

# Stone-Induced Purulent Choledocoduodenal Fistula Presenting with Ascending Cholangitis

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## ABSTRACT

A biliary enteric fistula (BEF) is a rare, abnormal communication between any segment of the biliary tree with any portion of the small or large intestine. BEF is more frequently diagnosed with the increasingly widespread use of endoscopic retrograde cholangiopancreatography. Different theories have been postulated regarding the etiology of this fistula formation, with the most likely cause being gallstones. Treatment modalities, ranging from conservative management to surgical reconstruction, show varying levels of success. We present a case of BEF secondary to large common bile duct stones, successfully treated with endoscopic sphincterotomy (EST) followed by papillary balloon dilatation, and we briefly discuss large stone retrieval in the setting of atypical anatomy.

## INTRODUCTION

A biliary enteric fistula (BEF) can form between any segment of biliary system and the large or small intestine; choledochoduodenal fistulas (CDFs) account for 62% of all BEFs.<sup>1</sup> A distal CDF connects the duodenum to a region within 2 cm of the distal common bile duct (CBD), whereas a proximal CDF drains  $\geq 2$  cm above the junction of CBD to the papilla.<sup>1,2</sup> Spontaneous CDFs are more common than post-surgical CDFs, and they are usually caused by stones in the gallbladder or CBD (90%), peptic ulcer diseases (6%), malignancy, trauma, Crohn's disease, or migration of a biliary clip or a metallic biliary stent.<sup>3-6</sup> There are no symptoms that are pathognomonic for CDF. CDF most commonly presents with epigastric pain (80.9%), cholangitis (54.2%), and nausea or vomiting (10.3%). Jaundice is not a typical finding, especially with large stones due to fistula drainage.<sup>3,7,8</sup>

Different management strategies, from conservative to surgical management, have been proposed depending on the size and morphology of the stone that precipitated the fistula. Multiple trials have confirmed that endoscopic sphincterotomy with large balloon papillary dilatation is the most effective approach in extracting complex biliary stones and in cases of complex anatomy.<sup>9</sup>

## CASE REPORT

A 93-year-old woman with a past medical history of biliary pancreatitis presented with epigastric pain, nausea, and emesis. Two weeks prior, she presented with acute cholecystitis and moderately severe biliary pancreatitis per revised Atlanta Criteria, and subsequent endoscopic retrograde cholangiopancreatography (ERCP) was unsuccessful due to papillary edema. She refused a cholecystectomy at index hospitalization given her advanced age and multiple comorbidities.

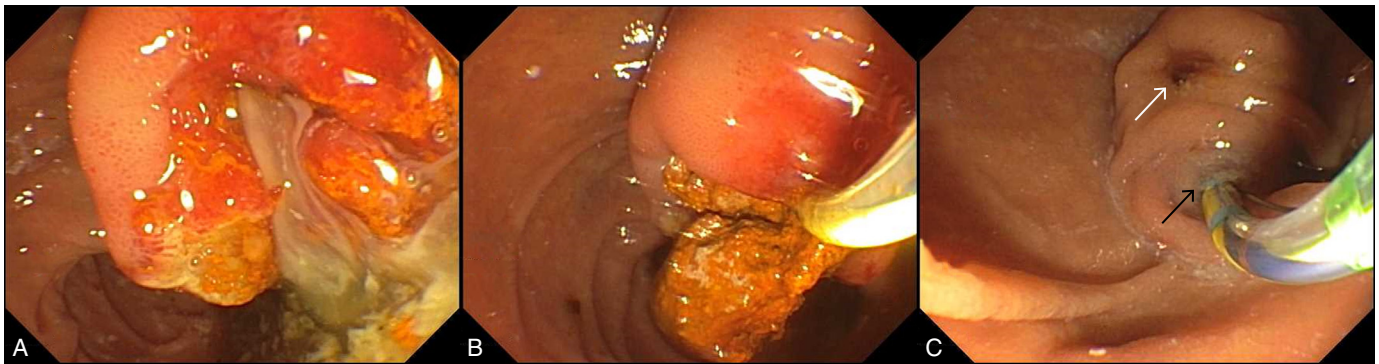
Differential diagnosis for the current presentation included acute cholecystitis, acute cholangitis, acute pancreatitis, and Mirizzi syndrome. Physical examination revealed mild scleral icterus and right upper quadrant tenderness

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**Figure 1.** (A) Purulent discharge from above the major papilla. (B) Stone concealing the choledochoduodenal fistula. (C) Exposed choledochoduodenal fistula (white arrow) with edematous papilla (black arrow).

with a positive Murphy's sign but no jaundice. Laboratory analysis was remarkable for leukocytosis  $25 \times 10^9/L$ , alanine transaminase 179 U/L, aspartate transaminase 390 U/L, alkaline phosphatase 173 IU/L, total bilirubin 2.4 mg/dL, and direct bilirubin 1.4 mg/dL. Abdominal ultrasound showed intrahepatic and extrahepatic biliary dilatation with a dilated CBD measuring 15 mm as well as a positive sonographic Murphy's sign, gallbladder wall thickening, pericholecystic fluid, and gallbladder distension consistent with cholecystitis and concomitant cholangitis.

