ACG CASE REPORTS JOURNAL
acgcasereports.gi.org

EDITORIAL BOARD

Editor in Chief
Mohammad Yaghoubi, MD, MSc, AFS
Medical University of South Carolina, Charleston, SC

Executive Editor
Manish Singla, MD
Walter Reed National Military Medical Center, Bethesda, MD

Associate Editors
Daniel E. Freedberg, MD
Columbia University Medical Center, New York, NY
Nazia Hasan, MD, MPH
New York University School of Medicine, New York, NY
Ryan Law, DO
Mayo Clinic, Rochester, MN
Kalyan Ray Parashette, MD
Indiana University School of Medicine, Indianapolis, IN
Andres J. Yarur, MD
University of Miami Miller School of Medicine, Miami, FL

EDITORIAL STAFF

Lindsey Topp
Editorial Advisor

Jenny Dunnington
Editorial Assistant

Theresa Bongorno
Graphic Designer

AIMS AND SCOPE

ACG Case Reports Journal, published by the American College of Gastroenterology and edited exclusively by GI fellows, provides a peer-reviewed publishing outlet for GI fellows, private practice clinicians, and other members of the healthcare team to share interesting case reports. This quarterly, open-access publication will make all content freely available online to all readers. ACG Case Reports Journal publishes case reports, images, and letters to the editor in all topics of gastroenterology and hepatology.

The ACG Case Reports Journal was created to help fulfill ACG’s commitment to providing growth and learning opportunities for GI fellows, and helps fellows meet core curriculum requirements for non-patient care activities. To this end, all case submissions must have a GI fellow or a resident interested in pursuing GI fellowship as the lead author. Cases authored by private practice clinicians and other members of the health care team who might traditionally face difficulty publishing with leading journals are also welcome.

PUBLISHER INFORMATION

Founded in 1932, the American College of Gastroenterology (ACG) is an organization with an international membership of more than 12,000 individuals from 86 countries. The College is committed to serving the clinically oriented digestive disease specialist through its emphasis on scholarly practice, teaching and research. The mission of the College is to serve the evolving needs of physicians in the delivery of high quality, scientifically sound, humanistic, ethical, and cost-effective health care to gastroenterology patients.

ACG Case Reports Journal is published online each quarter, and issues feature images, video clips, and multimedia content in addition to case descriptions. As an open-access publication, full-text articles are freely accessible for all readers in both HTML and PDF format immediately upon online publication. There is no print version of the Journal, but issue articles will be collated into an easily downloadable PDF for offline viewing and printing. ACG Case Reports Journal does not charge submission or publication fees for authors.

PREPARING FOR SUBMISSION

Manuscripts must be submitted online at mc.manuscriptcentral.com/acgcr. Questions regarding submission or site access should be sent to acgcasereports@gi.org. The ACG Case Reports Journal recommends that submitted manuscripts follow the general recommendations put forth by the ICMJE Uniform Requirements for Manuscripts.

PERMISSIONS

Authors are required to obtain permission to reproduce previously copyrighted materials from other sources in both print and electronic form. For questions regarding permissions for manuscripts published in ACG Case Reports Journal, please contact acgcasereports@gi.org.

ETHICS AND JOURNAL CONFLICT OF INTEREST

Authors must disclose all conflicts of interest, financial and otherwise, upon manuscript submission. Each year, the Editors publicly disclose their conflicts of interest on the ACG Case Reports Journal website.

When reporting on human or animal subjects, authors must state whether the work was approved by a local IRB or ethics committee, or in accordance with the Helsinki Declaration of 1975, as revised in 2008. Efforts should always be made to guarantee protection of patient privacy in submitted cases.

OPEN ACCESS AND CREATIVE COMMONS LICENSING

ACG Case Reports Journal content is licensed according to the Creative Commons Attribution-Noncommercial-No Derivative Works 3.0 Unported license, under which users are free to share (copy, distribute and transmit) the contribution under the following conditions:

• Attribution. Users must attribute the contribution in the manner specified by the author or licensor (but not in any way that suggests that they or their use of the contribution is endorsed by the author or licensor).
• Non-commercial. Users may not use this contribution for commercial purposes.
• No derivative works. Users may not alter, transform, or build upon this work.

For any reuse or distribution, users must make clear to others the license terms of this work. The best way to do this is with a link to the web URL of the published work. Any of the above conditions can be waived if users obtain permission from the copyright holder. The full legal terms of this license can be found on the Creative Commons website.

DUPLICATE PUBLICATION

Manuscripts must not be submitted to or previously published in any other journal. Any case that has been presented as a poster or oral presentation at any scientific meeting should contain a disclosure statement of this fact on the title page of the submission. Further, any cases published as an abstract related to a scientific meeting should be considerably expanded and enriched from the abstract version, and should contain a full disclosure of this former publication, including a full citation.

MANUSCRIPT ARCHIVE DEPOSITION

If your funding bodies and/or institution requires authors to self-archive articles in publicly accessible archives, then authors are responsible for depositing the accepted version of their manuscript into such an archive. ACG Case Reports Journal will register for indexing on PubMed as soon as possible.
Letter from the Editor

118 Tips for a Successful Case Report
Mohammad Yaghoobi, MD, MSc, AFS

Images

119 Unusual Finding of an Intact Moth During Routine Colonoscopy
Brijesh B. Patel, MD, Christian M. Andrade, MD, Marc J. Lajeunesse, PhD, and Reynaldo Geerken, MD

120 Emphysematous Gastritis: An Ominous Diagnosis Managed Conservatively
Brent E. Murchie, MD, Andrew C. Berry, BS, Andrew Ukleja, MD, Ryan McPherson, BA, Ariel Caplan, DO, and Warren L. Reuther III, MD

122 Endosonographic Findings in Colitis Cystica Profunda
Mohamed Sultan, MD, Walid Chalhoub, MD, Klaus Gottlieb, MD, and Gustavo Marino, MD

124 Hepatobiliary Fascioliasis: An Uncommon Cause of Biliary Obstruction in the United States
Jeff Basile, MD, M. Stanley Branch, MD, Svetang V. Desai, MD, Christopher Arnold, MD, Alastair Smith, CHB, FRCP, and Tzu-Hao Lee, MD

Case Reports

126 A Novel Approach to Management of Esophageal Pill Impaction
Brent W. Lacey, MD, Sean Caufield, MD, Eric Lavery, MD, and Brett Partridge, MD

128 Hepatic Portal Venous Gas: An Unusual Complication Following Upper Endoscopy and Dilation
Kristina Seeger, MD, and Sami R. Achem, MD

131 Acute Esophageal Necrosis: A Case of Black Esophagus Associated with Bismuth Subsalicylate Ingestion
Jean Abed, MD, Pavan Mankal, MD, Hani Judeh, MD, and Sang Kim, MD

134 A Treatment Option for Esophageal Intramural Pseudodiverticulosis
Amy Tyberg, MD, and Daniela Jodorkovsky, MD

137 Glass Microparticulate Ingestion: An Unusual and Difficult-to-Diagnose Cause of Chronic Abdominal Pain
R. Brooks Vance, MD, Marcus Mühlbauer, MD, PhD, Elizabeth B. Dreesen, MD, C. Robert Bagnell, Jr., PhD, Georgette A. Dent, MD, Hans Herfarth, MD, PhD, Christian Jobin, PhD, and Evan S. Della, MD, MPH
### TABLE OF CONTENTS

**ACG Case Reports Journal | Volume 1 | Issue 3**  
**April 2014**

#### Case Reports

<table>
<thead>
<tr>
<th>Page</th>
<th>Title</th>
<th>Authors</th>
</tr>
</thead>
<tbody>
<tr>
<td>140</td>
<td>Hemophagocytic Lymphohistiocytic Syndrome and Enteropathy-Associated T-cell Lymphoma in a Patient with Refractory Celiac Disease</td>
<td>Lucy Lu, MD, Shuoyan Ning, MD, Zain Kassam, MD, Richard Hunt, MB, MACG, and Marco Puglia, MD</td>
</tr>
<tr>
<td>143</td>
<td>Asymptomatic Duodenal Perforation from an Inferior Vena Cava Filter</td>
<td>Jean R. Park, MD, Veeral M. Oza, MD, and Somashekar G. Krishna, MD, MPH</td>
</tr>
<tr>
<td>145</td>
<td>Hematochezia Associated with Sevalamer-Induced Mucosal Injury</td>
<td>Preethi Chintamaneni, MD, Rohit Das, MD, Shih-Fan Kuan, MD, Taher R. Kermanshahi, MD, and Jana G. Hashash, MD</td>
</tr>
<tr>
<td>151</td>
<td>A Unique Case of Hematemesis in a 17-Year-Old Female</td>
<td>Tobias Zuchelli, MD, Eva Alsheik, MD, Bhavik Bhandari, MD, and Daniel Ringold, MD</td>
</tr>
<tr>
<td>154</td>
<td>Groove Pancreatitis: Four Cases from a Single Center and Brief Review of the Literature</td>
<td>Tyler P. Black, MD, Cynthia D. Guy, MD, Rebekah R. White, MD, Jorge Obando, MD, and Rebecca A. Burbridge, MD</td>
</tr>
<tr>
<td>158</td>
<td>Bouveret’s Syndrome with Severe Esophagitis and a Purulent Fistula</td>
<td>Rami Bonam, MD, Zahid Vahora, MD, Glenn Harvin, MD, and William Leland, MD</td>
</tr>
<tr>
<td>161</td>
<td>A Case of an Ectopic Ampulla of Vater in the Pyloric Channel</td>
<td>Sunil Dacha, MD, Xiao Jing Wang, MD, and Emad Qayed, MD</td>
</tr>
<tr>
<td>164</td>
<td>Sarcoidosis Presenting as Necrotizing Sarcoid Granulomatosis of the Liver, Sclerosing Cholangitis, and Gastric Ulcer</td>
<td>Njideka Momah, MD, Adetola Otesile, BSc, Rishi Pawa, MD, and Steve Shedlofsky, MD</td>
</tr>
<tr>
<td>167</td>
<td>Polymyositis Associated with Hepatitis B Virus Cirrhosis and Advanced Hepatocellular Carcinoma</td>
<td>Kessarin Thanapirom, MD, Satimai Aniwan, MD, and Sombat Treeprasertsuk, MD</td>
</tr>
<tr>
<td>170</td>
<td>Ceftriaxone-Induced Gallstones: Case Report and Literature Review</td>
<td>Aditi Nayak, MD, and Adam Slivka, MD, PhD</td>
</tr>
</tbody>
</table>

---

**Image 1:** Successful coil embolization of a 2.8-cm GDA pseudoaneurysm and outflow track of the right gastroepiploic artery. (Image from Zuchelli et al, page 151.)

**Image 2:** A balloon occlusion cholangiogram showing a distal common bile duct diameter of 7 mm and a round filling defect consistent with a bile duct stone. (Image from Dacha et al, page 161.)
Tips for a Successful Case Report

After a year of serving as the Editor-in-Chief of the *ACG Case Reports Journal*, I have been privileged to read hundreds of submitted case reports. Every submitted report has had its merits, and I wanted to highlight the characteristics of successful and well-written case reports that we have seen.

**Novelty**

Novelty is the single most important factor in writing an interesting case report. A comprehensive literature search can determine if the subject has been previously reported and can highlight novel aspects of your case to distinguish it from those in the literature. A specific and unique title to describe your case will grab the reader’s attention.

**Use Medical Language**

Make yourself familiar with the language of current medical literature before writing your manuscript. This can help you translate your case from a vernacular used in daily patient care to a more formal style of scientific writing. Manuscripts with simple text, active voice, and straightforward language attract readers, while unnecessary data and flowery language may be confusing and tedious.

**Identify Limitations**

Given the limitations of a case report compared to other forms of evidence-based documents, it is necessary to place your case in context. It is difficult to prove association with certainty in a case report; therefore, the discussion should avoid making wide-reaching conclusions based on a single experience. Advice from a senior colleague can help contextualize the role your case plays in the academic sphere.

**Remember the Author Instructions**

Prior to submitting a case report, remember to review the author guidelines and instructions. These details differ based on the target journal, but following them carefully will shorten the review process. Take this time to also have a colleague review your manuscript for spelling or grammatical errors, and clarifications of scientific writing. Reviewers enjoy reading clear and well-written manuscripts, and are often turned off by simple spelling and grammatical mistakes. One final review before submitting your manuscript can help identify small, overlooked mistakes and give your writing more polish.

**Confidentiality and Patients’ Rights**

Every effort should be made to obtain an informed consent from the patient, parents of a minor patient, or the next of kin of deceased patients. Make sure no identifying patient information is included in the text or in images. If consent cannot be obtained, provide a thorough description of the situation with your submission.

In the current era of evidence-based medicine, case reports are considered first-line evidence and might be the first academic contribution a young physician has in his or her career. I hope the above guidance and the opportunity provided by the *ACG Case Reports Journal* helps encourage our next generation of writers.

Mohammad Yaghoobi, MD, MSc, AFS
Editor-in-Chief
*ACG Case Reports Journal*
Unusual Finding of an Intact Moth During Routine Colonoscopy

Brijesh B. Patel, MD1,2, Christian M. Andrade, MD1,2, Marc J. Lajeunesse, PhD3, and Reynaldo Geerken, MD2

1Division of Digestive Diseases and Nutrition, University of South Florida, Tampa, FL
2Department of Gastroenterology, James A. Haley Veterans Affairs, Tampa, FL
3Department of Integrative Biology, University of South Florida, Tampa, FL

Case Report

There is scant literature describing inadvertent ingestion of insects visualized during endoscopy.1,2 Previously described insects include ants, wasps, bees, yellow jackets, and cockroaches. We present a case of a 55-year-old male with a normal colonoscopy except for the discovery of a lifeless winged insect between folds of the transverse colon (Figure 1). In the image, the insect is ventral side up on the colon lining. Two compound eyes and abdomen are visible, but the thorax and portions of the wings are overexposed. Six legs can be discerned, and the insect had roughly a 6-mm craniocaudal length and a 12-mm wingspan. The image was later identified by an entomologist as a moth belonging to order Lepidoptera. Moths typically have scales covering the body and wings, but these scales are easily removed when exposed to an acidic environment. A loss of these scales explains the whitish coloration of the moth, as most of the pigmentation is found on the scales. Although these ingestions are of little consequence to the patient, they are quite rare and may even be startling to the endoscopist. To our knowledge, this is the first case of a moth described within the gastrointestinal tract.

Disclosures

Author contributions: BB Patel, CM Andrade, and MJ Lajeunesse wrote and edited the manuscript. R. Geerken provided endoscopic images and reviewed and edited the final manuscript. BB Patel is the article guarantor.

Financial disclosure: None of the authors received financial support for the manuscript or express any personal or financial conflicts of interest.

Informed consent was obtained for this case report.

