Original Research Article

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Presentation and management of choledocal cyst

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ABSTRACT

Background: Choledochal cyst is congenital disease of biliary tract. There are five main types of choledochal cyst with few recognised sub types. The etiology of choledochal cyst is unclear.

Methods: Author here randomly studied patients from May 2012 to June 2015 from surgical department of the institute. Radical excision of cyst wall with reconstruction of biliary tract using a Roux-en-Y loop of jejunum is the treatment in a patient.

Results: Out of 30 patients, 24 were discharged as routine procedure. Out of 30, six patients had complications which were treated and cured, only one patient died due to ARDS post operatively.

Conclusions: Choledochal cyst is congenital disease. In the past, it was treated by drainage procedure. But nowadays, it is treated by radical excision with Roux-en-Y hepaticojejunostomy with minimum mortality. In present study, 3.33% (one patient) death occurred. So, in planned surgery results are good but in emergency (in this study one patient died) death can occur, inspite of good surgical approach.

Keywords: Choledochal cyst, Clinical presentation, Management

INTRODUCTION

Choledochal cysts are rare congenital. But not familial, anomalies of the intrahepatic or extrahepatic biliary tract. Choledocal cyst was first described by Vater and Ezler in 1723. 1,2 Cystic dilatation may affect every part of the biliary tree and may occur singly or in multiple numbers. Choledochal cysts are usually diagnosed in childhood and about 25% are detected in adult life.4 Choledochal cysts also have an unexplained female:male preponderance, commonly reported as 4.1 to 3:1.⁴ They are classified according to the location of biliary duct dilation as described by Todani et al.⁵ Complications of choledochal cysts include cholangitis, pancreatitis, secondary biliary cirrhosis, spontaneous rupture of cyst, and cholangiocarcinoma and mortality in case of choledochal cysts ranges from 10-15%. Improved imaging modalities have facilitated the diagnosis at any time from antenatal to adult life. Surgical management needs treatment ranges

from cysto-enterostomy to liver transplantation according to the type and presentation of choledochal cyst. In present study of presentation and management of choledochal cysts. Author had studied different views about the etiopathogenesis along with the natural course. Complications, diagnosis and surgical approach for choledochal cyst. It was an observational, interventional and open labelled and prospective study based on the disease, complications and follow up of the patients. The aim of the study was to study clinical cases of choledochal cysts with their demographic appearance, to study the clinical presentation and complications in cases of choledochal cysts in author's set up and to study results and outcome of this study.

METHODS

It was an observational, interventional and open labelled and prospective study based on the disease, complications and follow up of the patients. Present study was interventional, observational and open labelled. So, there was involvement and usage of surgical materials and drugs with availability from Government supply.

The data were collected according to demographic appearance (proforma) age, gender, presenting clinical symptoms, physical. Laboratory and histological findings, diagnostic methods, complications and treatment of choledochal cysts. Todani's classification was used for the determination of the cyst type.

Ultrasonography (USG), scintigraphy, computerized tomography (CT), percutaneous trans-hepatic cholangiography (PTC) and magnetic resonance (MR) cholangiography was used as diagnostic tools.

Histopathological confirmation of the diagnosis was obtained in all cases after excision, MRCP was done in most cases pre-operatively except in case where spontaneous rupture of cyst was operated in emergency setting. ERUP was done pre-operatively for diagnosis and relief of cholangitis in some selected cases.

This was a prospective, observational, interventional and open labelled study conducted in all surgical departments of this institute, where there was an availability of patients with choledochal cyst from May 2015 to June 2017.

Author studied 30 patients of choledochal cysts period ranges from May 2012 to June 2015.

Patient with age group, sex, those who willing for treatment and follow up and those patients fit for surgical intervention. Patient who was not willing for treatment and follow up, pregnant female, patient having cholangiocarcinoma and patient having life threatening morbidities were excluded.

