

Choledochal cyst of the cystic duct: Report of imaging findings in three cases and review of literature

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Abstract

The choledochal cysts, which refer to the cystic dilatation of the biliary duct, are rare lesions generally seen in children. Choledochal cyst of the cystic duct is an uncommon entity. Often, it is associated with the choledochal cyst of the rest of the biliary tree. Isolated cystic duct choledochal cyst is quite rare. Most of these lesions have been recognized only on surgery. Modern cross-sectional imaging methods have facilitated preoperative recognition and characterization of cystic duct cyst enabling formulation of appropriate management strategy. We report the imaging findings in three cases of choledochal cyst involving the cystic duct. All these cases were correctly diagnosed preoperatively. A review of cases reported in the literature and the role of imaging in guiding the management are also presented.

Keywords: Choledochal cyst; computed tomography; cystic duct cyst; magnetic resonance cholangiopancreatography

Introduction

Choledochal cysts of the biliary tract are well-known entities. However, choledochal cyst of the cystic duct is exceedingly rare. These lesions were often misdiagnosed in the past and were mostly recognized at surgery. Ultrasound is a readily available investigation and the cases of suspected choledochal cyst on ultrasonography (USG) can be further evaluated on magnetic resonance cholangiopancreatography (MRCP). With the advent of modalities like magnetic resonance imaging (MRI) and MRCP, more cases are being recognized preoperatively, which provides a road map to appropriate surgical management.

Case Reports

Case 1

A 26-year-old female presented with pain and jaundice.

Laboratory findings included mild elevation of total and direct serum bilirubin and raised levels of alkaline phosphatase. USG done previously had revealed a mass lesion in the gall bladder, with prominent hepatic ducts and the common bile duct (CBD). A tortuous cystic lesion was seen along the course of the cystic duct. The patient was referred for further radiological evaluation. Contrast-enhanced computed tomography of abdomen showed an enhancing mass lesion involving the gall bladder with regional lymphadenopathy. Cystic duct was dilated and tortuous and showed wide communication with CBD [Figure 1A]. A calculus was seen in distal CBD. The patient underwent endoscopic retrograde cholangiopancreatography (ERCP) to remove the obstruction. MRI and MRCP showed grossly dilated and tortuous cystic duct with a wide opening into a prominent common hepatic duct, in addition to gall bladder mass. The CBD was normal in caliber [Figure 1B and C].

The patient underwent laparoscopic radical cholecystectomy for the gall bladder mass. Concurrent excision of the cystic duct and partial excision of the CBD was performed, due to wide opening of the cystic duct into the CBD. The continuity was established by biliary enteric anastomosis. There was no immediate postoperative complication. The patient was discharged on the 7th postoperative day.

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Case 2

A 16-year-old male presented with intermittent upper abdominal pain, dark urine, and fever for 1 year. Hyperbilirubinemia was present on biochemical evaluation. Patient had an ultrasound done which indicated the presence of dilatation of the right hepatic duct. A cystic area was reported close to the liver hilum.

Abdominal MRI and MRCP showed fusiform dilatation of the cystic duct. The cystic duct opened into CBD through a wide opening. Fusiform dilatation of the right hepatic duct was seen. Left-sided ducts were mildly dilated [Figure 2A-C]. A diagnosis of choledochal cyst involving the cystic duct and intrahepatic ducts was made. The patient underwent open cholecystectomy with cyst excision and Roux-en-y hepaticojejunostomy.

Case 3

A 25-year-old male presented with intermittent right quadrant pain, fever, and jaundice for 5 years. He had been investigated in the past for his symptoms and was carrying multiple biochemical and ultrasound reports documenting the presence of hyperbilirubinemia and cholelithiasis with bilobar fusiform biliary dilatation. He was advised surgery in the past, but had refused to undergo the same. Increasing frequency of the symptoms forced him to seek a definitive therapy.

