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Original Article

Clinical Study and Management of Choledochal Cyst

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Abstract

Introduction: Choledochal cyst is defined as an isolated or combined congenital dilatation of the extra or intrahepatic biliary tree. Choledochal cyst is not an isolated entity but rather regarded as a constellation of pathological anomalies in the hepato-pancreatico-biliary system. In children, the classical findings include a right upper quadrant abdominal mass, jaundice, and abdominal pain. In the adult, however, it is more complicated. Hence, the diagnosis is often confused with benign disease of biliary tract or pancreas. The most reliable and primary mode of investigation is the ultrasonography. This is followed by cholangiographic modalities like magnetic resonance cholangiopancreatography, particularly in adults to confirm the diagnosis, to know the associated complications, to know the probable etiology, and to delineate the exact anatomy of the cyst and intra- and extra-hepatic biliary tree for surgical management. The management depends on the type of cyst and its associated complications. Choledochal cyst excision with reconstruction through biliary-enteric Roux-en-Y anastomosis rather than cyst drainage alone has become the mainstay of the treatment for most types of choledochal cysts. Exceptions to this are the treatment of Type-III and Type-V choledochal cysts, where they were managed conservatively with stenting, sphincterotomy, and hepatectomy for liver transplant, respectively.

Aim: This study aims to study the age and sex, clinical presentation, diagnostic modalities used in detection, complications, and surgical management of choledochal cyst in Osmania general hospital over the period of January 2016–November 2017.

Materials and Methods: The present prospective clinical study includes 20 patients of all ages having choledochal cyst presented to the department of general surgery and surgical gastroenterology in Osmania general hospital. All the patients have undergone both medical and surgical management. The medical management includes pre-operative stenting with 8F/10F double pigtail stent to reduce the levels of jaundice and control of cholangitis before surgery. Surgical includes complete excision of cyst with Roux-en-Y hepaticojejunostomy, end-to-side anastomosis was done. The patients are observed postoperatively for complications such as wound infections, bile leak, pancreatitis, and cholangitis and the results were analyzed.

Results and Conclusions: The incidence of choledochal cyst in Osmania general hospital (admission is 1:100,000) is comparable to Asian population statistics. Incidence among female:male is about 1.5:1. Mean age of incidence is 29 years. Recurrent cholangitis is the most common clinical presentation in this series. Ultrasound abdomen is the primary mode of investigation with 96% sensitivity; endoscopic retrograde cholangiopancreatography has 100% sensitivity. Type I choledochal cyst is the most common presentation; Types II and V are not encountered. Associated complication incidence in this series is 60%, comparable to western series. The incidence of malignancy is 0% in the present series against 3–8% in western series.

Key words: Choledochal cyst, Complications, Endoscopic retrograde cholangiopancreatography, Hepaticojejunostomy, Ultrasound

INTRODUCTION

Choledochal cyst is defined as an isolated or combined congenital dilatation of the extra- or intra-hepatic biliary tree.

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Choledochal cyst is not an isolated entity but rather regarded as a constellation of pathological anomalies in the hepato-pancreatico-biliary system.

The incidence of this disease is much more frequent in Asians, particularly Japanese than in Europe or North America.

This also shows sex bias being more predominant in females.

Bile duct cysts are typically a surgical problem of infancy and childhood. However, in nearly 20% of patients with bile duct cysts, the diagnosis is delayed until adulthood.

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In contrast of the pediatric patient, adults have increased rate of associated biliary pathology and they often present with complications of previous cyst-related procedures.

In children, the classical findings include a right upper quadrant abdominal mass, jaundice, and abdominal pain. In the adult, however, it is more complicated. Hence, the diagnosis is often confused with benign disease of biliary tract or pancreas.

