Biliary Ascites Caused by Perforation of Choledochal Cyst

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Choledochal cyst is no more considered a rare surgical entity with about 3000 cases already been reported in world literature(1). However, perforation of choledochal cyst is rare with only 56 cases recorded till date(2-17). The complication was first described by Weber in 1934(2). We report a case of biliary ascites caused by perforation of choledochal cyst in an infant.

Case Report

A 5-month-old male infant was admitted with abdominal distension of three weeks duration. The onset was accompanied with few episodes of non-bilious vomiting and sudden appearance of bilateral scrotal swellings. The baby had been passing acholic mucoid stools since two weeks. Physical examination revealed a pale, malnourished, anicteric infant having massive ascites and bilateral hydroceles. Abdominal paracentesis revealed free flow of bile.

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Initial investigations revealed low hemoglobin (6.4 g/dl) and deranged liver function tests (total protein 4.8 g/dl; serum albumin 3.6 g/dl; total bilirubin 2.6 mg/dl; direct bilirubin 1.68 mg/dl; and alkaline phosphatase 805 units/dl; GGT 208 units/1). Ultrasonography of the abdomen revealed a cystic swelling in the region of common bile duct measuring 3 x 3 x 1 . 7 cm in size; gall bladder was contracted and liver was normal. The abdominal paracentesis catheter drained 750 ml of bile over next 12 hours. The presence of bile salts and bile pigments were confirmed; total and direct bilirubin levels of the ascitic fluid were 16.6 mg/dl and 9.8 mg/dl, respectively. Bile was sterile on culture.

After initial resuscitation, the child was taken up for laparotomy which revealed biliary peritonitis due to a 5 mm wide perforation in the anterior wall of the choledochal cyst. In view of the acute inflammation of tissues, it was decided only to decompress the choledochal cyst with a T-tube through the perforation. Intra-operative cholangiography revealed fusiform dilatation of common bile duct. Intrahepatic biliary radical were normal. No dye was seen going down to the duodenum *(Fig. 1). A* tube drain was also put in the Morrison's pouch, and lesser sac, this was removed on third postoperative day. The child was discharged on fifth postoperative day. T-tube drained 100-200 ml of bile every day. The child continued to pass acholic stools. On 18th postoperative day, the patient was readmitted with acute intestinal obstruction. Laparotomy revealed multiple postoperative adhesions which were lysed. The postoperative period was uneventful.

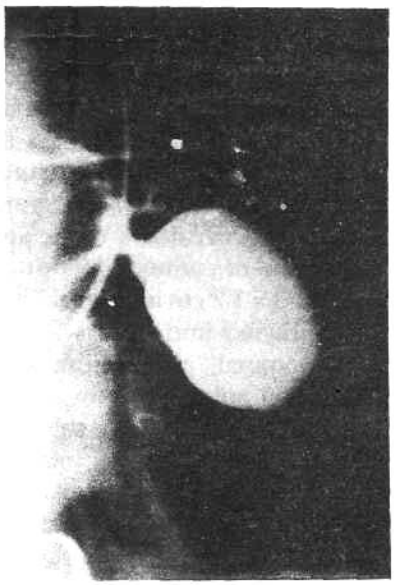

Fig. 1. Intra-operative cholangiogram demonstrating fusiform type of choledochal cyst with complete obstruction at the distal end. The intra-hepatic biliary radicals were normal.

The definitive surgery for choledochal cyst was performed after another 3 months, complete excision of choledochal cyst alongwith hepatocodochoduodenostomy using isolated jejunal conduit was performed. Posoperatively, the child developed biliary leak which stopped spontaneously by 12th postoperative day. Meanwhile the child was kept nil orally and was given parenteral nutrition. The child was discharged on 14th postoperative day and has remained well.

Discussion

Perforation of the choledochal cyst is a rare complication with its reported incidence being around 2%(l,10,ll). More than half of the cases have been reported from Japan(10-13). Although no definite cause could be ascertained in a majority of cases, trauma was responsible in a few(4-6). Perforation has been also reported to occur during pregnancy(7,16,17). Anomalous choledochopancreatic duct junction has been implicated in few cases(10,12).

The onset of symptoms is either acute or subacute. Vomiting, severe abdominal pain, with or without shock are the manifestations of acute form. The subacute form has been designated as 'biliary ascites' and is characterized by acholic stools, mild fluctuating jaundice, ascite and fluid herniae.

Definite diagnosis of the entity is difficult, even at laparotomy owing to the following factors: (a) a history suggesting a choledochal cyst is usually lacking, *(b)* collapse of the cyst following perforation may prevent its identification even at operation, (c) a relatively small cyst can also rupture; *(d)* perforation in the posterior wall may occur(8). In our case, ultrasonography of the abdomen had helped us to make a correct preoperative diagnosis of perforated choledochal cyst. 99 mTc sequential scintiphotography has also helped in the diagnosis of at least 2 cases previously(13,14).

Treatment of such a condition consists of drainage of the extrahepatic biliary system by cholecystostomy or T-tube choledochostomy initially. The definitive procedure for choledochal cyst; complete excision of the abnormal portion of extrahepatic biliary system and biliary reconstruction using Roux-Y jejunostomy or isolated jejunal conduit is best delayed till later.

It is pertinent to highlight the differ-

ence between the perforated choledochal cyst and the 'pseudo choledochal cyst' formation that takes place following spontaneous perforation of the extrahepatic biliary system(18). This distinction is important not only to know the true incidence of the two conditions and to avoid errors in the operative management. Intestinal anastomosis to the biliary pseudocyst is associated with significant mortality(18). Conversely, simple drainage of perforated choledochal cyst would not suffice.

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