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Acute Abdomen Secondary to a Spontaneous Perforation of the Biliary Tract, a Rare Complication of Choledocholithiasis



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ABSTRACT

INTRODUCTION: The spontaneous perforation of the biliary tract (SPBT) is an extremely rare cause of peritonitis, which was first described by Freeland in 1982, to date only around 70 cases have been reported. Here we present a case of spontaneous perforation of the biliary tract, in a patient with choledocholithiasis, which was treated with ultrasound-guided drainage and ERCP.

CASE REPORT: A 51-year-old male was admitted to the emergency room for 15-day evolution jaundice, localized pain in the right flank and hypochondrium of 3 days. He had a history of cholecystectomy 15 years ago and 4 episodes of cholangitis, the last one in 2015. A magnetic resonance imaging (MRI) was performed, that showed evidence of choledocholithiasis, in addition to a possible biliary leakage. The patient was treated with ultrasound-guided drainage and ERCP successfully.

DISCUSSION: Spontaneous perforation of the biliary tract is a disease entity in which wall of the extrahepatic or intrahepatic duct is perforated without any traumatic or iatrogenic injury. The clinical presentation varies from nonspecific abdominal pain to biliary peritonitis, in most of the cases forming bilomas. Universal management involves decompression of the biliary tree and repair of the leak site.

CONCLUSION: The spontaneous perforation of the biliary tract is a disease that represents a diagnostic challenge. The treatment in the patients with SPBT is not well established and has to be individualized for each case, depending on the history of the patient, the site of perforation, the time of evolution, the suspicion of infection, and the patient status.

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1. Introduction

Abdominal pain is one of the most common reasons for visits to the emergency room, comprising 7–8% of all visits [1,2]. Typically the patient is admitted to the emergency department with abdominal pain and a systemic inflammatory response, including fever, tachycardia, and tachypnea; abdominal rigidity suggests the presence of peritonitis [3]. The most common causes of peritonitis are appendicitis, cholecystitis, postoperative complications, and colonic non-diverticular perforation, among others.

The spontaneous perforation of the biliary tract (SPBT) is an extremely rare cause of peritonitis [4–7], which was first described by Freeland in 1982, to date only around 70 cases have been reported [5–7].

The precise cause of SPBT is unknown; it has been explained by obstruction leading to excessive pressure and duct compromise in areas of weakness [4,5,8,9], 70% of the cases are related to choledocholithiasis [14].

Since the symptoms and signs are not specific, the diagnosis is often delayed. So that, in most of the cases the diagnosis is made upon surgery [6,10–12]. Here we present a case of spontaneous perforation of the biliary tract, in a patient with choledocholithiasis, which was treated with ultrasound-guided drainage and ERCP. The following case has been reported in line with the SCARE criteria [13].

2. Case presentation

A 51-year-old male was admitted to the emergency room for 15-day evolution jaundice, localized pain in the right flank and hypochondrium of 3 days, and poor food tolerance. He had a history of two episodes of cholangitis resolved with ERCP, with subsequent cholecystectomy 15 years ago. In 2014 he developed severe cholangitis, entering the ICU, where another ERCP was performed. He was admitted again in 2015 with grade II cholangitis treated with ERCP and stent placement, which was withdrawn later that same year.

On physical examination, we found a conscious and oriented patient, tachycardic, with jaundice. The abdomen was distended, with decreased intestinal peristalsis, depressible, painful upon

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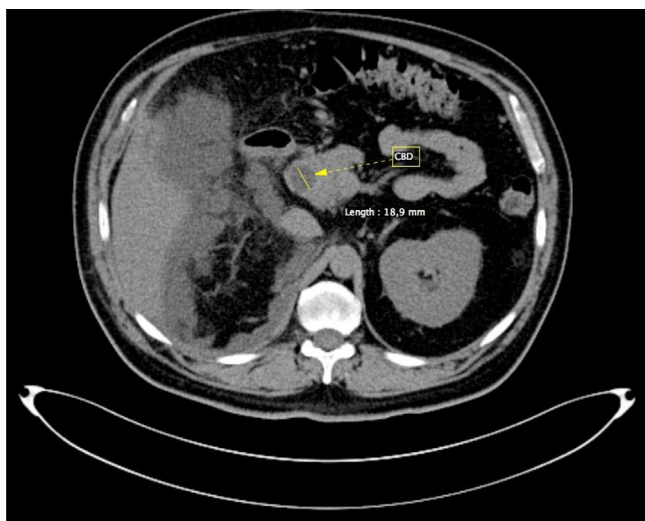


Fig. 1. CT scan shows free peri-renal fluid, in the Morrison space and, as well as dilation of the common bile duct (CBD).