Received: December 20, 2013; Accepted: February 18, 2014

References

Emphysematous Gastritis: An Ominous Diagnosis Managed Conservatively

Brent E. Murchie, MD¹, Andrew C. Berry, BS², Andrew Ukleja, MD¹, Ryan McPherson, BA², Ariel Caplan, DO³, and Warren L. Reuther III, MD⁴

¹Digestive Diseases Institute, Cleveland Clinic Florida, Weston, FL
²Kansas City University of Medicine and Biosciences, Kansas City, MO
³Internal Medicine Department, Palm Beach Centre for Graduate Medical Education, West Palm Beach, FL
⁴Department of Radiology, West Palm Hospital, West Palm Beach, FL

Case Report

A 54-year-old female with HIV, diabetes, and chronic obstructive pulmonary disease (COPD) presented with altered mental status, diabetic ketoacidosis, nonspecific gastrointestinal symptoms, and a buttock abscess. Initial abdominal and pelvic computed tomography (CT) without contrast demonstrated a small pericardial effusion, air in the gastric wall, and perianal abscess. Amid worsening leukocytosis (22,500/mm³), a wide excisional debridement of abscess was performed and later repeated. CT angiography of the chest demonstrated a markedly distended stomach with small amount of portal venous air (Figure 1). Abdominal X-ray of the kidney, ureters, and bladder (KUB) demonstrated a distended stomach with wall emphysema and gas collection within the gluteal region (Figure 2). Esophagogastroduodenoscopy (EGD) revealed black eschars and exudates in the stomach body and fundus (Figure 3). When gastric wall air is present, emphysematous gastritis—with a mortality rate of 50–80%—must be properly distinguished from the more common and less devastating gastric emphysema.¹,² Air within the

Figure 1. CT chest angiography demonstrating portal venous air and a mottled, non-linear air pattern in the gastric wall.

Figure 2. Abdominal X-ray of kidney, ureters, and bladder (KUB) showing gas/air within both the gastric lumen and the stomach wall.
gastric wall, together with portal venous air, leukocytosis, and a source of infection all support the diagnosis of emphysematous gastritis.\textsuperscript{3,4} Without evidence of sepsis or ischemia, surgical intervention was not indicated. Conservative management with bowel rest, parenteral nutrition, and broad-spectrum antibiotics was successful.\textsuperscript{5} The role of endoscopy in cases like this is strictly to monitor severity, identify gastric necrosis, and exclude other pathology.

**Disclosures**

Author contributions: All authors contributed to evaluating and managing the case and to writing the manuscript. AC Berry is the article guarantor.

Financial disclosure: No financial support or conflicts of interest to report.

Informed consent was obtained for this case report.

Received: November 18, 2013; Accepted: March 16, 2014

**References**

Endosonographic Findings in Colitis Cystica Profunda

Mohamed Sultan, MD¹, Walid Chalhoub, MD¹, Klaus Gottlieb, MD², and Gustavo Marino, MD²

¹Division of Gastroenterology, MedStar Georgetown University Hospital, Washington, DC
²Division of Gastroenterology, Veteran Affairs Medical Center, Washington, DC

Case Report

A 27-year-old male was referred to our institution for further evaluation of persistent rectal bleeding. A prior colonoscopy showed a sigmoid soft tissue lesion, and pathology revealed chronic active colitis and granulation tissue with ulcers and focal adenomatous changes. We performed a flexible sigmoidoscopy that showed a 4.5-cm multilobulated polypoid lesion approximately 45 cm from the anal verge (Figure 1). A 20 MHz Olympus endoscopic ultrasound (EUS) miniprobe showed hypoechoic lesion with cystic/spongy features involving the mucosa and submucosa (Figure 2). These features were thought to suggest colitis cystica profunda (CCP). Histologic examination of snare biopsies identified dilated glands with mucinous content, surrounded by variable degrees of fibrosis on a background of interspersed chronic inflammatory cells, with few colonic mucosal crypts and mild inflammatory cell infiltrate (Figure 3). The patient was instructed to follow up with gastroenterology if bleeding recurred.

CCP is a rare, benign disease of the colon and rectum often mimicking malignancy. It was first described in 1766 by Stark, who reported 2 cases associated with dysentery.¹ Histologically, it is characterized by dilated mucous glands mostly limited to the submucosa, but there are reported cases of penetration to the muscularis mucosa.¹ The etiology of CCP remains controversial; however, many consider solitary rectal ulcer syndrome (SRUS) and...
Sultan et al

Colitis Cystica Profunda

CCP to be different manifestations of the same pathology due to overlapping features.² Surface mucosal biopsies may rule out neoplasia, but only deep biopsies show characteristic histological features.¹ There are few case reports of the endosonographic features of CCP,³ which include multiple hypoechoic or anechoic lesions affecting mucosa or submucosa, with areas of echorefringent fibrosis between lesions in the absence of lymph node enlargement. EUS was instrumental in making our diagnosis and providing proper counseling to the patient.

Disclosures

Author contributions: All authors contributed equally to this article. M. Sultan is the article guarantor.

Financial disclosure: The authors have no financial disclosure.

Informed consent was obtained for this case report.

Received: December 9, 2013; Accepted: March 4, 2014

References


Figure 3. Hematoxylin and eosin stain section showing transmural dilated glands with mucinous content, surrounded by variable degrees of fibrosis on a background of interspersed chronic inflammatory cells and few colonic mucosal crypts, with mild inflammatory cells infiltrate.

Publish your work in ACG Case Reports Journal

ACG Case Reports Journal is a peer-reviewed, open-access publication that provides GI fellows, private practice clinicians, and other members of the health care team an opportunity to share interesting case reports with their peers and with leaders in the field. Visit http://acgcasereports.gi.org for submission guidelines. Submit your manuscript online at http://mc.manuscriptcentral.com/acgcr.
**Hepatobiliary Fascioliasis: An Uncommon Cause of Biliary Obstruction in the United States**

Jeff Basile, MD\(^1\), M. Stanley Branch, MD\(^1\), Svetang V. Desai, MD\(^1\), Christopher Arnold, MD\(^2\), Alastair Smith, MB, CHB, FRCP\(^1\), and Tzu-Hao Lee, MD\(^3\)

\(^1\)Division of Gastroenterology, Department of Medicine, Duke University Medical Center, Durham, NC  
\(^2\)Division of Infectious Diseases, Department of Medicine, Duke University Medical Center, Durham, NC  
\(^3\)Department of Internal Medicine, Duke University Medical Center, Durham, NC

**Case Report**

A 43-year-old woman presented with recurring upper abdominal pain. She had a 5-year history of symptomatic cholelithiasis without improvement following cholecystectomy. She had no prior history of elevated liver tests or jaundice. Her travel history was pertinent for annual trips to the Bahamas. On admission, the patient had a bilirubin of 4.7 mg/dL and liver enzymes more than 5 times the upper limit of normal. Abdominal computed tomography (CT) scan demonstrated a wedge-shaped area of decreased attenuation in liver segment III (Figure 1). Endoscopic retrograde cholangiopancreatography (ERCP) revealed a curvilinear filling defect within the distal common bile duct (Figure 2). Following papillary sphincterotomy, a living parasite was removed from the common bile duct (Video 1) and confirmed by pathology as *Fasciola hepatica* (Figure 3). Nitazoxanide was prescribed. Her liver enzymes normalized after 1 week of therapy, and symptoms resolved completely. Magnetic resonance imaging (MRI) 4 months later demonstrated resolution of all imaging abnormalities.

Figure 1. Abdominal CT scan showing a wedge-shaped area of decreased attenuation in liver segment III.  
Figure 2. Rotational fluoroscopic 3-D reconstruction image taken during ERCP showing a curvilinear filling defect in the distal common bile duct (white arrows) secondary to obstruction from *Fasciola hepatica*.
trematode have been noted.\textsuperscript{1,2} Further awareness of fascioliasis may help facilitate the diagnosis and management of this rare yet treatable cause of hepatobiliary disease in the United States.

**References**


**Figure 3.** Image of the liver fluke, *Fasciola hepatica*, captured during ERCP.

**Video 1.** Video demonstrating removal of *Fasciola hepatica* from the common bile duct after retrieval balloon sweep during ERCP. Please view the video at http://acgcasereports.gi.org/?p=1944.

**Disclosures**

Author contributions: All authors contributed equally to the preparation of this manuscript. J. Basile is the article guarantor.

Financial disclosure: The authors report no conflicts of interest or financial support for this article.

Informed consent was obtained for this case report.

Received: October 10, 2013; Accepted: January 16, 2014
A Novel Approach to Management of Esophageal Pill Impaction

Brent W. Lacey, MD, Sean Caufield, MD, Eric Lavery, MD, and Brett Partridge, MD

Naval Medical Center San Diego, San Diego, CA

Abstract

A 26-year-old male presented with symptoms of acute esophageal obstruction immediately after swallowing an 800-mg ibuprofen tablet. Multiple attempts to extract the pill with a variety of traditional endoscopic retrieval devices were unsuccessful. We successfully destroyed the pill using a threaded-tip biliary stent retrieval device to drill a hole in the center of the pill, which allowed us to use a rat-tooth forceps to crush the pill. This case report demonstrates a novel use of this device in a challenging esophageal pill extraction.

Introduction

Esophageal foreign body impaction can be managed with a variety of standard endoscopic retrieval devices, but anatomic features and the nature of the foreign body can make successful extraction difficult or impossible with standard techniques. We present this case to demonstrate a novel approach to the management of a challenging esophageal pill impaction.

Case Report

A 26-year-old male presented with symptoms of acute esophageal obstruction immediately after swallowing an 800-mg ibuprofen tablet. He could not swallow oral secretions and complained of focal anterior neck pain. During an urgent upper endoscopy, we encountered a circular, hard pill tightly impacted at a mid-esophageal stricture. The esophagus had a narrow caliber and the diagnostic endoscope could only be advanced to a point just proximal to the pill (Figure 1A). Multiple attempts to extract the pill with a variety of traditional endoscopic retrieval devices, including rat-tooth forceps, polypectomy snare, retrieval net, biopsy forceps, and three-pronged grasper, were unsuccessful.

After consideration of management options for refractory esophageal impaction (including surgical referral), we chose to attempt to disrupt the pill with a 7 French threaded-tip Soehendra biliary stent retriever (Cook Medical, Winston-Salem, NC). The stent retriever was centered in the lumen and placed gently against the center of the pill (Figure 1B). Clockwise rotation of the threaded tip created a central defect within the pill matrix (Figure 1C), then we used the rat-tooth forceps to rapidly crush the remaining pill, working off the central defect (Figure 1D). After pulverizing the impacted pill, we observed an esophageal stricture 8 mm in diameter (Figure 2), which we traversed with a narrow caliber (5.5 mm) upper endoscope and completed the examination. The patient recovered uneventfully. The endoscopic appearance and biopsies of the esophagus were consistent with eosinophilic esophagitis, which subsequently required esophageal dilation for treatment.

Discussion

The biliary stent retrieval device (Figure 3) has a threaded tip designed to be advanced over a guidewire into a metal biliary stent to facilitate extraction. Since its introduction, the device has been used for other purposes, such as expanding a hole in a metallic mesh stent for multiple stenting of a hilar biliary obstruction, EUS-guided drainage of a pancreatic pseudocyst, dilating refractory pancreatic duct strictures, and dilating pancreaticojejunosomy.


Correspondence: Brent W. Lacey, Naval Medical Center San Diego, 34800 Bob Wilson Dr., San Diego, CA, 92108 (brent.lacey@med.navy.mil).

Copyright: © 2014 Lacey et al. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.
Esophageal Pill Impaction

Lacey et al

strictures. This is the first published case using the device for management of an esophageal pill impaction. The threaded tip may cut rapidly, so it is important to drill through the pill carefully, checking progress frequently lest the device damage the esophageal wall. This device does not use electrocautery. Once a hole is created in the center of the pill, standard devices such as the rat-tooth forceps used in this case have the necessary leverage to crush the pill easily.

When encountering an esophageal pill impaction refractory to extraction with standard endoscopic retrieval devices, endoscopists may consider utilizing a threaded-tip biliary stent retriever. Further studies would be required to determine the efficacy and safety of the stent retriever in the removal of a variety of impacted foreign bodies within the gastrointestinal tract.

Disclosures

Author contributions: BW Lacey was the primary author of the final manuscript and the article guarantor. S. Caufield, E. Lavery, and B. Partridge contributed to and edited the final manuscript.

Financial disclosure: None to report.

Informed consent was obtained for this case report.

Received: February 10, 2014; Accepted: March 16, 2014

References

Hepatic Portal Venous Gas: An Unusual Complication Following Upper Endoscopy and Dilation

Kristina Seeger, MD, and Sami R. Achem, MD

1Department of Internal Medicine, Mayo Clinic, Jacksonville, FL
2Division of Gastroenterology and Hepatology, Mayo Clinic, Jacksonville, FL

Abstract

Hepatic portal venous gas (HPVG), a rare condition in which gas accumulates in the portal venous circulation, is often associated with a significant underlying pathology, such as intestinal ischemia, sepsis, and trauma. HPVG after endoscopy or dilation is an unusual complication. We report a case of HPVG following upper endoscopy and dilation for an esophageal stricture in a 34-year-old patient with eosinophilic esophagitis (EoE). The patient was treated conservatively, and his symptoms resolved. Follow-up computed tomography (CT) scan showed resolution of HPVG. This case highlights a rare and potentially ominous complication of upper endoscopy and dilation and underscores the role of conservative management.

Introduction

Hepatic portal venous gas (HPVG) is a rare condition in which intraluminal gas or gas produced by bacteria in the gut enters the portal venous circulation. Common precipitating factors favoring the development of portal venous gas include intestinal wall disruption, bowel distention, and sepsis, which typically suggest an ominous underlying pathology. We describe a previously unrecognized cause of HPVG following an upper endoscopy and dilation for a benign esophageal stricture in a young male patient with eosinophilic esophagitis (EoE).

Case Report

A healthy 34-year-old male with a past medical history significant for EoE and asthma presented with unexplained dysphagia. He underwent esophagogastroduodenoscopy (EGD), which revealed esophageal mucosal changes that were suggestive of EoE, including loss of vascular pattern and white exudates, and a benign esophageal stricture at the gastroesophageal junction. The stricture was dilated with a balloon size of 18 mm. Esophageal biopsies confirmed the diagnosis of EoE.

Within an hour after the procedure, the patient experienced nausea and epigastric pain. His physical exam was remarkable for diffuse epigastric tenderness with no guarding or rigidity. Laboratory tests were normal (complete blood count, liver, and pancreatic enzymes). Abdominal computed tomography (CT) demonstrated diffuse portal venous gas throughout the right hepatic lobe extending to the periphery of the liver (Figure 1). The patient was hospitalized and treated conservatively with intravenous fluids and pain control. He was discharged the following day after his symptoms had resolved. A follow-up repeat CT scan revealed no residual portal vein air. He continued to do well at a 6-month follow-up.

Discussion

HPVG has often been thought of as an ominous clinical finding; it is commonly the result of portomesenteric vein gas accumulation resulting from bowel ischemia. Other etiologies associated with HPVG are summarized...
Hepatic Portal Venous Gas

in Table 1. Only a small fraction of cases are attributed to radiologic and endoscopic procedures. Of these, HPVG has been reported following EGD in patients with a gastric ulcer and duodenal tumor,4 after corrosive acid ingestion,5 following esophageal variceal sclerotherapy and banding,6 after percutaneous endoscopic gastrostomy, and after ERCP.5

HPVG is best diagnosed with CT, where it appears as tubular areas of decreased attenuation in the liver. The low attenuation areas are caused by accumulation of gas in the intrahepatic portal veins, where it is carried by centrifugal blood to the hepatic periphery.2 Due to the high carbon dioxide content of portal venous gas, it is expected to last briefly in the vascular system before it is absorbed or removed by bulk flow, unless gas production persists.7

The mechanism for the formation of HPVG is unclear, but it has been hypothesized that certain factors allow gas to enter the portal circulation through veins or lymphatics of the intestinal wall and reach the hepatic veins through the hepatic sinusoids.7 These factors may include compromised bowel wall integrity, bowel distention, increased intraluminal pressure, and sepsis. Bacterial fermentation of carbohydrates in sepsis may also contribute to the development of HPVG.2

Endoscopic examination of our patient revealed esophageal inflammation with no gross ulcerations. It is possible this underlying esophageal inflammation contributed to compromised mucosal integrity. The esophageal inflammation coupled with the endoscopic procedure (air insufflation) and esophageal dilation likely facilitated diffusion of intraluminal air across the gastrointestinal mucosa. It has been proposed that endoscopic procedures facilitate diffusion in several ways. Specifically, shear pressure caused by dilation may compromise mucosal wall integrity, and gastric or lumen distention caused by insufflation may lead to increased intraluminal pressures, forcing air across the mucosal surface.5,7

The presence of HPVG may convey the notion of a life-threatening complication. However, HPVG is not necessarily an indication for surgical management and may require no treatment.7,8 The underlying etiology and clinical condition of the patient should dictate management. Surgical management is warranted in clinically unstable patients, or if there is evidence of peritonitis or bowel perforation. Conservative management may be used in clinically stable patients or those who have developed HPVG from an invasive procedure.2 Due to the likely iatrogenic-induced HPVG in our patient, he was treated conservatively with fluids and analgesics, which resolved his symptoms and CT findings.

