



Case series

Acute abdomen with jaundice: A clue to extrahepatic biliary tract perforation

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ABSTRACT

Introduction: Among a plethora of causes of acute abdomen, spontaneous common bile duct perforation (SCBDP) resulting in biliary peritonitis is almost never envisaged. Since the term SCBDP is often misconstrued as absence of an identifiable cause of perforation, 'nontraumatic perforation of CBD' is also in parlance to exclude relatively common causes such as trauma and iatrogenic injuries. In adults, choledochal cyst, cholangitis, infection, pancreatitis, pancreatobiliary maljunction have been identified as causes of perforation, however, choledocholithiasis remains the most common cause associated with spontaneous perforation of extra hepatic bile duct.

Case presentation: Here we present three cases of a spontaneous common bile duct perforation that presented as acute abdomen with peritonitis. Clinical presentation, biochemical abnormalities, imaging details, treatment options, and outcome were studied. This study has been reported in line with the SCARE 2020 Guideline [1].

Discussion and conclusion: Spontaneous perforation of the extrahepatic bile duct is a rare but important presentation of gallstones in adults and needs a high index of suspicion during day-to-day practice. Surgeons should seek out this uncommon diagnosis in the patient for early surgical intervention and appropriate drainage to ensure survival.

1. Introduction

Spontaneous perforation of the bile duct is a rare entity. It has been mostly reported in infants and children with anomalies such as choledochal cyst and pancreatobiliary maljunction [2]. However, in adults this entity is very rare and difficult to diagnose preoperatively. Such events are classified as the perforation of the intrahepatic bile duct and perforation of the extrahepatic bile duct. Intrahepatic bile duct perforations are located at the surface of the liver, while extrahepatic bile duct perforations can be located in four areas (the right hepatic duct, left hepatic duct, common hepatic duct, and common bile duct). Spontaneous perforations are difficult to diagnose preoperatively [3]. Many patients are diagnosed as having cholecystitis or choledocholithiasis preoperatively. According to the past reports of the adult cases, it may be related either to a single or multiple factors such as obstruction by multiple calculi, infective necrosis, and increased intraductal pressure [4]. Biliary ascites is a common presentation of spontaneous perforation of the biliary tract presenting with progressive abdominal distention, peritonitis that may be localized or generalized, jaundice or septic

shock, a fluctuating course, and evidence of cholestasis with derangement of liver function. On paracentesis, the presence of bile, the sign of peritonitis, and an absence of pneumoperitoneum on x-ray further emboldens the suspicion of SCBDP. A preoperative recognition or at least a strong suspicion for spontaneous perforation would help in early optimization and planned surgical management resulting in a favorable outcome. Surgical intervention ranging from percutaneous tube placement to complicated biliary procedures are required for favorable outcome [5].

The objective of this study was to determine the clinical presentation, treatment, and outcome of patients with spontaneous perforation of the bile duct.

2. Case presentation

We managed three patients with nontraumatic perforation of the bile duct in our institution between 2019 and 2021.

Patient's age ranged from 45 to 55 years and none of them had a previous history of operation on the hepatobiliary tract. Nontraumatic

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perforation of the bile duct was suspected with the observation of bile-tinged peritoneal fluid or after drainage of bile from the perihepatic 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 distension 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 perihepatic 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 2×2 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, pneumoperitoneum was absent. MRCP suggested chronic cholecystitis with cholelithiasis, choledocholithiasis with calculus 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 (Fig. 2). After clinical and radiological study suggestive of GB perforation, USG guided percutaneous abdominal drain was placed, the content was bilious, 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 2×2 cm was retrieved. 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).

2.3. Patient 3

46-year male with peritonitis, sepsis and acute renal failure, presented with no previous history of abdominal surgery, instrumentation, or trauma. Laboratory investigations revealed raised bilirubin and creatinine. USG abdomen suggested cholelithiasis. MRCP suggested cholelithiasis with solitary calculus in the cystic duct, mildly dilated IHBR, chronic pancreatitis with infected pseudocyst of pancreas (Fig. 5).