Data analysis

All observations were recorded, and results were analysed statistically. Data were expressed as mean of

retrospective review of patients admitted and operated in this institution and it will be compared with other studies done by Indian as well as foreign researchers.

Study end points

Study was of 30 patients of choledochal cysts who had fulfilled inclusion criteria of patient selection of point VII of study period ranging from 30 May 2015 to 30 June 2017.

It was from 1 to 30 patient's first time consultation in this institution with his/her history up to final result and outcome according to proforma.

RESULTS

Choledochal cysts is rare anomaly of the intrahepatic or extrahepatic biliary tract and should be considered in the differential diagnosis in all patients with a history of biliary colic, recurrent cholangitis or pancreatitis with associated dilatation of bile duct, particularly if they are <40 years of age (Table 1). Delay in the diagnosis increases the incidence of associated biliary pathology and suboptimal surgical therapy.

Table 1: Age distribution.

Age distribution	Values
Mean age	31.52 years
Age range	21 days-68 years

Due to better availability of imaging techniques and recent advances in radio-diagnosis and invasive techniques, pre-operative diagnosis of choledochal cyst becomes easier and which makes better plan of management for planned surgery. Surgery is the mainstay of treatment of choledochal cyst. Radical excision cyst wall with reconstruction of the biliary tract using a Rouxen-Y loop of jejunum is the treatment and complete resection of the cyst is necessary because of the association with the development of cholangiocarcinoma.

Table 2: Distribution according to sex: comparison of present study with other studies.

Sex distribution	Present study	Pereira et al ⁷ (n=18)	Jesudason et al ⁸ (n=57)
Male	11 (36.66%)	3 (16.66%)	24 (42.11%)
Female	19 (63.34%)	15 (83.33%)	33 (57.89%)
M:F ratio	1:1.72	1:5	1:1.38

In present study, out of total 30 patients, mean age was 31.52 years and age range were 21 days-68 years. Out of total 30 patients, there were 11 males and 19 females with a male: female ratio of 1: 1.72 (Table 2). The most frequent choledochal cyst types among these patients

were type I and IVa, in 21 (70%) and 5 (16.66%) patients respectively (Figure 1).

Abdominal pain is the main symptom in most types (19 out of 21). Jaundice is mainly seen in type I (13 out of 21

patients) and IV (4 out of 4 patients) cysts, both (partly) extrahepatic (Table 3).

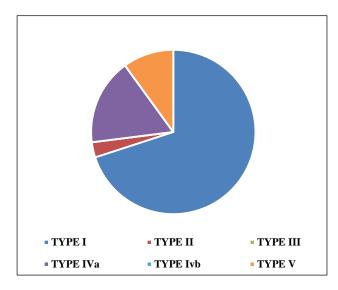


Figure 1: Types of choledochal cyst.

Table 3: Clinical presentation with associated pathologies-comparison of present study with other study.

Various presentations	Present study (n=30)	De Vries et al ⁶ (n=42)
Abdominal pain	27 (90%)	32 (76%)
Jaundice, alcoholic stools	18 (60%)	24 (57%)
Nausea/vomiting	17 (56.66%)	19 (45%)
Weight loss/ failure to thrive	11 (36.66%)	10 (23%)
Hepatomegaly	12 (40%)	14 (33%)
Palpable mass	4 (13.33%)	7 (16%)
Gall-stones	11 (36.66%)	10 (23%)
Cholangitis	9 (30%)	15 (35%)
Pancreatitis	7 (23.33%)	7 (16%)
Ruptured cyst	1 (3.33%)	0 (0%)

In liver function test, serum bilirubin and alkaline phosphatase were elevated in 60 % and 66.66%, respectively. Serum transaminase (SGOT and SGPT) were increased 53.33% and 56.66% respectively (Table 4).

Table 4: Liver function test: comparison of present study with other study.

