Trichobezoar Without Trichotillomania—a Case Report

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J Gen Intern Med 37(4):962–5 DOI: 10.1007/s11606-021-07194-6 © This is a U.S. government work and not under copyright protection in the U.S.; foreign copyright protection may apply 2021

INTRODUCTION

Since the advent of H2 blockers and proton pump inhibitors, the most common cause of gastric outlet obstruction has shifted from peptic ulcer disease to malignancy.[1] Bezoar (a mass of indigestible material that forms in the gastrointestinal tract) is a rare cause of gastric outlet obstruction in adults. Phytobezoars (vegetable matter) are the most common type of bezoars in adults (40%), followed by trichobezoars (20%). [2] Trichobezoars are generally found in patients with psychiatric disorders, most commonly trichotillomania (the urge to pull one's hair) and trichophagia (the urge to eat one's hair). [3] However, trichobezoars occur in only 1% of patients with these psychiatric disorders. [4] Trichobezoar without trichotillomania has been reported in girls and women, [5, 6] but to our knowledge has not been reported in an adult man. We present a man without trichotillomania who was found to have gastric outlet obstruction caused by a trichobezoar. In this patient, the diagnosis of trichobezoar was initially delayed and therefore, we examine the contribution of cognitive biases to this diagnostic delay.

CLINICAL PRESENTATION

A 32-year-old man presented to the emergency department with complaints of intractable nausea, vomiting, and chest pain. Post-prandial nausea began 6 days prior to presentation followed by non-bilious, non-bloody vomiting beginning 3 days prior to presentation. These symptoms were worse with solid compared to liquid foods. He reported epigastric pain that improved after emesis and early satiety with frequent eructation. He had not had a bowel movement for several days, denied diarrhea, and was able to pass flatus. His chest pain was described as a churning, stinging pain located in the mid sternum with radiation to the epigastrium that began 3 months prior to presentation. The pain was non-exertional

Prior Presentations: None. Received April 13, 2021 Accepted October 1, 2021 Published online January 3, 2022 and sometimes associated with food ingestion. He denied changes in diet, weight loss, melena, anorexia, dysphagia, odynophagia, fever, personal or family history of peptic ulcers, or shortness of breath.

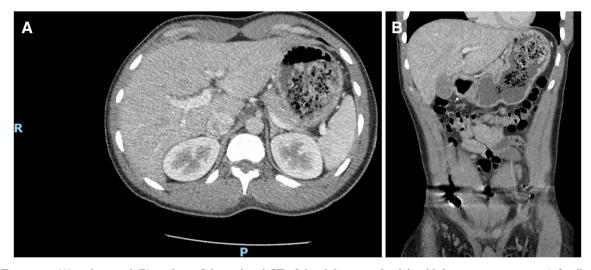
Medical history included treated chlamydia infection 3 years prior, several sprained wrists related to trauma from assaults, post-traumatic stress disorder, and a recent diagnosis of personality disorder with cluster A (paranoid) and B (borderline) traits. He did not have a history of *Helicobacter pylori* infection or gastroesophageal reflux and had no prior surgeries. Family history included diabetes in his parents. It was reported in the electronic health record that he had paranoia and frequent physical outbursts while recently incarcerated and was therefore started on olanzapine. He denied alcohol use in the 2 weeks prior to admission but previously consumed up to six beers a day for the past 4 years. He used marijuana infrequently over the past several years and denied other illicit drug use.

The patient was healthy appearing with normal vital signs. He was wearing a hospital gown and a beanie. He had a regular cardiac, pulmonary, and neurological exam. Abdominal exam revealed no tenderness to palpation without masses and normal bowel sounds. He had a paranoid effect, frequently asking, "Why do you need to know that?" in regards to questions. He endorsed a belief that he was poisoned in prison more than a month ago causing his vomiting and chest pain. He denied suicidal or homicidal ideation.

