

Endoscopic retrieval of gastric trichophytobezoar

Case report of a 12-year-old girl with trichophagia

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Abstract

Rationale: Trichophytobezoars, which are composed of hair and plant fibers, are usually located in the stomach. They are often associated with trichophagia and trichotillomania. The most commonly reported methods of trichophytobezoar treatment are open surgery and laparoscopic retrieval; there are few reports of endoscopic removal of trichophytobezoars.

Patient concerns and Diagnoses: Twelve-year-old girl presented with a 3-day history of increasing upper abdominal pain, anorexia, and postprandial emesis. She had a 3-year history of pulling out and eating her own hair. Endoscopic examination showed a large intragastric trichophytobezoar measuring 10.5 cm × 3.5 cm in size, with extension of a few hairs through the pylorus.

Interventions and Outcomes: The trichophytobezoar was packed with hair fibers and contained a hard core of mixed hair and vegetable fibers. After the core was cut, the trichophytobezoar was fragmented into pieces with the alternating use of a polypectomy snare and argon plasma coagulation. A small amount of hair and nondigestible food fibers was removed with grasping forceps during the initial procedure. The remaining hairball was loosened with biopsy forceps and was injected with sodium bicarbonate solution. The trichophytobezoar was removed completely at repeat endoscopy 5 days later. After 6 months of psychological intervention, the patient had no recurrence of trichophagia or trichophytobezoar.

Lessons: Endoscopy with sodium bicarbonate injection is an effective and minimally invasive method of retrieving a gastric trichophytobezoar.

Abbreviation: APC = argon plasma coagulation.

Keywords: endoscopy, pediatrics, trichophagia, trichophytobezoar

1. Introduction

Trichobezoars, or hair bezoars, are a rare type of bezoar, usually located in the stomach but sometimes extending through the pylorus into the small bowel and even reaching the transverse colon.^[1] This condition is known as Rapunzel syndrome, which

can cause intestinal obstruction, gastrointestinal perforation, pancreatitis, and other complications.^[2–4] Trichophytobezoars, which are mixtures of hair and plant fibers, are rarely reported in the literature and can also cause the aforementioned complications.^[5] Trichophytobezoars and trichobezoars are usually caused by trichophagia, a compulsive behavior characterized by eating hair, usually one's own.^[6,7] When ingested, hair, fruit fibers, and vegetable fibers resist digestion. They also resist peristalsis because of their smooth surface. These substances accumulate between the mucosal folds of the stomach. Over time, the continuous ingestion of hair and plant fibers leads to their impaction with mucus and food, causing the formation of a trichophytobezoar.^[8]

Herein, we report a case of a large gastric trichophytobezoar associated with trichophagia in a 12-year-old girl. The trichophytobezoar was successfully retrieved endoscopically using snare polypectomy, argon plasma coagulation (APC), and sodium bicarbonate injection.

2. Case report

A 12-year-old girl presented to the pediatric emergency department with a 3-day history of increasing upper abdominal pain, anorexia, and postprandial emesis. In addition, she indicated that she had experienced early satiety for more than 6 months. Her mother reported the girl's habit of hair pulling and hair eating since the age of 9 years. There was no history of other gastrointestinal illness. Physical examination revealed upper abdominal tenderness, most notably with palpation of a mobile lump in the epigastrium, but no muscular guarding. The patient's white blood cell count was 9.3×10^9 cells/L, with 69.0% neutrophils. Her hemoglobin concentration was 141 g/L. A

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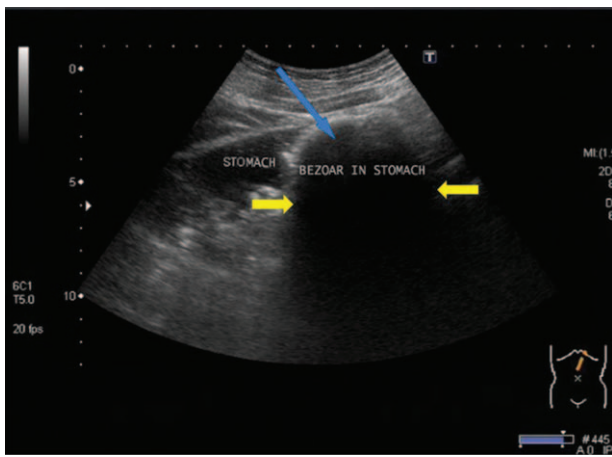


Figure 1. Gastric bezoar on ultrasonography. Gray-scale abdominal ultrasonography reveals an arc-shaped intraluminal mass (blue arrow) with a strong posterior acoustic shadow (yellow arrows).

peripheral blood smear revealed no abnormal blood cells. Abdominal ultrasonography revealed an arc-shaped, mobile, irregular, coherent, intraluminal mass with a strong acoustic shadow, 10.5 cm in its longest dimension, consistent with a gastric bezoar (Fig. 1). To distinguish gastric bezoar from undigested food mass, ultrasound examination was repeated after the patient drank 150 mL of warm water, with no change in the ultrasound findings.

