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Case Report

A case of contained jejunal perforation secondary to trichobezoar in a 65-year-old woman

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Abstract

Bezoars are conglomerations of undigested foreign material retained in the gastrointestinal tract. Trichobezoars consist of swallowed hair and constitute less than 6% of all bezoars. We present a case of jejunal trichobezoar with an intra-abdominal mass and contained jejunal perforation with a localized abscess in a 65-year-old female patient. She was scanned with contrast-enhanced abdominal CT. The patient underwent exploratory laparotomy with removal of the mass, abscess drainage, and jejunal resection with end-to-end anastomosis. Trichobezoars should be considered in the differential diagnosis of an intra-abdominal mass in older patients like ours. We are reporting this case as trichobezoar is a rare condition and it is even rarer in a 65-year-old woman without an underlying risk factor. So, we hope this will create some awareness among physicians of the condition.

Keywords: Bezoar, Trichobezoar, Rapunzel syndrome

Introduction

A bezoar is an accumulation of indigestible foreign material in the gastrointestinal tract (1). The term bezoar derives from the Arabic word "badzehr," which means antidote. Physicians used bezoars as antidotes against plague, snakebite, leprosy, and epilepsy between the years 1200 and 1800 (2, 3). Because of the stomach's large capacity, gastric bezoars do not become symptomatic until they are huge. "The masses are classified according to their contents. Phytobezoars include fruit fibers or plants, lactobezoars are composed of milk, trichobezoars are concretions of hair, and pharmacobezoars are composed of medications" (4). In humans, the most common type of bezoar is the trichobezoar, which is mostly made of hair. The prevalence rate varied from 0.06% to 4% (5). Trichobezoars are not associated with alterations in GI motility but are more common in patients with underlying psychiatric disorders. They often present in adolescence and during the second decade of life (6). The source of hair contributing to bezoar formation is usually the patient's own scalp hair, but it can come from the eyelashes, eyebrows, and pubic hair (3).

Trichobezoars are rare conditions that consist of hair bundles in the stomach or small bowel. In young women, trichobezoars are associated with psychiatric disorders such as trichotillomania (hair-pulling) and trichophagia (hair swallowing) (1). In adults, bezoars most often develop after gastric operations that alter the motility, emptying, and grinding of food in the stomach (7). Although the prevalence of bezoars in humans is low the associated mortality rates may be as high as 30% if left untreated, primarily due to gastrointestinal bleeding, destruction, or perforation (2). Human hair is indigestible and peristaltic movement resistant because of its smoothness. Continuous ingestion of hair, over a while, can lead to their impaction along with mucus and food material in the stomach. In some cases, however, the trichobezoar extends through the pylorus into the jejunum, ileum or even colon. This condition, known as Rapunzel syndrome, was initially described in 1968 by Vaughan et al. (8). Rapunzel was a long-haired female in a German fairy tale by the Grimm brothers (9). We are reporting this case as trichobezoar is a rare condition and it is even rarer in a 65-year-old woman without an underlying risk factor. We hope this will create some awareness among physicians of the condition.

Case Description

This is a 65-year-old female patient who presented with abdominal swelling and fever of 10 days duration. She came from the rural part of the southern Ethiopia. She was referred from a private hospital and admitted to the surgical emergency OPD with the impression of acute abdomen with an abdominal wall abscess. Her attendants claim that the swelling over the left side of the abdomen has been there for the last 7 months but had increased since the past month and for the last 10 days it was associated with abdominal pain and fever. She also had associated constipation but had no failure to pass feces or flatus and no episodes of vomiting. She had no history of abdominal trauma or abdominal surgery. She had no clear sign of psychiatric illness and had never been diagnosed with any psychiatric condition. However, her relatives claim that she doesn't communicate with them very well. She denied hair pulling or swallowing.

She had no chronic medical illness like diabetes mellitus or hypertension. On physical examination, she was acutely sick looking with BP of 90/60, PR of 120bpm, RR of 30 breaths/minute and body temperature of 38 ^{0}C. Her BMI was 23 Kg/M^{2}. On HEENT, she had slightly pale conjunctiva and dry buccal mucosa; there were no hairless regions on her scalp. On abdominal examination, the abdomen moved with respiration, there was about 20 x 15 cm intra-abdominal mass in the left mid hemi abdomen, it was globular- hard at the periphery and fluctuant, hot and tender at the center. On musculoskeletal examination, she had no edema. The following laboratory data of the patient were obtained after laboratory investigation (Table 1.)

