

## Case Report

# A case report of spontaneous biliary peritonitis with accessory liver lobe: rare presentation of a rare pathology

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## ABSTRACT

Spontaneous intrahepatic bile duct perforation with associated accessory liver lobe is an exceedingly rare condition with very few cases reported in the literature. Here we report a case of 76 years old man presenting with severe pain, abdominal distension since 3 days having jaundice and signs of peritonitis. Having undergone an exploratory laparotomy, 1 liter of bilious peritoneal fluid was seen with a perforation of left intra hepatic bile duct with a presence of an accessory lobe of liver. The gall bladder was contracted with multiple stones. Extrahepatic biliary duct was found normal intraoperatively. A cholecystectomy with resection of the accessory liver lobe was done. Post operatively patient had persistence of jaundice for which a magnetic resonance cholangiopancreatography was done, revealing a distal common bile duct calculus. Patient underwent endoscopic retrograde cholangiopancreatography with stone extraction on post operative day 10 relieving the symptoms. Patient is on regular follow up with no further complications. Awareness of such unusual occurrences is necessary for early diagnosis and instituting the appropriate treatment thereby reducing the mortality.

**Keywords:** Spontaneous biliary peritonitis, Accessory liver lobe, CBD stone, Cholangitis, Bile duct perforation, Case report

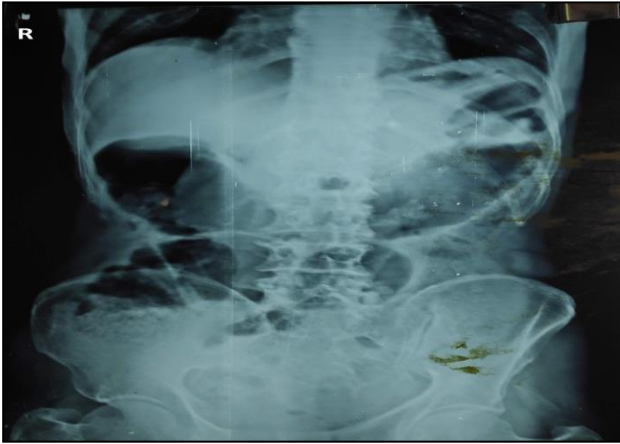
## INTRODUCTION

Spontaneous perforation of intrahepatic bile duct leading to biliary peritonitis is an extremely rare condition encountered in surgical practice. Though spontaneous bile duct perforation is encountered in infants, its occurrence in adults is seldom seen.<sup>1</sup> Such conditions progressing to biliary peritonitis presenting as acute abdomen requires high clinical suspicion for a timely diagnosis. Associated anatomical variants of the liver make the diagnosis more tedious. There are very few case reports in the literature of accessory liver lobe presenting as spontaneous biliary peritonitis. Here, we present such a case of spontaneous left intra hepatic duct perforation in a patient with an accessory left sided liver lobe due to a common bile duct (CBD) stone. This case report has been reported in line with the SCARE criteria.<sup>2</sup>

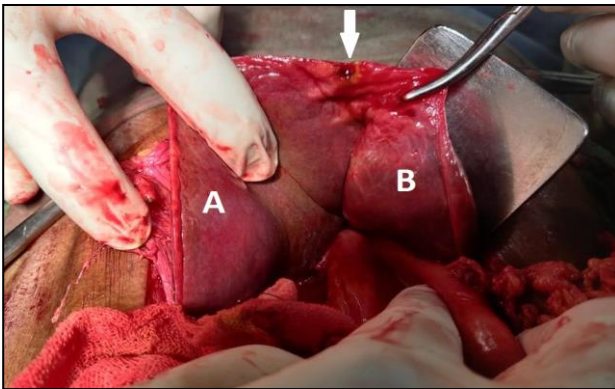
## CASE REPORT

The patient, a 76-year-old man presented with acute severe pain first localized in the right upper abdomen since 3 days and later extended to involve the entire abdomen. On examination patient was icteric with a pulse rate of 110/min and blood pressure of 104/66 mmHg. Abdominal examination revealed a distended tense abdomen with obvious tenderness and rigidity. Abdominal radiography was done and showed only features of ileus with no evidence of a perforated viscus as shown in figure 1. An ultrasound scan of the abdomen showed moderate peritoneal collection with internal debris, intrahepatic biliary radicles (IHBR) dilated and gall bladder contracted with multiple stones. Distal CBD could not be visualized. Liver function tests revealed an elevated total bilirubin

(4.5 mg/dl) with a direct bilirubin of 3.2 mg/dl. Alkaline phosphatase was found to be 280 mg/dl.



**Figure 1: Abdominal radiography showing no evidence of perforated viscous.**



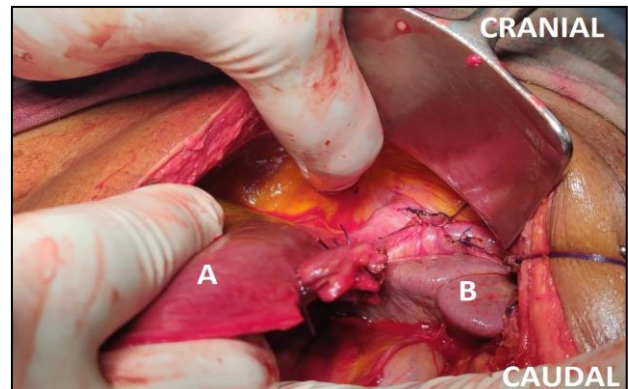
**Figure 2: Intraoperative photograph showing the site of perforation in the intra hepatic bile duct; (A) the left lobe of liver; (B) accessory liver lobe.**



