

# LILLY'S TECHNIQUE AS A PRIMARY PROCEDURE FOR COMPLICATED TYPE 1 CHOLEDOCHAL CYST

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## ABSTRACT

**Objective:** Objective: This study aimed to evaluate the efficacy, safety, and long-term outcomes of Lilly's technique in treating complicated Type 1 choledochal cysts.

**Materials and Methods:** A retrospective chart review was conducted from January 2014 to January 2024 on patients preoperatively diagnosed with complicated Type 1 choledochal cysts who underwent Lilly's technique as their primary surgical intervention. The study included 40 patients aged 2 to 12 years, treated over a 10-year period, with follow-up periods ranging from 3 to 8.5 years. Data on perioperative outcomes, postoperative complications, and long-term biliary function were collected and analyzed.

**Result:** Eight patients (20%) experienced early postoperative complications, and 3 cases (7.5%) had late complications. There were no instances of operative mortality or malignant transformation. Overall, 20% of patients had early post-operative complications, while 7.5% experienced late complications.

**Conclusion:** Lilly's approach remains the preferred procedure for complicated type 1 choledochal cysts. Additionally, this technique proves effective for type 1 choledochal cysts with dense adhesions caused by recurrent cholangitis, and it remains a safe method with favorable long-term results.

**Key Words:** Choledochal cyst, Lilly's procedure, Roux-en-Y hepaticojejunostomy.

**This article may be cited as:** Arshad M, Imran M, Ishfaq M, Jamil A, Khuroosh U, Khan R. Lilly's Technique As A Primary Procedure For Complicated Type 1 Choledochal Cyst. *J Med Sci* 2025 July-September;33(3):164-167

## INTRODUCTION

Choledochal cysts (CCs) are rare, inherited abnormalities characterized by cystic widening of the bile ducts, spanning from the intrahepatic ducts to the ampulla of Vater, without mechanical obstruction. <sup>1</sup> In 1959, Alonso-Lej et al. first divided choledochal cysts into three types based on the location of biliary duct dilation. This classification was changed in 1977 by Todani et al., who added two additional types, resulting in the five-category system commonly used by clinicians today. <sup>2</sup>

The occurrence of bile duct cysts ranges from 1 in 13,000 to 1 in 2 million births. Over 60% of these cases develop within the first year of life. Some cases emerge in adulthood, presenting with symptoms that differ from those typically seen in children. Females are affected more often than males, with a ratio of 4:1. The two most common clinical complaints are recurrent cholangitis and abdominal pain, followed by acute pancreatitis, a palpable mass in the right hypochondrium, jaundice, and broncho-

biliary fistula. <sup>3-5</sup>

A long common channel can permit pancreatic secretions to reflux into the CBD, activate proteolytic enzymes, and weaken and damage the duct wall. Another idea suggests that congenital weakening of the CBD is caused by defective bile duct epithelialization and recanalization during development. The abnormal function and spasm of the sphincter of Oddi have been recently associated. Long common channels can cause the sphincter of Oddi to behave abnormally, leading to high pancreatic duct pressure (30–50 cm) compared to CBD pressure (25–30 cm), which can cause reflux and choledochal cysts. <sup>6</sup>

Additionally, the presentation is often vague and broad. For patients without adhesions, complete cyst resection is ideal. However, for those with extensive adhesions, delayed hemorrhage, or malignancy risk, the Lilly technique with Roux-en-Y hepaticojejunostomy is a suitable alternative, enabling bile duct bypass and reducing complications while maintaining effective management. <sup>7</sup>

This article presents our expertise in managing complex type 1 choledochal cysts using Lilly's technique. The goal is to assess the safety, effectiveness, and complications of Lilly's technique in treating complicated type 1 choledochal cysts and to establish appropriate treatment recommendations for patients with choledochal cysts.

## MATERIALS AND METHODS

This retrospective chart review was conducted in

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**Date Received:** 28/03/2025

**Date Revised:** 19/08/2025

**Date Accepted:** 29/09/2025

the pediatric surgical unit at Khyber Teaching Hospital, Peshawar. Included patients were diagnosed with choledochal cyst and had intraoperative dense adhesions. Patients with abnormal hepatobiliary anatomy or without intraoperative dense adhesions were excluded. A convenience sampling was used, resulting in a sample size of 40. Ethical approval was obtained from the ethics committee of the Department of Medical Education at Khyber Medical College, Peshawar. Data on clinical symptoms, laboratory tests, previous treatments, associated conditions, operative details, and the preoperative and postoperative courses were recorded using a standardized form. Histopathological confirmation of the disease (chronic inflammatory changes) was obtained in all cases. Follow-up information was collected from outpatient records and through correspondence.