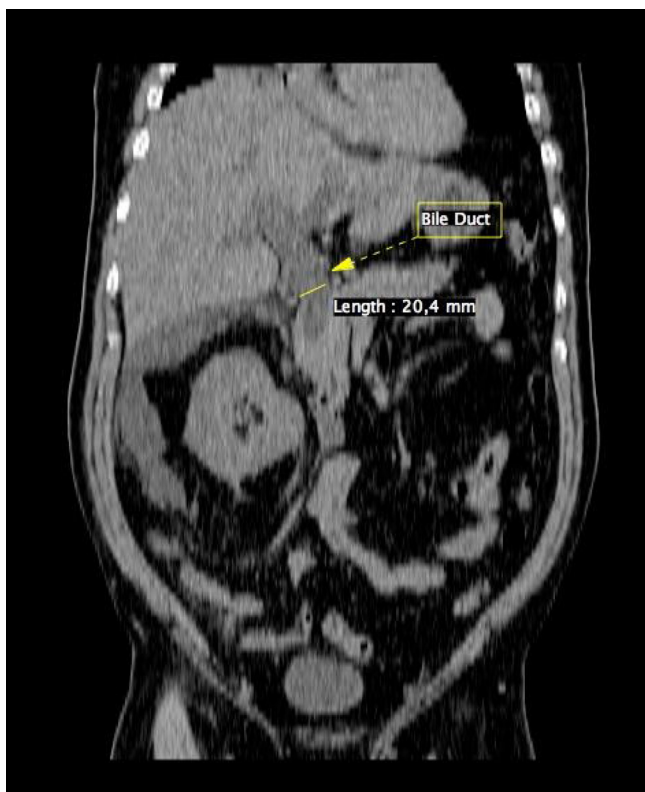


Fig. 2. CT scan coronal section. It shows dilation of the intra and extrahepatic biliary tract, as well as free fluid, in the right parieto-colic gutter.

superficial and deep palpation mostly in the right flank and hypochondrium, and rebound tenderness.

The laboratory test showed leukocytosis of 22,640/ μ l with 88.39% neutrophils. Procalcitonin of 9.26 ng/ml. Total bilirubin of 5.55 mg/dl, ALP 94 U/l, GGT 135 U/l; amylase and lipase within normal parameters.

A computed tomography (CT) scan was performed, which showed free peri-renal fluid in the Morrison space and parieto-colic gutter, as well as dilation of the intra and extrahepatic biliary tract (Figs. 1 and 2). Therefore it was decided to perform an ultrasound-guided drainage with a pigtail catheter, obtaining 200 ccs of biliary

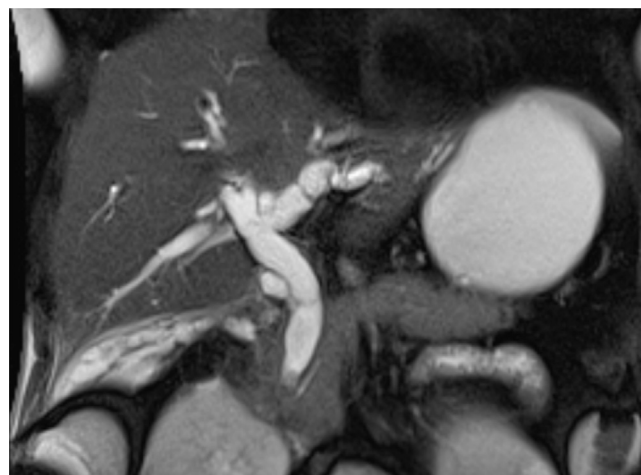


Fig. 3. MRC in T2 w shows choledocholithiasis in the distal third of the common bile duct, in addition to possible biliary leakage in the middle third.

fluid on the first day. The next day a magnetic resonance imaging (MRI) was performed, that showed evidence of obstruction of the biliary tract secondary to probable choledocholithiasis, in addition to a possible biliary leakage in the middle third (Fig. 3).

The patient was sent to ERCP, where the gallstone was removed with no evidence of biliary leakage. He was discharged 9 days after his admission without systemic inflammatory response and with a biliary drainage of about 50 ccs. The catheter was removed 7 days later during an outpatient visit, without further complications.

3. Discussion

Spontaneous perforation of the biliary tract is a disease entity in which wall of the extrahepatic or intrahepatic duct is perforated without any traumatic or iatrogenic injury [5]. The gall bladder, common bile duct, common hepatic duct and anomalous ducts of the liver are specific sites of biliary compromise; in most of the cases they are extrahepatic and are found frequently at the junction of the cyst duct and the common bile duct [6,9]. In our case, the leak was located in the middle third of the common bile duct.

Although the precise cause of SPBT is unknown, there are some factors that can be involved, such as ischemia, elevated pressure within bile ducts, congenital weakness of the bile duct wall and pancreaticobiliary reflux [4,5,8,9]; 70% of the cases are related to choledocholithiasis [14].

