Clinics in Surgery

Case Report
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A Rare Case of Sudden and Severe Abdominal Pain in Children

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Abstract

Introduction: Symptoms of choledochal cysts, constituting an uncommon congenital disease of bile duct anomaly, usually appear from the first year of life. The classic triad comprises intermittent abdominal pain, jaundice, and a right upper quadrant abdominal mass. However, this trial can be found only in a few patients. Herein, we describe the case of a young girl who experienced severe right upper quadrant abdominal pain that was diagnosed as choledochal-cyst-related biliary colic, for which she underwent surgery.

Case Report: A 10-year-old girl presented to Cathay General Hospital with sudden and severe epigastric pain lasting for 2 days. Additionally, her complaints included nausea and nonbilious vomiting. Laboratory data revealed total and direct bilirubin levels to be 1.1 and 0.6 mg/dL, respectively. The patient's condition had previously been diagnosed as gastrospasm by local doctors; however, the symptoms were persistent and even progressive. A diagnosis for type 1C choledochal cyst with biliary colic associated with distal common bile duct stone was made through abdominal computed tomography, sonography and magnetic resonance cholangiopancreatography. The patient was surgically treated through cholecystectomy, choledochal cyst excision, and Roux-en-Y hepaticojejunostomy. The patient recovered well without recurrence.

Conclusion: Differential diagnoses must be considered in cases involving persistent gastrospasm. Although most choledochal cysts are diagnosed at infancy, they can be diagnosed in older children as well and usually present as abdominal pain due to accompanying pancreatitis. However, choledochal cysts with abdominal pain due to biliary colic can also be one of the possible differential diagnoses for acute abdominal pain in children or adolescents.

Keywords: Choledochal cyst; Biliary colic; Abdominal pain; CBD stone

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Abbreviations

CBD: Common Bile Duct; IHD: Intrahepatic Duct; CHD: Common Hepatic Duct; CT: Computed Tomography; MRCP: Magnetic Resonance Cholangiopancreatography

Introduction

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. Choledochal cysts are congenital dilatations of the extrahepatic or intrahepatic biliary tree and are a rare cause of acute abdomen, and their prevalence is relatively high in Asian populations [1,2]. Although choledochal cysts may be discovered at any age and in any sex, it is mostly diagnosed in infants and young women. Surgery is usually required to remove the abnormal tissue and gallbladder. The classic clinical trial of choledochal cyst comprises intermittent abdominal pain, jaundice, and a right upper quadrant abdominal mass, but the symptoms and signs may vary from case to case. Choledochal cysts have recently been categorized into five major types and several subtypes in the Todani classification, which is the most commonly used classification method [3,4]. Choledochal cysts in older pediatric patients might cause abdominal pain due to acute pancreatitis. However, in the case reported herein, laboratory data for amylase and lipase were within normal ranges. We report are case of a 10-year-old girl diagnosed as having choledochal cysts with sudden onset of acute abdominal pain related to biliary colic caused by Common Bile Duct (CBD) stone.