Abdominal MRI and MRCP revealed cholelithiasis and gross fusiform dilatation of right, left, and common hepatic

ducts. Calculus was seen in the dilated right hepatic duct. The cystic duct was dilated, tortuous, and opened into normal caliber CBD with a narrow neck [Figure 3A-C]. A diagnosis of type IV choledochal cyst with cystic duct involvement and cholelithiasis was made. Open cholecystectomy with complete excision of the dilated extrahepatic biliary ducts was done and continuity was established with roux-en-y hepaticojejunostomy for right and left ducts separately. The hepatolithiasis were retrieved during surgery.

Discussion

Choledochal cyst refers to isolated or combined dilatation of extrahepatic and intrahepatic biliary system of congenital origin.^[1] The prevalence varies widely, ranging from 1:30,000-50,000 in the western population^[2-4] to 1:1000 in Asia.^[4] Females are more commonly affected. Majority of the cases present in childhood. Alonso-Lej and colleagues proposed the first classification for choledochal cyst in 1959,^[5] which was expanded by Todani in 1977.^[6]

Bode and Aust reported the first case of cystic dilatation of cystic duct in 1983.^[7] Since then, the description of this entity has largely been in the form of isolated case reports and case series. Several terms including choledochal cyst of cystic duct,^[8] cystic duct cyst,^[9] type VI choledochal cyst,^[10,11] choledochoceles of cystic duct,^[2] and cystic malformation of

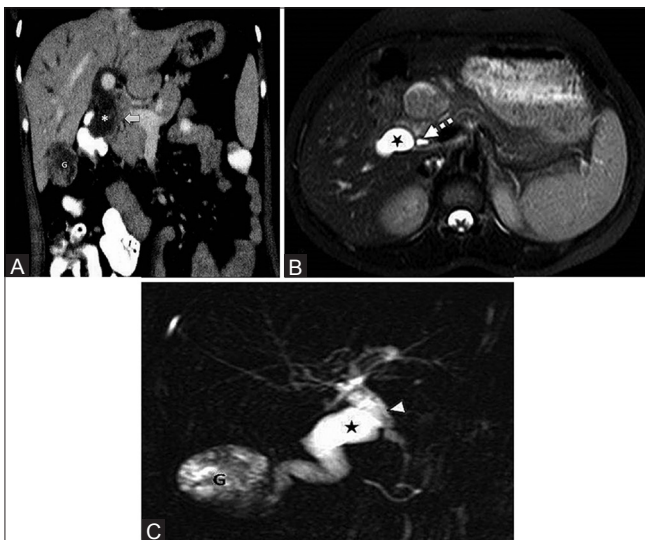


Figure 1 (A-C): Case 1: Cystic duct cyst in a case of carcinoma of gall bladder. (A) Coronal reformatted image from contrast-enhanced MDCT of abdomen done for evaluation of gall bladder carcinoma shows dilated and tortuous cystic duct (*). CBD distal to the opening of cystic duct is normal in caliber (arrow). Enhancing mass lesion in the gall bladder (G) is also seen. (B) Axial T2-weighted fat-suppressed MRI at the level of body of pancreas showing the dilated cystic duct (*). Normal caliber CBD is seen medially (arrow). (C) Thick slab coronal oblique MRCP image shows a dilated and tortuous cystic duct (*) with a wide neck insertion into the prominent common hepatic duct (arrow head). The distal common bile duct is of normal caliber. The gall bladder (G) shows intraluminal hypointense lesion

