



## SPONTANEOUS BILIARY PERFORATION IN CHILDREN: CLINICAL AUDIT OF A RARE ENTITY

### Pediatrics

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### ABSTRACT

Spontaneous or idiopathic biliary perforation is an infrequently encountered cause of acute abdomen. It is rarely suspected or correctly diagnosed preoperatively in most of the cases. Clinical presentation is that of a biliary peritonitis. The exact cause of the perforation remains unclear and the diagnosis is usually made at the time of laparotomy for an acute abdomen. Treatment is surgical and usually, consists of common bile duct (CBD) exploration and repair over a T-tube. This entity has an excellent outcome when intervened timely.

### KEYWORDS:

CBD, Spontaneous biliary perforation, biliary peritonitis, T- tube

### Introduction

Spontaneous common bile duct (CBD) perforation is a complication rarely seen in infants and children. Idiopathic perforation of nondilated CBD was first described by Caulfield in 1936<sup>1</sup>. The condition may present without previous signs of biliary tract disease. The child usually presents with abdominal distention, generalized peritonitis, mild fluctuating jaundice, acholic stools and septicemic shock. It is rarely suspected or correctly diagnosed preoperatively in most of the cases. The exact cause of the perforation remains still unclear and the diagnosis is made at the time of laparotomy for an acute abdomen. We present single center experience of four cases of spontaneous bile duct perforation in children and discuss the dilemmas in the diagnosis and management in the light of available literature.

### Aims and objectives

The present clinical study is aimed at:

- Evaluation of the total incidence, etiology, clinical feature and management of spontaneous CBD perforation.
- Enumerating the clinching features for the early diagnosis and management of spontaneous CBD perforation.

### Material and Methods

- **Study design:** This retrospective chart review study was carried out in the Department of Pediatric Surgery, at a tertiary care hospital in Jharkhand.
- **Subjects:** All children with feature of CBD perforation admitted between May 2012 to May 2017.
- **Inclusion Criteria:** Medical records of patients of CBD perforation who had completed treatment at our center, were recalled for the study in terms of incidence, etiology, clinical feature and management.
- **Exclusion criteria:** Patients whose records were unavailable were excluded.

### Result

A total of 4 patients of CBD perforation, presented in emergency at a tertiary care hospital, between May 2012 to May 2017, were included in the study. Out of the four patients, 2 were male and two females; age range of 1 to 3 years and presented with the clinical profile of acute abdomen (Table 1). The symptoms common to all patients were the presence of sign of generalized peritonitis, abdominal distention and shock. Radiography of abdomen was suggestive of free fluid without the gas under diaphragm. Mild icterus was seen in two of the patients. Ultrasonography of abdomen revealed free fluid with non-dilated bowel. All patients had undergone surgical exploration after initial stabilization. Intra-operative findings were common as bilious ascites and the common bile duct perforation (**Image :1**) with non-dilated CBD. In two of the patient, per-operative cholangiogram was obtained (**Image:2**); which was normal in one and dilated CBD seen in the other patient. All the patients were managed by flushing the CBD with normal saline via an infant feeding tube and primary repair of the perforation over a T- tube drainage. Normal cholangiograms were obtained after 2 weeks post-operatively, before removal of T- tube in three patients and minor leak was detected in one which was managed

conservatively. Recovery was satisfactory in all patients in follow up.

**Image:1**



**Intra-op Picture**

**Image : 2**



**Per-op Cholangiogram**

**Table 1: Clinical Profile**

| Serial No. | Age       | Sex | Clinical feature                                                  | Investigations                                                                                          | Treatment                                 | Post-op Cholangiogram              |
|------------|-----------|-----|-------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------|-------------------------------------------|------------------------------------|
| Patient 1  | 1 ½ yr.   | F   | Acute abdomen, ascites, septic shock                              | <b>X Ray-</b> ground glass appearance, no pneumoperitoneum<br><b>USG-</b> ascites and dilated bowel     | EL+ IO cholangiogram + repair over T tube | Normal                             |
| Patient 2  | 1 yr.     | F   | Acute abdomen, Ascites and Mild jaundice, Clay colored stool      | <b>X Ray-</b> ground glass appearance, no pneumoperitoneum<br><b>USG-</b> ascites, mild dilated bowel   | EL + repair over T tube                   | Dilated CBD                        |
| Patient 3  | 11 months | M   | Acute abdomen, Abdominal distention, Ascites                      | <b>X Ray-</b> ground glass appearance, no pneumoperitoneum<br><b>USG-</b> ascites and non-dilated bowel | EL+IO cholangiogram + repair over T tube  | Minor leak with normal caliber CBD |
| Patient 4  | 3 yrs.    | M   | Acute abdomen, mild jaundice, not passage of stool and distention | <b>X Ray-</b> ground glass appearance, no pneumoperitoneum<br><b>USG-</b> ascites and non-dilated bowel | EL+ repair over T tube                    | Normal                             |

S= Sex, EL= Exploratory Laparotomy, IO= Intraoperative

### Discussion

Perforation of the extrahepatic bile duct is extremely rare entity and is described mainly in infant due to congenital anomalies of common bile duct<sup>2</sup>. Spontaneous common bile duct perforation is commonly seen in

children less than four years of age<sup>3</sup> with peak incidence during first year of life. Banani et al<sup>4</sup> suggested that the peak age of occurrence was around 6 months of age and that it could range from 25 weeks of gestation to 7 years of age. The etiopathogenesis of spontaneous biliary perforations is still obscure. The causes for spontaneous perforation of bile duct reported in literature are, stone, previous biliary tract surgeries, tumors, congenital malformations like choledochal cysts, biliary diverticulum and sometimes idiopathic<sup>1,5</sup>.

Acute presentation is with abdominal distention, ascites and shock. Presence of jaundice is variable. Previous history pertaining to biliary tract disease is usually absent. The abdominal examination findings are that of a generalized or localized peritonitis with ascites. The pre-operative diagnosis of patients presenting in emergency is often difficult. In our study, all patients presented with sign of peritonitis, ascites, abdominal distention and no pneumoperitoneum on erect radiogram. The diagnosis of biliary perforation should be kept in mind in such type of presentation. After the initial patient, we were wiser by suspecting the entity even preoperatively. Ultra-sonographic findings in CBD perforation are of free intra-peritoneal fluid with normal intra and extra-hepatic ducts. In doubtful cases, the diagnosis can be confirmed with a radio-isotope scan. In two of the cases intra-operative cholangiogram was done, which indicated free flowing contrast in to the duodenum.

Surgical management is mandatory as soon as the diagnosis confirmed<sup>6</sup>. In the situation where the surgeon encounters biliary perforation without a pre-operative diagnosis, the safest policy is to drain the area and place a T-tube through perforation, a per-operative cholangiogram should be obtained if condition permits. Patients need post-operative antibiotics to counteract cholangitis and septicemia. Cholangiogram should be obtained after 2 weeks prior to removal of T-tube.

### Conclusion

Spontaneous perforation of the extra-hepatic bile duct is a rare but important condition in the children. The overall prognosis of the condition is good, provided an early and appropriate surgical management is instituted.

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- *Conflict of Interest: None*

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