Spontaneous perforation of common bile duct: a rare complication of choledocholithiasis

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Abstract A 35-year-old female presented with 48 hrs history of severe upper abdominal pain and abdominal distension. The patient was suffering from intermittent episodes of colicky upper abdominal pain and spiky fever with chill and rigor in association with yellowish discoloration of eyes and urine for last 3 months. On examination the lady was dehydrated, icteric, febrile and was having tachycardia and hypotension. Abdominal examination indicated features of generalized peritonitis. Hematological profile suggested neutrophilic leukocytosis, blood biochemistry suggested conjugated hyperbilirubinemia, raised alkaline phosphatase and transaminases. Skiagram of the abdomen showed ground glass opacity without any free gas under the diaphragm. Ultrasonography and contrast enhanced CT scan of the abdomen revealed multiple calculi within a distended, thick walled gall bladder, dilated common bile duct (CBD) with a 12.8 mm stone impacted at its lower end and free fluid in the lesser sac and rest of the peritoneal cavity. Diagnostic abdominal paracentesis showed heavily bile stained fluid. Exploration done for generalized biliary peritonitis with sepsis, revealed erosion at the posterior aspect of the retro-duodenal CBD over the site of the impacted stone. She underwent cholecystectomy, choledocholithotomy through a supra-duodenal choledochotomy, and T-tube drainage of the CBD. She made a slow but steady post operative recovery and was discharged from the hospital after 22 days in favorable conditions.

Keywords Bile duct · Perforation · Cholelithiasis · Surgery.

Introduction

Generalized biliary peritonitis following perforation of some portion of the biliary tract is commonly a sequel of rupture of an acutely inflamed gangrenous gall bladder [1]. Spontaneous perforation in the wall of the extra hepatic or intra-hepatic bile duct with biliary peritonitis, without any traumatic or iatrogenic injury, is an extremely rare event in adults. It was first described by Freeland in 1882 [2]. However, it is relatively more common in children [3]. Though termed “spontaneous”, it is mostly seen in association with choledocholithiasis in adults and the site of perforation is frequently extra-hepatic. The presentation of biliary peritonitis varies and because of its rarity the correct pre-operative diagnosis is often difficult and delayed. This along with the associated co-morbidity of the mostly elderly, patients in whom it is seen, results in a mortality rate of 30–50% [4].

We report a successfully managed, rare case of spontaneous extra hepatic bile duct perforations with choledocholithiasis, in a young adult female. Its clinical presentations, investigative findings and management are discussed and relevant literatures reviewed. The rarities of this case are the atypical site of CBD perforation and its occurrence at a relatively young age.

Case report

A 35-year-old female presented at the surgical emergency with 48 hrs history of severe upper abdominal pain and gradually increasing abdominal distension. For last 3 months she had been suffering from episodic colicky upper

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abdominal pain in association with yellowish discoloration of eyes and urine which fluctuated in its intensity, and spiky fever with chill and rigor. The abdominal pain increased in intensity, became continuous and more diffuse for the last two days. On examination, the patient was anxious, dehydrated, icteric, febrile (101° F) and was having a pulse rate 110/minute and blood pressure of 90/70 mm of Hg. The abdomen was distended with guarding and rebound tenderness mainly in the upper abdomen. There were evidences of free fluid in the abdomen. Liver dullness was not obliterated. Bowel sounds were absent.

Hematological profile revealed a total leucocyte count of 21,600 cells/mm³ (93% neutrophil). Total bilirubin 4.5 mg% (conjugated 2.8), alkaline phosphatase 220 IU/L (N: 35–130), transaminase 210 IU/L (N: 5–40 IU/L). Serum amylase and lipase levels were normal and so were the renal function parameters.

Diagnostic abdominal paracentesis revealed heavily bile stained peritoneal fluid. Straight x-ray of the abdomen (erect) did not show free gas under the diaphragm. Ultrasonography of the abdomen suggested free fluid in the peritoneal cavity, mildly enlarged liver with dilated intrahepatic biliary radicals. Gall bladder was distended and thick walled with multiple echo reflective calculi; common bile duct was 16 mm in diameter with a 21.8 mm calculus impacted at its lower end.

Contrast enhanced CT scan of the abdomen suggested cholelithiasis, choledocholithiasis, dilated intra and extra-hepatic biliary radicals, fluid collection in the lesser sac, ascites, omental thickening and mild bilateral pleural effusion (Fig. 1). The clinical and investigative findings (absence of free gas under diaphragm and distended gall bladder) excluded the possibilities of the two common causes of biliary peritonitis namely peptic perforation and gall bladder perforation. Definite preoperative diagnosis was not possible.

The lady underwent an exploratory laparotomy, after resuscitation and under broad spectrum antibiotic coverage, for biliary peritonitis with sepsis. Operative findings included more than 2 liters of bilious peritoneal fluid with fibrinous exudate accumulated within the lesser sac and other dependent parts of the peritoneal cavity. Gall bladder was distended, thick walled and contained multiple stones. No perforation was apparent either in the gall bladder or the gut wall. Kocherization of the duodenum confirmed the presence of an irregular, firmly impacted stone with a pointed end within the retro-duodenal portion of the CBD and also revealed bile trickling out from 3 mm erosion on the posterior wall of the duct near the pointed upper end of the stone (Fig. 2).

