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ORIGINAL ARTICLE

Surgical aspects of choledochal cyst in children and adults: an experience of 106 cases

Pornchai Achatsachat[1] · Chayutra Intragumheang[1] · Nimmita Srisan[1] · Katawaetee Decharun[1] · Prapapan Rajatapiti[1] · Somboon Reukvibunsi[1] · Krit Kitisin[1] · Supparerk Prichayudh[1] · Suppa-Ut Pungpapong[1] · Bunthoon Nonthasoot[1] · Pongserath Sirichindakul[1] · Paisarn Vejchapipat[1]

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Abstract

Purpose To describe clinical features of choledochal cyst (CC) patients in terms of demographic data, clinical presentation, investigations, treatment, and outcomes among children and adults.

Methods The medical records of patients undergoing choledochal cyst (CC) surgery from 2002 to 2021 at a university hospital were retrospectively reviewed. The patients were divided into two groups: children (< 15 years) and adults (≥ 15 years). Descriptive statistics were used.

Results There were 106 cases of CC (Female/male = 88/18, children/adult = 53/53). Abdominal pain was the predominant presenting symptom, followed by jaundice in both groups. Adults were significantly more prone to present with abdominal pain compared to children (86.8% vs. 52.8%; $p < 0.001$), while children were more likely to experience acholic stool than adults (22.6% vs. 3.8%; $p = 0.004$). Ultrasound was the preferred investigation screening modality (75.5%). Most patients were presented with type I CC (71.7%). Laparoscopic-assisted approach was performed in 8.5%. CC excision with roux-en-y hepatico-jejunostomy was the main procedure (88.7%). Adults had a higher incidence of post-op complications, including stones, anastomosis stricture, abdominal collection, and cholangitis. Adults were significantly more likely to require intervention after surgery, compared to children (26.4% vs. 5.7%; $p = 0.04$).

Conclusions Ultrasound was the most common screening tool for diagnosis. Postoperatively, adults with CC experience more serious post-op complications compared to children. This could be attributed to long-standing cystic inflammation. Therefore, prompt definitive surgery is recommended for CC patients.

Keywords Choledochal cyst · Children · Adults

Introduction

Choledochal cysts (CC) are congenital dilatations of the biliary tree. They predominantly occur in children, particularly in East Asian populations, with an incidence of 1:1000 compared to 1:100,000–150,000 in Western populations [1] Females face a higher risk, with nearly a 4:1 female preponderance over males [2, 3]. The exact etiology of CC remains incompletely understood. In 1969, Babitt described an anomalous pancreatobiliary union (APBDU) in three children with CC [4], hypothesizing it as a possible etiology leading to the reflux of pancreatic juice into the common bile duct. This process results in chronic inflammation, damage to the bile duct wall, and cystic changes. The Todani's classification is the most widely used to categorize CC [5, 6], with Type I being the most commonly encountered, followed by Type IV. Both types carry a higher risk of malignancy. As a consequence, prompt and complete surgical excision stands as the mainstay of treatment when feasible [2, 3].

The presentation of CC varies between children and adults, with resection associated with a degree of morbidity. Many series focusing on adults with CC have suggested an increased incidence of adult CC disease [7–10]. However, it remains unclear whether CC exhibits a slow

Pornchai Achatsachat is presenting author.

✉ Paisarn Vejchapipat
paisarnv@gmail.com

[1] Department of Surgery, Faculty of Medicine, Chulalongkorn University & King Chulalongkorn Memorial Hospital, Bangkok 10330, Thailand

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progression, stays undetected for some time, or develops later in life. Some studies suggest that CC in older adults is likely a different entity than that in childhood. Thus, close surveillance may be a preferable option over resection and reconstruction, considering the associated risks of long-term morbidity [11]. Several studies have explored CC in both adults and children. Soares et al. demonstrated that adults exhibited a higher readmission rate than children (32.1% vs. 22.2%; $p = 0.04$) and were more likely to undergo biliary procedures after resection (22.4% vs. 5.2%; $p < 0.001$) [7]. Additionally, Zheng et al. found that adults with Type IV-A CC had a higher morbidity of biliary stricture and/or lithiasis than children ($p = 0.041$) [8]. In a study by Senthilnathan et al., which focused on both children and adults undergoing laparoscopic management, three adults required conversion, and adults experienced a higher rate of post-operative complications (14.54% vs. 5.45%) [9].