Table 1: Incidence of clinical features in the present series of 15 cases

Incidence of clinical features		
Clinical features	**Number of cases**	**Percentage of cases (%)**
Pain abdomen	20	100
Fever	7	35
Jaundice	9	45
Mass abdomen	2	20
Nausea/vomiting	4	22
Hepatomegaly	4	22
Upper GI bleed	0	0

GI: Gastrointestinal

Table 2: Modes of imaging studies used in this series

Mode of Imaging	Number of cases
USG abdomen	20
USG alone only	0
ERCP	3
MRCP	20
CECT abdomen	15

USG: Ultrasound imaging, CECT: Contrast-enhanced computed tomography, MRCP: Magnetic resonance cholangiopancreatography, ERCP: Endoscopic retrograde cholangiopancreatography

Table 3: Laboratory investigations in the present series

Laboratory investigations	
Tests	**Number of cases**
Hyperbilirubinemia	8
Increased alkaline phosphatase	10
Increased serum amylase	3

The most reliable and primary mode of investigation is the ultrasonography. This is followed by cholangiographic modalities like (magnetic resonance cholangiopancreatography [MRCP]), particularly in adults to confirm the diagnosis, to know the associated complications, to know the probable etiology, and to delineate the exact anatomy of the cyst and intra- and extra-hepatic biliary tree for surgical management.

The management depends on the type of cyst and its associated complications. Choledochal cyst excision with reconstruction through biliary-enteric Roux-en-Y anastomosis rather than cyst drainage alone has become the mainstay of the treatment for most types of choledochal cysts. Exceptions to this are the treatment of Type-III and Type-V choledochal cysts, where they were managed conservatively with stenting, sphincterotomy, and hepatectomy for liver transplant, respectively.

The importance choledochal cyst lies in it being an integral part of differential diagnosis of the intermittent jaundice with pain abdomen in adults and as mass abdomen in the right hypochondrium with or without jaundice in children. As they are not frequently encountered, its importance is frequently undermined.

The presence of choledochal cyst also necessitates thorough the evaluation of the whole of hepato-pancreatico-biliary system as these organs may also be intrinsically involved or are affected due to contiguity.

Recent advances in imaging studies have facilitated increased detection of biliary tree anomalies, which led to increased incidence of choledochal cysts and have widened its spectrum of management. This paved a way for pioneering minimal access methods of management of choledochal cysts which led to improved patient outcomes; hence, revisiting the disease through this study gives us a bird's eye view as to the recent advances in this field.

Table 4: Different surgical procedures according to the type of cysts

Surgical, management			
Type of cyst	**Number of cases**	**Excision**	**Anastomosis**
Ia	1	15 cases complete excision one case Lilly's modification	Roux-en-Y end-to-side hepaticojejunostomy
Ib	8	Complete excision	Roux-en-Y end-to-side hepaticojejunostomy
Ic	5	Complete excision	Roux-en-Y end-to-side hepaticojejunostomy
III	1	Complete excision	Transduodenal excision
IVa	4	Complete excision	Roux-en-Y end-to-side hepaticojejunostomy
IVb	1	Complete excision	Roux-en-Y end-to-side hepaticojejunostomy

Table 5: Complications of surgery in number of cases

Complications of surgery		
Complications	**Number of cases**	**Percentage (%)**
Anastomotic leak	4	22
Wound infection	3	17
Atelectasis	4	22
DVT	1	5
Death	1	5

Table 6: Incidence of the type of cysts in the present series

Type of cysts in the present series		
Type	**Number of cases**	**Percentage (%)**
Type 1	14	70
Type II	0	0
Type III	1	5
Type IV	5	25
Type V	0	0

AIMS AND OBJECTIVES OF STUDY

Analysis of choledochal cyst cases presenting to Osmania general hospital over the period of January 2016–November 2017.

- To study the age and sex incidence of choledochal cyst.
- To study the clinical presentation of choledochal cyst.
- To study the diagnostic modalities used in detection of choledochal cyst.
- To study the complications of choledochal cyst.
- To study the surgical management of choledochal cyst in Osmania general hospital.

MATERIALS AND METHODS

Study Design

The present prospective clinical study includes 20 patients of all ages having choledochal cyst presented to the department of general surgery and surgical gastroenterology in Osmania general hospital. The present study period is between January 2016 and November 2017.

Inclusion Criteria

The following criteria were included in the study:

- Patients of all ages
- Both male and female patients
- Patients with clinical features and diagnosis suggestive of choledochal cyst.

Exclusion Criteria

- Patients with similar clinical features due to causes other than choledochal cyst such as biliary stone disease, benign or malignant stricture of biliary tree, extrahepatic biliary atresia, and periampullary carcinoma were excluded from the study.