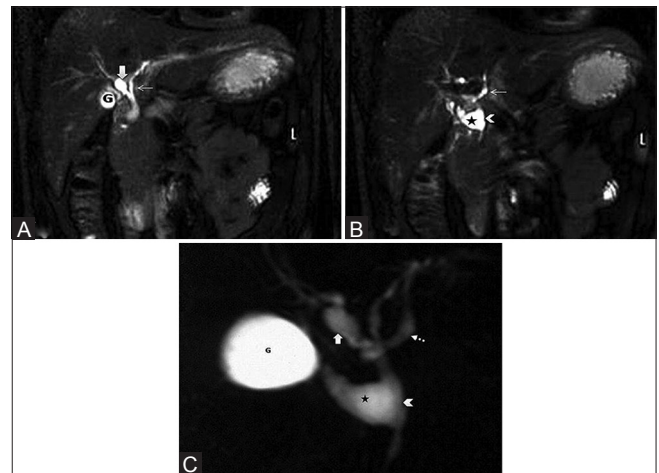


Figure 2 (A-C): Case 2: Cystic duct cyst with wide opening into the CBD associated with fusiform dilatation of the right and left hepatic ducts. (A) T2-weighted fat-suppressed (SPAIR) coronal MR images of abdomen show saccular dilatation of the right hepatic duct (bold arrow) with mild prominence of the left hepatic duct. (B) T2-weighted fat-suppressed (SPAIR) coronal MR images of abdomen show dilated cystic duct (*) with a wide neck insertion into the common hepatic duct (arrow head). Mild prominence of the left hepatic duct is seen (small arrow). Gall bladder is normal (G). (C) Coronal MR cholangiopancreatography image showing fusiform dilatation of the cystic duct (*) with a wide neck insertion (arrow head) into the normal caliber common bile duct. There is fusiform dilatation of the right hepatic duct (bold arrow). The left-sided biliary system is mildly dilated (dotted arrow). The gall bladder (G) is well distended

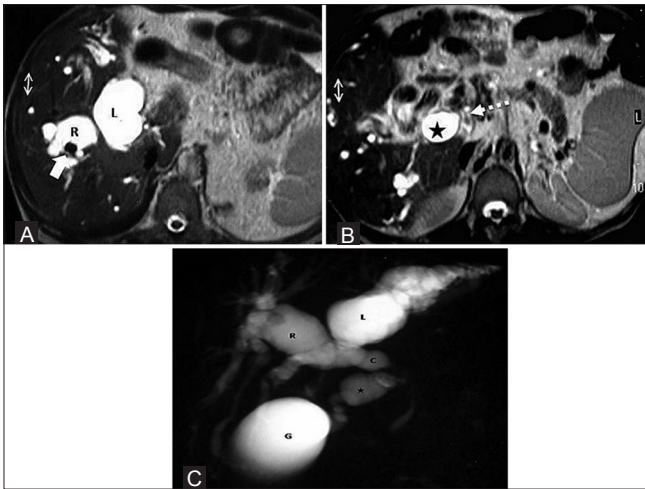


Figure 3: Case 3: Cystic duct cyst associated with type IV choledochal cyst. (A) T2-weighted axial MRI abdomen image shows gross fusiform dilatation of the right (R) and left (L) hepatic ducts. Calculus is seen in the dilated right hepatic duct (bold arrow) as an intraluminal signal void. (B) Another T2-weighted axial MRI abdomen image at the caudal level shows the dilated cystic duct (*). The common bile duct (dotted arrow) has a normal caliber. Multiple dilated intrahepatic biliary radicals are also seen. (C) Coronal MRCP image shows the dilated distal cystic duct with narrow opening into a normal caliber common bile duct. Gross fusiform dilatation of the right (R) and left (L) hepatic ducts is seen. The common hepatic duct (C) is also dilated. Gall bladder (G) is also visualized

cystic duct^[12,13] have been used. We carried out a search on the Medline database and Google search using the above terms and the term choledochal cyst. A total of 18 studies with 31 cases were found.^[1,2,7-22] Table 1 contains a summary of these cases, including our series.