The objective of our study was to describe the clinical features of CC patients in a university hospital in terms of demographic data, clinical presentation, investigations, treatment, and outcomes among both children and adults. The exploration of disparities in CC disease between these two age groups aims to enhance our comprehension of the natural progression of CC and the effectiveness of surgical interventions.

Methods

The medical records of patients undergoing CC surgery from 2002 to 2021 at a university hospital were retrospectively reviewed. The patients were divided into two groups: children (aged < 15 years) and adults (aged ≥ 15 years). The study gathered various data points, including patient age and gender, clinical presentation, pre-operative imaging, details of the surgical procedure, complications, and subsequent management, along with pathological results. The operation on the children's group was performed by pediatric surgeons (PA, CI, NS, KD, PR, SR, and PV), while the operation on the adult group was performed by adult general surgeons (KK, SP, S-UP, BN, and PS). A comparison was made between the data sets of children and adults. Categorical variables were presented as both whole numbers and percentages, and the comparison of these two independent groups was conducted using Chi-squared tests. The statistical analysis was performed using Stata 11 software for Windows, and statistical significance was determined based on a two-sided p-value of < 0.05. Data collection occurred after approval of the study protocol by the institutional review board No. 245/65.

Results

The study reviewed a total of 106 cases spanning a twenty-year period, from January 2002 to December 2021. Patients were evenly distributed between children and adults (53 in each group). Majority of patients were female in both cohorts (81.1% in children and 84.9% in adults out of 53 patients in each group). The mean (SD) age at the time of surgery for children and adults was 4.1 (3.9) and 34.3 (14.0) years, respectively. The average (SD) length of stay for children and adults was 15.7 (10.7) and 17.7 (11.5) days, respectively. The mean (SD) follow-up time for children and adults was 28.6 (35.3) and 38.1 (52.0) months, respectively.

Patient background and characteristics are detailed in Table 1. Abdominal pain emerged as the predominant presenting symptom, followed by jaundice in both groups. Remarkably, only 2 patients exhibited the classic triad of abdominal pain, jaundice, and a right upper quadrant mass. Adults were significantly more prone to present with abdominal pain compared to children (86.8% vs. 52.8%; $p < 0.001$), while children were more likely to experience acholic stool in contrast to adults (22.6% vs. 3.8%; $p = 0.004$).

Figure 1 provides a comprehensive overview of the investigation modalities employed in the study. Children were markedly more likely to have undergone ultrasonography compared to adults (96.2% vs. 54.7%). In contrast, adults were more to have undergone computed tomography (CT) (39.6% vs. 22.6%), magnetic resonance imaging (MRI) or magnetic resonance cholangiopancreatography (MRCP) (43.4% vs. 13.2%), and endoscopic retrograde cholangiopancreatography (ERCP) (22.6% vs. 13.2%). Some patients did

Table 1 comparison of the background and characteristics of children and adults undergoing choledochal cyst surgery

	Children (N = 53)	Adult (N = 53)	P-value
Initial clinical presentation			
Abdominal pain	28 (52.8%)	46 (86.8%)	< 0.001
Jaundice	23 (43.4%)	24 (45.3%)	0.845
Vomit	17 (32.1%)	13 (24.5%)	0.388
Fever	15 (28.3%)	13 (24.5%)	0.659
Acholic stool	12 (22.6%)	2 (3.8%)	0.004
Cholangitis	3 (5.7%)	7 (13.2%)	0.184
Anorexia	5 (9.4%)	4 (7.5%)	0.727
Pancreatitis	3 (5.7%)	4 (7.5%)	0.696
Distended abdomen	6 (11.3%)	1 (1.9%)	0.051
Diarrhea	4 (7.5%)	1 (1.9%)	0.169
Mass	2 (3.8%)	2 (3.8%)	1.000
Preoperative imaging findings			
Stone	11 (20.8%)	20 (37.7%)	0.055
PBM	N/A	7 (13.2%)	–

