

Gastrointestinal Bezoars in Pediatrics: Case Series and Literature Review

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Research Article

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Abstract

Purpose:

To present data, management and outcome of cases with trichobezoars in our institute for whom surgery was done.

Methods:

We documented a retrospective review of trichobezoars done in our hospital between 2016 and 2022. All demographic data collected included gender and age of cases, composition and extent of bezoar, clinical presentation, imaging modalities, endoscopic trial, surgical approach and outcome.

Results:

Five cases of gastrointestinal trichobezoars underwent surgery. All cases were females between (13 & 16 years). Trichobezoars were three gastric, one ileal and one of combined gastric and colonic. Complaints were abdominal pain, vomiting, weight loss, and halitosis. Three cases had a palpable abdominal mass. Different Radiological modalities were performed. Endoscopic retrieval was tried in one patient and the laparoscopic approach in another one, but the first route failed. Laparotomy followed by gastrotomy, enterotomy and colotomy were done without complications.

Conclusions:

Trichobezoars should be suspected in any child with unexplained abdominal complaints, or with a palpable abdominal mass, especially in girls. Imaging can be done in different modalities for diagnosis. Endoscopic retrieval could be tried; however its failure is common, necessitating laparotomy, which has an excellent outcome.

Introduction:

Bezoars are indigestible hard lumps which are usually found in the stomach as pylorus is a natural obstacle for distal propagation. [1] The stomach find a difficulty to expel either the hair or other substances out of its lumen because the surface of friction is not enough for propulsive peristalsis. [2] There are different types of bezoar; phytobezoar, trichobezoar, lithobezoar, pharmacobezoar, plasticobezoar, lactobezoar and metal bezoar. [3]

Trichobezoars constitute \approx 6% of all bezoars cases in humans which mostly affect females in the twenties and thirties of life with a rarity in pediatrics. [4–6] Trichotillomania (hair pulling) and trichophagia (hair eating) are commonly associated in about 10% of cases presenting with trichobezoars. [7]

Abdominal pain, nausea, vomiting, weight loss, constipation, Small bowel obstruction, occasional hematemesis with a palpable mass are commonly found in these patients. [8]

Disturbance of gastric physiology as gastric emptying due to gastric operations, gastroparesis or a gastric outlet obstruction is a well-known cause of bezoars. [9] “Rapunzel syndrome” is a variant of trichobezoar in which the mass extends past the pylorus with difficult endoscopic removal that mandates the operative removal of it. [10, 11]

Variable imaging scans have been recommended for diagnosis of bezoars as an opaque soft tissue in a dilated stomach. [12–14] Ultrasound shows it curvilinear with bright echogenic band generating a shadow over the left upper quadrant. [12, 13] Ultrasound has a limited ability in detecting trichobezoars due to the high echogenicity of hair and the multiple acoustic interfaces caused by trapped air and food. [15]

Both upper GIT contrast radiography and endoscopy are considered the ideal choice for confirmation of the diagnosis. [13, 15] Contrast radiography detects the trichobezoar and its related complications. In addition, endoscopy is the definitive diagnostic tool for trichobezoar and might be used for endoscopic retrieval of small trichobezoars located proximally. [13]

CT-scan is the diagnostic tool of choice in the localization of bezoars with a characteristic mottled gas pattern in the mass which is supposed to be air bubbles retained within the bezoar [15] while the bezoar appears as a mass in the small bowel having mottled and confluent low signal intensities on both T1 and T2- weighted magnetic resonance images. [16]

The treatment involves retrieval of the mass through a single enterotomy or resection of the bowel or multiple enterotomies in the Rapunzel syndrome. It is mandatory to perform a thorough exploration of all the small intestine and the stomach searching for retained bezoars. [17]

Laparoscopy has the advantage of fewer intraabdominal adhesions versus open laparotomy. Additionally, the psychological counselling is crucial to prevent recurrences. [18]

Herein we present our experience regarding the diagnosis and treatment of gastrointestinal trichobezoars.

Patients And Methods:

Bearing on approval from our Institutional Review Board after assuring of an informed consent from all parents for possible further publication conditioning of data confidentiality, we retrospectively identified all patients at our institutional hospital that required surgical treatment for gastrointestinal trichobezoars between January 2016 and January 2022. The patients’ data were collected including age, gender, symptoms, physical signs in addition to the presence of bowel obstruction, perforation, and the composition, location and extent of the bezoars. Management items included the investigations, history of previous endoscopic intervention, and operative technique. Outcome involved the presence of

complications either early or late were collected by reviewing inpatient records as well as outpatient follow-up records.

