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Spontaneous common bile duct perforation—A rare clinical entity

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ABSTRACT

INTRODUCTION: Spontaneous common bile duct perforation is an uncommon clinical entity in both adults and children. Few case reports have been published since the first clinical description in 1882. Our work has been reported in line with SCARE criteria.
PRESENTATION OF CASE: Herein, we describe the case of a 28 year-old female who suffered spontaneous common bile duct perforation while admitted for choledocholithiasis.
DISCUSSION: The perforation occurred while in-hospital, and extensive imaging and laboratory tests characterized the disease in detail. To our knowledge, this is the first report of spontaneous common bile duct perforation witnessed from pre-perforation through definitive management.
CONCLUSION: Physicians and Surgeons should seek out this uncommon diagnosis in the patient with suspected Choledocholithiasis who suddenly become peritoneal on physical exam so that definitive care can be expedited.

1. Introduction

Spontaneous common bile duct perforation is an uncommon clinical entity in both adults and children. Few case reports have been published since the first clinical description in 1882.

Because of its relative rarity, diagnosis has typically been confirmed during surgical exploration of the abdomen, often undertaken for other suspected pathology such as a perforated viscus. Mortality is often elevated as the diagnosis is most commonly reported in frail and elderly individuals under going surgical exploration.

In this case report, we describe spontaneous choledochal perforation believed to be due to impacted choledocholithiasis in a young woman with diagnosis made prior to surgical exploration.

2. Presentation of case

A 28 year-old female presented to the emergency department, complaining of postprandial epigastric pain that was associated with nausea and emesis. She denied fever and chills. Her past medical history was significant for gastroesophageal reflux disease as well as hypothyroidism. Abdominal exam was non-tender with negative Murphy's sign. Laboratory studies were significant for transaminitis: AST 343 mg/dL and ALT 490 mg/dL. However, canalicular and pancreatic enzyme and leukocyte levels were within normal levels.

Computed tomography (CT) revealed dilated common as well as extra- and intrahepatic ducts without free fluid or other acute abdominal findings. Right upper quadrant ultrasound demonstrated cholelithiasis without cholecystitis and 10 mm dilated common bile duct (CBD).

On hospital day one, her symptoms markedly worsened with new abdominal distension, diffuse abdominal tenderness, and voluntary guarding in the left lower and left upper quadrants. Magnetic resonance cholangiopancreatography revealed pericholecystic fluid, CBD reduced to 6 mm diameter, resolved biliary ductal dilation, edema within the head of the pancreas, and no evidence of choledocholithiasis. Complex ascites was also now present in the upper abdomen and left paracolic gutter (Fig. 1).

Laboratory studies also demonstrated a marked change. Canalicular enzymes were slightly elevated: total bilirubin 1.4 mg/dL, direct bilirubin 0.8 mg/dL and alkaline phosphatase 135 IU/L. Complete blood count demonstrated significant leukocytosis of 13,100 WBCs/uL with a left shift of 91.5%. Amylase and lipase were also elevated at 1702 IU/L and 1737 IU/L, respectively.

Plain radiographs were negative for pneumoperitoneum. Repeat CT abdomen and pelvis with intravenous and oral contrast demonstrated a large volume of ascites, thickened gallbladder wall, pericholecystic fluid, resolved gallbladder distension and CBD dila-

