



Trichobezoar – A Case Report

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Abstract

Trichobezoar is a term used to describe trapped hairball masses seen in gastrointestinal tract of, commonly, young adolescent females with underlying psychiatric illness. This is a case report on a 14-year-old Indian female who presented with abdominal pain and features of intestinal obstruction, diagnosed with trichobezoar on CT scan. She underwent laparotomy and removal of large gastric trichobezoar and satellite trichobezoar in jejunum. This report also includes a review of recent literature on investigations and treatment of trichobezoar and highlights the necessity of psychiatric evaluation.

Keyword: *Trichobezoar, trichotillomania, trichophagia, intestinal obstruction, Rapunzel syndrome, endoscopy, laparoscopy, open laparotomy in trichobezoar.*

Introduction

Trichobezoar is a term, coined from Greek word thrix/ trichos meaning hair and Persian word pad-zahr meaning stone or concretion used as an antidote, to describe ingested hairball masses seen in gastrointestinal tract. It is commonly seen in young adolescent females with underlying psychiatric illness^[1]. It was first described by Baudomont in 1779^[2]. Prevalence of this disease is 0.4-1%^[3].

It is seen in patients with an underlying impulse control psychiatric disorder-trichotillomania and trichophagia whereby the patient feels relief on plucking hair and eating the same^[4]. Persistent eating of hair over a period of time results in

accumulation of tight hair balls with mucus, blood and food, in gastrointestinal tract, as the smooth hair resists digestion and peristalsis^[5]. It is most commonly seen in the stomach but may rarely extend through the pylorus to reach upto the colon – called the Rapunzel Syndrome^[6].

Complications include mucosal erosions, ulcerations, perforation, obstruction, malabsorption etc^[7,8].

It is usually asymptomatic and presents with complications once the trichobezoar reaches larger size^[9]. A careful history taking, examination and imaging with CT scan reveals the diagnosis. Upper GI endoscopy is considered the gold standard investigation for diagnosis but CT scan is

necessary to rule out Rapunzel syndrome and satellite lesions^[10,11].

Various modalities described for treatment of trichobezoar include minimally invasive techniques like endoscopic, laparoscopic, robotic assisted extraction and open surgical- laparotomy. Success rates of laparotomy of 100%, less complication and ease of screening of entire bowel for satellite lesions makes laparotomy the treatment of choice. Psychiatric evaluation and treatment of underlying disease is imperative to avoid relapses^[10,12-14].

Case Presentation

A 14-year-old female with no known past illness or previous surgeries presented with complaints of on and off colicky upper abdominal pain for the past 6 months which aggravated over past 1 week. For the past 1 week the pain had been associated with multiple episodes of bilious vomiting. She also had constipation for past 5 days. She had no history of hematemesis/ melaena. She had history of eating hair and thread since 7 years of age.

On examination, she had a BMI - 17, ECOG- 0, no pallor. Her vitals were stable. P/A- soft, not distended, with a 4x 4 cm well defined non tender firm mobile intraperitoneal mass in the umbilical region. Per rectal examination was normal.

She was evaluated with X-ray abdomen which showed distended small bowel loops with multiple air fluid levels s/o Small bowel obstruction. She was further evaluated with CECT abdomen which showed relatively well defined heterogeneous low density mottled appearing non-enhancing intraluminal lesion noted within gastric lumen (20x5 cm) and jejunal loop (15x5 cm) possibly trichobezoar with dilated and fluid filled duodenal and jejunal loop with transition point at the site of intraluminal mass s/o Small bowel obstruction with Gastric and jejunal trichobezoar.

All pre-anaesthetic workup was done and she underwent exploratory laparotomy with an upper midline incision, gastrotomy was done and around 20x6 cm trichobezoar in gastric antrum extending into pylorus and first part of duodenum extracted.

15x 5 cm jejunal trichobezoar with luminal obstruction and proximal bowel dilatation, 65cm from duodenojejunal flexure retrieved through enterotomy. Gastric and jejunal trichobezoar appeared to be separate bezoars – with the distal bezoar being a detached satellite fragment of proximal one of them being two distinct primary bezoars.

Rest of the entire bowel was screened for more satellite bezoars and was found to be normal.