Table 1: Patient's laboratory examination results

CBC	Values
WBC	14000/µL with Neutrophil of 70%
Hgb	10.9 g/dL
MCV	82
MCH	26
MCHC	31
Platelet	420000/µL
Blood group & Rh	O^{+}
RFTs	Values
Serum creatinine	0.8
Serum BUN	14.5

The patient was referred from a private hospital with contrast-enhanced abdominal CT and did not undergo an abdominal ultrasound.

Contrast-enhanced abdominal CT findings: There was a well-defined rounded complex left side mid-level intra-abdominal mass grossly measuring 11.4 cm X 10.7 cm X 8 cm, with dense thick peripheral calcification and internal significant air loculation with dependent part hypo-dense fluid with air bubbles forming air-fluid level. The mass was adherent to the adjacent anterior abdominal wall on the left side, which was markedly thickened with rim enhancing hypo-dense intra-abdominal wall collections, surrounding edematous tissue, and fat stranding. The mass had also abutted and displaced regional bowel loops apart (see the scout film, Figures 1 2)

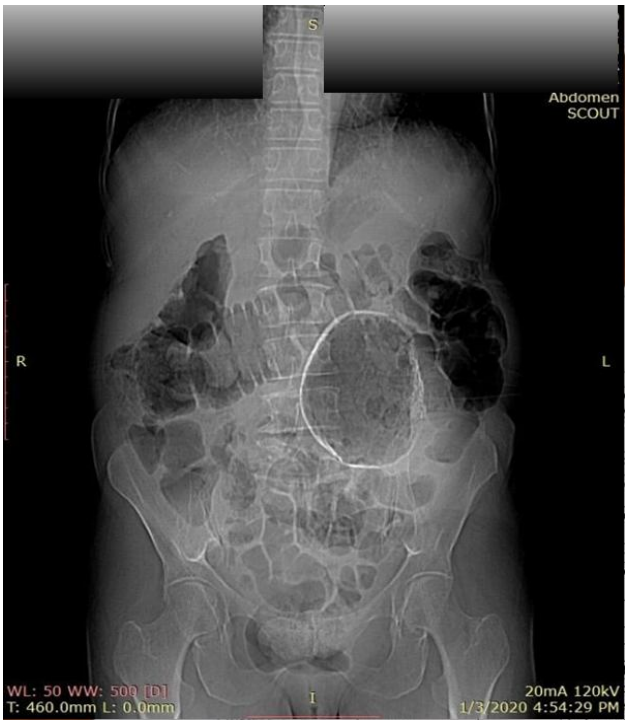

Figure 1: Patient's scout film of CT

Conclusion of CT findings: Well defined left midlevel intra-abdominal mass with an air-fluid level, peripheral wall dense calcification adherent to the left anterior abdominal wall with rim enhancing irregular abdominal wall significant collection/abscess, associated fat stranding, and tissue oedema.

With the above impression, the patient was resuscitated and as she produced adequate urine, she was taken to the OR for exploratory laparotomy. Under general anesthesia, the abdomen was cleaned and draped then entered through midline vertical incision.

Figure 2: Contrast-enhanced abdominal CT of the patient

Findings from exploratory laparotomy and procedures done

Findings: There was an intrabdominal mass in the left mid-level hemi abdomen, which had eroded the adjacent anterior abdominal wall with overlying intact skin. It was attached to the omentum, transverse colon, small bowel and small bowel mesentery, there was about 300 ml of pus localized to the left hemiabdomen. The stomach and the rest of the viscera appeared normal.

Procedure: The mass was mobilized from its attachments (see Figure 3). In doing so there was an existing contained jejunal perforation at its

mesenteric border about 70 cm from the ligamentum Treitz. The mass had a calcified shell with bilious matter inside (see Figure 4).

The abscess was drained; the shell was excised along with the perforated bowel. An end-to-end jejuno-jejunal anastomosis was created. Finally, we did thorough lavage with warm saline and closed the abdominal wall in layers. The patient was transferred to the recovery room with stable vital signs. The specimen was not subjected to Histopathological examination.

