Original Article

Gastrointestinal Bezoars in Paediatrics: Case Series and Literature Review

Mohamed Ahmed Elghazeery, Alhassan Mohamed Hassan[1]

Department of General Surgery, Pediatric Surgery Unit, Faculty of Medicine, Tanta University, [1]Department of General Surgery, Gastrointestinal Surgery Unit, Faculty of Medicine, Tanta University, Tanta, Egypt

Abstract

Background: Bezoars are indigestible lumps which are usually found in stomach. Types of bezoar include phytobezoar, trichobezoar, lithobezoar, pharmacobezoar, plasticobezoar, lactobezoar and metal bezoar. Trichobezoars mostly affect females in 20s and 30s with a rarity in paediatrics. Unexplained complaints with a palpable mass are commonly found in these patients. Treatment involves retrieval of mass with searching for others. The purpose of this study was to present data and surgical management of cases with trichobezoars. **Materials and 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 tract (GIT) trichobezoars underwent surgery. All cases were females between (13 and 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 was 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.

Keywords: Abdominal pain, epigastric mass, gastrointestinal bezoars, trichobezoars, trichophagia, trichotillomania

Introduction

Bezoars are indigestible lumps, usually found in the stomach as pylorus prevents distal propagation.[1] The stomach has not enough friction surface for these substances to expel out of its lumen.[2] There are phytobezoar, trichobezoar, lithobezoar, pharmacobezoar, plasticobezoar, lactobezoar and metal bezoar.[3]

Trichobezoars constitute <6% of bezoars affecting females in 20s and 30s.[4-6] Trichotillomania and trichophagia are associated in 10% of cases having trichobezoars.[7]

GIT upset, weight loss, hematemesis and a palpable mass are common complaints.[8] Disturbed emptying due to gastric operations, gastroparesis or outlet obstruction is a cause of bezoars.[9] 'Rapunzel syndrome' is a trichobezoar variant, that extends past the pylorus that mandates surgery.[10,11]

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Radiography is recommended for diagnosis.[12-14] Ultrasound shows a limited ability in detecting trichobezoar due to its high echogenicity of hair and multiple acoustic interfaces.[12,13,15]

GIT contrast and endoscopy are ideal for diagnosis and possible retrieval of small proximal trichobezoars.[13,15] Computed tomography (CT)-scan detects a trichobezoar with a characteristic mottled gas pattern of retained air bubbles,[15] whereas trichobezoar appears as a mottled mass with confluent low signals in magnetic resonance imaging.[16]

The treatment involves retrieval of the mass besides exploration of the gut for retained bezoars.[2] Psychological counselling is

Address for correspondence: Dr. Mohamed Ahmed Elghazeery, Pediatric Surgery Unit, Faculty of Medicine, Tanta University, Tanta, Egypt.
E-mail: elghazeery@hotmail.com

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mandatory to prevent recurrences.[17] Our aim is to present the management of gastrointestinal trichobezoars.

Materials and Methods

Bearing on approval from our institutional review board after assuring of 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 and physical signs in addition to the presence of bowel obstruction, perforation, the composition, location and extent of the bezoars. Management items included the investigations, a history of previous endoscopic intervention and operative technique. The outcome involved the presence of complications either early or late was 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 female; their age range was 13–16 years. Of the five patients, one case had psychological issues of paternal divorce and four had a history of trichotillomania and trichophagia. In addition, two patients had an extension of the trichobezoar past the pylorus (Rapunzel syndrome) as shown in Figures 1 and 2, one case had small gut trichobezoar as shown in Figure 3 in addition to another case had combined gastric and colonic trichobezoars as shown in Figures 4 and 5 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).



