finishing procedure.

The mean duration of surgery 135 min using laparoscopic approach including complete intracorporeal suturing.

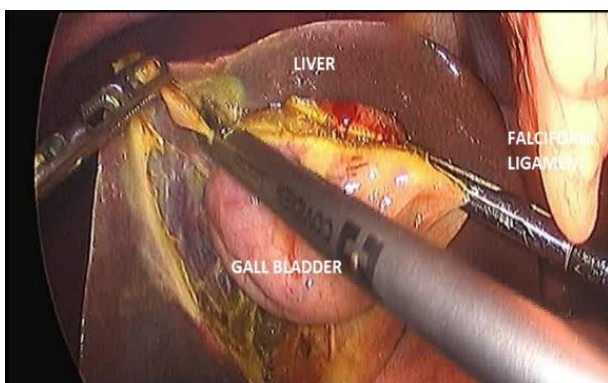


Figure 5: Intraoperative image of separating the gall bladder from the liver bed.

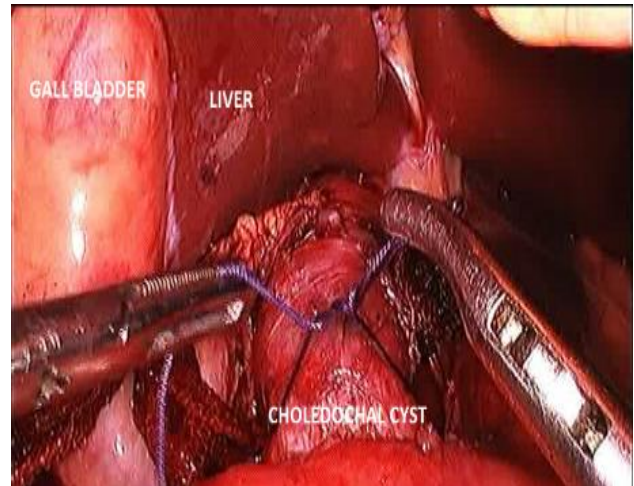


Figure 6: Intraoperative image depicting ligation of choledochal cyst.

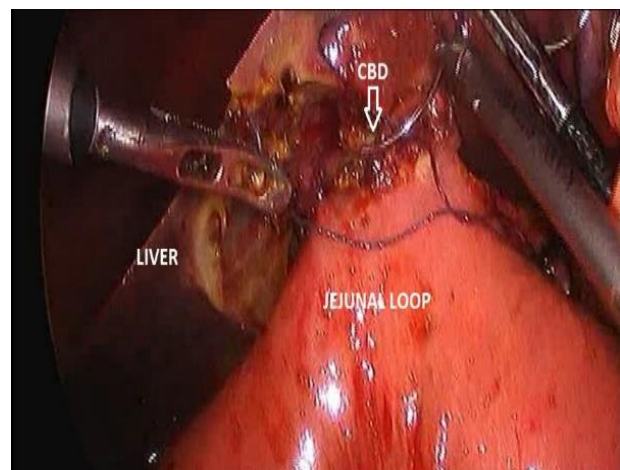


Figure 7: Intraoperative image depicting creation of hepaticojejunostomy.

Postoperative findings

Postoperatively, ambulation was started within 8 hours post-surgery in all of the cases, oral feed was started in patients on the mean postoperative day 4. A visual analog scale was used to assess the level of pain experienced by the patients, mean score had risen from 5 in the preoperative period to 8 in the immediate postoperative period, followed by a drop to a mean VAS score of 2 at the time of discharge. Eleven patients recovered without any complications. Two of the patients developed coughs on day 2 of the postoperative period, which revealed right-sided pleural effusion on the chest radiograph in both cases. They were managed with chest physiotherapy and incentive spirometry. An anastomotic leak was observed in two cases, which were managed conservatively using intravenous fluids and antibiotics. Of these four patients, three developed a fever which was managed using intravenous antipyretic i.e. paracetamol (Table 2). The mean total serum bilirubin level dropped from 3.4 (in the preoperative period) to 1.25 (at the time

of discharge). All the patients were discharged after drain removal on the mean of the 8th postoperative day. Patients returned to routine functioning by day 2 of surgery, with early return to work. No mortality was observed in our study. Follow-up for a period of three months revealed no fresh complaints.

Table 2: Post-operative complications.

Complication	N	Percentage (%)
Anastomotic leak	2	13.3
Pleural effusion	2	13.3
Fever	3	20
Surgical site infection	0	0
Cholangitis	0	0
Sepsis	0	0

The Table 2 illustrates the complications observed in the post-operative period in the patients that underwent Roux-en-Y hepaticojejunostomy in our study. Out of the fifteen patients, anastomotic leak was observed in two patients. Both these patients were managed conservatively. Two of the patients were found to have developed Pleural Effusion. Three out of these four patients developed fever which was managed using antipyretics. All of the patients in the study recovered and no further intervention was required. No mortality was observed in the study.

DISCUSSION

Choledochal cyst is a rare entity primarily seen in the paediatric age group. Although uncommon, it can also present in early adulthood.⁹ Our study observed the female-to-male ratio to be 4:1, which was similar to the study done by Wiseman et al.¹⁰ The median age of presentation in our study was 21.5 years. The study done by Machado et al had a median age of presentation of 31 years in females and 36 years in males.¹¹ The most common type in our study was found to be type 1 choledochal cyst (38.8%), whereas the study conducted by Özsoy et al found type 1 choledochal cyst in 67.7% of the cases.¹² The median total bilirubin level dropped from 3.4 in the preoperative state to 1.25 at the time of discharge. The short-term complications of our patients are generally good, with low early complication rate similar to other studies. The study done by Wong-Hoi She et al had an early complication rate of 13.3%.¹³ Anastomotic leak rate is found to be between 0 to 20%, in our study the leak rate was 13.3%, while in the study done by Palanivelu et al the leak rate was 0%.¹⁴ The study conducted by Lipsett et al observed cholangitis in the postoperative period, no such complication was observed in our study.¹⁵ Overall, a minimally invasive approach provides the patient with better cosmesis, little post-operative pain, and early resumption of work. In expert hands, laparoscopy is an excellent tool for the management of choledochal cysts, although it is associated with a learning curve that requires to be mastered. In our institute, because of the huge experience

with minimal access surgery, laparoscopic management of choledochal cysts is preferred over the open technique.

CONCLUSION

Earlier, choledochal cysts were treated using the open technique. Laparoscopic Roux-en-Y hepaticojejunostomy has become a very suitable alternative to the open technique, providing the patient with the benefits of minimal access surgery. Like all laparoscopic procedures, the long learning curve has to be completed for favourable outcomes in these procedures. Faster recovery, better cosmesis, and less postoperative pain have made the laparoscopic approach preferable when in expert hands.

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