Table 7: Complications of cyst in percentage of cases

Complication of cyst itself		
Complications	**Number of cases**	**Percentage (%)**
Cystolithiasis	3	16
Choledocholithiasis	4	20
Cholelithiasis	1	4
Pancreatic leak	2	9
Cholangiocarcinoma	0	0
Cirrhosis	0	0
Intrahepatic abscess	0	0
Spontaneous perforation	0	0

Table 8: Sex variation in different series

Series	Female: male ratio
Present series	78:22
Wani[19]	65:35
Kandelwal[20]	56:44
56: 44 Chijiiwa and Koga[21]	89:11

Table 9: Mean age in different series

Age presentation	
Series	**Mean age (years)**
Present series	29
Wani[19]	27
Chijiiwa and Koga [21]	24
John Hopkins[16]	23

Methods

Clinical features

The clinical features of patient are compared to the statistics of standard textbooks and original series of different authors.

The clinical features considered are as follows:

- Pain abdomen
- Jaundice
- Fever
- Mass abdomen
- Nausea and vomiting
- Upper gastrointestinal (GI) bleeding.

Biochemical Tests

These tests are done to know the complications associated with choledochal cyst and to prepare the patients for surgery. Tests that are done - liver function tests, serum amylase, serum proteins, blood grouping and typing, blood sugar, blood urea, etc.

Imaging Studies

Ultrasonography of abdomen is done to all patients by Esaote MyLab Classic C, with 3.5 MHz curvilinear probe.

Table 10: Clinical features in the percentage in different series

Presenting symptoms				
Symptoms	**Present series (%)**	**Wani[16]**	**Chijii and Koga[18]**	**John Hopkins[15]**
Pain abdomen	100	85%	78%	87%
Fever	35	42.8%	28%	26%
Jaundice	45	35%	43%	42%
Mass abdomen	20	17.8%	13%	33%
Classical triad	5	7%	15%	0%

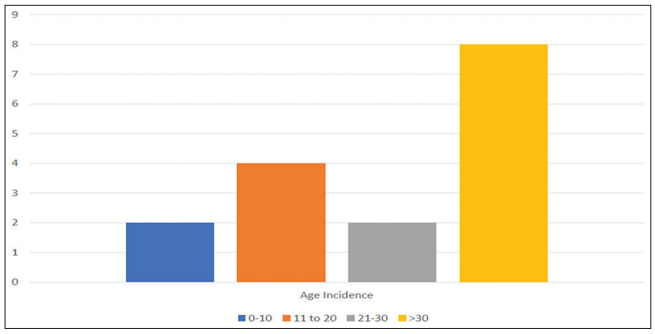

Figure 1: Bar diagram showing incidence

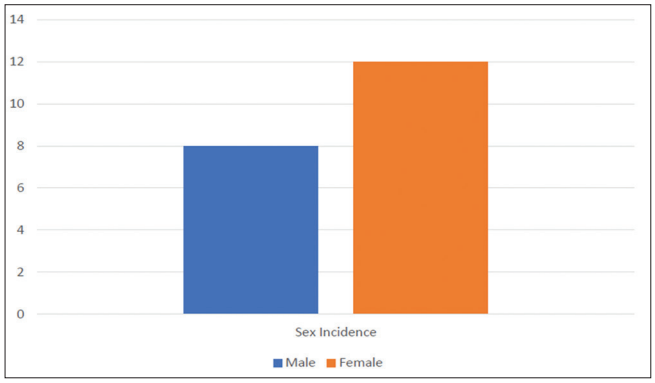

Figure 2: Bar diagram showing sex incidence

Some of these patients were subjected to endoscopic retrograde cholangiopancreatography (ERCP) with side view Duodenoscope, OLYMPUS JF 30 or TJF-20 Videoscope with the help of TOSHIBA fluoroscopy.

Some of these patients were subjected to computerized tomographic scan and MRCP.

Management

All the patients have undergone both medical and surgical management.

The medical management includes pre-operative stenting with 8F/10F double pigtail stent to reduce the levels of jaundice and control of cholangitis before surgery.

Surgical Management

The patients are prepared preoperatively with stenting and antibiotics, Vitamin K injection, and fresh frozen plasma to control jaundice and cholangitis.