The most widely accepted theory for occurrence of choledochal cyst postulates that an anomalous pancreatic biliary ductal junction (APBDJ) with long common channel is the primary abnormality.^[7,16,23] APBDJ has been reported in 33-90.2% of choledochal cysts.^[23,24] It has been suggested that abnormal APBDJ results in reflux of the pancreatic enzymes in the CBD. Due to loss of sphincteric action of the duodenum, the pancreatic enzymes cause weakening of the walls of the biliary channels ultimately leading to dilatation. This has mostly been seen with type I cyst and supported by the level of pancreatic enzymes in biliary secretions. However, this does not completely explain the pathogenesis of the cysts where the CBD is normal. Another popular hypothesis includes a ganglionosis with proximal biliary dilatation.^[25] According to De *et al.*,^[18] cystic duct cyst may result from a combination of APBDJ, acute angulation, and wide orifice of cystic duct hepatic duct junction. However, APBDJ has been observed only in three cases.^[11,14,21] Also, wide opening of the cystic duct into hepatic duct was not found in all cases. APBDJ was not seen in our cases. Hence, in our view, other factors may be responsible for cystic duct dilatation. Congenital stricture^[26] and aganglionic segment with dilatation of proximal segment^[4,18] have also been reported as the possible etiologies. The evaluation of

other factors which might be responsible requires further research.

The clinical presentation of patients with cystic duct cyst was generally the same as seen with other types of the choledochal cyst, and included abdominal pain, fever, and jaundice. Most of the patients also had associated complicating biliary tract disease ranging from cholangitis, cholecystitis, cholelithiasis, gall bladder polyp, cystolithiasis, etc.^[1,10,12,13,17-22] Gall bladder carcinoma was seen in three cases^[9,12] including one of our cases.

Association of cystic duct cysts with other congenital biliary abnormality is well documented,^[8,11,12,14,16,17,19,21] which makes the case for inclusion of this entity within Todani's classification. However, due to wide variation in the appearance, exact classification has been a matter of debate. Serrena *et al.*^[1] proposed the inclusion of cystic duct cyst as type VI in Todani's classification. Loke *et al.*^[9] suggested that these lesions may be variant of type II 2 choledochal cysts. Recently, Mishra *et al.*^[21] have proposed a new modification, wherein cystic duct cysts are included in type II and mixed variants are grouped as type VI.

We studied the age and sex distribution of the disease. Of the 28 cases for which information regarding age was available, 17 were adults^[1,2,11-15,17-19,22] and 11 were children.^[7,12,17-19,24] Data regarding sex were available for 25 patients. Seventeen patients were females^[1,2,7,9,11,13,19-21] and eight were males.^[17,19,22] The clinical presentation of these patients was same as in those with choledochal cysts.

A plethora of imaging tools have been used for evaluation in these cases, and include USG, nuclear imaging, multidetector computed tomography (MDCT), ERCP, and MRI with MRCP. The liver-specific contrast agents show biliary excretion and are used to acquire T1-weighted MR cholangiogram which has excellent spatial resolution. Hepatobiliary imino di acetic acid (HIDA) scan is used to study the biliary anatomy and diagnose obstruction of the biliary tree. In most of the early reports, the correct diagnosis was made only at surgery.^[12,18,19] This is most likely due to lack of awareness of the entity and limitations of earlier imaging tools. With modern imaging, these cases are being correctly recognized preoperatively.

USG is generally the first modality employed for investigation of biliary symptoms. A cyst of variable size is seen in relation to portahepatis or CBD. Communication of the cyst with both gall bladder/cystic duct and CBD is seldom seen^[19] owing to the cyst being larger and additional abnormalities like dilated extrahepatic biliary tree and shadowing from calculi. Moreover, ultrasound is operator dependent. In the series of Maheshwari *et al.*,^[19] definitive sonographic diagnosis was made in only one out of six cases where USG was used.