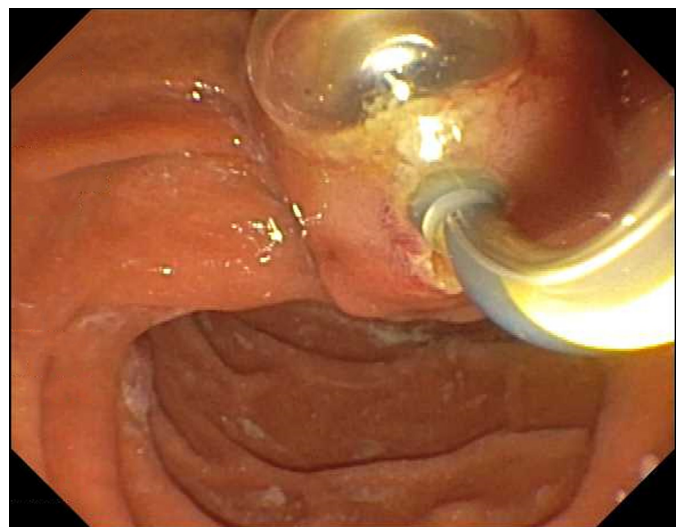
The diagnosis of ascending cholangitis was made, and the patient was put on a nothing-per-oral diet in addition to intravenous antibiotics. A same-day ERCP demonstrated an edematous major papilla with bilious and purulent discharge from above the major papilla orifice, determined to be the site of a CDF (Figure 1). Injected contrast demonstrated a dilated biliary tree and multiple CBD-filling defects, measuring up to 10 mm, consistent with complex biliary stones. A generous biliary sphincterotomy was made with an extension close to the adjacent horizontal duodenal fold, connecting the major papillary opening to the fistula, and large balloon papillary dilation to 11 mm was performed (Figure 2). Although it is difficult to guarantee no recurrence, it was felt that an opening of  $>1$  cm was sufficient to ensure adequate passage of stones given that the largest filling defect was 10 mm in size. Because the duct was subsequently cleared of all debris and purulent material, a stent was not deemed necessary. A final occlusion cholangiogram was negative for filling defects in the biliary tree. The patient tolerated the procedure well and had no subsequent complications from the initial presenting complaint or intervention. She remained asymptomatic after 2 years of follow up.

## DISCUSSION

BEFs are usually diagnosed incidentally on radiography.<sup>10</sup> Management strategies for BEF vary because there are no established treatment guidelines. In general, treatment depends on correcting the underlying cause. As this

condition is most commonly stone-induced, different treatment modalities are utilized depending on the severity of the disease.<sup>8</sup>

Endoscopic sphincterotomy, first described in 1974 by Kawai et al, is considered the gold standard for CBD stone retrieval, however this approach has been found to be successful in only 12% of cases of large stone retrieval, usually requiring supplemental therapies.<sup>11,12</sup> Endoscopic papillary balloon dilation (EPBD) is an alternative approach to preserve sphincter function and to reduce the morbidity associated with traditional endoscopic sphincterotomy.<sup>13</sup> Recently, the approach of sphincterotomy followed by balloon dilation has offered promising outcomes. In a prospective randomized controlled trial, outcomes from sphincterotomy followed by balloon dilation were found to be similar to those from sphincterotomy followed by mechanical lithotripsy and were found to have fewer complications.<sup>9</sup> Furthermore, in cases requiring stenting, the group undergoing balloon dilation had no retained



**Figure 2.** Inflated balloon through choledochoduodenal fistula prior to sphincterotomy.

fragments on repeat ERCP for stent retrieval, unlike those who underwent lithotripsy.<sup>9</sup>

In cases where these traditional methods prove unsuccessful, such as cases in which the stone is impacted in the cystic duct or the stone is located above a strictured CBD segment, electrohydraulic lithotripsy provides an alternative option. Although electrohydraulic lithotripsy is a successful alternative in cases with difficult to extract stones (i.e., 96% of stones are removed in the first attempt), 18% of these cases recur.<sup>14</sup> Furthermore, the high cost and difficult maneuverability proves to be a significant deterrent for its use. Likewise, the cost, fragility, and difficult maneuverability of laser lithotripsy in conjunction with the need for multiple treatments for stone extraction render this a less desirable option.

It has been reported that approximately 10–15% of complex stones cannot be retrieved by conventional ERCP modalities.<sup>15</sup> Cholangioscopy-guided lithotripsy has been proposed as an alternate technique that allows for fragmentation through electrohydraulic waves, allowing for easy retrieval, with success rates ranging from 80–90% in 1 session.<sup>14–16</sup> This reduces the risk of bile duct injury and the need for mechanical lithotripsy. However, its relatively rare availability compared to more traditional methods makes this a less viable option in complex stone retraction.<sup>17</sup>

In our case, the CDF was managed by EST followed by EPBD. The patient remains asymptomatic 24 months post-intervention without any complication from the CDF or the procedure.

## DISCLOSURES

Author contributions: C. Shams and S. Hakim wrote the manuscript. M. Cannon and J. Bortman edited the manuscript. C. Shams is the article guarantor.

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Informed consent was obtained for this manuscript.

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