Case Report

Huge simultaneous trichobezoars causing gastric and small-bowel obstruction

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Abstract

Bezoars are concretions of foreign materials that impair gastrointestinal motility or cause intestinal obstruction in the stomach, small intestine or bowel of humans or animals. There are many types of them such as phyto, lacto and trichobezoars. Although bezoars are not rare, multiple giant bezoars which totally fill the stomach lumen and have extension to the small intestine (Rapunzel syndrome) are very rare. This is a case report of a young girl who had a history of trichophagia and presented with partial gastric and intestinal obstructive signs. The patient was healthy, and her physical exam was almost normal and the only positive thing in her past medical history was trichophagia from several years ago. She had a big trapped bobble in her stomach and several air-fluid levels in abdominal radiograph and was investigated with endoscopy which confirmed the diagnosis of a huge gastric trichobezoar.

KEYWORDS: Bezoars, Gastric Outlet Obstruction.

Bezoars are a collection of foreign materials or indigestible organic substances in the alimentary tract of humans and certain animals such as ruminants. There are several types of bezoars; phyto, tricho, and lacto-bezoars are some samples. Indeed bezoars can be categorized based upon their location. Gastric bezoars mostly occur as a complication of gastric operations although, they may also be seen in the healthy individuals as a result of ingestion of various objects which conglome-rate in stomach and do not pass through the pylorus. A small number of patients have been reported in whom the gastric trichobezoar has a long tail and extends throughout the small bowel. This condition, known as the Rapunzel syndrome, occurs almost exclusively in young girls.\textsuperscript{1}

Case Report

A 19-year-old girl presented with colicky abdominal pain; distension and vomiting for more than 3 months. The pain increased in severity, became localized in the periumbilical area and was associated with nausea, vomiting and constipation. Previous attacks of similar pain had resolved with antispasmodics. The family said that the girl had pulled out and ingested her hair since several years ago.

On examination the abdomen was found to be slightly distended with epigastric tenderness. A large mass was palpable at the right iliac fossa. Bowel sounds were hyperactive.

The abdominal radiograph showed multiple air-fluid levels with distended small-bowel loops, a big bobble trapped at the fundus of stomach and a dome shape shadow were seen in her right upper quadrant (Figure 1). During endoscopy of the gastrointestinal tract we faced a huge trichobezoar which its tail was not visible (Figure 2). Exploration of the abdomen revealed distended stomach and proximal small-bowel loops with two masses identified causing the small-bowel obstruction. Gastric longitudinal
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Figure 1. Abdominal radiograph of the patient before upper GI endoscopy
A big bobble at fundus of stomach, several air-fluid levels and distended loops at small intestine can be seen.

Figure 2. Gastric trichobezoar confirmed by upper GI endoscopy
The endoscopic view of the patient's stomach lumen; a mass made up of hair and other fibrous material was directly visualized to occupy the entire stomach. It is taken at the admitting time and before surgery.
incision was made on the anterior wall. Hairball mass was extracted completely. The mass had completely filled the stomach lumen and was about 22 cm long and its maximum diameters were 7.5 × 6.5 cm. Then proximal ileum entrotomy was done and a mass of trichobezoar (14 × 4.5 × 4 cm$^3$) removed from the last 30 cm of the terminal ileum (Figure 3; A and B).

**Figure 3.** Stomach and ileum bezoars (extracted after gastro entrotomy)
The gross view of the stomach and ileum bezoars; these pictures were taken in the operating room after gastro-entrotomy.  
A) The trichobezoar, which cast, in the shape of the stomach; its size was 22 × 7.5 × 6.5 cm$^3$ and it was extracted by gastrectomy.  
B) Photograph shows trichobezoar that was removed through entrotomy. It was located in the terminal ileum and its size was 14 × 4.5 × 4 cm$^3$. 
The patient was tolerating a general diet until 5th day after the operation and was discharged to her home, with psychiatric follow-up.