A basic metabolic profile and complete blood count were unremarkable. His liver function tests revealed an aspartate transaminase level of 136 U/L (normal 0–50 U/L) and alanine transaminase 52 U/L (normal 0–39 U/L), with unremarkable albumin, alkaline phosphatase, and bilirubin levels. The lipase was 69 U/L (normal 13–60 U/L). Alcohol level was undetectable. Urine toxicology screen was positive for cannabinoids. A D-dimer was 0.66 ug/mL (normal 0.00–0.50 ug/mL). Hemoglobin A1C was normal. Troponin level was undetectable. An EKG performed revealed normal sinus rhythm without evidence of ischemia. Chest x-ray was unremarkable. CT angiogram of the chest revealed no filling defects in the pulmonary arteries or esophageal abnormalities.

He was admitted to the hospital from the emergency department after an oral challenge with antacids, oral lidocaine, and water resulted in forceful vomiting. The initial working diagnosis from the emergency department was alcoholic pancreatitis given his abdominal pain and alcohol use history. On hospital day 2, a CT of the abdomen and pelvis with





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Fig. 1 Transverse (A) and coronal (B) sections of the patients' CT of the abdomen and pelvis with intravenous contrast. A focally dilated duodenum and a proximal jejunum with a mottled appearance of the non-enhancing gastric contents were present. No inflammatory changes of the pancreas were present

intravenous contrast revealed a focally dilated duodenum, proximal jejunum, and stomach without abnormalities to the pancreas (Fig. 1). The study was interpreted by a teleradiologist overnight, with whom the team did not speak. The written report noted that the findings were likely due to peristalsis and a mechanical obstruction was felt to be unlikely with clinical correlation warranted. The patient declined nasogastric tube placement. Despite a normal lipase, normal findings of the pancreas on CT, and an AST:ALT ratio not > 2:1, the team's leading diagnosis persisted as alcoholic pancreatitis. The differential diagnosis for gastric outlet obstruction was discussed (including trichobezoar, which was initially dismissed due to lack of signs of trichotillomania and the patient being male), and ultimately more common diagnoses were focused on compared to rare diagnoses.

On hospital day 3, the patient had forceful vomiting after a trial of a clear liquid diet. Nasogastric tube was offered and declined. After he was asked, "Do you feel you are improving?" he replied with, "No, you do know I ate my own hair a week ago, right?" He endorsed shaving and ingesting all of his hair over several days prior to his admission. The patient stated his goal by eating the hair was to have it grow back better. He denied neurovegetative symptoms and excessive anxiety or worry. Examination of his scalp revealed a recently shaved head without signs of patchy alopecia. He noted he had done the same 2 years prior without associated gastrointestinal symptoms at that time. Otherwise, he denied ingesting or pulling his own hair. Further review of his imaging with radiology after obtaining this information confirmed a nonenhancing mottled appearance to the gastric contents, consistent with a trichobezoar.

An esophagogastroduodenoscopy (EGD) was performed on hospital day 4, revealing a large trichobezoar occupying a majority of the stomach extending into the duodenal bulb (Fig. 2). Endoscopic removal of the trichobezoar was attempted, but the mass was too large to be pulled across the gastroesophageal junction. Only 30% of the trichobezoar was fragmented and removed using a combination of endoscopic devices. General surgery evaluated the patient and opted for conservative management to see if partial fragmentation

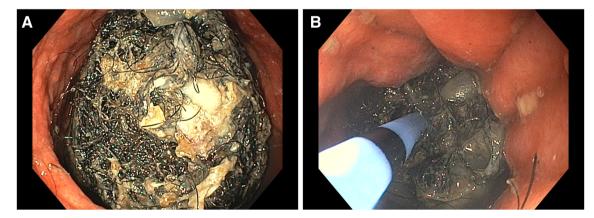


Fig. 2 A Endoscopic view of the large trichobezoar mixed with food occupying the stomach. B Attempt of fragmentation using argon plasma coagulation on the trichobezoar

would allow the remaining trichobezoar to pass through the gastrointestinal system. Over the next several days, the patient's diet was advanced and he began to have small bowel movements with hair contents visible. His presenting symptoms resolved. He declined an inpatient psychiatry evaluation but was amenable to outpatient visits with psychiatry and a substance abuse counselor. He endorsed an understanding that his trichophagia led to his hospitalization and stated he would not ingest his hair again because of his symptoms and the hospitalization that resulted from it. As of 9 months after hospital discharge, the patient was lost to follow-up.