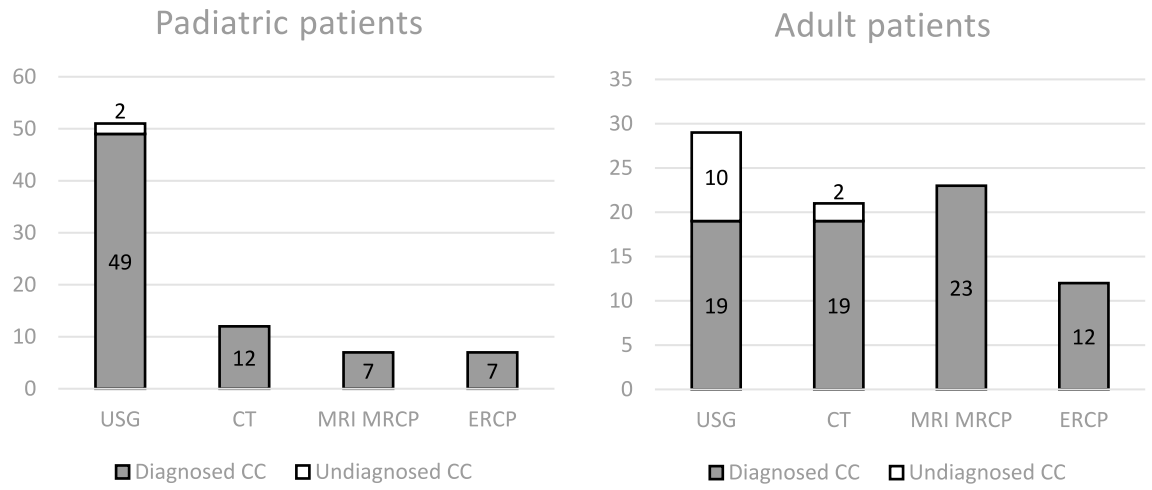

Fig. 1 Pre-operative imaging in a cohort of 106 patients, including both adult and pediatric populations

not receive a diagnosis through ultrasonography, with 2 out of 51 cases (3.9%) in children and 10 out of 29 cases (34.5%) in adults remaining undiagnosed through this modality. Additionally, pancreaticobiliary maljunction was exclusively identified in the adult group (13.2%), with 6 cases diagnosed through ERCP and 1 through MRCP.

Figure 2 outlines the CC subtypes according to the Todani's classification within this cohort. Most patients were presented with a type I CC (71.7%). Type I CCs were more prevalent in children compared to adults (79.3% vs. 66.0%), while type IV CCs were more predominant in the adult population (32.1% vs. 17.0%). Turning to the operations for the treatment of CC, as depicted in Fig. 3, a significant proportion of patients (87.7%) underwent a laparotomy approach for cyst excision with Roux-en-Y hepaticojejunostomy. Nine out of 106 patients underwent a laparoscopic-assisted approach for cyst excision with Roux-en-Y hepaticojejunostomy, comprising six children (11.3%) and three adults (5.7%). As shown in Table 2, adults demonstrated a higher likelihood of having a subcutaneous Roux limb created (22.6% vs. 3.8%; p = 0.004) and undergoing hepatectomy (11.3% vs. 0.0%; p = 0.012). The indications for hepatectomy included 5 cases of type IV CC, with the remaining one performed for the removal of a stone. Drain placement was a routine procedure in every case.

The post-operative complications are outlined in Table 3, revealing that adults had a higher incidence of stones, anastomosis or bile duct stricture, abdominal collection (not caused by anastomotic leakage), and cholangitis. Adults were significantly more likely to need intervention compared to children (26.4% vs. 5.7%; p = 0.004). Furthermore, abnormal histologic findings were exclusive to adult patients. These findings included 2 cases of adenocarcinoma, 1 case of cholangiocarcinoma, 1 case of focal adenomatous hyperplasia, 1 case of intestinal metaplasia, 1 case of high-grade

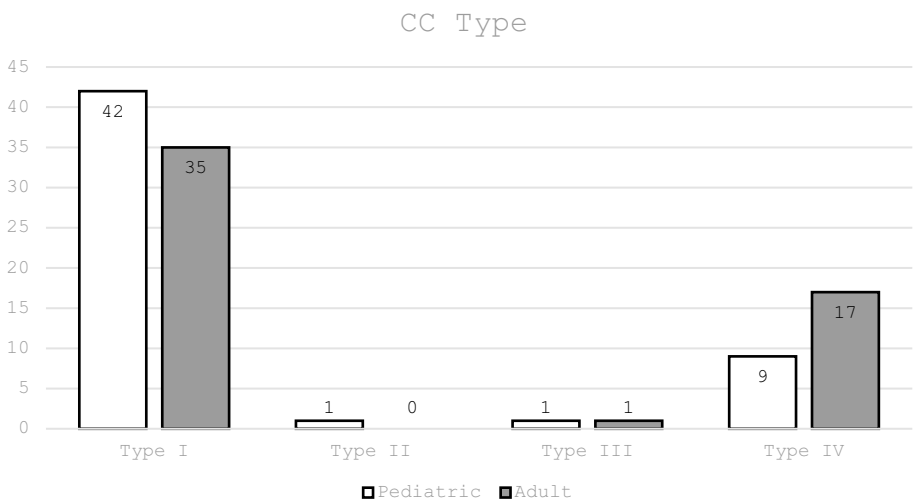

Fig. 2 the types of choledochal cysts in a cohort of 106 patients, including both adult and pediatric populations

