First case of a primary biliary phytobezoar

Fahad Albogami, Alan N. Barkun, Kevin Waschke

ABSTRACT

Introduction: We present a patient with an unusual cause of biliary obstruction.

Case Report: A 50-year-old male was presented with a five-month history of worsening recurrent biliary abdominal pain and fevers. There was no previous biliary surgery. His workup revealed a normal bilirubin with elevation of other liver tests. Abdominal ultrasound demonstrated a common bile duct (CBD) diameter of 6 mm and cholelithiasis. A magnetic resonance cholangiopancreatography was unremarkable. An endoscopic ultrasound showed gallbladder sludge and stones, as well as CBD wall thickening with sludge in its mid to distal segments. At endoscopic retrograde cholangiopancreatography, a CBD filling defect was noted. After sphincterotomy, a balloon catheter extracted what looked like a cast occupying the entire lower CBD, extending into the cystic duct. This was retrieved in one piece using a rat tooth forceps and sent for pathology. The patient was discharged without complication. Cholecystectomy was recommended. Pathological analysis revealed the concretion was made of vegetable material. There have only been eight cases of biliary phytobezoar described in the modern English medical literature. Most reports describe the occurrence of a biliary phytobezoar presenting up to 40 years following a surgical bilioenteric anastomosis either with associated choledocholithiasis or alone. There exist only two case reports of patients having developed a biliary phytobezoar in the absence of any bilioenteric anastomosis or fistula. In both, the bezoar acted as a nidus for CBD stone formation, although the mechanism for developing a phytobezoar is not completely understood.

Conclusion: We describe the first reported case of an isolated biliary phytobezoar in the absence of previous biliary surgery or bilioenteric fistula.
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ABSTRACT

Introduction: We present a patient with an unusual cause of biliary obstruction. Case Report: A 50-year-old male was presented with a five-month history of worsening recurrent biliary abdominal pain and fevers. There was no previous biliary surgery. His workup revealed a normal bilirubin with elevation of other liver tests. Abdominal ultrasound demonstrated a common bile duct (CBD) diameter of 6 mm and cholelithiasis. A magnetic resonance cholangiopancreatography was unremarkable. An endoscopic ultrasound demonstrated a common bile duct (CBD) diameter of 6 mm and cholelithiasis. A magnetic resonance cholangiopancreatography was unremarkable. An endoscopic ultrasound showed gallbladder sludge and stones, as well as CBD wall thickening with sludge in its mid to distal segments. At endoscopic retrograde cholangiopancreatography, a CBD filling defect was noted. After sphincterotomy, a balloon catheter extracted what looked like a cast occupying the entire lower CBD, extending into the cystic duct. This was retrieved in one piece using a rat tooth forceps and sent for pathology. The patient was discharged without complication. Cholecystectomy was recommended. Pathological analysis revealed the concretion was made of vegetable material. There have been eight cases of biliary phytobezoar described in the modern English medical literature. Most reports describe the occurrence of a biliary phytobezoar presenting up to 40 years following a surgical bilioenteric anastomosis either with associated choledocholithiasis or alone. There exist only two case reports of patients having developed a biliary phytobezoar in the absence of any bilioenteric anastomosis or fistula. In both, the bezoar acted as a nidus for CBD stone formation, although the mechanism for developing a phytobezoar is not completely understood. Conclusion: We describe the first reported case of an isolated biliary phytobezoar in the absence of previous biliary surgery or bilioenteric fistula.

Keywords: Endoscopic retrograde cholangiopancreatography (ERCP), Phytobezoar, Post-biliary surgery

INTRODUCTION

Bezoar is defined as a foreign body resulting from accumulation of ingested material and classified according to its composition. A phytobezoar is the most common type of bezoar, and is composed of indigestible vegetable-like material.
Phytobezoars are commonly reported in patients who have had previous gastric surgery [1, 2]. They can occur at any site in the gastrointestinal tract, but most commonly are found in the stomach. However, biliary phytobezoars are extremely rare. We describe, for the first time, a case of a patient presenting with an obstructing biliary phytobezoar causing cholangitis in the absence of a history of abdominal surgery, sphincterotomy, spontaneous biliary-enteric fistula, or associated choledocholithiasis.

**CASE REPORT**

A 50-year-old male was presented with a five-month history of worsening recurrent biliary abdominal pain and fevers. There was no significant past medical history, including no previous biliary surgery. Physical examination showed only right upper quadrant tenderness.

