Case Report

Management of Choledochal Cyst of Cystic Duct in Adult: A Very Rare Case Report

Arnetta Naomi Louise Lalisang[1,2], Ocsyavina Ocsyavina[1], Patricia Rosalind Ismantara[2,3]

1. Digestive Surgery Division, Department of Surgery, cipto mangunkusumo hospital, Jakarta, Indonesia; 2. Faculty of Medicine, Faculty of Medicine, Universitas Indonesia, Cipto Mangunkusumo Hospital, Indonesia; 3. cipto mangunkusumo hospital, Jakarta, Indonesia

Choledochal cyst is a rare congenital biliary anomaly, with the cystic duct variant being extremely uncommon and often difficult to diagnose due to nonspecific symptoms. This case report highlights a rare occurrence of a cystic duct choledochal cyst in an adult. We collect patient's data through examination reports, hospitalization and surgery reports documented in the medical record. Patients presented with fever, epigastric pain, nausea, and reduced oral intake, with tenderness in the epigastric and right hypochondriac regions. Imaging confirmed a cystic duct choledochal cyst, a CBD stone, and a liver abscess, prompting initial ERCP for stone clearance and stent placement, followed by laparoscopic cholecystectomy, cyst excision, and abscess drainage. The patient recovered well postoperatively. Choledochal cysts are rare in adults and often present with nonspecific symptoms such as abdominal pain, fever, and jaundice, making diagnosis challenging. Imaging modalities like MRCP and MDCT play a crucial role in identifying cystic duct cysts and associated biliary abnormalities, while early diagnosis is essential due to the risk of complications, including cholangitis, cirrhosis, and malignancy. The preferred treatment involves complete cyst excision, cholecystectomy, and biliary reconstruction, with surgical approach depending on factors like cyst anatomy and associated anomalies, followed by long-term monitoring to prevent complications. A cystic duct choledochal cyst complicated by a CBD stone and liver abscess, emphasizing the importance of early diagnosis and surgical intervention to prevent serious complications.

Corresponding author: Arnetta Naomi Louise Lalisang, arnetta.naomi01@ui.ac.id

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Introduction

Choledochal cyst (CC) is a rare congenital anomaly characterized by cystic dilatation of the biliary tract, which can involve both intrahepatic and extrahepatic bile ducts. While the classification has evolved over time, with the widely accepted Todani classification categorizing it into five types, there remains ongoing debate regarding its pathogenesis, natural course, and management. Among the various types, choledochal cyst of the cystic duct is exceedingly rare and often presents a diagnostic challenge due to its nonspecific symptoms, such as jaundice, abdominal pain, and a palpable mass. However, advancements in imaging modalities have significantly improved preoperative diagnosis and surgical planning. Given the potential risk of malignant transformation, early recognition and appropriate surgical management are crucial for optimal outcomes.[1][2] This case report presents a rare instance of choledochal cyst of the cystic duct in an adult.

Case presentation

A 64-year-old male presented with fever and epigastric pain since the previous night, accompanied by nausea without vomiting, bloating, and reduced oral intake. On examination, he was hemodynamically stable (GCS 15, BP 116/82 mmHg, HR 87 bpm, RR 20, T 36.7°C) with tenderness in the epigastric and right hypochondriac regions. Abdominal examination revealed tenderness in the epigastric and right hypochondriac regions without signs of peritonitis. Laboratory tests were within normal limits, except for an elevated erythrocyte sedimentation rate (ESR) of 109 mm/hour. Imaging studies confirmed a choledochal cyst originating from the cystic duct, along with a common bile duct (CBD) stone and a liver abscess.