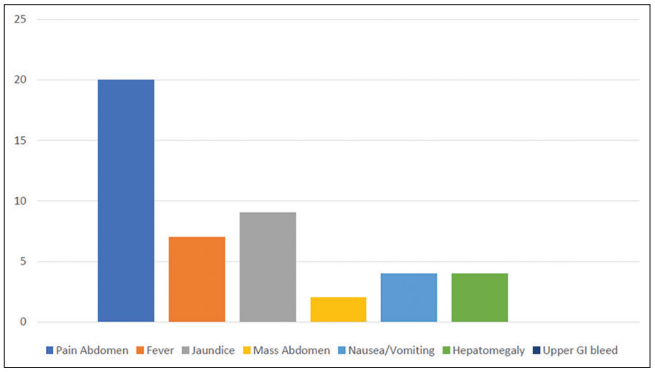

Figure 3: Bar diagram showing incidence of clinical features

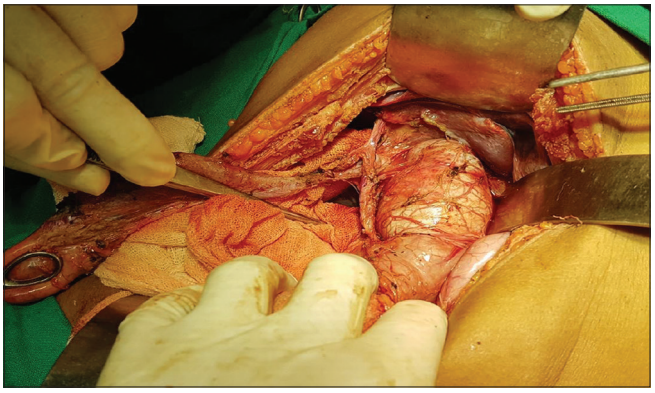

Figure 4: Dissection and exposure of choledochal cyst Type Ic

Complete excision of cyst with Roux-en-Y hepaticojejunostomy (HJ), end-to-side anastomosis with 4-0 vicryl was done. The patients had a drain kept in Morrison's pouch.

Complications during surgery looked for are primary hemorrhage (> 100 ml) and injury to major structures.

The patients are observed postoperatively for complications such as wound infections, bile leak, pancreatitis, and cholangitis. The post-operative period is about 15–20 days.

The specimens of cysts in all patients are subjected to histopathology examination to diagnose cyst itself and particularly for evidence of malignancy. Hematoxylin and eosin stains are used routinely.

The complications of cyst are diagnosed by clinical features, biochemical studies, imaging, surgical exploration, and histopathological examination of cyst.