Figure 1: Rapunzel syndrome; case 4



Figure 2: Rapunzel syndrome; case 5

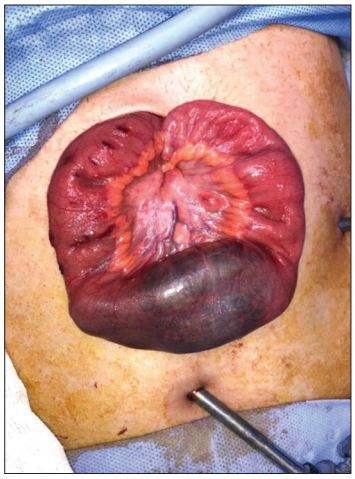


Figure 3: Extracorporeal view of ileal trichobezoar; case 1



Figure 4: Operative findings case 3



Figure 5: Operative findings case 3

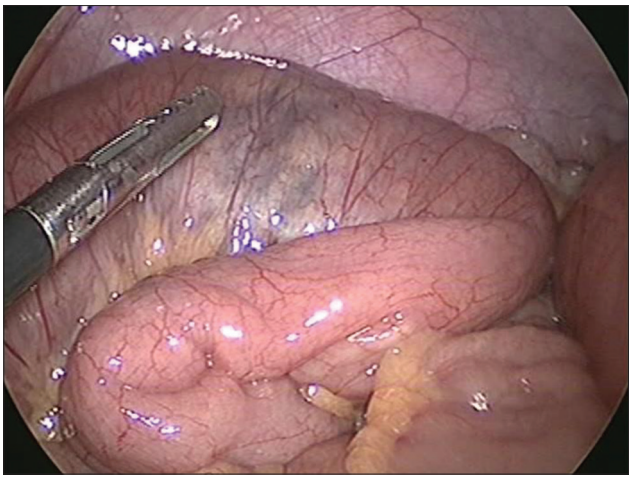


Figure 6: Laparoscopic view of ileal trichobezoar; case 1



Figure 7: CT findings; case 3. CT: Computed tomography



Figure 8: Ovarian cyst; operative finding of case 3

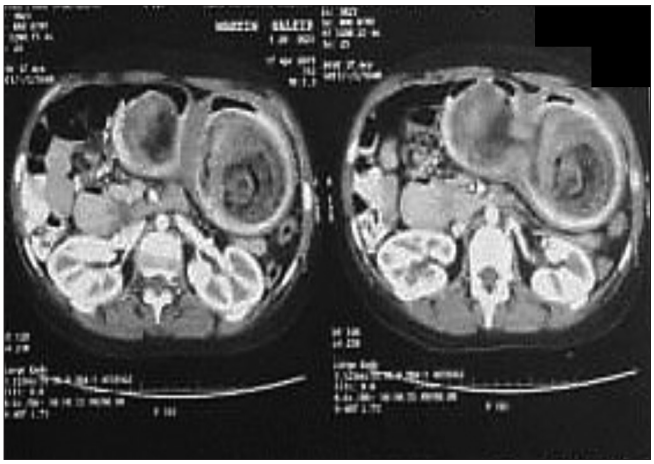


Figure 9: CT finding; Case 4. CT: Computed tomography

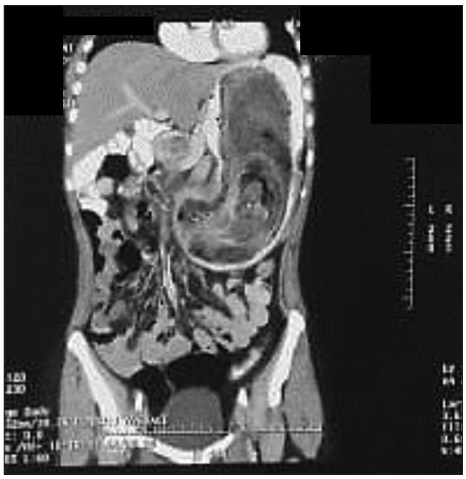


Figure 10: CT finding; Case 4. CT: Computed tomography

In one patient, endoscopic retrieval of the trichobezoar was attempted before 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 and then retrieve ileal trichobezoar [Figure 6]. All patients recovered without complications except the case that was done laparoscopically showed umbilical wound infection in the follow-up visit.