Qualitative data, such as gender, symptom types, and early and late complications, were reported as frequencies and percentages. Quantitative data were presented as mean ± SD. Patients were stratified by age and gender, and a chi-square test was used for comparison at a 5% level of significance. SPSS v23 was employed for statistical analysis. Microsoft Office 2013 was used to prepare the article.

**RESULTS**

There were 40 patients in this cohort, including 26 females (65%) and 14 males (35%). The average age of the patients was 4.5 years, with a standard deviation of 1.4 years. The distribution of presenting symptoms is shown in Table 1. The most common symptom was jaundice. Abdominal pain and palpable mass were also observed. Fifteen (37.5%) patients had abnormal liver function tests at admission, which were corrected before proceeding with the procedure. Abdominal ultrasonography diagnosed 34 (85%) patients. Computed tomography (CT) scans, along with magnetic resonance imaging (MRI), were performed in all patients to confirm the diagnoses. Two patients underwent endoscopic retrograde cholangiopancreatography to relieve obstructive jaundice with stenting. A liver biopsy was performed in 18 (45%) of the patients.

The frequency of different early postoperative complications is shown in Figure 1. Of the eight early postoperative complications, one patient experienced a burst abdomen, which was managed with tension suturing. Five patients developed surgical site infections, treated medically with culture-sensitive antibiotics and wound washes, followed by delayed closure. Two patients had biliary leaks; one was managed in our unit by keeping the sub-hepatic drain for two weeks. The other patient was referred to a specialized hepato-biliary center after conservative management failed.

Three patients (7.5%) experienced late complications (table 2): two with abnormal liver functions and one with recurrent abdominal pain; all were managed medically. Seven patients were lost to follow-up after 1 to 1.5 years of consultations. No mortality was observed during the follow-up period.

There was no significant difference in early complications between males and females ( $X^2 = 0.154$ ,  $df = 1$ ,  $p > 0.05$ ). Likewise, no difference was observed in the late complication rate between genders ( $X^2 = 0.004$ ,  $df = 1$ ,  $p > 0.05$ ). Patients were divided into two age groups: 45% were under 4 years old and 55% were over 4 years old. No statistical difference was found in the early and late complication rates (late complications;  $x^2=2.65$ ,  $df=1$ ,  $p>0.05$ ; early complications;  $x^2=0.925$ ,  $df=1$ ,  $p>0.05$ ).

**DISCUSSION**

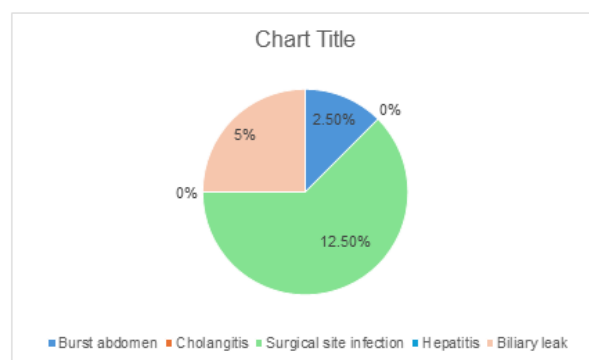
Lilly's technique is the standard surgical approach for treating type 1 choledochal cysts. The method focuses on completely removing the cyst, which is essential to reduce the risk of malignancy from leftover cystic tissue. After excision, a Roux-en-Y hepaticojejunostomy is performed to restore bile flow. This approach is preferred because it can decrease the chances of postoperative complications, such as biliary stricture and cholangitis, which

**Table No 1: Presentation of choledochal cyst**

Mode of Presentation	Number (%age)
Jaundice	24 (60%)
Dark Urine	16 (40%)
Acholic feces	9 (22%)
Right Hypochondrium Mass	18 (45%)
Cholangitis	(100%)
Charcot's Triad	24 (60%)
Antenatal ultrasonography	8 (20%)
Portal hypertension	4 (10%)
Pancreatitis	35 (40%)

**Table No 2: Late post-operative complications (3 months to 3 years)**

Late Complications	Frequency (%age)
Liver dysfunction	5%
Repeated pain abdomen	2.5%
Intra-hepatic duct stones	0%
Chronic Pancreatitis	Nil
Portal hypertension	Nil
small bowel obstruction	Nil



