#### **Case Report**

# A Rare Case of Choledochal Cyst Presented as Sudden and Severe Abdominal Pain



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### Abstract

We report a pediatric case of Type 1C choledochal cyst presented as acute biliary colic pain. Choledochal cyst is an abnormal expansion of the biliary tract. The Type 1 choledochal cyst is the cystic dilation of the common bile duct. The Type Ic choledochal cyst is the fusiform dilatation of the common bile duct with an anomalous pancreaticobiliary junction. Patients with choledochal cysts may have the formation of cholelithiasis due to chronic biliary stagnation and repeated inflammation. Cholelithiasis may fall into the common bile duct to cause biliary colic. Abdominal sonography can be used as a preliminary examination.

Keywords: Choledochal cyst, cholelithiasis, laparotomy, sonography

## INTRODUCTION

Choledochal cysts are often diagnosed before the age of 10 years and are more common in females. The common symptoms are repeated abdominal pain, jaundice, and abdominal masses. Patients with choledochal cysts, due to chronic biliary stagnation and repeated inflammation, will experience the formation of biliary sludge, which will then lead to cholelithiasis.<sup>[1]</sup> Cholelithiasis may fall into the common bile duct to cause biliary colic. If these symptoms are portrayed, an abdominal sonography can be used as a preliminary examination. We report a pediatric case of choledochal cyst presented as acute biliary colic pain.

## **CASE REPORT**

This is a 10-year-old Taiwanese female with normal growth and development. The vaccinations were administered as scheduled. The family history was not remarkable. According to the patient and her family, she experienced with sudden acute epigastric colic and stinging pain shortly after dinner. She needed to bent her body in the waist and knee to relieve the pain, and she could hardly walk. The patient neither had nausea, vomiting, nor fever. She was initially diagnosed by the clinic as gastrospasms. Intravenous analgesics were

Received: 25-12-2019 Revised: 30-01-2020 Accepted: 14-02-2020 Available Online: 12-09-2020

Access this article online	
Quick Response Code:	Website: www.jmuonline.org
	<b>DOI:</b> 10.4103/JMU.JMU_27_20

administered for pain control. However, she continued to have persistent epigastric pain. Two days later, she was sent to another clinic with the same diagnosis gastrospasm, and intravenous analgesics were again administered. However, the severe epigastric pain and symptoms remained. She was sent to our emergency department on the same night.

Physical examination showed apparent tenderness and knocking pain over the right upper quadrant abdomen. There was neither rebounding pain nor jaundice. Abdominal roentgen revealed no specific findings. A computed tomography (CT) scan showed narrowing of the distal common hepatic duct with dilatation of intrahepatic bile ducts, gallbladder, and proximal common bile duct [Figures 1 and 2]. The patient was admitted for further evaluation and treatment.

The blood chemistries on admission showed hemoglobin 14.2 g/dL, leukocytes  $8.46 \times 1000/uL$ , total bilirubin 1.1 mg/dL, direct bilirubin 0.6 mg/dL (H), aspartate aminotransferase 58 IU/L, alanine aminotransferase 49 IU/L, alkaline phosphatase 180 IU/L, amylase 50 IU/L, lipase 11 IU/L, blood urea nitrogen 9 mg/dL, creatinine 0.62 mg/dL, and C-reactive protein 0.885 mg/dL.

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as sudden and severe abdominal pain. J Med Ultrasound 2020;28:256-9.

Lin: choledochal cyst, cholelithiasis, gastrospasm

An abdominal sonography showed the Type 1C choledochal cyst and narrowing of the distal common hepatic duct with dilatation of intrahepatic bile ducts, gallbladder, and proximal common bile duct [Figure 3]. A subsequent magnetic resonance cholangiopancreatography (MRCP) showed a small 0.4 cm liver cyst at segment 2 and narrowing of distal common bile duct with dilation of bilateral intrahepatic bile ducts, gallbladder, and proximal common hepatic duct. A 0.8 cm stone was found in the distal common bile duct [Figure 4]. A common channel was found outside the duodenal wall [Figure 5], between the pancreas duct and the common bile duct.

A laparotomy was performed 3 days later. An intraoperative cholangiography showed an abnormal long common channel >1.5 cm between the pancreatic and common bile duct outside the duodenal wall, compatible with Type 1C choledochal cyst. The surgical operation included hepaticojejunostomy, Roux-en-Y hepaticojejunostomy, and jejunojejunostomy with excision of choledochal cyst and cholecystectomy. The common bile duct cyst was excision

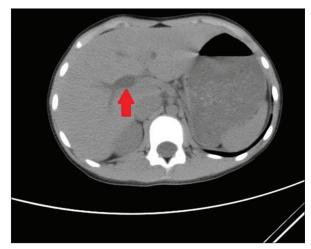


Figure 1: Abdominal computed tomography showing dilation of bilateral intrahepatic bile ducts



Figure 3: Abdominal sonography showing dilated intrahepatic bile duct, common hepatic duct, and common bile duct. (Type 1C choledochal cyst)

with the reconstruction of the common hepatic duct for a bypass connecting jejunum. The pathology revealed chronic active cholecystitis, acute and chronic cholangitis, and gallbladder and common bile duct stones [Figures 6 and 7]. The patient started food intake on the postoperative day 3 and discharged on the postoperative day 10. The patient was followed up at the outpatient clinic with satisfied recovery without complications.