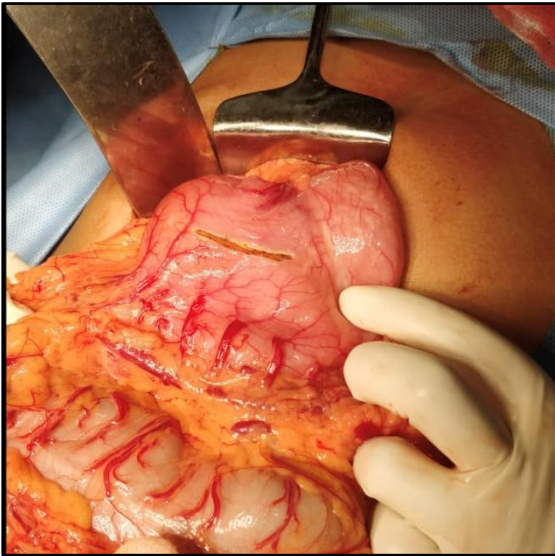
The postoperative period was uneventful. Oral feeds were started on postoperative day 3 and she improved. Psychiatry consultation was done, and advice followed. Patient was discharged on post op day 5 with no complaints and was followed up in Psychiatry and General Surgery OPD.



1. CT abdomen showing gastric trichobezoar



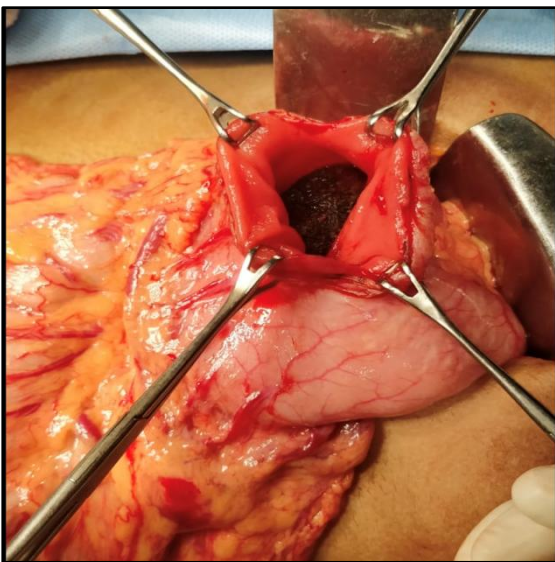
2. CT abdomen showing jejunal trichobezoar



3. Gastrostomy incision



6. Jejunal trichobezoar



4. Gastric trichobezoar



7. Gastric and Jejunal distinct primary trichobezoars



5. Jejunal trichobezoar

Discussion

Introduction

Bezoar is an indigestible stony concretion or conglomerate mass trapped in gastrointestinal tract, which was accidentally or intentionally ingested. Bezoars can be mainly of 4 types depending on the substance ingested^[15] –

- Phytobezoars- plant materials like fiber, seeds, skin of vegetables and fruits.
- Pharmacobezoars- medications like bulk forming laxatives – peridium, psyllium.
- Lactobezoars – milk protein from synthetic milk.

- Trichobezoar – ingested hair.

Trichobezoar was coined from the Greek word *thrix* or *trichos* meaning hair and the Persian word *Pad-zahr* meaning a stone or concretion which was used as an antidote. A trichobezoar is a rare condition in which hair bundles are trapped in stomach and intestines over a period of time. It is commonly seen in pediatric age group and young adolescent females – 80%^[1]. It was first described by Baudomont in 1779^[2]. Prevalence of this disease is 0.4- 1%^[3].

Pathophysiology & complications

It is associated with psychiatric conditions like trichotillomania and trichophagia. Trichotillomania is an impulse control psychiatric disorder within the group of conditions known as body-focused repetitive behaviors (BFRBs)^[4]. The patients usually have an underlying depression, anxiety and poor self image and feels relief on pulling out their hair. Human hair escapes digestion due to its enzyme-resistant properties and slippery surface. Continuous ingestion of hair over a period of time results in accumulation of hair with food, blood and mucus forming tight hairballs within mucosal folds which remain stagnant in the gastrointestinal system^[5].

Trichobezoar can be found anywhere in the gastrointestinal tract, but it is most commonly seen in stomach – 50% cases^[3]. Sometimes they can extend along the pylorus into the duodenum and jejunum and may vary rarely even reach upto the colon. This condition is called Rapunzel Syndrome and was first described by Vaughan et al in 1968^[6]. Rarely a section may detach from the rest and can get dislodged distally causing obstruction^[5]. Large trichobezoars can cause complications like mucosal erosions, ulcerations, gastrointestinal bleeding, intestinal obstruction(25.9%) and perforation(18.3%)^[7]. Other reported complications include anemia (IDA & Megaloblastic anemia), obstructive jaundice or pancreatitis due to duodenal extension, intussusception(7.4%^[7]), protein losing enteropathy and edema^[8].

