

Gastric Trichobezoar in the Absence of Psychosocial Stressors: a Report of Two Cases

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Abstract

Bezoars are accumulations in the gastrointestinal tract caused by intentional or unintentional consumption of indigestible materials. After food and mucus impaction they become solid masses with different sizes, presenting with various non-specific symptoms. Current treatment options for bezoars include enzymatic dissolution, endoscopic fragmentation and removal, and surgical removal.

Herein, we report two cases with an abdominal trichobezoar due to trichophagia presenting with mild atypical symptoms and requiring surgical removal. The patients had no underlying psychological and behavioral deviations, psychiatric disorders, stressful or traumatic life events that may trigger the condition. We present our approach to making the diagnosis, the challenges we met, and our treatment strategy.

Trichobezoar should not be forgotten as part of the differential diagnosis of abdominal pain and abdominal mass in children and adolescent.

Keywords

bezoar, endoscopy, gastric surgery, pediatric gastroenterology, pediatric gastroscopy, trichobezoar, pediatric surgery

INTRODUCTION

Bezoars are accumulations in the gastrointestinal tract caused by intentional or unintentional consumption of indigestible materials. Indigestible materials may include vegetable or fruit fibers (phytobezoar), hair or fur (trichobezoar), undissolved medications (pharmacobezoar), milk protein and mucus (lactobezoar) and foreign body bezoars such as parasitic worms.^[1] After food and mucus impaction the initial small accumulations become solid masses with different sizes, presenting with various non-specific symptoms. Bezoars may be formed in any

segment of the gastrointestinal tract. However, the stomach is the most common organ of bezoar formation. Most bezoars in children are trichobezoars from swallowed hair.^[2] The accumulated hair within the stomach is resistant to digestion and may extend beyond the stomach. A rare condition when a gastric trichobezoar extends to or beyond the ileocecal valve, causing a gastric outlet or an intestinal obstruction is the Rapunzel syndrome.^[2,3]

We report 2 cases of an abdominal trichobezoar due to trichophagia, presenting with mild non-specific symptoms and treated surgically. The peculiarity of our cases is the absence of underlying psychiatric disorders and psychoso-

cial stressors. We present the difficulties and the challenges we met, our diagnostic algorithm and treatment strategy.

CASE REPORT

Case 1

A 5-year-old girl presented to our pediatric gastroenterology department with a history of persistent mild epigastric pain without heartburn, nausea, vomiting or loss of appetite. Acute gastritis and gastroesophageal reflux were initially suspected, while they were not supported by the symptoms and therapy history. Besides a family history of stomach ulcer, there was no relevant family history of gastrointestinal illnesses or psychiatric conditions.

The patient appeared constitutionally well and had normal vital signs. Her abdomen was soft but there was a mild tenderness in the left upper quadrant without peritonism or a sign of a rebound. We performed an abdominal ultrasound that demonstrated a hyperechogenic abdominal mass with posterior acoustic shadow, located in the left upper quadrant (Fig. 1). Our initial diagnosis was a fecal impaction, and we



Figure 1. An ultrasound of the bezoar in the gastric outlet. An arc-shaped intraluminal mass with an acoustic shadow.

prescribed a laxative (PEG 3350) to the patient for several days. Five days later there was no change in the ultrasound findings, and we considered further diagnostic investigations.

A computed tomography (CT) scan of the abdomen was performed to provide more detailed information about the structure and the anatomy of the detected abdominal formation. It revealed a heterogeneous mass in the stomach, with fluid and gas (Fig. 2). The next step of our diagnostic algorithm was to do an upper gastrointestinal endoscopy. It demonstrated a large thick accumulation composed of hair bundles within the stomach cavity – a trichobezoar (Fig. 3). An endoscopic removal was not possible because of the mass' large size.

Upon closer questioning of the patient's family after the endoscopy, it was revealed that the patient had had the habit of eating her hair since she was 4 years old. In addition, the patient's grandmother reported that sometimes she had found hairs in the stool of her granddaughter. However, by the initial physical examination no signs of alopecia were found.

The patient was referred for psychiatric consultation and assessment. The results were consistent with a healthy 5-year-old child and highlighted no areas of concern. They showed normal childhood development milestones, a normal IQ score, absence of depressive symptoms and no history of abuse or other traumatic events.



Figure 2. The abdominal computed tomography revealed a heterogeneous mass in the stomach cavity.

