

Research Article

Congenital Cystic Dilatations of the Bile Ducts in Adults

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Abstract

Introduction: Cystic dilatation of the bile ducts, or choledochal cyst, is a rare congenital disorder of the biliary tree, usually discovered in childhood. It is uncommon and only presents in adulthood in 20% of cases. **Material and methods:** Over a 23-year period from 2003 to 2026, we operated on 12 consecutive patients with congenital cystic dilatation of the bile ducts. There were 2 men and 10 women with a mean age of 49.1 years (range: 21–78 years). The cystic dilatation of the bile ducts was classified according to the Todani classification as type I in 9 cases, type II in 1 case, type IVa in 1 case, and type V in 1 case. Complete resection of the cyst was performed for types I, II, and IV. In one case, an adenocarcinoma of the common bile duct cyst was discovered upon examination of the surgical specimen. Drug treatment was prescribed for type V. **Results:** All our patients were symptomatic. The isolated or combined clinical signs leading to the discovery of the lesion were abdominal pain in all patients, jaundice and fever in 75% (9 cases) and 41.7% (5 cases), respectively. Postoperative complications were marked by episodes of acute cholangitis, which were more or less successfully controlled by percutaneous drainage combined with antibiotic therapy. **Conclusion:** The treatment of congenital cystic dilatations of the bile ducts must follow a strategy adapted to each type of malformation. Cystoenteric anastomosis is contraindicated, and resection should be as complete as possible.

Keywords

Congenital Cystic Dilatations, Bile Ducts, Surgery, Excision

1. Introduction

Cystic dilatations of the bile ducts, or choledochal cysts, are congenital lesions of the bile ducts generally discovered in childhood and only manifest in adulthood in 20% of cases. They are characterized by one or more communicating saccular or fusiform dilatations of the intra- and/or extrahepatic bile ducts. They are the second most common congenital malformation of the bile ducts after biliary atresia. Todani [1-3] grouped them into five types based on the location, extent, and type of bile duct dilatation. Their incidence varies from

1/100,000 to 1/200,000 births and is seen primarily in Asian countries, particularly Japan, but no etiological factor explaining this difference has yet been identified [4]. There is a clear female predominance with a sex ratio of 4 women to 1 man [4-8].

The anomaly of the biliopancreatic junction plays a crucial role in the development of this malformation and in the secondary degeneration of the biliary wall. Surgical resection is the standard treatment. The aim of this retrospective study is

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Received: 14 September 2026; **Accepted:** 23 September 2026; **Published:** 23 October 2026



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to analyze the results of our series and to review the literature in order to propose a therapeutic approach for this condition.

2. Material and Methods

This was a retrospective, single-center study of 12 cases of cystic dilatation of the bile ducts. These patients were collected from the visceral and digestive surgery department of the Tizi Ouzou University Hospital Center, Algeria, over a 23-year period from January 2003 to April 2026. There were 2 men and 10 women. The mean age was 49.1 years (range: 21–78 years).

The type of dilatation, associated complications, and the presence of a biliopancreatic junction anomaly (BPJA) were determined using imaging techniques: ultrasound, computed tomography (CT), magnetic resonance imaging (MRI), endoscopic retrograde cholangiopancreatography (ERCP), and intraoperative cholangiography. Two classifications were used: the Todani classification (Figure 1) for bile duct dilation, and the Kimura classification (Figure 2) [9] and the Komi classification (Figure 3) [10, 11] for biliopancreatic junction abnormalities.

Treatment methods and short- and long-term outcomes were recorded.