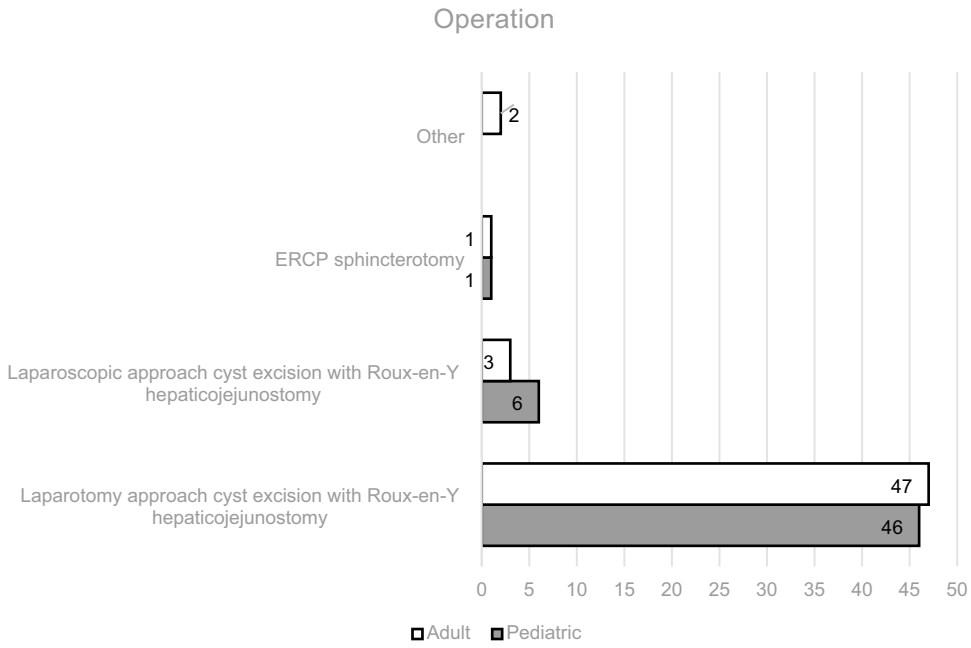

Fig. 3 the operations performed for choledochal cyst surgery in a cohort of 106 patients

Table 2 comparison of the operative results of children and adults undergoing choledochal cyst surgery

	Children (N = 53)	Adult (N = 53)	P-value
Operation			
Subcutaneous limb	2 (3.8%)	12 (22.6%)	0.004
Hepatectomy	0 (0.0%)	6 (11.3%)	0.012
Intra-operative complication			
Portal vein injury	1 (1.9%)	2 (3.8%)	0.558
Pathology			
Abnormal histological finding	0 (0.0%)	9 (17.0%)	0.002

Table 3 comparison of the post-operative complications of children and adults undergoing choledochal cyst surgery

	Children (N = 53)	Adult (N = 53)	P-value
Post-operative complications			
Stone	0 (0.0%)	13 (24.5%)	< 0.001
Anastomosis or bile duct stricture	1 (1.9%)	8 (15.1%)	0.015
Abdominal collection	1 (1.9%)	8 (15.1%)	0.015
Pancreatitis	1 (1.9%)	4 (7.5%)	0.169
Cholangitis	0 (0.0%)	4 (7.5%)	0.041
Bile leakage	1 (1.9%)	3 (5.7%)	0.308
Gut obstruction	2 (3.8%)	2 (3.8%)	1.000
Wound complication	0 (0.0%)	3 (5.7%)	0.079
Remnant	0 (0.0%)	1 (1.9%)	0.315
Complication management			
Intervention needed	3 (5.7%)	14 (26.4%)	0.004
Re-operation	4 (7.5%)	8 (15.1%)	0.22

dysplasia, 2 cases of adenocarcinoma of the gallbladder, and 1 case of adenocarcinoma of the ampulla.

