Management of biliary perforation in children

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ABSTRACT

Background: To study the aetiology, management and outcome of biliary perforations in paediatric age group. **Patients and Methods:** In a retrospective study, the records of patients presented with biliary peritonitis due to biliary perforations, managed from March 2006 to July 2009, are reviewed. **Results:** Eight male patients with biliary peritonitis due to biliary perforation were managed. These patients were divided in two groups, A and B. Group A, (n = 3) patients, had common bile duct (CBD) perforation, and Group B (n=5) patients had gallbladder perforation. The presenting features were abdominal pain, fever, abdominal distension, vomiting, constipation, jaundice and signs of peritonism. The management of CBD perforations in Group A was by draining the site of perforation and biliary diversion (tube cholecystostomy). In Group B, the gallbladder perforations were managed by tube cholecystostomy in four patients and cholecystectomy in one patient, however, one patient had to be re-explored and cholecystectomy performed due to complete necrosis of gall bladder. There was no mortality in our series. All patients were asymptomatic on regular follow-up. **Conclusion:** Early optimal management of biliary perforations remarkably improved the very high mortality and morbidity that characterised this condition in the past.

Key words: Common bile duct, gallbladder, perforation, biliary peritonitis

DOI: 10.4103/0189-6725.70413

INTRODUCTION

Biliary peritonitis following common bile duct (CBD) or gallbladder perforation is a rare entity in paediatric age group, compared with the adult population.[1,2] In paediatric patients, the CBD may perforate spontaneous, or due to anomalous union of pancreatico-biliary ductal system, congenital weakness of CBD, trauma, associated with choledochal cyst, viral infection, stenosis of CBD, necrotizing enterocolitis, intramural thrombosis, iatrogenic, or stone in the CBD.[3,4] The causes of gallbladder perforation in paediatric patients include acalculous cholecystitis, trauma, associated typhoid fever, gall stones, ischemic necrosis of the wall due to septicemia or disseminated intravascular coagulation; and it could be spontaneous.[5-8]

Previously, the reported mortality of biliary peritonitis due to biliary perforations was very high (> 40%) but latest reports described lower rates (12%). Some recent studies even showed no mortality at all.[9-13] This retrospective study analysed the aetiology, management and outcome of patients presenting with biliary perforation.

PATIENTS AND METHODS

Three patients with CBD perforation (Group A) and five with gallbladder perforation (Group B) formed the basis of this study. These patients presented at the Department of Paediatric Surgery, The Children's Hospital and The Institute of Child Health Lahore, Pakistan, from March 2006 to July 2009. Each patient's case notes were carefully examined and information on their biodata, presentation, mode of treatment, complications and outcome of management retrieved.

RESULTS

There were a total of eight male patients, in the age group of 2 to 10 years (mean 5.5), with biliary perforations, who presented with biliary peritonitis. The mean duration of illness before hospitalisation was 10.87 days (range of 2 to 30 days). The main presenting feature in all patients were abdominal pain, fever (in seven), abdominal distension (in seven), vomiting (in four), constipation (in two), jaundice (in one) and signs of peritonism in six patients. Six patients (75%) presented with acute fulminating biliary peritonitis whereas 2 (25%) patients presented with sub-acute features.

Two patients in Group A had blunt trauma abdomen,

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resulting in isolated CBD perforation and biliary peritonitis; in one patient CBD perforation was spontaneous [Figure 1]. In Group B, three patients had enteric fever-related gallbladder perforations, and one in patient each the gallbladder perforation was spontaneous and due to blunt trauma abdomen [Figure 2].

In all patients, total leucocytes count (TLC) was elevated, and ranged between 11000 and 20000 (mean 14500). Liver function tests (LFTs) were slightly deranged in two patients, although clinical mild jaundice was present in only one patient. Widal agglutination test was positive in three patients in Group B despite the enteric fever related gallbladder perforations. Haemoglobin level ranged between 9 and 11 g/dl (mean 10.5). In one patient, serum calcium was low. Plain abdominal x-ray showed air fluid levels in two patients. Abdominal ultrasound revealed mild to moderate pelvic collections in three patients, pericholecystic and sub-hepatic collections in two patients.

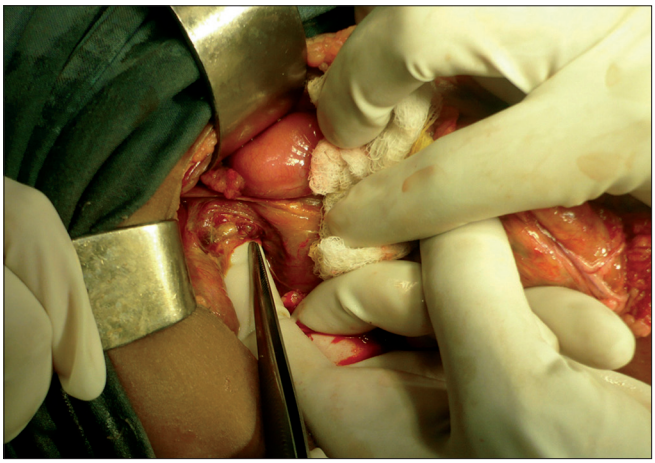


Figure 1: CBD perforation in a four-year-old child. This perforation was spontaneous in nature.