No pneumoperitoneum was seen. Based on imaging finding, the patient was subjected to USG guided pigtail catheter drainage of the collection, which drained about 150 ml bile during 1st 24 h. Minimal drainage was observed thereafter. The patient underwent elective laparotomy after 3 episodes of hemodialysis for anuria. A large CBD perforation around 2×1 cm with three calculi, largest 2×1 cm was identified at the lateral border of the CBD with 2.5 l of bile in the peritoneal cavity with massive staining of all abdominal organs. Peritoneal lavage was given and CBD was repaired over T-tube. The post-operative period was uneventful, and the t-tube was removed on the 21st day after normal T tube cholangiogram.

Histopathology study of the gall bladder showed chronic inflammation. The patient was discharged on the 24th post-operative day.

Morbidity occurred in this patient, involving pneumonia and hepatic abscess. Hepatic abscess resolved with percutaneous catheter drainage. All patients have been well and without disease recurrence for average duration of 12 months on follow-up.

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 perihepatic abscess or peritonitis is combined with a biliary calculus [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 20 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 bilious 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
Hematologic Investigation on admission.

Characteristic	Patient 1	Patient 2	Patient3
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	33/46	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

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

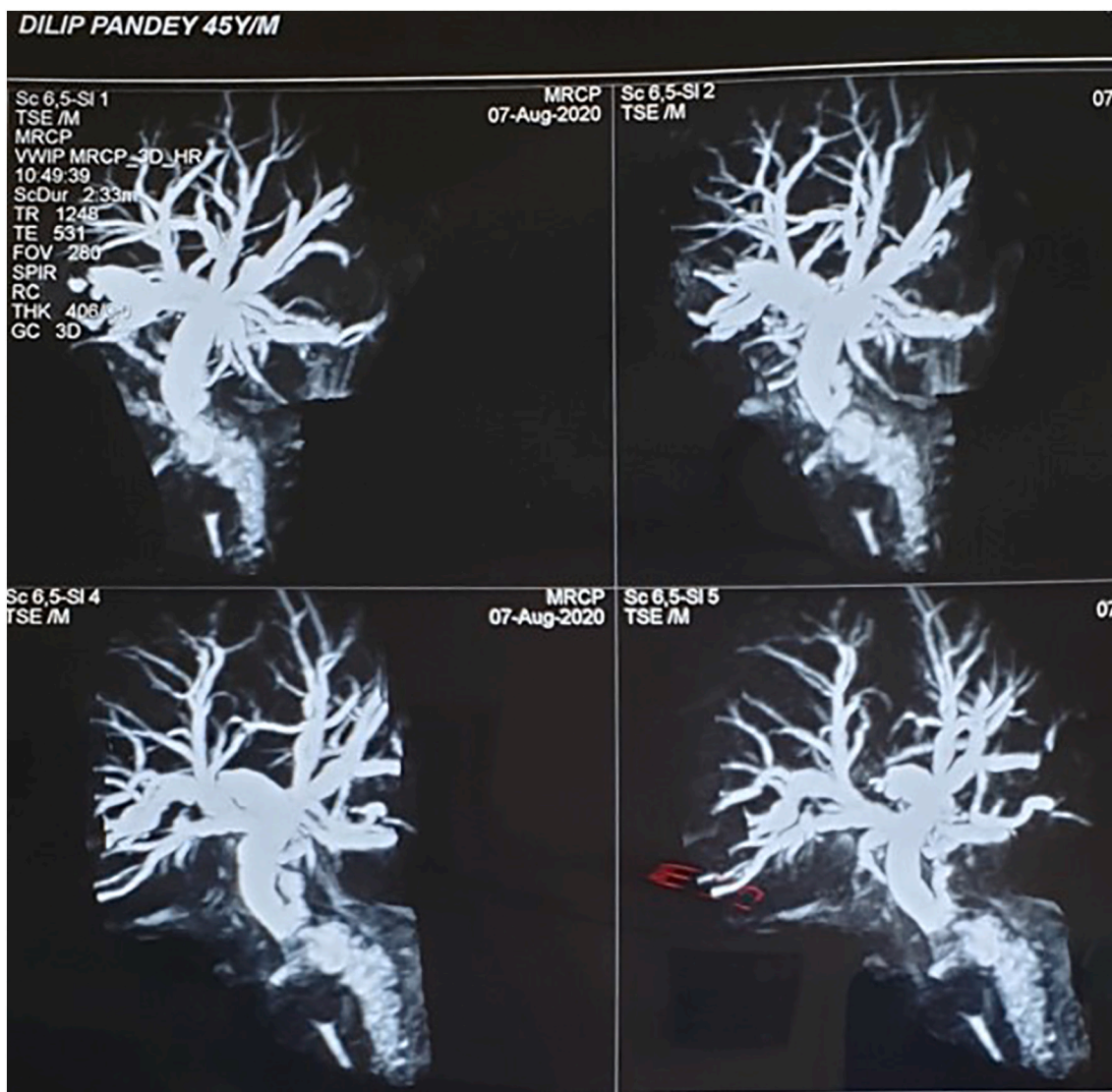


Fig. 1. MRCP suggested chronic cholecystitis with cholelithiasis, choledocholithiasis with calculus at distal end of common bile duct resulting into proximal dilated CBD and IHBR dilated.