Endoscopy was performed with a standard gastroscope (FUJINON EG-410HR/D, Fuji Photo Optical Co. Ltd, Saitama-ken, Japan) under general anesthesia. The endoscopic examination revealed a large intragastric trichophytobezoar approximately 10.5 cm \times 3.5 cm in size, with extension of a few

hairs through the pylorus (Fig. 2A). The trichophytobezoar was packed with hair fibers and had a hard core of mixed hair and vegetable fibers. After the hard core was cut with endoscopic scissors, the trichophytobezoar was fragmented into pieces with alternating use of a polypectomy snare and APC (ERBE VIO 300D/APC2, ERBE Elektromedizin GmbH, Tübingen, Germany; power setting: 40 W). Some of the hair and nondigestible food fibers were retrieved with grasping forceps (Fig. 2B). The remaining bezoar was loosened with biopsy forceps and was injected with 50 mL of 5% sodium bicarbonate solution through the biopsy channel. Routine fasting and intravenous fluid replacement therapy were instituted for 24 hours after endoscopic therapy, and a no-fiber diet was implemented.

A second endoscopic therapy session was conducted 5 days later. Gastroscopy revealed that the gastric trichophytobezoar was significantly looser than it had been previously (Fig. 2C). The hairball was cut with scissors and then fragmented into pieces with the alternating use of a polypectomy snare and APC, as in the initial procedure. The remaining trichophytobezoar was completely endoscopically retrieved through the pylorus in under an hour (Fig. 2D). Five days later, a third endoscopic examination revealed no remaining material in the stomach (Fig. 3). After this treatment the patient received 6 months of psychological intervention for the treatment of trichophagia. She did not exhibit any further hair-pulling or hair-eating behavior problems in 6 months of follow up. This child's clinical course is summarized in the Supplemental Time-line (see Supplemental Fig. 1, Supplemental Content, <http://links.lww.com/MD/B527>, which illustrates the clinical course of the child described in this case report).

3. Discussion

Bezoars are masses of indigestible or inedible exogenous material found in the gastrointestinal tract. According to their composi-

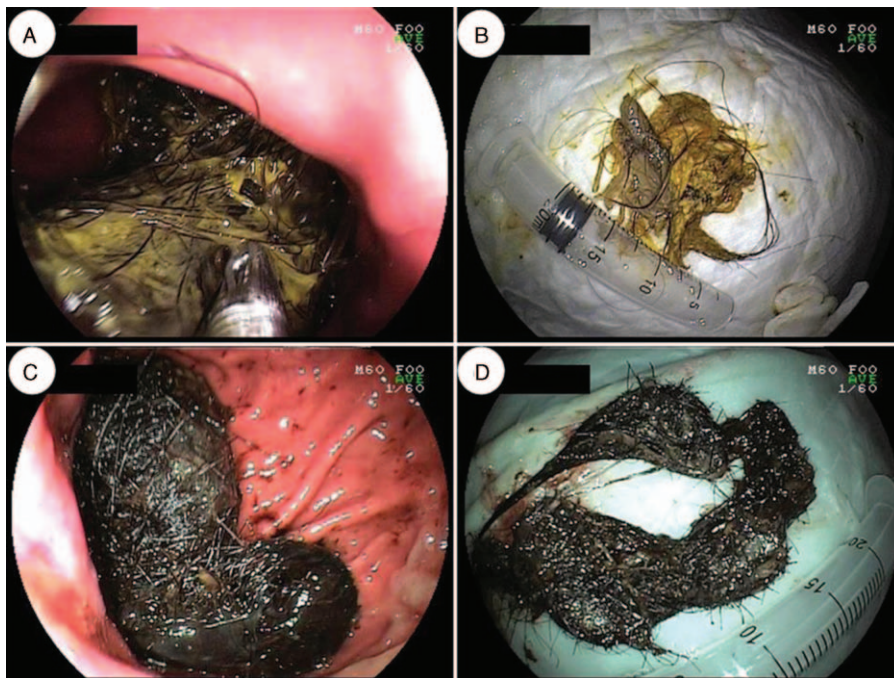


Figure 2. Endoscopic therapy. (A): Gastric trichophytobezoar at the first endoscopic treatment. (B) Some hair and nondigestible food fibers are retrieved during the initial endoscopy. (C) The gastric trichophytobezoar has loosened 5 days after initial treatment. (D) The trichophytobezoar is completely removed during the second endoscopy.