Figure 3: The intra-abdominal mass (intraoperative).

Post-op Progress: On the 3rd post op day, she started to desaturate and with the impression of hospital-acquired pneumonia she was treated with ceftazidime, vancomycin, and metronidazole and oxygen supplementation, for 18 days. And on the 5th post –op day, she began having purulent discharge from the incision site; with the impression of superficial surgical site infection, local wound care was started with dressing changes daily. Finally, she was discharged improved on her 24th -post-op day. The patient did not come for follow-up, even though she was appointed to both surgical clinic and psychiatric clinic. She is from a remote area and that may be the reason for not showing up.

Figure 4: A gross image of the excised mass/shell and ball of hair inside it (see also Figure 5).

Figure 5: The trichobezoar removed from the shell.

Discussion

Currently, bezoars are known to be harmful and should be removed (1). Affected patients often remain asymptomatic after several years. Symptoms develop as the bezoar increases in size to the point of obstruction (6).

Bezoars may present with abdominal pain, nausea, vomiting, early satiety, weight loss, intestinal obstruction and ulceration leading to bleeding and/or perforation. Rarely, intussusception can also occur (3,9). malabsorption–related complications include protein-losing enteropathy, iron deficiency, and megaloblastic anemia (6). Most cases of trichobezoar are reported in females, which may be attributed to the traditional long hair in females. Most cases present between age 13 and 20 years (6). Our patient, a 65-year-old female, presented with progressively increasing abdominal swelling with pain and fever. An upper abdominal mass remains the most common presenting sign (9). In this study, there was a palpable intrabdominal mass in the left mid hemi abdomen that was non-tender peripherally and warm and tender at the center.

Diagnostic modalities include US, CT scan and upper endoscopy. CT scans have a high accuracy rate, but the accuracy of US in such cases is not so high. The diagnosis is made easily at endoscopy or at times from a plain radiograph (10). The gold standard for diagnosis is upper gastrointestinal endoscopy. In addition to providing direct visualization, this procedure allows sample taking and potential therapeutic intervention (6). The most common diagnostic tool used in the literature is a CT scan, with a typical image showing a well-defined intraluminal ovoid heterogeneous mass with interspersed gas (11,12). In this study, contrast-enhanced abdominal CT was performed and the diagnosis was confirmed during exploratory laparotomy.

A firm or hard mass in the mid-upper abdomen is usually suggestive of a malignant process, particularly in adults and the elderly (9). The characteristics of the mass in the present case was initially concerning for cancer, but the intraoperative findings proved it to be a contained jejunal perforation with jejunal trichobezoar forming a calcified mass.

The management and treatment of a bezoar include the removal of the mass and prevention of recurrence by addressing the underlying physical or psychiatric causes (6). The method of bezoar removal depends on its component, size, and location. In phytobezoars, Coca-Cola irrigation or administration is effective for fragmentation (4). Trichobezoars, or hairballs, are not easily dissolved with pharmacotherapy or using endoscopic trials. Laparotomy has been the treatment of choice for large trichobezoars (4). Since the advent of minimally invasive surgery, surgeons now use laparoscopic techniques for small to moderate-sized bezoars (6). We did not find any reports of trichobezoar in Ethiopia.

There are few reports in Africa and throughout the world. However, given the age of the patient and the location of the trichobezoar, our case is quite unique.

Conclusion

Most of the reports of trichobezoars occur in adolescent females with an underlying psychiatric disorder, with the stomach being the most common site. Our case is unusual due to the patient's age and the site of her bezoar. There is no report of this condition in an elderly woman. We conclude that, although rare, trichobezoars should not be forgotten in the differential diagnosis even in an elderly patient like ours with no documented history of psychiatric condition and no history of hair swallowing. Management always requires surgical removal.

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Consent for publication

The patient consented to the publication of this case report.

Data availability

Not applicable.

Data Availability statement

Not applicable.

Conflict of interest

The authors declared no conflicts of interest exist.

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Authors' Contribution

ABA is the PI who took part in the entire process from the inception of the case report to its final write up. Additionally, he took part in the operation of the case. AYK took part in the operation and provided the patient's detailed history, investigation results.

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