CASE REPORT

Acute spontaneous perforation of the common bile duct in a toddler

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Introduction

Acute spontaneous perforation of the common bile duct (CBD) causing an acute abdomen in a previously well infant is extremely rare. Only few case reports of this potentially fatal condition are recorded in the literature. The majority of affected children are, aged between 6 months and 7 years with a female preponderance [1,2,3,4]. Identified aetiological factors include congenital biliary tree anomalies, blunt abdominal trauma, distal CBD obstruction and viral infections. Some perforations are of idiopathic origin [1,5].

Spontaneous CBD perforations are of acute (20%) or insidious (80%) origin. Acute spontaneous CBD perforations may present with features of an acute abdomen and shock resulting from biliary peritonitis. In such emergencies, appropriate emergency surgery is life-saving [1,2,3]. Perforations of insidious onset are characterised by progressive jaundice, painless abdominal distension and acholic stools [1, 2].

In stable children with slow biliary leaks, CT/MRI scan or, MRCP would be valuable in deciding on the mode of interventions such as; image-guided biliary stenting and suturing of the defect [1, 2, 3, 4].

Case study

One year and four months old previously healthy girl was admitted with a one day history of non-bilious vomiting, lethargy and poor sucking with no history of abdominal trauma or ill health. She was haemodynamically unstable having tensed abdomen and absent bowel sounds. Since clinical features were suggestive of intestinal perforation with hypovolaemic and septic shock, resuscitation was commenced.

X-ray abdomen showed 'white halo' like outline of bowel wall which is suggestive of free fluid in the abdomen. Emergency Ultrasound Scan of abdomen by consultant radiologist identified moderate ascites and sluggish bowel movements. Sonography did not identify any pathology in the gallbladder and biliary tree. Rest of viscera was sonographically normal. Full blood count and electrolytes were unremarkable.

Figure 1 : Pre operative supine X-ray abdomen

After resuscitation, an emergency standard right upper transverse laparotomy was made. On entering the peritoneal cavity, around 200ml of greenish ascitic fluid, indicative of diffuse biliary peritonitis found. No intestinal perforation identified. After peritoneal lavage, about 4mm long, a fairly fresh perforation was identified in the medial aspect of the CBD, approximately 2mm inferior to the union of the cystic duct with the CBD. Liver, spleen and other abdominal viscera appeared normal.

Repair of CBD perforation over a T tube/ stent was planned but, a suitable size stent was not available at this newly established paediatric surgical unit. A cholecystostomy catheter drain insertion was therefore selected. CBD perforation repaired with 6/0 interrupted PDS sutures. Gallbladder mobilised to facilitate attachment to abdominal wall. Size 8 Foley's catheter inserted as cholecystostomy drain.

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Patency of the CBD repair was confirmed by injecting saline through the cholecystectomy drain. Free flow of saline downwards CBD noted excluding distal CBD obstruction. The anatomy of the biliary tree and macroscopic appearance appeared normal. A tube drain was inserted to the hepatorenal pouch and standard laparotomy closure was performed.

She was observed for 2 days in the intensive care unit. She made an uneventful recovery. With reduction of cholecystostomy catheter drainage and normal coloured urine and stools, gradual oral feeding commenced.

USS abdomen on post-operative day 14 was normal. Contrast study via cholecystostomy catheter drain confirmed the integrity of the CBD repair and free flow of contrast down the biliary tree.

Figure 2: Postoperative contrast study

The cholecystostomy drain was removed on post-operative day 14. An ultra sound scan of the abdomen, performed 24 hours later was normal. Abdominal drain was removed on post operative day 15. With the establishment of regular feeding, she was sent home 18 days after the laparotomy. USS abdomen performed during paediatric surgical clinic follow-up at 1 week, 1 month, three months and six months after discharge were normal.

About one year after laparotomy, she was hospitalised with an episode of bilious vomiting with clinical and x-ray evidence of adhesive intestinal obstruction. At laparotomy a fibrous band obstructing mid-ileum was identified and separated. Integrity of the CBD was found to be normal. Baby made an uneventful recovery and remains well nearly two years after the laparotomy for adhesive intestinal obstruction.

Discussion

Common clinical features of infants with acute spontaneous perforation of the CBD such as nausea, vomiting, fever and abdominal distension are similar to clinical features of intestinal perforation [1,4]. In very sick children having an acute abdomen presenting to resource-poor settings as in this baby, performing pre-operative diagnostic tests such as CT/MRI scans is difficult. Radiological diagnosis is often limited to x-ray and an ultrasound scan abdomen.

Emergency laparotomy was performed in this baby based on clinical features such as a tense abdomen with absent bowel sounds, x ray findings of small bowel loops with a white halo usually seen with ascites, and USS indicates ascites and sluggish bowel movement.

The mid and distal extra-hepatic biliary tree is supplied by the branches of the posterior superior pancreaticoduodenal artery(around 60%) and hepatic artery(around 38%). Both vessels run along the course of the CBD. The area of demarcation of their blood supply lies at the region of union of the cystic duct and the CBD. This anatomical region is considered as congenitally weak and more prone to perforations [1,5].

In acute spontaneous CBD perforations, surgical management would depend on the child's clinical status. Acute biliary leaks may rapidly progress to biliary peritonitis and irreversible shock necessitating emergency surgical intervention. Recommended surgical options for visible CBD perforation at laparotomy include: CBD repair over a T tube or, CBD repair with a cholecystostomy drain when a suitable stent is not available [1, 2].

Literature describes few case appropriate surgical interventions in the management of this rare pathology [1, 2, 3, 4). Some case reports on stable children with CBD perforations have described initial laparotomy/laparoscopy and intra-operative cholangiography. Some have performed subsequent MRCP followed by ERCP and image-guided percutaneous stenting.

When CBD perforation is found to be sealed off at laparotomy, peritoneal lavage and placing of an abdominal drain alone is an option.

Clinical stability and the passage of urine and stools of normal colour and consistency after surgery indicates free biliary drainage. Integrity of extrahepatic-biliary tree anatomy and exclusion of distal CBD obstruction was confirmed by

contrast study via cholecystostomy catheter in this baby. Her follow up Ultrasound scans of the abdomen were normal. The post operative adhesive intestinal obstruction would have resulted from initial peritoneal contamination.

Conclusion

Managing acute surgical abdomen in previously healthy babies in resource poor surgical settings is challenging. In such emergencies, performing laparotomy based on clinical and available radiological imaging such as X-ray and USS can be regarded as safe practice.

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Learning Points:

- Pre-operative accurate diagnosis of rare abdominal pathologies such as acute spontaneous CBD perforation is difficult.
- In resource poor settings, case appropriate emergency laparotomy and corrective surgery would save life.