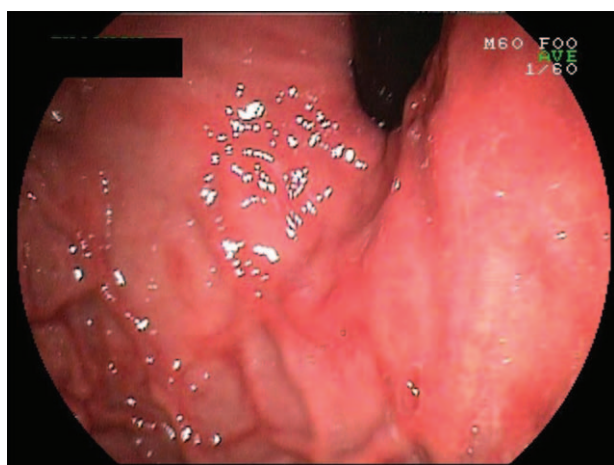


Figure 3. Nothing is present in the stomach at the third endoscopic examination.

tion, bezoars are classified as phytobezoars (composed of indigestible food particles found in vegetable or fruit fibers), lactobezoars (composed of undigested milk and mucus), pharmacobezoars (concretions of various medications), trichobezoars (composed of hair), worm bezoars (composed of dead ascaris), and others, such as those composed of a mixture of hair and plant fibers.^[9–11]

Among these classifications, phytobezoars are the most common type but trichophytobezoars are rarely reported.^[12] A small amount of hair can be excreted in the feces; trichophytobezoars and trichobezoars, unlike other types of bezoar, are not associated with alterations in gastrointestinal motility but are instead associated with underlying psychiatric disorders.^[13] Rapunzel syndrome, originally described by Vaughan et al in 1968,^[1] is a rare type of trichobezoar extending into the small intestine.

Most trichophytobezoars and trichobezoars are caused by trichophagia, which is a manifestation of pica.^[14] Trichophagia is usually accompanied by trichotillomania, a rare psychiatric disorder characterized by the impulse to pull out one's own hair. The causes of trichotillomania and trichophagia are not completely understood. Recent studies have suggested that patients are more likely to be females and that appropriate psychotherapeutic interventions should be recommended.^[15]

Bezoars can be asymptomatic or patients can present with various nonspecific gastrointestinal symptoms.^[16] Abdominal ultrasonography, computed tomography, gastrointestinal radiography, and endoscopy are useful for detecting bezoars. Among these diagnostic methods, endoscopic examination plays the greatest role in making a definitive diagnosis of the composition of gastrointestinal bezoars, as well as in determining their treatment.^[17–20] Surgical, laparoscopic, or endoscopic trichobezoar removal is chosen based on the size and composition of the trichobezoar and the clinician's experience. Because gastric trichophytobezoars and trichobezoars are generally more difficult than other types to remove endoscopically, surgical and laparoscopic removal are most commonly reported.^[20,21]

A comprehensive literature search was performed using PubMed and EMBASE database for articles with focus on trichophytobezoar, which were published up until October 2016. A search strategy that Title using the term "trichophytobezoar" was used. Retrieval results showed that only 8 articles on human trichophytobezoar were reported. Of these articles, Scorpio and

Sharma^[5] reported a 5-year-old girl with large terminal ileal trichophytobezoar causing perforation peritonitis, Ciampa et al^[6] reported a case of a 13-year-old girl with a 19 × 11 × 2-cm³ trichophytobezoar, Malik et al^[16] reported a case of trichophytobezoar without gastrointestinal complaints in a 42-year-old woman, Lee et al^[22] reported a 73-year-old male with gastric cancer and trichophytobezoar, and Capeche et al^[23] reported a 20 years old man with duodenal trichophytobezoar. All of these cases were treated with laparotomy.

Sodium bicarbonate^[24] and some carbonated beverages^[9,25–27] have been widely reported as treatments for bezoars, either as sole treatment or in combination with endoscopy. The mechanism of these treatments may be that these substances increase bezoar digestion due to dissolution by sodium bicarbonate and loosening of the bezoar structure by carbon dioxide bubbles.^[25,26]

In our patient, endoscopic removal of the trichophytobezoar was achieved with the combined use of a polypectomy snare and APC, performed twice, 5 days apart. At the first treatment, only a small amount of the hair was removed because of the hard core of the trichophytobezoar containing hair and vegetable fibers. Sodium bicarbonate solution was injected into the trichophytobezoar to loosen and dissolve the structure. At repeat endoscopy 5 days later, the gastric trichophytobezoar had loosened and was successfully removed.

4. Conclusions

Trichophytobezoars, a rare category of bezoar, are often found in young girls who have trichophagia and trichotillomania. Trichophytobezoars can be successfully retrieved by combining endoscopy and sodium bicarbonate injection, offering an effective and minimally invasive therapy for clinical practice. For the general physicians, it can be carried out first, rather than surgical treatment. However, this treatment may not be effective for larger or more complex trichophytobezoars in patients with Rapunzel syndrome. Therefore, further research is needed.

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