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Malrotation-associated cholecystoduodenal fistula

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Patient: Female, 16
Final Diagnosis: Malrotation and cholecystoduodenal fistula
Symptoms: Abdominal pain • anorexia • fever • nausea • vomiting
Medication: —
Clinical Procedure: —
Specialty: Gastroenterology and Hepatology

Objective: Anatomical anomaly/variation





Background: Cholecystoduodenal fistula (CDF) is the most common cholecystenteric fistula. It is a late complication of gallbladder disease with calculus and is mainly encountered in the elderly and females.

Case Report: We report the case of a teenage patient with cholecystoduodenal fistula and malrotation. Direct plain abdominal x-ray demonstrated air in the biliary system. Computed tomography revealed CDF-associated with an anomaly of intestinal malrotation. She had gallstones (with a few stones in the gallbladder) and cholecystitis. CDF is caused by malrotation, and cholecystitis has not been reported before. In this regard our patient is the first and youngest reported case.

Conclusions: We suggest that CDF is probably a consequence of malrotation. The patient's clinical features and operative management are presented and discussed with current literature.

MeSH Keywords: cholecystoduodenal fistula • cholecystitis • malrotation

Full-text PDF: <http://www.amjcaserep.com/download/index/idArt/889903>

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Background

Cholecystoduodenal fistula (CDF) is the most common cholecystenteric fistula and is predominantly a delayed complication of cholecystitis and cholelithiasis in women [1]. Malrotation is a rare anomaly, occurring in about 1 in 6000 live births, and is often diagnosed in the neonatal period [2]. Symptoms and signs may be nonspecific, and clinical signs may be seen later in life or may not occur at all [3].

This is the youngest case of a teenage that not only had CDF, but also an intestinal anatomic variant that consequently involved a long and difficult treatment requiring surgical re-intervention. A search of published literature in English revealed that a similar CDF due to malrotation has not yet been described. Our patient is the first CDF case to have concomitant malrotation.

Case Report

A 16-year-old girl presented with blunt and continuous right upper abdominal pain and high fever with 1-day duration. Her history revealed anorexia, nausea, and 3 times/day vomiting. She had a previous history of similar pain, a vesicoureteral reflux and recurrent abdominal pain attacks. She was usually treated under the assumption of kidney stone or urinary tract infection. Physical examination revealed a slim body with no acute distress or jaundice. Vital signs revealed a temperature of 39.1°C; blood pressure of 105/65 mmHg; heart rate of 79 beats/min; and respiratory rate of 22 breaths/min. The abdomen was soft and not distended but revealed light tenderness in the right upper quadrant. There was no rigidity or rebound. Hematological laboratory results revealed a slightly elevated white blood cell count of $11.6 \times 10^3/\text{UL}$, hemoglobin 10.6 g/dL, platelet count $225,000/\text{mm}^3$, erythrocyte sedimentation rate (ESR) 44 mm/h, and C-reactive protein 17.1 mg/dL. There was no abnormality in biochemical parameters including glucose 84 mg/dL, blood urea nitrogen of 5 mg/dL, creatinine 0.4 mg/dL, lactate dehydrogenase 240 U/L, sodium 138 mEq/L, potassium 4.6 mEq/L, chloride 102.4 mEq/L, calcium 9.7 mg/dL, phosphorous 4.3 mg/dL, amylase 46.6 IU/L, lipase 30.3 IU/L, total bilirubin 0.87 mg/dL, direct bilirubin 0.54 mg/dL, alkaline phosphatase 280 U/L, GGT 193 U/L, aspartate aminotransferase 21 U/L, and alanine aminotransferase 24 U/L. An upright plain abdomen X-ray revealed some air in the biliary system with no additional intestinal obstruction findings (Figure 1). Due to the character of the pain and excessive air in the biliary system, a computed tomography (CT) of the abdomen was subsequently performed. CT revealed a contracted and atrophic gallbladder with a few stones. Air was visualized within the gallbladder, common bile duct, and the intrahepatic biliary ductal system (Figure 2). In addition, the CT scan was unexpectedly suggestive of intestinal malrotation, with the colon located in the left abdomen, and the small bowel



Figure 1. Abdominal radiograph demonstrating air in the liver.



Figure 2. Contrast-enhanced CT demonstrates gas in the biliary system. Coronal reconstruction of CT scan showing reliable findings with malrotation, with the colon predominantly located in the upper abdomen and left flank, and multiple gas filled loops of the small bowel located in the right lower quadrant. No intra-abdominal free air and portal venous gas was seen.

located in the right side (Figure 2). The proximal duodenum did not cross the midline, and the region of the ligament of Treitz was located in the right upper abdomen, creating a mobile duodenum. An intestinal malrotation was consequently diagnosed; Figure 3 provides an illustrated schematic view.

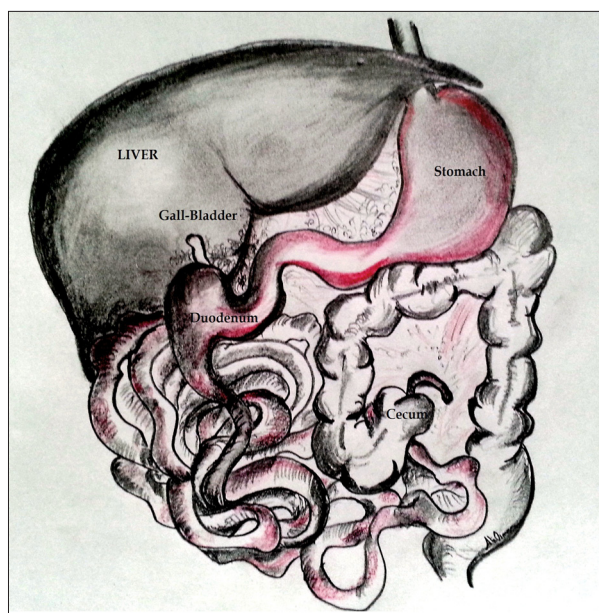


Figure 3. The schematic view.