Clinical Features

They remain asymptomatic for years and symptoms arise only when the hair ball accumulates to a very large size and cause complications^[9]. Patients usually present with abdominal pain(37%), palpable abdominal mass, nausea, vomiting(33.3%) and weight loss (7.4%)^[7]. On examination, there can be evidence of malabsorption, a mobile, well-defined mass may be palpable in the epigastrium. In cases with intestinal obstruction patient would present with an acute abdomen^[10] and a high index of suspicion is required to consider the diagnosis.

Investigations

1. X-ray abdomen erect – features suggestive of intestinal obstruction- multiple air fluid levels, dilated proximal bowel loops and features suggestive of perforation- air shadow under diaphragm^[11]. Rarely concretion of hairs has other associated foreign body with air trapping, which appears as a mottled, soft tissue opacity in the shape of the distended stomach. Contrast radiograph is helpful and diagnostic to delineate the anatomical abnormality created by the trichobezoar^[11]. But in case of Rapunzel syndrome Barium meal is not recommended as it can precipitate obstruction and perforation^[7].
2. USG abdomen- heterogenous hyperechoic band like areas can be seen, but is often not diagnostic^[11]. Distended stomach or bowel loops can be identified.
3. CT scan of abdomen – it is superior to USG in diagnosing the heterogenous non enhancing masses consisting of ‘mottled gas pattern’ or ‘compressed concentric rings’ pattern due to the presence of entrapped air and food debris^{[16][17]}.
CT is superior to Upper GI endoscopy in showing the size and configuration of the trichobezoar^[16], differentiates it from a neoplasm^[18] and most accurately identifying its location and in diagnosing Rapunzel syndrome^[7].

4. Upper GI endoscopy – considered to be the Gold standard in diagnosing trichobezoar but it cannot prove existence of Rapunzel syndrome^[7]. But endoscopy has the advantage of being a diagnostic as well as therapeutic tool^[10]. It can differentiate trichobezoar from other foreign bodies^[9].

Treatment

Several treatment modalities have been described for treatment of bezoars with varying degrees of success. Modalities include pharmacotherapy, endoscopic removal, laparoscopy and laparotomy. Pharmacotherapy with Coca-cola and enzymatic dissolution of proteins with papain has been described for phytobezoars^[1,15] but are mostly ineffective^[13]. Trichobezoars are resistant to pharmacotherapy^[15].

1. Endoscopic removal

It is useful in gastric trichobezoars which are smaller in size^[13]. Endoscopic removal of larger bezoars may be attempted with fragmentation using devices like a polypectomy snare, a basket catheter, biopsy forceps, alligator forceps, an argon plasma coagulation device, electro-surgical knife and an electrohydraulic lithotripsy device^[15]. It is most usually unsuccessful^[13] with only 5% success rates^[10].

Disadvantage

- It is a time consuming procedure and most usually unsuccessful.
- Multiple repeated introduction of the endoscope might be required for complete removal of all fragments^[13].
- Pressure ulceration, esophagitis and oesophageal perforation may occur.
- Fragments may migrate distally through the pylorus, causing intestinal obstruction.
- Not recommended in case of Rapunzel Syndrome.
- Examination of the intestine for satellites cannot be done via endoscopy^[13].

2. Laparoscopic Extraction

Laparoscopic removal of gastric trichobezoar through a gastrotomy was first described by Nirasawa in 1998^[14]. Laparoscopic assistance may be used in endoscopic removal to aid fragmentation^[13]. 75% success rates have been reported for laparoscopic extraction^[10].

Advantage

- Smaller incision
- Lesser pain
- Lesser duration of hospital stay.

Disadvantage

- Less successful in large trichobezoar
- Longer operation duration
- Intraperitoneal spillage and contamination.
- Need for extension of port incision^[12].
- Screening of rest of the intestine for satellite masses in laparoscopy is challenging^[13].

3. Laparotomy

It is still considered to be the treatment of choice as laparotomy has 100% success rates, relatively low complications, ease of examining the rest of the bowel for satellites^[10,13].

It is also the only recommended modality for cases with multiple satellite masses in intestine, Rapunzel syndrome, failure of minimally invasive techniques and in recurrent trichobezoar^[18].

Rare complications include perforation of intestine, pneumonia, paralytic ileus and wound infections^[13].

Recently minimally invasive techniques like endoscopic, laparoscopic and robotic assisted extraction are gaining favour over the conventional open laparotomy technique^[10] and appears to be the future treatment modality. However, open surgery still holds the upper hand in cases of trichobezoar with complications like perforation and Rapunzel syndrome^[13].

Treatment of trichobezoar is not complete without psychiatric treatment of the underlying pathology in order to prevent relapses^[13].

