

# Spontaneous Common Bile Duct Perforation in Full Term Pregnancy: A Rare Case Report and Review of Literature

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## Case report

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# Abstract

**Background:** Spontaneous biliary system perforation is a rare presentation in clinical practice. It is even rare in adult population than infants. The condition is rarely suspected and diagnosed preoperatively because of small number of cases, vague sign and symptoms and ambiguous presentation.

**Case presentation:** we are presenting an interesting case of spontaneous perforation of common bile duct in a 16 years old lady presented a week after her first delivery to the emergency department with complain of diffuse abdominal pain, abdominal distention, fever, vomiting and constipation. She was having generalize peritonitis but the cause of peritonitis was unknown despite of all available investigations performed. She underwent exploratory laparotomy and a perforation in the supra duodenal region of common bile duct was found intraoperatively. Common bile duct repair over T-tube and cholecystectomy performed and patient recovered.

**Conclusion:** Spontaneous biliary perforation is a rare cause of acute abdomen in adults and extremely rare in pregnancy and its belated diagnoses and management is leading to high morbidity and mortality. All physicians especially surgeons should be aware of its possibility and consider it a cause of peritonitis on differential diagnosis especially when there is no apparent etiology available for the presentation.

## Introduction

Although most injuries to the bile duct are iatrogenic after some kind of interventions like endoscopic retrograde cholangiopancreatography (ERCP) and open or laparoscopic cholecystectomy[1]. Spontaneous bile duct perforation can also occur and is a rare disease in adults. Aetiologies include increase pressure in the bile duct secondary to obstruction by stones, stricture, tumors, parasites; intra mural infection, necrosis of bile duct wall secondary to thrombosis of intramural vessels, direct erosion by stones, cirrhosis and weakness of duct wall by multiple reasons. It can also occur without known cause but calculi were found in common bile duct (CBD) in 70 % reported cases. It is comparatively common in infants and aetiologies are congenital biliary anomalies most of the time[2,3]. Here we present a case of a young lady who was diagnosed with spontaneous CBD perforation during surgery just after a week she gave birth to her baby. We also performed an extensive literature review and found that only 5 cases of bile duct perforation during pregnancy or in early postpartum period have been reported in literature till now.

## Case Presentation

A 16 years old female Gravida 1 para 1 presented to the emergency department with no past medical and surgical history except present one. According to her history she developed intermittent fever and chills, right upper quadrant pain and urinary retention from last 2 weeks and underwent normal vaginal delivery 1 week ago. She was hospitalized in another province for the given complain. The presenting complain now were diffuse abdominal pain, abdominal distention and continuous fever from last 3 days; vomiting and constipation from last 2 days. The vital signs were BP 133/97 mmHg, RR 20/m, PR 136/m, Temp 39 C<sup>0</sup> and Spo2 90 %. On physical examination the patient was ill looking having distended abdomen with generalize tenderness, rebound tenderness and guarding. Fluid thrill and shifting dullness was positive too. Bowel sound were not audible on auscultation. Blood investigation results were TLC 21000/cmm (normal 4000-11000), neutrophil 89 % (normal 40-75), Hb 8.2g/dl (normal 11.5-16.5), serum creatinine 0.86 mg/dl (normal 0.6-1.2), total bilirubin 0.71 mg/dl (normal 0.1-1.2), SGOT 20 IU/ml (normal 0-40), SGPT 34 U/L (normal 5-40), alkaline phosphatase 109 IU/L (normal 40-240), blood group AB +ve, HbsAg, anti HCV Ab and HIV were negative. She was received by emergency doctor and was labelled postpartum sepsis as she was having signs and symptoms of sepsis. Ultrasound showed distended bowel loops floating in free fluid in peritoneal cavity. Erect chest X-ray shows no air under right hemidiaphragm. CT with contrast was performed which revealed gross ascites considering peritonitis but

could not find the cause of ascites. Keeping hollow viscous perforation in mind an exploratory laparotomy was performed after initial management. Biliary peritonitis was found with about 4 liters of bile-stained fluid in the peritoneal cavity. Stomach, small intestine (distended) and colon was normal. A perforation of about 7-8 mm was found on anterolateral wall of supra duodenal segment of CBD below the junction of cystic duct and common hepatic duct as shown in fig 1. Gall bladder and rest of biliary tree was normal without any calculus. The facility of intra operative cholangiogram was not available, a 10 French feeding tube was passed to the duodenum through perforation to confirm the distal obstruction which passes without any resistance. Peritoneal lavage with warm normal saline, Cholecystectomy and CBD repair over T-tube was done. Post-operative stay was uneventful and patient fully recovered and discharged home on 7th Post-operative day. T- tube removed on 15th post-operative day after cholangiogram showed no filling defect and normal contrast flow to the duodenum. The patient is doing well after one year follow up.