Case Presentation

The patient was born at 38 weeks of gestation. At birth, she was healthy and had no known systemic diseases. She received scheduled vaccinations and achieved appropriate developmental milestones. She was taken to a local clinic due to sudden-onset abdominal pain and nausea and suspected stomach spasms. However, her symptoms persisted or even worsened; subsequently, she was brought to the emergency department of Cathay General Hospital, Taiwan. She complained of sudden onset of severe cramping abdominal pain lasting for 2 days and nonbilious vomiting once with food content. She had neither fever nor diarrhea. Her physical examination revealed acceptable vital signs (body temperature: 36.3°C; heart rate: 68 beats/min; respiratory rate: 16 breaths/min; and blood pressure: 104/63 mmHg) but a distended abdomen, a positive Murphy's sign, and knocking pain over the right upper abdomen. Laboratory tests revealed normal white blood cell (8.46×10^3 cells/mm^3), amylase (50 IU/L), and lipase (11 IU/L), and elevated C-reactive protein (0.885 mg/dL), aspartate aminotransferase (58 IU/L), alanine aminotransferase (49 IU/L), total bilirubin (1.1 mg/dL), direct bilirubin (0.6 mg/dL), and *alkaline phosphatase (180 IU/L) levels*. A plain radiograph of the abdomen revealed distended bowel loops with fecal matter. Abdominal computed tomography (Figure 1 and 2) and sonography (Figure 3) revealed dilatation of the gallbladder with wall thickening and dilatation of the Intrahepatic Duct (IHD), Common Hepatic Duct (CHD), and CBD. Magnetic resonance cholangiopancreatography (Figure 4) revealed a 0.8-cm distal CBD stone with bilateral IHD, CBD, and gallbladder dilatation. Her condition was diagnosed as choledochal cyst type 1C with biliary colic due to CBD stone; thereafter, surgical intervention was performed. Intraoperative cholangiography (Figure 5) confirmed that the junction of the biliary-pancreatic duct was abnormal and formed a long fistula (>1.5 cm). Subsequently, laparoscopic cholecystectomy, choledochal cyst excision, and Roux-en-Y hepaticojejunostomy were performed successfully. A pathology report (Figure 6 and 7) showed chronic cholecystitis with cholelithiasis and acute and chronic cholangitis with choledocholithiasis. This case report was approved by the Institutional Review Board of Cathay General Hospital, Taiwan, and patient consent was obtained.

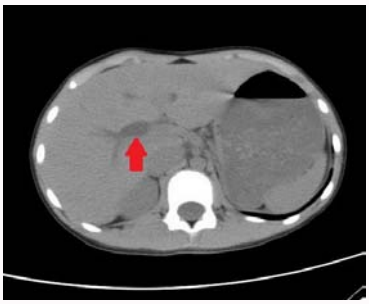

Figure 1: Abdominal computed tomography revealing bilateral IHD dilatation.

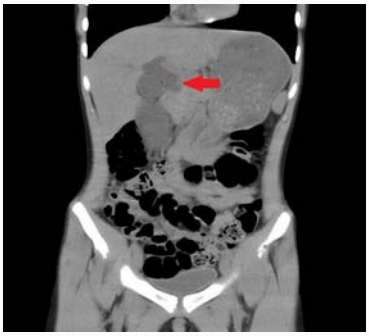

Figure 2: Abdominal computed tomography revealing distended gallbladder with wall thickening and CHD and CBD dilatation.

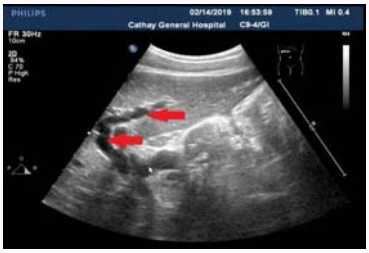

Figure 3: Sonography revealing dilatation of the gallbladder with wall thickening and dilatation of the IHD, CHD, and CBD.

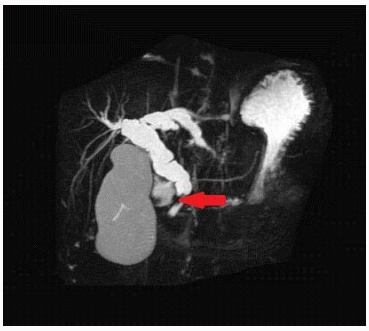

Figure 4: MRCP showing a 0.8-cm distal CBD stone with bilateral IHD, CBD, and gallbladder dilatation.

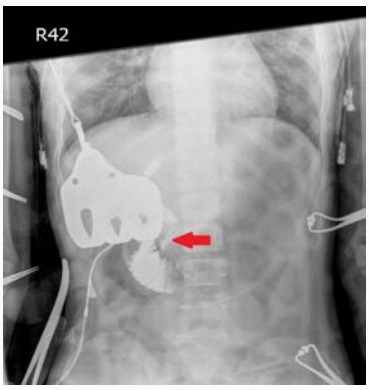

Figure 5: Intraoperative cholangiography confirmed that the junction of the biliary-pancreatic duct was abnormal and formed a long fistula (>1.5 cm).

