

ORIGINAL ARTICLE

Re-operative Surgery for Choledochal CystMd. Mohsen Chowdhury¹, Zulfikur Rhaman Khan², Ruksana Karim³**Abstract:**

Excision of a choledochal cyst is possible and desirable even after a previous drainage procedure. In severely ill patients with complication of choledochal cyst, external drainage may be a preferable initial maneuver. External drainage of choledochal cyst is a safe option and secondary excision can be done later. The presence of external catheter ensures the patient to return for definitive surgery. This is a retrospective study with seventy patients and fifteen had a previous cyst drainage operation. Eight patients had undergone external drainage of the choledochal cyst, five had choledochal-jejunostomy and two had received a choledochal-cysto-duodenal anastomosis. Later, re-operative surgery was done for all these patients.

Introduction:

Choledochal cysts are uncommon and of unknown cause with an incidence of 0.1% even among adults referred for ERCP investigation¹. There is higher incidence in female^{2,3,4} and they are more common in the far east than in population of the western European origin⁵. The presentation is often vague and non-specific, impending prompt diagnosis. However, the diagnosis is facilitated by modern imaging technique and may be made at any time from the antenatal period to adult life. The more severe complications of choledochal cyst disease include pancreatitis, cholangitis, biliary cirrhosis and cholangiocarcinoma^{5,6}.

Choledochal cysts are best managed by total excision, although in the past internal drainage into the jejunum or duodenum were recommended¹⁻⁵. Internal drainage is often associated with anastomotic narrowing, because of various factors, including lack of a proper mucosal lining in the cyst and recurrent infection and inflammation due to reflux of pancreatic enzymes⁶⁻⁸. Reoperation may be necessary to excise this choledochal cyst. Revisional surgery is complicated by adhesions and by the sequelae of inadequate biliary drainage, some of which, such as malignancy, may even be fatal. This article gives an experience with reoperative surgery for the choledochal cysts following previous cyst drainage operations. The presentation, problems and difficulties encountered during surgery were studied.

Materials and method:

Seventy patients had surgery for choledochal cysts at Bangabandhu Sheikh Mujib Medical

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Figure-1: Tube cholangiogram showing a choledochal cyst [with external drainage]

University in the seven years from 2001 to June 2007. Fifteen (21.43%) had a previous cyst drainage operation. The records of these patients were reviewed. Eleven women and four men of mean age 36 (range 16-52) years had their initial operation six months to 11 years (mean 29 years) before presentation. Eight patients had undergone external drainage of the choledochal cyst, five had choledocho-cysto-jejunostomy and two had a choledocho-cysto-duodenal anastomosis.

Ultrasonography was done in all patients and barium meal examination was done in those who previously had cyst drainage into the duodenum. Cholangiography through the drainage catheter was done in patients who had external drainage (Figure-1) and the remaining patients underwent endoscopic retrograde cholangiopancreatography (ERCP).

The cysts were grouped according to Todani et al's modification of Alonso Lej's classification¹: type I, solitary extrahepatic cyst; type II, extrahepatic supraduodenal diverticulum; type III, choledochocoele; type IVa, extrahepatic and intrahepatic cysts; type

IVb, multiple extrahepatic cysts; and type V, Caroli's disease.

Regular follow-up was done after surgery to assess post-operative morbidity, regression in size of intrahepatic cysts, recurrence of cholangitis and other features of biliary obstruction.

Results:

External drainage of the choledochal cyst was done in eight patients, of whom three had extensive pericystic adhesions making the excision difficult; in two patients, general condition was not good, and in three the cyst was mistaken for duct dilatation secondary to choledocholithiasis and T-tube drainage was done.

Seven patients had previous cystoenteric drainage operations; two were asymptomatic. Presenting complaints in the five symptomatic patients were abdominal pain, jaundice and recurrent attacks of cholangitis before admission. Three patients had raised serum amylase levels after episodes of acute epigastric pain. Surgical excision of the cysts

was advised, even in the asymptomatic patients, after discussion of the possible long-term complications.