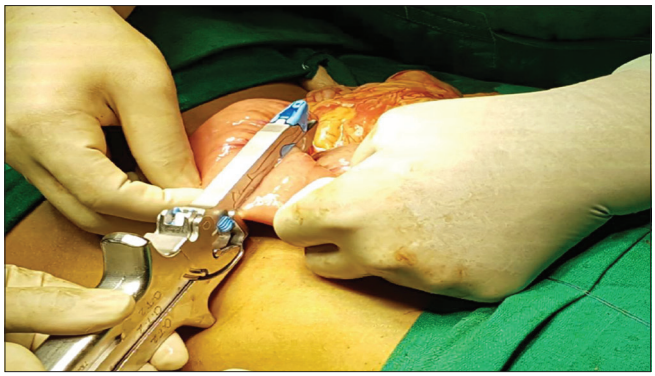

Figure 5: Creation of Roux-en-Y jejunal limb

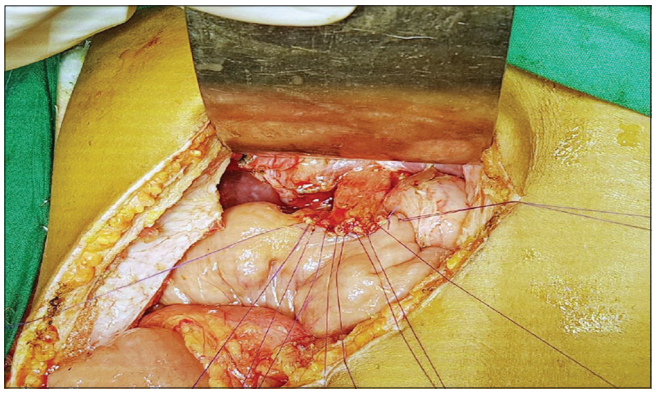

Figure 6: Roux-en-Y hepaticojejunostomy

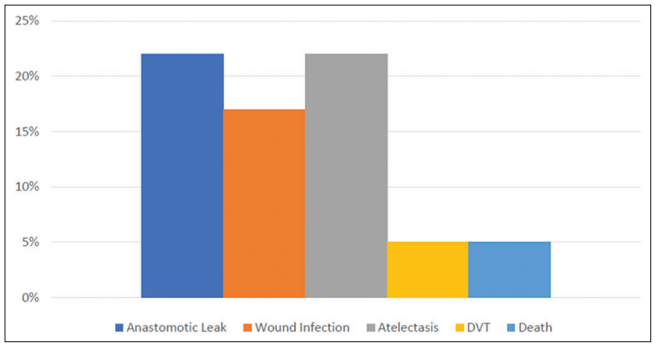

Figure 7: Bar Diagram showing post-operative complications

RESULTS AND ANALYSIS

Age Incidence

In the present series, age group is extending from 2 years to 65 years. The youngest patient is 2 years old and the oldest is 65 years old. The total number of patients in 0–10 years age group is 2, between 11 and 20 years age group is 4, between 21 and 30 years is 6, and above 30 years of age is 8.

The mean age in the present series is 29 years.

SEX INCIDENCE

In this series, 12 female patients and eight male patients are present. Female-to-male ratio is 1.5:1.

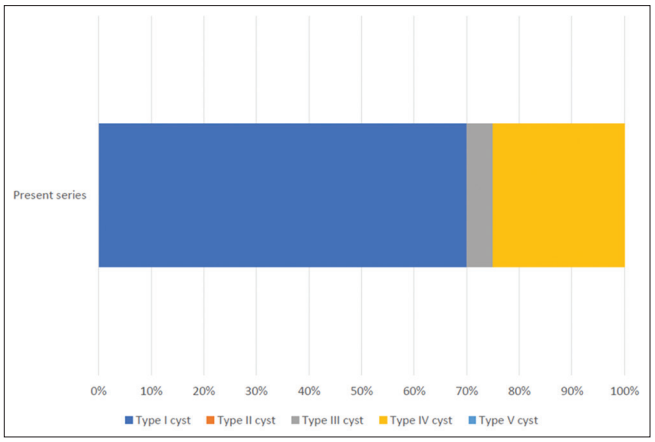

Figure 8: Stack graph of incidence of the types of cysts in the present series

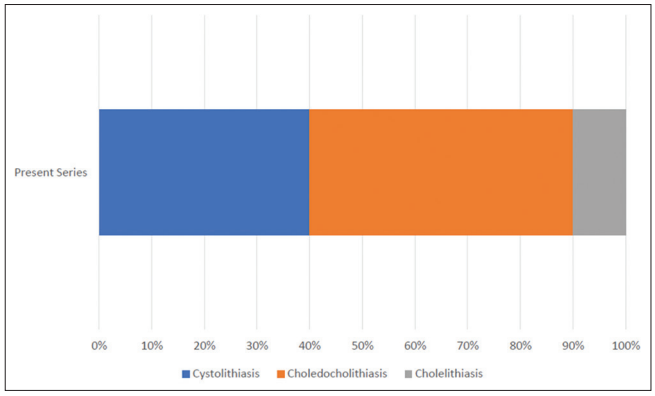

Figure 9: Stack graph of various complications of the choledochal cyst in the present series

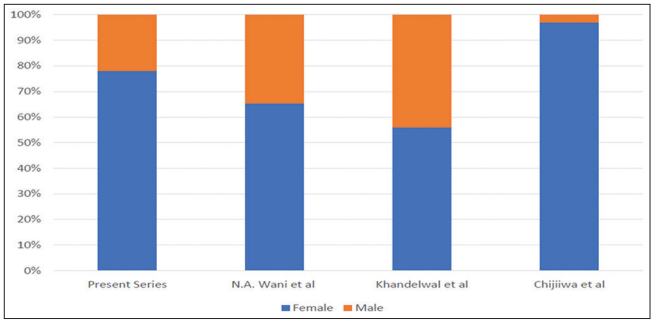

Figure 10: Graph comparing sex incidence in different series

All the patients in the present series had pain abdomen which is usually recurrent, episodic, dull aching type, in the right hypochondrium, and epigastric region with or without radiation.

Next common symptom is jaundice, seen in 14 patients and is usually associated with pain abdomen.

Fever is seen in 11 patients and is usually associated with pain abdomen and jaundice.