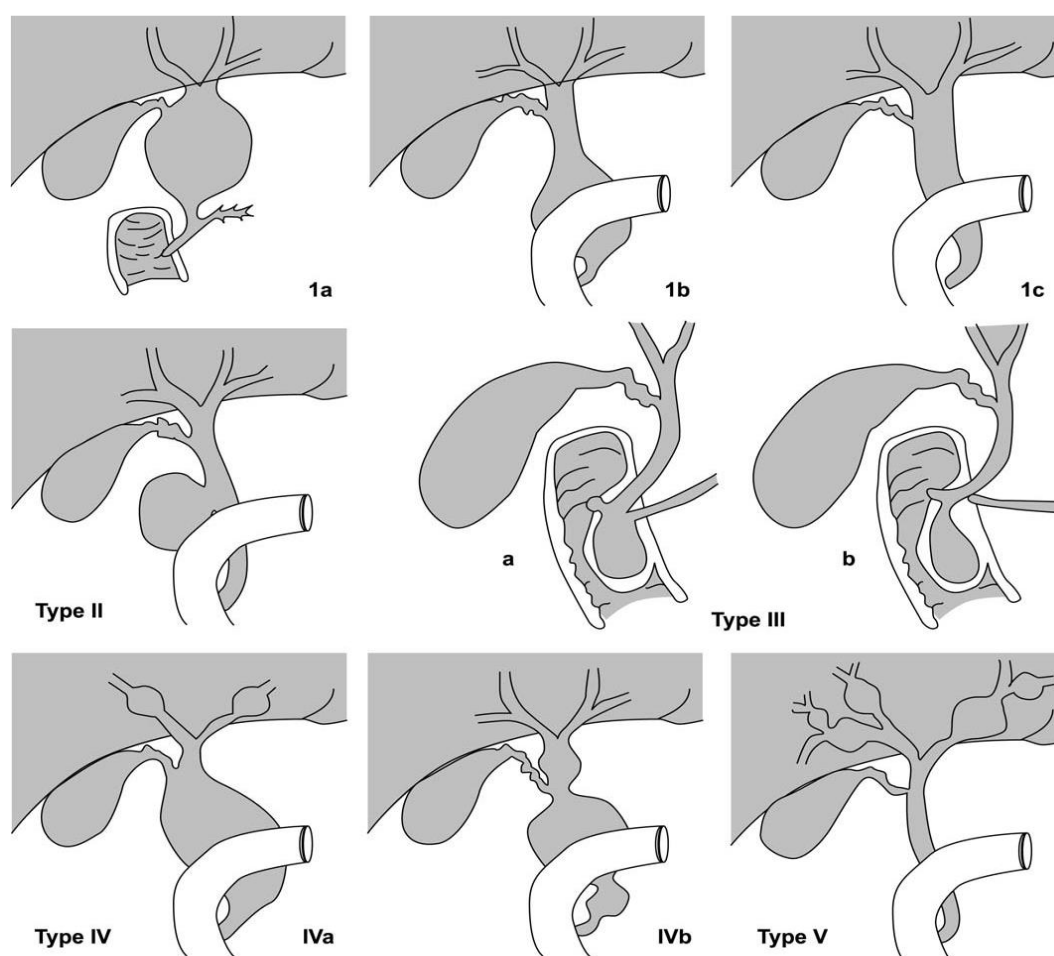


Figure 1. Todani Classification.

Type I (80%): Cystic dilatation of the common bile duct.

Type II (10%): supraduodenal diverticulum of the common bile duct.

Type III (4%): Choledochocoele.

Type IV (11%).

IVa: dilation of the intra/extrahepatic bile ducts.

IVb: multisegmental dilatation of the main bile duct.

Type V (< 1%): Isolated dilatation of the intrahepatic bile ducts.

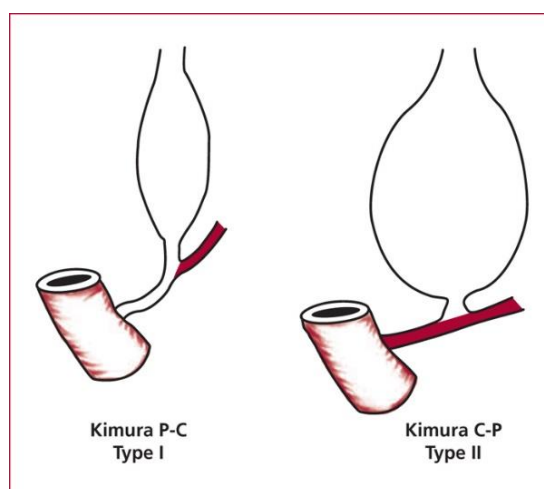


Figure 2. Kimura's classification of the anomaly of the biliopancreatic junction.

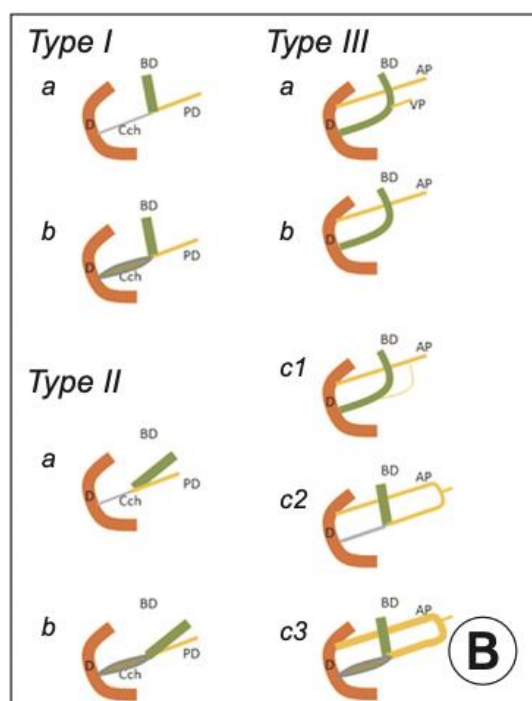


Figure 3. Komi's Classification of anomalies of the biliopancreatic junction (BD, bile duct; PD, pancreatic duct, Cch, common channel; D, duodenum; AP, accessory pancreatic duct; VP, ventral pancreatic duct.).