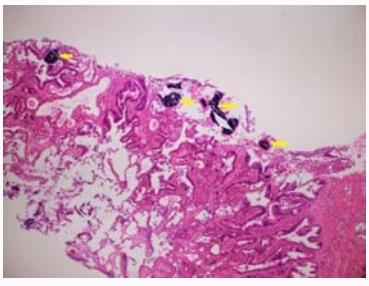

Figure 6: Pathology report showing chronic cholecystitis with cholelithiasis.

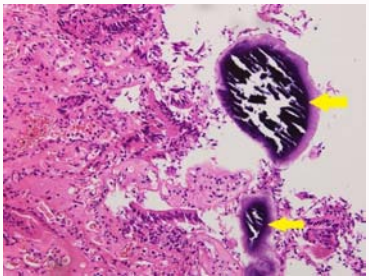

Figure 7: Pathology report showing acute and chronic cholangitis with choledocholithiasis.

Discussion

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. Biliary colic is rarely observed in children. Colic (sudden pain) occurs when a gallstone temporarily blocks the cystic duct. Biliary colic can range from mild, such as dull abdominal pain, to severe cramping pain. It is most frequently caused by the obstruction of the CBD or cystic duct by a gallstone. The pain is typically in the right upper part of the abdomen and usually lasts from 15 min to a few hours. Acute episodes of biliary pain may be induced or exacerbated by certain foods, most commonly those high in fat [5-8]. Choledochal cysts constitute a rare disease usually diagnosed in children under the age of 1 year. Although the incidence of choledochal cysts varies by country, it is remarkably higher in Asian populations, with the reported incidence being 1 per 1,000 live births [9]. In the Todani classification, choledochal cysts are categorized into five major types and several subtypes based on bile duct dilatation and pancreatic junctional abnormalities [10]. Patients with choledochal cysts may present with fever, jaundice, abdominal pain, abdominal mass, nausea and vomiting, or any symptoms secondary to cholangitis and pancreatitis. Choledochal cysts usually cause cholestasis, but clinical manifestations differ between infants and older children owing to different pathogeneses. Studies by Chen and Niramis have reported that painless jaundice usually occurs in infants and abdominal pain is more likely in older patients [11]. In most patients with choledochal cysts who have abdominal pain, the cysts are connected to the pancreatic duct. Laboratory data usually reveal elevated amylase and lipase levels. However, in this our patient, laboratory data indicated that the amylase and lipase levels were within the normal range. The final diagnosis revealed that the cause of abdominal pain was biliary colic due to CBD stone instead of stomach spasms, which is a very rare clinical manifestation in pediatric patients with choledochal cysts and can be easily overlooked by pediatricians. Early diagnosis and appropriate therapy are essential to prevent morbidity and mortality. Therefore, our case presents an experience for pediatricians that persistent stomach spasms can be an indication of other underlying diseases and choledochal cysts may be a diagnosis for older children presenting with acute abdomen without pancreatitis.

Conclusion

Persistent stomach spasms may indicate an underlying condition and must be carefully examined and diagnosed. Biliary colic is usually overlooked because it is rarely observed in pediatric patients and might be misdiagnosed as other disease. Although most choledochal cysts are diagnosed during infancy, some cases can be diagnosed at a later age due to different pathogeneses. Older children with choledochal cysts present abdominal pain due to pancreatitis, which can be cross-referenced with amylase and lipase data. However, pancreatitis is not the only cause of abdominal pain in patients with choledochal cysts; biliary colic may also cause acute abdomen in children, particularly those with severe cramping pain. Gallstone-related biliary colic should be carefully considered for persistent abdominal pain in children even when there is no obvious jaundice.

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