Ultrasonography revealed a dilated bile duct in all patients; 10 had calculi in the cyst. Two patients with a choledocho-cysto-duodenostomy had a barium meal investigation; the contrast entered the cyst in only one patient.

Based on tube cholangiography and ERCP findings, nine patients were thought to have a type I cyst, five had type IVa cysts and one had type IVb cysts.

The pancreatic duct could be evaluated in 12 patients, of whom three had an abnormal pancreatic biliary duct junction. ERCP was done in patients with previous cystocentric drainage. In all but one no contrast was seen to enter the bile duct, indicating anastomotic stenosis; this included the two asymptomatic

patients. Calculi were evident in eight patients who had internal drainage and two with external drainage only.

At operation, mobilization of the cyst was complicated by pericystic adhesions especially in patients with a previous choledocho-cysto-duodenostomy; the entire hepatoduodenal ligament in these patients was inflamed with dense vascular adhesions around the choledochal cyst. The duodenum around the cyst-duodenal anastomosis was also involved in the inflammatory response. The anastomosis between the cyst and the bowel was either blocked completely or reduced to a small lumen with multiple stones packed in the cyst. Separation of the cyst from the portal vein was difficult. Two patients with choledocho-cysto-jejunostomy had pericholedochal varices due to portal hypertension. Excision of the cyst was possible in only one case.

Table-I: Indications for re-operative surgery following internal drainage procedures for choledochal cyst

Reference	Year	Number of patients	Indication for reoperation	Nature of reoperation
Takiff et al	1985	16 operations	Cholangitis (14) Pain and jaundice (1) Recurrent pain (1)	Excision (5) Bilioenteric drainage procedure (11)
Todani et al	1988	08	Cholangitis (5) Malignancy (3)	Excision and hepaticoduodenostomy (3) Excision and Roux-en-Y hepaticojejunostomy (3) Palliative bypass alone (1) Laparotomy alone (1)
Tan and Howard	1988	07	Cholangitis (5) Portal hypertension (2)	Excision (5) Cystojejunostomy (2)
Present series	2006	07	Pancreatitis (3) Cholangitis (2) Asymptomatic (2)	Excision (5) Cystojejunostomy (2)

Table-II: Experience with reoperation following external drainage of choledochal cyst

Reference	Year	Number of patients	Indication for external drainage	Outcome after excision
Todani et al	1987	08	Sever cholangitis (4) Acute pancreatitis (1) Bile peritonitis (2) Indication (1)	Favourable
Tan and Howard	1988	04	Cyst rupture (1) Cholangitis (3)	Favourable
Present series	2006	08	Pericyclic adhesions (3) Mistaken diagnosis (3) Perforation of cyst (2)	Favourable

hypertension, as seen in two patients in this series. The cyst could be excised in only one; large pericholedochal varices rendered excision difficult in the second patient. A portasystemic shunt is necessary before excision can be attempted⁴. Reversal of secondary biliary cirrhosis has been reported with drainage of the choledochal cyst. Because of the possibility of portal hypertension, upper gastrointestinal endoscopy should always be performed in patients who had a previous cyst drainage operation so that adequate preparation can be made for constructing a portasystemic shunt.

Pancreatitis is also more likely in patients who have had internal drainage operations, because the abnormal lower end of the cyst continues to cause reflux into the pancreatic duct. In one study¹⁷, 80% of patients had pancreatitis. Recurrence of pancreatitis can be prevented by excision of the cyst.

