

Research Article

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Bezoars: a rare cause of Rapunzel syndrome and large bowel obstruction by sigmoid volvulus

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ABSTRACT

Background: Bezoars are conglomerates of indigested materials that accumulate in the gastrointestinal tract. The aim of this work was to study our cases of bezoars and its different management modalities.

Methods: This is a retrospective study of the patients diagnosed to have gastrointestinal bezoars that were admitted to our institute from the start of 2008 to the end of 2014. The clinical data, management, and outcomes of these patients were studied.

Results: Bezoars were present in 8 patients. There were 5 female patients (62.5%) and 3 males, and the median age was 28 years (range: 16-54). Gastric bezoars were present in 5 patients, all were females and all were trichobezoars. One of them was extending to the duodenum with a tail of hair, making what is called rapunzel syndrome. Two were treated by endoscopy, two needed open laparotomy with gastrotomy and one treated conservatively. Small bowel bezoars were phytobezoars that caused small bowel obstruction in two male patients. One treated conservatively, and laparotomy and enterotomy done in another patient. One male patient with sigmoid bezoars caused volvulus and ischemia of the sigmoid colon. This was treated with laparotomy, sigmoid resection and colostomy. There were no mortality and very low morbidity.

Conclusions: Bezoars are uncommon causes of gastrointestinal diseases. Presentations depend on the site and size of the bezoars. Gastric bezoars are usually trichobezoars and are more common in young females. Rapunzel syndrome is a rare presentation of trichobezoars. Small bowel bezoars are usually phytobezoars, and their usual presentation is small bowel obstruction. Sigmoid volvulus is a very rare presentation of colonic bezoars. Different treatment modalities are needed according to its type and location.

Keywords: Bezoars, Trichobezoars, Phytobezoars, Rapunzel syndrome, Sigmoid volvulus

INTRODUCTION

The term bezoar refers to an intra-luminal mass in the gastrointestinal system caused by the accumulation of indigestible ingested materials, such as vegetables, fruits, and hair. Bezoars are named according to the material they are made of: a trichobezoar consists of hair; a phytobezoar of vegetable and fruit residues; a lactobezoar is formed from dairy products; a pharmacobezoar is caused by medications and a polybezoar is caused by ingested foreign bodies. The most common type of

bezoar is the phytobezoar, which consists of indigestible food residue, such as cellulose and hemicellulose.¹

The stomach is the most common site of trichobezoars, while phytobezoars are more common in the small intestine and usually presented with small bowel obstruction (SBO). Colonic bezoars are very rare and usually in the form of lithobezoars.²

Trichobezoars are unusual and are usually found in young psychiatric females, who often deny eating their own hair

(trichophagy). It is caused by ingestion of hair, which remains undigested in the stomach. Human hair is resistant to digestion and peristaltic movement because of its smoothness. Continuous ingestion of hair can lead to their impaction along with mucus and food materials into the stomach. In some cases, the trichobezoar extends through the pylorus into the small intestine. This condition, called Rapunzel syndrome, was first described by Vaughan et al, in 1968.^{3,4}

Trichobezoars may present with abdominal pain, nausea/vomiting, early satiety, weight loss, intestinal obstruction, ulceration leading to bleeding and/or perforation. Rarely intussusception can also happen. An upper abdominal mass remains the commonest presenting sign. The diagnosis is made easily by upper endoscopy or by CT scan. Management options of trichobezoars include endoscopic removal, laparoscopic removal, or via laparotomy with anterior gastrotomy. Medical treatment as enzyme therapy with papain, cellulase, or acetylcysteine may be tried but usually ineffective.⁴ Cola was used for dissolution of bezoar since the start of 2000. The mechanism of bezoar dissolution by cola has not been well explained, but having an acidity of pH 2.6 due to carbonic and phosphoric acid, it resembles gastric acid which is thought to be important for dissolution of bezoar. In addition, NaHCO₃ mucolytic effect and CO₂ bubbles enhance the dissolving mechanism.⁵ Ladas et al suggested that cola alone could be effective in gastric phytobezoar dissolution in half of the cases and combination with additional endoscopic methods, was successful in more than 90% of cases.⁶ Surgery is indicated when a very large or solid bezoar causes perforation or hemorrhage, or in the case of Rapunzel syndrome, when there is significant extension of the bezoar.⁷