Blood chemistry	Present study (n=30)	Jesudason et al ⁸ (n=57)
S. bilirubin (increased)	18 (60%)	16 (28%)
S. ALP (increased)	20 (66.66%)	35 (61.4%)

In present study, 25 out of 30 (83.33%), patients were diagnosed by ultrasonography (Table 5).

Table 5: Diagnostic tool: ultra-sonography comparison with other study.

Different studies	Ultrasonography as a diagnostic tool
Present study	83.33%
Pereira et al ⁷	77%
Singham et al ¹⁰	71%

MRCP was done in most cases pre-operatively except in case where spontaneous rupture of cyst was operated in emergency setting. ERCP was done in 6 out of 30 (20%) patients pre-operatively for diagnosis and relief of cholangitis. Procedure-related complications were noted in 6 out of 30 (20%) of the patients. Mortality was observed in 3.33% patient i.e. one patient was due to pneumonia on 15th post-operative period (Table 6).

Table 6: Post-operative complications: comparison of present study with other study.

Complications	Present study (N=30)	De Vries et al ⁶ (n=43)
Wound infection	1 (3.33%)	2 (4.76%)
Wound hematoma	1 (3.33%)	1 (2.38%)
Cholangitis	1 (3.33%)	1 (2.38%)
Bile leak	1 (3.33%)	1 (2.38%)
Incisional hernia	1 (3.33%)	1 (2.38%)
Death	1 (3.33%)	1 (2.38%)

Table 7: Age distribution-comparison of present study with other study.

Age group	Present study (n=30)	De Vries et al ⁶ (n=42)
Infants (<1 yr)	1 (3.33%)	10 (24%)
Children (1-16 yrs)	10 (33.33%)	11 (26%)
Adults (>16 yrs)	19 (63.33%)	21 (50%)

DISCUSSION

In this study of age distribution for all age group, out of total 30 patients mean age was 31.52 years and age range were 21 days-68 years.

In this study, out of 30 patients, one (3.33%) patient was infant, thirteen (43.33%) patients were children and sixteen (53.34%) patients were adults in compared to De Vries JS et al, where out of 42 patients.⁶ 10 (24%), 11 (26%), 21 (50%) patients were infants, children and adults respectively.

The most frequent choledochal cyst types out of 30 patients were type 1 and IVa in 21 (70%) and 5 (16.66%) patients, respectively. There was only one patient with

choledochal cyst type II, and 3 patients with choledochal cyst type V (Caroli's disease).

Present study was compared with De Vries et al, Periera et al, Jesudason et al. ⁶⁻⁸ Where in De Vries et al, study out of 42 patients most frequent choledochal cyst types were type I and type V. ⁶ 30 (71%) and 5 (12%) patients, respectively. There were 2 (5%) patient with choledochal cyst type 11, and 1 (2%) patient with choledochal cyst type III, and 4 (10%) patients with type IVa.

Where in Periera et al, study out of 18 patients most frequent choledochal cyst types were type I and type IVa, 09 (50%) and 7 (38%) patients, respectively. There were 1 (5%) patient with choledochal cyst type II and 1 (5%) patient with choledochal cyst type V.

Where in Jesudason et al, study out of 57 patients most frequent choledochal cyst is types ere type I and type Iva, in 41 (71.92%) and 13 (22.80%) patients, respectively. There were 2 (3.5%) patients with choledochal cyst type IVb, and 1 (1.75%) patient with choledochal cyst type V.

Abdominal pain is the main symptom in most types in this study. Jaundice is mainly seen in type I (13 of 21 patients) and IV (4 of 5 patients) cysts, both (partly) extrahepatic. Where in other study, abdominal pain is the symptom in most types along with jaundice is seen in type I (17 of 30 patients) and IV (4 of 4 patients).

Present study compared with Jesudason et al, in liver function test, serum bilirubin and alkaline phosphatase were elevated in 57% and 64.28%, respectively. Where in this study, S. bilirubin and S. AIPO were 60% and 66.66%, respectively.