Results:

We identified five patients who underwent surgery for the treatment of gastrointestinal trichobezoars between 2016 and 2022. All patients were females; their age range was 13–16 years. The five patients, one case had psychological issues of paternal divorce and four had a history of trichotillomania and trichophagia. Additionally, two patients had extension of the trichobezoar past the pylorus (Rapunzel syndrome) as shown in figure (A,B), one case had small gut trichobezoar figure (C) in addition to another case had combined gastric and colonic trichobezoars figure (D,E) and one had gastric trichobezoar.

The presenting symptoms were abdominal pain and vomiting and a palpable abdominal mass was detected in three patients as illustrated in table (1). Physical examination revealed a large epigastric mass in two cases and another in the left hypochondrium. Preoperative imaging was obtained in all cases and included plain films (all cases), abdominal ultrasound (all cases) and abdominal CT (four cases).

In one patient, endoscopic retrieval of the trichobezoar was attempted prior to surgery but failed. Six patients underwent laparotomy then either gastrotomy or enterotomy or colotomy for removal of the bezoar. In one case, the laparoscopic approach was attempted and was used to assist transumbilical exteriorization of the bowel then retrieve ileal trichobezoar. All patients recovered without complications except the case was done laparoscopically showed umbilical wound infection in follow up visit. All cases were reviewed by a psychiatrist for further management.

Cases Report:

Case 1:

A 13-year old girl had a recurrent abdominal pain for two months; she presented with symptoms of intestinal obstruction. On examination, the patient was anxious with tender abdomen and rebound tenderness mainly in right iliac fossa. Plain abdominal x-ray revealed multiple air-fluid levels of small bowel obstruction and abdominal ultrasound revealed bowel dilatation. The patient was then explored using laparoscopy through optical umbilical, suprapubic, left lumbar ports which revealed significantly dilated small bowel loop with a bowel mass in the mid ileum measuring 7×5×3 cm with collapse of the distal bowel and no other masses were detected on exploration of the remaining gut as in Figure (F). Exteriorization of the loop containing the mass through umbilical port with extension of skin and abdominal wall incision. Extracorporeal enterotomy was done and a mass of hair threads in the form of a bunch was detected. The mass was removed through transverse enterotomy and then was closed vertically in two layers. The postoperative course was smooth and the patient was counseled by the psychiatrist for further treatment after notification of recent paternal divorce.

Case 2:

A 14-year-old girl had a history of trichotillomania, trichophagia and obsessive compulsive disorder, presented with a 3-day history of epigastric pain in association with vomiting. It had worsened over the past three days. Ultrasound revealed an echogenic mass suspicious of a large gastric trichobezoar. Trial of endoscopic retrieval was done but failed. Laparotomy followed by gastrotomy was performed to remove the bezoar. She was assessed by the psychiatrist for etiological treatment and behavior modification.

Case 3:

A 15 year old girl had a history of trichotillomania, trichophagia and previous retrieval of gastric trichobezoar 1 year ago, who complained of abdominal pain and vomiting within the last two days with a palpable abdominal mass in the left hypochondrium. Ultrasound revealed suspected gastric trichobezoar and interestingly, CT abdomen and pelvis showed gastric, colonic trichobezoars in addition to right ovarian cyst 5 cm as illustrated in Figures (G). Laparotomy followed by gastrotomy and colotomy were performed to extricate the bezoars then ovarian sparing cystectomy was also done (H). The patient was discharged without symptoms. She was counseled psychologically for etiological treatment.

Case 4:

A 16 year old girl had trichotillomania and depression due to domestic problems without clinically significant medical or surgical issues. She presented to the out-patient clinic with chronic upper abdominal pain increasing with eating associated with infrequent vomiting of gastric content and food material, she also had 1 stone of weight loss during the complaint period. On examination she was vitally stable, apparently ill, and a left hypochondrium tender mass was detected. Plain X-ray abdomen was unremarkable, abdominal ultrasound showed suspicious intragastric mass, CT abdomen and pelvis with contrast was done showing intragastric mass reaching duodenum with thickened gastric wall and dilated stomach suggesting of trichobezoar bearing on the above mentioned history figure (I). Exploration followed by wide longitudinal gastrotomy was done revealing a huge trichobezoar reaching D1 mandating its extraction. Formal exploration till rectum was done in addition to closure of gastrotomy with PDS 2/0 interrupted suture single layer. Patient recovered smoothly and discharged to the ward, oral feeding resumed 48 hrs after surgery and she was discharged home 5 days post-operatively.