Phytobezoar-induced SBO occurs mostly in the distal small intestine.⁸ The greater width of the colonic lumen reduces the possibility of colonic mechanical obstruction due to bezoars, although a few rare cases of colon obstruction have been reported in children, usually in the

form of lithobezoars.¹ Laparoscopic interventions are being performed increasingly. However, open surgery is still the most common method used for the surgical treatment of bezoar-induced SBO. For the patients with ischemia and perforation caused by bezoars, anastomosis or stoma procedures should be performed with segmental small bowel resection.¹

The aim of this work was to study retrospectively our cases of bezoars and to study its different types, presentations, and different management modalities.

METHODS

This is a retrospective study of the patients diagnosed to have gastrointestinal bezoars that were admitted to our institute from the start of 2008 to the end of 2014. The charts were retrospectively reviewed and the clinical data, operative and non-operative treatment, and outcomes of these patients were collected and studied. The study was in accordance with the ethical standards of our Institutional Ethical Committee. Informed consent was taken from all the patients. The data collected included age and gender, symptoms and signs, the presence of acute complications (bowel obstruction and perforation), and the composition, location and extent of the bezoars. Management parameters including the diagnostic workup and different treatment modalities were collected. Outcome parameters in the form of early and late complications and any mortality or recurrence were reviewed.

RESULTS

Bezoars were present in 8 patients. There were 5 female patients (62.5%) and 3 males, and the median age was 28 years (range: 16-54). Gastric bezoars were present in 5 patients (62.5%), all were females (100%) and all were trichobezoars (100%). One of them was extending to the duodenum with a tail of hair, making what is called Rapunzel syndrome.

Table 1: Patients characteristics according to the subgroups of bezoars.

Patients characteristics	Gastric bezoars	Small intestinal bezoars	Colonic bezoars
Number of patients	5 (62.5%)	2 (25%)	1 (12.5%)
Median age and range	19 (16-25)	51 (48-54)	44
Sex ratio (M/F)	5 females	2 males	1 male
Nature of bezoars	Trichobezoars	Phytobezoars	Polybezoars
Previous abd. surgery	0	2	0
Psychiatric diseases	Trichotillomania (3) Trichophagia (5)	0	Mental retardation
Presentations	Vomiting, nausea, pain, weight loss, epig mass	SBO	Acute abdomen, septic shock

Presenting symptoms included abdominal pain, vomiting, nausea, weight loss and anorexia. Most of the patients had a palpable epigastric mass (4 patients). Most of the female patients with trichobezoars were suffering from trichotillomania (pulling out of her own hair) and trichophagia (swallowing of hair). Table 1 shows the patients characteristics according to the subgroups and location of the bezoars.

A large variety of imaging modalities were used. These included abdominal x-rays, abdominal ultrasound, CT abdomen. The diagnosis was confirmed in all the cases of trichobezoars with upper endoscopy. Figure 1 shows the CT picture of giant gastric trichobezoar.

Two of the patients with trichobezoars were successfully treated by endoscopy, one treated conservatively as it was small one, and two needed open laparotomy with anterior gastrotomy for hair removal (Figures 2-4).

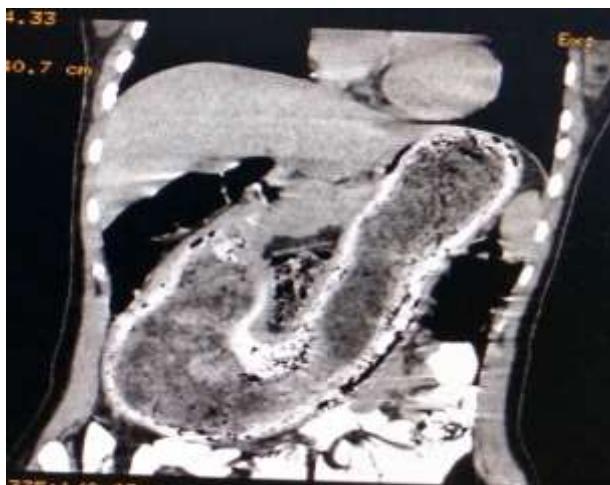


Figure 1: CT scan picture of giant gastric trichobezoar.