3. Results

All our patients were symptomatic. The isolated or combined clinical signs leading to the discovery of the bile duct malformation were abdominal pain in all patients, jaundice in 9 patients (75%), and fever in 5 other patients (41.7%).

Of our 12 patients, 9 had a type I malformation according to the Todani classification, associated in 4 cases with an anomaly of the biliopancreatic junction; 1 had a type II saccular dilatation; 1 had a type IVa malformation; and 1 had diffuse cystic dilatation of the intra- and extrahepatic bile ducts,

type V.

Five patients (41.7%) had autochthonous lithiasis, located within the common bile duct cyst in two cases, in the non-cystic common bile duct in two other cases, and in the intrahepatic bile ducts in one case.

A cholecystitis with calculi was observed in one patient with a type I malformation. Stage C necrotizing pancreatitis on CT scan was associated with a type I malformation in one patient. Malignant transformation was observed in only one patient, unsuspected during surgery but revealed after systematic histological examination of the surgical specimen from the resection of a type I common bile duct cyst. This was a well-differentiated adenocarcinoma in situ.

The anatomy of the biliopancreatic junction was specified in seven cases. An anomaly of the biliopancreatic junction was observed in four patients with a type I biliary tract malformation. This anomaly was type I according to Kimura in one case and type II in three cases. The diagnosis was made in two cases by MRI, in one case after intraoperative transcystic cholangiography, and in one case after ERCP.

Treatment of type I cystic malformations of the bile ducts (Figure 4) consisted of total cyst resection, cholecystectomy, and a Roux-en-Y hepaticojejunostomy in eight patients. The ninth patient, a 77-year-old woman whose cyst was discovered during an episode of severe necrotic pancreatitis, did not undergo secondary resection because she was lost to follow-up. There was no perioperative mortality.

Three external biliary fistulas were also observed and treated with external drainage. A 65-year-old woman on anticoagulants, who underwent surgery for a malignant cyst, died two years after her procedure from a myocardial infarction,

with no signs of neoplastic recurrence.

The patient with the type II lesion (Figure 5) also underwent resection of the common bile duct, including the choledocele, combined with cholecystectomy and a Roux-en-Y hepaticojejunostomy. The postoperative course was uneventful.

The patient with a type IVa cystic dilatation of the bile ducts underwent total resection of the common bile duct with a Roux-en-Y hepaticojejunostomy, separate from the two hepatic cannons. No liver resection was performed. The postoperative course was complicated by the development of a well-positioned external biliary fistula. The median follow-up duration was 3 years (range: 2–11 years).

The patient, class V (diffuse Caroli disease), had only received treatment for lithiasis of the common bile duct (choledochotomy, extraction of stones and placement of a Kehr drain) followed by drug treatment such as urodeoxycholic acid for intrahepatic lithiasis.

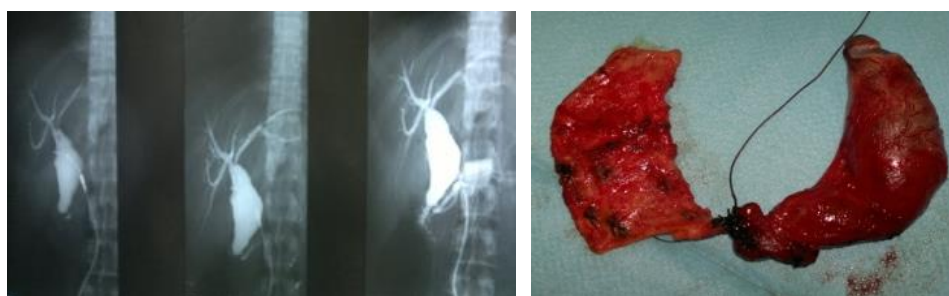


Figure 4. Intraoperative appearance of Todani type I (intraoperative cholangiography, surgical resection).

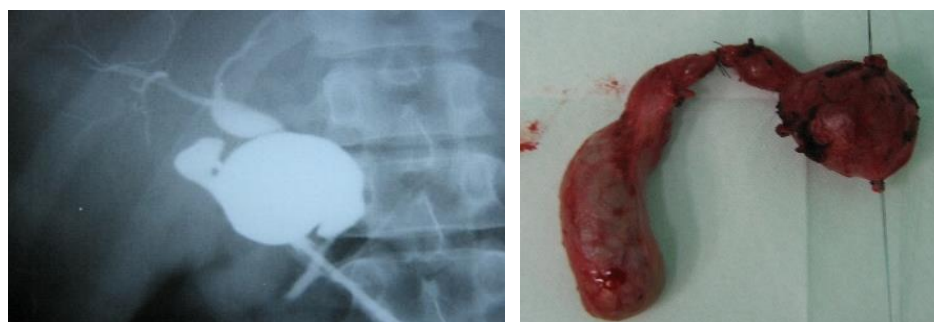


Figure 5. Intraoperative appearance of Todani type II (intraoperative cholangiography, surgical resection).