Case 5:

16 year old girl had trichotillomania and psychiatric issues. She presented with recurrent vomiting with a long history of chronic upper abdominal pain and weight loss two months ago. On examination she was vitally stable, apparently ill, and a left hypochondrium tender mass was detected. Plain X-ray abdomen was unremarkable, abdominal ultrasound showed suspicious intragastric mass, CT abdomen and pelvis with contrast was done showing intragastric mass reaching duodenum with thickened gastric wall and dilated stomach suggesting of trichobezoar bearing on the above mentioned history. Exploration followed

by wide longitudinal gastrotomy was done revealing a huge tricobezoar reaching D2 mandating its extraction. Formal exploration till rectum was done in addition to closure of gastrotomy with PDS 2/0 interrupted suture single layer. Patient recovered smoothly and discharged to the ward, oral feeding resumed 48 hrs after surgery and she was discharged home 5 days post-operatively.

Table 1. Summary of 5 cases of surgically treated gastrointestinal bezoars

Item	Case 1	Case 2	Case 3	Case 4	Case 5
Sex	Female	Female	Female	Female	Female
Age	13	14	15	16	16
Chronic Abd. pain	no	no	yes	Yes	Yes
Vomiting	yes	yes	yes	Yes	Yes
Weight Loss	no	no	yes	Yes	Yes
Alopecia	yes	no	no	No	Yes
Palpable Mass	no	no	yes	Yes	Yes
Peritonitis signs	no	no	no	No	No
Psychiatry Comorbidity	yes	no	yes	Yes	yes
Laboratory Results	CRP TLC	no	Iron deficiency anemia	Low K, iron deficiency anaemia.	
Abdominal x ray	Air fluid levels	yes	yes	yes	yes
Ultrasound results	Bowel dilatation	Free fluid	Echogenic mass in the stomach suspicious of trichobezoar	Echogenic mass suspicious of trichobezoar.	Echogenic mass suspicious of trichobezoar
CT Abdomen Results	No	Trichobezoar In the stomach	Trichobezoar in stomach & colon Right Ovarian cyst 5cm	Trichobezoar in stomach & 1 st part of duodenum (Rapunzel)	Trichobezoar in stomach & 2 nd part of duodenum (Rapunzel)
Endoscopic trial	no	yes	no	No	No
Preoperative Diagnosis	no	yes	yes	Yes	yes
Location	Proximal ileum	stomach	Stomach colon	Stomach	Stomach
Treatment	Laparoscopic assisted enterotomy	Laparotomy gastrotoomy	Laparotomy Gastrotoomy	Laparotomy gastrotoomy	Laparotomy gastrotoomy

			colotomy		
Successful	yes	yes	yes	Yes	Yes
Conventional Laparotomy	no	yes	yes	Yes	yes
Composition	trichobezoar	trichobezoar	trichobezoar	trichobezoar	trichobezoar
Associated Pathology			Right ovarian cyst 5 cm Corpus luteum hemorrhagic cyst	No	no
Size	6×5×3 cm	10×6×4 cm	11×7×4 cm 8×5×3 cm	15x8x5 cm	25x12x8 cm
Postoperative complications	Port site infection	no	no	No	No
Recurrences	no	no	no	No	No
Additional psychiatric consultation	yes	yes	yes	yes	Yes

Discussion:

The origin of bezoar is either from the Arabic term “badzehr” or possibly the Persian word “padzahr”, both meaning to expel poison, or antidote. These words were described a greenish, hard concretion found in the animal stomach, which was formerly considered a useful medication, sometimes with magical properties. In the mid-1890s, Quain, an Irish surgeon and anatomist at the University of London, reported a mass in the stomach, found on autopsy, which he called a “bezoar” [19, 20].

Our 6 year retrospective review showed that surgical treatment for gastrointestinal bezoars is rare. As regard trichobezoar, we have reported our experience of five adolescent patients (three gastric, one both gastric and colon and one ileum) who were treated for trichobezoar. Gorter et al. [21], reported four cases of gastric bezoars were identified over an 18 year period and Fallon et al. [22] identified seven cases that occurred over an 8 year period. However, we should suspect of gastrointestinal bezoars as the etiology of abdominal cramps, vomiting, and loss of weight or abdominal mass despite of its rarity. [23] We found epigastric mass in two patients in addition to a mass in the left hypochondrium in one case, and this finding in patients with the above mentioned symptoms should raise the suspicion of gastrointestinal bezoar. In girls, history of trichotillomania and trichophagia, can lead to the correct diagnosis and workup. Additionally, mental retardation is another medical condition that should raise the suspicion of gastrointestinal bezoars.

Various imaging modalities have proved to be sensitive in demonstrating gastrointestinal bezoars and their complications especially of the gastric ones. Therefore, we recommend starting with plain films, and performing more advanced imaging only if the above are equivocal.

Endoscopy is rare to be successful in removal of gastric bezoars, while reports of unsuccessful endoscopic attempts are more common [24–25]. Moreover, attempted endoscopic fragmentation might lead to complications such as gastric and esophageal perforations [26, 27] and distal “embolization” of fragments causing bowel obstruction [28].

