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Trichobezoar presenting as a gastric outlet obstruction: A case report

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ABSTRACT

INTRODUCTION: Rapunzel syndrome is a rare intestinal condition that starts with the ingestion of a trichobezoar. The condition is predominately found in females and can be associated with trichotillomania, or the compulsive urge to pull one's own hair out. There are less than 40 cases described in the literature with the prevention of recurrence aimed at psychological treatment.
PRESENTATION OF CASE: The patient is a 7 year-old girl with a history of trichotillomania with trichophagia as a young child who presented with abdominal pain, nausea, and vomiting, consistent with a gastric outlet obstruction. She had an exploratory laparotomy with gastrostomy performed revealing a 18 cm by 18 cm trichobezoar with extension into the small bowel.
DISCUSSION: Bezoars, an already rare entity, can occasionally lead to gastric and small bowel obstructions. Small collections of ingested hair build up in the intestinal tract causing significant symptoms. These obstructions can sometimes be treated through minimally invasive techniques but, in our case described, it is unlikely to have been treated any other way due to the substantial size of the trichobezoar.
CONCLUSION: Early consideration of Rapunzel syndrome is important in young females presenting with a gastric outlet obstruction.

1. Introduction

Rapunzel syndrome is a rare intestinal condition that starts with ingestion of a trichobezoar. The syndrome is predominately found in females and can be associated with trichotillomania (the compulsive urge to pull one's own hair out). Many patients who present with this syndrome have other significant mental or behavioral disorders including neglect, family discord, and bereavement [1,2]. Rapunzel syndrome, first documented in 1968, can be described as a trichobezoar starting in the stomach and extending into the small intestine. Patients can present with a wide variety of symptoms, ranging from early satiety to peritonitis. Diagnosis can be made with computed tomography (CT) or esophagogastroduodenoscopy (EGD). The treatment of this disease is exclusively surgical when causing a gastric outlet obstruction. These trichobezoars are indigestible and removal through a gastrotomy is recommended. Prevention of recurrence is intimately linked with psychological treatment. The syndrome is considered to be the most severe form of trichophagia, with fewer than 40 cases described in the literature. Although most cases of a Rapunzel syndrome are present in toddlers, adolescents, or young adults, recent literature indicates that the prevalence in adulthood is increasing [3].

2. Presentation of case

The patient is a 7-year-old girl with a history of trichotillomania with trichophagia as a young child. The parents were aware, but denied observation of this behavior after the age of four. She presented to the emergency department with symptoms of abdominal pain, nausea, and vomiting, consistent with a gastric outlet obstruction. On examination, her abdomen was firm but nontender, with a large mass palpated in the epigastrium. An abdominal x-ray was performed, showing a large gastric mass, concerning for a trichobezoar [Fig. 1]. The patient was taken to the operating room where she underwent an exploratory laparotomy with gastrostomy (Figs. 2 and 3). During the procedure, an 18 cm × 18 cm trichobezoar with extension into the small bowel was found [Figs. 4 and 5]. Postoperatively the patient recovered without complication and was discharged home safely.

3. Discussion

Bezoars are a rare and fascinating entity that can cause gastric and small bowel obstructions. Several types have been reported, including phytobezoars (plant material), trichobezoars (hair fibers), and lactobezoars (milk). Trichobezoars are most often reported in young females with histories of emotional trauma or psychological disorders. These "small" collections of hair build up in the intestinal tract, most often the stomach, and can cause a significant amount

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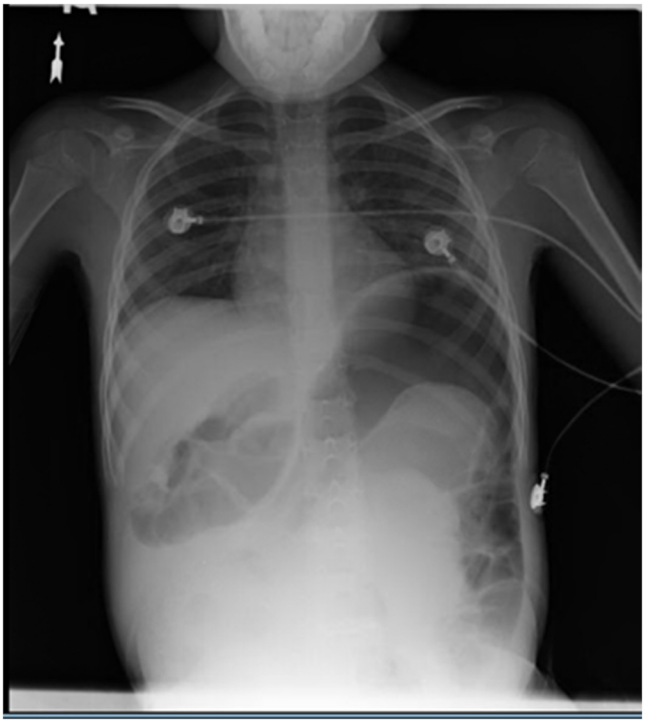

Fig. 1. Upright X-ray of patient presenting with Trichobezoar.