We describe a case of HPVG following EGD and dilation in a young patient with EoE. Only a handful of cases have mentioned HPVG following endoscopic procedures, and a large national database from the American Society for Gastrointestinal Endoscopy does not mention it as a complication.
after EGD or dilation. This case illustrates the need to recognize a rare complication following EGD and dilation. In this setting, HPVG can be effectively managed conservatively.

Disclosures
Author contributions: K. Seeger completed the literature search and review, and wrote the manuscript. SR Achem evaluated the patient, designed the study, wrote and edited the manuscript, and is the article guarantor.

Financial disclosure: None to report.

Informed consent was obtained for this case report.

Received: November 21, 2013; Accepted: January 27, 2014

References
Acute Esophageal Necrosis: A Case of Black Esophagus Associated with Bismuth Subsalicylate Ingestion

Jean Abed, MD¹, Pavan Mankal, MD¹, Hani Judeh, MD¹, and Sang Kim, MD²

¹Department of Medicine, Icahn School of Medicine, Mount Sinai St. Luke’s and Roosevelt Hospitals, New York, NY
²Department of Medicine, Division of Gastroenterology, Weill Cornell Medical College, New York, NY

Abstract
We present a case of acute esophageal necrosis (AEN) likely caused by chronic use of bismuth subsalicylate, an active ingredient in over-the-counter Pepto-Bismol®, which contains 220 g of salicylic acid in each 30 mL quantity. While aspirin is known to cause gastritis and gastric ulcers, this is the first case, to our knowledge, reporting AEN after chronic bismuth subsalicylate use.

Introduction
Acute esophageal necrosis (AEN), or black esophagus, is a rare entity caused by variety of factors. One theory is that the relatively low perfusion state in the distal areas of the esophagus make it susceptible to mucosal injury. We present the first case report of AEN after using large doses of over-the-counter Pepto-Bismol® (bismuth subsalicylate).

Case Report
An woman in her early 80s with past medical history significant for ulcerative colitis and gastroesophageal reflux (GERD) with use of bismuth subsalicylate was admitted to the hospital for 2 days of constant, diffuse abdominal pain with melena. She denied any tobacco, alcohol, illicit drug, or any nonsteroidal anti-inflammatory drug (NSAID) use. On admission, the patient was hemodynamically stable; her physical exam was notable for a slightly distended abdomen with diffuse tenderness to palpation without any rebound or guarding. She had positive guaiac stool with negative stool cultures. Laboratory tests revealed leukocytosis and hemoglobin of 10.1 g/dL (baseline: 14 g/dL). The basic metabolic panel, hepatic function panel, coagulation studies, and urinalysis were all normal.

On the second day of admission, the patient underwent an esophagogastroduodenoscopy (EGD) that revealed an ulcerated and darkened necrotic esophageal mucosa extending continuously from 20 cm beyond the incisors to the gastroesophageal junction (Figure 1). A biopsy revealed extensive necrosis, marked inflammation, and absence of esophageal epithelium consistent with AEN (Figure 2).

The patient reported consuming 40–50 antacids per day for 8–10 years for her acid reflux disease. Total parenteral nutrition was started and the patient was managed conservatively. A repeat EGD 10 days after admission demonstrated diffuse esophageal edema and pink granulation tissue consistent with healing esophageal mucosa (Figure 3). The patient’s diet was slowly advanced and she was discharged in stable condition 2 weeks after admission.
AEN, or black esophagus, is a rare clinical entity with an incidence ranging from 0.01% to 0.2% in autopsy studies and clinical trials. Prior case studies have shown a predilection for AEN in individuals who suffer from cardiovascular ischemic events, acute alcohol intoxication, and caustic injury from alkaline compounds, but AEN is thought to be a result of many factors. The most common presentation is hematemesis and melena, with endoscopy demonstrating friable or macerated mucosa involving the distal two-thirds of the esophagus with a very sharp demarcation at the gastroesophageal junction. The reason for the location may be due to the relatively low perfusion state compared to more proximal areas of the esophagus that are often affected in patients with vasculopathy. Histopathology often shows mucosal and submucosal necrosis with heavy leukocyte infiltration, as seen in our patient, though biopsy is not required for diagnosis. Management is conservative, but patients require close monitoring due to the risk of complications such as superimposed infections, stenosis, strictures, and perforations. Mortality can reach as high as 32% (often in cases presenting with other comorbidities), but mortality specific to AEN is around 6%.

The exact etiology of AEN in this case is unclear in the absence of vascular disease. AEN in this patient might be secondary to the chronic ingestion of a large dose of antacids (i.e., bismuth subsalicylate) causing chemical injury and necrosis. Bismuth subsalicylate, an active ingredient in over-the-counter Pepto-Bismol®, is used widely to treat symptomatic inflammation of gastric and intestinal epithelium. The reactivity of bismuth with sulfur molecules ingested from food can lead to linguinal hyperpigmentation (black tongue),
which is often benign and reversible with medication discontinuation. The high doses of an alkalinizing medication such as bismuth subsalicylate, which contains 220 g of salicylic acid per 30 mL quantity, may have caused chemical injury to the esophageal epithelium to develop even without the use of other NSAIDs. The chemical reaction between bismuth and sulfur may have created the dark pigmentation of the esophagus classic to AEN. To our knowledge, no other case report has reported a black esophagus after prolonged, heavy use of over-the-counter bismuth subsalicylate with rapid recovery after the discontinuation of the causative agent.

Disclosures

Author contributions: J. Abed and P. Mankal wrote the manuscript. H. Judeh and S. Kim provided the case history and photographs. S. Kim is the article guarantor.

Financial disclosure: No conflicts of interest or financial disclosures to report.

The patient consented in writing to all procedures. The authors made every effort to contact the patient and her next of kin, but due to the length of time between the case and this report, neither the patient nor the next of kin could be reached for consent to publish. However, the authors feel that the patient information is sufficiently anonymous and that the patient would not object to this publication.

Received: December 10, 2013; Accepted: February 20, 2014

References

A Treatment Option for Esophageal Intramural Pseudodiverticulosis

Amy Tyberg, MD, and Daniela Jodorkovsky, MD

Division of Gastrointestinal and Hepatobiliary Diseases, New York Medical College, Westchester Medical Center, Valhalla, NY

Abstract
Esophageal intramural pseudodiverticulosis (EIPD) is a rare condition often presenting with esophageal strictures. Treatment is often limited to endoscopic dilatation and treatment of the underlying esophageal pathology. We present a case of a patient with longstanding GERD on famotidine (she experienced anaphylaxis with proton pump inhibitors [PPIs]) who presented with dysphagia and weight loss. Work-up revealed a diagnosis of EIPD with a 5-mm mid-esophageal stricture. Therapy with dilatation was unsuccessful until the addition of sucralfate, after which dilatation was successful and symptoms resolved. In patients who are unable to take PPIs, the addition of sucralfate may enhance the success of dilatations of esophageal strictures and EIPD.

Introduction
Esophageal intramural pseudodiverticulosis (EIPD) is a rare condition of unclear pathogenesis that was first described in 1960. Since then and through 2011, only about 200 cases have been reported worldwide. The true incidence of EIPD is unknown, though in a retrospective review of esophageal radiograms from 1986, EIPD was found to have a prevalence of approximately 0.15%. The primary symptom of this condition is dysphagia, usually due to the presence of an esophageal stricture that often accompanies diagnosis. Associated conditions reported in the literature include diabetes, alcoholism, gastroesophageal reflux disease (GERD), fungal infections, and esophageal neoplasms. Dysmotility disorders are occasionally associated with EIPD, including 2 reported cases of achalasia and 1 case of nutcracker esophagus, though no causal relationship has been elucidated. Treatment is often limited to endoscopic dilatation and treatment of the underlying esophageal condition, such as acid suppression therapy or treatment of fungal infections. In rare cases, esophagectomy has been required. We present a case of a patient with EIPD managed successfully with dilatation and sucralfate.

Case Report
A 58-year-old female was admitted to the hospital with several months of progressive dysphagia and a 20-lb weight loss. Her medical history was significant for GERD, for which she was only prescribed famotidine due to a prior anaphylactic allergy to proton pump inhibitors (PPIs). A barium esophagram showed a smooth mid-esophageal stricture and several intramural diverticula and intramural tracts distal to the stricture (Figure 1).

An esophagogastroduodenoscopy (EGD) was done to examine the stricture. The luminal diameter was approximately 5 mm, and length was 6 mm. There was no desquamation, ulceration, or furrowing of the mucosa. The stricture could only be traversed with an XP 180 endoscope. Distal to the stricture, the esophagus was found to have innumerable shallow depressions (Figure 2). Multiple biopsies were taken from within and adjacent to the stricture. Pathology showed histologic evidence of esophagitis with neutrophils and lymphocytes, and rare
Esophageal Intramural Pseudodiverticulosis

Tyberg et al

Figure 1. Barium swallow showing intramural diverticula and intramural tracts.

Figure 2. EGD showing esophagus with innumerable shallow depressions.

eosinophils without evidence of malignancy or an alternative diagnosis. The patient was diagnosed with EIPD.

A series of near-weekly balloon dilatations were performed with minimal success. The first dilatation session used a 6-mm, followed by a 7.5-mm, through-the-scope (TTS) balloon. Ten days later, bougie dilatation was performed using a 27 French dilator, and moderate force was required. A TTS balloon was then used to dilate from 9 mm to 10 mm. Two weeks later, the stricture was still unable to be traversed with a GIF-160, and the diameter was estimated to have returned back to 8 mm. A TTS balloon was used to dilate from 8 mm to 12 mm. The fourth dilatation session showed that the stricture still could not be traversed; the diameter had narrowed again. A TTS balloon was used to dilate from 12 mm to 14 mm.

The patient was then prescribed sucralfate suspension 4 times daily. The next dilatation started with a 12-mm balloon, and the stricture was dilated to 15 mm. Dysphagia symptoms largely resolved at this time, though the patient noted on follow-up that she had to be “very careful” eating solid food and chased all oral intake with liquid. One final dilatation session was performed 1 month later. The stricture could be easily traversed with a GIF-160, and the luminal diameter was estimated to be 12 mm. The stricture was dilated with a 15–18-mm controlled radial expansion (CRE) balloon. After these dilatations, the patient’s dysphagia completely resolved, and she has since gained 21 lbs. She has not required further dilation at 1 year follow-up.

Discussion

EIPD is a rare disorder with an unclear pathogenesis. It is often associated with esophageal strictures, and therapy consists mainly of endoscopic dilatation. There have been no reports of the use of esophageal stents for the treatment of EIPD in the literature. EIPD can be associated with other conditions, the most common of which are GERD and fungal infections. Treatment of these other underlying conditions, such as acid suppression in the setting of GERD, is often required. In patients who are unable to tolerate standard acid suppression therapy with proton-pump inhibitors (PPIs), the addition of sucralfate may enhance the success of dilatations of esophageal strictures, as our case illustrates.

A recent Cochrane review showed sucralfate to be associated with a trend towards esophagitis healing in the setting of GERD, though the effect was modest and not statistically significant. Sucralfate functions by forming a physical barrier between esophageal mucosa and harmful agents, promoting mucosal healing, and decreasing the inflammatory response. All of these mechanisms may explain its beneficial role in this case. Regardless of the mechanism, this case suggests that in patients with EIPD and stricture formation who are unable to tolerate PPI therapy, sucralfate may be a viable alternative.

Disclosures

Author contributions: A. Tyberg collected data, wrote the manuscript, and is the article guarantor. D. Jodorkovsky edited the manuscript.

Financial disclosure: None to report.

Informed consent was obtained for this case report.

Received: November 20, 2013; Accepted: March 18, 2014
References

Glass Microparticulate Ingestion: An Unusual and Difficult-to-Diagnose Cause of Chronic Abdominal Pain

R. Brooks Vance, MD1, Marcus Mühlbauer, MD, PhD1, Elizabeth B. Dreesen, MD2, C. Robert Bagnell, Jr., PhD3, Georgette A. Dent, MD3, Hans Herfarth, MD, PhD1, Christian Jobin, PhD1, and Evan S. Dellon, MD, MPH1

1Center for Gastrointestinal Biology and Disease, Division of Gastroenterology and Hepatology, Department of Medicine, University of North Carolina School of Medicine, Chapel Hill, NC
2Department of Surgery, University of North Carolina School of Medicine, Chapel Hill, NC
3Department of Pathology and Laboratory Medicine, University of North Carolina School of Medicine, Chapel Hill, NC

Abstract

In the absence of overt structural abnormalities, the diagnostic approach to chronic abdominal pain can be challenging. Occupational particulate inhalation causing injury to an organ other than the lung is rare. We report a case of inadvertent glass microparticulate ingestion causing chronic abdominal pain with altered local and systemic inflammatory responses.

Introduction

Occupational particulate inhalation causing injury in an organ other than the lung is rare, and when present, can be difficult to diagnose.1,2 We report a case of inadvertent glass microparticulate ingestion causing chronic abdominal pain with altered local and systemic inflammatory responses. We also describe the specialized etiologic evaluation, including x-ray microanalysis, immunohistochemistry, and peripheral blood assays, that were used to confirm the diagnosis, and suggest that altered local and systemic inflammatory response might be responsible.

Case Report

The patient is a previously healthy 38-year-old man who developed chronic recurrent abdominal pain after starting work as a manager at a glass-cutting factory. He was present on the factory floor while glass was being cut, ground, and smoothed, and did not routinely use protective gear. The abdominal pain was episodic, periumbilical, lasted several days, and would resolve spontaneously. He was asymptomatic between episodes.

Physical exam was unremarkable both during and between pain episodes. An extensive laboratory work-up revealed only a mildly elevated C-reactive protein. Blood counts, electrolytes, liver and kidney function, thyroid tests, and iron studies were normal. Blood cultures were negative. Testing for HIV, C1 esterase, complement levels, porphyrins, heavy metals, and paroxysmal nocturnal hemoglobinuria was unrevealing. Upper endoscopy, colonoscopy, and capsule endoscopy were non-diagnostic. Abdominal CT during a pain flare showed mild thickening in the appendix and mesenteric stranding with sub-centimeter lymph nodes. Diagnostic laparoscopy revealed vermiform-appearing adhesions on the abdominal wall (Figure 1). Mesenteric biopsy showed chronic inflammation with evidence of foreign body reaction (Figure 2).
To investigate these histopathologic findings in more detail, specialized testing was performed. X-ray microanalysis revealed the presence of silicon (Figure 3), silver chloride, tungsten, titanium, and vanadium, which are metals commonly used in glass manufacturing. On immunohistochemistry, there was scant staining for IL-1ß. Lipopolysaccharide (LPS)-induced IL-1ß protein secretion was also significantly lowered in this patient compared to healthy controls (Figure 4).

On clinical follow-up of 3 years, exposure avoidance with a respirator mask at work led to significant symptom improvement, and lapses in exposure avoidance led to recurrent symptoms.

