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Case Reports

Spontaneous Perforation of Extra Hepatic Bile Duct in a Child

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Abstract Spontaneous perforation of biliary tree is rare. It mostly occurs in infants. Most cases are associated with biliary stone or other factors related to the bile flow. We report a case in six-year-old girl, hailing from eastern part of India, with SBP without any predisposition. She presented with acute abdomen for three days. Investigations revealed free fluid in abdomen but no free gas under diaphragm. An exploratory laparotomy showed perforation at the supraduodenal portion of common bile duct. No stone/stricture or any other apparent cause of the perforation was found. It was repaired over T-tube and abdomen was drained. Postoperatively the patient made an uneventful recovery. T-tube cholangiogram done on day fourteen was normal and the tube was removed. At one year, the patient is doing well. We include a review of the literature with emphasis on the management of this rare condition.

Key words Bile duct perforation; Biliary peritonitis; Idiopathic common bile duct perforation; Spontaneous bile duct perforation

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Introduction

Spontaneous biliary perforation (SBP) is rare. If trauma and choledochal cysts are excluded, stones are the most commonly associated factor. The first case described was in 1932. Since then fewer than 150 cases have been reported, mostly in infants. The commonest site of perforation is at the junction of the cystic and common bile duct (CBD) anteriorly. We report an unusual case of supraduodenal SBP in a six-year-old. A literature review is included.

Case Report

A six-year-old girl was admitted with three-day history of abdominal pain and distention. No history of hepato-biliary disorder or trauma was noted. She was anicteric, febrile, tachycardic and tachypneic. The abdomen was distended with evidence of guarding, free fluid and paralytic ileus.

X-ray abdomen did not show any free gas. Ultrasonography confirmed free fluid in abdomen. On

exploration one liter of free bile was drained. Her gall bladder was normal with no stones. A 0.5-centimeter perforation at the supraduodenal part of CBD anteriorly was found. Intraoperative cholangiogram was not available but meticulous manual exploration of biliary tract showed no stone, obstruction or cystic dilatation and normal pancreas. As there is a high prevalence of worm infestation in this part of the world that can precipitate cholangitis and biliary obstruction, a thorough search for parasites in the intestinal and biliary tract was performed with negative result.

The perforation was closed over number twelve French T-tube. Tissue from bile duct wall was sent for biopsy and a drain was placed in the hepato-renal pouch. Postoperatively, patient made a rapid recovery. Her liver function test, amylase, and lipase were normal and apart from neutrophillia that returned to baseline in a few days, her blood count was unremarkable. T-tube cholangiogram obtained two weeks postoperatively showed free flow of bile into duodenum with no stones, obstructions or anomalous pancreatobilliary junction (Figure 1). Histopathology study did not show any evidence of tuberculosis or any chronic infective/granulomatous/neoplastic aetiology. An Ultrasonography at three months showed normal hepatobiliary and pancreatic anatomy and repeat liver function test, amylase, and lipase were normal.