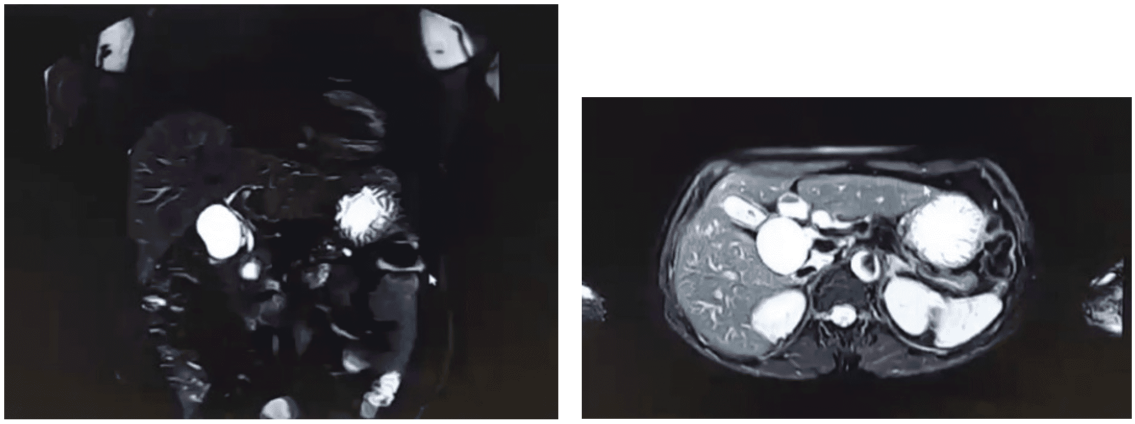

Figure 1. Confirmed a choledochal cyst originating from the cystic duct found from imaging.

The patient underwent an initial endoscopic retrograde cholangiopancreatography (ERCP) for CBD stone clearance and plastic stent insertion on February 20th. Following ERCP, a laparoscopic cholecystectomy, cyst excision, and drainage of the liver abscess were performed on February 22nd. Intraoperatively, the gallbladder showed signs of chronic inflammation, and a cystic dilation of the cystic duct was identified with no dilatation of CBD.

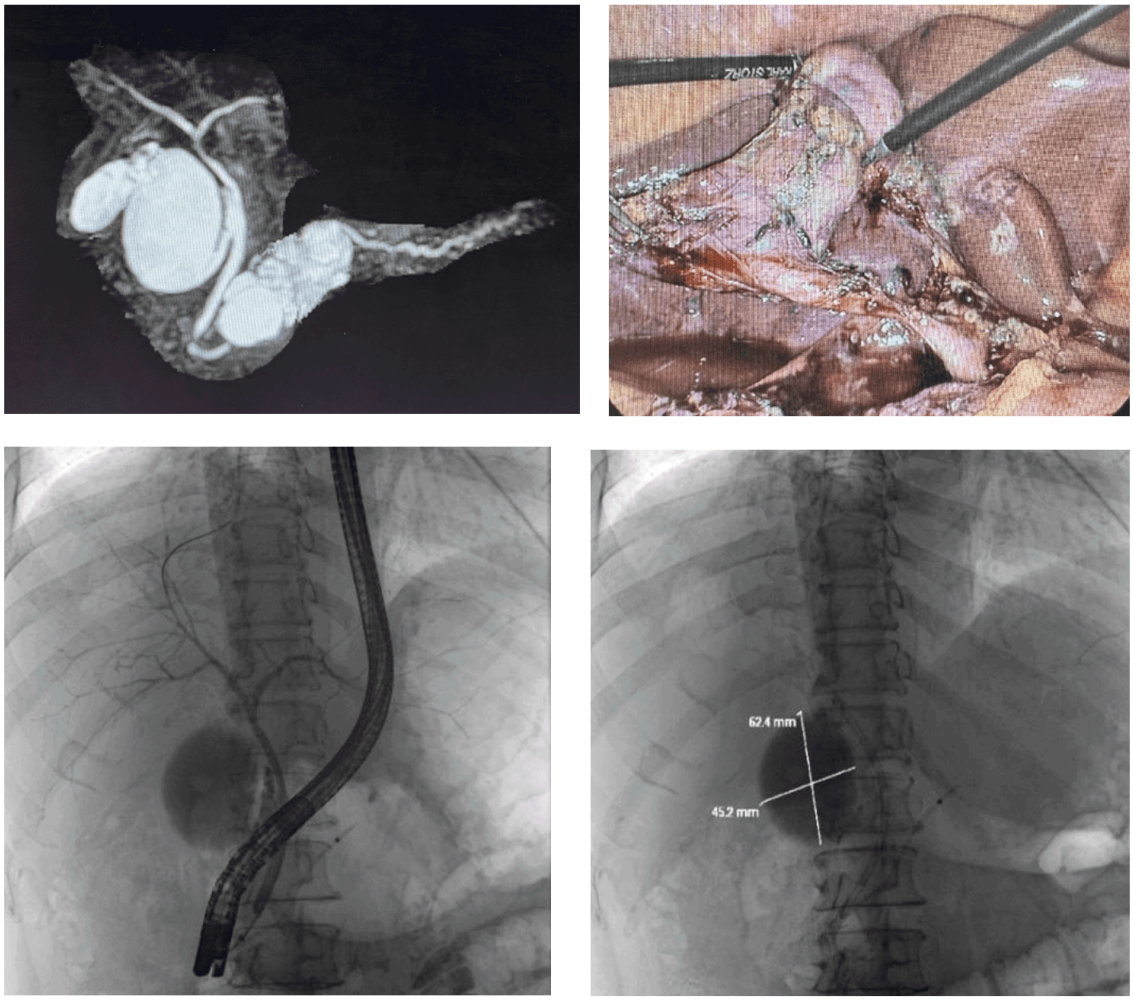

Figure 2. ERCP, laparoscopic cholecystectomy, cyst excision, and drainage of the liver abscess.