Table 1: Summary of reported cases of cystic duct cysts

Author and year of publication	Number of cases	Imaging modalities used	Cystic duct abnormalities	Associated biliary tract abnormalities	Diagnosis preoperative/intraoperative	Management
Bode <i>et al.</i> , 1983 ^[7]	One	USG, Tc99 m PIPIDA scan, PTC	Fusiform dilatation with narrow neck	Obstructed CBD, cholangitis	Intraoperative	Cholecystectomy, cyst excision, choledochoduodenostomy
Champetier <i>et al.</i> , 1987 ^[8]	Two	USG, IVC, ERCP	Not known	Cyst of common bile duct: Case 1 Cholelithiasis: Case 2	Preoperative	Excision of cyst with bile duct cyst and cholecystectomy: Case 1 Excision of cyst with cholecystectomy: Case 2
Serena Serradel <i>et al.</i> , 1991 ^[11]	One	OCG, USG	Cystic dilatation of cystic duct with redundant cystic duct	Cystolithiasis	Intraoperative	Cholecystectomy, cystic duct excision
Loke <i>et al.</i> , 1999 ^[9]	One	USG, CT, ERCP	Dilated cystic duct with wide opening into the CBD	Cystolithiasis	Intraoperative	Cholecystectomy, cyst excision with RYHJ
Bresciani <i>et al.</i> , 1998 ^[10]	One	USG	Cyst of cystic duct with anomalous duct joining the cyst to right hepatic duct	Chronic calculous cholecystitis	Intraoperative	Video laparoscopic <i>en bloc</i> resection of cyst and gall bladder with ligature with a clip of the cystic duct and anomalous duct
Baj <i>et al.</i> , 2002 ^[2]	One	USG, oral CT cholangiography	Fusiform dilatation, wide opening	None	Preoperative	Patient refused surgery
Weiler <i>et al.</i> , 2003 ^{[11]**}	One	USG, ERCP	Not known	APBDJ	Preoperative	Excision of cyst, CBD with cholecystectomy and RYHJ
Manickam <i>et al.</i> , 2004 ^[12]	One	USG, HIDA scan, CT, MRC	Not known	APBDJ	Not known	Excision of cyst with cholecystectomy
Yoon <i>et al.</i> , 2009 ^[3]	Three		Case 1: Fusiform dilatation	Advanced carcinoma gall bladder with lymphadenopathy	Preoperative	Not known
			Case 2: Fusiform dilatation joining by a wide opening	Fusiform dilatation of CBD, cholelithiasis chronic cholecystitis	Preoperative	Refused surgery
			Case 3: Fusiform dilatation with wide opening in the CBD	CBD dilatation, gall bladder polyps	Preoperative	Cyst excision, RYHJ ¹
Chan <i>et al.</i> , 2009 ^[14]	One	CT, MRCP	Fusiform dilatation with narrow opening in CBD	Cholelithiasis, chronic cholecystitis	Intraoperative	Laparoscopic excision of the cyst
Conway <i>et al.</i> , 2009 ^[15]	One	CT, ERCP, HIDA	Fusiform dilatation with narrow opening in CBD	-	Intraoperative	Excision of cystic duct cyst
Ghatak <i>et al.</i> , 2010 ^[16]	One	Not known	Saccular dilatation	Fusiform dilatation of CBD	Not known	Excision of cyst, CBD, RYHJ ¹
Khanna <i>et al.</i> , 2010 ^[17]	One	USG, MRCP	Cystic dilatation with wide opening into the common bile duct	Dilatation of common hepatic duct and CBD Carcinoma gall bladder	Preoperative	Excision of cyst, gall bladder, and common hepatic duct with hepaticojejunostomy
De <i>et al.</i> , 2011 ^[18]	One	USG, CT, MRC	Cystic duct cyst with wide opening into CBD and normal distal CBD	Cholecystitis	Intraoperative	Excision of cyst, gall bladder, and distal CBD, hepaticoenterostomy
Maheshwari <i>et al.</i> , 2012 ^[19]	Ten	USG, CT, MRCP	Fusiform dilatation in six, saccular dilatation in four	Fusiform CBD dilatation: Two cases Cystic duct calculi and malignancy: One case	Preoperative	Surgical management of cyst: Five cases, details of surgery not known Surgery for other indications, no intervention for cystic duct cyst: One case Expectant management: Three cases Refused follow-up: One case
Shah <i>et al.</i> ^[20] 2013	One	USG, MRCP	Cystic dilatation with wide opening	Cholecystitis	Preoperative	Excision of cystic duct and part of CBD with RYHJ
Mishra <i>et al.</i> , 2013 ^[21]	Two	USG, MRCP	Case 1: Fusiform dilatation with wide opening Case 2: Fusiform dilatation of CBD with a wide opening	Case 1: CBD, diverticulum, choledochocoele, and cholelithiasis Case 2: Dilated CBD, right and left hepatic ducts	Preoperative Preoperative	Case 1: Excision of CDC with RYHJ ¹ , deroofting of the choledochocoele Case 2: CDC excision with RYHJ ¹
Kesici <i>et al.</i> , 2013 ^[22]	One	USG, MRCP	Fusiform dilatation	Cholelithiasis	Preoperative	Elective excision of gall bladder and cystic duct cyst offered to the patient