One of the mainstays in the treatment of choledocholithiasis is the ERCP with endoscopic sphincterotomy. Even though the endoscopic sphincterotomy brings a risk of long term complications (5.8–24%), including recurrent duct stones and cholangitis [15,16], the late complication rate after repeat ERCP is higher representing 36% [16]. Our patient had a history of 4 previous ERCP and previous episodes of cholangitis, which could have caused weakness of the bile duct wall. In addition to a current diagnosis choledocholithiasis and a probable cholangitis as a risk factor, which may have triggered the SPBT.

The clinical presentation of SPBT varies greatly from nonspecific abdominal pain to biliary peritonitis, without a characteristic history [4,9,10,17]. In most of the cases, the encapsulation of the bile within the omentum and mesentery prevents generalized peritonitis, forming bilomas; which are generally localized in the right upper quadrant of the abdomen. As the bile is sterile and is absorbed by the peritoneum, the patients may not present symptoms for weeks, until the bile becomes superinfected [10,18]. Thus the diagnosis of biliary tract perforation is often delayed, which results in high morbidity [6].

Several imaging modalities can be used in the diagnosis of biliary leaks and bilomas. These modalities include ultrasound (US), computed tomography (CT), Magnetic resonance imaging (MRI), and nuclear medicine hepatobiliary cholescintigraphy [18,19].

The US is often the initial diagnostic modality and can show an anechoic well-circumscribed fluid collection. A complex loculated collection with internal septations is suggestive of infection [18,20]. The CT can show discrete fluid collections with or without surrounding peripheral capsule; commonly the density is less than 20 Hounsfield units. Furthermore, the CT is an excellent study to identify a collection and assess the surrounding anatomy [18–20].

On an MRI, a biliary leak or a biloma appears as a hypointense signal on T1 w and a hyperintense signal on T2w. The Magnetic Resonance Cholangiography (MRC) can also be helpful in demonstrating communications between the biliary ducts and a fluid collection. The accuracy rate of diagnosis and localization of an extravasation of bile by T2 w in MRC is within the range 70–74% [21]. The Hepatobiliary cholescintigraphy is able to demonstrate continuity of fluid collections with the biliary tree, nevertheless, this modality does not provide highly detailed anatomy; so identifying the precise location of the leak can be difficult [20].

The differential diagnosis should include hematoma, seroma, liver abscess, cyst, pseudocyst, and lymphocele. Percutaneous aspiration, which is usually performed utilizing an 18–22 gauge co-axial needle under CT or US guidance, can also aid in diagnosis and treatment; but in most of the cases the diagnosis is made upon surgery [10–12,20].

Universal management involves decompression of the biliary tree and repair of the leak site [9]. Formerly the treatment used to be an open or laparoscopic surgical approach [5,8]. Nowadays, the percutaneous drainage of the collection and endoscopic modalities are preferable to surgery as the first step in treatment for patients who are stable and without peritonitis [5,8,10,22]. The procedure for repairing the perforation is not always necessary; spontaneous closure has been observed in case reports [5], such is the case of our patient.

4. Conclusion

The spontaneous perforation of the biliary tract is a disease with nonspecific symptoms and represents a diagnostic challenge. The SPBT should be suspected in a patient with a history of biliary disease that presents an acute abdominal pain and a CT scan or US with compatible findings of biloma. The performance of an RMC is helpful for identifying the site of injury in the biliary tract and can be determinant to decide the treatment modality, so it must be done if the resource is available. The treatment in the patients with SPBT is not well established and has to be individualized for each case, depending on the history of the patient, the site of perforation, the time of evolution, the suspicion of infection, and the patient status. In our patient, we performed drainage of the biloma and ERCP as the treatment because of the history of previous cholecystectomy, the presence of choledocholithiasis and hemodynamic stability. In a patient with gallbladder disease, the laparoscopic modality can be used to perform a cholecystectomy and a bile duct exploration, with or without placing of a “t” tube. In all cases, the treatment must involve the decompression of the biliary tract and drainage of the biloma. As to our knowledge, this is the first case of spontaneous perforation of the biliary tract treated with ultrasound-guided drainage and ERCP successfully reported in México.

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Conflicts of interest

None.

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Ethical approval

The written consent was sign by the patient

Consent

The written consent was sign by the patient.
No personal information is given nor modified.

Author contribution

Gómez-Torres GA.- writing the paper, final decision to publish, data collection and analysis.
Rodríguez-Navarro FM.- data collection, and analysis.
López-Lizárraga CR.- data collection, and analysis.
Bautista López CA.- data collection, and analysis.
Ortega-García OS.- writing the paper, data collection and analysis.
Becerra-Navarro G.- data collection, and analysis.
Águila-Barragán A.- data collection, and analysis.
Ploneda-Valencia CF.- writing the paper, data collection.

Guarantor

Gustavo Ángel Gómez Torres.

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