DISCUSSION

This patient presented with gastric outlet obstruction secondary to a trichobezoar from trichophagia in the absence of trichotillomania. To our knowledge, a diagnosis of trichobezoar from trichophagia, specifically in the absence of trichotillomania, has not been reported in an adult man. In addition to an opportunity to review, in brief, the management of bezoars in adults, this case also provides important lessons on the recognition of cognitive biases.

Treatment options for gastric bezoar include dissolution, endoscopic, and surgical interventions. Dissolution has been described using Coca-Cola, cellulase, papain, and other products. [7, 8] Trichobezoars, unlike phytobezoars, are generally resistant to dissolution treatments and typically require endoscopic or surgical treatment. [9] Endoscopic fragmentation and removal of trichobezoars is frequently attempted but is time-consuming and often ineffective. [10, 11] Fragmentation has been associated with migration of portions of the hair mass distally leading to bowel obstruction. [9] A review of pediatric patients with trichobezoars revealed 5% were successfully managed with endoscopy alone, highlighting the frequent need for surgical intervention. [9] Indications for surgery via laparotomy or laparoscopy [12, 13] are not well-defined for adults, and management should be discussed with an interdisciplinary team of gastroenterologists and surgeons. Treatment of trichophagia and trichotillomania is centered around bodyfocused repetitive behavior reversal using cognitive behavioral therapy. [14, 15] While this was the patient's second time ingesting his hair, given his intact insight and decision-making capacity, consideration for inpatient psychiatric admission was not felt to be appropriate. Follow-up in the clinic was scheduled.

In this case, diagnosis was delayed for several days even after CT imaging of the abdomen was performed. Several forms of cognitive bias may have contributed to this delay including anchoring, confirmation, blind obedience, frequency gambling, and gender bias. Anchoring bias (the most common cognitive bias identified, occurring in 5.9–87.8% of cases) [16, 17] occurs when individuals rely on their initial diagnostic impression despite subsequent information to the contrary. This patient's recorded history of alcohol use disorder in the presence of a slightly elevated lipase and AST:ALT ratio of greater than 2:1 led to an initial differential diagnosis of alcoholic or gallstone-related pancreatitis, gastritis, or a combination of the two. The leading diagnosis was alcoholic pancreatitis from the emergency department's signout to the Medicine team. Despite an illness script that was not consistent for alcoholic pancreatitis (the patient did not consume alcohol in the 2 weeks prior to admission, reported non-classical epigastric pain, and had a lipase that was not three times the upper limit of normal without a CT), the subsequent workup and management over the next 2 days was focused on alcoholic pancreatitis. The delays were further compounded by confirmation bias (the tendency to look for confirmatory evidence while disregarding invalidating evidence) as the CT for abdomen and pelvis was ordered to further evaluate for evidence of pancreatitis rather than for causes of gastric outlet obstruction. Further complicating the diagnosis was a CT dictation that was nonspecific for a bezoar and gastric outlet obstruction, to which blind obedience bias (reliance on an authoritative source on the presence or absence of information) [18] occurred. While CT is the imaging of choice for bezoars, its diagnostic accuracy is 73-95% and can be less accurate early in the formation of a bezoar. [19] The classic findings of a trichobezoar on imaging include a dilated stomach with a mottled appearance of the mass material, [20] which was not reported on initial dictation. After discussion with radiology in concordance with the patient's clinical symptoms and additional history, it was felt the findings on the CT did support a finding of a bezoar. This stresses the importance of clinical correlation when reading radiology reports. Frequency gambling (the tendency in equivocal or ambiguous presentations to opt for a benign diagnosis on the basis it is more likely than a rare one) and gender bias (the belief that gender is a determining factor in the probability of diagnosis of disease when no such pathophysiological basis exists) [21] may have influenced the medical decision-making of this patient. When the differential diagnosis of gastric outlet obstruction included a trichobezoar, the treatment team dismissed this in favor of more common diagnoses given the rarity of trichobezoars in adults, men, and individuals without signs of trichotillomania. Gender bias has been noted to cause differences in diagnostic timing and treatment modalities with well-researched diseases including coronary artery disease, [22] Parkinson's disease, [23] irritable bowel syndrome, [24] neck pain, [25] tuberculosis, [26] and joint arthrosis [27, 28]. While review of 152 trichobezoar cases (including cases with and without trichotillomania) revealed 91.4% of cases were in females [29], dismissing trichobezoar as a diagnosis based on the patient's gender alone was ultimately a large source of cognitive bias causing delayed diagnosis in this patient.

