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Trichobezoar - A hair-raising surgical emergency

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CASE STUDY

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ABSTRACT

A trichobezoar is a mass of undigested hair in the proximal gastrointestinal tract. It is a rare condition seen in young females with psychiatric disorders of trichotillomania and trichophagia or mental retardation. We present a case of an intellectually impaired 14-year-old girl with a gastric trichobezoar discovered during an emergency laparotomy. We will discuss the limited but salient findings on history and examination. Through a review of the literature, imaging modalities and treatment options for the early versus late stage of trichobezoar presentations will be identified. Through our case, subsequent holistic management will also be highlighted to prevent recurrence.

Key Words
Trichobezoar, laparotomy, perforation

Implications for Practice:

1. What is known about this subject?
Trichobezoars are commonly associated with adolescent females suffering from trichotillomania and trichophagia. In late presentations such as perforation, they require explorative laparotomy.

2. What new information is offered in this case study?
Trichobezoars of this size have rarely been reported in the literature. The subsequent holistic management is essential in both post-surgical recovery and preventing recurrence.

3. What are the implications for research, policy, or practice?
The differential of trichobezoars should be considered in patients with recurrent abdominal pain. Complications often necessitate operative management; however early presentation and accurate diagnosis could prevent surgery.

Background

Trichobezoars are the second most common type of bezoar, its prevalence rates ranging from 0.06 per cent to 4 per cent in abdominal pain presentations.[1] With more advanced imaging modalities over the past decades, the diagnostic rate of trichobezoars has increased[1]. However, we suspect awareness of the condition is still limited.

It is important for clinicians to recognise the associated psychiatric behaviours causing trichobezoars and subsequently linking the patient to psychiatric resources. This will prevent further progression and recurrence of the disease.[2]

Given the dire complications associated with trichobezoars, early recognition of this condition is critical. This will prevent prolonged recovery post-surgery for perforation complications and/or death.

Other than a detailed history and examination, choosing the appropriate imaging to investigate and provide therapeutic management according to the stage of presentation is important. In the early stages of a trichobezoar; endoscopy and laparoscopic removal has some benefit. However, in the late stage, proceeding to laparotomy may be the only course of action.

Hence awareness of the differential of trichobezoars will enable the surgeon to take a detailed holistic history regarding trichotillomania/trichophagia, as well as conduct tests and procedures according to clinical severity of trichobezoar in a hospital versus clinical setting.

Case details

A 14-year-old girl was brought in to the emergency department by ambulance with epigastric pain and a rigid distended abdomen. Her mother reported that she has been unwell for the past five months with early satiety and had loss of appetite in the past week. She had not opened her bowels for the past five days. She was otherwise healthy with a mild cognitive impairment. She showed signs of shock and peritonitis. While being resuscitated, she had an erect chest x-ray, which demonstrated a large amount of free gas under the diaphragm. Based on these clinical findings, a gastrointestinal tract perforation was suspected, and the patient was consented for an emergency explorative laparotomy.

Intra-operatively we found a 2cm ulcer in the greater curvature of the stomach with hair visible, and a distended, rigid stomach. The ulcer was incised and a large trichobezoar of 24×12×9cm was removed (Figure 1). It did not extend past the pylorus. The gastrostomy was closed with interrupted sutures. The patient recovered slowly due to her poor pre-operative nutritional status. Her inpatient stay was complicated by recurrent wound infections. It was noticed that as she was waking up from the anaesthetic, she reached for her hair and pulled it into her mouth. Inpatient psychiatric input was required to counsel her. After consultation with her mother, a hair cut was performed to a length that she was no longer able to grip onto. Three weeks post-surgery, she was discharged home with outpatient psychiatric reviews.

Discussion

First reported by Baudamant in a human autopsy case in 1779,[2] Trichobezoars are the second most common type of bezoar, often associated with trichotillomania (hair pulling), and trichophagia (hair swallowing). This impulse control disorder is almost exclusively seen in adolescent females, of which 1 per cent then go on to develop trichobezoars.[2] The parents of our intellectually impaired patient did not report this behaviour pattern. However immediately post-operatively, she was observed reaching for her hair.

Human hair is indigestible due to its smooth slippery surface and enzyme-resistant properties. It escapes peristalsis and is trapped in the gastric mucosal folds.[2] Continuous ingestion of hair mixed with food, results in the formation of a black trichobezoar; the hair's proteins denatured to a black colour by the stomach's acidic environment. Trichobezoars are usually located in the stomach, rarely extending past the pylorus. The phenomenon of the trichobezoar extending into the jejunum, ileum or even colon, was first described in 1968 by Vaughan et al. as Rapunzel syndrome.[3]

Patients can present with non-specific symptoms ranging from epigastric pain (70.2 per cent), nausea, vomiting (64 per cent), haematemesis (61 per cent) and weight loss (38 per cent).[2] This vague presentation often creates a diagnostic challenge, resulting in delayed treatment and subsequent disease progression/ complications. 70 per cent of cases in the Debakey et al. series also had a mobile epigastric mass on examination.[4] Our patient reported a five-month history of the above symptoms in a mild form as well as halitosis and early satiety. As her trichobezoar was massive (24×12×9cm), we were not able to clearly differentiate the palpable epigastric mass from voluntary guarding.