## Discussion

Spontaneous perforation of common bile duct was first described by John Freeland in 1882 when he found multiple diverticula around CBD with stones on autopsy[4]. Most common site of biliary system perforation is gallbladder accounting for 91% of cases followed by common bile duct (4.4%), cystic duct (3.3%) and common hepatic duct (1.1%) [5]. Commonest site of extrahepatic biliary system perforation excluding gallbladder is the junction of cystic duct with hepatic duct[6]. First case in infants was presented by Dijkstra in 1932[7]. Incidence of spontaneous biliary perforation is 1.5 in 1000,000 live births over the first year of life, the most common site in infants below age one was the junction of common hepatic and cystic duct (43%), common bile duct (23%), gall bladder 12%, hepatic duct 9 % and cystic duct 5% [8]. The incidence of overall biliary system disease in pregnancy is ranging from 0.05-0.3 % [9]. The first case of spontaneous bile duct perforation in pregnancy which was associated with gall stone was reported by Piotrowski in 1990 but bile duct perforation due to congenital anomaly in pregnancy has been reported before that in literature. 70 % spontaneous cases of biliary perforation were associated with gall stones where stone were found during surgery [2]. The prevalence of spontaneous bile duct perforation is ultra-rare in pregnancy with only few cases reported in literature. To the best of our knowledge all the cases of normal anatomy biliary system perforation (Including perforation of gallbladder and bile duct) in pregnancy or early postpartum period reported in literature are listed in Table 1.

Table 1: All reported cases of spontaneous biliary system perforation in pregnancy or early postpartum period

Name and publication year	Age/GP	No of cases	Site of perforation	cause of perforation	Surgery time	Treatment	references
Chaimoff et al 1973	?	1	Gall bladder	?	?	?	[10]
Piotrowski et al 1990	23 G1P0	1	CHD	Gall stone	concomitant with C section	Hepaticojejunostomy	[2]
Behera A et al 1991	?	1	Gall bladder	Acute cholecystitis	?	?	[11]
Goodlin et al 1991	?	4	Gall bladder	Gangrene of Gall bladder	postpartum	Cholecystectomy	[12]
Petrozza et al 1995	28 G7P6  23 G4P4	2	Gall bladder	Gall stone	postpartum	Cholecystectomy	[13]
McGrath et al 2005	34 G1P0	1	CBD	Gall stone	postpartum	Sphincterotomy + stent placement	[14]
Talwar et al 2006	28 G2P1	1	Gall bladder	Gall stone	during pregnancy	Cholecystectomy	[15]
Talwar et al 2006	21 G1P0	1	CBD	Idiopathic	during pregnancy	Over T-tube repair	[15]
Dabbas et al 2008	20 G1P1	1	CBD	Gall stone	postpartum	Choledocholithotomy +Over T-Tube repair	[16]
Bediako-Bowan et al 2013	29 ?	1	CBD	Idiopathic	postpartum	Cholecystectomy +Over T-Tube repair	[17]

CHD: common hepatic duct, CBD: Common bile duct, GP: Gravid and Para

Strictly speaking most of the cases are not spontaneous because they are secondary to some other underlying pathology. Out of all these spontaneous biliary system perforations cases given in the table, only 5 cases of bile duct perforation have been reported; 4 in common bile duct and 1 in common hepatic duct. We are presenting the truly spontaneous common bile duct perforation case which we think is the 6<sup>th</sup> case of bile duct and 5<sup>th</sup> case of CBD perforation during pregnancy in literature.

According to our analysis the commonest site of biliary tract perforation during pregnancy is gall bladder 9/15 (60 %), CBD 5/15 (33.3%) and hepatic duct 1/15 (6.66%). The cause for spontaneous perforation was gall stones 6/9 (66.6 %) and idiopathic 3/9 (33.3 %). The cause for 6 other cases mentioned in the table has not been clearly described and for the purpose of accuracy been excluded from the analysis.

