Case Report

Recurrent Gastric Trichobezoars - A Case Report and Literature Review

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A trichobezoar is a hairball usually confined to the stomach of almost exclusively young females, and typically results from the patient pulling out her hair (trichotillomania) and swallowing it (trichophagia). We describe a case of recurrent gastric trichobezoars in a young female with trichophagia who presented at the ages of 18, 21, and 25 years old with similar complaints of epigastric discomfort, early satiety, loss of appetite, intermittent nausea, and very foul smelling burps. At each presentation, endoscopy revealed a gastric trichobezoar and endoscopic fragmentation efforts were unsuccessful. The first two trichobezoars were removed via laparoscopic gastrotomy. The most recent and largest of the three trichobezoars required laparotomy secondary to dense adhesions. The patient and her parents persistently refused psychiatric consultation and outpatient follow-up which is an integral aspect of preventing trichobezoar recurrence.
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INTRODUCTION

A bezoar is a buildup of undigested exogenous material that coalesces in the gastrointestinal tract of ruminant animals and humans. The etymology of the term "bezoar" is from the Arabic bazahr or Persian padzahr which literally mean "counterpoison" or "antidote".[1,2,6] Historically, they were thought to have magical properties and were used medicinally as early as 1000 BC in Western Persia.[2,3] Bezoars eventually made their way to Europe from the Middle East in the 11th century and were utilized as treatment for a diverse group of maladies: plague, dysentery, epilepsy, vertigo, arsenic poisoning, venomous bites, etc.[3] Interestingly, they were often adorned with gold and ornately decorated which reflects their cultural and perceived medicinal value of the time and gave rise to the term "bezoar stone".[3] In modern times, the mystical properties and medicinal value of bezoars have largely been replaced by their recognition as a potentially serious medical problem.

Bezoars are generally classified into four types based on their composition: phytobezoars, lactobezoars, pharmacobezoars, and trichobezoars.[3,5] Phytobezoars are the most common type in modern times and are comprised of indigestible food particles such as cellulose, hemicellulose, and lignin found in fruit and vegetables.[3] Lactobezoars are composed of undigested milk curds and are traditionally associated with pre-term infants fed with highly concentrated formula, although the etiology is likely multifactorial as they also occur in patients who consume breast milk, commercial infant formula, and cow's milk.[3,4] Pharmacobezoars are comprised of pills or capsules and are often associated with extended release products secondary to the indigestible, semi-permeable tablet coating composed of cellulose acetate.[3] Trichobezoars are generally comprised of hair and occur most frequently in young females with trichotillomania and/or trichophagia.[3,5,6] Other types of bezoars have also been described: trichophytobezoars (composed of hair, fruit, and vegetable fibers), diospyrobezoars (unripe persimmons), worm bezoars (dead ascaris), a toilet paper bezoar,[7] and more.[3]

Trichobezoars are so infrequent their true incidence is unknown. Trichotillomania affects approximately 1% of the population and about one third of these patients develop trichophagia.[8] Of patients with trichophagia, somewhere between 1% and 37% form a clinically evident trichobezoar.[8,10] Recurrent trichobezoars are rarer with less than fifteen cases reported in the English literature. The reported cases of recurrence are generally similar and occur in young females with trichotillomania and refusal or poor psychiatric follow-up. Here, we report an analogous case of recurrent gastric trichobezoars and highlight the salient clinical features and management.

CASE REPORT

An 18-year-old white female with a history of mild spastic cerebral palsy and normal IQ presented in May 2007 with a 2-month history of epigastric discomfort, early satiety, loss of

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appetite, weight loss of 7 pounds, intermittent nausea, and very foul smelling burps. A firm, nontender mass was palpated in the epigastrium. Physical exam was otherwise unremarkable with no evident alopecia or scalp scaling. A complete blood count, basic metabolic panel, liver function tests, and lipase were within normal limits. A CT scan revealed a large bezoar present in the stomach. Subsequent endoscopy showed a 15 x 5 cm gastric trichobezoar. Numerous attempts to fragment the bezoar via endoscopic snare cautery were unsuccessful. Given the size of the bezoar and progressive worsening of symptoms over the previous couple of months, a laparoscopic gastrotomy was utilized for removal. Upon further clinical examination, the patient admitted to chewing her hair (trichophagia) since middle school. Psychiatric follow-up was recommended but refused by the patient and her parents. The patient had an uneventful recovery and was discharged home.

