

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.



International Journal of Case Reports and Images (IJCRI)



International Journal of Case Reports and Images (IJCRI) is an international, peer reviewed, monthly, open access, online journal, publishing high-quality, articles in all areas of basic medical sciences and clinical specialties.

Aim of IJCRI is to encourage the publication of new information by providing a platform for reporting of unique, unusual and rare cases which enhance understanding of disease process, its diagnosis, management and clinico-pathologic correlations.

IJCRI publishes Review Articles, Case Series, Case Reports, Case in Images, Clinical Images and Letters to Editor.

Website: www.ijcasereportsandimages.com

CASE REPORT

PEER REVIEWED | OPEN ACCESS

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.

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

How to cite this article

Albogami F, Barkun AN, Waschke K. First case of a primary biliary phytobezoar. Int J Case Rep Images 2018;9(1):43–46.

Article ID: Z01201801CR10875FA

doi: 10.5348/ijcri-201806-CR-10875

Fahad Albogami^{1,3}, Alan N. Barkun^{1,2}, Kevin Waschke¹

Affiliations: ¹Division of Gastroenterology, The McGill University Health Centre, Montreal General Hospital site, Montréal, Canada; ²Epidemiology and Biostatistics and Occupational Health, McGill University Health Center, McGill University, Montreal, Canada; ³Division of Gastroenterology, King Fahad Specialist Hospital, Dammam, Saudi Arabia.

Corresponding Author: Dr. Alan N. Barkun, Division of Gastroenterology, The McGill University Health Centre, Montreal General Hospital site, 1650 Cedar Avenue, Room D7-346, Montréal, Canada H3G 1A4; Email: alan.barkun@muhc.mcgill.ca

Received: 13 May 2017
Accepted: 14 August 2017
Published: 01 January 2018

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).

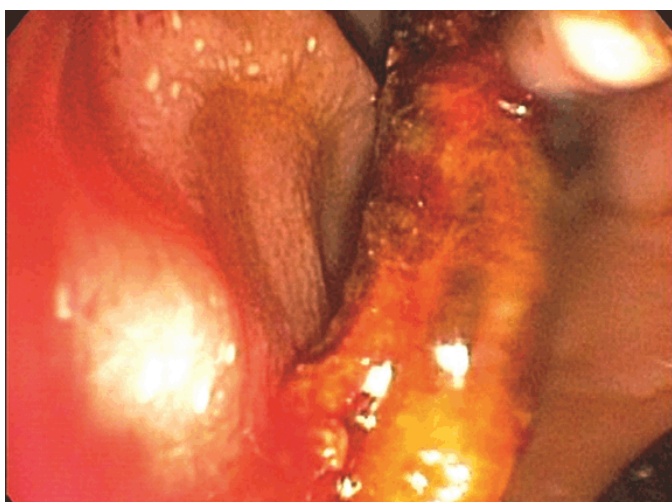


Figure 1: Endoscopic image of the common bile duct cast removed at endoscopic retrograde cholangiopancreatography.



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

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 choledochoduodenal 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.

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

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

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

1. Angelelli G, Magliocca M, Zaccheo N, Vinci R, Rotondo A. Intestinal obstruction caused by phytobezoar: Computerized tomography findings. Report of 3 cases. [Article in Italian]. Radiol Med 1997 Jun;93(6):789–91.
2. Kim Y, Park BJ, Kim MJ, et al. Biliary phytobezoar resulting in intestinal obstruction. World J Gastroenterol 2013 Jan 7;19(1):133–6.
3. Lamotte M, Kockx M, Hautekeete M, Holvoet J, Hubens H. Biliary phytobezoar: A medical curiosity. Am J Gastroenterol 1995 Aug;90(8):1346–8.
4. Moghaddam JA, Amini M, Adibnejad S. Development of bile duct bezoars following cholecystectomy caused by choledochoduodenal fistula formation: A case report. BMC Gastroenterol 2006 Jan 5;6:1.
5. Bae JM, Lee YK. Extremely rare case of extrahepatic duct phytobezoar treated with intraoperative transenteral endoscopy. Ann Surg Treat Res 2014 Aug;87(2):100–3.
6. Ban JL, Hirose FM, Benfield JR. Foreign bodies of the biliary tract: Report of two patients and a review of the literature. Ann Surg 1972 Jul;176(1):102–7.
7. Cetta F, Lombardo F, Rossi S. Large foreign body as a nidus for a common duct stone in a patient without spontaneous biliary enteric fistula or previous abdominal surgery. HPB Surg 1993;6(3):235–42.
8. Kim TO, Lee SH, Kim GH, et al. Common bile duct stone caused by a phytobezoar. Gastrointest Endosc 2006 Feb;63(2):324; discussion 324.
9. Procházka V, Krausová D, Kod'ousek R, Zámečníková P. Foreign material as a cause of choledocholithiasis. Endoscopy 1999 Jun;31(5):383–5.
10. McMahon GS, Attar S, Dennison AR. Bile duct “clip-stones”: Why a stitch in time could save nine. Hepatogastroenterology 2010 Sep–Oct;57(102–103):1037–9.

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.

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.

Copyright

© 2018 Fahad Albogami et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

Access full text article on
other devices



Access PDF of article on
other devices



Edorium Journals: An introduction

About Edorium Journals

Edorium Journals is a publisher of international, high-quality, open access, scholarly journals covering subjects in basic sciences and clinical specialties and subspecialties.

Invitation for article submission

We sincerely invite you to submit your valuable research for publication to Edorium Journals.

Why should you publish with Edorium Journals?

In less than 10 words: "We give you what no one does".

Vision of being the best

We have the vision of making our journals the best and the most authoritative journals in their respective specialties. We are working towards this goal every day.

Exceptional services

We care for you, your work and your time. Our efficient, personalized and courteous services are a testimony to this.

Editorial review

All manuscripts submitted to Edorium Journals undergo pre-processing review followed by multiple rounds of stringent editorial reviews.

Peer review

All manuscripts submitted to Edorium Journals undergo anonymous, double-blind, external peer review.

Early view version

Early View version of your manuscript will be published in the journal within 72 hours of final acceptance.

Manuscript status

From submission to publication of your article you will get regular updates about status of your manuscripts.

Our Commitment

Six weeks

We give you our commitment that you will get first decision on your manuscript within six weeks (42 days) of submission. If we fail to honor this commitment by even one day, we will give you a 75% Discount Voucher for your next manuscript.

Four weeks

We give you our commitment that after we receive your page proofs, your manuscript will be published in the journal within 14 days (2 weeks). If we fail to honor this commitment by even one day, we will give you a 75% Discount Voucher for your next manuscript.

Favored author program

One email is all it takes to become our favored author. You will not only get 15% off on all manuscript but also get information and insights about scholarly publishing.

Institutional membership program

Join our Institutional Memberships program and help scholars from your institute make their research accessible to all and save thousands of dollars in publication fees.

Our presence

We have high quality, attractive and easy to read publication format. Our websites are very user friendly and enable you to use the services easily with no hassle.

Something more...

We request you to have a look at our website to know more about us and our services. Please visit:
www.edoriumjournals.com

We welcome you to interact with us, share with us, join us and of course publish with us.



Edorium Journals: On Web



Browse Journals

CONNECT WITH US

