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Spontaneous Gallbladder Perforation in a Preterm Neonate

Abstract

Spontaneous gallbladder perforation among the pediatric population is a rare occurrence. The authors present a 26 week gestation white male with a history of Spontaneous bowel perforation and repair that presented with recurrent pneumoperitoneum. The patient was found upon laparotomy to have a perforation at the gallbladder fundus which was treated with tube cholecystostomy. A review of the current literature is provided.

Keywords: Gallbladder; Perforation; Neonate

Case Report

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Introduction

Spontaneous gallbladder perforation among the pediatric population is a rare occurrence. To date only 8 cases have been described in the English literature in neonates and infants [1-8]. Presenting signs and symptoms vary, as do treatment methodologies. Patients have presented with bilious drainage from peritoneal catheters [1] abdominal distention with discoloration [2] discolored communicating hydroceles [3, 4] gastric outlet obstruction [5] and acute abdomen [6]. We present a premature neonate with a history of spontaneous intestinal perforation that presented with pneumoperitoneum. At laparotomy he was found to have a perforation of the gallbladder fundus which was successfully treated with tube cholecystostomy.

Case Presentation

A 26 week gestation, 950 gm, white male was delivered after premature labor, resuscitated, and required emergent intubation due to respiratory distress. Deteriorating respiratory status, requiring hydrocortisone and oscillatory ventilation, complicated the first days of life. On day of life 7, the patient developed a distended abdomen with blue discoloration of the abdominal wall. Pneumoperitoneum was identified on abdominal x-ray. The patient was otherwise hemodynamically stable. He received emergent laparotomy demonstrating a spontaneous intestinal perforation of the mid jejunum that was treated with segmental resection and primary anastamosis.

On day of life 10 the patient's abdomen became distended and once again pneumoperitoneum was demonstrated on abdominal x-ray. Exploratory laparotomy revealed bilous ascites. The bowel was examined from the gastro esophageal junction to the rectum and confirmed an intact small bowel anastomosis from the previous surgery without additional intestinal perforations. The right upper quadrant was examined revealing a perforation at the fundus of the gallbladder. The perforated segment was excised and sent to pathology. No gallstones were identified and a cholecystostomy tube was placed. Pathology reported necrotic and hemorrhagic tissue of the gallbladder fundus.

On day of life 13 the abdomen became distended and pneumoperitoneum was again demonstrated by abdominal x-ray. Emergent laparotomy revealed a second spontaneous bowel perforation at the ligament of Trietz. Due to the extremely proximal location of the small bowel perforation, we opted to repair the intestine primarily. The patient's condition continued to improve over the following month. The cholecystostomy tube was removed on day of life 32 without complication and he was later discharged home after 3 months in the neonatal intensive care unit.

Discussion

Extra hepatic biliary perforations in neonates and infants typically occur along the common bile duct. Several etiologies have been described in the literature and range from anomalous union of the pancreatico-biliary ductal system, congenital weakness of the ductal system, choledochal cysts and trauma [9]. Gallbladder perforation is rare and has been shown to result from trauma, gallstones, distal obstruction, and typhoid fever [9]. The fundus is the most common site of perforation and is suspected to result from local ischemia from a tenuous blood supply. Various locations along the extra hepatic biliary tree have been reported in the English literature as potential sites of perforation (Table 1). Our case describes a unique complication of multiple spontaneous bowel perforations and a gallbladder perforation. Spontaneous intestinal perforation has been linked to steroid use and local ischemia [10]. We suspect that the etiology of the multiple intestinal perforations and the gallbladder perforation were secondary to the use of hydrocortisone in this patient, which has not been previously reported as a complication in this population. Though there have not been studies evaluating spontaneous intestinal perforation with gallbladder perforation, we suspect the risk factors may be similar. Steroid administration and in particular hydrocortisone, has been shown to decrease mucin production in the gallbladder thereby removing the protective coat for the gallbladder epithelium against bile [11,12]. Coupled with prematurity and periods of asphyxia which can decreases splanchic blood flow may contribute to gallbladder perforation [2]. Thermal injury

from previous surgery is also a possibility, though we feel this is less likely.

Radionuclide hepatobiliary imaging [7], CT scan [3], ultrasound and paracentesis [4] have all been used as adjuncts to laboratory tests to assist in the diagnosis of gallbladder perforation in neonates. Despite this, the discovery of spontaneous gallbladder perforation is most often made at the time of surgery [1-2]. Pneumoperitonium, as was found in our case, has not been previously described as a diagnostic modality for gallbladder perforation in the neonatal population. The presence of gas forming bacteria may offer an explanation for the pneumoperitoneum however this patient's cultures were negative.

The reported management of spontaneous perforation of the gallbladder among infants and neonates include cholecystectomy [1], cholecystectomy with hepaticojejunostomy [7], primary repair [2] and partial cholecystectomy with external drainage [3,4]. Sharma et al. [4] has suggested surgical intervention with the aim of external biliary drainage and routine intraoperative cholangiogram to evaluate for distal obstruction [4]. We performed tube cholecystostomy after removing the perforated segment of the gallbladder. We abstained from cholecystectomy due to the marked inflammation at the porta hepatis which is also supported in the literature [3,4]. We found this treatment to be successful and decrease the risk of injury to the porta hepatis and surrounding structures.

Table 1: Summary of present literature.

Article	Age	Sex	Presentation	Gallstones	Perforation	Treatment
Ying-Yi et al. [1]	60 day	Male	Bile drainage from peritoneal dialysis tube	No	Gallbladder neck	Cholecystectomy
Gull et al. [2]	4 day	Male	Abdominal distension, green discoloration to the abdomen, tenderness	Not stated	Gallbladder neck	Primary repair with drain in the sub hepatic pouch
Rhoads et al. [3]	3.5 month	Male	Jaundice, bile peritonitis, bilateral inguinal hernia	Yes	Fundus to infundibulum	Drain at the porta hepatis
Sharma et al. [4]	3 month	Male	Abdominal distension, fever, acholic stool, bilateral scrotal swelling with green discoloration	Not stated	Gangrenous**	Partial cholecystectomy with drain at the remnant
Nambirajan et. al. [5]	10 day*	Female	Gastric outlet obstruction	Not stated	Gallbladder neck	Cholecystostomy tube
Shukla et al. [6]	4 month	Male	Acute abdomen	Not stated	Perforation **	Cholecystectomy
Sharma et al. [7]	2 month	Female	Acholic stool, jaundice, abdominal distension	Not stated	Gallbladder fundus	Cholecystectomy with hepaticojejunostomy
Snyder et al. [8]	6 week	Female	Constipation, abdominal distension	Yes	Junction of gallbladder to cystic neck	Cholecystostomy tube
Presented case	10 day	Male	Pneumoperitoneum	No	Gallbladder fundus	Cholecystostomy tube

Conclusion

Spontaneous gallbladder perforation in infants and neonates is a rare occurrence. The etiology is not yet clear and further research is warranted. Multiple modalities have been used to assist in diagnosis yet most diagnoses are still made at the time of laparotomy. Although treatment options vary depending on the site of perforation, we recommend biliary diversion by cholecystostomy for perforations occurring on the gallbladder to avoid injury to the porta hepatis. Postnatal steroid use in this population should be used with caution and one must have a low index of suspicion for intra-abdominal complications.

Conflict of Interest

There are no conflicts of interest.

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