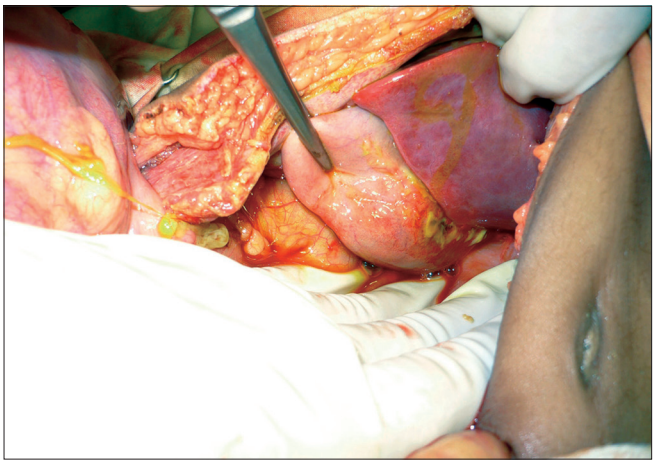


Figure 2: Gallbladder perforation in a six-year-old child. This perforation was spontaneous in nature.

The pre-operative diagnosis was secondary peritonitis due to perforated appendicitis in five patients, typhoid fever related peritonitis in one patient and suspicion of typhoid fever related gallbladder perforation in one patient. All patients had exploratory laparatomy. Group A *(n=3)*: CBD perforation was close to its union with cystic duct in all patients. Drainage of local site and biliary diversion (tube cholecystostomy) was performed. Postoperative recovery was uneventful in all cases. Retrograde cholangiogram, through tube cholecystostomy, performed on postoperative day 15, showed no leakage and the contrast material was safely passed to the duodenum. Drains and cholecystostomy tube were removed and patients discharged. All the patients were healthy after a minimum of six months follow-up.

Group B (n=5): Gallbladder perforation was found at fundus in two patients, body in two and at multiple sites in one patient. Tube cholecystostomy was performed in four cases while cholecystectomy was performed in one case with multiple perforations. In one patient who had prolonged history (one month), adhesions were present between loops of bowel, necessitating adhesionolysis and ileostomy, in addition to tube cholecystostomy performed because of iatrogenic serosal tears. One patient continued to have persistent peritonism and fever after the tube cholecystostomy. He had a re-operation during which the gallbladder was found to be completely necrosed for which he had cholecystectomy performed.

In cases with tube cholecystostomy, retrograde cholangiogram was performed on postoperative day 15 to rule out any distal obstruction [Figure 3]. Patients were discharged after removing the cholecystostomy tubes. Only one patient has had a follow-up of one month, others have had more than a year's symptom-free period of follow-up. In each patient with tube cholecystostomy the bilio-cutaneous fistula closed spontaneously within few days of removing the tube.

Postoperative complications were found in two patients. These included wound infection in one patient, and persistent collection and peritonism in other. The hospital stay was between 9 to 25 days (mean: 16.5 days). The summary of patients with reference to their aetiology and management is presented in Table 1.

DISCUSSION

Biliary peritonitis due to CBD or gallbladder perforation is scarcely reported in paediatric age group as compared

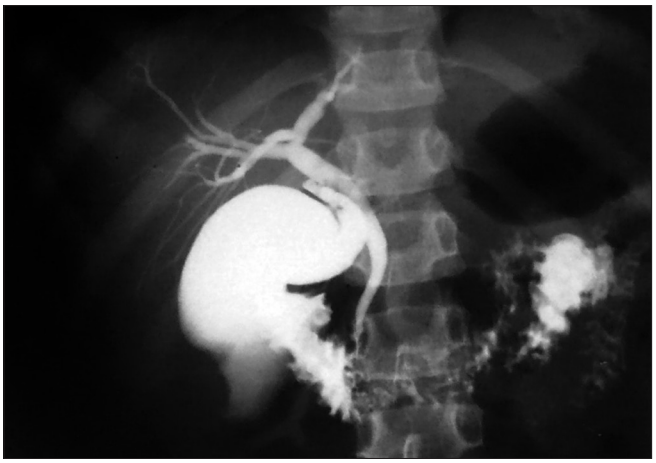


Figure 3: Retrograde cholangiogram through tube cholecystostomy. Note that there is no leakage, filling defect or abnormal constriction anywhere in the biliary tract.

to the adult population. Very few case reports and limited case studies have been published indicating the rarity of the condition in this age group. CBD perforation is commoner than gallbladder perforation in paediatric patients.[1,2,6,7,14] In our series, however, there were more cases of gallbladder perforations than CBD perforations, probably because of the high incidence of typhoid fever and its related surgical complications in our region. Our institution receives a lot of referrals from all over Pakistan and some times, Afghanistan. This might explain the reason for the high number of patients with biliary perforation seen in this three-year period.