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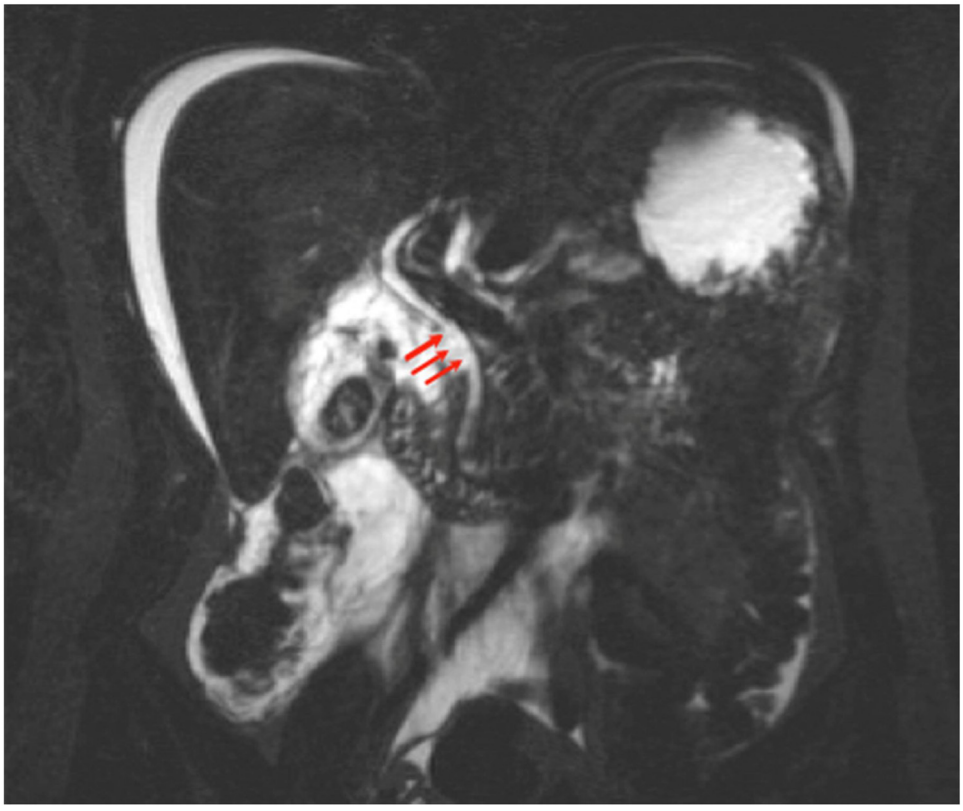

Fig. 1. MRCP showing probable CBD necrosis (RED ARROWS) with bilious ascites.

tion, possible perforation of the CBD, as determined by incomplete visualization of circumferential CBD with large amount of fluid in lesser sac. There was no evidence of oral contrast extraluminal extravasation (Fig. 2A and B).

Hepatobiliary scintography (Fig. 3) revealed tracer accumulation in the subhepatic and perihepatic spaces, indicating bile leak. Image guided paracentesis aspirated 30cc of bilious fluid, and an 8.5 French catheter was placed for further drainage. Fluid studies on the aspirated fluid revealed amylase of 9750 IU/L with negative cultures.

Endoscopic retrograde pancreatography could not localize biliary ductal perforation to either the CBD or common hepatic duct. A covered stent placed across the perforation allowed adequate bile efflux from the liver without extravasation of injected contrast.

Two weeks of nonoperative management led to minimal improvement in clinical condition, and subsequent operative exploration revealed necrotic CBD with an intact, healthy gallbladder. The CBD was resected and a roux-en-y choledochojejunostomy with common hepatic duct to jejunum anastomosis was created. The patient recovered well.

3. Discussion

Non-traumatic perforation of the extrahepatic bile ducts in adults is an uncommon clinical entity. The disease more commonly occurs in infants due to associated congenital anomalies such as choledochal cysts [8] with an incidence of 1.5 per 1,000,000 live births; from 1990 to 2015, 90 cases were reported in infancy [5].

As of 2004, non-traumatic perforation of the extrahepatic bile duct among adults had been reported only 70 times in the English literature since its first description by Freeland in 1882 [6]. A Pubmed search of reports published after 2004 revealed an additional 27 cases, bringing the total case reports to approximately 97 cases in the past 135 years. With very few adult cases reported, prevalence among adults remains uncertain [10]. However, one researcher approximates the mortality rate to be 30–50%, owing to comorbidities in the elderly who comprise the majority of case reports [1].

The diagnosis of spontaneous CBD perforation is difficult to establish and often delayed due to nonspecific symptomatology [4] and often undiagnosed prior to laparotomy [3,6]. Despite advances in intensive care management, delayed treatment still results in high morbidity and mortality [6,10].

Our case represents a unique presentation in that CBD perforation occurred within the hospital after admission for choledocholithiasis, allowing for multiple imaging studies (CT scan, HIDA scan, and MRCP) and treatment modalities from pre-perforation to definitive management of spontaneous CBD perforation; to our knowledge, all previous reports were performed in patients arriving to the hospital after perforation of the biliary system. This is an exclusive presentation for this condition as many of the case reports occur in frail elderly patients at the time of laparotomy and are not diagnosed prior to surgical intervention.

