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TITLE: First case of a primary biliary phytobezoar

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ABSTRACT

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

Case report
A 50 year-old man presented with a 5-month history of worsening recurrent biliary abdominal pain and fevers. There was no previous biliary surgery. His work-up revealed a normal bilirubin with elevation of other liver tests. Abdominal ultrasound demonstrated a common bile duct (CBD) diameter of 6mm and cholelithiasis. A magnetic resonance cholangio-pancreatography was unremarkable. An endoscopic ultrasound showed gall bladder 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 bilio-enteric anastomosis either with associated choledocholithiasis or alone. There exist only 2 case reports of patients having developed a biliary phytobezoar in the absence of any bilio-enteric 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 bilio-enteric fistula.

Keywords: ERCP, PHYTOBEZOAR, POST-BILIARY SURGERY
**TITLE:** First case of a primary biliary phytobezoar

**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 man presented with a 5-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.

His work-up 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 6mm, pneumobilia and choledolithiasis. A magnetic resonance cholangio-pancreatography (MRCP) from another institution was reported as unremarkable. An endoscopic ultrasound (EUS) showed gall bladder sludge and stones, as well as CBD wall thickening with sludge in its mid to distal segments. At endoscopic retrograde cholangio-pancreatography (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 bilio-enteric anastomosis either with associated choledocholithiasis [9], or alone [2, 3, 5] - in one case as a result of a choledochoduodenal fistula 12 years post cholecystectomy [4]. There exist only 2 case reports of patients having developed a biliary phytobezoar in the absence of any bilio-enteric 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 manometric abnormalities were noted in one patient, and remain of unclear clinical significance [9]. In the patient presented herein, although cholelithiasis was noted, no CBD stone was present in the phytobezoar. 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 bilio-enteric fistula.

CONFLICT OF INTEREST

None
AUTHOR’S CONTRIBUTIONS

Conception and design, Acquisition of data, Analysis and interpretation of data: Fahad Albogami, Alan N Barkun, Kevin Waschke

Drafting the article, Critical revision of the article: Fahad Albogami, Alan N Barkun, Kevin Waschke

Final approval of the version to be published: Fahad Albogami, Alan N Barkun, Kevin Waschke

REFERENCES


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>
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<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>hepaticojejunostomy (remote)</td>
<td>no</td>
<td>yes</td>
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<tr>
<td>Current report: Albogami et al.</td>
<td>2015</td>
<td>no</td>
<td>None</td>
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FIGURE LEGENDS

Figure 1: Endoscopic image of the common bile duct cast removed at ERCP

Figure 2: Appearance of the biliary cast in pathology specimen container

FIGURES

Figure 1: Endoscopic image of the common bile duct cast removed at ERCP
Figure 2: Appearance of the biliary cast in pathology specimen container