The clinical features of choledochal cyst - pain abdomen, jaundice, and mass per abdomen are present in six patients, of which four patients are <15 years.

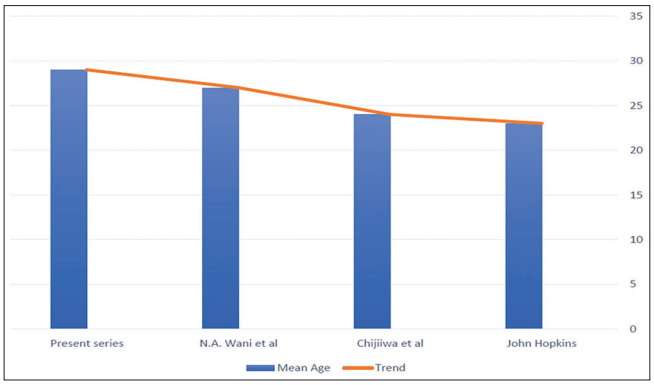

Figure 11: Bar diagram showing mean age presentations in different series

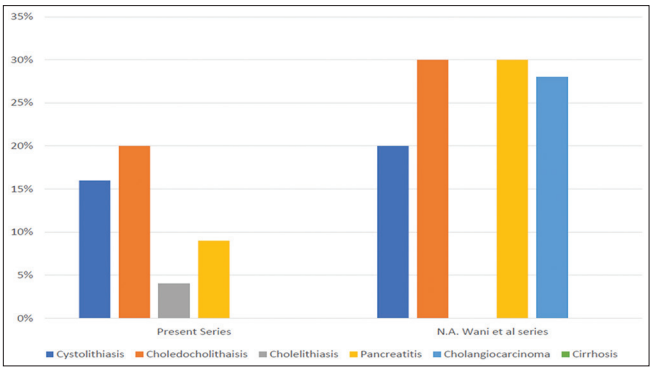

Figure 13: Bar diagram comparing complications of cysts in different series

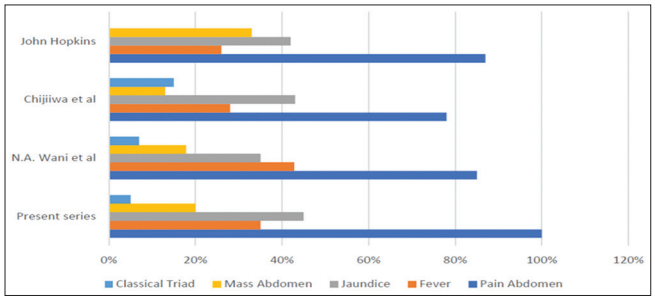

Figure 12: Graph comparing incidence of clinical features in different series

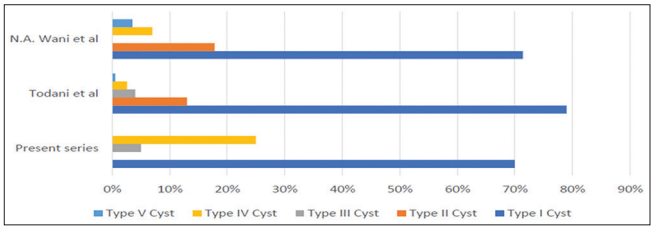

Figure 14: Bar diagram showing incidence of the types of choledochal cysts

The upper GI bleeding with features of portal hypertension is not seen in this series.

Three patients presented with fever, pain abdomen, and jaundice for which ERCP was done and stenting was done to control obstructive jaundice.

Four patients presented with choledocholithiasis and two presented with pancreatitis.

The provisional diagnosis before investigations in most of the patients is either acute or chronic cholecystitis or obstructive jaundice due to common bile duct (CBD) stones. In most of the patients, choledochal cyst was considered as a differential diagnosis.

Ultrasonography of abdomen was done in all patients as a primary mode of imaging. Of 20 cases, 16 cases were diagnosed as choledochal cyst.

In other cases, further investigation with ERCP, MRCP, or computed tomography scan abdomen was done to confirm and defined anatomical variations and extent of choledochal cyst.

ERCP was done in the three patients after ultrasonography. It diagnosed choledochal cyst in all the three patients. In two patients, CBD stricture was noted at the lower end. Stenting was done for three patients.

All patients were managed by IV antibiotics. Nil oral for 2 days. No complications were noted.

