Introducción: Entre un amplio abanico de causas de abdomen agudo, la perforación espontánea de las vías biliares (PECBD) resultante en peritonitis biliar a menudo no se ha considerado. Desde que el término PECBD se ha malinterpretado como falta de una causa identificable de perforación, ‘perforación no traumática de la vía biliar’ también es en parlance para excluir las causas relativamente comunes como trauma e heridas operatorias. En adultos, el cistitis choledócalica, la cholangitis, la infección, la pancreatitis, la malposición biliar pancreática han sido identificados como causas de perforación, sin embargo, la choledoco litiasis sigue siendo la causa más común asociada con la perforación espontánea de las vías biliares extrahepáticas.

Presentación del caso: Aquí presentamos tres casos de perforación espontánea de la vía biliar común que se presentaron como abdomen agudo con peritonitis. Se estudiaron los detalles clínicos, los hallazgos bioquímicos, los detalles de la imagen, las opciones de tratamiento y el resultado. Este estudio ha sido reportado de acuerdo con la guía SCARE 2020 [1].

Discusión y conclusión: La perforación espontánea de las vías biliares extrahepáticas es un evento raro pero importante de las gotas de las vesículas en los adultos y requiere un alto índice de sospecha durante la práctica diaria. Los cirujanos deben buscar este diagnóstico poco común en los pacientes para una intervención quirúrgica temprana y un drenaje apropiado para garantizar la supervivencia.

1. Introducción

La perforación espontánea de la vía biliar es un hecho raro. Ha sido principalmente reportado en lactantes y niños con anormalidades como cistitis choledócalica y malposición pancreática biliar [2]. Sin embargo, en adultos esta entidad es muy rara y difícil de diagnosticar preoperatoriamente. Tales eventos se clasifican como la perforación de las vías biliares intrahepáticas y perforación de las vías biliares extrahepáticas. Las vías biliares intrahepáticas pueden estar localizadas en la superficie del hígado, mientras que las vías biliares extrahepáticas pueden estar localizadas en cuatro áreas (la vía ductal derecha hepática, la vía ductal izquierda hepática, la vía ductal común hepática y la vía ductal biliar común). Las perforaciones espontáneas son difíciles de diagnosticar preoperatoriamente [3]. Muchos pacientes son diagnosticados como teniendo queratitis o choledoco litiasis preoperatoriamente. Según los informes anteriores de los casos adultos, puede haber relación con un solo o múltiples factores que incluyen obstrucción por calculos, necrosis infectiva y aumento de la presión intraductal [4]. El ascitis biliar es un evento común de perforación espontánea de la vía biliar reuniendo con pringamiento abdominal progresivo, ascitis de las vesículas que se pueden localizar o generalizar, ictericia o septic shock, una fluctuación del curso, y evidencia de ácido bilis con desequilibrio de la función hepática. El lavado peritoneal, la presencia de bilis, el signo de peritonitis y la ausencia de ascitis del peritoneo en el rayo X favorecería la sospecha de PECBD. Una recuperación preoperatoriamente o al menos una fuerte sospecha para perforación espontánea debería ser bien descrita en el paciente para el tratamiento quirúrgico adecuado y asegurar la supervivencia.

2. Presentación del caso

Manejamos a tres pacientes con perforación espontánea de la vía biliar no traumática en nuestro centro entre 2019 y 2021. El rango de edad del paciente oscilaba entre 45 y 55 años. Ninguno de ellos tenía una historia de operación previa en el tracto hepatobiliar. No traumática...
perforation of the bile duct was suspected with the observation of bile-tinged peritoneal fluid or after drainage of bile from the perihpatic space.

All the patients had acute abdominal pain, which was diffuse in nature with distention due to the biliary ascites in one patient or localized due to abscess in one patient (Tables 1 and 2).

2.1. Patient 1

55 year old woman presented in emergency with diffuse abdominal pain and distention of 1 week duration. Per abdomen revealed tenderness of whole right abdomen. Ultrasound abdomen suggested cholelithiasis with choledocholithiasis with proximal Common bile duct and Intra hepatic biliary radicle dilated with collection in perihpatic space. Contrast enhanced computed tomography abdomen revealed thickened GB wall, choledocholithiasis, dilated proximal CBD and left IHBR. Patient underwent exploratory laparotomy wherein omentum was found adhered to contracted gall bladder and a CBD perforation of size $\frac{1}{2} \times \frac{1}{2}$ cm at anterolateral surface of supraduodenal part was present. On enlarging the perforation, single calculus of size $2 \times 2$ cm was delivered. Multiple small calculi present in gall bladder, surrounding organs were bile stained.