After temporary improvement on intravenous antibiotics and fluids therapy, fever and the general condition recovered. On the fifth day a surgical treatment was scheduled. At first, a laparoscopic approach was attempted but was unsuccessful due to the adhesions in the region of the gallbladder, thus an open cholecystectomy was performed. Laparotomy and exploration revealed a malrotation such that the patient did not have a Ligament of Treitz. The duodenum did not run to the left side; instead, it descended to the right directly. Although the cecum and colon were loose and located on the left side, Ladd bands were not present. However, she had no vascular compromise. There were severe adhesions between gallbladder and duodenum. The gallbladder had contracted due to the cholecystitis and was atrophic, together with a fistula between the gallbladder and duodenum. Atrophic and adherent distal gallbladder was subtotally removed. The fistula was hardly separated, and was repaired at the same time. To avoid creating new problems, “Ladd’s procedure” and an appendectomy were not performed. But early in the post-op period (the third day), leakage from the repaired fistula was detected. Her abdomen started to fill with intestinal contents, and consequently, distention and peritonitis occurred. When the abdomen was re-explored, many intestinal adhesions were realized, and the fistula’s area was re-opened. After abdominal cleaning due to fistula leakage, it was restored. Two drains were placed in the abdominal and duodenal fistula’s area, and the surgical procedure was terminated. The patient’s post-operative course was not satisfactory, and the bilious contents of the bowels started to come out from the drain. With extra care, the patient was followed-up for 15 days, at the end of which the leakage stopped. She was discharged from the hospital with total recovery.

Discussion

Gallstone is a common disease in developed countries, with 10% prevalence. Complications of asymptomatic cholelithiasis are generally rare, with an incidence of <1% per year. The most common complications are acute cholecystitis, acute pancreatitis, ascending cholangitis, and gangrenous gallbladder. Less frequent ones include Mirizzi syndrome, cholecystenteric fistula, and gallstone ileus [4]. Several isolated cases have been described of cholecystocolonic fistula and gallstone ileus [5].

The cholecystenteric fistula was first described by Courvoisier in 1890. It is believed that these fistulas occur as a result of inflammation in the gallbladder due to cholecystitis [6]. However, other causes have been reported, such as cancer, trauma, amebiasis, peptic ulcer, and diverticulitis. It has been suggested that the pathophysiology of cholecystoenteric fistula involves the formation of a fistula as acute/chronic cholecystitis with stones causes inflammation and adhesions [7]. Nonetheless, the true causes of the fistula remain unknown, with continuing speculation [6]. In the present case, malrotation is considered to predispose to CDF. Although a CDF fistula thought lead to ascendant infection of the biliary system, they are rarely reported, and thus the patient is prone to high fever [8].

Evidence of air in the biliary tree on plain abdominal X-ray points to the presence of a bilioenteric fistula; still contrast radiology by barium meal, barium enema or both is essential for the definition of the fistulous connections [9]. However, MRI cholangiography is unconditionally selective in detecting CDFs.

Intestinal malrotation is a congenital anomaly that results from abnormal or incomplete rotation and fixation of the midgut during embryonic development. About 75% to 85% of these patients are diagnosed during infancy, whereas the diagnosis in the rest can be delayed to childhood or even to adulthood. The true incidence of malrotation is difficult to estimate in the older population since most remain asymptomatic [10]. It was reported that if the existence of malrotation is confirmed, surgical correction is compulsory to prevent volvulus and intestinal obstruction. In all cases, Ladd’s procedure with appendectomy is suggested. Nevertheless, exactly how to deal with different atypical presentations of intestinal malrotation will remain an enigma for diagnosis and treatment [3]. In the present case, a Ladd procedure was not performed due to the uncomplicated intestinal malrotation.

Different management strategies exist, with no agreed-upon best approach [6]. In the present case, a sub-total cholecystectomy was performed. With inflammatory conditions affecting the gallbladder and consequences of the veiled anatomy, dissection of Calot’s triangle can be difficult and it is unsafe to continue with a complete cholecystectomy. In such circumstances,

some authors propose a subtotal procedure for cholecystectomy. However, these patients often have high operative risk, as in the present case. Traditional operations help decrease the risk of cholangitis from colonic ascending bacterial translocation through the fistula into the biliary tree as the fistula is closed. This risk has been reported to be as high as 5% in experimental animal models. Furthermore, biliary sepsis in these patients is associated with 13% mortality. Despite all these factors, some authors support the patient's decision to choose observation alone [6]. When the recovery process is prolonged, we also think that non-operative management and clinical observation is an appropriate treatment.

Conclusions

No previous report has shown that rotational anomalies provide additional problems in cases of CDF, as in the present

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Acknowledgments and conflict of interest

Written informed consent was obtained from the patient for publication of this case report and accompanying images. The authors declare to no conflict of interests.