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Case Report

Biliary Peritonitis Due To Spontaneous Rupture of Type II Choledochal Cyst in an Adult

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Abstract

Rupture of Bile duct cyst has been widely reported in children. Although, it is very uncommon in adults few cases have been reported after trauma, pregnancy and intervention. Spontaneous rupture of Choledochal cyst (CC) is rare and spontaneous rupture of type II CC is unreported. The management of the CC during the ensuing biliary peritonitis is challenging. Excision of the cyst is recommended but may not always be appropriate in a sick patient with peritonitis. We report a case of spontaneous rupture of type II CC in an adult lady presenting with biliary peritonitis. Staged management comprising initial laparoscopic lavage and drainage of the peritoneal cavity followed by biliary stenting allowed for delayed elective resection without morbidity. Type II CC is infrequent and the high type seems to be most common. Ruptured type II CC presenting in adult with is extremely rare. Initial management of biliary peritonitis is of priority and the excision of the CC can be safely delayed till the patient is recovered.

Keywords: Biliary peritonitis; Bile duct cyst; Choledochal cyst

Introduction

In about 20% of cases, the presentation of Choledochal Cyst (CC) is delayed until adult life [1]. Adult CC are associated with a higher incidence of complications like stones, cholangitis, cirrhosis, and malignancy as compared to the pediatric age group. Although spontaneous perforation of CC has been reported in 2% to 18% of childhood presentations, it is an infrequent complication in adults [2]. Adult cases of CC rupture have been reported after trauma, during pregnancy, and after intervention [3-5]. Spontaneous rupture seems to be rare [6]. Choledochal cyst type 1 and type 4 are most common and are also the usual types reported with rupture. We report a rare case of a young female who presented with peritonitis due to perforated Type II CC.

Case Presentation

A 35-year-old female with no significant past medical history presented to the emergency department with complaints of non-bilious vomiting and general abdominal pain for 1 day. On inquiring, she also mentioned having vague upper abdominal pain for the last 6 years. Examination revealed maintained vitals with generalized abdominal tenderness and rebound suggestive of peritonitis. Labs showed leukocytosis (22.21 × 10^3/µL), mildly elevated total bilirubin (24 µmol/L). Abdominal Computerized Tomography (CT) revealed a well-defined cystic lesion in segment 4 of the liver measuring 11 × 9.5 × 15.5 cm with suspected rupture of the inferior aspect of the lesion and abdominopelvic ascites with enhanced peritoneal reflections. She underwent urgent Magnetic Resonance Imaging (MRI) of the liver, which confirmed a cystic lesion in segment 4 of the liver with rupture and perihepatic fluid (Figures 1 and 2). An impression of biliary cystadenoma with rupture was made, and the patient was planned for emergency laparoscopy. Surprisingly, at surgery, 3 liters of bilious fluid were drained from the peritoneal cavity. The cystic lesion was noted in segment 4, which seemed to be communicating with the left hepatic duct. Copious lavage of the peritoneal cavity was done with warm saline, a small part of the cyst wall near the rupture site was excised for pathology, and drains were placed in the peritoneal cavity, subhepatic space, and the cyst itself. Her postoperative period was uneventful. Two days after her surgery, she underwent Endoscopic Retrograde Cholangiopancreatography (ERCP), sphincterotomy, and stenting of both the right and left ducts. Pathology of the cyst wall was reported as an inflamed cyst wall with abscess formation possible, representing a choledochal cyst. By day 7 her drains were removed except for the cyst drain, which continued to have minimal bilious output. She was discharged on day 8 with the drain and planned for elective surgery.

4 weeks later, the patient was readmitted for left hepatectomy. CT abdomen prior to surgery redemonstrated the cyst, with a reduction in ascites and peritoneal inflammation. Open Left hepatectomy was performed through a midline incision. Her post-operative period was uneventful, and she was discharged by day 6. Pathology confirmed the diagnosis of Choledochal cyst communicating with the left duct. A final diagnosis of Type 2 CC from left hepatic duct complicated by rupture was made. Subsequently, she had removal of the biliary stents and MRI liver at 3 months, which was normal without any biliary dilation or strictures.