In most cases, surgery was done 3–5 days after ERCP, which should be done within 24 h.

All patients underwent MRCP for proper delineation of biliary tree anatomy and plan for surgery.

Ten patients showed raised serum alkaline phosphatase levels, as high as 1843 Kaw/dl. The three patients showing high level of serum amylase indicate possible presence of pancreatitis.

Pre-operative management included usage of antibiotics, stenting, Vitamin K injections, and bowel preparation.

In all patients, the right subcostal incision was taken.

In 17 patients, cholecystectomy was done. Pre-operative stenting was done in three patients.

In Type IV cases, extrahepatic cysts were completely excised and HJ was done.

Kehr's T-tube was kept in five cases for which T-tube cholangiogram was done on the 10th post-operative day

Table 11: The complications of cyst at presentation in percentages

Associated pathology of cyst		
Complication of cyst	**Present series (%)**	**Wani[19] series (%)**
Cystolithiasis	16	20
Choledocholithiasis	20	30
Cholelithiasis	4	0
Pancreatitis	9	30
Cholangiocarcinoma	0	28
Cirrhosis	0	0

Table 12: Incidence of the types of cysts in different series

Type of cyst			
Type	**Present series (%)**	**Todani (%)**	**Wani[19] (%)**
1	70	79	71. 4
II	0	13	17 8
III	5	4	0
IV	25	2. 6	7
V	0	0. 5	3. 5

Table 13: Type of surgery performed in different series

Type of surgery performed	
Series	**Type of surgery**
Present series	Complete cyst excision with Roux-en-Y hepaticojejunostomy
Narayan *et al*.[22]	Hepaticoduodenostomy and hepaticojejunostomy
Li *et al*.[23]	Laparoscopic Roux-en-Y hepaticojejunostomy

and was blocked, and then, T-tube was completely removed in 6 weeks.

Anastomotic leak was seen in four cases which were managed conservatively, and which subsided spontaneously in 5–10 days.

Wound infection was seen in three cases managed by daily dressings, drainage of abscess, and secondary suturing.

One case developed choledochal deep vein thrombosis, a patient of 56 years for which low-molecular-weight heparin was given.

One patient developed shock postoperatively and succumbed to death on the day of surgery. Significant intraoperative bleeding was not seen in this series.

In this series, type of cyst is determined by imaging studies and surgical exploration.

16 patients had Type I cyst - specifically Type Ib cyst.

One patient had Type 3 cyst

Five patients had Type IV cyst - which was diagnosed preoperatively and confirmed during surgery

Type II and V cysts are not encountered.

Cystolithiasis is seen in three patients.

Choledocholithiasis is seen in four patients.

Cholelithiasis is seen in one patient.

Pancreatitis is seen in two patients.

Associated hepatobiliary malignancy is not seen in the present series [Figures 1-14 and Tables 1-13].

ANALYSIS AND DISCUSSION

The present series of 20 cases from January 2016 to November 2017 is compared with different series as presented in different journals and textbooks.

The incidence in the present series is comparable to published data to be at 3–4:1. The present series has 3.5:1 or 78:22 for 100 cases, which is same as other series.

The mean age in the present series is 17.24 years, which is less compared to the published data. The present series has age group ranging between 7 and 56 years.

These data of present series can be subjected to selection bias in view of lack of the patients of pediatric age group.

In the present series, pain abdomen is present in all patients, whereas in other series, it is only 85%, cholangitis with fever is seen in 35% of cases, which is comparable to other series.

The classical triad presentation in this series is 5%, which is comparable to other published data.

The presentation of choledochal cyst as mass abdomen in the present series is 20%, comparable to other Indian series.

After pain abdomen, jaundice is the most common presentation in this series with 45%.

These clinical parameters are subject to detection bias if jaundice is not present in the patient which may diverge from the diagnosis of choledochal cyst.

The total percentage of complications associated with the cyst at presentation in this series is 55.5%, which is

high when compared to western series but is same when compared to Indian series.

The high rate in the present series may be due to high rate of gallbladder disease and cholangitis associated with choledochal cyst.

In the present series, the incidence of malignancy and associated cirrhosis is zero, whereas in western series, the malignancy rate is 20%.

The complication of cystolithiasis is more common in this series.

This series is subject to attrition bias as some of the data is lost due to lack of follow-up during the study.