4. Discussion

Cystic dilatations of the bile ducts are congenital malformations characterized by one or more communicating cystic dilatations of the intra- and/or extrahepatic bile ducts. They are uncommon and usually manifest before the age of 10 [9]. This condition is rare in Western countries, with an incidence of 1/100,000 to 1/175,000 births. It is 100 times more frequent in Southeast Asia and Japan [9, 12]. In the twenty years from 2003 to 2026, we recorded only 12 cases. It is the second most

common congenital malformation of the bile ducts after biliary atresia [9]. Recent improvements in imaging techniques have led to a resurgence in cases of cystic dilatations of the bile ducts discovered in adulthood, the frequency of which was probably underestimated due to more subtle symptoms than in children [13]. Currently, 20% of these malformations are revealed in adulthood [9].

The first description of ductal dilatations of the biliary tree dates back to 1723 by Vater. It was only in 1959 that Alonso-Lej [1] proposed the first classification of these anomalies. Currently, the most widely used classification is that of Todani,

proposed in 1977 [14, 15], which comprises 5 types. Type I, representing approximately three-quarters of cases, is congenital dilatation of the common bile duct. Type II, which accounts for three-quarters of cases, corresponds to a diverticulum of the common bile duct; type III to a choledochocoele; and type IV to multiple intra- and extrahepatic cysts. Finally, type V corresponds to Caroli's disease.

The pathophysiology of congenital bile duct dilatations remains poorly understood. Numerous theories have been proposed to explain the origin of these malformations [16-18]. The most widely accepted is that put forward by Babbitt in 1969, which implicated an anomaly of the biliopancreatic junction (BPJ) [19]. This anomaly of the biliopancreatic junction (BPJ) is found in more than 90% of patients with cystic dilatation of the common bile duct. BPJ results from an embryological anomaly of fusion of the primitive common bile duct with the ducts of the ventral and dorsal pancreas. According to Japanese guidelines, BPJ is defined by the presence of a common biliopancreatic duct of abnormal length (> 10 mm) associated with a biliopancreatic junction located outside the duodenal wall [20]. Acute biliary edema (ABPE) is responsible for the reflux of pancreatic enzymes into the common bile duct, leading to increased intrabiliary pressure, weakening of the biliary epithelium, and then dilation of the common bile duct. An abnormal level of pancreatic enzymes in the bile ducts, measured by biliary catheterization, can also lead to the diagnosis of ABPE if imaging is inconclusive. Abnormalities of the sphincter of Oddi or of the autonomic innervation of the common bile duct (hypoganglionosis), and more generally any factor responsible for obstruction of the common bile duct, could also explain the development of the common bile duct cyst [21-23]. In this case, it would be a secondary lesion developing on an already complete bile duct, sometimes before birth, rather than a developmental defect of a malformative type. Three types of pancreatic junction obstruction (PJO) are distinguished according to the type and angle of the junction between the bile and pancreatic ducts (Komi classification, Figure 3). In Japanese series, cystic malformations of the bile ducts are associated with PJO in almost all cases [10-14, 24]. In our series, the biliopancreatic junction anomaly was found in four patients.

Choledochal cysts are diagnosed in childhood in more than 80% of cases because the symptoms are often pronounced, in the form of jaundice associated with abdominal pain and hepatomegaly [25, 26]. In our series, this triad was never found complete. However, there are forms diagnosed later in life, in adulthood, where the clinical presentation may be less specific and limited to right upper quadrant pain [25, 27], as was the case for all our patients. Common bile duct cysts can also be diagnosed following a biliary lithiasis complication such as cholecystitis or cholangitis, following a liver abscess, or even at the initial stage of secondary biliary cirrhosis with portal hypertension [25, 27]. Biliary peritonitis due to cyst rupture is a serious but rare complication ($<2\%$), which can classically

occur during pregnancy [25, 28, 29]. There is also a high prevalence of acute pancreatitis (70%) in this population, the origin of which may be gallstones or related to bile reflux into the main pancreatic duct [27].

The clinical presentation of common bile duct cysts can differ depending on their morphology. Type II cysts are most often asymptomatic, while type III cysts more frequently present with an episode of acute pancreatitis or with a high intestinal obstruction due to duodenal lumen blockage. Type IVa and V cysts appear to be most associated with biliary lithiasis complications.