Discussion

Choledochal cysts (CC) can be diagnosed in patients of all ages and carry a risk of malignancy as well as serious complications. Our study focused on demographic data, clinical presentation, investigations, treatment, and outcomes among both children and adults at a university hospital in Thailand, where CC is more predominant than in Western countries [2, 12].

From our study, we affirmed a female predominance in CC disease, with a male-to-female ratio of 1:4–5 observed in both children and adults [2, 3, 7–9, 12, 13]. Abdominal pain emerged as the most prevalent presenting symptom in both age groups. However, adults were more likely to present with abdominal pain than children, while children exhibited a higher propensity for presenting with acholic stool than adults. These clinical presentations were consistent with findings from other studies [7–9]. Only two patients exhibited the classic triad of CC. These findings align with other studies that have demonstrated the rarity of patients presenting with the classic triad [7, 9, 13]. This is likely due to the early concern and the use of screening ultrasound in the diagnosis of CC.

Pancreaticobiliary maljunction (PBM) is a congenital anomaly, usually forming a markedly long common channel. The cystic type of CC is almost always associated with PBM [12, 14]. PBM causes the reflux of pancreatic juice into the common bile duct, leading to chronic inflammation, damage to the bile duct wall, and cystic changes [2, 12]. However, in our study, we identified only 7 cases (13.2%) of adult patients with CC and PBM because ultrasound is unable to accurately identify PBM; the diagnosis of PBM primarily relies on ERCP, with some cases diagnosed through MRCP [1, 2, 14, 15]. According to Saito et al.'s study, ERCP demonstrates a higher visualization rate of PBM compared to MRCP (82% vs. 57%, respectively; $p = 0.006$) [16]. Consequently, PBM was exclusively found in adult patients. We acknowledge that the incidence of PBM in our study may be lower than the true incidence because ultrasound (US) is the primary imaging modality, especially in children [2, 12, 15, 17].

In the context of CC subtypes based on Todani's classification, we found that Type I CC is the most prevalent in both children and adults. Nevertheless, Type IV CC is more predominant in the adult group. These findings align with results from other studies. In the investigation by Soares et al., among 394 CC patients, 70.1% had Type I CC. Type I CCs were more frequently observed in children compared to adults (79.7% vs. 54.9%; $p = 0.003$), while Type IV CCs prevailed in the adult population (23.9% vs. 12.0%; $p = 0.006$) [7]. In the study by Senthilnathan et al., out of 110 CC patients, 71.8% had Type I CC. Type I CCs were more commonly observed in children compared to adults (74.6% vs. 69.1%), while Type IV CCs predominated in the adult population (30.9% vs. 25.5%) [9].

The most common procedure performed in our study is cyst excision with Roux-en-Y hepaticojejunostomy, which is the preferred method for biliary reconstruction at Juntendo [1]. However, extrahepatic cyst excision and Roux-en-Y hepaticojejunostomy are not satisfactory for many patients with complex type IV CC. In our study, six cases underwent hepatectomy, including five with type IV CC, and one performed for stone removal. Xia et al. reported results from 59 patients with type IV-A CC who underwent combined extrahepatic cyst excision, partial hepatectomy, and Roux-en-Y hepaticojejunostomy. Long-term biliary function was excellent in 33 (67.4%) patients, good in 9 (18.4%), fair in 5 (10.2%), and poor in 2 (4.1%) [10]. Additionally, Zheng et al. found that for adult patients with type IV-A CC, the morbidity of biliary stricture and/or lithiasis and the reoperation rate in the extrahepatic cystectomy group were significantly higher than those in the liver resection group (34.3% vs. 10.3%; $p = 0.037$) [8]. However, studies by Friedmacher et al. demonstrated that over 40 children with type IV CC who were regularly followed up by ultrasonography showed a considerable reduction in intrahepatic duct dilatation, typically within the first year after surgery [6]. These findings correlated with the study by Kronfli and Davenport, suggesting that type IV CC represents a progression from originally type I CC [18]. Thus, partial hepatectomy may be effective for treating complex type IV-A CC in adults with substantial intrahepatic bile duct involvement and hilar bile duct stenosis [8], while conservative surgery may be more appropriate in children [18]. In addition, some adult patients in our study underwent subcutaneous Roux limb creation. This technique is also reported in the study by Htut Saing et al., where it was used for stone extraction in patients after the operation for CC who had hepatolithiasis [19].