Hence, malignancy in cases following surgery or in those patients who have not consented for surgery could not be evaluated.

Type I cyst is the most common type of cyst in this series, about 88%, which is not comparable to other series.

Incidence of Type III cyst is 5%.

The incidence of Type IV cyst in this series is 25%, which is high when compared to other series. The incidence of Types II and V cyst is 0% in the present series, whereas in western series, it is 13%, 4%, and 0.5%, respectively.

The two most commonly performed methods of reconstruction after cyst excision are hepaticoduodenostomy (HD) and Roux-en-Y HJ, of which the HJ is favored by most surgeons. Evidence concerning the optimal method of reconstruction is, however, sparse.

In the present series, all the cases were operated using the more popular anastomotic method, that is, Roux-en-Y HJ.

In the meta-analysis study of Narayan *et al*., six retrospective studies were included, comprising a total of 679 patients, 412 of whom (60.7%) underwent HD, and the remainder, 267 (39.3%) underwent HJ. Although HD group had slightly shorter hospital stay (MD: 0.30; 95% confidence interval [CI]: −0.22–0.39; $P < 0.00001$), it showed a higher incidence of post-operative reflux/gastritis (odds ratios: 0.08; 95% CI: −0.02–0.39; $P = 0.002$). However, the other outcomes such as bile leak, cholangitis, anastomotic stricture, bleeding, operative time, reoperation rate, and adhesive intestinal obstruction did not differ between HD and HJ groups.[25]

HD shows higher post-operative reflux/gastritis than HJ but a shorter hospital stay. There are few good quality studies that compare the outcomes from HD and HJ, meaning that caution should be exercised in the generalization of the results of this meta-analysis, which suggests HD to be comparable with HJ in terms of other complications, operative benefits, and outcomes.[19]

Minimal access surgery has considerably decreased post-operative hospital stay as explained by the case series of Li *et al*.[20]

Thirty-five patients with choledochal cyst were studied. Their age ranged from 3 months to 9 years (average age, 3.6 years). The choledochal cysts were cyst type in 33 cases and fusiform type in the other two cases. Four trocars were utilized with 3–5-mm instrumentation. Under laparoscopic guidance, the gallbladder and the dilated bile duct were excised completely. The Roux-en-Y jejunojejunostomy was performed extracorporeally by exteriorizing the jejunum through the extending umbilical incision (1.5–2 cm), and an end-to-side HJ was carried out intracorporeally by the hand suture methods.

Average duration of operation was 4.3 h (range, 3.5–7.6 h), intraoperative blood loss was 5–10 mL, and eight of the 35 patients had associated hepatic ductal stenosis and underwent laparoscopic excision of the cyst and ductoplasty. In one of the eight cases, bile leak was noticed from day 1 through 26 postoperatively. The post-operative course was uneventful in the other 34 patients with a hospital post-operative stay ranging from 3 to 6 days. There were no post-operative complications in 3-month–12-year follow-up.

As HD and laparoscopic HJ were not performed in cases of the present series, the differences in outcomes cannot be outlined.

Limitations of Study

- The present study is limited to patients presenting to a teaching hospital. Hence, the incidence and other statistics cannot be interpolated to general population.
- A larger series is needed to postulate definite recommendations of the management of choledochal cyst.
- Loss of follow-up can lead to attrition bias.
- Outcomes of minimal access invasive procedures like laparoscopic Roux-en-Y HJ could not be evaluated.

CONCLUSIONS

- The incidence of choledochal cyst in Osmania general hospital (Admission is 1:100,000) is comparable to Asian population statistics.

- Incidence among female:male is about 1.5:1.
- Mean age of the incidence of choledochal cyst is 29 years.
- Recurrent cholangitis is the most common clinical presentation in this series.
- The incidence of classical triad of symptoms (pain abdomen, jaundice, and mass abdomen) is 5%, which is comparable to other series.
- Ultrasound abdomen is the primary mode of investigation with 96% sensitivity, ERCP has 100% sensitivity.
- Type I choledochal cyst is the most common presentation; Types II and V are not encountered.
- Associated complication incidence in this series is 60%, comparable to western series.
- The incidence of malignancy is 0% in the present series against 3–8% in western series.
- Surgical excision of cyst is the main modality of treatment, more so in our population due to poor follow-up.
- Complication after surgery is 60%, which is high.

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