Mean S. bilirubin was 3.58 mg/dl (normal range 0-1.0 mg/dl). Mean S. alkaline phosphatase was 102.41 U/L (normal range 38-94 U/L). Mean S. ALT (SGPT) and S. AST (SGOT) were 57.14 U/L (normal range O-42U/L) and 50.1U/L (normal range O-37 U/L) respectively. In this study, S. ALT (SGPT) and S. AST (SGOT) were elevated in 53.33% and 56.66% patients which were compared with Savić et al, study in which both are increased 69%. 9

In present study, 25 out of 30 (83.33%) patients were diagnosed by ultrasonography which was compared with others studies Pereira et al, Singham et al, 77% and 71 % respectively. MRCP was done in most of cases preoperatively except in case where spontaneous rupture of cyst was operated in emergency setting. ERCP was done in 6 out of 30 (20%) patients pre-operatively for diagnosis and relief of cholangitis in some cases.

In treatment modility, surgery is the only choice of management and complete removal of cyst wall is the mainstay of treatment Roux-en-Y- Hepatico-Jejunostomy is the surgical management of choice in most of the cases, while cyst excision is the management for Type-II choledochal cysts. In this study, out of 30 cases, most of cases 26 (86.66%) were operated by Roux-en-Y-Hepatico-jejunostomy as a surgical procedure and only one patient (3.33%) was operated with simple cyst excision and 3 (10%) patients were managed conservatively with Caroli's disease as there are no facilities of hepatic lobectomy or liver transplantation available in this institution.

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Ethical approval: The study was approved by the

Institutional Ethics Committee

REFERENCES

- Vater A. Dissertatioo Inaugualis Medica. Proes. Diss. Qua Scirrhis Vicerum Disseret C. S Ezerlus, 4 Wittembergaes Pamphlets. Another copy H22, 70, 19, Edinburaglh, University Library, 1723;881:1,22.
- Edil BH, Cameron JL, Reddy S, Lum Y, Lipsett PA, Nathan H, et al. Choledochal cyst disease in children and adults: a 30-year single-institution experience. J Am Coll Surg. 2008;206(5):1000-5.
- 3. Waidner U, Henne-Bruns D, Buttenschoen K. Choledochal cyst as a diagnostic pitfall: a case report. J Med Case Rep. 2008;2(1):5.
- 4. Liu CL, Fan ST, Lo CM, Lam CM, Poon RT, Wong J. Choledochal cysts in adults. Arch Surg. 2002;137(4):465-8.
- Todani T, Watanabe Y, Narusue M, Tabuchi K, Okajima K. Congenital bile duct cysts: classification, operative procedures, and review of thirty-seven cases including cancer arising from choledochal cyst. Am J Surg. 1977;134(2):263-9.
- 6. De Vries JS, De Vries S, Aronson DC, Bosman DK, Rauws EA, Bosma A, et al. Choledochal cysts: age of presentation, symptoms, and late complications related to Todani's classification. J Pediatric Surg. 2002;37(11):1568-73.
- 7. Pereira LH. Bustorff-Silva JM, Sbraggia-Neto L, Bittencourt DG, Hessel G. Choledochal cyst: a 10 year experience. Jornal de Pediatria. 2000;76(2):143-8.
- 8. Jesudason SB, Jesudason MR, Mukha RP, Vyas FL, Govil S, Muthusami JC. Management of adult choledochal cysts-a 15-year experience. HPB. 2006;8(4):299-305.
- 9. Savić DJ, Milovanović D, Jovanović D. Congenital dilatation of the common bile duct (congenital choledochal cyst). Srpski Arhiv Za Celokupno Lekarstvo. 2001;129:47-50.
- Singham J, Schaeffer D, Yoshida E, Scudamore C. Choledochal cysts: analysis of disease pattern and optimal treatment in adult and paediatric patients. HPB. 2007;9(5):383-7.

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