Over the past decade, there has been a noticeable shift towards utilizing laparoscopic techniques in the management of CC. Pediatric surgeons from Asian countries such as Japan, China, and Vietnam, are often considered to have extensive experience in minimally invasive CC surgery [6]. A retrospective review of 93 laparoscopic CC excisions conducted at Juntendo and Monterrey hospitals between 2009 and 2022 revealed no intraoperative complications [1]. Selthilnathan et al. explored the outcomes of laparoscopic CC management in both pediatric and adult populations, noting significantly reduced intraoperative blood loss, shorter operative times, and shorter hospital stays in pediatric patients compared to adults. Additionally, three patients (2.73%), all adults, required conversion to laparotomy [9].

In our study, laparoscopic techniques were employed in both pediatric and adult patients, albeit infrequently. However, due to our ongoing learning curve and the limited dataset, we refrained from conducting a comparative analysis between laparotomy and laparoscopic approaches. We think that laparoscopic surgery for CC will gain more and more popularity.

According to a meta-analysis study, the incidence of postoperative anastomotic stricture was found to be 2.1%. with a higher occurrence in type IV-A (10.1%) compared to type I (2.0%) ($p=0.001$) [20]. In our study, we observed that adults with CC faced more serious complications than children. Among the post-operative complications occurring more frequently in adults were stones, anastomosis strictures, abdominal collections, and cholangitis. This could be attributed to long-standing cystic inflammation. Furthermore, cholangitis and stone formation have been reported to be associated with dilatation of the peripheral portion of the intrahepatic bile duct [1]. These findings align with the study conducted by Zheng et al., which demonstrated a significantly higher morbidity of biliary stricture and/or lithiasis in adults compared to children (23.4% vs. 5.9%; $p=0.041$). Specifically, in the extrahepatic cystectomy group, the reoperation rate for adults was notably higher than that for children (40.0% vs. 6.3%; $p=0.019$) [8]. Additionally, the study by Soares et al. revealed that children were more likely to experience anastomosis leakage (3% vs. 0%; $p=0.01$), whereas adults were more prone to perihepatic abscess (7.7% vs. 0%; $p<0.001$) and fistula (2.7% vs. 0%; $p=0.05$) [7].

The data from the Japanese Study Group on Pancreatico-Biliary Maljunction (JSGPM) registry demonstrate an increased risk for biliary tract cancer in CC cases with age [1]. Adults with CC face a malignancy risk ranging from 6 to 30%, while in children, it is sporadically identified. The predominant histologic subtype is adenocarcinoma (73% to 84%). After the operation, there is a lifelong elevated risk of up to around 4% for cancer development. Consequently, post-operative follow-up, including annual checks of CA 19–9 and abdominal ultrasound, is recommended [21–23]. Okazaki et al. conducted a comprehensive review on premalignant/malignant histology (PMMH) in excised CC specimens from children. Among the 20 reports analyzed, 4 cases were identified as adenocarcinoma, 4 as sarcoma, and 12 as dysplasia [24]. Our study aligns with these findings, revealing that abnormal histologic findings were exclusive to adult patients. Adenocarcinoma is the most common subtype, occurring in various locations such as extrahepatic bile ducts, gallbladder, and ampulla.

This study has, however, some limitations. Firstly, it is retrospective in nature, which has resulted in some bias and missing data. Secondly, the sample size is relatively small. To enhance accuracy in future research endeavors, it may be necessary to adopt a prospective design that encompasses multiple institutional centers. Finally, the 20-year retrospective study carries the burden of inevitable significant changes in clinical practice during the studied period.

Conclusions

Abdominal pain is the primary presentation of choledochal cysts and is more prevalent in adults. Type I choledochal cyst is the most common type and is more frequently found in children. Ultrasound is the most used screening tool for diagnosis. Post-operatively, adults with choledochal cysts experience more severe complications compared to children, possibly due to long-standing cystic inflammation. Therefore, prompt definitive surgery is recommended for choledochal cyst patients.

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Author contributions P.A., C.I., and P.V. wrote the main manuscript. P.A. prepared figures. All authors reviewed the manuscript.

Data availability No datasets were generated or analysed during the current study.

Declarations

Conflict of interests We would like to declare that the abstract entitled above has been previously presented as an oral presentation at the 48th Annual Scientific Congress of the Royal College of Surgeons of Thailand, held from 27 to 29th July 2023 at PEACH, Royal Cliff Hotels Group, Pattaya, Thailand. The meeting is an annual national surgical meeting. Only the abstract (not full manuscript) will be published soon in the Thai Journal of Surgery in 2024. However, the full manuscript of the abstract has never been previously considered or published in any other journal.

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