As trichobezoars increase in size, the risks of complications such as gastric/intestinal obstruction and perforation are higher. In our case, it is likely the reduced blood supply to the gastric mucosa that led to erosion and subsequent perforation. Infrequently intussusception, obstructive jaundice, pancreatitis and even death have been reported.[4]

Blood tests usually reveal an iron deficiency anaemia.[3] Diagnostic modalities in a non-acute setting are ultrasonography, contrast radiography and upper gastrointestinal endoscopy. Endoscopy is considered the gold standard for diagnosis as it allows for direct visualization and biopsies to determine the composition of the bezoar and subsequent treatment. Phytobezoars and lactobezoars can be treated endoscopically.[5] Computed tomography (CT) scan is commonly used in the acute setting to identify the location of obstruction, also showing a well-circumscribed intraluminal mass with interspersed gas.[6]

A review of 108 trichobezoar cases by Gorter et al.[7] evaluated the treatment options of endoscopic versus laparoscopic removal versus laparotomy. Five per cent of attempted endoscopic removals were successful, compared to 75 per cent success rate of attempted laparoscopies. Small trichobezoars can be removed in pieces via endoscopy, but bore the risk of esophagitis with multiple scope insertions.[7] Given our patient's peritonitic signs and trichobezoar size, endoscopy would not have been a feasible or safe treatment option. Fragments could also migrate distally and cause a bowel obstruction. Laparoscopy had a risk of exposing contaminated hair fragments in the abdominal cavity. Gorter et al. concluded by recommending laparotomy with its 100 per cent success rate in 100 patients, and advantage of short amount of operating time

in examining the entire gastrointestinal tract for residual fragments.[7] Post laparotomy, our patient also underwent a successful albeit slow recovery.

The success rates of enzymatic dissolution and extracorporeal shock wave fragmentation are yet to be defined in the literature and conventional laparotomy remains the gold standard.[8] Given our patient's presentation with shock and peritonitis, we bypassed further imaging and proceeded to a laparotomy as an erect chest X-ray had demonstrated free gas under the diaphragm. In addition to a short haircut, our patient underwent psychiatric consultation for behavioural therapy. This was essential to prevent recurrence of her condition.[5]

The recurrence rate of trichobezoars in patients who underwent surgery is unknown.[9] Seven case reports have been published to date,[9-15] all concluded that post-surgery psychiatric consultation is integral to prevent recurrence. Majority of these patients have psychiatric disorders such as anxiety, depression, anorexia nervosa and obsessive compulsive disorder.[9-15] Trichotillomania/trichophagia has links to childhood neglect, mental retardation, and bereavement, hence parental counselling is useful.[16] A 1989 double-blind trial comparing clomipramine (serotonin reuptake blocker) and desipramine (tricyclic antidepressant) found clomipramine more effective in decreasing the intensity of compulsion in trichotillomania.[16] Due to lack of evidence for one particular treatment over the other, there are no clear management guidelines.[17] Psychotherapy and cognitive behavioural therapy for habit-reversal techniques are often used initially due to its lack of side effects; with subsequent addition of selective serotonin reuptake inhibitors (fluoxetine) or clomipramine if comorbid psychiatric disorders are present.[17] Thus, we feel that the risk of recurrence is reduced if compliance to psychiatric reviews and treatment is achieved.

Conclusion

The differential of trichobezoars should be considered in young females with chronic and recurrent abdominal pain. Endoscopy is the best diagnostic modality, with limited therapeutic potential in the early stage. The role of laparoscopy and other novel options are still debated in the literature, with laparotomy still the treatment of choice in large trichobezoars and presence of complications. Of course, psychiatric counselling and treatment is essential for underlying psychiatric disorders so as to prevent relapse.

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PEER REVIEW

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CONFLICTS OF INTEREST

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PATIENT CONSENT

The authors, *Tan E, Durgakeri P, Penington B,* declare that:

1. They have obtained written, informed consent for the publication of the details relating to the patient(s) in this report.
2. All possible steps have been taken to safeguard the identity of the patient(s).
3. This submission is compliant with the requirements of local research ethics committees.

Figure 1: Demonstrates the large Trichobezoar extracted from the stomach. Note how it has taken up the shape of the stomach