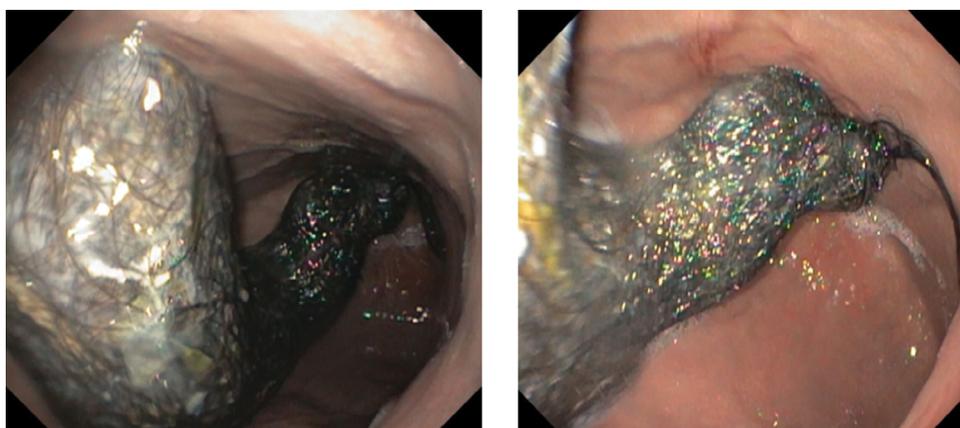


Figure 3. Upper gastrointestinal endoscopy confirming the diagnosis and showing a moderate luminal blockage.

The laboratory evaluations revealed a normal complete blood count and no abnormalities in the peripheral blood smear. Other laboratory findings, including C-reactive protein, electrolytes, liver function tests, and renal function tests, were also within normal limits.

Based on the clinical, imaging, and endoscopic outcomes, our final diagnosis was an abdominal trichobezoar due to trichophagia. The differential diagnosis included a non-Hodgkin lymphoma, neuroblastoma, and carcinoma of the stomach. Other possible conditions such as postprandial food and intramural mass were also considered and ruled out within the diagnostic process.

Since the bezoar had a large size and the endoscopic removal was not possible, we decided to remove it surgically (Fig. 4). The patient underwent a gastrotomy and the bezoar was extracted without complications. The postoperative period was uneventful. A routine followup check-up 2 weeks after surgery revealed a patient without gastrointestinal complaints, with normal defecation and progression of recovery.

Case 2

A 15-year-old girl presented to our pediatric gastroenterology department with a 10-day history of mild to moderate abdominal pain. The family had been visiting a general practitioner due to the girl's complains, but nothing abnormal had

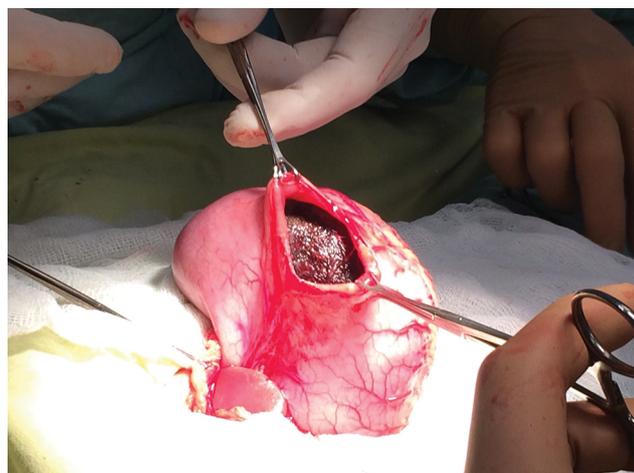


Figure 4. Surgical extraction of the trichobezoar in Case 1.

been found at the physical examination. The mother of the patient described darker stools in the last several days, since the abdominal pain started. Her father indicated that she had a habit of hair pulling and eating for many years. In addition, the patient reported to have fatigue and general weakness. There was no history of other gastrointestinal illness and no relevant family history of medical or psychiatric conditions.

The general physical examination revealed a well-nourished and weighed girl with extremely pale skin and mucous membranes. The abdominal examination found a solid palpable mass in the epigastrium. The initial laboratory evaluations showed a severe iron-deficiency anemia (Hgb 33 g/l, RBC $2.72 \times 10^{12}/l$, Hct 0.13 l/l, MCV 49 fl). The findings of microcytic anemia, low serum iron and blood urea excluded acute hemorrhage as a possible cause for the anemia.

Other laboratory findings, including electrolytes, inflammatory markers, liver function tests, were within normal ranges. The hematological parameters and part of the biochemical values of our patient are presented in Table 1. Three hemotransfusions were needed to achieve the normal hemoglobin levels for the age and sex.

As a next step in our diagnostic process, we performed some diagnostic imaging. The abdominal ultrasound and the CT scan demonstrated a large heterogeneous mass filling the stomach cavity from the fundus to the pylorus and extending to the duodenum (Fig. 5).