Discussion
Historically the kings of ancient Persia were often targets of poisoning. It was therefore their practice to place a calculus from the intestinal tract of the Persian mountain goat at the bottom of their wine cups, attributing magical protective power to it and believing that the porous structure absorbed poisons. It was called "Padzahr", the Persian word for anti-poisoning. The derived word bezoar is still used to designate large, unpleasant gastrointestinal concretions. The first reported description of a post-mortem human bezoar was given by Swain in 1854.2

Several components may be found in bezoars. Trichobezoars are an infrequent form of bezoars formed from ingested hair. Lactobezoars are compact masses of undigested milk concretions located within the gastrointestinal tract; they are most frequently found in infants and can cause gastric outlet obstruction.3

Trichobezoars occur almost exclusively in females and 80% of these cases have a psychiatric disorder. Apart from obstruction, trichobezoars may also cause ulceration with haematemesis, perforation or peritonitis.4

Phytobezoars are firm masses of undigested fruit or vegetable fiber, which can cause gastric or small-bowel obstruction. Dietary behavior, previous gastric surgery, particularly partial gastrectomy or truncal vagotomy with pyloroplasty are some of the predisposing factors of bezoars.5 In adults, bezoars are most frequently encountered after gastric operations. In children they are associated with pica, mental retardation and psychiatric disorders.6

Although trichobezoars are usually seen in the stomach, they may also be found in other parts of the alimentary track such as duodenum, ileum, jejunum, colon or even Meckel's diverticulum. They can be extremely large, cause a wide variety of symptoms and can be fatal. Bezoars are known to cause gastric ulcers, intestinal obstruction or perforation, hemorrhage, peritonitis, anemia and malabsorption syndromes.7 Rarely, jaundice, pancreatitis, or colonic obstruction may occur if large bezoars develop.8

Rapunzel syndrome is a rare clinical scenario that arises when a large stomach trichobezoar mass extends through the duodenum and into the small intestine with or without mechanical intestinal obstruction. It derives its name from the long-haired heroine in the fairy tale by the Grimm brothers.9

Patients with bezoars may present gastrointestinal obstruction that may involve any part of the bowel. In childhood, undiagnosed gastric bezoars may result in serious complications. A past medical history of foreign body ingestion, especially in children and mentally impaired patients, is important. Ultrasonography and computed tomography (CT) scan are reliable methods for diagnosing gastrointestinal tract bezoars.10

Recently, magnetic resonance imaging (MRI) is recommended for the evaluation of small-bowel diseases. It can be useful in determining the site and the cause of small-bowel obstructions. MRI shows the bezoar as a mass containing mottled and confluent low signal intensities on both T1 and T2 images.11

Management of trichobezoars depends largely on the presentation of the patient. Although there are some medical treatments, they mostly used in cases with mild symptoms and are applied when there is no sign of acute abdomen. Most of these methods refer to ordering "cola" for dissolving phytobezoars.12-15 Surgical intervention should be reserved for those who have acute abdominal conditions or large bezoars. Nirasawa et al were the first who reported the laparoscopic treatment of bezoars in 1998.16 After that, a few surgeons reported laparoscopic removal of bezoars with almost similar techniques.17 Laparoscopy is associated with less postoperative pain, faster recovery, reduced rate of wound complications and very good cosmetic results. But endoscopy is used just for small bezoars and those with softer construction (most of the bezoars specially trichobezoars are very big and have a
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stony structure) thus has a high failure rate and is associated with significant complications such as esophageal rupture. The treatment consists of removing the mass by entrotomy or when it is not feasible, resection of the obstructed part.4,18

Laparoscopic approach could be problematic in cases of giant trichobezoars. In some reports performing minilaparotomy is recommended because of its improved cosmetic results especially in young girls.19

In the Rapunzel syndrome it is recommended to extract the bezoars by multiple entrotomy.9 De Baker et al reported its surgical mortality rate to be 10.4%.4 It is obligatory to do a thorough exploration of all the small intestine and the stomach searching in retained bezoars.20

Recurrent bezoars have also been reported, especially in women and psychiatric follow-up is necessary to prevent recurrences.21,22

Conclusions

Diagnosis of bezoars needs high index of suspicion and it should be considered in the differential diagnosis of bloating in young females. Trichobezoars are usually seen in healthy patients with normal digestive system but with psychiatric problems such as trichophagia, trichotillomania and/or mental retardation. Determining the cause of trichobezoars in patients without a known psychiatric history is very difficult but it seems that most of the times there are some degrees of behavioral disturbance linked with them. So in such cases psychiatric evaluations is strongly recommended.

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Conflict of Interests

Authors have no conflict of interests.

Authors' Contributions

FMG carried out the design, coordinated the study, supervised all the experiments and participated in manuscript preparation. MH coordinated and carried out all surgical management. MSR participated in manuscript preparation and did the final revision. MLM provided assistance in the design of the study, coordinated and carried out all the medical experiments and participated in manuscript preparation. FJ provided assistance in all experiments. All authors have read and approved the content of the manuscript.

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