Our attempt of endoscopic retrieval failed, whereas in the series reported by Gorter, endoscopy was attempted in two of four cases and failed in both [21]. Therefore, failure of endoscopy should prompt surgical intervention. Moreover, surgical intervention may be suitable in cases where the trichobezoar is strongly suspected, especially with large and extended trichobezoars or in cases where gastric perforation is suspected.

Surgical removal is generally accomplished via laparotomy then gastrotomy or enterotomy with removal of the bezoar [29]. Rapunzel syndrome, defined as extension of the bezoar beyond the pylorus [19], it can generally be removed through the gastrotomy only. However, one should be aware of possible small bowel obstruction due to fragmentation of the bezoar. Therefore, extracting the bezoar through the gastrotomy should be done very carefully.

Gastrotomy and colotomy in our series were performed via laparotomy, with one case of enterotomy was done laparoscopically assisted. In the literature, laparoscopic removal of bezoars is controversial. In 1998, the laparoscopic trial of a gastric trichobezoar removal was described [30], and then more cases were also illustrated especially in adults and adolescents. Cintolo and team [31] mentioned success of laparoscopical removal a large gastric trichobezoar from a 4 year old girl, during which fragmentation of the bezoar was done allowing removal through the 12 mm port site. In 2010, Gorter and colleagues [21] found only six case reports of the laparoscopic removal of gastric bezoars since the initial trial in 1998, two cases failed and conversion was done. Till now, the open approach is still the best choice due to the difficult removal of a large bezoar and the risk of spillage in laparoscopy. Recently reported, however, was the successful laparoscopy-assisted removal of gastric trichobezoars in five patients. The technique involves temporary gastrocutaneopexy, with or without the use of Alexis wound protector. [32, 33]

The outcome expected after surgical removal via laparotomy then gastrotomy or enterotomy, as found in our series as well as in the literature, is excellent. In our series we did not encounter any complications except one case had port site infection. The reported rate of postoperative complications in the literature according to the review by Gorter et al. [21] was also very low – 8 of 100 patients, with mostly minor complications (minor wound infections in three, pneumonia in two, postoperative ileus in two, and fecal leakage into the wound in one). In the case study reported by Fallon and co-authors [22], the rate of postoperative complications was also similar – 1 case (14%) of wound infection [22].

Conclusion:

Gastrointestinal bezoars should be considered in cases of abnormal abdominal complaints and mass, especially in girls with a history of trichotillomania with trichophagia. Even in the absence of this history, bezoars may occur in mentally retarded children. Plain films can aid in the diagnosis before using UGI contrast, ultrasound or CT.

Endoscopic retrieval could be tried in selected cases despite the preference of surgery, which shows better prognosis as the last option after failure of the dissolving agents and endoscopic to removal.

In all types of bezoars, prevention can be implemented with comprehensive psychiatric evaluation and management to decrease recurrence that has been reported to be 14% in the literature.

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Figures



Fig. (A): Rapunzel syndrome; case 4.



Fig. (B): Rapunzel syndrome; case 5.



Fig. (C): Extracorporeal view of ileal trichobezoar; case 1.

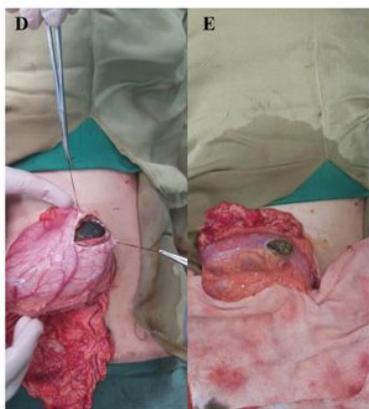


Fig. (D & E): Operative findings case 3.



Fig. (F): Laparoscopic view of ileal trichobezoar; case 1

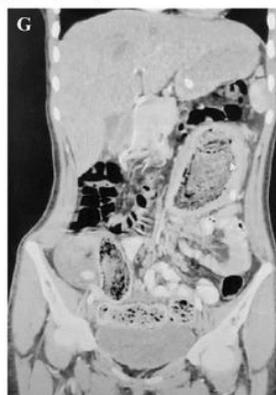


Fig. (G): CT findings; case 3



Fig. (H): Ovarian cyst; case 3

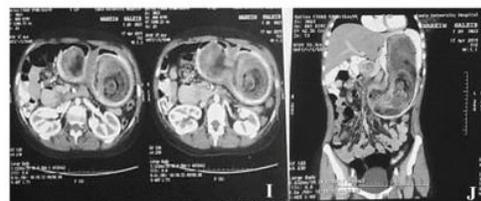


Fig. (I & J): CT finding; Case 4

Figure 1

See image above for figure legend.