Having in mind the given anamnestic data for trichophagia, the clinical and the radiographic findings, we suspected a trichobezoar and our patient was referred to the Department of Pediatric Surgery for operative treatment - surgical extraction of the bezoar. The postoperative course was uneventful, and the girl was discharged with normal defecation, normal hemoglobin levels and normal progression of recovery. During the six-month follow-up period post discharge the hemoglobin levels remained within the reference ranges.

DISCUSSION

Bezoars are aggregates of inedible or undigested materials located within the gastrointestinal tract, usually in the stomach. The exact mechanism for their formation is unknown. Different theories suggest that it initially starts with

Table 1. Complete blood count and blood biochemistry of case 2

| | 14.11.2019 | 15.11.2019 | 16.11.2019 | 18.11.2019 |
|----------------------------|------------|------------|------------|------------|
| Hgb (g/l) | 28.0 | 54 | 98 | 121 |
| RBC ($10^{12}/l$) | 2.57 | 3.23 | 4.89 | 5.84 |
| Hct (l/l) | 0.123 | 0.194 | 0.34 | 0.436 |
| MCV (fl) | 47.8 | 60.1 | 69.5 | 69.4 |
| Ret (%) | 2.02 | | | |
| Urea (mmol/l) | 2.8 | | | |
| Iron ($\mu\text{mol}/l$) | 1.3 | | | |
| TBIC ($\mu\text{mol}/l$) | 86.2 | | | |

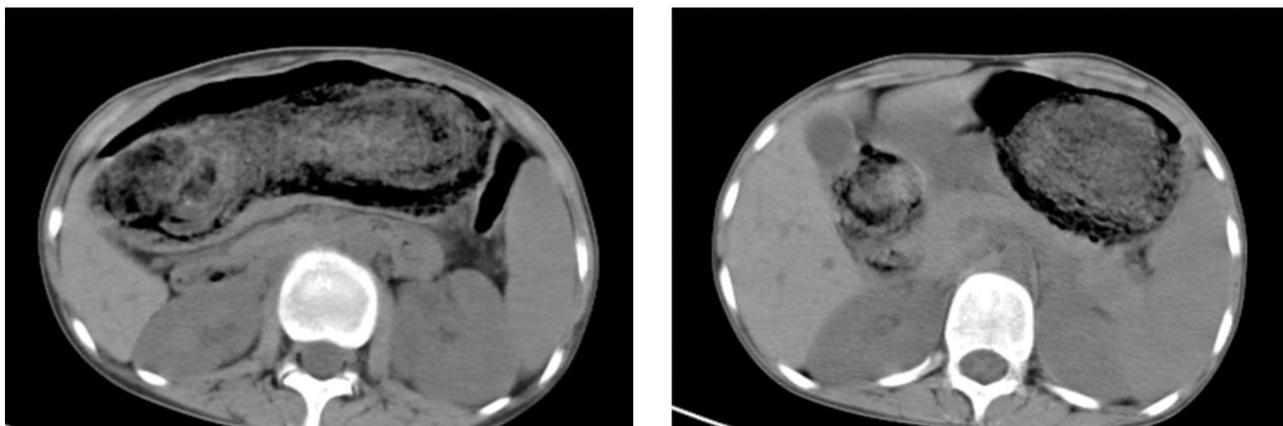


Figure 5. CT scan of abdomen revealed a heterogeneous mass in the stomach cavity and the duodenum.

the collection of trapped materials within gastric folds or that it is a consequence of gastrointestinal dysmotility or postoperative delayed gastric emptying.^[4,5]

Bezoars are initially asymptomatic until they grow to a relatively large size. Then they may produce a variety of symptoms such as pain, nausea, vomiting, delayed gastric emptying, and weight loss. Malabsorption of vitamins and of trace elements may cause fatigue and anemia, especially iron deficiency anemia.^[6,7] Other signs and complications of bezoars include bowel obstruction, gastric mucosal erosion, pressure necrosis, gastrointestinal perforation and bleeding, pancreatitis, intussusception, and peritonitis.^[3,8,9] A correct and timely diagnosis is the key to prevent the complications that sometimes may be fatal. However, it is usually missed or delayed as the condition is rare and asymptomatic in the initial stages.