This case demonstrates the importance of considering the diagnosis of a trichobezoar in adult men who present with symptoms of gastric outlet obstruction and a history of psychiatric illness. Careful attention to avoidance of cognitive biases is imperative to reach a timely diagnosis. Overreliance on radiology reports without consideration of clinical correlation can compound other forms of bias. Endoscopic or surgical treatment are frequently required for trichobezoars, as dissolution therapy is rarely effective. Psychiatric intervention with cognitive behavioral therapy is warranted to prevent further episodes.

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Author contributions None.

Declarations:

Conflict of Interest: The authors declare that they do not have a conflict of interest.

REFERENCES

- Shone DN, Nikoomanesh P, Smith-Meek MM, Bender JS. Malignancy is the most common cause of gastric outlet obstruction in the era of H2 blockers. Am J Gastroenterol. 1995 Oct;90(10):1769-70. PMID: 7572891.
- Eng K, Kay M. Gastrointestinal bezoars: history and current treatment paradigms. Gastroenterol Hepatol (N Y). 2012;8(11):776-778.
- Gorter RR, Kneepkens CM, Mattens EC, Aronson DC, Heij HA. Management of trichobezoar: case report and literature review. Pediatr Surg Int 2010;26(5):457-463.
- Gonuguntla V, Joshi DD. Rapunzel syndrome: a comprehensive review of an unusual case of trichobezoar. Clin Med Res 2009;7(3):99–102.
- Jain A, Agrawal A, Tripathi AK, Bansod RK, Jain G, Yadav KS. Trichobezoar without a clear manifestation of trichotillomania. *J Family Med Prim Care.* 2020;9(5):2566-2568. Published 2020 May 31. doi:https://doi.org/10.4103/jfmpc.jfmpc_11_20.
- Mehra A, Avasthi A, Gupta V, Grover S. Trichophagia along with trichobezoar in the absence of trichotillomania. J Neurosci Rural Pract. 2014;5(Suppl 1):S55-S57. doi:https://doi.org/10.4103/0976-3147. 145204.
- Ladas SD, Damberoglou D, Karamanolis G, Vlachogiannakos J, Zouboulis-Vafiadis I. Systematic review: Coca-Cola can effectively dissolve gastric phytobezoars as a first-line treatment (2013). *Alim Pharm Ther* 37:169-173.
- Khan S, Jian K, Zhu L, et al. Upper gastrointestinal manifestation of bezoars and the etiological factors: a literature review (2019). Gastro Research and Practice 2019:1-13.
- Gorter RR, Kneepkens CMF, Mattens ECJK, Aronson DC, Heij HA. Management of trichobezoar: case report and literature review (2010). Pediatr Surg Int 26:457-463
- Hall JD, Shami VM. Rapunzel's syndrome: gastric bezoars and endoscopic management (2006). Gastrointest Endoscopy Clin N Am 16:111-119
- 11 Iwamuro M, Okada H, Matsudea K, et al. Review of the diagnosis and management of gastrointestinal bezoars (2015). World J Gastrointest Endos 7(4): 336-345.
- Javed A, Agarwal AK. A modified minimally invasive technique for the surgical management of large trichobezoars. J Minim Access Surg. 2013;9(1):42-44. doi:https://doi.org/10.4103/0972-9941.107142.
- Nirasawa Y, Mori T, Ito Y, Tanak H, Seki N, Atomi Y. Laparoscopic removal of a large gastric trichobezoar. J Pediatr Surg. 1998;33:663–665. doi: https://doi.org/10.1016/S0022-3468(98)90342-6.