Furthermore, the lab abnormalities present during work up after the perforation occurred are not surprising after the diagnosis of perforation to the biliary tree. However these lab results also commonly occur in other clinical entities such as gallstone pancreatitis and perforated peptic ulcerations. Imaging abnormalities witnessed in our patient and other previous case reports in conjunction with this pattern of laboratory parameters (canilicular enzyme elevations in combination with elevated amylase and lipase) should doubly prompt suspicion for a primary perforation of the biliary tree.

The pathogenesis of spontaneous bile duct perforation is poorly understood, likely related to its rarity. It is currently thought to be related to multiple factors including increased intraductal pressure, fluid stasis, dilation of the bile duct (due to distal obstruction or spasm of the sphincter of Oddi), diverticulum, abnormal glands

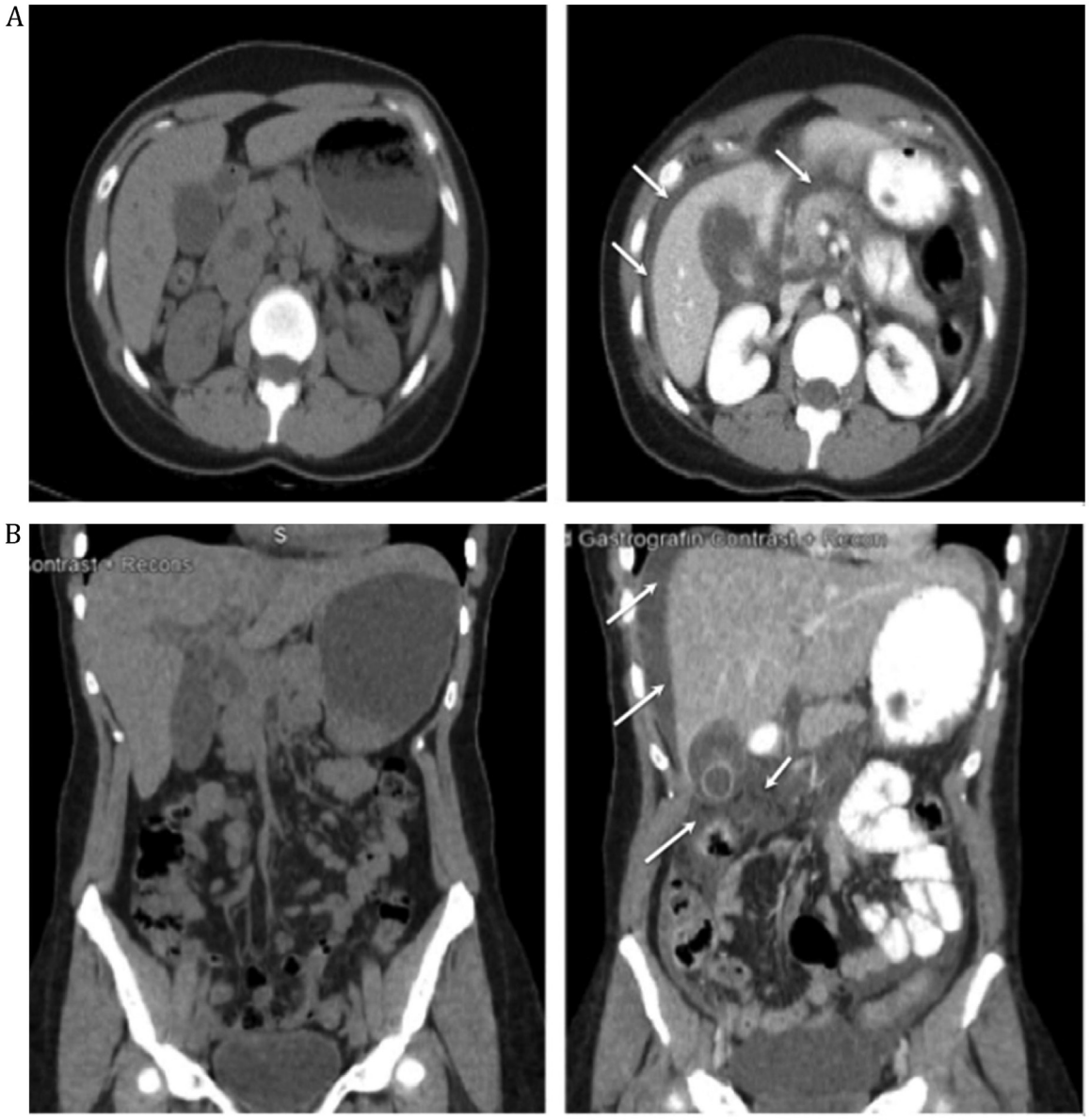

Fig. 2. A: Side by side comparison of axial slices of abdominopelvic computed tomography (CT A/P). Left image is admission CT scan, Right image is repeat CT after common bile duct perforation with new demonstration of ascites (WHITE ARROWS). B: Side by side comparison of coronal cuts of CT A/P. Left image from admission CT and Right image from repeat CT after perforation with ascites in lesser sac (WHITE ARROWS).

in the bile duct wall, infection of the bile duct, a connective tissue defect, or ischemic compromise, and occasionally malignancy [1,4,6,11].

In 70% of reported cases, ductal stones are associated with the perforation [1,11] In our case, we attribute the perforation to probable ischemic compromise of the duct from an impacted stone within the CBD, despite the patient not showing signs of cholestasis on laboratory studies.