Discussion

Occupational particulate ingestion causing inhalational lung injury is well reported. However, particulate involvement of other organ systems is rare, and the clinical significance of such exposures is not well understood. The inflammatory response and cellular changes generated by foreign body reaction have been recently reported for inhalational exposures in coal miners, ship-builders, and glass manufacturers. Ferrira et al described a glass laminator with inflammatory lung granulomas consisting of silica-predominant particulate. However, only 1 report has described similar exposures involving the GI tract in “glass dust esophagitis” in a glass grinder who presented with chest pain.

Dysfunction of the intestinal inflammasome, particularly with altered levels of cytokines such as IL-1ß, may contribute to many gastrointestinal conditions, including inflammatory bowel disease, colon cancer, and functional bowel disorders. In our patient, we detected a number of mesenteric glass microparticulates suggestive of inadvertent ingestion and subsequent microperforation. We also found lower IL-1ß levels locally in mesenteric tissue and systemically with blunted IL-1ß secretion after LPS stimulation. There is evidence that silica can suppress murine macrophage produc-
Vance et al
acgcasereports.gi.org
Glass Microparticulate Ingestion

ACG Case Reports Journal | Volume 1 | Issue 3 | April 2014

We hypothesize that inadvertent glass microparticulate ingestion and microperforation led to a silica-induced impairment of IL-1ß secretion and subsequent chronic inflammation and pain. It is important to consider occupational exposures in patients with unexplained chronic gastrointestinal complaints that do not fit with symptom patterns in other established disorders.

Disclosures

Author contributions: RB Vance collected and interpreted data, and drafted, critically revised, and approved the manuscript. M. Mühlbauer collected, interpreted, and analyzed data, and critically revised and approved the manuscript. EB Dreesen provided patient care and critically revised and approved the manuscript. CR Bagnell and GA Dent provided pathologic analysis and interpretation, and critically revised and approved the manuscript. H. Herfarth and C. Jobin designed the study, interpreted data, and critically revised and approved the manuscript. ES Dellon designed the study, provided patient care, collected and interpreted data, drafted, critically revised, and approved the manuscript, and is the article guarantor.

Financial disclosure: None to report.

Informed consent was obtained for this case report.

Previous Presentation: This paper was presented as a poster at the 2012 ACG Annual Meeting.

Received: January 20, 2014; Accepted: March 4, 2014

References


Figure 4. White blood cells from the patient display impaired LPS-induced IL-1ß protein secretion compared to healthy controls (n=3). Error bars represent the standard deviation of the measurements (each experiment was performed in triplicate for each of the samples).
Hemophagocytic Lymphohistiocytic Syndrome and Enteropathy-Associated T-cell Lymphoma in a Patient with Refractory Celiac Disease

Lucy Lu, MD1, Shuoyan Ning, MD1, Zain Kassam, MD2, Richard Hunt, MB, MACG3, and Marco Puglia, MD3

1Department of Medicine, McMaster University, Hamilton, Ontario, Canada
2Harvard School of Public Health, Harvard University, Boston, MA
3Division of Gastroenterology, McMaster University, Hamilton, Ontario, Canada

Abstract
A 70-year-old woman with celiac disease presented with weight loss and diarrhea unresponsive to gluten-free diet (GFD) and prednisone. Diagnosis of type 2 refractory celiac disease (RCD) was made by small intestinal biopsies showing severe villous blunting and intraepithelial lymphocytosis. She was diagnosed with hemophagocytic lymphohistiocytic syndrome (HLH) after developing fever, pancytopenia, hypofibrinogenemia, elevated ferritin, and demonstration of hemophagocytosis on her bone marrow biopsy. An expert pathologist on lymphoma reviewed her biopsies and revised the final diagnosis to type 1 enteropathy-associated T-cell lymphoma (EATL) based on large T-cells infiltrating the lamina propria. We describe the first case of HLH associated with localized EATL and RCD.

Introduction
Hemophagocytic lymphohistiocytic syndrome (HLH) is a rare, rapidly progressive and potentially fatal syndrome characterized by overactive histiocytes. HLH has been described in advanced enteropathy-associated T-cell lymphoma (EATL), a type of non-Hodgkin’s T-cell lymphoma associated with celiac disease. We report the first case of HLH associated with localized EATL in the context of refractory celiac disease (RCD).

Case Report
A 70-year-old woman with a 4-year history of celiac disease was referred for RCD unresponsive to strict gluten-free diet (GFD) and 1 month of treatment with prednisone. She initially presented with a 25-lb weight loss over 4 months, non-bloody diarrhea, and abdominal bloating, and had been diagnosed via duodenal biopsies showing villous atrophy. Since then, she had followed a strict GFD.

Prior to referral, she had negative evaluations for metabolic and infectious causes of diarrhea. Her blood work showed increased anti-tissue transglutaminase IgA, antigliadin antibody IgG, and antigliadin IgA, with normal total IgA levels. An abdominal computed tomography (CT) showed inflammation in the small bowel with loss of the normal jejunal mucosa. Five days into her admission, she developed melena; esophagogastroduodenoscopy (EGD), colonoscopy, and push enteroscopy did not identify a source of bleeding. She was diagnosed with type 2 RCD based on duodenal and jejunal biopsies, which demonstrated severe villous blunting, intraepithelial lymphocytosis, and lymphoplasmacytic infiltration. Her diarrhea persisted despite a strict GFD and prednisone. Cyclosporine 60 mg IV was started but discontinued due to drug-related fever. She was treated empirically with piperacillin/tazobactam and transferred to our center.
On referral, the patient was cachectic, tachycardic, and hypotensive with evidence of ongoing gastrointestinal bleeding. A repeat abdominal CT showed no small bowel abnormality, hepatosplenomegaly, or lymphadenopathy. HLH was suspected after the patient developed pancytopenia, hypofibrinogenemia, elevated liver enzymes, and hyperferritinemia (19,574 ug/L; normal: 51–400 ug/L) in the context of ongoing fever. Bone marrow biopsy confirmed the diagnosis, revealing prominent hemophagocytosis (Figure 1). An HLH treatment protocol was initiated with dexamethasone 10 mg IV twice daily, cyclosporine 100 mg IV twice daily, anakinra 100 mg subcutaneously daily, 1 dose of etoposide 100 mg IV, and intravenous immunoglobulin (IVIG), along with transfusions of blood products. She was unresponsive to treatment and remained pancytopenic while her ferritin increased to 60,552 ug/L. Persistent diarrhea and GI bleeding were suspicious for a small bowel EATL. A pathologist experienced in lymphoma reviewed her previous small intestinal biopsies and revised the final diagnosis to include type 1 EATL based on the high proportion of large T-cells with prominent nucleoli infiltrating the lamina propria and the abnormal T-cell marker profile (Figure 2). Unfortunately, on the day of the diagnosis, the patient passed away from a small intestinal bleeding.

**Discussion**

We describe a rare case of EATL-associated with RCD and subsequent development of HLH, of which there are very few reported cases. RCD is a diagnosis of exclusion defined by ongoing symptoms and persistent villous atrophy despite a strict GFD for 1 year. RCD is classified into type 1 (normal intraepithelial lymphocyte morphology) and type 2 (abnormal intraepithelial lymphocyte morphology). Type 2 RCD, often diagnosed in elderly women, is more commonly associated with serious complications, with 60–80% of patients developing EATL within 5 years. It carries a 5-year survival rate of 40–58%. HLA-DQ2 haplotype is present in up to 98% of cases.

EATL is a rare form of non-Hodgkin’s T-cell lymphoma that is associated with celiac disease in up to 70% of cases. EATL usually develops in the jejunum or ileum, but can arise in any part of the gastrointestinal tract. Two types of EATL exist. Type 1 EATL is strongly linked to celiac disease and RCD and is characterized by large cells or non-monomorphic cytology with negative CD56 and positive CD30 T-cell marker expression. Type 2 EATL has a monomorphic cytology with CD56 expression. The prognosis of EATL is poor. Median progression-free survival ranges between 3 and 6 months, with 5-year progression-free survival of 3.2–18%. Mortality typically results from perforation, bleeding, and malabsorption.

EATL may not be identified on standard imaging. CT enteroclysis may detect tumors as small as 5 mm, with sensitivity and specificity of 95–100%. Positron emission tomography (PET) scans may be more sensitive than CT for detecting EATL, but experience with magnetic resonance imaging (MRI) for diagnosing EATL is limited. Validated magnetic resonance (MR) enteroclysis scores have been developed for identification of type 2 RCD and CD-related small bowel non-Hodgkin’s lymphoma (NHL). The presence of less than 10 folds per 5 cm of jejunum, mesenteric fat infiltration, and bowel wall thickening are associated with type 2 RCD. The diagnosis of EATL relies primarily on histology. Treatment options for EATL include surgical resection and anthracycline-based chemotherapy with modest survival benefits. Emerging therapies utilizing high-dose induction chemotherapy and autologous stem cell transplantation have demonstrated improved survival.

HLH is characterized by the activation, proliferation, and abnormal function of histocytes. It requires at least 5 of the following 8 criteria: fever, splenomegaly, peripheral cytopenia of 2 lineages, hypertriglyceridemia and/or hypofibrinogenemia, hyperferritinemia, elevated soluble CD25, absent natural killer cell activity, and histological hemophagocytosis in the bone marrow, lymph nodes, or spleen. Primary HLH is an autosomal recessive genetic disorder. Secondary HLH is associa-
Hemophagocytic Lymphohistiocytic Syndrome

There is no published case report of RCD associated with HLH. Only 7 cases have been reported of EATL-associated HLH, of which all patients had metastatic EATL before development of HLH and died within 3 months. Our patient rapidly declined after developing HLH and died shortly after a confirmed diagnosis of HLH.

This is the first case of EATL-associated HLH treated with HLH-specific therapy. Since vinblastine resulted in temporary improvement in a previous case report, perhaps these patients should be treated more aggressively for their underlying EATL as opposed to HLH-directed therapy. Our patient did not receive EATL-specific therapy because the diagnosis of EATL was delayed. Our case demonstrates that HLH can develop with RCD and early-stage EATL. Gastroenterologists should consider screening patients with RCD and EATL for HLH. Prompt referral is crucial for early diagnosis and treatment of EATL and HLH. When suspicion is high and EATL is not apparent on standard imaging, clinicians should remember that EATL requires histological diagnosis by an experienced pathologist.

Disclosures

Author contributions: L. Lu and S. Ning drafted the manuscript. Z. Kassam, R. Hunt, and M. Puglia reviewed and edited the manuscript. All authors approved the final manuscript. M. Puglia is the article guarantor.

Acknowledgements: Dr. Denis Bailey and Dr. Catherine Ross for providing the pathology slides and assisting in their interpretation.

Financial disclosure: None to report.

Informed consent was obtained for this case report.

References

Asymptomatic Duodenal Perforation from an Inferior Vena Cava Filter

Jean R. Park, MD, Veeral M. Oza, MD, and Somashekar G. Krishna, MD, MPH

Division of Gastroenterology, Hepatology and Nutrition, Department of Medicine, The Ohio State University, Columbus, OH

Abstract

Recent exponential increase in inferior vena cava (IVC) filter placements has led to a higher rate of filter complications. A 46-year-old man with a past history of IVC filter placement for bilateral deep vein thrombosis presented with lower abdominal pain. Imaging studies demonstrated IVC filter strut penetrations into multiple structures. Upper endoscopy confirmed an uncomplicated single IVC filter strut penetration into the duodenal wall. The abdominal pain was determined to be unrelated to IVC filter strut penetration, and the patient was managed conservatively. Although IVC filter strut penetrations can cause significant complications, current guidelines remain unclear for management of asymptomatic enteric IVC filter strut penetrations.

Introduction

Over the last decade, inferior vena cava (IVC) filter placement has increased dramatically. Complications due to IVC filter include perforation of the aorta, the psoas muscles, and the gastrointestinal (GI) tract.1,2 These perforations are exceedingly rare and usually symptomatic.3 Existing literature describes cases of symptomatic duodenal perforations from IVC filters managed by surgical procedures.2 However, there is a paucity of literature on the treatment of asymptomatic penetrations. We present a patient with an asymptomatic duodenal penetration by an IVC filter.

Case Report

A 46-year-old man with a history of paraplegia from prior trauma, bilateral lower extremity thrombus, and placement of an IVC filter 6 years ago underwent a computed tomography (CT) for evaluation of lower abdominal pain. The patient also complained of daytime urinary frequency, incontinence, nocturia, urgency, and weak urinary stream, and had evidence of microscopic hematuria. The CT scan was remarkable for IVC filter strut penetrations into the right psoas muscle, L3 vertebral body, and possibly the duodenum (Figure 1). The patient denied symptoms of GI bleeding and a complete blood count was unremarkable. An esophagogastroduodenoscopy (EGD) confirmed a single IVC filter strut penetrating into the second part of the duodenal wall. The intraluminal strut measured 1.5 cm in length, and the end of the strut was hooked, providing a soft end (Figure 2). There was no ulceration or bleeding at the site of penetration. The vascular surgeons felt that the potential complications from IVC filter removal would outweigh the benefits in an otherwise asymptomatic patient. Meanwhile, laboratory evaluation revealed that the patient had a urinary tract infection, and urology service diagnosed benign hypertrophy of prostate. His abdominal pain was attributed to this cause. He was started on tamsulosin with antibiotics and later reported resolution of lower abdominal pain and urological symptoms. After 1 year of follow-up, the patient remains asymptomatic and free of complications secondary to duodenal penetration by the IVC filter.
Asymptomatic Duodenal Perforation from an IVC Filter

Discussion

Caval filters are classified by their anatomical site: infrarenal or suprarenal. Further categories include permanent filters (indicated for patients with life-long risk of pulmonary embolism) or retrievable/optional filters (for perioperative use). The optional filters have a retrieval hook facilitating easy removal, and the majority of filters currently being used are the retrievable type. Interventional radiology guidelines list access site thromboses, IVC occlusions, and IVC penetration as the most frequently reported complications. Other complications include filter movement and tilt, fracture or embolization, deployment outside the target area, and recurrent pulmonary embolism. Filter tilt is the resultant angulation after insertion and is usually minimal after placement in the IVC. A tilt of >15° away from the long axis of the IVC reduces efficacy and predisposes to caval perforation. To reduce risk of filter tilt, manufacturers have implemented ‘hooks’ and ‘legs’ at the end of the filter. The overall incidence of filter perforation through the IVC wall is approximately 0.3%. Currently, there are no guidelines for the management of patients with asymptomatic IVC filter penetrations into the duodenum. As the risks of removing the filter outweigh the any likely benefits in such cases, asymptomatic patients require a multidisciplinary approach involving interventional radiology, gastroenterology, and vascular surgery, and a closely monitored non-operative follow-up. In symptomatic patients, removal of the filter is difficult due to exposure of the IVC bounded by inflammatory reaction, so surgical treatment is recommended only after definitive failure of conservative management and if symptom relief is certain. When removal is contemplated, CT and EGD should be used to evaluate filter strut penetrations, and an open procedure by exploratory laparotomy is the typical removal technique. Venotomy is performed to remove the filter, and if the filter struts are not extractable, they should be trimmed with wire cutters. In our patient, a non-surgical approach was successfully adopted for the management of IVC filter strut penetration through the duodenal wall.

Disclosures

Author contributions: JR Park and VM Oza completed the literature review and wrote the manuscript. SG Krishna participated in clinical management of the patient, reviewed the manuscript, and is the author guarantor.

Financial disclosure: None to report.

Informed consent was obtained for this case report.