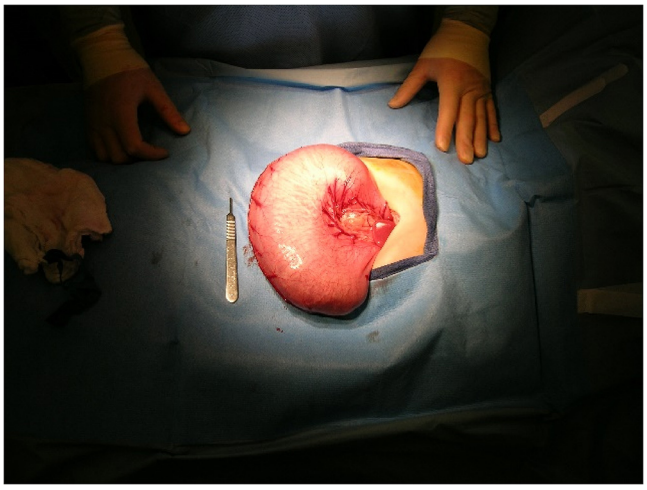

Fig. 2. Extracorporeal stomach filled with trichobezoar (continued).

of symptoms. Recently, the literature indicates that trichobezoars could be the rare cause of jejunal intussusception, appendicitis, nephrotic syndrome, and biliary obstruction [4–8]. Most patients present with abdominal pain, nausea, and vomiting and, therefore, it is an important diagnosis to consider in this setting. They can even rarely cause peritonitis and perforation [2,9–14]. The clinical scenario is most common in teenage girls with the diagnosis in a child very rare.

A majority of the literature describes minimally invasive techniques for trichobezoar removal. Other treatments described include laparotomy, laparoscopy-assisted, or endoscopy [15,16]. In one review, it was shown that there was a 75% success rate for laparoscopic treatment versus a 99% success rate with laparotomy. Although success is greater in laparotomy, there is a higher chance of complications. As a surgeon faced with this rare entity, it is important to take into consideration the clinical status of the patient and severity of the obstruction when choosing which ther-

Fig. 3. Gastrotomy with trichobezoar.

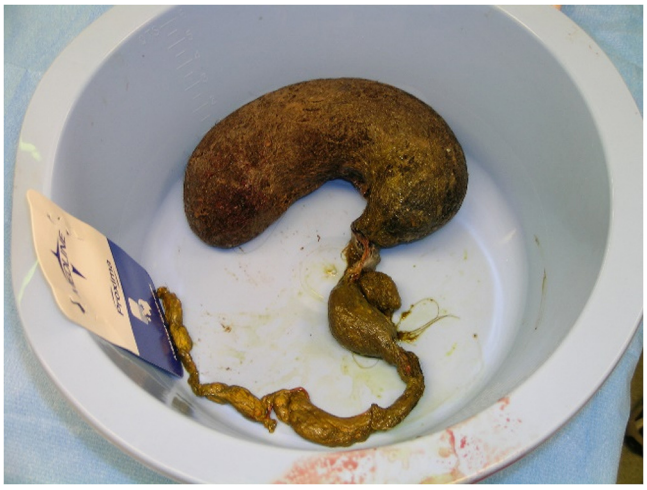

Fig. 4. Trichobezoar specimen with tail removed from duodenum.

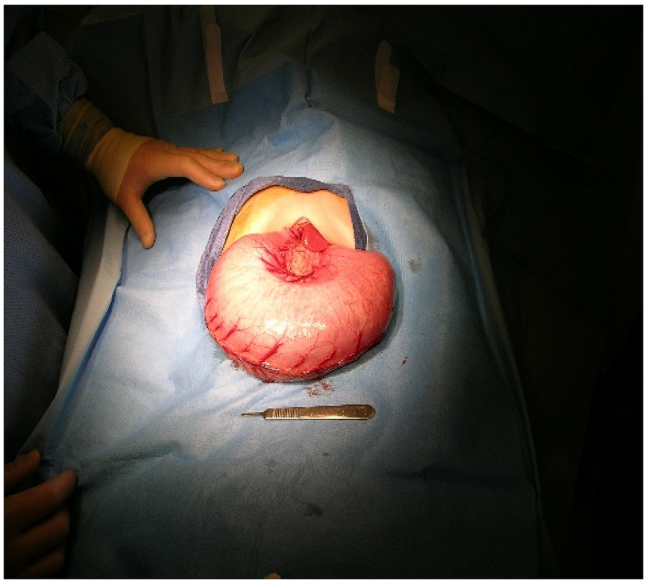

Fig. 5. Extracorporeal stomach filled with trichobezoar.

apeutic modality. In a patient who only has a partially obstructing bezoar, the benefit of laparoscopy is clear. Given the clinical severity of the gastric outlet obstruction as well as the size of the bezoar, it is unlikely that treatment with laparoscopy would have been successful in the case presented.

Rapunzel syndrome has been clearly described in the literature. The existing information on the topic has a lack of intra-operative photographs and diagnostic abdominal x-rays, which we have provided in the text. It is important to learn that in the given clinical scenario, especially in a pediatric patient, ingested materials including hair should be carefully considered. As hair is indigestible, lack of consideration of the diagnosis could potentially delay life saving surgical intervention.

4. Conclusions

Trichobezoar is a rare entity that should be carefully considered in young female patients presenting with abdominal pain and symptoms of a gastric outlet obstruction. We hope that education on the topic and the clear intraoperative photographs presented remind surgeons to keep this on their differential diagnosis when faced with this clinical scenario. All work presented is in line with the SCARE criteria [17].

Conflicts of interest

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Ethical approval

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Consent

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Author contributions

Eugene Nwankwo – Author, data collection
Edward Daniele – Editor, contributor
Erin Woller – contributor
John Fitzwater – contributor
Thomas McGill – contributor
Steve E. Brooks – contributor

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