The most common site of extrahepatic bile duct perforation (excluding the gallbladder which complicates up to 10% cases of acute cholecystitis [2]) is the CBD (42 patients) followed by the common hepatic duct (28 patients) [6,11]. In one case series, nine of the 11 patients had intrahepatic ductal perforation [6]. When the perforation is described in the CBD, it is usually on the anterolateral surface of the supraduodenal portion of the CBD distal to the confluence of the cystic duct and the common hepatic duct [7–9].

Other case reports describe ascites of a similar distribution to our case, in the omental bursa [4,8], a location where ascites does not typically accumulate. In our patient this was seen on post-perforation CT scan as well as on MRCP. We suggest that evidence of ascites within the lesser sac should trigger an elevated suspicion for this rare diagnosis. Again similar to our case, in these cases, a diagnostic paracentesis was useful for establishing the diagnosis [4,8]. We suggest that when doubt exists clinically for perforation of the biliary tree, a paracentesis be performed in clinically stable patients prior to laparotomy to maximize effective surgical planning.

A high degree of suspicion must be maintained for this unusual clinical entity, especially for the patient that presents with abrupt change in exam and lab parameters with suspected choledocholithiasis as delay in diagnosis is associated with increased morbidity and mortality.

4. Conclusion

Physicians and Surgeons should seek out this uncommon diagnosis in the patient with suspected Choledocholithiasis who suddenly becomes peritoneal on physical exam so that definitive care can be expedited.