CBD was repaired over a T-Tube. The postoperative recovery was uneventful. A T-tube cholangiogram was performed on the 14th post-operative day, which showed normal free flow of bile into the duodenum and jejunum without any filling defect. Histopathology study of the gall bladder showed chronic inflammation. Patient was discharged on the 20th post-operative day.

2.2. Patient 2

45 years male presented with complaints of jaundice for 6 months, pain abdomen and fever for 5 days. Per abdomen examination revealed features of peritonitis. However, pneumoperitonium was absent. MRCP suggested chronic cholecystitis with cholelithiasis, choledocholithiasis with calculi at distal end of common bile duct resulting into proximal dilated CBD and IHBR dilated (Fig. 1).

CECT W/A suggested cholelithiasis with the collapsed wall showing irregular margins with moderate ascites suggestive of GB perforation. After clinical and radiological study suggestive of GB perforation, USG guided percutaneous abdominal drain was placed, the content was bile stained. About 1.5 l bile was drained. Due to persistent pain and progressively deteriorating clinical condition, a decision to explore was made.

On exploration, about 0.5 l of bile in peritoneal cavity along with pus flakes and staining of whole of small bowel was found. The gall bladder was thickened with a large calculus within (Fig. 3).

Perforation of size $\frac{1}{2} \times \frac{1}{2}$ cm in diameter was detected at anterolateral surface of supra duodenal part of common bile duct with 2 calculi, largest of $2 \times 2$ cm was removed. Cholecystectomy was done and CBD was repaired over T-tube as the CBD was too fragile and friable and thickened. The postoperative recovery was uneventful.

A T-tube cholangiogram was performed on the 16th post-operative day, which showed normal free flow of bile into the duodenum and jejunum without any filling defect (Fig. 4).

3. Discussion

Spontaneous perforation of the common bile duct is a rare cause of peritonitis. It is seldom suspected or diagnosed preoperatively owing to its uncommon occurrence. Perforation of the biliary system is a recognized complication of cholelithiasis and choledocholithiasis; the diagnosis should be suspected if a perihpatic abscess or peritonitis is combined with a biliary calculous [6].

Spontaneous perforation of the bile duct unrelated to trauma or surgical complications is a rare condition that is more often seen in infants and children [7]. Such events are classified into perforation of the intrahepatic bile duct and perforation of the extrahepatic bile duct. Freeland reported the first case of spontaneous hepatic duct perforation in 1882 in a case that was diagnosed on autopsy [8]. To date, only about 22 cases of intrahepatic bile duct perforation and 50 cases of extrahepatic bile duct perforation have been reported [9]. The etiologic factors proposed to be causative of spontaneous perforation of extrahepatic bile duct are erosion by biliary stones directly through the duct wall, obstruction of the distal bile duct and increased intraductal pressure, vascular thrombosis supplying the duct wall, intramural infection of the duct as a result of cholangitis, diverticulitis of the bile duct, and carcinomas arising in the hepato-biliary-pancreatic organs [10]. A combination of these factors is probably responsible for most bile duct perforations. As seen in literature, choledocholithiasis is the most frequent cause spontaneous perforation.

Perforation of the common bile duct was most probably related to the abrupt increase in local intraluminal pressure causing erosion of the stone. This results in an abrupt increase in intraluminal pressure and decreased blood flow in the vessels which run along the lateral border of the bile duct resulting in ischemia on the anterior surface of the bile duct [11]. This might explain the site of supraduodenal CBD perforation in our patients [12].

A pattern of presentation in all our patients was in the form of abdominal distention and ascites. Aspiration of ascites revealed bile-stained fluid which was confirmed to be bilius on dipstick examination for biliary and lab evaluation of fluid. All patients had tense tender abdomen, abdominal ultrasonography of all patients suggested gallstone and ascites being the most consistent finding.