The theories behind spontaneous bile duct perforation in adults are obstruction distal to perforation leading to high canalicular pressure; weakness of bile duct wall or combination of both. The aetiologies leading to these phenomena,

mentioned in literature are impacted stones or erosion of bile duct wall by stone without impaction, intramural infection, stricture, stone, tumor, parasites, spasm of sphincter of Oddi, necrosis of wall of bile duct due to thrombosis of bile duct blood vessels, cirrhosis, birth trauma, biliary tract congenital anomalies like choledochal cyst or biliary diverticulum, connective tissue diseases and previous biliary tract surgeries. Some other comorbidities associated with spontaneous biliary perforation like HIV infection, tuberculosis of CBD, Hodgkin's Lymphoma and severe necrotizing enterocolitis involving duodenum or viral infection of the bile duct are also reported in literature [2,3,6]. Spontaneous bile duct perforation sometimes can be idiopathic like in our case. But the possible reason for idiopathic cases maybe thrombosis of those small blood vessels leading to ischemia and necrosis of bile duct wall and finally perforation.

The presentation of the patient can be different from case to case, it can have both acute and insidious onset. Most of the patients having insidious onset (80%) may present with abdominal distention without abdominal pain, clay color stool and progressive jaundice may follow[3,6,18]. In acute cases which are about 20% the signs and symptoms of acute abdomen like generalize abdominal pain, abdominal distention due to bilious ascites, vomiting, fever, jaundice, high level of bilirubin or even shock may develop. The patient can present with perihepatic abscess instead of generalize peritonitis if the bile localized in the area [3,19].

We believed our case was insidious onset and developed CBD perforation two weeks before she presented to us but she was misdiagnosed by less experience local health care workers in rural area maybe because she was a full term pregnant and they misinterpret the symptoms as labor pain. She gave birth to her baby just a week after her symptoms starts. As a case of negligence, she finally developed generalized abdominal pain which maybe because of rupture of walled off biloma leading to fulminant peritonitis, infection with high fever and high leukocyte count or the symptoms may get evident when enough bile accumulated and infected in the peritoneal cavity. There is less chance of first scenario because we could not found and walled up cavity during surgery. The symptoms of bowel obstruction (abdominal pain, distention, vomiting and constipation) can be explained by paralytic ileus caused by infection and peritonitis.

Owing to the rarity of the case, suspicion and preoperative diagnosis of the condition is difficult and diagnosis is mostly made during surgery. As perforation of biliary system is a known complication of cholelithiasis or choledocholithiasis, it should be suspected if patient presented with perihepatic abscess or signs and symptoms of peritonitis with the history of biliary stone disease [19]. If suspected, pre-operative diagnosis can be made by hepatobiliary iminodiacetic acid scan (HIDA scan), magnetic resonance cholangiopancreatography (MRCP), Endoscopic retrograde cholangiopancreatography (ERCP), Magnetic resonance imaging (MRI) and Computerized tomography (CT scan). Because these facilities are expensive and not widely available especially in low-income countries or in rural areas of many other countries, an easy and cheaper biochemical test from the ascitic tap "ascitic fluid bilirubin concentration and ascitic fluid: serum bilirubin ratio" can help in diagnosis of preoperative biliary peritonitis. The normal range of ascitic fluid bilirubin is 0.7-0.8 mg/dl, concentration above 6 mg/dl support the diagnosis of choleperitoneum[18]. A study by Darwin et al shows that the peritoneal fluid to serum bilirubin ratio (FSBR) greater than 5 is 100 % specific and sensitive for prediction of bile leak [20]. Ultrasonography maybe the first investigation to perform because its cheap, readily available and can show free fluid in abdominal cavity, fluid and pus collection in perihepatic area and biliary system pathologies. In case of CBD stones the dilated biliary tree proximal to obstruction can be well visualized. So, ultrasound may not find the exact site of perforation but still is a helpful investigation especially in low equipped settings. chest X-ray in erect or left lateral decubitus position will not reveal gas under right hemidiaphragm which is expected in many gut perforation cases. So, the suspicion should be more if the patient presented with peritonitis with no gas under right hemidiaphragm on X ray and other positive marker like bilious peritoneal tap with history showing any biliary disease. Sharma et al considering peritonitis with bilious peritoneal tap, no pneumoperitoneum and acholic stool pathognomonic for spontaneous biliary perforation [21].

The condition may be confused and challenging to diagnose because of similar presentation with other pregnancy related diseases like pregnancy induced hypertension (PIH) preeclampsia and hemolysis, elevated liver enzymes and low platelets (HELLP) syndrome. McGrath et al and Goodlin et al discussed about biliary system perforation confused with or misdiagnosed as pregnancy associated problems [12,14]. This confusion may lead to delayed diagnosis which can be the cause of high morbidity and mortality. Spontaneous perforation of biliary tree should be considered in differential diagnosis in such cases to avoid such catastrophic events.