The development of bile duct cancers (cholangiocarcinoma and adenocarcinoma of the gallbladder) is the most serious complication of common bile duct cysts. The overall incidence of bile duct cancers in this population is estimated at 10.7% [30]. However, the incidence varies with age: it is very low in childhood ($<1\%$) and increases progressively with age, reaching 40% after age 60 [31]. The median age at diagnosis is 49 years. Chronic inflammation of the biliary epithelium resulting from the reflux of pancreatic enzymes into the bile ducts appears to be the main etiological factor explaining the high incidence of cancer in these patients. Type I and IV cysts are at the highest risk of malignant transformation and account for 90% of cancer cases [30]. In our series, we have a single case of a choledochal cyst that degenerated into a well-differentiated adenocarcinoma. The diagnosis was made based on systematic histological examination of the surgical specimen. It was a type I Todani choledochal cyst. In cases of acute biliary stenosis without an associated choledochal cyst, the risk of cancer is concentrated in the gallbladder, with an overall incidence exceeding 40% [20]. In approximately one-third of cases, the cancer is located within the cyst walls themselves, in the form of an adenocarcinoma. The other histopathological forms, in order of frequency, are cholangiocarcinomas located outside the cyst (43%), gallbladder carcinomas (9%), anaplastic carcinomas (7%), pancreatic adenocarcinomas (3%), followed by rare biliary sarcomas [32], squamous cell carcinomas, and adenoacanthomas [33].

Magnetic resonance cholangiopancreatography (MRCP) is the gold standard for diagnosing choledochal cysts. It allows confirmation of the diagnosis by visualizing a communication between the cystic dilation and the bile ducts and by ruling out the main differential diagnoses, particularly non-communicating extrabiliary cysts. Liver MRI also allows for the assessment of the cyst's impact on the liver parenchyma (liver dysmorphism, abscess) and the screening for complications (cancers, portal hypertension). Visualization of a biliary-patellar-containing adenoma (BPCA) on MRI is essential to distinguish a common bile duct cyst from an enlarged common bile duct in elderly patients or secondary to cholecystectomy. If there is doubt about the presence of a BPCA, which can be difficult to visualize on MR cholangiopancreatography (MRCP), endoscopic ultrasound of the biliary-pancreatic duct (EUS-CP) generally confirms the diagnosis. Given the significant risk of cholangitis, diagnostic ERCP should be reserved

for suspected cholangiocarcinoma in order to obtain histological samples. No imaging examination can confirm early malignant transformation of a cystic dilatation of the common bile duct [34]. The only malignant case in our study was discovered during routine histological examination of the surgical specimen.

The treatment of choice for common bile duct cysts is surgical excision, which prevents degeneration of the cyst wall and bile ducts. Cholecystectomy should be performed systematically in cases of associated biliopancreatic junction abnormalities, given the high risk of gallbladder epithelial degeneration [9]. Cystodigestive bypasses are contraindicated [35-37]. The management of common bile duct cysts differs depending on the cyst type.

For types I and IV: Total resection of the cyst (and therefore of the common bile duct) with Roux-en-Y hepaticojejunostomy is the treatment recommended by the French Association for the Study of the Liver (AFEF) [38]. Cholecystectomy should also be systematically performed at the same time. Given the morbidity associated with pancreatic surgery, resection of the intrapancreatic common bile duct by pancreaticoduodenectomy (PD) is not recommended in the absence of signs of cholangiocarcinoma of the lower common bile duct. Similarly, for type IVa cysts with unilateral involvement of the intrahepatic bile ducts, partial hepatectomy is not routinely performed but may be considered in cases of intrahepatic lithiasis complicated by recurrent cholangitis (with stones that cannot be extracted endoscopically) or in cases of intrahepatic cholangiocarcinoma. Short-term complications of surgery can be pancreatic (fistulas, acute pancreatitis) or biliary (anastomotic fistulas, biliary peritonitis) [39]. Long-term complications of surgical resection, occurring in approximately 15% of cases, are primarily biliary anastomosis strictures and lithiasis, particularly in type IV cysts [40]. In rare cases (<10%), surgical revision may be necessary to repair the biliary-enteric anastomosis or remove residual cystic dilatations [40]. Although surgical resection significantly reduces the risk of biliary epithelial degeneration, a risk of cancer remains. In a retrospective Japanese series of 94 patients, 4 developed cholangiocarcinoma some time after surgery, with a cumulative incidence of 11% at 25 years [41]. In this series, all cancers occurred in unresected biliary segments (intrahepatic, hilar, intrapancreatic). Therefore, excision of common bile duct cysts should be discussed as soon as the diagnosis is made to avoid complications related to chronic inflammation, particularly neoplastic ones.

Laparoscopic resection of this type of dilation has been described [42].

For type II and III cysts, given the low risk of cholangiocarcinoma, complete resection of the common bile duct is not necessary.

In type II, excision of the entire diverticulum combined with cholecystectomy and hepaticojejunostomy is the preferred procedure [27].