Table 1: Summary of five 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 abdominal 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 anemia	
Abdominal X-ray	Air fluid levels	Yes	Yes	Yes	Yes
Ultrasound results	Bowel dilatation	Free fluid	Echogenic mass in the stomach suspicious of tricobezoar	Echogenic mass suspicious of tricobezoar	Echogenic mass suspicious of tricobezoar
CT abdomen results	No	Trichobezoar In the stomach	Trichobezoar in the stomach and colon Right ovarian cyst 5 cm [Figure 7]	Tricobezoar in the stomach and 1st part of duodenum (rapunzel) [Figure 9]	Tricobezoar in the stomach and 2nd part of duodenum (rapunzel) [Figure 10]
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 Gastrotomy	Laparotomy Gastrotomy Colotomy	Laparotomy Gastrotomy	Laparotomy Gastrotomy
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 [Figure 8]	No	No
Size	6 cm × 5 cm × 3 cm	10 cm × 6 cm × 4 cm	11 cm × 7 cm × 4 cm 8 cm × 5 cm × 3 cm	15 cm × 8 cm × 5 cm	25 cm × 12 cm × 8 cm
Postoperative complications	Port site infection	No	No	No	No
Recurrences	No	No	No	No	No
Additional psychiatric consultation	Yes	Yes	Yes	Yes	Yes

CRP: C-reactive protein, TLC: Total leukocyte count

All cases were reviewed by a psychiatrist for further management.

Discussion

The origin of bezoar is either from the Arabic term 'badzehr' or possibly the Persian word 'padzahr', both meanings to expel poison or antidote. These words were described as 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'.[18,19]

Our 6-year retrospective review showed that surgical treatment for GIT bezoars is rare. As regards 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*.[20] reported four cases of gastric bezoars were identified over 18 years and Fallon *et al*.[21] identified seven cases that occurred over 8 years. However, we should suspect of gastrointestinal bezoars as the aetiology of abdominal cramps, vomiting and loss of weight or abdominal mass despite of its rarity.[22] We found an epigastric mass in two patients in addition to a mass in the left hypochondrium in one case, and this finding in patients with the abovementioned symptoms should raise the suspicion of gastrointestinal bezoar. In girls, a history of trichotillomania and trichophagia can

lead to the correct diagnosis and workup. In addition, 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 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 the removal of gastric bezoars, whereas reports of unsuccessful endoscopic attempts are more common.[23-24] Moreover, attempted endoscopic fragmentation might lead to complications such as gastric and oesophageal perforations[25-27] and distal 'embolization' of fragments causing bowel obstruction.[28]

Our attempt at endoscopic retrieval failed, whereas in the series reported by Gorter *et al*., endoscopy was attempted in two of four cases and failed in both.[20] 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 the gastric perforation is suspected.

Surgical removal is generally accomplished through laparotomy then gastrotomy or enterotomy with the removal of the bezoar.[28] The Rapunzel syndrome is defined as an extension of the bezoar beyond the pylorus,[18] and it can generally be removed through gastrotomy only. However, one should be aware of possible small bowel obstruction due to fragmentation of the bezoar. Therefore, extracting the bezoar through the gastrostomy should be done very carefully.

Gastrotomy and colotomy in our series were performed through 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,[29] and then more cases were also illustrated, especially in adults and adolescents. Cintolo *et al*.[30] mentioned the success of laparoscopical removal of 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 *et al*.[20] 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; however, it is recently reported the success of laparoscopic-assisted removal of gastric trichobezoars in five patients. The technique involves temporary gastrocutaneopexy, with or without the use of Alexis wound protector.[31,32]

The outcome expected after surgical removal through 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 in one case had port site infection. The reported rate of postoperative complications in the literature according to the review by Gorter *et al*.[20] 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 *et al.*,[21] the rate of postoperative complications was also similar –1 (14%) case of wound infection.[21]

Conclusions

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 upper gastrointestinal tract contrast, ultrasound or CT.

Endoscopic retrieval could be tried in selected cases despite the preference for 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 which has been reported to be 14% in the literature.

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Conflicts of interest

There are no conflicts of interest.

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