### Table 1

| Characteristic | Patient 1 | Patient 2 | Patient 3 |
|---------------|-----------|-----------|-----------|
| Hb            | 9.7       | 9.8       | 12.6      |
| WBC           | 8300      | 12,086    | 12,500    |
| T. Bilirubin(D) | 1.9(1.4)  | 14.5(13.5)| 23.4(16.7)|
| ALP           | 565       | 804       | 1060      |
| AST/ALT       | 36/46     | 343/490   | 250/390   |
| Amylase/Lipase| 64/32     | 150/120   | 250/160   |
| PT-INR        | 14/1.2    | 18/1.5    | 19/1.5    |
| S.urea/S.creatinine | 1.8/0.6 | 20/1.3   | 33/8.7    |
Choledocholithiasis is the most common underlying cause found in spontaneous bile duct perforation in adult population. Loculated intra-peritoneal collection was a consistent finding. CT scan was done in all patients, it confirmed the presence of free fluid and dilated gallbladder in one patient while the other two cases showed loculated collection; facilities for the intraoperative cholangiogram or ERCP were not available. MRCP was not able to conclusively diagnose CBD perforation. ERCP may also not be able to suggest perforation of extrahepatic biliary tree with authority at times, leaving the surgeon in a lurch whether to go ahead with exploration [13]. All patients underwent t-tube drainage of the common bile duct along with cholecystectomy because of friability and inflammation around the perforation site. Intense inflammation around the perforation site associated with edematous and friable CBD makes primary repair of CBD over T-Tube a quick and safe procedure.

It appears prudent to avoid biliary enteric procedures, if possible, as widespread peritonitis coupled with friable CBD would result in inferior outcomes.

In addition, a patient of sepsis with a lengthy intraoperative procedure would be associated with septicemic complications in the post-operative period.

The goal of the treatment, which should be tailored to individual patients, is to stop bile leakage, address choledocholithiasis and cholangitis and divert the bile duct. An optimal operation should consist of a cholecystectomy, intraoperative cholangiogram, bile duct exploration,

Table 2
Clinical characteristic and operative management.

| Case/age/sex | Manifestation | Primary diseases | Perforation site/size(cm) | Management |
|--------------|--------------|-----------------|--------------------------|------------|
| 1/55YRS/F    | Acute abdomen| Gall stone with CBD stones | Anterolateral wall CBD/0.5*0.5 | CBDE, T-Tube |
| 2/45/M       | Bile peritonitis | CBD stone | Anterolateral CBD/0.5 *0.5 | CBDE, T-Tube |
| 3/46/M       | Subcapsular bilioma with peritonitis; imaging suggestive of infected pseudocyst | CBD stone | Lateral wall CBD/2*1 | CBDE, T-Tube |

Fig. 1. MRCP suggested chronic cholecystitis with cholelithiasis, choledocholithiasis with calculus at distal end of common bile duct resulting into proximal dilated CBD and IHBR dilated.
repair of the perforated site, and t-tube insertion. The site of t-tube insertion depends on the accessibility of perforation. T-tube insertion is crucial for the successful treatment of spontaneous perforation of the bile duct. Most such perforations occur on the anterior wall of the bile duct [14,15]. Bile duct stone can be easily removed after enlarging the perforation and the t-tube inserted during the treatment of such perforation. Surgical management is more difficult when the perforation occurs on the other side of the wall. If a large defect is present, a more invasive operation may be considered such as choledochojejunostomy, hepaticojejunostomy, hepaticoduodenostomy, or Gall bladder wall flap [16-18].

Until the 1960s, the mortality rate for bile duct perforation was estimated to be 50%. The introduction of current examination, progress in endoscopic treatment, interventional radiology, surgical techniques, and antibiotics have contributed to a significant decrease in the mortality rate since the 1970s [19,20].

4. Conclusion

The presence of jaundice, rather deep jaundice and cholestasis associated with an acute abdomen should alert the clinician to be more inquisitive and be aware that biliary system may be the culprit. Spontaneous CBD perforation is a rare but potentially fatal condition in adults. When SCBDP is an intraoperative diagnosis, it catches the surgeon unawares and the daunting task of dealing with a bile duct perforation in a hostile abdomen is challenging. An acute abdomen accompanied with jaundice or cholestasis should ring a bell that

Fig. 2. CECT W/A suggested cholelithiasis with the collapsed wall showing irregular margins with moderate ascites suggestive of GB perforation.

Fig. 3. Intraoperative image revealed perforation at Anterolateral surface of supraduodenal CBD with bile-stained small bowel and peritoneal cavity.
additional investigations are mandated before a decision to explore is made. Deteriorating clinical condition of the patient may be the only clue to go ahead with exploration. Awareness of the clinical presentation and ultrasound examination demonstrating cholelithiasis/cholecystolithiasis are important adjuncts in the diagnosis. Early surgical intervention is probably the only measure to prevent mortality and morbidity.

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Dr. Anjay Kumar—constructing an idea or hypothesis for manuscript, analysis and interpretation, critical review.

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**Declaration of competing interest**

The author declared that there is no conflict of interest.

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