For type III cysts, endoscopic sphincterotomy is often recommended to prevent the risk of obstructive bile duct stones [27]. In cases of associated acute biliary stenosis, preventive cholecystectomy remains justified due to the high risk of gallbladder cancer. In cases of incidental discovery of an asymptomatic choledochocoele, simple monitoring, combining retrograde cholangiopancreatography with biopsies and biliary and pancreatic blood tests, is proposed by some authors [43, 44].

Histological analysis of the intracystic epithelium could play an important role in therapeutic decision-making. The epithelium can be of three types: duodenal, biliary, or undifferentiated [44, 47]. Biliary-type epithelium is thought to be more common in adults [48]. Several hypotheses have been proposed to explain the presence of duodenal epithelium at the terminal portion of the common bile duct. Following an anomaly in the migration of the common bile duct and pancreatic junction or a failure of involution of the ampulla of Vater before the 8th week of gestation, the common bile duct and pancreatic junction would remain outside the duodenal lumen, which could be covered either by biliary epithelium or by migration of the duodenal epithelium [48]. Another hypothesis describes choledochocoele as a congenital lesion of the duodenal duplication type, communicating with the duodenum via a fistula and receiving the papilla [49]. The duodenal epithelium is thought to be less prone to degeneration than the biliary epithelium usually present in choledochocoeles [9], thus explaining the lower risk of degeneration in choledochocoeles. In cases of duodenal lining, a simple bypass would suffice, whereas in cases of biliary or undifferentiated epithelium, complete resection of the cyst with reimplantation into the common bile duct and pancreatic duct may be necessary [48]. Overall, the conservative approach usually recommended should probably be adjusted according to the type of intracystic epithelium and does not preclude long-term monitoring to avoid overlooking degeneration [21].

The treatment for type IVB is the same as for type I.

In type V, resection of the common bile duct is not necessary. Localized Caroli disease type V does not pose a therapeutic problem. Hepatic resection allows for a definitive cure.

The problem arises when bile duct involvement is diffuse. Transplantation is the intervention of choice [45, 46]. It is discussed based on the patient's age, the duration of the disease, the frequency of cholangitis, and the presence of associated portal hypertension.

There are no recommendations detailing the monitoring of surgically removed common bile duct cysts. Nevertheless, given the risk of postoperative complications, particularly gallstones, and the risk of residual cancer, annual monitoring by magnetic resonance cholangiopancreatography (MRCP) should be maintained.

5. Conclusion

Congenital cystic dilatations of the bile ducts in adults are

a rare malformation. Diagnosis is greatly facilitated by modern imaging techniques, primarily cholangiopancreatography - MRI. Treatment should follow a strategy tailored to each type of malformation, with the most complete possible resection of the dilated portion followed by a Roux-en-Y biliary anastomosis being the surgical procedure of choice. This should be offered as soon as the diagnosis is established, before the onset of complications. Prolonged radiological and laboratory monitoring is necessary to watch for the development of stenosis of the biliary anastomosis or progression to liver cirrhosis.

Abbreviations

CT	Computed Tomography
MRI	Magnetic Resonance Imaging
ERCP	Endoscopic Retrograde Cholangiopancreatography
MRCP	Magnetic Resonance Cholangiopancreatography
EUS-CP	Endoscopic Ultrasound of the Biliary-Pancreatic Duct
BPJA	BilioPancreatic Junction Anomaly
BPJ	BilioPancreatic Junction
ABPE	Acute Biliary Edema
PJO	Pancreatic Junction Obstruction
BPCA	Biliary-Patellar-Containing Adenoma
AFEF	French Association for the Study of the Liver

Author Contributions

Habarek Meziane: Conceptualization, Data curation, Formal Analysis, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing

Dali Katia: Data curation, Formal Analysis, Writing – review & editing

Bentabet Amar: Data curation, Formal Analysis, Software, Supervision, Writing – review & editing

Merzouki Sonia: Investigation, Software, Supervision, Writing – review & editing

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Ladjouzi Fares: Methodology, Software, Writing – review & editing

Ait Gharbi Saliha: Visualization, Writing – review & editing

Saad Saliha: Visualization, Writing – review & editing

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Conflicts of Interest

The authors declare no conflicts of interest.