Contd...

Table 1: Contd...

Author and year of publication	Number of cases	Imaging modalities used	Cystic duct abnormalities	Associated biliary tract abnormalities	Diagnosis preoperative/intraoperative	Management
Present series	Three	USG, CT, MRCP	Case 1: Cystic dilatation of cystic duct with wide opening Case 2: Fusiform dilatation of cystic duct with wide opening Case 3: Cystic dilatation of cystic duct with narrow opening	Carcinoma gall bladder Fusiform dilatation of hepatic duct Fusiform dilatation of both hepatic duct and common hepatic duct	Preoperative Preoperative Preoperative	Cholecystectomy with cystic duct cyst excision, removal of CBD with RYHJ Cholecystectomy with cystic duct excision, and CBD excision with RYHJ Open cholecystectomy with complete excision of extra hepatic biliary ducts with RYHJ with right and left hepatic ducts separately

USG: Ultrasound, Tc PIPIDA: Technitium para isopropyl iminodiacetic acid, PTC: Percutaneous transhepatic cholangiography, IVC: Intravenous cholangiography, ERCP: Endoscopic retrograde cholangiopancreatography, OCG: Oral cholecystography, CT: Computed tomography, HIDA: Hepatobiliaryiminodiacetic acid, MRCP: Magnetic resonance cholangiopancreatography, CBD: Common bile duct, APBDJ: Aberrant pancreatobiliary duct junction, RYHJ: Roux-en-y hepaticojejunostomy, CDC: Choledochal cyst, **Article is in german

Abdominal MRI with MRCP is preferred for further evaluation of sonographic abnormalities due to lack of radiation and high contrast resolution which results in excellent depiction of biliary anatomy and relationships. Origin of cyst from the cystic duct can be easily ascertained and the associated biliary abnormalities are well seen. Normal cystic duct diameter has an upper limit of 5 mm.^[27] The cystic duct dilatation may be fusiform or saccular; the fusiform variety is more common. The width of opening of cyst into the CBD is important in cases of isolated cyst due to management implications^[9] and must be specifically mentioned in the report. According to Maheshwari *et al.*, cystic duct cyst with a wide opening cannot be distinguished from type II choledochal cyst. However, we feel that with the use of MRI and MRCP, such problem seldom arises.

Management of cystic duct cyst is influenced by multiple factors. These include association of other anomalies, type of opening of the cyst into the CBD, and surgical expertise available. According to Bode *et al.*,^[7] the management should aim at prompt diagnosis, examination of the entire biliary tree, complete cyst excision, establish continuity of the biliary tree and intestines, and cholecystectomy. Some authors advocate laparoscopic resection in cases where the isolated cyst has narrow opening into the CBD.^[9,15] For cases with a wide opening, associated dilatation of other parts of biliary tree and complete resection of the dilated segment with re-establishment of biliary intestinal communication is the preferred treatment.^[9,15] In most cases, an open surgery was performed.^[7-9,11,12,15,16,18,21] Bresciani *et al.*^[10] performed video laparoscopic excision. Laparoscopic method was used in our first case as well.

Conclusion

In conclusion, we have reported three different variants of cystic duct cyst. Ultrasound and abdominal MRI with MRCP are helpful for preoperative diagnosis and mapping other biliary abnormalities in most cases, which is crucial for appropriate surgical management.

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