Discussion

Bile duct cysts are congenital biliary tract anomalies with a prevalence of 1 in 100000 to 1 in 150000 live births [7]. They are more frequently observed in Asians, especially in Japan [8]. The reason for Asian preponderance and higher incidence in females reported at 4:1 is still unclear [8]. The prevalence of Todani type II CC is difficult to determine and varies between 0.8 and 5% in large reported series of

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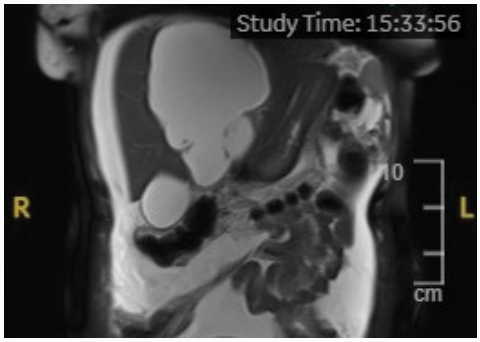

Figure 1: Pre-operative T2 Half-Fourier-Acquired Single-shot Turbo Spin Echo (HASTE) Coronal images of the segment 4 cyst and free peritoneal fluid.

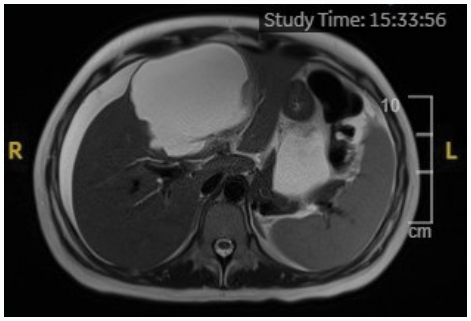

Figure 2: Pre-operative T2 Half-Fourier-Acquired Single-shot Turbo Spin Echo (HASTE) Axial images of the cyst relation to left hepatic duct and peri-hepatic fluid.

CC [9]. Contrary to popular belief, the most common site for Type II CC is at the level of the confluence of hilar ducts and above [9]. The European Multicenter Study which is the largest series of Type II CC defined 3 subtypes according to the location of the diverticulum in relation to the biliary tree [9]. Superior when located at the level of the main biliary convergence and common hepatic duct, middle when located at the level of the common bile duct, and low when located in the intrapancreatic portion of the extrahepatic bile duct. The study identified 58% of their 19 cases as being high level. None of the patients in the series presented with cyst rupture and biliary peritonitis. Rupture of CC in adults is as such rare. Most cases have been reported during pregnancy, postpartum, or after abdominal trauma [3,4,5]. Our case is unusual as it is possibly the first report of spontaneous rupture of superior type II CC in an adult. Since adult rupture of CC is very infrequent, there is no guideline for management. Most agree that surgical resection of the cyst is necessary. However, in a sick patient with biliary peritonitis, the risk of surgical complications may be too high [6]. Furthermore, the type of resection for Type II CC varies from excision of diverticulum to liver resection and Pancreaticoduodenectomy [9]. A simple excision of the middle subtype is probably safe during emergency surgery. On the contrary, a major biliary resection or hepatectomy may be fraught with complications and is better delayed. In this case we demonstrated that initial emergency management of the biliary peritonitis followed by endoscopic biliary drainage with delayed definitive surgery is a safe and effective option. It manages the acute emergency and allows definitive surgery in an elective controlled setting resulting in optimal outcomes.

Conclusions

This case reports a patient with spontaneous rupture of superior type II CC. It highlights the benefit of initial conservative surgical management with laparoscopic lavage and drainage to tide over the acute crisis followed by endoscopic sphincterotomy and stenting to decompression and temporary seal of the rupture. The benefit of elective surgery after resolution of peritonitis may be significant to reduce complications and improve long-term outcomes.

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