After four years of being asymptomatic, the patient was again admitted in January 2011 with epigastric discomfort, abdominal pain, nausea, and foul smelling burps. An upper GI series identified the presence of a large bezoar. Subsequent endoscopy showed an 11 x 5 cm gastric trichobezoar with no evidence of pyloric obstruction. Multiple attempts to fragment the bezoar via endoscopic water jet and snare cautery were unsuccessful and a laparoscopic gastrotomy was performed for removal. Discussion with the patient and her parents with regard to psychiatric follow-up were again unsuccessful; they were, however, agreeable to interval follow-up endoscopic evaluations every 3-6 months to establish the continuity of her behavior and remove smaller trichobezoars before they resulted in more serious clinical sequelae. An additional recommendation to cut her hair short to minimize the available hair for swallowing and theoretically the size of the resulting trichobezoar was offered. An esophagogastroduodenoscopy in June 2011 showed a well healed gastrotomy scar, no ulcerations or erosions, and no evidence of foreign body or trichobezoar in the gastric corpus. A stomach biopsy at that time showed a fundic gland polyp.

After being lost to follow-up for three years and now 25 years old, the patient was admitted in March 2014 with similar symptoms and physical exam as previous admissions. Upper GI endoscopy showed a large trichobezoar filling the entire stomach and a single shallow ulcer on the anterior wall of the greater curvature (**Figure 1**). The distal esophagus showed concentric rings and longitudinal furrows "consistent with eosinophilic esophagitis". An esophageal biopsy showed changes compatible with eosinophilic esophagitis but noted the necessity of clinical correlation (**Figure 2**). Numerous attempts to fragment the bezoar via endoscopic snare cautery in April were unsuccessful. A laparoscopic gastrotomy was attempted but ultimately aborted secondary to dense adhesions. An uncomplicated open gastrotomy was performed resulting in extraction of a 22 x 5 x 6 cm trichobezoar with two smaller trichobezoars measuring 1.9 x 1.5 x 1.2 cm and 1.1 x 1 x 0.9 cm also found in the gastric body (**Figure 3**). The importance of follow-up psychiatric care was again emphasized but openly declined by the patient. She had an uneventful recovery and was discharged home on postoperative day 8 with prescription for a proton pump inhibitor. No additional endoscopies or recurrence of symptoms are documented in the eight months of recorded follow-up.