Received: January 23, 2014; Accepted: March 16, 2014

References

Hematochezia Associated with Sevalamer-Induced Mucosal Injury

Preethi Chintamaneni, MD1, Rohit Das, MD2, Shih-Fan Kuan, MD3, Taher R. Kermanshahi, MD3, and Jana G. Hashash, MD2

1Department of Internal Medicine, University of Pittsburgh Medical Center, Pittsburgh, PA
2Department of Gastroenterology, Hepatology, and Nutrition, University of Pittsburgh Medical Center, Pittsburgh, PA
3Department of Pathology, University of Pittsburgh Medical Center, Pittsburgh, PA

Abstract
We present a case of a 61-year-old woman with end-stage renal disease (ESRD) who developed painless hematochezia following initiation of anticoagulation. Work-up revealed a large ulceration in the sigmoid colon, and histologic images revealed sevelamer crystals embedded in the colonic mucosa, consistent with sevelamer crystal-mediated injury. This is a novel cause of gastrointestinal hemorrhage that has not previously been described in the literature. Physicians should be aware of the potential for sevelamer-induced injury.

Introduction
Non-absorbable drugs such as sequestering agents exert their effect in the gastrointestinal lumen by binding a target molecule and forming an insoluble complex preventing absorption. This class of drugs includes many commonly used medications such as sodium polystyrene sulfonate, cholestyramine, and sevelamer. While the potential for mucosal injury of the gastrointestinal tract has been demonstrated in the literature with the use of cholestyramine and sodium polystyrene sulfate, there have been few reports of sevelamer-associated mucosal injury.

Case Report
A 61-year-old African-American woman with a history of end-stage renal disease (ESRD) requiring chronic hemodialysis presented to our hospital for management of a thrombosed arteriovenous graft. After mechanical thrombectomy, the patient was started on an IV heparin infusion with transition to warfarin. Two days post-operatively, she developed painless hematochezia. Her medications included calcium acetate, cinacalcet, diltiazem, glipizide, hydroxyzine, losartan, omeprazole, pravastatin, and sevelamer 2,400 mg by mouth 3 times daily. She denied any recent nonsteroidal anti-inflammatory drug use. She denied having problems with constipation. Prior to surgery, the patient received a brief course of high-dose steroids as premedication for a contrast allergy.

Physical examination revealed a hemodynamically stable obese woman in no acute distress. Her abdomen was soft, non-tender, and non-distended. Rectal exam revealed dark red blood with no hemorrhoids or anal fissures. Laboratory evaluation revealed hemoglobin 10.8 g/dL (baseline: 11.1 g/dL). Colonoscopy revealed a 5-cm semi-circumferential ulceration in the sigmoid colon 25 cm from the anal verge (Figure 1 and Figure 2). The ulcer was not actively bleeding, so no endoscopic intervention for hemostasis was performed. The ulcer border was biopsied, and histology revealed an ulcer bed with attached fibrin, hemorrhage, and entrapped pale yellow-to-or-
ange crystals, which is a finding associated with sevelamer-induced mucosal injury (Figure 3). Sevelamer was discontinued with no recurrence of hematochezia, and a heparin drip was restarted 6 hours after the colonoscopy without any subsequent gastrointestinal bleeding. After discussion with the vascular surgery team, the patient was switched to clopidogrel for long-term anticoagulation. Repeat endoscopy is planned 6 months after this bleeding episode to check for mucosal healing after discontinuation of sevelamer.

Discussion

Sevelamer is an anion exchange resin that is approved for use in the United States and Europe to treat hyperphosphatemia in patients with ESRD who are on hemodialysis. It is a nonabsorbable polymer that dissociates from its anion (either carbonate in Renvela® or hydrochloride in Renagel®) and binds to dietary phosphate within the gastrointestinal tract preventing reabsorption. The most common side effects associated with sevelamer use are gastrointestinal, including vomiting (22%), nausea (20%), diarrhea (19%), dyspepsia (16%), and constipation (8%). The most concerning adverse effect is metabolic acidosis, which is seen with sevelamer hydrochloride (Renagel®) but not with sevelamer carbonate (Renvela®). Sevelamer use is contraindicated in the presence of bowel obstruction. Sevelamer crystals are non-polarizable and are characterized as having broad, curved, irregular fish scales. Sevelamer crystal deposition in the colon may contribute to mucosal inflammation and ulcer formation. On hematoxylin and eosin stain, the crystals are two-toned in appearance with bright pink accentuations on a rusty yellow background.

There is little existing literature regarding sevelamer crystal-mediated mucosal injury. A prospective series identified 15 pathology specimens from throughout the gastrointestinal tract containing sevelamer crystals, of which 14 had abnormal background mucosal findings. While some of these cases were felt to include incidental sevelamer crystal deposition in the context of other comorbidities, there were also patients with findings suggestive of direct sevelamer crystal-mediated injury as evidenced by acute ulcerations in various parts of the gastrointestinal tract. They also found a dose-dependent relationship with an increased degree of mucosal injury in patients receiving higher doses of sevelamer.

Madan et al described an incident of sevelamer use causing lower gastrointestinal bleeding through the formation of a stercoral ulcer. Stercoral ulceration is defined as an ulcer caused by pressure necrosis from a fecaloma. In this

Figure 1. A 5-cm semi-circumferential ulceration in the sigmoid colon 25 cm from the anal verge revealed by colonoscopy.

Figure 2. Ulcer in the sigmoid colon.

Figure 3. Hematoxylin and eosin stain of ulcer bed with sevelamer crystals.
case, sevelamer caused severe constipation, which in turn resulted in formation of a fecaloma and subsequently a stercoral ulceration in the rectum. In contrast to this report, our patient did not report constipation and her ulcer was in the sigmoid colon, thus stercoral ulceration unlikely to be the cause of her colonic mucosal injury.

With the growing use of sevelamer in patients with ESRD, it is important to recognize the potential clinical impact of mucosal injury associated with sevelamer crystal deposition. Unfortunately, at this time, there are no known strategies described in the literature to prevent sevelamer-associated mucosal injury. Our case suggests that there may be an association between sevelamer use and mucosal injury, but further evidence is necessary to indicate causation. Sodium polystyrene sulfonate is a cation-exchange resin used in the treatment of hyperkalemia that is believed to be directly toxic to the colonic mucosa, resulting in colonic ischemia and ulceration. We postulate that sevelamer potentially mediates mucosal injury under a similar mechanism, and that sevelamer crystals deposit in the mucosa of the colon, where they are directly toxic and result in ischemic changes and subsequent ulcer formation.

Disclosures

Author contributions: P. Chintamaneni and R. Das acquired and interpreted the data, and drafted the manuscript. SF Kuan and TR Kermanshahi acquired and interpreted the data. JG Hashash interpreted the data, and drafted and critically revised the manuscript for important intellectual content. P. Chintamaneni is the article guarantor.

Financial disclosure: None to report.

Informed consent was obtained for this case report.

Received: January 15, 2014; Accepted: March 16, 2014

References

Use of Serum Infliximab Level Prior to Cyclosporine Salvage Therapy in Severe Ulcerative Colitis

Christopher G. Chapman, MD, Ashley Bochenek, MSN, APN/FNP-BC, Adam C. Stein, MD, and David T. Rubin, MD

Inflammatory Bowel Disease Center, University of Chicago Medical Center, Chicago, IL

Abstract
Medical treatment options for severe, steroid refractory ulcerative colitis (UC) include infliximab (IFX) or cyclosporine (CSA), but general consensus has been that both agents should not be used together or even successively. We report a case of a 17-year-old male with severe UC refractory to IV steroids with successful sequential salvage therapy guided by serum IFX level. After primary lack of response to IFX, an undetectable serum IFX level and elevated IFX antibodies were followed by immediate transition to IV CSA. This case demonstrates the possibility of therapeutic drug monitoring of IFX levels when calculating the risk/benefit ratio for patients with steroid-refractory UC failing primary salvage therapy.

Introduction
Both infliximab (IFX) and cyclosporine (CSA) are effective for the treatment of severe ulcerative colitis (UC) that is refractory to IV steroids, but general consensus has been that both agents should not be used together or even successively. A recent randomized trial of these agents in IV steroid-refractory patients found no difference in efficacy, but 40% of patients did not respond to the therapy in either arm. Case series have reported significant infectious events when the 2 agents are used for salvage therapy in close succession. We report a case of severe UC with successive salvage therapy guided by serum IFX level.

Case Report
A 17-year-old male with a 6-month history of pan-UC presented to our center. He had primary non-response to 8 weeks of mesalamine and continuous oral prednisone. He was started on combination therapy with azathioprine (AZA) 50 mg per day and IFX 5 mg/kg IV at 0 and 2 weeks, but due to lack of response, received 7.5 mg/kg IV at 6 weeks. Despite this approach, he was unable to wean from prednisone with persistent disease activity characterized by 6–8 bloody loose stools per day with associated severe urgency and nocturnal bowel movements. He received his next IFX at 7.5 mg/kg 6 weeks later. When this failed to control his colitis, he transferred care to our institution and was admitted. At that time (2 weeks after his last IFX dose) admission labs (Table 1) and serum IFX level and antibodies to IFX (ATI) were ordered (electrochemiluminescence immunoassay [ECLIA]; Esoterix Endocrinology, Calabasa Hills, CA).

He was found to have Clostridium difficile (C. difficile) infection and was treated with oral vancomycin 125 mg 4 times daily, metronidazole 500 mg IV every 8 hours, and IV methylprednisolone 20 mg every 12 hours. He was maintained on dual therapy for C. difficile for 12 days with continuation of oral vancomycin monotherapy for an additional 18 days. Despite clearance of C. difficile (by negative PCR 7 days after initiating therapy), he continued to have frequent bloody stools and urgency. Colonoscopy confirmed the presence of diffuse moderate-
Cyclosporine Salvage Therapy in Severe UC

Chapman et al

Moderate-to-severe active colitis (Figure 1). Biopsies demonstrated no evidence of viral cytopathic change, and immunostains were negative for cytomegalovirus infection. Eight days after admission, IFX and ATI results showed that serum infliximab was undetectable, and ATI was 265 ng/mL (reference: <22 ng/mL). After discussion with the patient, we decided to pursue further medical management. CSA 2 mg/kg IV continuous infusion was started (goal serum level: 300–400 ng/mL) with standard antibiotic prophylaxis for Pneumocystis pneumonia with trimethoprim/sulfamethoxazole (TMP/SMZ) and continued IV steroids. After 4 days of CSA, the hematochezia had stopped and the diarrhea started to resolve. He was discharged from the hospital on oral CSA, prednisone, TMP/SMZ, and AZA (Figure 2). At the time of this report 9 months later, he remains in steroid-free remission only on AZA, and there were no adverse events.

Discussion

Medical options for hospitalized patients with severe, corticosteroid-refractory UC include IFX or CSA. Initial response rates for both CSA and IFX in this challenging patient population are similar, ranging from 62% to 83%, respectively, and 71% of patients at 3 months. A recent parallel, open label, randomized control trial comparing the 2 therapies found IV CSA and IFX to be no different for achieving short-term remission and colectomy-free survival, with similar rates of serious adverse events.

In patients treated with either CSA or IFX who do not respond to therapy, options include colectomy or, in a selective cohort, successive salvage therapy with the other agent. Published experience with successive salvage medical therapy has been limited to several small case series. Manosa et al reported that 6 of 16 patients receiving IFX salvage after CSA (38%) required colectomy after a median follow-up time of 195 days, while in a similar cohort, Chaparro et al reported that 29% progressed to colectomy at 1 year. Maser et al reported a retrospective study of 19 patients with steroid-refractory UC who were treated with CSA or IFX within 30 days as sequential rescue therapy. At 1 year, 8 (42%) patients required colectomy; steroid-free remission was achieved in 40% of the IFX-salvage cohort and 33% in the CSA-salvage cohort without statistical difference between them. In the largest cohort published to date, Leblanc et al published a retrospective study of 86 patients in which 48 (56%) patients had a colectomy despite sequential salvage therapy.
at 1 year, while 22% of patients had steroid-free remission.

The decision to pursue sequential medical therapy must account for the risks of delaying surgical intervention and that dual immunosuppression raises the risk of infection. Maser et al reported 3 (16%) serious infectious adverse events, including 1 death from gram-negative sepsis in the IFX salvage group. Leblanc et al reported that their IFX salvage group suffered 9 (10.5%) serious infectious complications as well as 1 death due to post-colectomy pulmonary embolism, possibly related to delayed surgical intervention. Chaparro et al also reported an infection adverse event rate of 8.4%, including 1 death due to post-colectomy nosocomial pneumonia.

The significantly longer half-life for IFX (8–10 days) versus CSA (6–8 hours) has led some to suggest the use of CSA as a first approach to steroid-refractory UC; if CSA fails, a shorter wash-out period may allow for safer IFX use. A lack of experience with CSA by gastroenterologists has made this option impractical. There is increasing evidence that the primary non-response to IFX in severe colitis may be partially attributed to rapid metabolism or rapid clearance of the drug, with emerging reports of fecal elimination of the drug as a potential explanation.12,13

This is the first report of the use of therapeutic drug monitoring to minimize toxicity and guide IFX-CSA salvage therapy. We believe that this patient’s lack of response to IFX was due to rapid clearance and anti-drug antibodies. We do not know if he was losing IFX through a protein-losing colopathy and this “pseudo-episodic” therapy resulted in immunogenicity against the drug, or if he already had ATI that neutralized the IFX. In either explanation, it was clear that the subtherapeutic drug level was contributing to his lack of response to this therapy. The undetectable IFX level provided reassurance for use of salvage CSA. We propose that therapeutic monitoring may be used in the assessment of salvage therapies for selected patients.

A practical limitation to this approach is the time required to obtain IFX and ATI levels with currently available clinical assays, which delays clinical and therapeutic decisions for the patient. We believe that waiting for these results is worthwhile, considering the risks of surgery that may be averted. In the future, we hope “point of service” assays and improved turnaround times—driven by volume and demand—will reduce waiting time and make this approach more practical.

**Disclosures**

Author contributions: CG Chapman and AC Stein were involved with patient care and wrote the manuscript. A. Bochenek was involved with patient care. DT Rubin was involved with patient care, reviewed and revised the manuscript, and is the article guarantor.

Financial disclosure: No financial support was received, and the authors have no conflicts of interest to report.

Informed consent was obtained for this case report.

Received: November 3, 2013; Accepted: December 16, 2013

**References**


A Unique Case of Hematemesis in a 17-Year-Old Female

Tobias Zuchelli, MD, Eva Alsheik, MD, Bhavik Bhandari, MD, and Daniel Ringold, MD

Division of Gastroenterology and Hepatology, Drexel University College of Medicine, Philadelphia, PA

Abstract

Hemosuccus pancreaticus (HP) is a rare cause of gastrointestinal bleeding (GIB) that should be considered in a patient with a history of pancreatitis and GIB. A 17-year-old female presented with nausea followed by an episode of hematemesis. Fourteen weeks prior to presentation, she had 3 episodes of vomiting within a week. Six weeks prior to presentation, she developed abdominal pain and was diagnosed with acute idiopathic pancreatitis. Computed tomography (CT) revealed a cystic lesion arising in the gastroduodenal artery (GDA), and coil embolization was performed. There are no reported cases of HP in an adolescent with acute idiopathic pancreatitis.

Introduction

Hemosuccus pancreaticus (HP) was defined by Sandblom in 1970 as gastrointestinal bleeding secondary to rupture of a pseudoaneurysm into the pancreatic duct.1 This condition is exceedingly rare in the adolescent population. We describe an unusual presentation of HP in a 17-year-old female with a history acute idiopathic pancreatitis.

Case Report

A 17-year old white female presented with a sudden feeling of nausea followed by a single episode of reddish-brown emesis. The patient felt the reddish color was related to her breakfast of red licorice and cherry-flavored breakfast tarts. She reluctantly presented to the emergency room of our tertiary care center for further evaluation.