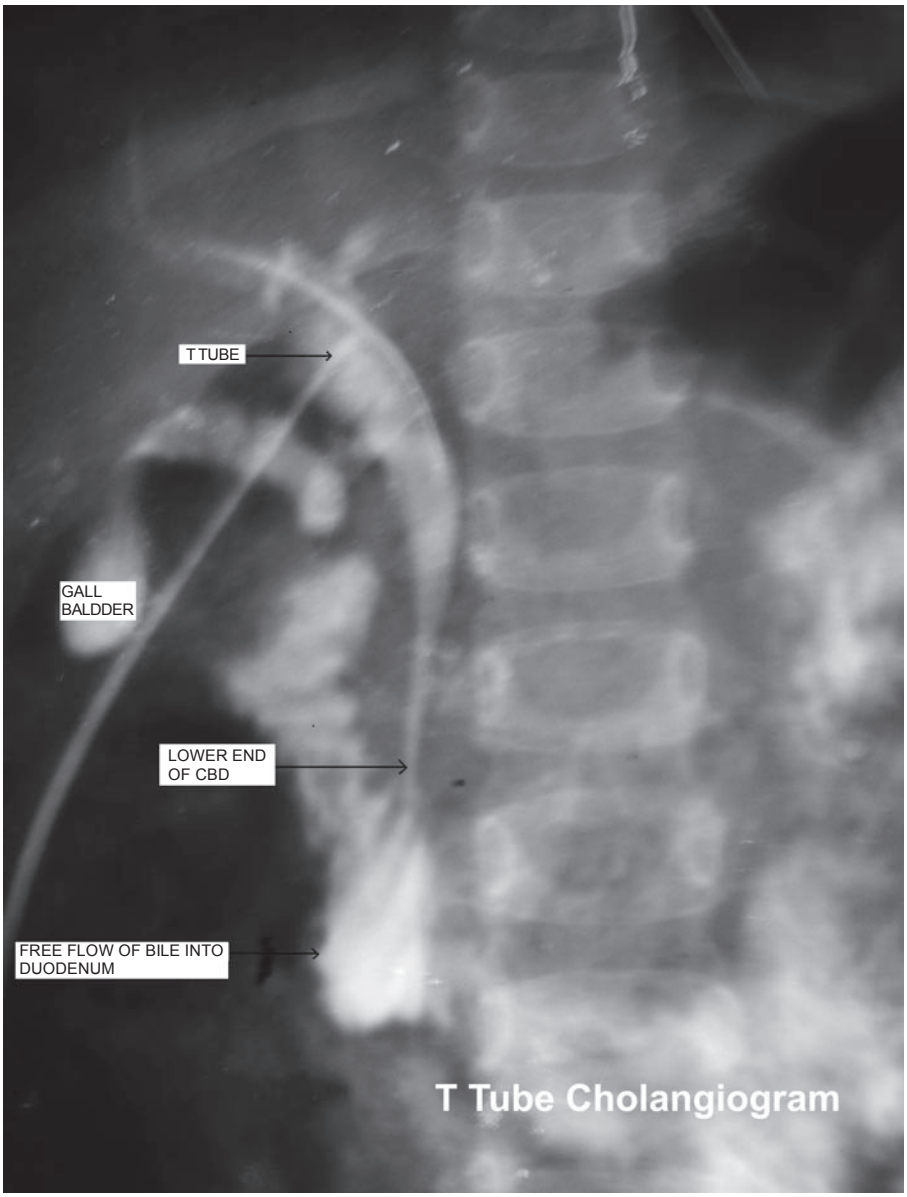

Figure 1 T-tube cholangiogram.

Discussions

It is difficult to define the etiology of the billiary perforation here. Most reports in the paediatric literature are about infants. Congenital mural weakness at the junction of the cystic duct and CBD makes it the commonest site. Transient increase in intrabiliary pressure causing a "blowout" is a possibility. Congenital embryonic weakness or malformation of the bile duct wall and ischemia of CBD because of thrombosis in the microcirculation are also put forward as etiology. Associated factors include inspissated bile, necrotising entero-colitis in the context of neonatal susceptibility to ischemia, distal bile duct stenosis in a preexisting choledochal malformation and anomalous pancreatobilliary junction (APBJ) with long common channel causing pancreatitis.[1,2] In cases of posterior perforation of bile duct or hepatic duct, the diagnosis of the exact site is difficult. Patients with AIDS or lymphoma on chemotherapy can also develop SBP.[3]

Adult SBP is rarer. A Korean report identified 11 adults with nontraumatic bile duct perforation largely due to gallstones obstructing the CBD but examples of a perforated choledochal cyst and an obstructing bile duct phytobezoar arising as a complication of choledocho-duodenostomy are also described.[4]

The clinical presentation is usually insidious with increasing abdominal distension due to biliary ascites, mild jaundice and pale stool. Acute presentation without jaundice is rarer. The triad of bilious abdominal paracentesis, peritonitis and absent free gas on X-ray indicates SBP.[5] Billiary scintigarphy is a good investigation in subacute cases with jaundice.

If distal billiary obstruction is excluded, simple drainage of bile and optional sphicteroplasty with or without primary closure of peroration is also an option. Bilio-enteric diversion is advised especially where perforation is large or distal obstruction cannot be corrected.[6] Only ERCP (endoscopic retrograde cholangio-pancreatogram) with sphincterotomy with or without stenting, as is sometimes done with traumatic billiary tract injuries, is not advisable. External or internal drainage procedure should be added to it. We did not have intraoperative cholangiogram facility and so we chose the safer option of repair over T-tube. We did not find intraoperative CBD exploration harmful as suggested in some papers.[7]

One case reported eosinophilia associated with SBP.[8] In our case parasitic infection especially roundworm as a contributory factor was ruled out by meticulous search in the intestine and biliary tree. In addition, biopsy proved negative for tuberculosis or any granulomatous disease.

Portal vein thrombosis and chylous ascitis are known complications especially in posterior peroration.[9] Our patient, now in one-year postoperative follow-up is doing well.

In our case, no associated cause of SBP was found. We can hypothesize stone impaction in CBD with perforation and stone passing out, but the short history and normal gall bladder make it unlikely. Although we did not do a measurement of amylase or trypsin in the bile to rule out reflux of pancreatic enzyme into the bile from APBJ, the normal postoperative choloangiogram without any reflux into pancreatic duct was assuring. Biopsy from the perforation site also did not show any degenerative changes typically seen in pancreaticobiliary malformations. As there was no further history of abdominal pain and amylase level done at three months was normal, abnormally long common channel as a cause of peroration was less likely. In contrast study bile duct showed slight dilatation. With T-tube in situ, it is not unusual. Ultrasound at three months showed normal anatomy of the area ruling out suggestion of choledochal cyst. To completely rule out APBJ that is commonly identified as the cause of SBP in Oriental patients, ERCP (endoscopic retrograde cholangiopancreatogarm) or MRCP (magnetic resonance cholangiopancreatogram) would have been helpful. In this particular case, normal amylase and ultrasonography at three months follow-up and no symptom even after a year of clinical follow up, weighed against the small but definite risk of complications from ERCP, and the cost factor of MRCP. Repair over T-tube is safe and recommended after dealing with precipitating factors if present. We did not do cholecystectomy, as gall bladder was normal.

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