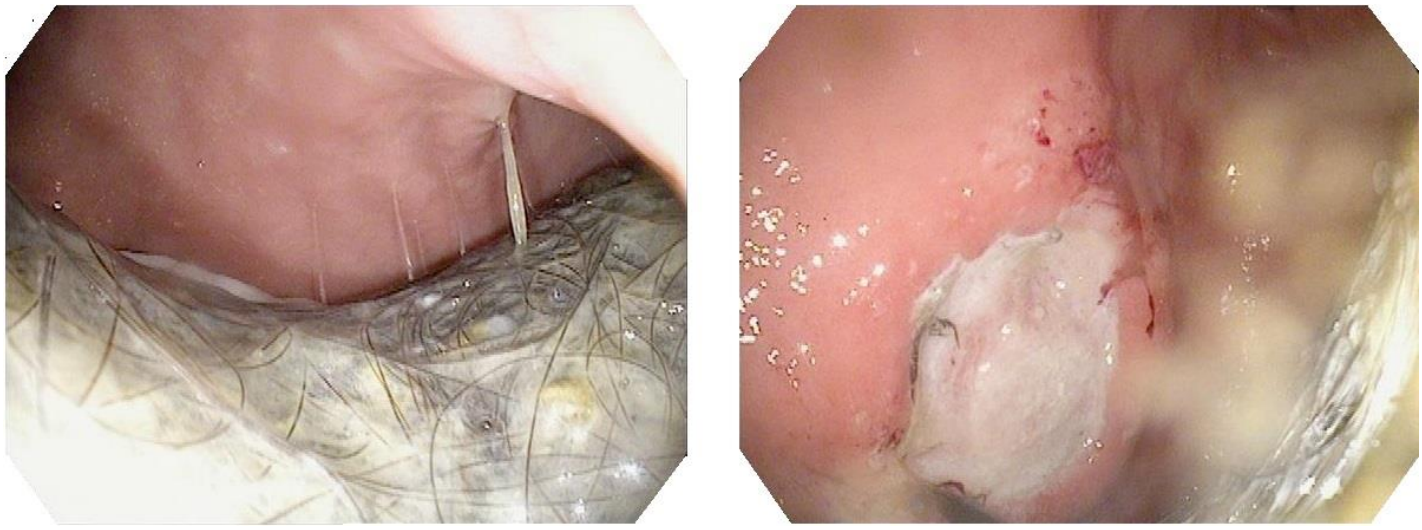

Figure 1. Endoscopic images showing **(a)** trichobezoar in the gastric body and antrum and **(b)** gastric mucosa with a shallow ulcer on the anterior wall of the greater curvature.

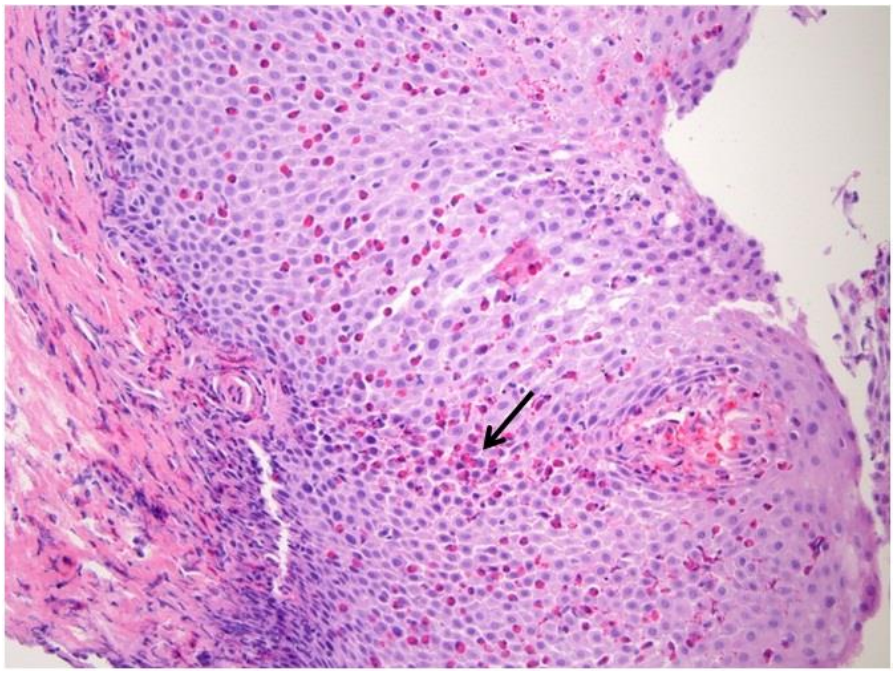

Figure 2. Esophageal biopsy showing diffusely increased intraepithelial eosinophils with eosinophilic microabscesses (arrow).