Fourteen weeks prior, she had 3 episodes of vomiting and abdominal pain and was prescribed a proton pump inhibitor (PPI). Two weeks after stopping the PPI (6 weeks prior to presentation) she experienced recurrent vomiting and abdominal pain requiring admission. Lipase was elevated to 3 times the upper limit of normal, but right upper quadrant ultrasound and laboratory tests, including liver function tests, calcium, and lipid panel, did not reveal an etiology. She was discharged with a diagnosis of acute idiopathic pancreatitis, although endoscopic ultrasound (EUS) and magnetic resonance cholangiopancreatography (MRCP) were not performed.

On this presentation, she had no episodes of nausea or vomiting beyond her first episode and her review of systems was unremarkable. She denied nonsteroidal medications, herbals, alcohol, illicit drug use, recent travels, trauma, and sick contacts. In the emergency room, her vital signs were normal and physical exam was unremarkable, including rectal exam, which revealed brown stool. Admission labs showed hemoglobin 9.0 mg/dL, mean cell volume (MCV) 89 FL, blood urea nitrogen (BUN) 12 mg/dL, and creatinine 0.7 mg/dL. Liver function tests, amylase, and lipase were normal. An esophagogastroduodenoscopy (EGD) performed for anemia and hematemesis showed a large deformity in the antrum and duodenal bulb thought to be secondary to extrinsic compression. There was a deep duodenal ulcer in the area of extrinsic compression without evidence of active bleeding (Figure 1). Given the presence of extrinsic compression, we were concerned that this ulcer was an extrinsic process that warranted further radiologic imaging.
A contrast computed tomography (CT) scan of the abdomen was subsequently performed that revealed a 4.6 x 4.4 x 6.8-cm mixed attenuated cystic lesion with a thin enhancing rim (likely a pancreatic pseudocyst with mass effect on the antrum and duodenum) and an ovoid focus extending from the gastroduodenal artery (GDA) suggestive of pseudoaneurysm formation (Figure 2). The patient was immediately referred to interventional radiology and underwent angiogram, which revealed a 2.8-cm GDA pseudoaneurysm. Successful coil embolizations of the GDA and outflow track of the right gastroepiploic artery were performed (Figure 3). Her remaining hospital stay was uneventful. A follow-up CT scan 10 weeks later showed resolution of the pseudocyst and pseudoaneurysm. Genetic testing for CFTR and PRSS-1 did not reveal an etiology for the pancreatitis, and she currently remains asymptomatic.

Discussion

Hemosuccus pancreaticus (HP) was defined by Sandblom in 1970 as gastrointestinal bleeding secondary to rupture of a pseudoaneurysm into the pancreatic duct. The most common cause of HP is chronic pancreatitis with pseudocyst erosion into a peripancreatic artery and formation of a pseudoaneurysm. Although HP only accounts for 1/1,500 cases of gastrointestinal bleeding, it should be considered in a patient with a history of pancreatitis. Idiopathic pancreatitis, which is diagnosed when no underlying cause is found on routine investigation, comprises an estimated 30% of cases of acute pancreatitis in the adult population. It is exceedingly rare in adolescents. Pseudocysts occur in approximately 10% of all cases of acute pancreatitis, of which 10% can develop HP. The most common arteries implicated in HP are the splenic (45%), gastroduodenal (17%), and pancreaticoduodenal (16%) arteries. Due to the high concentration of digestive enzymes within pseudocysts, pseudoaneurysms can eventually rupture into the ductal system. Bleeding is intermittent due to clot formation in the main pancreatic duct, and active bleeding from the papilla is visualized on upper endoscopy in only 30% of patients. However, upper endoscopy should always be performed to exclude other sources of gastrointestinal bleed. Point ulcers in the gastric or duodenal wall due to aneurysm can also be seen, as was the case in our patient.

Because of the intermittent nature of bleeding and difficulties in determining the bleeding source endoscopically, diagnosis of HP is frequently delayed. The diagnosis is generally made with abdominal CT and/or selective angiography of the celiac trunk and superior mesenteric artery; the latter is 96% sensitive in detecting pseudoaneurysms. Interventional radiology is the primary modality for confirming diagnosis and treatment with coil embolization. If bleeding persists or is life threatening, treatment with a Whipple procedure or pseudocyst resection and ligation of the culprit vessel is indicated.

This case is unique in that the patient presented with acute
Idiopathic pancreatitis followed by a bleed from HP 6 weeks later. There are no similar reported cases of HP in an adolescent with acute idiopathic pancreatitis. In a patient with a history of pancreatitis and gastrointestinal bleed, HP must be considered in the differential to ensure timely diagnosis and treatment of a potentially lethal condition.

**Disclosures**

Author contributions: T. Zuchelli and E. Alsheik drafted and critically revised the manuscript for important intellectual content. B. Bhandari critically revised the manuscript for important intellectual content. D. Ringold drafted and critically revised the manuscript for important intellectual content and provided endoscopic images. T. Zuchelli is the article guarantor.

Financial disclosure: None to report.

The IRB of Drexel University College of Medicine reviewed and approved this case report for publication. This institution deems IRB approval as sufficient consent.

Received: December 13, 2013; Accepted: March 16, 2014

**References**

Groove Pancreatitis: Four Cases from a Single Center and Brief Review of the Literature

Tyler P. Black, MD\(^1\), Cynthia D. Guy, MD\(^2\), Rebekah R. White, MD\(^3\), Jorge Obando, MD\(^4\), and Rebecca A. Burbridge, MD\(^4\)

\(^1\)Department of Internal Medicine, Duke University Medical Center, Durham, NC
\(^2\)Department of Pathology, Duke University Medical Center, Durham, NC
\(^3\)Department of Surgery, Duke University Medical Center, Durham, NC
\(^4\)Department of Gastroenterology, Duke University Medical Center, Durham, NC

Abstract
Groove pancreatitis is a rare form of chronic pancreatitis that affects the groove anatomical area between the head of the pancreas, duodenum, and common bile duct. We provide a summary of the clinical findings of 4 groove pancreatitis cases diagnosed at a tertiary academic medical center over a 5-year period. A detailed review of the current literature surrounding this clinical entity is also provided. Although rare, groove pancreatitis should be considered in the differential diagnosis of patients presenting with pancreatic head mass lesions, as appropriate diagnosis can help avoid unnecessary surgical procedures.

Introduction
Groove pancreatitis is a rare condition characterized by fibrotic inflammation affecting the anatomical area between the head of the pancreas, duodenum, and common bile duct.\(^1\) This condition can often be difficult to differentiate from adenocarcinoma of the pancreatic head, but certain clinical, radiological, and histological features can be useful in making a diagnosis. The prevalence of groove pancreatitis has been difficult to establish, but has been reported as high as 24.5% in pancreaticoduodenectomy specimens from patients with chronic pancreatitis.\(^2\) It often presents in middle-aged males (aged 40–50 years) with a history of alcohol consumption, recurrent or chronic pancreatitis, and pancreatic head mass lesions on imaging.\(^3\) Its presentation is similar to adenocarcinoma of the pancreatic head or, more rarely, autoimmune pancreatitis, but can often be cured by conservative management.\(^4,5\) We present a series of 4 cases of groove pancreatitis diagnosed at a tertiary medical center over a 5-year period, with a detailed review of the current medical literature.

Case Report
Four cases of suspected groove pancreatitis were identified over a 5-year period. Two cases had histological confirmation of groove pancreatitis. The other 2 cases were considered to be likely diagnoses based on the clinical presentation and imaging findings. Table 1 shows a summary of the findings for each case. The average age was 56.5 years. Two patients were African American and 2 were female. All patients presented with abdominal pain and were given a diagnosis of pancreatitis based on an elevation of lipase and/or amylase. Two patients had both computed tomography (CT) and magnetic resonance imaging (MRI), while the other 2 patients had either CT or MRI. A representative CT image from patient 3 is shown in Figure 1. Patients 1 and 4 were given a diagnosis of groove pancreatitis based on their imaging findings, whereas patients 2 and 3 were diagnosed by...
a combination of imaging and histological findings. Histology was obtained for patient 1, but was non-diagnostic. The pancreatic head mass seen on the imaging from patient 1 resolved on a follow-up MRI 2 months later. Histology was not obtained for patient 4. Patients 1 and 3 presented with recurrent episodes of pancreatitis, which is often seen before a diagnosis of groove pancreatitis is established. All 4 patients were active or former smokers. One patient had a significant alcohol use history.

### Discussion

Groove pancreatitis was first described by Becker in 1973 in the German literature as “segmentare pancreatitis.” In 1982, Stolte et al defined groove pancreatitis as a special form of segmental pancreatitis characterized by fibrous scars of the anatomic space between the dorsocranial part of the head of the pancreas, the duodenum, and the common bile duct. In the early 1990s, Becker and Mischke further classified groove pancreatitis into pure and segmental forms. The pure form involves the groove area only, with preservation of the pancreatic parenchyma and main pancreatic duct. The segmental form involves both the groove and the head of the pancreas with stenosis of the pancreatic duct causing upstream dilatation.

The prevalence of groove pancreatitis varies widely. In 3 surgical series, groove pancreatitis was diagnosed in 2.7%, 19.5%, and 24.5% of pancreaticoduodenectomy specimens taken from patients with chronic pancreatitis. Groove

### Table 1. Patient Information and Diagnostics Summary

<table>
<thead>
<tr>
<th>Patient 1</th>
<th>Patient 2</th>
<th>Patient 3</th>
<th>Patient 4</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age, y</td>
<td>53</td>
<td>58</td>
<td>47</td>
</tr>
<tr>
<td>Race</td>
<td>White</td>
<td>Black</td>
<td>Black</td>
</tr>
<tr>
<td>Sex</td>
<td>Female</td>
<td>Male</td>
<td>Male</td>
</tr>
<tr>
<td>Symptoms</td>
<td>Abdominal pain, nausea/vomiting, recurrent episodes of pancreatitis</td>
<td>Abdominal pain, nausea</td>
<td>Abdominal pain, recurrent episodes of pancreatitis</td>
</tr>
<tr>
<td>Tobacco</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>Alcohol</td>
<td>No</td>
<td>No</td>
<td>Yes</td>
</tr>
<tr>
<td>CT</td>
<td>Low-density mass in the region of the pancreatic head adjacent to the duodenum, with surrounding inflammatory changes</td>
<td>None</td>
<td>Multiloculated cystic structure interposed between the pancreatic head and duodenum</td>
</tr>
<tr>
<td>MRI</td>
<td>Pancreatic head lesion without corresponding pancreatic ductal dilatation</td>
<td>Pancreatic head is enlarged and inhomogeneous</td>
<td>Same findings as CT</td>
</tr>
<tr>
<td>Pathology</td>
<td>Non-diagnostic</td>
<td>Pancreatic head and duodenum: chronic pancreatitis with features suggestive of groove pancreatitis (parampullary duodenal wall cyst noted)</td>
<td>Pancreas negative for malignancy</td>
</tr>
</tbody>
</table>

CT=computed tomography; MRI=magnetic resonance imaging.
pancreatitis often presents in alcoholic men aged 40–50 years.7,8 Presenting symptoms consist of abdominal pain, nausea, vomiting, and weight loss.7,8 Laboratory evaluation often reveals an elevation in pancreatic enzymes and rarely an elevation in hepatic enzymes.10 Carcinoembryonic antigen and carbohydrate antigen 19-9 can also be elevated.9

A diagnosis of groove pancreatitis is often difficult to establish. The most important distinction to make is between groove pancreatitis and adenocarcinoma of the head of the pancreas. Ultrasound will typically show a hypoechoic mass, and CT will often reveal a hypodense, poorly enhancing mass between the head of the pancreas and a thickened duodenal wall.11 The most characteristic MRI finding is a sheet-like mass between the head of the pancreas and the duodenum associated with duodenal wall cystic changes and thickening.5 Endoscopic retrograde cholangiopancreatography (ERCP) and endoscopic ultrasonography (EUS) are often useful in the differentiation of groove pancreatitis from pancreatic adenocarcinoma, and should be used early in evaluation even when cross-sectional imaging is not suggestive of a mass.12 Table 2 lists characteristic imaging features differentiating groove pancreatitis from pancreatic adenocarcinoma.

The pathogenesis of groove pancreatitis remains unclear, and several mechanisms have been proposed. The most accepted mechanism is a disturbance of pancreatic outflow in the duct of Santorini through the minor papilla.13,14 Chronic alcohol consumption is felt to contribute by increasing the viscosity of the pancreatic juice, leading to Brunner gland hyperplasia and causing occlusion or dysfunction of the minor papilla.13,14 Another proposed mechanism is pancreatic heterotopia in the minor papilla causing dysfunction and occlusion of the papilla.13 Other factors, such as fibrous scarring secondary to peptic ulcers, gastric and duodenal resections, or true duodenal wall cysts may also be related.5

The treatment of choice is conservative in the acute phase of groove pancreatitis and includes bowel rest, analgesia, and intravenous fluids.10 Patients may have a protracted course requiring supplemental nutrition with tube feeding or total parenteral nutrition. One report demonstrated successful treatment of groove pancreatitis by endoscopic drainage of the accessory pancreatic duct via the minor papilla in patients where obstruction of the minor papilla appears to play a role in the pathogenesis.15 Groove pancreatitis can be resistant to medical treatment and require surgical intervention. The surgical treatment of choice is a Whipple procedure (pancreaticoduodenectomy) or a pylorus-preserving pancreaticoduodenectomy.2

In our series of 4 patients, a combination of clinical and radiological features was used to make the diagnosis of groove pancreatitis. Although other case reports show common alcohol use and a male predominance, 2 of our patients were female and only 1 patient had a history of significant alcohol use. Although rare, this condition should be considered in the differential diagnosis of pancreatic head masses with signs of duodenal wall infiltration. Alternatively, a careful and thorough evaluation to rule out adenocarcinoma should be performed before a definitive diagnosis of groove pancreatitis is established.

Disclosures

Author contributions: All authors contributed equally to the preparation of this manuscript. RA Burbridge is the author guarantor.

Financial disclosure: None to report.

Informed consent was obtained for this case report.

Received: December 20, 2013; Accepted: March 4, 2014

Table 2. Differentiation of Groove Pancreatitis and Pancreatic Adenocarcinoma Based on Imaging Features

<table>
<thead>
<tr>
<th>Imaging</th>
<th>Groove Pancreatitis</th>
<th>Pancreatic Adenocarcinoma</th>
</tr>
</thead>
<tbody>
<tr>
<td>CT</td>
<td>Hypoechoic mass between pancreatic head and duodenum, duodenal wall thickening, cystic changes, and stenosis, normal-to-mildly dilated common bile duct</td>
<td>Pancreatic head mass, with or without dilation of common bile duct and/or pancreatic duct</td>
</tr>
<tr>
<td>MRI</td>
<td>Sheet-like mass in groove, duodenal thickening, common bile duct stenosis</td>
<td>Enlarged mass mostly in pancreatic head, with widening of the space between the distal pancreatic duct and common bile duct and duodenal lumen</td>
</tr>
<tr>
<td>ERCP/EUS</td>
<td>Smooth tubular common bile duct stenosis</td>
<td>Irregular common bile duct stenosis, vascular encasement</td>
</tr>
</tbody>
</table>

CT = computed tomography; ERCP = endoscopic retrograde cholangiopancreatography; EUS = endoscopic ultrasonography; MRI = magnetic resonance imaging

Adapted from Levenick et al.10 and Malde et al.11

References

Bouveret’s Syndrome with Severe Esophagitis and a Purulent Fistula

Rami Bonam, MD¹, Zahid Vahora, MD², Glenn Harvin, MD², and William Leland, MD²

¹Mercer University School of Medicine, Macon, GA
²Internal Medicine, Brody School of Medicine at East Carolina University, Greenville, NC

Abstract

Bouveret’s syndrome is a rare variant of gallstone ileus with an overall incidence of 1–3%. It is a rare cause of gastric outlet obstruction resulting from the passage and impaction of a large gallstone through a cholecystoduodenal fistula. A combination of diagnostic modalities is often required for a diagnosis. Management options include endoscopy and surgery. The most commonly performed procedures are enterolithotomy or gastrostomy, either alone or with cholecystectomy and fistula repair. We describe a unique variant of chronic Bouveret’s syndrome with the unusual associations of severe esophagitis and a purulent fistula.