References

- [1] Alonso-Lej F, Rever Jr WB, De Pessagno DJ. “Congenital choledochal cyst with a report of analysis of 94 cases. Surg Gynecol Obstet Int 1959, 108: 1-30.
- [2] Babbitt D, Starshak R, Clemett A. “Choledochal cysts: a concept of etiology.” Am J Roentgenol Radium Ther Nucl Med 1973, 119: 57-62.
- [3] Baison G, Bonds M, Helton W, Kozarek R. “Choledochal cysts: similarities and differences between Asian and Western countries.” World J Gastroenterol 2019, 25: 3334-43.
- [4] Soares KC, Kim Y, Spolverato G, Maithel S, Bauer TW, Marques H, *et al.* Presentation and Clinical Outcomes of Choledochal Cysts in Children and Adults: A Multi-institutional Analysis. JAMA Surg. 2015; 150(6): 577-84.
- [5] Dhupar R, Gulack B, Geller DA, Marsh JW, Gamblin TC. The changing presentation of choledochal cyst disease: an incidental diagnosis. HPB Surgery 2009 [Article ID 103739, 4 pages].
- [6] Stringer MD, Dhawan A, Davenport M, Mieli-Vergani G, Mowat AP, Howard ER. Choledochal cysts: lessons from a 20-year experience. Arch Dis Child 1995; 73: 528-31.
- [7] Liu-Binshi, Shu-You Peng, Xing-Kai Meng *et al.*: diagnosis and treatment of congenital choledochal cyst: 20 years' experience in China. World J Gastroenterol 2001; 7(5): 732-734.
- [8] Erlinger S. dilatations kystiques des voies biliaires. Revue du praticien 2000; 50: 2136 – 2141.
- [9] Kianmanesh R, Régimbeau JM, Belghiti J. Anomalies de la jonction biliopancréatique et dilatations kystiques congénitales des voies biliaires. J Chir (Paris) 2001; 138–4: 196–204.
- [10] Komi N, Tamura T, Miyoshi Y, Kunitomo K, Udaka H, Takehara H. Nationwide survey of cases of choledochal cyst. Analysis of coexistent anomalies, complications and surgical treatment in 645 cases. Surg Gastroenterol 1984; 3: 69-73.
- [11] Komi N, Takehara H, Kunitomo K, Miyoshi Y, Yagi T. Does the type of anomalous arrangement of pancreaticobiliary ducts influence the surgery and prognosis of choledochal cyst? J Pediatr Surg 1992; 27: 728-731.
- [12] Weyant MJ, Maluccio MA, Bertagnolli MM, Daly JM. Choledochal cysts in adults: a report of two cases and review of the literature. Am J Gastroenterol 1998; 93: 2580-3.
- [13] Lipsett PA, Pitt HA, Colombani PM, Boitnott JK, Cameron JL. Choledochal cyst disease. A changing pattern of presentation. Ann Surg 1994; 220: 644-52.
- [14] Todani T, Wanatabe 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: 263–9.