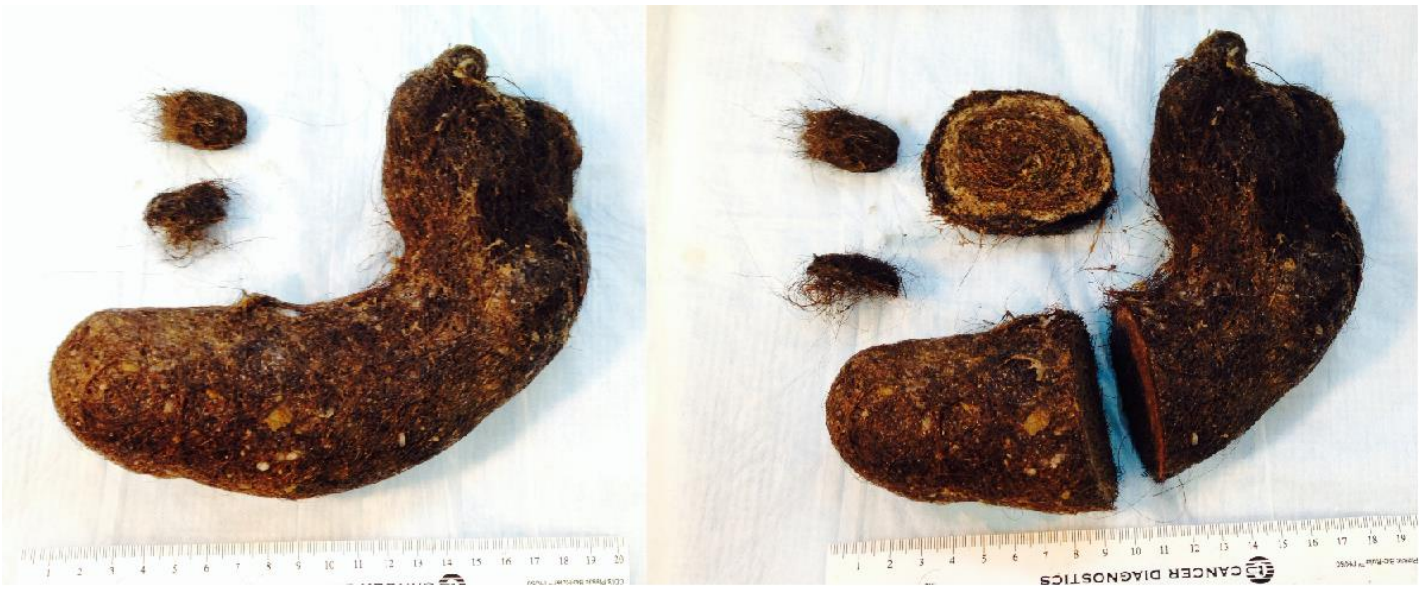

Figure 3. Extracted trichobezoar before **(a)** and after **(b)** sectioning showing the characteristic black hair intermixed with mucus and decaying food particles.

DISCUSSION

Trichobezoars, colloquially referred to as "hairballs", generally result from ingesting large quantities of hair but may also result from ingestion of carpet fibers, rope, string, or clothing.[5] The first reported case of a trichobezoar in humans was described in a 1779 autopsy of a 16-year-old male patient who had been eating hair since infancy and developed gastric perforation and peritonitis.[11] Although gastric acid denatures hair proteins resulting in a characteristic black color, the keratinous constitution of hair makes it poorly digestible.[3,7,11] Hair is ineffectively moved by peristalsis secondary to its smooth surface.[7,9] Consequently, the hairs coalesce, trap undigested fat and mucus, and can reach sufficient sizes to cause stomach distention and inhibit gastric emptying.[3,5,11] While they are usually confined to the stomach, they rarely have an attached tail extending through the pylorus into the small bowel which is known as "Rapunzel Syndrome".[6,9] Occasionally, pieces of the tail can detach and wander through the pylorus resulting in intestinal obstruction.[9]

Trichobezoars occur more frequently in children and young adult females with 80% of cases under 30 years of age.[3,5,6] Most patients have an underlying psychiatric disorder, the most common of which is the urge to pull out one's own hair (trichotillomania) and/or swallow it (trichophagia).[5,6,9,10] Other factors associated with trichobezoar formation include pica, mental retardation, motility disorders, abuse, obsessive compulsive disorder, depression, and anorexia nervosa.[5,8,9] Cases of bezoars have been described in patients with cerebral palsy and concomitant neurologic disability.[15] Patients may be asymptomatic for many years and early symptoms may be vague and indistinguishable from other gastrointestinal disorders: discomfort, abdominal bloating, nausea/vomiting, anorexia, weight loss, and halitosis.[3,6,9,11] The decomposition of food particles and fermentation of fats trapped in the trichobezoar, as seen in this case, result in halitosis and putrid smelling eructation.[3,11] If not clinically recognized, they continue to increase in size and eventually result in obstruction. For larger trichobezoars, the gastric and/or small intestinal blood supply can become focally compromised and result in ulceration and perforation requiring open surgical intervention.[5-7,9,11] When bezoars are suspected clinically, especially in young females, a good history elucidating the presence of trichotillomania and/or trichophagia is integral to making a timely diagnosis.