The excised tissue was sent for histopathological examination. Histopathological examination of the gallbladder tissue revealed chronic active cholecystitis with Rokitansky-Aschoff sinuses extending into the subserosal layer, moderate chronic inflammatory cell infiltration, fibrosis, and evidence of cholelithiasis.

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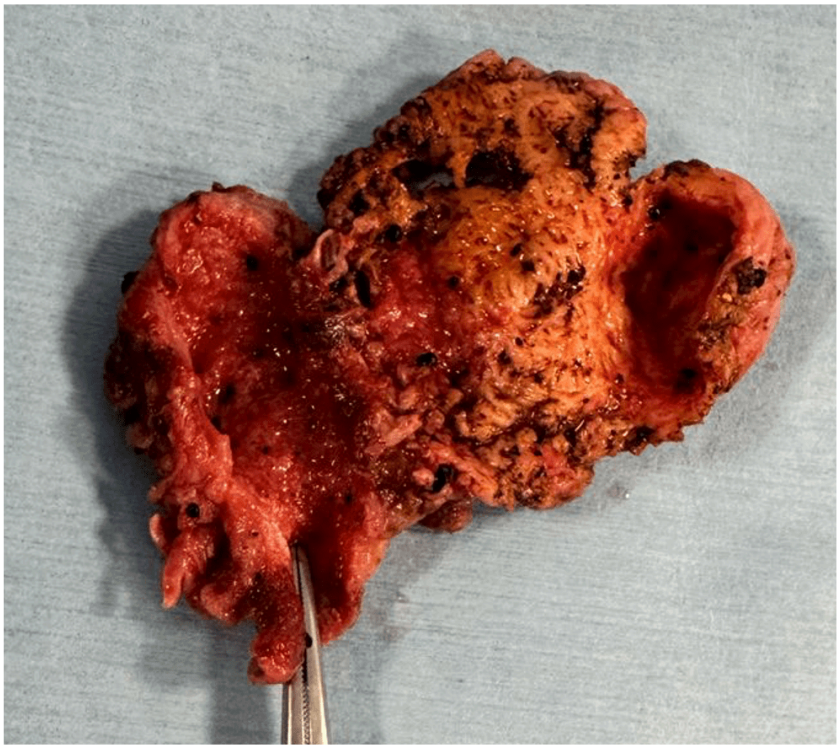

Figure 3. Choledochal cyst.

Postoperatively, the patient received intravenous antibiotics (daryaven 500 mg), proton pump inhibitors (esocazel 40 mg), antiemetics (invomit 4 mg), tomit 10 mg/2 ml IV, laxon powder 17 g sachet, microlax suppository, opilax 120 ml syrup, vectrine 300 mg capsule. The patient showed good condition and was discharged on the 3rd day.

Discussion

Choledochal cysts are congenital anomalies. The prevalence varies widely, ranging from 1 in 30,000-50,000 in Western populations to 1 in 1,000 in Asia. The condition is rare, with an estimated incidence of 1 in 13,000 to 1 in 2 million live births. It is more commonly found in females than in males. The majority of cases present during childhood. Although most cases are diagnosed in childhood, approximately 25% of patients are first diagnosed in adulthood.[2][3][4][5]

Choledochal cysts are suspected to occur due to an anomalous pancreaticobiliary ductal junction (APBDJ). It is thought that APBDJ allows pancreatic enzymes to reflux into the common bile duct (CBD). The loss of

sphincter function at the duodenum leads to weakening of the bile duct wall, eventually resulting in ductal dilation. Another etiological hypothesis involves ganglionosis, causing proximal biliary duct dilation.[2][5]