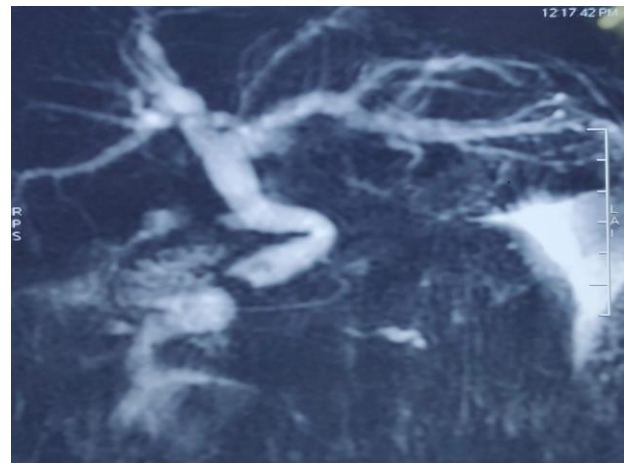
**Figure 3: Resected accessory lobe of liver.**

With a clinical diagnosis of peritonitis secondary to cholangitis, patient underwent exploratory laparotomy after initial resuscitation. Intraoperative findings revealed 1000ml of bilious fluid in peritoneal cavity; an accessory liver lobe attached to the left lobe of liver; a perforation of

the intra hepatic bile duct from the accessory lobe as shown in figure 2. Gall bladder was found to be thick-walled containing multiple stones. Rest of the abdominal viscera including the inspected CBD, liver surface and right lobe of liver was found to be normal. A cholecystectomy was performed. The accessory liver lobe with the perforated bile duct was resected as shown in figure 3 and sent for histopathological examination. The operative field after resection of the accessory liver lobe is shown in figure 4. Due to unavailability of an intraoperative choledochoscopy, the distal CBD could not be examined for any stones. A thorough abdominal wash was done prior to closure.



**Figure 4: Operative field after resection of accessory liver lobe; (A) left lobe of liver; (B) Spleen.**



**Figure 5: MRCP showing distal CBD calculus with proximal dilatation.**

No immediate post operative complications were noted. The histopathological examination of the resected accessory liver lobe was consistent with that of a hepatic tissue. With the persistence of jaundice on post operative day 5 and to find the probable cause of biliary obstruction leading to the perforation, patient underwent a magnetic resonance cholangiopancreatography (MRCP) only to reveal a distal CBD stone of size 8 mm with proximal dilatation as shown in figure 5. Patient underwent endoscopic retrograde cholangiopancreatography (ERCP)

with stone extraction on post operative day 10. A repeat liver function test done 4 days later showed significant improvement and patient was discharged. Patient is on regular monthly follow up with no complications till date.

## DISCUSSION

Spontaneous bile duct perforation is an uncommon entity encountered in surgical practice. With the first case published by Freedland in 1882, there have been many attempts at explaining the pathophysiology of such occurrences to aid in early diagnosis and treatment.<sup>3</sup> Though encountered occasionally in infancy and childhood as a cause of jaundice, its occurrence in adults is extremely rare.<sup>4,5</sup> It is categorized into perforation of the intrahepatic bile duct and perforation of the extrahepatic bile duct of which the extrahepatic variety is much more common.<sup>5,6</sup>

There are many etiological proposals given to explain the occurrence of spontaneous bile duct perforation such as; bile duct obstruction due to stones, tumors or stricture with raised intraductal pressure; stones or tumors directly eroding the bile duct wall; bile duct diverticulum; vascular insufficiency to biliary tree.<sup>7-10</sup> It is seen that bile duct stones is the most common cause of bile duct perforation.<sup>11</sup> The raised intraductal pressure proximally in the biliary tree leads to spontaneous perforation at vulnerable points.

In our case, we propose that the occurrence of CBD stone with an accessory liver lobe predisposed the patient for spontaneous intrahepatic bile duct perforation. Progression of this pathology to biliary peritonitis and sepsis increases the morbidity and mortality. A high index of clinical suspicion in patients presenting with severe right upper quadrant abdominal pain, distension and jaundice with signs of peritonitis should warrant a diagnosis of biliary peritonitis due to bile duct perforation. Ultrasound findings would indicate peritoneal collection with decompression of the gall bladder due to bile leak through the perforation. Patients with established biliary peritonitis require exploratory laparotomy with a CBD exploration and T tube drainage followed by a cholecystectomy.

An intraoperative choledocscopy may facilitate the identification of CBD stones which may go unnoticed. The site of perforation in the intrahepatic bile duct can be sutured with delayed absorbable sutures. The presence of an accessory liver lobe predisposing to the spontaneous perforation makes this case unique. Such occurrences warrant a resection of the accessory lobe including the site of biliary perforation.

With the advent of endoscopy techniques, a noninvasive approach to the management of bile duct obstruction is preferred.<sup>12</sup> But the presence of biliary peritonitis warrants a laparotomy with the intraoperative finding guiding the surgery. Follow up of such patients with liver function tests is essential to note the decline in bilirubin levels,

failure of which would necessitate an MRCP to confirm the pathology.

## CONCLUSION

Spontaneous intrahepatic biliary perforation with an accessory liver lobe is an exceedingly rare condition making preoperative diagnosis difficult leading to delay in management. Awareness of such occurrences is necessary to diagnose and manage such patients without delay. All patients with intrahepatic bile duct perforation must be investigated for the primary pathology. Early diagnosis and prompt treatment gives a favorable prognosis.

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