Our case has endoscopic and histopathologic evidence of eosinophilic esophagitis (**Figure 2**). Eosinophilic esophagitis (EoE) generally has a male preponderance and is more common in adults.[12,13] The clinical presentation varies by age: younger children are more likely to present with nonspecific gastrointestinal symptoms, and older children and adults more commonly present with food impaction and dysphagia with a history of allergy (including atopic disease).[12] While the etiology remains unknown, the association with hypersensitivity makes allergy the most likely possibility.[12,13] The distinction of EoE from GERD is challenging and requires an integrative clinical and pathologic correlation. The endoscopic concentric rings and linear furrowing of the distal esophagus in this case suggests a diagnosis of EoE, but these findings are not specific and do not definitively differentiate EoE from GERD.[12] The diagnosis of EoE essentially requires the presence of an esophageal eosinophilic infiltrate on biopsy, normal pH monitoring, and persistence even with PPI therapy.[12,13] In the eight months of documented follow-up, however, there has been no documented pH monitoring and no additional notes since starting PPI therapy. Therefore, no clear distinction between the two entities can be made. The presence of a large trichobezoar occupying her entire stomach, however, would likely result in consumed food and beverage being propelled retrograde with peristaltic action. Thus, the case likely represents GERD with an unusually high eosinophilic component.

Gastric trichobezoars can be confirmed by a variety of imaging techniques: swirled configuration with or without calcification on plain films, outline of contrast medium on upper GI series, echogenic arc of air between the bezoar and gastric wall on ultrasound, and free-floating filling defect on CT with contrast.[5,6,11,14,15] The gold standard for diagnosis and differentiation from tumors and other entities is direct visualization via upper gastrointestinal endoscopy.[5,11] Effective therapy necessitates removal of the mass and close psychiatric follow-up to prevent relapse.[3-11]

Effective management techniques for removal depends largely on the size, location, and consistency of the trichobezoar.[9,11] As reported by Gorter et al,[9] about 95% of endoscopic removal attempts are unsuccessful due to large size, density and hardness. Additionally, endoscopic fragmentation and retrieval efforts increases the risk of esophagitis, pressure ulceration, perforation, and distal migration of satellites resulting in intestinal obstruction.[5,9] Surgical intervention via laparoscopy or laparotomy is usually required. Laparoscopy is generally successful for small to intermediate-size bezoars but is more technically challenging than conventional laparotomy: it requires a longer operative time and there is risk of spilling contaminated hair fragments into the abdominal cavity resulting in infection.[5,9,11] Laparotomy is less complex, has a low complication rate, allows for careful examination for satellites in less time, and is generally utilized in cases that have perforated or when the trichobezoar is too large to be managed safely via less invasive methods.[5,9] Other therapies demonstrating various efficacy have been described mostly in smaller trichobezoars: extracorporeal shock wave lithotripsy, enzymatic fragmentation, and prokinetic medications (metoclopramide, acetylcysteine).[5,9,11] Few recurrences have been reported following the initial removal of trichobezoars.[11]

Psychiatric consultation and follow-up are an integral component of management to prevent recurrence of trichobezoars.[3-11,14] Trichotillomania is classified as an impulse control or obsessive compulsive disorder and is characterized by a feeling of tension followed by relief after pulling out the hair.[10] Trichophagia has been linked to childhood neglect or abuse, mental retardation, bereavement, and psychiatric disorders.[11] The mainstay of management is habit reversal therapy, although no formal guidelines for treatment have been established.[6,8,10,11,14] A selective serotonin reuptake inhibitor or clomipramine may be added depending on the individual effectiveness of behavioral therapy or the presence of comorbid depression, anxiety, or obsessive compulsive disorder.[8,10,11,14] Parental counseling is typically included and is important in preventing relapses.[6] Our case exemplifies the importance of psychiatric follow-up and parental counseling as an integral aspect of long-term relapse prevention. The prognosis is excellent and the likelihood of recurrence is low if psychiatric follow-up is maintained.[8,10,11,14]

In summary, trichobezoars occur most frequently in young females and are closely associated with trichotillomania and/or trichophagia. The clinical presentation can be indistinguishable from other gastrointestinal disorders early in the course. Trichobezoars should always be considered in

young females complaining of chronic abdominal pain, nausea/vomiting, and halitosis especially if psychiatric comorbidities are present. A probing clinical history elucidating the presence of trichophagia is integral in making a timely diagnosis. If not clinically recognized, more serious sequelae such as obstruction, ulceration, and perforation can occur as the trichobezoar increases in size. Various imaging modalities can confirm the presence of a bezoar, but endoscopy is the gold standard for diagnosis. Retrieval can be accomplished via endoscopy, laparoscopy, or laparotomy depending largely on the size. Psychiatric follow-up is necessary to prevent recurrence.

CONFLICT OF INTEREST

The authors have no conflict of interest to disclose.

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