In neonates or children, choledochal cysts usually present with an abdominal mass or abdominal pain. Initial presentation in adults is rare and often presents with nonspecific symptoms, such as abdominal pain, fever, and jaundice. The clinical symptoms of patients with cystic duct cyst were generally similar to those observed in other types of choledochal cysts. Most adult patients also have concomitant biliary diseases, including cholangitis, cholecystitis, cholelithiasis, gallbladder polyps, and cystolithiasis. The clinical presentation may be episodic and nonspecific, with 69% of patients showing abnormal liver function test results. Diagnosis is often delayed.[2][5]

The association between cystic duct cysts and other congenital biliary abnormalities is well established, supporting their inclusion within Todani's classification. However, due to significant variations in their presentation, their precise classification remains a topic of debate. Serrena et al. advocated for categorizing cystic duct cysts as type VI within Todani's system, while Loke et al. suggested that these cysts may represent a variant of type II choledochal cysts. More recently, Mishra et al. proposed a revised classification in which cystic duct cysts are grouped under type II, while mixed variants are designated as type VI.[2]

Various imaging modalities have been used to evaluate choledochal cysts of cystic duct, including ultrasound (USG), nuclear imaging, multidetector computed tomography (MDCT), endoscopic retrograde cholangiopancreatography (ERCP), and magnetic resonance imaging (MRI) with magnetic resonance cholangiopancreatography (MRCP). Ultrasound is generally the first-line investigation in biliary symptoms and may reveal cystic structures of varying sizes around the porta hepatis or the CBD. However, ultrasound findings are highly operator-dependent. In one study, only one out of six cases could be definitively diagnosed by ultrasound.[2][5]

ERCP is traditionally considered the gold standard for diagnosing choledochal cysts. However, it carries the risk of morbidity due to its invasiveness. Alternatively, MRCP is a valuable, noninvasive diagnostic tool with good accuracy for detecting and classifying choledochal cysts. MDCT with image reconstruction is another important technique that provides detailed anatomical information about the biliary tree and the pancreaticobiliary ductal union. Liver-specific contrast agents exhibit biliary excretion and are utilized to obtain T1-weighted MR cholangiograms, which provide exceptional spatial resolution. A hepatobiliary imino diacetic acid (HIDA) scan is employed to assess biliary anatomy and detect obstructions in the

biliary system. Nevertheless, both MRCP and MDCT have limitations in detecting minor ductal anomalies.[2][5][6]

Abdominal MRI with MRCP is the preferred modality for further evaluating sonographic abnormalities, as it avoids radiation exposure and offers high contrast resolution, allowing for detailed visualization of biliary anatomy and its relationships. The cyst's origin from the cystic duct can be clearly identified, along with any associated biliary abnormalities. The normal cystic duct diameter has an upper limit of 5 mm. Dilatation of the cystic duct can be either fusiform or saccular, with the fusiform type being more common.[5][6]

In the past, choledochal cysts were often treated with drainage procedures. However, it is now known that 30% to 50% of patients undergoing such procedures develop long-term complications, including acute cholangitis, secondary biliary cirrhosis, and even cholangiocarcinoma. Biliary tract malignancy has been reported to occur in 2.5% to 28% of patients with choledochal cysts, a risk approximately 20 times higher than in the general population. The risk increases with age, reaching 14.5% in patients over the age of 20. The malignancy risk is also higher in patients with a history of internal drainage procedures (such as cystoduodenostomy).[4]

The preferred treatment for choledochal cysts involves complete excision of the extrahepatic bile duct, cholecystectomy, and reconstruction using Roux-en-Y hepaticojejunostomy, which carries low morbidity and mortality rates. Management of cystic duct cysts depends on several factors, including associated anomalies, the type of cyst opening into the common bile duct (CBD), and available surgical expertise. Bode et al. emphasize the importance of early diagnosis, thorough evaluation of the biliary system, complete cyst excision, restoration of biliary-intestinal continuity, and cholecystectomy. Some experts suggest laparoscopic resection for isolated cysts with a narrow opening into the CBD. However, for cysts with a wide opening, or when other parts of the biliary system are dilated, complete resection of the affected segment with biliary-intestinal reconstruction is preferred. In most cases, open surgery has been performed. Long-term monitoring is recommended for all patients with choledochal cysts.[2][5]

Conclusion

Choledochal cysts, though rare congenital anomalies, should be considered in adult patients presenting with nonspecific biliary symptoms. This case highlights a rare presentation of a choledochal cyst originating from the cystic duct in an adult male, complicated by CBD stone and liver abscess. Early and

accurate diagnosis through advanced imaging modalities, followed by appropriate surgical intervention, is essential to prevent severe complications, including recurrent cholangitis and malignancy.

Statements and Declarations

Informed Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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Declarations

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