From experience it is seen that excision of the choledochal cyst was most difficult in patients who had previous drainage into the duodenum, and easiest in those who had external drainage. In the former patients, the hepatoduodenal ligament was fibrosed and shortened with vascular adhesions; the duodenum was also involved in the inflammatory process. Patients with previous

external drainage had adhesions around the track of the drain tube but the planes around the cyst could be dissected with ease. Although the drain is an avenue for extraneous infection, adequate biliary decompression prevents the development of cholangitis and secondary biliary cirrhosis. The choledochal cyst could be excised in all patients who had external drainage, but here it was done in only five of the seven who had internal drainage. In Todani et al's review¹⁰, in the eight patients, with external drainage excision was achieved with good results (Table-II), and of 19 patients with enteric drainage five underwent excision and in three only a palliative bypass could be performed because malignancy had developed. The remaining 11 patients were lost to follow up. Patients who have revision surgery may have higher incidence of bile leak after cyst excision than those who have primary cyst excision, possibly because proximal ducts are unhealthy as a result of cholangitis and calculi.

Cyst excision can be done safely after a cyst drainage operation in the majority of cases. In severely ill patients, external drainage of the choledochal cyst is a safe option and secondary excision can be done later. The presence of all external catheter ensures that the patient will return for definitive surgery.

References:

1. Todani T, Watanabe Y, Narasue M, et al. Congenital bile duct cysts. Classification, operative procedures, and review of thirty-seven cases including cancer arising from choledochal cyst. *Am J Surg* 1977; 134: 263-9.
2. Kasai M, Akasura Y, Taira Y. Surgical treatment of choledochal cyst. *Am Surg* 1970; 172: 844-51.
3. Trout HH III, Longmire WP Jr. Long-term follow-up study of patients with congenital cystic dilatation of the common bile duct. *Am J Surg* 1971; 121: 68-86.
4. Flanigan DP. Biliary cyst. *Am Surg* 1975; 182: 635-43.
5. Yamaguchi M. Congenital choledochal cyst. Analysis of 1433 patients in the Japanese literature. *Am J Surg* 1980; 140: 653-7.
6. Powell CS, Sawyers JL, Reynolds VH. Management of adult choledochal cysts. *Am Surg* 1981; 193: 666-76.
7. Deziel DJ, Rossi RL, Munson JL, et al. Management of bile duct cysts in adults. *Arch Surg* 1986; 121: 410-15.
8. Matsuyama S, Matsushima M. A long term follow up study of choledochal cysts undergoing cystoduodenostomy. *Jap J Pediatr Surg* 1970; 6: 293-303.
9. Nagorney DM, McIlrath DC, Adson MA. Choledochal cysts in adults. Clinical management. *Surgery* 1934; 96: 656-63.
10. Todani T, Watanabe Y, Toki A, Urushihara N, Sato Y. Reoperation for congenital choledochal cyst. *Ann Surg* 1988; 207: 142-7.
11. Nakaomra S, Kimura S, Ishitoya T. Reoperation of the choledochal cyst. *Zeitschrift fur Kinderchirurgie* 1977; 22: 41-50.
12. Takiff H, Stone M, Fonkilsrud FW. Choledochal cysts: results of primary surgery and need for reoperation in young patients. *Am J Surg* 1985; 150: 141-6.
13. Tan KC, Howard ER. Choledochal cyst: a 14-year surgical experience with 36 patients. *Br J Surg* 1988; 75: 892-5.
14. Flanigan DP. Biliary carcinoma associated with biliary cysts. *Cancer* 1977; 40: 880-3.
15. Todani T, Watanabe Y, Toki A, Urushihara N. Carcinoma related to choledochal cysts with internal drainage operation. *Surg Gynecol Obstet* 1987; 164: 61-4.
16. Yeong ML, Nicholson GI, Lee SP. Regression of biliary cirrhosis following choledochal cyst drainage. *Gastroenterology* 1982; 82: 332-5.
17. Swisher SG, Cates JA, Hunt KK, et al. Pancreatitis associated with adult choledochal cysts. *Pancreas* 1994; 9: 633-7.
18. Chaudhury A, Dhar P, Sachdev A. Reoperative surgery for choledochal cyst. *Brit J Surg* 1997; 84: 781—784.