Various factors have been identified to cause biliary perforations including, distal obstruction, and congenital weakness of wall, anomalous union of pancreatico-biliary ducts, infections, trauma, inflammation, stone-related and iatrogenic injury.[3-5,8,15] In our series, the main causes of biliary perforations are trauma and typhoid infection. Enteric fever is almost an exclusive affliction of the developing world, caused mainly by unhygienic conditions and poor sanitation, and presents with a wide variety of systemic manifestations. Some of its surgical importance relates to its serious abdominal complications such as intestinal perforation, bleeding, cholecystitis, gallbladder perforation and pancreatitis.[16] Each year we managed at least one patient of typhoid-related gallbladder perforation in our institution.

Patients with biliary perforations can present acutely or insidiously. Acute presentation includes fever, severe pain abdomen, peritonism, vomiting and shock. The insidious or sub-acute presentation includes progressive jaundice, painless abdominal distension and clay coloured stools.[17,18] In our series, six patients presented with acute fulminant biliary peritonitis and two patients with insidious mode of presentation.

Peritonitis due to biliary perforations is a very rare disease causing difficulties in pre-operative diagnosis. Various studies depicted only a few cases in which pre-operative diagnosis was possible.[5,19-21] In our study, we suspected biliary perforation in only one out of eight patients. In most of cases, our pre-operative diagnosis was secondary peritonitis due to perforated appendicitis.

Abdominal X-ray is not a modality of choice to diagnose this condition. Abdominal ultrasound, CT scan, MRI and radionuclide scans are important tools in the diagnosis.[5,19-21] However, operation should not be unduly delayed due to unavailability of these diagnostic tools if there are established features of generalised peritonitis.

The surgical management options in CBD perforation are simple drainage at the site of perforation, with or without biliary diversion and with or without closing the perforation. Many authors are not in favour of closing the perforation. Previously cholecystectomy with CBD excision and Roux-en-y hepatico-enterostomy was performed for CBD perforation. But now, it is preferred and reserved for non-treatable distal obstruction, persistent bilio-cutaneous fistula, persistent biliary leak, choledochal cyst associated CBD perforation etc.[2,3,7,8,17,18,22] The management options for gallbladder perforation are cholecystectomy

Table 1: Summary of aetiology, management performed for biliary perforations

Pt.	Age	Aetiology	Operative Findings	Procedure
A1	2y	Traumatic	CBD perforation	Drain + Tube cholecystostomy
A2	4y	Spontaneous	CBD perforation	Drain + Tube cholecystostomy
A3	5Y	Traumatic	CBD perforation	Drain + Tube cholecystostomy
B1	6Y	Spontaneous	GB perforation	Tube cholecystostomy
B2	5Y	Typhoid fever	GB perforation	Tube cholecystostomy
B3	6Y	Typhoid fever	GB perforation	Cholecystectomy (CC)
B4	10Y	Typhoid fever	GB perforation	Tube cholecystostomy then CC
B5	6Y	Traumatic	GB perforation	Tube cholecystostomy

CBD: Common bile duct

and tube cholecystostomy. Cholecystectomy is the preferred method but due to inflammation in that region, sometimes operative morbidity can rise due to difficulty in identifying structures. There are various studies showing successful management of acute phase of biliary perforations by percutaneously placement of ultrasound / CT guided drains.[1,9-11]

We successfully managed the CBD perforations by local drainage and biliary diversion. Gallbladder perforations were managed by performing tube cholecystostomy preferably in most of our patients, although in case with multiple perforations and complete necrosis of gallbladder, cholecystectomy was preferred. Tube cholecystostomy, as the primary modality to manage gallbladder perforation, is preferred in our series because it is a simple surgical technique and even residents can perform it with ease; it is safer in cases where identification of various important structures in that area is precluded by inflammation and adhesions.

Postoperative complications of biliary perforations are wound infection, burst abdomen, persistent peritonitis, prolonged ileus, ligation of CBD during cholecystectomy, haemobilia, gallbladder necrosis etc.[9,10,12,21] We were lucky to have only a few postoperative complications.

In 1942, Glenn *et al*. reported 42% mortality rate in biliary peritonitis. This was further reduced to 12-16% in some other studies, and more recent studies have reported zero mortality rate.[9-11] This is also true in our study. The main reasons for this reduction in mortality are early surgical intervention and development of better intensive care modalities.

To conclude, biliary peritonitis due to biliary perforation is a manageable condition in paediatric patients. Early diagnosis and surgical intervention can reduce mortality and morbidity and ensure a good prognosis.

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Source of Support: Nil, **Conflict of Interest:** None declare.