- Grant JE, Odlaug BL. Clinical characteristics of trichotillomania with trichophagia. *Compr Psychiatry*, 2008;49(6):579-584. doi:https://doi. org/10.1016/j.comppsych.2008.05.002.
- Reddy YCJ, Sudhir PM, Manjula M, Arumugham SS, Narayanaswamy JC. Clinical Practice Guidelines for Cognitive-Behavioral Therapies in Anxiety Disorders and Obsessive-Compulsive and Related Disorders. *Indian J Psychiatry*. 2020;62(Suppl 2):S230-S250. doi:https://doi.org/10. 4103/psychiatry.IndianJPsychiatry_773_19.
- Saposnik, G., Redelmeier, D., Ruff, C.C. *et al.* Cognitive biases associated with medical decisions: a systematic review. *BMC Med Inform Decis Mak* 16, 138 (2016). https://doi.org/https://doi.org/10.1186/s12911-016-0377-1. Accessed March 4, 2021.
- Kassirer JP, Kopelman RI. Cognitive errors in diagnosis: instantiation, classification, and consequences. Am J Med. 1989 Apr;86(4):433-41. doi: https://doi.org/10.1016/0002-9343(89)90342-2. PMID: 2648823.
- Wellberry C. Flaws in Clinical Reasoning: A Cause of Diagnostic Error. AAFP. 2011 Nov 1;84(9):1042-1048.
- Ho TW, Koh DC. Small-bowel obstruction secondary to bezoar impaction: a diagnostic dilemma. World J Surg 2007;31:1072–8.
- Newman B, Girdany BR. Gastric trichobezoars-sonographic and computed tomographic appearance. Pediatr Radiol. 1990;20(7):526-7. doi: https://doi.org/10.1007/BF02011382. PMID: 2216587.
- 21 Croskerry, Pat MD, PhD The Importance of Cognitive Errors in Diagnosis and Strategies to Minimize Them, Academic Medicine: August 2003 -Volume 78 - Issue 8 - p 775-780
- 22 Daly C, Clemens F, Lopez Sendon JL, et al. Euro Heart Survey Investigators. Gender differences in the management and clinical outcome of stable angina. Circulation. 2006 Jan 31;113(4):490-8. doi: https://doi. org/10.1161/CIRCULATIONAHA.105.561647. PMID: 16449728.
- 23. Gun-Marie Hariz, Takeshi Nakajima, Patricia Limousin, et al (2011) Gender distribution of patients with Parkinson's disease treated with subthalamic deep brain stimulation; a review of the 2000-2009 literature, Parkinsonism & Related Disorders, Volume 17, Issue 3. https://doi. org/10.1016/j.parkreldis.2010.12.002
- Hamberg K, Risberg G, Johansson EE. Male and female physicians show different patterns of gender bias: A paper-case study of management of irritable bowel syndrome. Scandinavian Journal of Public Health. 2004;32(2):144-152. doi:https://doi.org/10.1080/ 14034940310015401.
- Hamberg K, Risberg G, Johansson EE, Westman G. Gender bias in physicians' management of neck pain: a study of the answers in a Swedish national examination. J Womens Health Gend Based Med. 2002 Sep;11(7):653-66. doi: https://doi.org/10.1089/152460902760360595. PMID: 12396897.
- 26 Fazlul Karim, Md. Akramul Islam, AMR Chowdhury, Eva Johansson, Vinod K Diwan. Gender differences in delays in diagnosis and treatment of tuberculosis. *Health Policy and Planning*. Volume 22, Issue 5, September 2007, Pages 329–334. doi.org/https://doi.org/10.1093/heapol/czm026.
- Katz JN, Wright EA, Guadagnoli E, Liang MH, Karlson EW, Cleary PD. Differences between men and women undergoing major orthopedic surgery for degenerative arthritis. Arthritis Rheum. 1994 May;37(5):687-94. doi: https://doi.org/10.1002/art.1780370512. PMID: 8185695.
- Hamberg K. Gender Bias in Medicine. Women's Health. May 2008:237– 243. https://doi.org/10.2217/17455057.4.3.237.
- Debakey M, Ochsner A. Bezoars and concretions: A comprehensive review of the literature with an analysis of 303 collected cases and a presentation of 8 additional cases. Surgery. Volume 5, Issue 1. Pages 132-160. January 1939. doi: https://doi.org/10.5555/ uri:pii:S0039606039904289.

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