Workup of the patient revealed a total bilirubin of 7.4 mg/L, direct bilirubin 4.8 mg/L, aspartate aminotransferase 71 U/L, alanine aminotransferase 103 U/L, alkaline phosphatase 292 U/L, and gamma-glutamyl transferase 992 U/L. Abdominal ultrasound demonstrated a common bile duct (CBD) diameter of 6 mm, pneumobilia and cholelithiasis. A magnetic resonance cholangiopancreatography (MRCP) from another institution was reported as unremarkable. An endoscopic ultrasound (EUS) showed gallbladder sludge and stones, as well as CBD wall thickening with sludge in its mid to distal segments. At endoscopic retrograde cholangiopancreatography (ERCP), a CBD filling defect was noted. After sphincterotomy, a balloon catheter extracted what looked like a cast occupying the entire lower CBD, extending into the cystic duct (Figure 1). This was retrieved in one piece using a rat tooth forceps and sent for pathology. The patient was discharged without complication. Pathological analysis revealed the cast was made of vegetable material (Figure 2).

**DISCUSSION**

A literature search revealed only eight cases of biliary phytobezoar described in the modern English medical literature. Four were isolated biliary phytobezoars [2–5], while the phytobezoar acted as a nidus for CBD stones in the other patients [2, 6–8] (Table 1).

In 1972, Ban et al. [6] described a biliary phytobezoar acting as nidus for symptomatic CBD stones that had developed in a patient two years post-choledochojejunostomy. Most reports describe the occurrence of a biliary phytobezoar presenting up to 40 years following a surgical bilioenteric anastomosis either with associated choledocholithiasis [9], or alone [2, 3, 5]— in one case as a result of a choledochooduodenal fistula 12 years post cholecystectomy [4]. There exist only two case reports of patients having developed a biliary phytobezoar in the absence of any bilioenteric anastomosis or fistula. In both, the bezoar acted as a nidus for CBD stone formation [7, 8] as has also been noted with foreign bodies such as surgical clips [10].

Although the mechanism for developing a phytobezoar is not completely understood, a main contributing factor relates to ablation or bypass of the sphincter of Oddi due to surgical manipulation or fistula formation. The mechanism in the absence of any such altered anatomy remains unclear and some have suggested that intermittent stone passage may contribute [7]. Sphincter of Oddi manometry abnormalities were noted in one patient, and remain of unclear clinical significance [9]. In the current patient, although cholelithiasis was noted, no CBD stone was present in the phytobezoar. So, cholecystectomy was recommended.
CONCLUSION

In conclusion, we describe the first reported case of an isolated biliary phytobezoar in the absence of previous biliary surgery or bilioenteric fistula.

REFERENCES


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Author Contributions
Fahad Albogami – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Alan N. Barkun – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Kevin Waschke – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor of Submission
The corresponding author is the guarantor of submission.

Table 1: Characteristics of patients reported in reported cases of biliary phytobezoar

<table>
<thead>
<tr>
<th>Authors</th>
<th>Year</th>
<th>Presence of prior sphincterotomy</th>
<th>Prior biliary enteric anastomosis or fistula</th>
<th>Presence of common bile duct stones</th>
<th>Presence of a sole phytobezoar</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ban JL et al. [6]</td>
<td>1972</td>
<td>No</td>
<td>Choledochojunostomy (2 years prior)</td>
<td>Yes</td>
<td>No</td>
</tr>
<tr>
<td>Cetta F et al. [7]</td>
<td>1993</td>
<td>No</td>
<td>None</td>
<td>Yes</td>
<td>No</td>
</tr>
<tr>
<td>Lamotte M et al. [3]</td>
<td>1995</td>
<td>No</td>
<td>Cholecystogastrostomy (15 years prior)</td>
<td>No</td>
<td>Yes</td>
</tr>
<tr>
<td>Procházka V et al. [9]</td>
<td>1999</td>
<td>No</td>
<td>None</td>
<td>Yes</td>
<td>No</td>
</tr>
<tr>
<td>Moghaddam JA et al. [4]</td>
<td>2006</td>
<td>No</td>
<td>cholecystectomy (6 years prior) with subsequent choledochoduodenal fistula</td>
<td>No</td>
<td>Yes</td>
</tr>
<tr>
<td>Kim TO et al. [8]</td>
<td>2006</td>
<td>No</td>
<td>None</td>
<td>Yes</td>
<td>No</td>
</tr>
<tr>
<td>Kim Y et al. [2]</td>
<td>2013</td>
<td>No</td>
<td>Cholecystectomy (12 years prior) with subsequent choledochoduodenal fistula</td>
<td>No</td>
<td>Yes</td>
</tr>
<tr>
<td>Bae JM et al. [5]</td>
<td>2014</td>
<td>No</td>
<td>hepaticojunostomy (remote)</td>
<td>No</td>
<td>Yes</td>
</tr>
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<td>Current report: Albogami et al.</td>
<td>2015</td>
<td>No</td>
<td>None</td>
<td>No</td>
<td>Yes</td>
</tr>
</tbody>
</table>
Source of Support
None

Consent Statement
Written informed consent was obtained from the patient for publication of this case report.

Conflict of Interest
Authors declare no conflict of interest.
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