- [15] Mercadier M, Chigot JP, Clot JP, Langlois P, Lansiaux P. Caroli disease. *World J Surg* 1984; 8: 22–9.
- [16] Nagata E, Sakai K, Kinoshita H, Hirohashi K. Choledochal cyst: complications of anomalous connection between the choledochus and pancreatic duct and carcinoma of the biliary tract. *World J Surg* 1986; 10: 102–10.
- [17] Lacheretz M, Debeugry YP, Bonneville M. Étiologie de la dilatation congénitale du cholédoque chez l'enfant. *Chirurgie* 1986; 114: 111–4.
- [18] Desmet VJ. Congenital diseases of intra hepatic bile duct: variation on the term “ductal plate malformation”. *Hepatology* 1992; 16: 1069–83.
- [19] Babbitt DP. Congenital choledochal cysts: new etiological concepts based on anomalous relationship of the common bile duct and pancreatic bulb. *Ann Radiol* 1969; 12: 231–40.
- [20] Kamisawa T, Ando H, Suyama M, Shimada M, Morine Y, Shimada H, *et al.* Japanese clinical practice guidelines for pancreaticobiliary maljunction. *J Gastroenterol.* 2012; 47(7): 731–59.
- [21] Metcalfe MS, Wemyss-Holden SA, Maddern GJ. Management dilemmas with choledochal cysts. *Arch Surg* 2003; 138: 333–9.
- [22] Kimura K, Ohto M, Ono T, *et al.* Congenital cystic dilatation of the common bile duct: relationship to anomalous pancreaticobiliary ductal union. *Am J Roentgenol* 1977; 128: 571–7.
- [23] Miyano T, Yamataka A, Li L. Congenital biliary dilatation. In: *Process Citation. Sem Pediatr Surg* 2000; 9: 187–95.
- [24] Tanaka K, Nishimura A, Ishibe R, *et al.* Clinical abnormalities in adults with pancreaticobiliary maljunction with and without dilatation. *Can J Surg* 1995; 38: 351–4.
- [25] Yamaguchi M. Congenital choledochal cyst: analysis on 1433 patients in the Japanese literature. *Am J Surg* 1980; 140: 653–7.
- [26] Chijiwa K, Kimura H, Tanaka M. Malignant potential of the gallbladder in patients with anomalous pancreaticobiliary ductal junction. The difference in risk between patients with and without choledochal cyst. *Int Surg* 1995; 80: 61–4.
- [27] Brown ZJ, Baghdadi A, Kamel I, Labiner HE, Hewitt DB, Pawlik TM. Diagnosis and management of choledochal cysts. *HPB.* 2023; 25(1): 14–25.
- [28] Cazares J, Koga H, Yamataka A. Choledochal cyst. *Pediatr Surg Int.* 2023; 39(1): 209.
- [29] Wu DQ, Zheng LX, Wang QS, Tan WH, Hu SJ, Li PL. Choledochal cysts in pregnancy: case management and literature review. *World J Gastroenterol.* 2004; 10(20): 3065–9.
- [30] Ten Hove A, de Meijer VE, Hulscher JBF, de Kleine RHJ. Meta-analysis of risk of developing malignancy in congenital choledochal malformation. *Br J Surg.* 2018; 105(5): 482–90.
- [31] Sastry AV, Abbadessa B, Wayne MG, Steele JG, Cooperman AM. What is the incidence of biliary carcinoma in choledochal cysts, when do they develop, and how should it affect management? *World J Surg.* 2015; 39(2): 487–92.
- [32] Nonomura A, Mizukami Y, Matsubara F, Ueda H. A case of choledochal cyst associated with adenocarcinoma exhibiting sarcomatous features. *J Gastroenterol* 1994; 29: 669–75.
- [33] Fieber SS, Nance FC. Choledochal cyst and neoplasm: a comprehensive review of 106 cases and presentation of two original cases. *Am Surg* 1997; 63: 982–98.
- [34] Nicolet, J., Moutardier, V., & Belghiti, J. (1996). Carcinomatous degeneration of a choledochal cyst. *Gastroenterol Clin Biol*, 20(3), 324–326.
- [35] Todani T, Watanabe Y, Urushihara N, *et al.* Choledochal cyst, pancreaticobiliary maljunction, and cancer. *J Hepatobiliary Pancreatol Surg* 1994; 1: 247–51.
- [36] Chijiwa K, Koga A. Surgical management and long-term follow-up of patients with choledochal cysts. *Am J Surg* 1993; 165: 238–42.
- [37] Nagorney DM, MacLlath DC, Adson MA. Choledochal cysts in adults: management. *Surgery* 1984; 96: 656–63.
- [38] Neuzillet, C., *et al.* (2024). Management of intrahepatic and perihilar cholangiocarcinomas: Guidelines of the French Association for the Study of the Liver (AFL). from <https://afef.asso.fr/recommandation/recommandations-afef-sur-prise-en-charge-des-cholangiocarcinomes-intrahepatiques-et-perihilaires/>
- [39] Jabłońska B. Biliary cysts: Etiology, diagnosis and management. *World J Gastroenterol WJG.* 2012; 18(35): 4801–10.
- [40] Urushihara N, Fukumoto K, Fukuzawa H, Mitsunaga M, Watanabe K, Aoba T, *et al.* Long-term outcomes after excision of choledochal cysts in a single institution: operative procedures and late complications. *J Pediatr Surg.* 2012; 47(12): 2169–74.
- [41] Ohashi T, Wakai T, Kubota M, Matsuda Y, Arai Y, Ohyama T, *et al.* Risk of subsequent biliary malignancy in patients undergoing cyst excision for congenital choledochal cysts. *J Gastroenterol Hepatol.* 2013; 28(2): 243–7.
- [42] Tan HL, Shankar KR, Ford WD. Laparoscopic resection of type I choledochal cyst. *Surg Endosc* 2003; 17(9): 1495.
- [43] Tajiri H. Choledochocoele containing stones. *Am J Gastroenterol* 1996; 91: 1046–8.
- [44] Jordan PH, Jr., Goss JA, Jr., Rosenberg WR, Woods KL. Some considerations for management of choledochal cysts. *Am J Surg* 2004; 187: 790–5.
- [45] Ammori BJ, Jenkins BL, Lim PC, *et al.* Surgical strategy for cystic diseases of the liver in a Western hepatobiliary center. *World J Surg* 2002; 26 (4): 462–9.
- [46] Waechter FL, Sampalo JA, Pinto RD, *et al.* the role of liver transplantation in patients with caroli's disease. *Hepatogastroenterology* 2001; 48 (39): 672–4.
- [47] Heikkinen ES, Salminen PM. Congenital choledochal cyst opening into the intraduodenal part of the common bile duct and complicated by cholelithiasis and acute pancreatitis. *Acta Chir Scand* 1984; 150: 183–5.

- [48] Schimpl G, Sauer H, Goriupp U, Becker H. Choledochocoele: importance of histological evaluation. *J Pediatr Surg* 1993; 28: 1562-5.
- [49] Sarris GE, Tsang D. Choledochocoele: case report, literature review, and a proposed classification. *Surgery* 1989; 105: 408-14.