Figure 2: Case of trichobezoar treated by laparotomy and anterior gastrotomy.



Figure 3: A trichobezoar after its delivery from a small gastrotomy with extension of hair into the duodenum (Rapunzel Syndrome).



Figure 4: The stomach after closure of the gastrotomy with one layer of interrupted absorbable sutures.

Small bowel bezoars were phytobezoars that caused small bowel obstruction in two male patients. They were successfully treated conservatively in one patient, and with laparotomy and enterotomy in another one male patient. Both patients had history of previous abdominal surgery with history of recurrent colicky abdominal pain. They were presented with picture of adhesive SBO with multiple air fluid levels on abdominal x-ray. The patient treated conservatively passed a large amount of undigested orange fibers as a phytobezoar after swallowing of gastrograffin for follow through series. The patient treated with laparotomy failed conservative treatment and developed severe abdominal pain and vomiting after gastrograffin swallowing. A large amount of undigested peaches were delivered from his ileum by small enterotomy.

There was one mentally retarded male patient with sigmoid bezoars that caused volvulus and sigmoid ischemia with acute abdomen and septic shock. His abdominal x-ray shows huge distention of the sigmoid colon. He was resuscitated in the intensive care unit with vigorous intravenous fluids and antibiotics. Urgent abdominal exploration was done rapidly after the resuscitation. The sigmoid colon was severely distended with volvulus and ischemia mainly at the anti mesenteric border. It was filled completely with ingested pieces of foreign bodies in the form of gauzes and pieces of clothes and bed sheets (Figures 5-6).



Figure 5: The sigmoid colon markedly distended with volvulus and ischemia of the anti-mesenteric border.



Figure 6: The opened distended sigmoid colon with the foreign bodies inside.

This was treated successfully with sigmoid resection and colostomy. Re-anastomosis and closure of the colostomy was performed after 4 months without complications. Table 2 shows the diagnostic procedures and various treatment modalities for each group of patients. There were no mortality and very low morbidity. No recurrence has been documented so far. Only one female patient developed wound infection after laparotomy and gastrotomy. After treating her wound successfully, there was regain of fever with high total leucocytic count. There was pus collection anterior to the stomach which was drained percutaneously by pig-tail catheter guided with ultrasonography. Follow up with CT scan shows minimal fluid collection which was treated conservatively with antibiotics.

Table 2: The diagnostic procedures and various treatment modalities for each group of patients.

Management	Gastric bezoars	Small intestinal bezoars	Colonic bezoars
Diagnostic modality	Endosc., CT	X-ray, gastrogr. series	X-ray, exploration
Conservative	1	1	0
Endoscopy	2	0	0
Open surgery	2 exploration and gastrotomy	1 exploration and enterotomy	Sigmoid resection and colostomy

DISCUSSION

Bezoar is the accumulation of undigested foreign bodies or nutrients in the gastrointestinal tract. These foreign bodies can be hair (trichobezoar), fibers or seeds of vegetables and fruits (phytobezoar) or remnants of milk (lactobezoar) and stones (lithobezoar). Although the stomach is the predilection site for bezoar, it can be rarely seen in the colon and it may cause mechanical intestinal obstruction.⁹

In present study, most of the cases were trichobezoars located in the stomach in young females. Only one patient had the rare Rapunzel syndrome. Two male patients had phytobezoars causing SBO. One mentally retarded male patient was presented with acute abdominal pain and septic shock, with volvulus of the sigmoid colon due to ingestion of foreign materials in the form of pieces of gauze, bed sheets and clothes.