Conclusion

Trichobezoar is a relatively rare disease, seen in young adolescent females with underlying psychiatric illness. It remains asymptomatic and presents very late with complications like anemia, obstruction, perforation or intussusception. Diagnosis is made by careful history taking, examination, imaging- CT scan and Upper GI endoscopy. Treatment includes minimally invasive techniques like endoscopic, laparoscopic and robotic assisted extraction and open laparotomy for complicated cases and in Rapunzel syndrome. Psychiatric evaluation and treatment is an integral part of treatment of this condition.

In this patient, open laparotomy was considered because of the large size of the gastric trichobezoar and the satellite mass in jejunum. Surgery was successful with no postoperative complications. Patient was then referred for further psychiatric evaluation and treatment.

Reference

1. Cannalire G, Conti L, Celoni M, Grassi C, Cella A, Bensi G, et al. Rapunzel syndrome: an infrequent cause of severe iron deficiency anemia and abdominal pain presenting to the pediatric emergency department. *BMC Pediatr* 2018;18(1):125.
2. Shorter E, Shorter HC in the H of M and P of PE. *A Historical Dictionary of Psychiatry*. Oxford University Press, USA; 2005.
3. Ahmad Z, Sharma A, Ahmed M, Vatti V. Trichobezoar Causing Gastric Perforation: A Case Report. *Iran J Med Sci* 2016;41(1):67–70.
4. Trichotillomania (Hair-Pulling Disorder) DSM-5 312. 39 (F63.2) - Therapedia [Internet]. [cited 2020 Jul 23]; Available from: <https://www.theravive.com/therapedia/tric>
5. Al-Janabi IS, Al-Sharbaty MA, Al-Sharbaty MM, Al-Sharifi LA, Ouhtit A. Unusual trichobezoar of the stomach and the intestine: a case report. *Journal of Medical Case Reports* 2014;8(1):79.
6. Vaughan ED, Sawyers JL, Scott HW. The Rapunzel syndrome. An unusual complication of intestinal bezoar. *Surgery* 1968;63(2):339–43.
7. Wang Z, Cao F, Liu D, Fang Y, Li F. The diagnosis and treatment of Rapunzel syndrome. *Acta Radiol Open* [Internet] 2016 [cited 2020 Jul 23];5(11). Available from: <https://www.ncbi.nlm.nih.gov/pmc/articles/PMC5122172/>
8. Wadlington WB, Rose M, Holcomb GW Jr. Complications of trichobezoars: a 30-year experience. *South Med J*. 1992;85(10):1020-1022. doi:10.1097/00007611-199210000-00024
9. Jatal SN, Jamadar NP, Jadhav B, Siddiqui S, Ingle SB. Extremely unusual case of gastrointestinal trichobezoar. *World Journal of Clinical Cases* 2015;3(5):466–9.
10. Al-Osail EM, Zakary NY, Abdelhadi Y. Best management modality of trichobezoar: A case report. *International Journal of Surgery Case Reports* 2018;53:458–60.
11. Yik YI. Stomach trichobezoar (rapunzel syndrome) with iatrogenic intussusception. 2016;71(2):3.
12. Vepakomma D, Alladi A. Complete laparoscopic removal of a gastric trichobezoar. *J Minim Access Surg* 2014;10(3):154–6.
13. Gorter RR, Kneepkens CMF, Mattens ECJL, Aronson DC, Heij HA. Management of trichobezoar: case report and literature review. *Pediatr Surg Int* 2010;26(5):457–63.

14. Nirasawa Y, Mori T, Ito Y, Tanaka H, Seki N, Atomi Y. Laparoscopic removal of a large gastric trichobezoar. *Journal of Pediatric Surgery* 1998;33(4):663–5.
15. Iwamuro M, Okada H, Matsueda K, Inaba T, Kusumoto C, Imagawa A, et al. Review of the diagnosis and management of gastrointestinal bezoars. *World Journal of Gastrointestinal Endoscopy* 2015;7(4):336–45.
16. Rapunzel syndrome | Radiology Reference Article | Radiopaedia.org [Internet]. [cited 2020 Jul 23]; Available from: <https://radiopaedia.org/articles/rapunzel-syndrome-1>
17. Ripollés T, García-Aguayo J, Martínez M-J, Gil P. Gastrointestinal Bezoars: Sonographic and CT Characteristics. *American Journal of Roentgenology* 2001;177(1):65–9.
18. Palanivelu C, Rangarajan M, Senthilkumar R, Madankumar MV. Trichobezoars in the stomach and ileum and their laparoscopy-assisted removal: a bizarre case. *Singapore Med J.* 2007;48(2):e37-e39.