Management of spontaneous bile duct perforation is ranging from minimal invasive intervention to more aggressive surgical management. Based on the patient condition, different treatment modalities maybe choose. The management mainstay in case of suspected calculi in biliary tree is to 1) evaluate stone probability in biliary tree, 2) choledocholithotomy or removal of stones if present and 3) removal the source of stone (cholecystectomy) [22]. ERCP is of diagnostic and therapeutic advantage and spontaneous biliary perforation cases can be diagnosed and treated at the same time with removal of stone and placement of stent in the bile duct [14,21]. Failed ERCP cases or where ERCP facility is not available, laparoscopic or open exploration should be considered. If there is biloma and localized perihepatic abscess secondary to CBD obstruction by stone, it can be drained percutaneously concomitantly with stones in CBD removed with endoscopic sphincterotomy[19].

Patients presented with generalize biliary peritonitis need prompt exploration, thorough peritoneal toilet and drainage and management of perforation site accordingly [21]. An intraoperative cholangiogram should be performed if available to check for stones and pathologies of biliary tree. A conservative management of abdominal drainage and biliary tree decompression is recommended if there is no post perforation obstruction and perforation will heal automatically once the biliary tree decompressed [19]. Primary closure of the perforation site is hard and dangerous to perform because of inflammation around the perforation site and most of the patients will recover after external decompression and treating the primary pathology[19,21].

Some authors believe that if the facility of intraoperative cholangiogram is available and there is no distal obstruction, primary closure of bile duct perforation can be performed. But if there is distal obstruction like stricture or atresia, a biliary enteric anastomosis is required to avoid portal hypertension and biliary cirrhosis[3,23]. Spigland et al also suggested only external drainage for biliary perforation without ductal abnormalities, but he added the cholecystostomy may help in healing of duct perforation and also guide when to remove peritoneal drain[24]. Gurusamy et al in a review study of 2007 suggested that primary closure of choledochotomy is as effective as closure over T-tube in open exploration of bile duct but in review of 2013 they suggested that choledochotomy closure over T-tube in open exploration of bile duct increase operative time and hospital stay without any benefit, they believed routine T-tube drainage for CBD stones should be avoided as there is no justification for its use based on current available evidences. They also suggested that T-tube drainage in laparoscopic exploration of CBD also increase surgery time and post-operative hospital stay without significant difference in morbidity than primary closure without stent [25]. Simple T-tube peritoneal drainage is also acceptable even in the presence of distal obstruction when the exploration of porta hepatis is very risky in inflamed condition and may worsen the scenario. This mini surgery is less morbid and have good chances for healing or at least stabilizing the patient for second look to have definitive surgery if the condition has not cured[23]. we recommend the use of peritoneal drainage and percutaneous cholecystostomy in cases where porta hepatis exploration is not feasible for any reason even if distal obstruction is present. Roux-en-Y bilioenterostomy was performed previously for biliary perforation but now only limited to untreatable distal obstruction, persistent biliocutaneous fistula or biliary leakage and CBD perforation associated with choledochal cyst [21]. If the facility of the cholangiogram is not available then the patient may best managed by closure over T- tube and cholecystectomy as in our case [3,18,23].

## Conclusion

Spontaneous bile duct perforation is very rare in adults and even rare in pregnancy. Preoperative diagnosis is hard to make because of its rarity and similar presentation with many other diseases. Physicians especially surgeons should be aware of this condition and should consider it a cause of peritonitis on differential diagnosis. High index of suspicion should be made in patients presented with peritonitis in pregnancy or in early post-partum period with bilious peritoneal tap, without pneumoperitoneum and no apparent cause for peritonitis. Peritoneal tap bilirubin concentration and its ratio with serum bilirubin are very helpful in diagnosis. The purpose of the management is to halt the catastrophic event, remove the cause and source of perforation whenever possible and decompress the biliary tree. Simple peritoneal drainage maybe done in patients unfit for porta hepatis exploration followed by another surgery after stabilization.

## Abbreviations

CBD: common bile duct

HIDA: Hepatobiliary iminodiacetic acid

MRCP: Magnetic resonance cholangiopancreatography

ERCP: Endoscopic retrograde cholangiopancreatography

MRI: Magnetic resonance imaging

CT: Computerized tomography

PIH: Pregnancy induced hypertension

HELLP: Haemolysis, elevated liver enzymes, Low platelets

## Declarations

Ethics approval and consent to participate: The study was approved by the Ethics committee of Amiri Medical Complex, Kabul, Afghanistan.