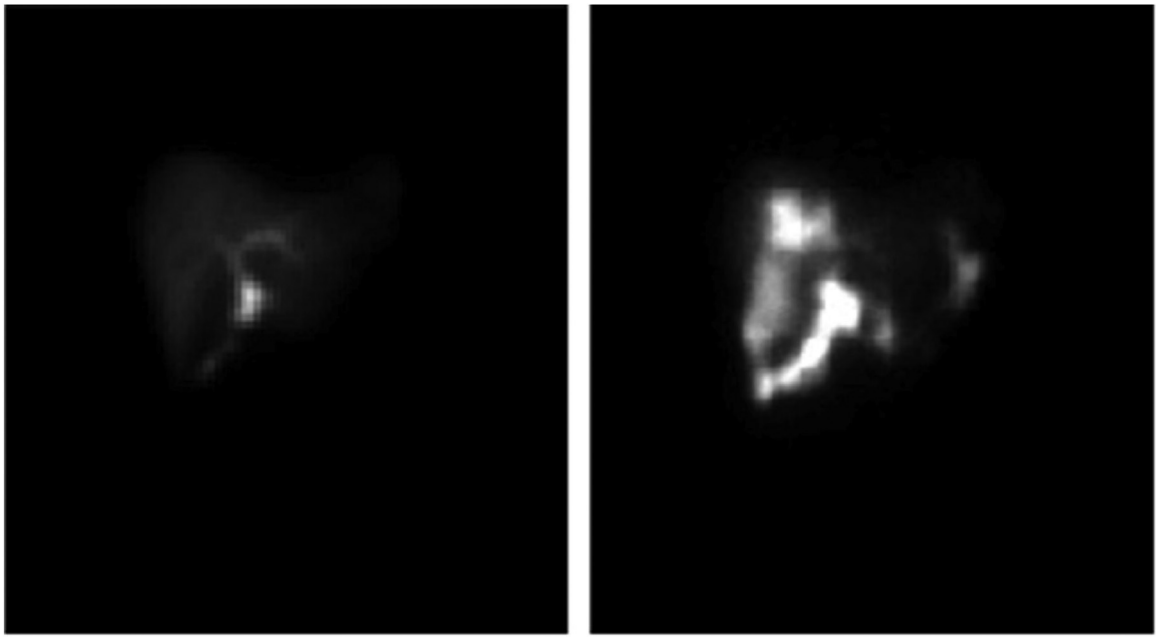

Fig. 3. Selected images from hepatobiliary scintography. Left image showing initial tracer filing right and left hepatic ducts. Right image with extravasation of tracer into morrison's pouch (hepatorenal recess) and into the right paracolic gutter.

Conflicts of interest

Each author declares no conflicts of interest.

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Ethical approval

Institutional Review Board approval was obtained for this research project (SBH IRB #2018.2).

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

MA is the primary author and contributed to writing, study concept and data collection.

NB and GT are the secondary author and contributed to writing and data collection.

GB is the tertiary author and contributed to writing, data collection and data interpretation.

SP is the primary investigator and contributed to study concept, data collection, data interpretation and writing the manuscript.

Guarantor

Shani Palmer MD, FACRS.

References

[1] Prosanta Kumar Bhattacharjee, Durjoy Choudhury, Himanshu Rai, Navin Ram, Debarati Chattopadhyay, Rajendra Prashad Roy, Spontaneous perforation of common bile duct: a rare complication of choledocholithiasis, Indian J. Surg. 71 (2) (2009) 92–94.

[2] N. Dabbas, M. Abdelaziz, K. Hamdan, B. Stedman, M. Abu Hilal, Gallstone-induced perforation of the common bile duct in pregnancy, HPB Surg. (2008) (2008) 1–3.

[3] Ambika P. Dash, Tapas Mishra, Manoranjan Mohapatra, M.D. Ibrarullah, Spontaneous perforation of bile duct, Trop. Gastroenterol. 33 (2) (2012) 151–153.

[4] Hamura Ryoga, Koichiro Haruki, Jun Tsutsumi, Sumio Takayama, Hiroaki Shiba, Katsuhiko Yanaga, Spontaneous biliary peritonitis with common bile duct stones: report of a case, Surg. Case Rep. 2 (1) (2016).

[5] Cerine Jeanty, S. Christopher Derderian, Shinjiro Hirose, Hanmin Lee, Benjamin E. Padilla, Spontaneous biliary perforation in infancy: management strategies and outcomes, J. Pediatr. Surg. 50 (7) (2015) 1137–1141.

[6] Sung-Bum Kang, Ho-Seong Han, Min Seog Ki, Hyeon Kook Lee, Nontraumatic perforation of the bile duct in adults, Arch. Surg. 10 (2004) 1083.

[7] Rahul Khanna, Nikhil Agarwal, Ajay Kumar Singh, Seema Khanna, Som Prakas Basu, Spontaneous common bile duct perforation presenting as acute abdomen, Indian J. Surg. 72 (5) (2010) 407–408.

[8] Mushtaq A. Laway, Iftikhar H. Bakshi, Mubbashir Shah, Showkat A. Paray, Mohd Sadiq Malla, Biliary peritonitis due to spontaneous perforation of choledochus: a case report, Indian J. Surg. 75 (S1) (2011) 96–98.

[9] D. Paramhans, Sapna Shukla, Jitendra Grover, Spontaneous perforation of the common bile duct in an adult, Indian J. Surg. 75 (S1) (2012) 376–378.

[10] Pulat Huseyin, Oktay Karaoke, Mehmet Fatih Benzin, Mehmet Safer Sabuncuoglu, Recep Metin, A rare cause of acute abdomen: spontaneous common hepatic duct perforation, Ulus. Travma Acil Cerrahi Derg. 22 (January (1)) (2016) 103–105.

[11] Subasinghe Duminda, Edippuli Arachchige Don Udayakumara, Upul Somathilaka, Milinda Huruggamuwa, Spontaneous perforation of common bile duct: a rare presentation of gall stones disease, Case Rep. Gastrointest. Med. (2016) (2016) 1–3.