Introduction

Bouveret’s syndrome is a cause of gastric outlet obstruction that results from the passage and impaction of a large gallstone through a cholecystoduodenal fistula. Bouveret first published 2 cases of this syndrome in 1896.¹ It tends to occur more commonly in women, has a mean age of 74 years, and has an overall incidence of 1–3%.² Various endoscopic and surgical techniques have been utilized in the management of Bouveret’s syndrome, and a number of variants of the syndrome have been reported. We describe a unique variant: chronic Bouveret’s syndrome characterized by the unusual associations of severe esophagitis and a purulent fistula.

Case Report

An 81-year-old Caucasian female with a history of gastroesophageal reflux disease (GERD) and atrial fibrillation was admitted to hospital with a 1-week history of worsening emesis and 2 days of inability to tolerate oral intake. She denied any sick contacts, fever, chills, diarrhea, melena, or abdominal pain. The patient related a similar episode 4 months prior that resolved spontaneously. Work-up at that time included a negative abdominal x-ray. Since then, she had reduced her meal sizes to avoid symptoms and had lost 30 lbs.

Her abdominal exam was unremarkable. Laboratory examination showed normal hemoglobin, white blood cell 20,000/uL, normal liver enzymes, and hypokalemic hypochloremic metabolic alkalosis. An abdominal x-ray showed pneumobilia with no small bowel obstruction. An abdominal computed tomography (CT) confirmed the pneumobilia and revealed a 2.5-cm outpouching in the duodenal bulb suspicious for duodenal diverticula or duodenal ulcer (Figure 1). Esophagogastroduodenoscopy EGD showed severe esophagitis (Figure 2) and a large gallstone completely obstructing the proximal duodenum with surrounding purulent material (Figure 3).

The area of obstruction was impassable endoscopically. Endoscopic lithotripsy was performed by breaking the stone and removing debris via forceps, water pick, snares, balloon sweeps, and a variety of nets. The scope was then advanced, and multiple pieces of stones fell into the distal duodenum. We noted a fistula extending...
into the gallbladder, and multiple stones were seen in the gallbladder. Endoscopic manipulation in the area caused several stones and purulent material to pass through the fistula into the duodenum. The quantity of stones led to aborting the procedure, starting piperacillin/tazobactam, and consulting surgery. At surgery, a gastrostomy allowed for the manual and complete removal of multiple remaining large gallstones. The fistula and gallbladder were left intact due to adhesions in the right upper quadrant and because the cystic duct was patent with no residual gallstones remaining. The patient made an uneventful recovery.

Discussion
Gallstone disease is a common digestive disease with a prevalence of 6% in men and 9% in women in the United States. Gallstone ileus is an unusual complication of cholelithiasis, occurring in less than 0.5% of patients with gallstones. Bouveret’s syndrome is a rare variant and is responsible for 1–4% of all cases of mechanical obstruction. It is caused by the passage and impaction of a large gallstone through a cholecystoduodenal fistula that impacts in the proximal duodenum. After passing through the fistula, most of the gallstones pass asymptptomatically through the bowel. Impaction is most likely to occur in the ileum and less likely to occur in the duodenum and the proximal stomach.

A combination of diagnostic modalities is often required for diagnosis. An abdominal plain film in 30–35% of cases shows the Rigler triad (bowel obstruction, pneumobilia, and a calcified ectopic gallstone). Abdominal ultrasound and CT are the preferred noninvasive diagnostic tests for delineating the gastroduodenal anatomy, and can demonstrate a cholecystoduodenal fistula with ectopic stone location. CT scans show the exact location of the ectopic stone and the site of obstruction and often visualize the biliary–enteric fistula. However, in 15–25% of patients, the stone is isoaattenuating, so differentiation of the stone from the surrounding bile and fluid is not possible. In such cases, magnetic resonance cholangiopancreatography (MRCP) can be helpful to distinguish isoaattenuating stones from fluid and to visualize the fistula.

The management of Bouveret’s syndrome includes endoscopy and surgery. Endoscopic removal should be tried first because it is minimally invasive and has a low rate of complications. Many endoscopic techniques have been described, including mechanical lithotripsy, net extraction, electrohydraulic lithotripsy, intracorporeal laser lithotripsy, or combinations of these techniques. These techniques are
more successful for proximal gallstone obstruction. The success rate of these time-consuming procedures is only 9%. Failed endoscopic attempts and distal migration of gallstones after dislodgement are indications for surgery. The most commonly performed procedures are enterolithotomy or gastrostomy, either alone or with cholecystectomy and fistula repair. A fistula may spontaneously close, and repair may be unnecessary if the cystic duct is patent and no residual gallstones are present.

We suspect that this patient developed a partial obstruction approximately 4 months prior to our intervention. The prolonged partial obstruction led to significant weight loss and the development of esophagitis. The chronic course of this patient’s illness is a unique presentation of Bouveret’s syndrome. Perhaps the development of inflammation from the purulent fistula and elevated WBC count led to edema and complete obstruction. In published case reports of gallstone ileus and Bouveret’s syndrome, patients present with acute symptoms of small bowel obstruction typically within 1 week. Weight loss in Bouveret’s syndrome is reported in less than 20% of patients. A PubMed review of 20 published cases and a comprehensive review of 128 cases revealed only 2% of Bouveret’s syndrome cases presented with esophagitis and none of them had a purulent fistula.

Gallstone-related obstruction of the small bowel is a rare occurrence. Chronic Bouveret’s syndrome is possible when an elderly patient presents with upper gastrointestinal obstruction symptoms, pneumobilia, significant weight loss, and esophagitis. In this patient, a purulent fistula and elevated WBC count was associated with the worsening of clinical symptoms. Antibiotics were instituted to prevent further complications associated with endoscopic instrumentation. Chronic Bouveret’s syndrome should be added to the growing list of variants that have been reported in the literature.

Disclosures
Author contributions: All authors contributed equally to the manuscript. R. Bonam is the article guarantor.

Financial disclosure: No conflicts of interest or funding to disclose.

Informed consent was obtained for this case report.

Received: November 24, 2013; Accepted: February 18, 2014

References
CASE REPORT | BILIARY

A Case of an Ectopic Ampulla of Vater in the Pyloric Channel

Sunil Dacha, MD¹, Xiao Jing Wang, MD², and Emad Qayed, MD¹

¹Division of Digestive Diseases, Department of Internal Medicine, Emory University School of Medicine, Atlanta, GA
²Department of Internal Medicine, Emory University School of Medicine, Atlanta, GA

Abstract

A 51-year-old male presented with abdominal pain and jaundice. He was subsequently diagnosed with cholestatic jaundice and cholangitis. A side-viewing duodenoscope failed to identify the ampulla of Vater in the second portion of duodenum. A regular gastroscope was used, and an ectopic ampulla of Vater was identified in the pyloric channel.

Introduction

The ampulla of Vater encompasses the openings of both the common bile duct and pancreatic duct. It is typically located within the wall of the duodenum, surrounded by the small circular and longitudinal muscular segments that comprise the sphincter of Oddi. We report the case of a man who presented with cholestatic jaundice and cholangitis, and was found to have an ectopic ampulla of Vater in the pyloric channel.

Case Report

A 51-year-old male presented with complaints of intermittent abdominal pain and chills for a few days prior to admission. He was afebrile with normal vital signs. His physical examination was significant for scleral icterus and a benign abdominal exam. Laboratory data were significant for an aspartate transaminase (AST) of 153 IU/L, alanine transaminase (ALT) of 177 IU/L, alkaline phosphatase of 169 IU/L, total bilirubin of 5.5 mg/dL, and a direct bilirubin of 3.5 mg/dL.

Computed tomography (CT) suggested possible annular pancreas with pneumobilia. Right upper quadrant ultrasound revealed cholelithiasis, pneumobilia, and a common bile duct diameter of 8 mm. Magnetic resonance cholangiopancreatography (MRCP) showed a possible annular pancreas with hepatic and pancreatic duct dilation. On attempted endoscopic retrograde cholangiopancreatography (ERCP), the ampulla of Vater was not visualized in the second portion of the duodenum. A regular gastroscope was used for visualization and revealed a 2-cm gastric ulcer in the distal antrum with significant surrounding inflammation (Video 1). Distal to the ulcer in the pyloric channel, two orifices were identified. One of them was draining bile and the other one was draining clear liquid. This was consistent with the biliary orifice and pancreatic orifice, respectively (Figure 1). A sphincterotome with a 0.035 wire was used to enter the orifice that was draining bile, which resulted in deep common bile duct cannulation.

A cholangiogram showed mild intrahepatic duct dilation, common bile duct dilation (to 1 cm), and a “hook-shaped” configuration of the distal common bile duct. These findings were consistent with an ectopic ampulla within the pylorus. A balloon occlusion cholangiogram showed a distal common bile duct diameter of 7 mm and a round filling defect consistent with a bile duct stone (Figure 2). A 3-mm limited sphincterotomy was performed at the ampullary orifice, followed by balloon dilation to 10 mm for 45 seconds under endoscopic and fluoroscopic guidance.


Correspondence: Sunil Dacha, 615 Michael Street Ste. 200, Atlanta, GA 30322 (sdacha@emory.edu).

Copyright: © 2014 Dacha et al. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.
Ectopic Ampulla of Vater in the Pyloric Channel

Dacha et al

1 Most of the reported cases suggest the presence of the major papilla in the stomach. The minor papilla is reportedly found in the distal duodenum. Based on few published reports, there appears to be an increased risk for choledocholithiasis due to anomalous bile drainage and lack of sphincter control mechanisms. Pancreatobiliary secretions can lead to mucosal damage with ulcer formation. The anomalous location in the pylorus can facilitate reflux of gastric contents into the biliary tree, predisposing to biliary tree injury and cholangitis.

Endoscopists who perform ERCP should be aware of this rare anomaly. In cases where the ampulla cannot be seen in a normal anatomical location, an ectopic ampulla should be suspected, and the endoscopist must carefully examine the stomach and the duodenum. As in our patient, a regular, forward-viewing gastroscope can be used to better visualize the stomach. A large sphincterotomy should be avoided due to increased risk of perforation. Ampullary balloon dilation is recommended to extract large bile duct stones.

Disclosures
Author contributions: All authors contributed equally to the manuscript. S. Dacha is the article guarantor.

Financial disclosure: The authors disclose no conflict of interest or financial support.

Informed consent was obtained for this case report.
Received: August 29, 2013; Accepted: January 16, 2014

References


Sarcoidosis Presenting as Necrotizing Sarcoid Granulomatosis of the Liver, Sclerosing Cholangitis, and Gastric Ulcer

Njideka Momah, MD¹, Adetola Otesile, BSc², Rishi Pawa, MD¹, and Steve Shedlofsky, MD¹

¹University of Kentucky, Digestive Diseases and Nutrition, Lexington, KY
²University of Kentucky, College of Medicine, Lexington, KY

Abstract
Sarcoidosis is a multisystem granulomatous disease. The liver is affected in up to 50–90% of cases. Sarcoidosis typically presents as non-necrotizing epithelioid granuloma. The occurrence of non-infective necrotizing sarcoid granuloma (NSG) is infrequent, and the finding of NSG in the liver is rare. We report a case of NSG of the liver and lymph nodes, granulomatous gastric ulcer, and secondary cholangitis coexisting in a patient. We discuss the clinical features of the case and briefly review NSG. There is only 1 previously reported case of NSG of the liver in literature.

Introduction
Sarcoidosis is a multisystem granulomatous disease that has a prevalence of approximately 3–5 per 100,000. The incidence is highest among young adults aged 25–40 years old. The immunopathologic basis is complex and involves interplay between genetic predisposition, sarcoid antigen, and immune cells. The liver is affected in to 50–90% of cases. Sarcoidosis typically presents as non-necrotizing epithelioid granuloma, and presentation as non-infective necrotizing sarcoid granuloma (NSG) is infrequent. The finding of NSG in the liver is rare.

Case Report
A 26-year-old African-American male was referred for epigastric and right upper quadrant (RUQ) abdominal pain and pruritus for 1 year, with associated nausea and intermittent vomiting. He denied nonsteroidal anti-inflammatory use. Physical exam revealed scleral icterus, generalized lymphadenopathy, and tender hepatosplenomegaly. To evaluate his symptoms, an esophagogastroduodenoscopy (EGD) was done and showed esophageal varices and a gastric ulcer. Gastric ulcer biopsy showed chronic active gastritis and acute inflammation with focal granuloma, with stains negative for Helicobacter pylori. RUQ ultrasound showed gallbladder wall thickening and a cholecystectomy was planned; during the operation, the gallbladder was noted to be adherent to the liver and could not be dissected. The liver appeared nodular, so a wedge biopsy was performed, which demonstrated granulomatous inflammation with areas of necrotizing granulomata and portal triads that were difficult to locate.

Laboratory tests showed cholestatic liver pattern (Table 1). He had normal lipase, with negative tests for viral hepatitis and HIV. Ferritin, ceruloplasmin, and alpha-1 antitrypsin serum levels were normal. Cytoplasmic anti-neutrophil cytoplasmic antibodies (C-ANCA), perinuclear anti-neutrophil cytoplasmic antibodies (P-ANCA), anti-nuclear, anti-mitochondrial, anti-smooth muscle, and anti-liver kidney antibodies were negative. Tests for Toxoplasma gondii, Bartonella, Brucella, Treponema pallidum, and Histoplasma were negative. Purified protein derivative (PPD) skin test was non-reactive. Chest computed tomography (CT) showed hilar and mediastinal lymphadenopathy. Abdominal CT showed hepatomegaly with asymmetric left lobe enlargement, splenomegaly,
Sarcoidosis Presenting as NSG

Momah et al

165

and retroperitoneal and periportal lymphadenopathy. A magnetic resonance cholangiogram showed sclerosing cholangitis (Figure 1). An endoscopic retrograde cholangiogram (ERCP) was done to evaluate for extrahepatic biliary obstruction and showed a normal common bile duct, though the left hepatic duct showed diffuse rarefaction and the right hepatic duct did not opacify. To confirm that the lymphadenopathy was part of the same disease process, a right cervical lymph node biopsy was done, which showed necrotizing and non-necrotizing granulomatous inflammation (Figure 2). Gram stains, acid fast bacilli (AFB), and Warthin-Starry stains were negative for microorganisms. Cultures for AFB, anaerobic, and fungal organisms were negative. Given these findings, the patient was diagnosed with sarcoidosis.

The patient was started on 40 mg of prednisone. He showed an improvement in abdominal pain and a decrease in total bilirubin. He was discharged with a plan for outpatient management. His total bilirubin decreased (Table 1) and he continued to require prednisone therapy.