Consent for publication: A written informed consent has been obtained from both the patient and her husband for publication.

Availability of data and material: The datasets used during the current study are available from the corresponding author on reasonable request.

Competing interest: The authors have no competing interests to declare.

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Author contributions: MM and MAS designed and conceptualized the study. Both authors did the literature review, collected and analyzed the data, wrote the manuscript, read and approve the final version of the manuscript.

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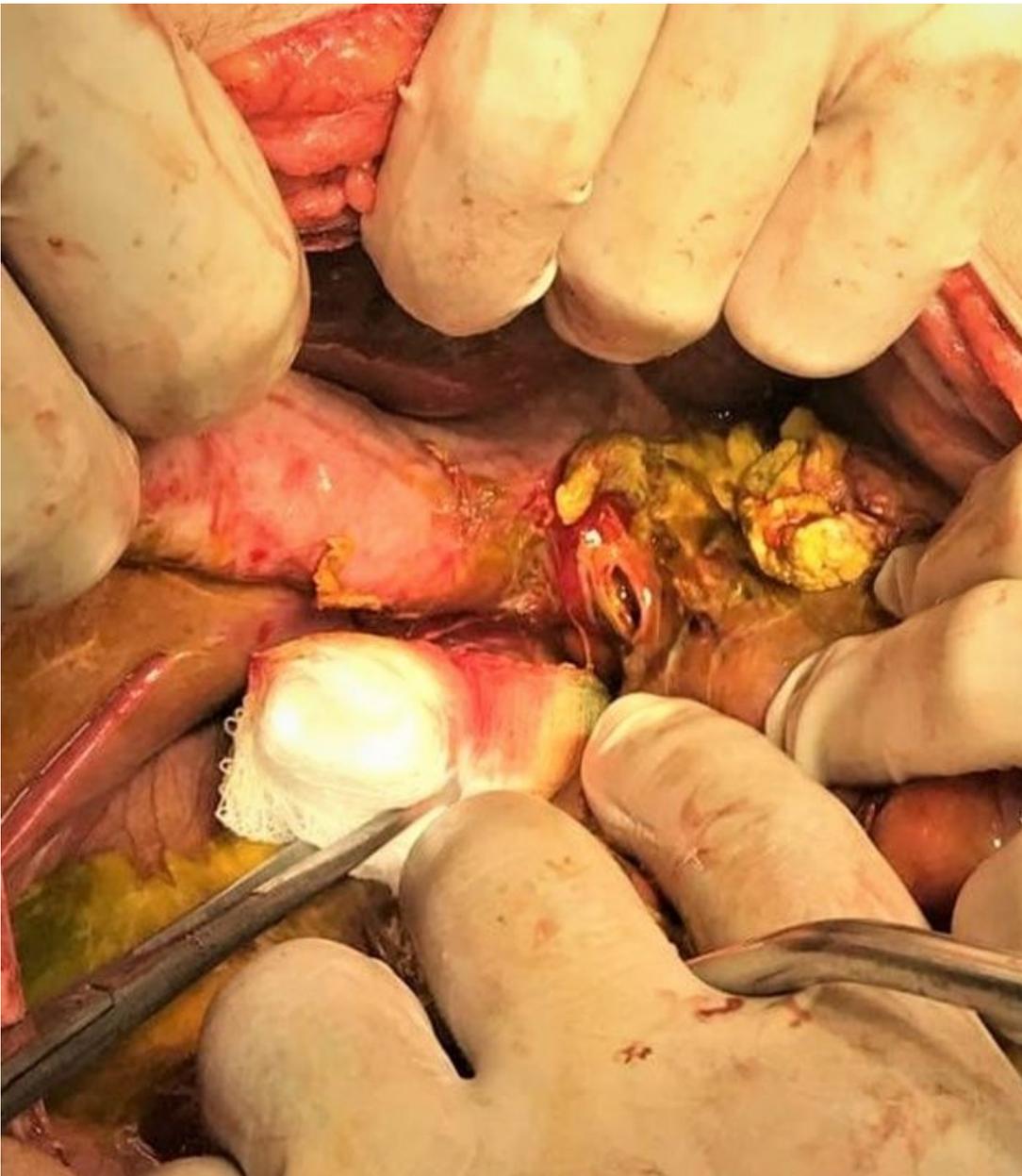
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## Figures



**Figure 1**

Perforation of supra duodenal segment of common bile duct