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,

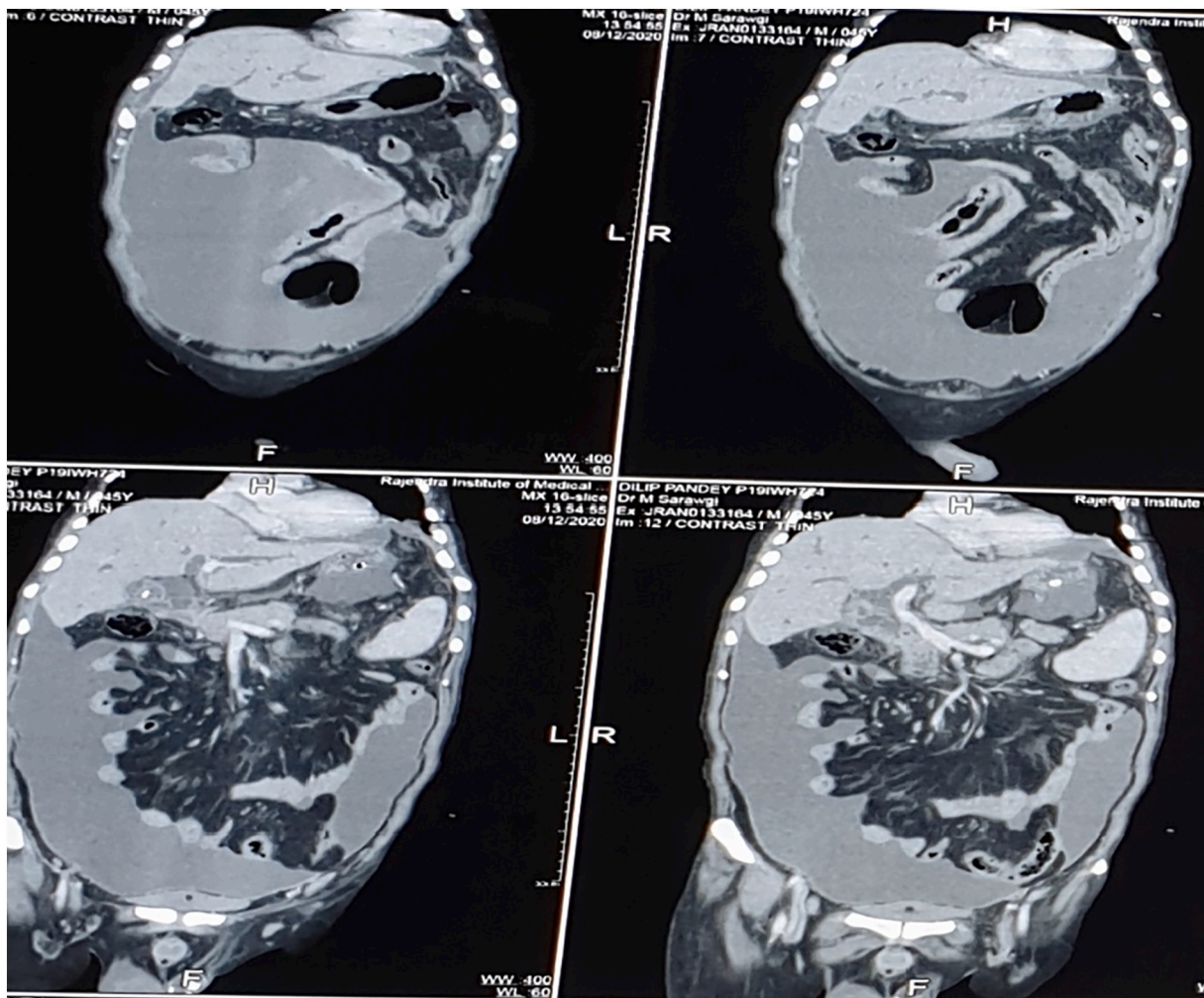


Fig. 2. CECT W/A suggested cholelithiasis with the collapsed wall showing irregular margins with moderate ascites suggestive of GB perforation.

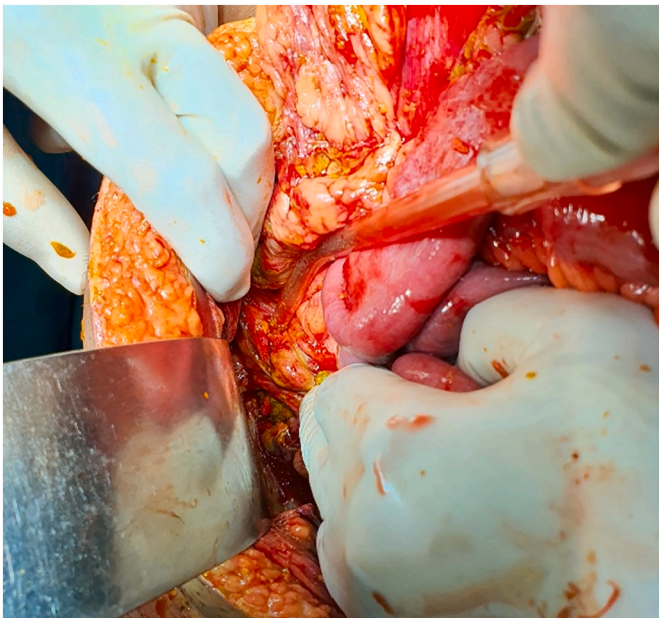


Fig. 3. Intraoperative image revealed perforation at Anterolateral surface of supraduodenal CBD with bile-stained small bowel and peritoneal cavity.

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. 4. T tube cholangiogram showing free flow of bile into duodenum and jejunum without any filling defect.

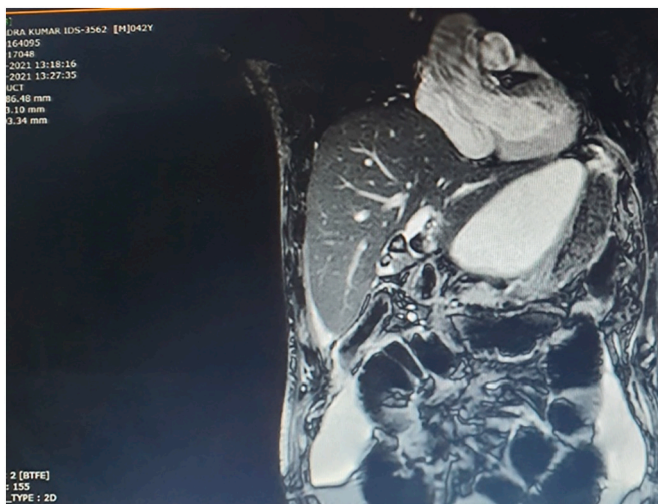


Fig. 5. MRCP suggested cholelithiasis with solitary calculus in the cystic duct, mildly dilated IHBR.

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/choledocholithiasis are important adjuncts in the diagnosis. Early surgical intervention is probably the only measure to prevent mortality and morbidity.

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Ethical approval

Approval taken.

Consent

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CRedit authorship contribution statement

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

Dr. Shalini Sonali-design, data collection and processing, literature review.

Santosh Kumar Pandit-data collection, analysis.

Dr. M. Sarawgi-critical review, supervision.

Declaration of competing interest

The author declared that there is no conflict of interest.

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