## DISCUSSION

The patient is a case of choledochal cysts presented as biliary colic with chronic active cholecystitis and gallbladder stones. The abdominal colic pain was likely due to passing gallbladder stones after a meal with high fat.<sup>[2]</sup> The common bile duct was not completely obstructed without tea-color urine and clay-colored stool. The patient did not develop severe infection without fever and chillness.

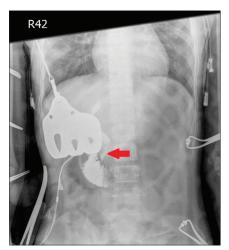


**Figure 2:** Abdominal computed tomography showing dilation of the gallbladder, common hepatic duct, and common bile duct

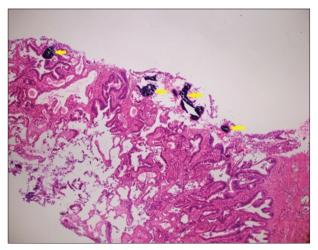


**Figure 4:** Magnetic resonance cholangiopancreatography showing dilation of the gallbladder, bilateral intrahepatic bile ducts, common hepatic duct, common bile duct, and a 0.8 cm stone located at the distal common bile duct

Lin: choledochal cyst, cholelithiasis, gastrospasm



**Figure 5:** Intra-operative cholangiography showing the abnormal connection between pancreatic and common bile duct outside the duodenal wall and formation of long common channel >1.5 cm



**Figure 6:** Pathological report showing chronic active cholecystitis and gallbladder stones

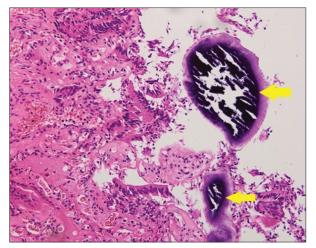


Figure 7: Pathological report showing acute and chronic cholangitis and common bile duct stones

This patient was previously diagnosed with gastrospasm. Stomach spasms are contractions of the abdominal muscles, including that of the stomach and intestines. Depending on the affected muscle group and the intensity of the spasm, affected individuals may feel either a slight muscle twitch or cramp. Stomach spasms are mostly harmless, but they could represent a symptom of an underlying condition. Stomach spasms occur when the muscles of the stomach or intestines contract. Most cases of stomach spasms are harmless but may indicate an underlying condition that requires attention.<sup>[3]</sup>

Cholelithiasis is the presence of solid concretions in the gallbladder. Gallstones form in the gallbladder but may exit into the bile ducts (choledocholithiasis).<sup>[4]</sup> Symptoms ensue if a stone obstructs the cystic, bile, or pancreatic duct. Biliary colic is a common presentation of a stone in the cystic duct or common bile duct of the biliary tree. Colic refers to the type of pain that "comes and goes," typically after eating a large, fatty meal which causes contraction of the gallbladder.<sup>[5]</sup>

Choledochal cyst is an abnormal expansion of the biliary tract. The formation of a common channel between the common bile duct and pancreatic duct where they enter the duodenum is an important factor in the development of a choledochal cyst. Pancreatic juice with digestive enzymes can flow retrograde into the common bile duct to cause damages, and the lumen wall becomes fragile and cystic or shuttle change to form choledochal cysts.

Alonso-Lej divided the choledochal cysts into three main types. The first type is the cystic dilation of the common bile duct, the second type is the formation of a diverticulum in the common bile duct, and the third type is a protrusion of the common bile duct. In 1977, Todani proposed Type 4a as common bile duct cyst combined with intrahepatic bile duct dilation, Type 4b as multiple extrahepatic choledochal cysts, and Type 5 as multiple intrahepatic bile duct dilation. The cystic dilatation of Type I choledochal cyst is further classified as diffuse (1a), focal (1b), and fusiform with anomalous pancreaticobiliary junction (1c). Our case is consistent with Type 1C choledochal cyst.

Choledochal cysts are more likely to occur in children younger than 10 years old, with only a few of them progressed into malignancy. Typical symptoms of choledochal cysts include abdominal pain, jaundice, and a palpable mass at the right upper abdomen.<sup>[6]</sup> Infants may develop clay-colored stool. The abdominal sonographic findings of the biliary tract can be confirmed by the subsequent CT or MRCP. The main primary treatment is surgical resection, and the prognosis is usually good.

## CONCLUSION

We report a pediatric case of Type 1C choledochal cyst presented as acute biliary colic pain. Patients with choledochal cysts may develop cholelithiasis. Cholelithiasis may fall into the common bile duct to cause biliary colic. Abdominal sonography can be used as a preliminary examination for the differential diagnosis. Lin: choledochal cyst, cholelithiasis, gastrospasm

#### **Declaration of patient consent**

The authors certify that they have obtained appropriate patient and the legal guardian consent form. In the form, the patient and the legal guardian have given their consent for the patient's images and other clinical information to be reported in the journal. The patient and the legal guardian understand that the name and initial will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

# Financial support and sponsorship Nil.

#### **Conflicts of interest**

There are no conflicts of interest.

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