Cholecystectomy, choledocholithotomy through a supra duodenal choledochotomy was performed. Operative cholangiogram excluded any distal obstruction and the bile duct was drained by a T-tube. No attempt was made to repair the perforation because of its thinned out margins, friability and presence of infection. Abdomen was closed after thorough peritoneal toileting with a sub-hepatic drain.

Fig. 1 CT scan of the abdomen showing distended gall bladder with calculi, dilated CBD and cystic duct with calculus, encysted collection in the lesser sac and ascites

Fig. 2 Shows a probe passed through the perforation in the posterior wall of CBD via the anterior choledochotomy

The patient made a slow postoperative recovery. Drainage of bile through the sub-hepatic drain stopped after 8 days. T-tube cholangiogram performed on the 20th post operative day showed no filling defects or spillage of dye and its free flow into the duodenum. T-tube was removed after clamping for 24 hours. Patient was discharged from hospital on the 22nd day in favorable condition and was doing well at the time of the first follow up visit after 2 weeks.

Discussion

Spontaneous perforation of the bile duct means perforation of any portion of the extra-hepatic or intra-hepatic bile duct without any traumatic or iatrogenic injury. It is a rare entity and described mainly in infants due to congenital weakness of the CBD seen in conditions like choledochal cyst [3]. It is extremely rare in adults with less than 90 cases reported in English literature [1, 5–7]. Most cases are not “spontaneous” in the true sense but secondary to some underlying pathology. 70% of the reported cases in adults are associated with ductal stones [8]. It has been rarely reported in adults

in association with pregnancy, [8] acalculous cholecystitis, [9] choledochal cyst [10]. Some other interesting co-morbid conditions and diseases reported to be associated with spontaneous bile duct perforation are HIV infection and Hodgkin's lymphoma, [11] tuberculosis, [12] and severe necrotizing enterocolitis involving the duodenum, caused by paracolon bacteria – providence alcalifaciens, with spontaneous complete avulsion of the CBD [12].

Other still rare causes are phytobezoars in CBD in patients with previous chledochooduodenostomy [5] or weakness at the stomal site in patients with previous choledochoduodenostomy [6].

The site of perforation is nearly always extra-hepatic though a few cases of intra-hepatic ductal rupture have also been reported in association with calculous disease, stenosis of the papilla and Caroli's disease [1]. Commonest site of extra-hepatic biliary perforation (excluding gall bladder) is the junction of the cystic duct and CBD / hepatic duct [1].

Though the pathogenesis of the spontaneous perforation of the bile duct in adults is not clearly established it has been suggested that any condition causing increase in intra-ductal pressure like choledocholithiasis causes dilatation of the bile duct, infection and thrombosis of the intra mural vessels leading to local infarction and perforation.⁵ Multiple factors like stasis, infection, inflammation and gradual pressure necrosis of the duct wall are responsible for the perforation occurring in patients with large impacted stone, as in this reported case. Slow erosion by a calculus often leads to fistula formation, but the posterior location of the perforation in this reported case prevented this sequel but explains the notable amount of collection in the lesser sac. Weakened and dilated extra-hepatic duct common in advanced age explains the predilection of such perforations among the elderly.

Obstruction of the bile duct due to tumor generally do not cause perforation because of the slow rise of intra ductal pressure in contrast to the sudden increase that occurs in stone disease [5].

The presentations may be acute with features of infective peritonitis as in this reported case or more commonly insidious with jaundice, clay colored stool and abdominal distension [6].

In the unlikely event of this condition being suspected preoperatively, a hepatobiliary scintigraphy with iminodiacetic acid showing the extravasation of bile in conjunction with an ultrasonography could make a precise preoperative diagnosis. Magnetic resonance cholangiopancreatogram (MRCP) is a useful investigation in the diagnostic armamentarium. However, in the reported case MRCP was not advised as the ultrasonography and CT scan had suggested a dilated CBD and choledocholithiasis and MRCP was unlikely to add any extra information. MRCP can also be useful to assess the intra and extra-hepatic biliary tree postoperatively prior to the T-tube removal but obviously a costlier choice than a T-tube cholangiogram. Endoscopic retrograde cholangiopancreatogram (ERCP) can also diagnose the presence of an obstructing calculus and the site of the leak. Intra-operative cholangiogram before T-tube inser-

tion is imperative to evaluate the condition of the bile duct and exclude distal obstruction [13].

Endoscopic stone retrieval and stenting / sphincterotomy followed by laparoscopic cholecystectomy, peritoneal toiletting and drainage are alternatives which were not tried in our patient because of the large size of the stone. It can be considered as an alternative in patients unfit for surgery. Laparoscopic common duct exploration, T-tube drainage and cholecystectomy have also been described in similar conditions [7].

Awareness of this entity as a rare cause of biliary peritonitis avoids undue delay in the diagnosis and thus improves prognosis. Our patient who was relatively young underwent prompt operative intervention after resuscitation, before gross biliary / systemic sepsis could supervene and this played a major role in the positive outcome for this patient.

Conflict of interest The authors do not have any disclosable interest

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