**Fig 1: Early Post-op Complications (<3 months)**

are more frequent in non-excisional methods.<sup>8,9</sup>

Numerous international studies have confirmed the safety and effectiveness of Lilly's technique for managing type 1 choledochal cysts. The complication rates in our study are similar to those reported in the literature. A thorough review by Todani et al. (2020), which included over 500 patients, showed a low complication rate, with fewer than 5% experiencing postoperative morbidity. The review highlighted the importance of complete cyst removal, showing that patients who underwent this procedure had a significantly lower occurrence of recurrent cholangitis and stricture formation compared to those with incomplete excision.<sup>10,12</sup> In a similar study, a multicenter observational study in Japan conducted by Tashiro et al. (2018), involving 300 patients, found that Lilly's approach delivered excellent long-term results, with 95% of patients remaining symptom-free over a 10- to 12-month period. This finding emphasizes the method's success in preventing cancer, as no cases of bile duct cancer have been observed in the group, similar to our cohort.<sup>11</sup> Likewise, our results align with those of Kim et al. (2017), who reported positive outcomes with few complications, especially among pediatric patients.<sup>13-15</sup>

At the same time, Lilly's method is powerful, but it isn't always without risks. The most common complications associated with this technique include bile leakage, anastomotic stricture, and contamination. Bile leakage is a significant concern, occurring in about 3-7% of cases, as reported in a meta-analysis by Khan et al. (2019). If not promptly addressed, this can lead to biliary peritonitis. To lower the risk of bile leakage, intraoperative cholangiography is recommended to confirm the complete removal of the cyst and to verify the patency of the biliary-enteric anastomosis.<sup>16-18</sup>

Anastomotic stricture is another complication, with an occurrence rate reported to be around 2-5% in the literature. Stricture formation can lead to recurrent cholangitis and may require further intervention. The risk of stricture can be reduced by ensuring a tension-free anastomosis and using fine, absorbable sutures for hepaticojejunostomy. Postoperative monitoring with imaging is crucial for the early detection and management of anastomotic strictures.<sup>19</sup> We did not encounter any strictures; however, this may be due to the shorter follow-up period in our study population.

Cholangitis, though much less common, is a serious problem. Using prophylactic antibiotics and careful surgical strategies is crucial in reducing the risk of postoperative infections. A study by Li et al. (2020) emphasized the importance of perioperative antibiotic prophylaxis, showing that it can decrease the incidence of postoperative cholangitis by up to 50%.<sup>20</sup>

The long-term outcomes of Lilly's approach to managing type 1 choledochal cysts are generally positive, with several studies highlighting its effectiveness in providing lasting symptom relief and reducing long-term headaches. Additionally, this study observed a significant decrease in the risk of developing cholangiocarcinoma,

a serious concern linked to untreated choledochal cysts. This decline in malignancy risk further emphasizes the technique's success in addressing the underlying issues related to type 1 choledochal cysts.<sup>21</sup>

In our study, 5% of patients developed persistent liver dysfunction after surgery, which is similar to a longitudinal study by Ueda et al., who followed patients for 12-20 months. They found that over ninety percent of the patients maintained normal liver features and showed no signs of biliary obstruction. These long-term results support Lilly's approach as a durable and reliable option for treating type 1 choledochal cysts.<sup>17,18</sup>

## CONCLUSION

Lilly's approach remains the procedure of choice for complicated type 1 choledochal cyst, and our study findings are consistent with the favorable outcomes published in different studies.

Furthermore, this technique is effective for type 1 choledochal cysts with dense adhesions caused by recurrent cholangitis, and it remains a safe procedure with favorable long-term results.

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**CONFLICT OF INTEREST:** Authors declare no conflict of interest

**GRANT SUPPORT AND FINANCIAL DISCLOSURE:** NIL

**Authors Contribution:**

Following authors have made substantial contributions to the manuscript as under

Authors	Conceived & designed the analysis	Collected the data	Contributed data or analysis tools	Performed the analysis	Wrote the paper	Other contribution
Arshad M	✓	✗	✓	✗	✗	✓
Imran M	✓	✓	✗	✓	✓	✗
Ishfaq M	✓	✗	✓	✗	✗	✓
Jamil A	✓	✓	✗	✗	✓	✗
Khuroosh U	✓	✗	✓	✗	✗	✓
Khan R	✓	✓	✗	✓	✓	✗

Authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

**Ethical Approval:**

**This Manuscript was approved by the Ethical Review Board of Khyber Teaching Hospital, Peshawar. Vide No. 232/DME/KMC.**

**Dated: 27 03 2024**



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