Our results were different from previous reports that shows higher prevalence of phytobezoars.⁶ However, this

could be from the small number of our patients and the fact that the prevalence of bezoars varies among ethnic groups and geographic locations, since the occurrence rate of phytobezoars is mostly reflected by food cultures. For example, multiple cases of persimmon phytobezoar (diospyrobezoar) have been reported in regions where the residents frequently consume fresh persimmon fruits and dried persimmons.¹⁰ Our results of higher prevalence of trichobezoars in young females with trichotillomania and trichophagia were resembling other previous reports which show that trichobezoars are nearly always gastric and diagnosed in young females.¹¹

The clinical presentation of bezoars in our study was not different from other studies, except for the case of sigmoid colon bezoar caused by a very rare type of foreign body bezoar that presented with sigmoid volvulus and acute abdomen.^{1,12} For our knowledge, this is the first case reported so far with sigmoid volvulus due to bezoars of ingested pieces of gauze and bed sheets and clothes that was localized only to sigmoid colon.

The most common source of fibers in the phytobezoars in the literature was the persimmon. However, this was not the case in our study which showed the orange fibers and undigested peaches as the cause of our cases. The previous abdominal surgery and adhesive type of mechanical SBO was a predisposing factor in our study. It is believed that this is a complication of delayed gastric emptying due to different causes.¹³

The role of upper gastrointestinal endoscopy is crucial in the diagnosis and management of trichobezoars. In our study, the diagnoses were confirmed by endoscopy in all the cases of trichobezoars, and two cases were successfully treated with it. However, the success of endoscopic removal is not similar in all the previous reports. While all the attempts failed in one study, it was successful in many others.^{10,12,14}

Laparoscopic removal of bezoars continues to be controversial.¹² The first described case of laparoscopic removal of a gastric trichobezoar was in 1998 and since then a few case reports described successful laparoscopic removal of gastric bezoars, generally in adults and adolescents.¹⁵ In their literature review in 2010, Gorter and colleagues found only six case reports of the use of laparoscopy to remove gastric bezoars since the original description in 1998, two of which failed and resulted in conversion to laparotomy.¹¹ To date, an open approach is still preferred by many because of the difficulty in removing a large bezoar and the risk of spillage in a laparoscopic setting.¹² It was found that our cases of open laparotomy were not suitable for laparoscopy. The cases of gastric trichobezoars were huge, and the cases of small intestinal and colonic bezoars were markedly distended. However, recent studies emphasized the importance of laparoscopy in the management of gastrointestinal bezoars.^{16,17}

We have one patient with small gastric trichobezoar that was treated conservatively with psychiatric and endoscopic follow up. Spontaneous disappearance of a bezoar under the absence of specific treatment was also observed in previous reports.¹⁸ The etiology of the bezoars and the mechanisms underlying how the bezoars were digested in these patients remain to be determined. However, careful follow-up without any specific treatment is a possible option in the management of bezoar patients, if they are in stable condition.¹³ Cola beverages were not used for the dissolution of bezoars as was shown in many studies with different success rates. The main reason was that most of our cases were trichobezoars, while cola is more suitable for phytobezoars.

There are minimal complications in present study with no mortality. Only one young female patients with gastric trichobezoar developed wound infection and intra-peritoneal abscess formation which was drained percutaneously. Even the case of peritonitis with septic shock and sigmoid volvulus passed smoothly without any complications. This was in accordance with most of the published studies.^{11,19}

CONCLUSION

Bezoars are uncommon causes of gastrointestinal diseases. Presentations depend on the site and size of the bezoars. Gastric bezoars are usually trichobezoars and are more common in young females. Rapunzel syndrome is a rare presentation of trichobezoars. Small bowel bezoars are usually phytobezoars and their usual presentation are SBO. Sigmoid volvulus is a very rare presentation of colonic bezoars. Different treatment modalities are needed according to the type and location. However, larger case series and randomized studies (if possible) are needed for better understanding the etiology and how to manage this rare disease.

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