Case Report

Management of Spontaneous Perforation of the Bile Duct in an Infant in a Semi-Urban Setup: A Case Report

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Abstract

Spontaneous perforation of the extrahepatic bile duct leading to biliary peritonitis is a rare occurrence once other causes of biliary peritonitis, such as trauma, choledochal cyst, stone diseases, and distal atresia of the bile duct, are ruled out. A 7-month-old male infant was brought to the hospital in critical condition with distension of the abdomen. He had a history of vomiting and diarrhoea, low-grade fever, and refusal to feed for 2 days. Signs of peritonitis were found upon examination. Due to the poor general condition of the patient, the case was taken up for laparotomy, and a diagnosis of spontaneous extrahepatic bile duct perforation was made intra-operatively. In the present case, the cause was idiopathic. An external drain was placed near the site of the leak for 2 weeks. The patient recovered well and was discharged on post-operative day 16. Disease awareness for correct pre-operative diagnosis and interventional planning is required to reduce mortality, morbidity, and complications in spontaneous perforation of the common bile duct.

Keywords: *bile duct diseases, extrahepatic bile duct, gut, perforation, peritonitis, spontaneous rupture*

Introduction

Biliary peritonitis is a serious intra-abdominal emergency. Spontaneous idiopathic perforation of the non-dilated extrahepatic bile duct is a rare finding in infants. The condition, first described by Caulfield in 1936 (1), may present acutely without any sign of previous biliary tract disease. The aetiology of perforation of the common bile duct (CBD) leading to biliary peritonitis, although unclear, is thought to include increased intraductal pressure, calculus erosion, necrosis of the duct wall secondary to the thrombosis, stenosis of the duodenal papilla, and abdominal trauma (2). Several techniques have been applied for pre-operative diagnosis, yet most cases are diagnosed intra-operatively (3).

Case Report

A 7-month-old male infant was brought to the Aakash hospital with complaints of distension of the abdomen, vomiting, intermittent diarrhoea, refusal to feed, and low-grade fever for the past 2 days, for which he was treated by a paediatric physician before coming to the hospital. There was no history of direct or indirect trauma to the abdomen. Upon examination, the patient was found to be lethargic, irritable, and pale, with mild icterus but without any signs of dehydration. Breathing was laboured with a respiratory rate of 50 breaths per minute, and pulse rate was 170 per minute. The abdomen was distended and tense, with shiny skin displaying superficial veins. Upon palpation, the abdomen was tender and rigid; rebound tenderness and guarding were also present. The lab findings revealed anaemia (haemoglobin level of 9 g/dL), leucocytes (total leucocyte count of 12.3×10^9/L), and mild jaundice (serum bilirubin level of 1.5 mg/dL, serum alkaline phosphatase level of 321.0 IU/L, serum glucose phosphatase level of 31.4 IU/L). A plain X-ray of the abdomen, with the patient in an erect posture, revealed the absence of pneumoperitoneum. A paracentesis was performed, and the aspirated fluid was found to be bilious. Due to a lack of modern diagnostic techniques and the poor general condition of the patient, the case was undertaken for an emergency surgery. Approximately 600 mL of bilious fluid was aspirated, and the peritoneal cavity was mopped dry. Aspirated fluid was sent for laboratory examination and was found to be sterile. After exploration, a small biliary leak was found at the junction of the cystic duct and the CBD (supraduodenal part). Thorough peritoneal lavage was performed with a large volume of warm saline. An external drain was placed near

the site of the leak in the subhepatic space. Post-operatively, the patient was managed on adequate intravenous fluids and parenteral antibiotics (ceftriaxome sodium 100 mg/kg body weight, given intraperitoneally in 2 divided doses). On post-operative day 13, the drain ran nearly dry. The drain was removed on post-operative day 16. The patient was discharged with satisfactory recovery.

Discussion

Spontaneous perforation of the bile duct (SPBD) is a rare occurrence and most often seen in early infancy (2 to 20 weeks) with an almost equal sex ratio. However, a few cases have been reported in late infancy (4). The disease typically presents itself in previously healthy infants with unremarkable pre-natal and post-natal histories (5). Perforation commonly occurs in the anterior wall of the CBD near its junction with the cystic duct (4), as in our case, and is believed to occur due to congenital bile duct weakness, possibly due to a mural malformation during early embryogenesis (6). Eighty percent of cases present subacutely with fluctuating mild jaundice, normal to acholic stool, slowly progressive ascites, and abdominal distension, often associated with anoraexia, failure to thrive, fever, dark-coloured urine, and the development of umbilical, inguinal, or scrotal hernia. A history of biliary tract disease was absent; this absence of prodrome is notable and helps to distinguish SPBD from other causes of biliary peritonitis (7). Less commonly (20%), the disease presents acutely with abdominal distension of sudden onset, fever, vomiting, and signs of severe peritonitis (4).

In a number of cases, the diagnosis is made upon laparotomy, as in the present case, but can be easily substantiated pre-operatively (8).

Bilious ascitis, which is indicative of rupture of extrahepatic bile duct, can easily be diagnosed with the help of paracentesis (higher bilirubin level in the ascetic fluid than in the serum). Various investigations such as cholangiography, endoscopic retrograde cholangiopancreaticography (ERCP), magnetic retrograde cholangiopancreaticography, and biliary scintigraphy can reveal the site of the leak. ERCP has an additional interventional advantage, as stenting can be performed, and pancreatitis can simultaneously be ruled out (9). Early surgical intervention reduces mortality and morbidity (6). The aim of the treatment is to rule out distal obstruction and to establish adequate bile drainage. Various treatment modalities such as laparotomy with open drainage, percutaneous drainage, laparoscope guided drainage, and ERCP can be used, depending on the availability of the facilities, the presence of bile duct obstruction distal to the site of perforation, and the condition of the case. If the bile duct is obstructed distal to the site of perforation, T-tube insertion into the bile duct and open drainage can be performed with or without the help of a laparoscope. Alternately, ERCP with bile duct stenting may be performed (10). If the bile duct is patent distal to the site of perforation, any of the said modalities can be chosen, depending on the facility and the expertise available. Simple cholecystectomy can be performed if there is perforation in the cystic duct. In patients with biliary stricture, biliary-enteric bypass can be performed to avoid delayed sequelae, such as biliary cirrhosis and portal hypertension. Similar work has been reported in Northern region of India but in an urban, well-equipped setup (6,7,10). In rural or semi-urban setups, as in our case, where limited facilities are available, laparotomy with open drainage can safely be performed to save the patient's life. If the volume of drained fluid does not show a decreasing trend within 10 days, the patient may be shifted to a tertiary medical care centre, where ERCP and other modern facilities are available.

SPBD should be suspected in a patient presenting signs of peritonitis or bilious ascites, those without pneumoperitoneum (as determined by a plain X-ray of the abdomen taken with the patient in the erect position), and those without a history of biliary disease.

To conclude, disease awareness for correct pre-operative diagnosis and surgical treatment is central in reducing mortality, morbidity, and complications in SPBD patients.

Authors' Contribution

Conception and design: MJ, LS
Provision of patient, drafting of the article: SJ, MJ, DK
Collection and assembly of data: MJ, DK
Analysis and interpretation of the data: SJ, MJ, LS
Critical revision of the article: MJ, DK, LS
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