### Discussion

Sarcoidosis is a multisystem disease. The liver is commonly affected, but hepatic presentations are usually mild.¹ Up to 3% of patients develop severe hepatic involvement with portal hypertension and esophageal varices.² Sarcoidosis rarely involves the luminal gastrointestinal tract and is usually asymptomatic. It is diagnosed on the basis of clinical presentation, biochemistry, radiology, and histopathology. The features depend on the affected organ. Hepatic sarcoidosis may present as fatigue, RUQ abdominal pain, pruritus, jaundice, weight loss, and fever.³ Features of esophageal sarcoidosis include dysphagia from impaired motility, esophagitis, stenosis, or extrinsic compression from paraesophageal nodes.⁴ Gastric presentation includes epigastric pain, ulcers, gastric polyps/nodules, or lymphoma-like lesions.⁴ Ten percent of individuals with sarcoidosis have gastric involvement with normal appearing mucosa.⁵ Involvement of the small bowel may clinically present with abdominal discomfort, malabsorption, protein losing enteropathy, gastrointestinal bleed, or intestinal obstruction. Colonic manifestations include colitis, polyposis, or a mass.⁴

Our patient had compelling features of sarcoidosis, including anergy to PPD, elevated serum angiotensin-converting enzyme (ACE) levels, a cholestatic liver biochemistry, hilar and mediastinal lymphadenopathy, and histologic granulomas. The histology of sarcoidosis is typically described as non-necrotizing epithelioid granulomas, though necrotizing granuloma in the setting of sarcoidosis has been described.⁶
There are 3 histologic categories of hepatic sarcoidosis: cholestatic, necroinflammatory, and vascular patterns.\(^7\) Our patient falls in to the necroinflammatory pattern, which has chronic portal inflammation and spotty necrosis.

Liebow first described non-infective necrotizing granulomas and coined the term necrotizing sarcoid granuloma (NSG) in 1973.\(^8\) The majority of reported cases of NSG affect the lungs, but cases involving the skin, subcutaneous tissues, kidney, colon, and orbit have been reported. There is only 1 report of NSG involving the liver,\(^7\) but the clinical presentation of the patient was not described. There is currently no consensus on whether NSG is a manifestation of sarcoidosis or a distinct vasculitic entity. Saldana et al believed that NSG was a different entity. Their patients with NSG lacked extra-thoracic involvement, had normal serum ACE levels, and only a few had hilar adenopathy.\(^9\) Churg et al concluded that NSG was a histologic manifestation of nodular sarcoid.\(^10\)

There is no confirmatory test for sarcoidosis. The diagnosis is based on excluding other causes of granulomatosis, such as primary biliary cirrhosis, Crohn’s disease, tuberculosis, brucellosis, viral hepatitis, fungal infections, lymphoma, and drugs. The treatment of NSG is empiric. Oral prednisone 40–60 mg daily for 8 weeks with a slow taper based on clinical response has been recommended.\(^11\) The unique features of this case are 2 rare manifestations of sarcoidosis—NSG and a gastric ulcer—in the same patient, which has not been previously reported.

Disclosures
Author contributions: N. Momah designed the study and revised and critically reviewed the manuscript for content. A. Otesile obtained and edited the images. R. Pawa critically reviewed the manuscript. S. Shedlofsky revised and critically reviewed the manuscript for content, and is the article guarantor.

Financial disclosure: The authors do not have any financial conflict of interest to disclose.

Informed consent was obtained for this case report.

Received: November 28, 2013; Accepted: March 18, 2014

References
Polymyositis Associated with Hepatitis B Virus Cirrhosis and Advanced Hepatocellular Carcinoma

Kessarin Thanapirom, MD, Satimai Aniwan, MD, and Sombat Treeprasertsuk, MD

Department of Medicine, Chulalongkorn University, Bangkok, Thailand

Abstract

Polymyositis (PM) is an inflammatory condition of skeletal muscle and is believed to be a paraneoplastic syndrome associated with various types of cancer. PM associated with chronic hepatitis B virus (HBV)-related hepatocellular carcinoma (HCC) is very rare. We report a case of advanced HCC with chronic HBV cirrhosis that presented with proximal muscle weakness. Further investigation showed elevation of muscle enzymes, myopathic pattern of electromyography (EMG), and evidence of myositis compatible with PM. Lamivudine and 1 mg/kg of oral prednisolone were given. Two sessions of transcatheater arterial chemoembolization (TACE) were performed and sorafenib was started. Muscle enzymes normalized after 6 weeks of treatment. Unfortunately, 5 months after treatment, patient was readmitted and died of severe bacterial pneumonia.

Introduction

Polymyositis (PM) is an idiopathic inflammatory myopathy. It is a systemic disease that affects skeletal muscles and results in proximal muscle weakness. PM is associated with malignancy in 10–15% of patients. The 3 most commonly associated cancers are nasopharyngeal, lung, and breast cancer. Hepatocellular carcinoma (HCC)-associated PM is quite rare. We report a case of hepatitis B virus (HBV) cirrhosis with advanced HCC presenting with PM.

Case Report

A previously healthy 56-year-old male presented with a 6-week history of fever. Two weeks prior to admission, he developed progressive proximal muscle weakness. Through work-up, he was diagnosed with chronic HBV cirrhosis (Child-Pugh B; MELD 7) with advanced HCC. On physical examination, body temperature was 38°C, blood pressure was 120/75, and pulse rate was 92 bpm. Examination of the limbs showed normal tone without muscle wasting or tenderness. There was bilateral proximal weakness in both upper and lower limbs, with grade 3 of 5 in strength of both flexion and extension based on the Medical Research Council scale (MRC), with normal strength in distal limbs. His neck muscles were also weak. Deep tendon reflexes and sensation were normal. He had hepatomegaly and signs of chronic liver stigmata, but no ascites and no signs of hepatic encephalopathy. His Eastern Cooperative Oncology Group (ECOG) performance status score was 2.

Laboratory revealed albumin 2.2 g/dL, total protein 4.4 g/dL, aspartate aminotransferase (AST) 724 IU/L, alanine aminotransferase (ALT) 236 IU/L, total bilirubin 1.0 mg/dL, alkaline phosphatase 148 U/L, prothrombin time (PT) 14.1 s, international normalized ratio (INR) 1.14, and creatinine 0.8 mg/dL. His creatinine phosphokinase was 17,963 IU/L. Serum electrolytes and thyroid function tests were normal. His viral profiles were positive for HBV with DNA polymerase chain reaction (PCR) of 17,460 IU/mL. Tests for hepatitis C virus and HIV were negative. Alpha-fetoprotein (AFP) was 56,310 ng/mL (normal: <25 ng/mL). Autoantibodies including anti-dsDNA, anti-Jo-1, anti-neutrophilic cytoplasmic antibody, anti-RNP, anti-SSA, and anti-SSB were negative.
Abdominal computed tomography (CT) showed liver cirrhosis with an ill-defined 12 x 7-cm, arterial-enhancing, heterogeneous, hypodense lesion in the left lobe of the liver with increased enhancement on venous and delay phase and central necrosis. The CT showed 2 additional arterial-enhancing lesions, size 6 x 7 cm and 3 x 3 cm, with contrast washout in the venous phase at hepatic segments V/VIII and VII and a left main portal vein thrombosis (Figure 1). Liver biopsy of the largest lesion was performed to exclude secondary liver neoplasm. Histology showed poorly differentiated carcinoma positive for glypican 3 (GPC3) and negative for CK7, CK20, and hepatocyte paraffin 1 (Figure 2), which was consistent with the diagnosis of HCC.

Electromyography (EMG) showed low-amplitude, short-duration action potentials with an early recruitment pattern, normal nerve conduction study, and repetitive nerve stimulation. These findings were highly suggestive of a myopathic pattern (Figure 3). Muscle biopsy showed increased endomysial connective tissue and lymphocyte infiltration with necrotic and regenerating myofibers (Figure 4). No vasculitis or perifascicular pattern was seen, and a diagnosis was polymyositis (PM) was confirmed. Prednisolone 1 mg/kg for PM treatment and lamivudine for preventing hepatitis B reactivation were given. Two sessions of transcatheter arterial chemoembolization (TACE) were performed. His creatinine kinase level decreased to normal after 6 weeks of treatment, but his muscle strength did not improve. Unfortunately, 5 months after treatment, he was readmitted with severe bacterial pneumonia and died after 16 days of hospitalization.

**Discussion**

HCC-associated PM is a rare condition. Only 4 cases of PM associated with HCC have been reported (Table 1). A previous study showed that large tumor size and a high AFP level were commonly found in HCC patients who had paraneoplastic syndromes. Our patient had both a large tumor and a very high AFP level. The pathogenesis of PM has not been identified. A possible mechanism is that an autoimmune process triggered by the tumor leads to clonally expanded CD8-positive cytotoxic T-cells that invade muscle fibers and express major histocompatibility complex (MHC) class 1 antigen and release cytokines, causing muscle inflammation. HBV-associated PM has also been reported, so it is possible that HCC, HBV, or both may have caused PM in this patient.

The role of steroids for treating HCC-associated PM is controversial. Physicians should be aware of an increased risk of infection when using high-dose corticosteroids in patients with advanced HCC and cirrhosis. Our patient's ability to maintain daily life was limited by his weakness; therefore, cor-
Table 1. Summary of Baseline Characteristics, Treatment, and Outcome of Patients with HCC-Associated Polymyositis

<table>
<thead>
<tr>
<th>Case 1</th>
<th>Case 2</th>
<th>Case 3</th>
<th>Case 4</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sex</td>
<td>M</td>
<td>F</td>
<td>M</td>
</tr>
<tr>
<td>Age, y</td>
<td>72</td>
<td>50</td>
<td>37</td>
</tr>
<tr>
<td>Largest tumor size, cm</td>
<td>12</td>
<td>4.8</td>
<td>7</td>
</tr>
<tr>
<td>AFP, ng/mL</td>
<td>Elevated</td>
<td>Elevated</td>
<td>N/A</td>
</tr>
<tr>
<td>PM therapy</td>
<td>Corticosteroid</td>
<td>Corticosteroid</td>
<td>Corticosteroid, methotrexate</td>
</tr>
<tr>
<td>HCC therapy</td>
<td>Chemoembolization</td>
<td>Surgery</td>
<td>Bed supportive care</td>
</tr>
<tr>
<td>Outcomes after therapy</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Clinical weakness</td>
<td>Not improved</td>
<td>Improved</td>
<td>Improved</td>
</tr>
<tr>
<td>Serum CPK</td>
<td>N/A</td>
<td>Decreased</td>
<td>Decreased</td>
</tr>
<tr>
<td>HCC-related death</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>Follow-up, mo</td>
<td>2</td>
<td>2</td>
<td>3</td>
</tr>
</tbody>
</table>

AFP = alpha-fetoprotein; CPK = creatinine phosphokinase; HCC = hepatocellular carcinoma; N/A = not applicable

ticosteroids were given after discussion with him. As a result of corticosteroid therapy, lamivudine was needed for prevention HBV reactivation. Lamivudine has good efficacy for this indication, particularly when the HBV DNA is low, as in our patient.13,14 Most patients with HCC-associated PM have had poor prognosis and treatment outcomes. Despite high-dose corticosteroids and surgery/chemoembolization for HCC management,5–8 few patients had improvement of muscle weakness and all died from HCC-related causes within 6 months after diagnosis.

Disclosures

Author contributions: K. Thanapirom wrote the first draft, collected the data, and conducted the literature research. S. Aniwan conducted the literature research and drafted the article. S. Treeprasertsuk reviewed the final draft and is the article guarantor.

Financial disclosure: No conflicts of interest or sources of funding to report.

Informed consent was obtained for this case report.

Received: January 4, 2014; Accepted: March 18, 2014

References

Ceftriaxone-Induced Gallstones: Case Report and Literature Review

Aditi Nayak, MD, and Adam Slivka, MD, PhD

Division of Gastroenterology, Hepatology and Nutrition, University of Pittsburgh Medical Center, Pittsburgh, PA

Abstract

We report a case of gallbladder and common bile duct stones occurring in a 14-year-old male who was exposed to ceftriaxone for 6 weeks. Ceftriaxone-induced gallstones are under-reported and remain an important cause of gallstones in patients exposed to this antibiotic. Gallstone development should be considered in the appropriate clinical context.

Introduction

Gallstones are most commonly seen in middle-aged multiparous women who are obese. Cholesterol-containing stones are most common. Liver disease and hemolysis are risk factors for black pigment stones, and biliary infections can cause mixed pigment stones. Gallstones in children are relatively uncommon. We report a case of a young male with antibiotic-induced gallstones after exposure to ceftriaxone for 6 weeks.

Case Report

A 14-year-old male sustained a right anterior cruciate ligament (ACL) tear during a soccer match and underwent a partial physeal-sparing anatomic ACL reconstruction using his own hamstrings. Two weeks later, he presented with an infected joint and underwent incision and drainage with hardware removal. Cultures grew Serratia marcescens; the patient had a peripherally inserted central catheter (PICC) line placed and was discharged on 6 weeks of ceftriaxone 2 gm IV daily.

During the last week of his ceftriaxone therapy, he developed intermittent right upper quadrant (RUQ) pain without nausea. His third attack in 72 hours prompted an emergency room visit. On exam, he was fit and thin with a non-toxic appearance, without fever, tachycardia, or hypotension. He was anicteric and had RUQ tenderness. The remainder of his exam was normal. He had no history of liver disease or hemolysis. There was no family or personal history of gallstones.

His laboratory tests showed a normal hemogram without leukocytosis. His electrolytes, blood urea nitrogen (BUN), and creatinine were normal. His total bilirubin was 1.2 mg/dL, alanine transaminase (ALT) 270 IU/L, and aspartate transaminase (AST) 463 IU/L. His amylase and lipase were normal. CT scan showed several densely calcified gallstones in the gallbladder (Figure 1) and 1 stone in the distal common bile duct (Figure 2) without evidence of acute cholecystitis. He was admitted to the hospital and underwent an ERCP with biliary sphincterotomy and extraction of yellow-green stone material (Figure 3), which did not appear like typical cholesterol, mixed pigment, or black pigment stones. He had no anatomic biliary abnormalities. He did well and underwent uneventful laparoscopic cholecystectomy the next day.
Discussion

The radiologic and phenotypic appearance of this patient's stones and the timing of prolonged ceftriaxone therapy—along with absence of any risk factors for cholesterol, mixed pigment, or black pigment stones—strongly suggest that his stones were secondary to the antibiotic treatment. Ceftriaxone has been associated with the formation of biliary precipitates, which Park et al identified as the calcium salt of ceftriaxone in 4 gallbladder specimens using thin-layer chromatography, high-performance liquid chromatography (HPLC), and electron microprobe analysis. The high levels of calcium in ceftriaxone leads to high density on imaging in contrast to typical cholesterol stones. Forty percent of the drug is excreted into the bile, where concentration and dehydration can occur in the gallbladder and lead to precipitation. In animal models, ceftriaxone may inhibit gallbladder contractility, further favoring the formation of precipitates. The incidence of precipitates may be up to 46% and appears to be higher in the pediatric population. Although there are no time studies or dose–response relationship studies, biliary precipitates can occur after relatively short exposures to the drug (weeks) and can be asymptomatic or lead to symptoms including biliary colic, cholecystitis, pancreatitis, and cholangitis. Spontaneous resolution has been reported.

Ceftriaxone-induced gallstones are an uncommon but recognized complication of therapy with this antibiotic. Patients should be counseled regarding the possibility of this complication and informed to recognize and report biliary-type symptoms early during the course of treatment. Doctors need to be vigilant for these symptoms in patients on prolonged therapy.

Disclosures

Author contributions: A. Nayak prepared the manuscript. A. Slivka supervised the clinical care of this patient and the creation of this manuscript, and is the article guarantor.
Financial disclosure: Neither author has a conflict of interest related to this report.

Informed consent was obtained for this case report.

Received: January 16, 2014; Accepted: March 4, 2014

References