Trichobezoar is an uncommon entity observed mostly in young female patients under the age of 20 and occurring most commonly in the stomach.^[10] The majority of the cases are linked to psychiatric conditions - trichophagia or trichotillomania^[11], but can also be found associated with a number of other conditions such as pica (psychological disorder characterized by tendency or craving to eat substances other than normal food), often caused by iron deficiency anemia, steatorrhea and protein-losing enteropathy.^[8,12] There are no signs and symptoms pathognomonic for trichobezoar. However, typical symptoms, which resulted in a trichobezoar diagnosis include epigastric pain, nausea, loss of appetite, and bowel or gastric outlet obstruction.^[13]

In our cases, the leading symptom reported from our patients was a mild to moderate epigastric pain. In the second case, it was accompanied by fatigue and general weakness due to the iron deficiency anemia. However, in both cases the symptoms were discrete and non-specific. During the physical examination, we discovered an abdominal mass and conducted the further investigations based on this finding. Our diagnostic algorithm was consistent with the standard diagnostic approach to trichobezoars that includes a detailed patient history, a brief family history, physical examination, laboratory tests, and radiological im-

aging - computer tomography scan, abdominal ultrasound, and upper gastrointestinal endoscopy.^[14]

The most common treatment option for trichobezoars is surgical removal by laparotomy. Although there are other possible treatments, such as endoscopic or laparoscopic removal, this method is favoured as it reduces the risk of fragmentation that may cause further bowel obstruction.

The treatment modality used in a specific case depends on the type of the bezoar. Trichobezoars are resistant to enzymatic degradation and pharmacotherapy. The enzymatic dissolution is a therapeutic option by phytobezoars. The trichophytobezoar's therapy typically involves size reduction by a sodium bicarbonate injection and then a segmental endoscopic retrieval. Pharmacobezoars, on the other hand, may dissolve spontaneously.^[15]

Upper gastrointestinal endoscopy is an important and powerful tool in the management of pediatric bezoars that can be both a diagnostic and a therapeutic procedure. In our first case, the bezoar was too hard and large, and the only possible treatment was surgical extraction by gastrotomy, but in other similar cases, the removal was carried out via endoscopy.^[16]

Essential in the therapy of the pediatric bezoars is the prevention of bezoar recurrence. Direct bezoar removal is only a part of it that does not cancel the necessity of an appropriate psychiatric counseling and treatment.^[13]

CONCLUSIONS

Trichobezoars are impactions of swallowed hairs usually found in patients with a history of psychological or psychiatric disorders, or another underlying medical conditions. Clinical manifestations vary from being asymptomatic to a full-blown acute abdomen, resulting in a comatose state. Treatment can be conservative, medical, or surgical. A recurrence is possible depending on the initial cause.

These cases highlight the fact that not all trichophagia-caused bezoars can be associated with psychological disorders, mental retardation or a behavioural disorder. Unfortunately, the exact causes of their occurrence in our

patients remained unknown. Although trichobezoars are rare conditions, they should not be forgotten as part of the differential diagnosis in a patient presenting with nonspecific abdominal pain and abdominal mass.

Author contributions

I.Y. managed the patient in the Department of Pediatrics, performed the ultrasound and endoscopy procedures, wrote the manuscript, contributed to the ultrasound and endoscopy images. V.T. managed the patient in the Department of Pediatric Surgery, performed the operation, contributed to the intraoperative images. D.M. performed the operation as vice-operator. P.K. performed the CT-scan and reviewed pictures from all imaging studies. R.S. reviewed and participated in writing the manuscript. D.M. managed the patient in the Department of Pediatric Surgery, performed the operation as a second operator. N.B. reviewed the images from the endoscopy procedure.

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Competing Interests

The authors have declared that no competing interests exist.

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Желудочный трихобезоар в отсутствие психосоциальных стрессоров: отчёт о двух случаях

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Резюме

Безоары представляют собой скопления в желудочно-кишечном тракте, вызванные преднамеренным или непреднамеренным потреблением неперевариваемых материалов. После попадания пищи и слизи они превращаются в твёрдые массы разной величины, проявляющиеся различными неспецифическими симптомами. Текущие варианты лечения безоаров включают ферментативное растворение, эндоскопическую фрагментацию и удаление, а также хирургическое удаление.

Здесь мы сообщаем о двух случаях абдоминального трихобезоара из-за трихофагии, проявляющейся лёгкими атипичными симптомами и требующей хирургического удаления. У пациентов не было сопутствующих психологических и поведенческих отклонений, психических расстройств, стрессовых или травмирующих жизненных событий, которые могли бы спровоцировать данное состояние. Мы представляем наш подход к постановке диагноза, трудности, с которыми мы столкнулись, и нашу стратегию лечения.

Не следует забывать о трихобезоаре как части дифференциальной диагностики болей в животе и новообразований в животе у детей и подростков.

Ключевые слова

безоар, эндоскопия, хирургия желудка, детская гастроэнтерология, детская гастроскопия, трихобезоар, детская хирургия
