Case Series

Management Of Trichobezoar With And Without Rapunzel Syndrome In Tertiary Care Centre

Ashutosh Talwar[1], Amandeep Singh[1], Manpreet Kaur[1], Haramritpal Kaur[2]

Abstract

Trichobezoar is a rare pathology in which swallowed hairs accumulate in the stomach. An unusual form of bezoar extending from the stomach to the small intestine or beyond has been described as Rapunzel syndrome. Trichobezoars typically cause abdominal pain and nausea, but can also present as an asymptomatic abdominal mass, progressing to abdominal obstruction and perforation. Trichobezoar with Rapunzel syndrome is an uncommon diagnosis. It is predominantly found in emotionally disturbed or mentally retarded young people. The diagnosis may be suspected in young females with abdominal pain, epigastric mass and malnutrition, who have a history of trichophagia.

The Authors presents a series of three cases treated successfully by laparotomy and removal of trichobezoars. Psychodynamic aspects, clinical manifestations, diagnosis and therapeutic strategies are discussed.

Keywords : Females; Psychiatric disorder; Trichobezoar; Trichophagy; Trichotillomania.

Author affiliations:
[1] Department Of Surgery, Guru Gobind Singh Medical College, Faridkot 151203, Punjab India.
[2] Department Of Anesthesia, Guru Gobind Singh Medical College, Faridkot 151203, Punjab India.

Correspondence:
Dr. Ashutosh Talwar,
Department Of Surgery
Guru Gobind Singh Medical College,
Faridkot 151203 Punjab India.

Email: ashutoshtalwar@yahoo.co.in

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Introduction

A trichobezoar is an unusual condition of hair bundles in stomach and small intestine, leading to intestinal obstruction usually affecting younger females.[1,2] It is mainly associated with a psychiatric disorder, trichotillomania having tendency of pulling hairs and significant hair loss. A large number of patients with this disorder have depression, anxiety and poor self-image.[3] They feel pleasant while pulling the hairs involving the scalp, eyebrows, eyelashes or elsewhere in the body, which leads to noticeable hair loss. The prevalence of the condition is 0.06% to 4%.[4]

When ingested, because of its smooth surface, human hair resists both digestion and peristalsis, and accumulates between the mucosal folds of the stomach. Continuous ingestion of hair over a period of time leads to the impaction of hair together with mucus and food, causing the formation of a trichobezoar. In most cases, the trichobezoar is confined within the stomach. In some cases, however, the trichobezoar extends through the pylorus into the jejunum, ileum or even colon. This condition is called Rapunzel syndrome and was first described by Vaughan et al in 1968.[4] In addition, parts of the tail can break off and migrate to the small intestine, causing intestinal obstruction.[5-7] Trichobezoars may not be recognized in their early stages because of their nonspecific presentation, or even lack of symptoms.

They can be classified into four types based on their location in the body:
•Gastric Trichobezoar: This is the most common type and is found in the stomach. It is typically caused by a disorder known as trichotillomania, which is characterized by the obsessive pulling out of one's hair.
•Intestinal Trichobezoar: This type of bezoar is found in the small intestine and can cause obstruction and severe abdominal pain.

•Cecal Trichobezoar: This type of bezoar is found in the cecum, or beginning of the large intestine, and can cause nausea, vomiting, bloating, and abdominal pain.
•Colonic Trichobezoar: This type of bezoar is located in the colon (the large intestine) and can cause obstruction as well as abdominal pain and discomfort due to its size or shape blocking food passage through the intestines

We describe a series of cases of trichotillomania comprising of a rare case of recurrence in a 36 years old male and two cases of young females that led to the formation of a trichobezoar that needed emergent surgical intervention and follow-up psychiatric treatment.

Case Reports

We, at our institute encountered three cases of trichobezoar in the last seven years, out of which two were found to be of Rapunzel syndrome and one case of Trichobezoar. All these patients were managed successfully by open surgical interventions in view of large sized mass in the gut.

CASE 1

A rare case presentation of a 36 years old male having recurrence of trichobezoar is reported. This male had recurrence and gave the history of trichophagy,(**Figure 1**) symptoms of early satiety, nausea and generalised weakness when reported.

Patient was brought with complaints of dull ache in epigastrium for eight months, mass in abdomen for six months, vomiting for 5 months and associated decrease of appetite for 4 months. Patient had edema of legs and feet for one and a half month.

Abdominal examination revealed mildly tender intrabdominal lump 10 cm x 7 cm occupying epigastrium, left lumbar/ hypochondrial area and a part of umbilicus. The mass was non pulsatile, had irregular well defined margin with partial mobility transversely. Abdomen showed previous upper midline scar extending from xiphisternum to umbilicus with history of previous laparotomy 5 years back. Patient was anemic with hemogram 6 gm%. Xray abdomen showed soft tissue shadow in the epigastrium with crescent like gas transonic features, situated in the upper abdomen. Barium meal showed barium diffused on either side of non-opaque foreign body following regular contour of greater and lesser curvature and in the duodenum (**Figure 2**). Anterior gastrostomy was done under general anaesthesia and foul smelling mucous covered trichobezoar with a tailed hairy extension into the duodenum and jejunum was removed (**Figure 3**). Operative diagnosis of trichobezoar with Rapunzel syndrome was made.

CASE 2

A mentally retarded adolescent girl (14 years age) presented in emergency with history of trichotillomania and trichophagia. She had been eating her own hair from the age of 10 years. She had chronic constipation with bowel movements every 2 days and pellet stools for the last 2 years. A palpable mass was identified in the epigastrium that extended into the right hypochondrium, mildly painful to palpation. She was conscious, with signs and symptoms of mild dehydration, anemic and had no signs of jaundice. The systemic parameters were normal.

An abdominal X-ray showed a radio-opaque image from the epigastrium to the mesogastrium, with displaced transverse colon and no evidence of intestinal air. An abdominal ultrasound showed evidence of a significant dilatation of the intestinal loops at the level of the epigastrium and the left side. The abdominal computed tomography (CT) showed that the gastric lumen was occupied by heterogeneous material, mostly hypodense, with concentric rings of different density which extended into the first duodenal segment and was surrounded by contrast medium allowing for distal passage that was consistent with Trichobezoar. Accordingly, patient was planned for emergency laparotomy. Trichobezoar extended from the stomach through the pylorus into the jejunum so the diagnosis of Rapunzel syndrome was made. (**Figure 4**)

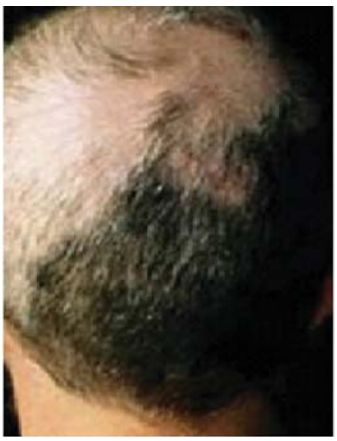

Figure 1. Showing sparicity of hair on the scalp

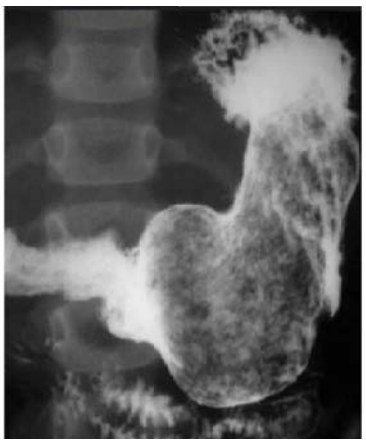

Figure 2. Barium meal showing non opaque foeign body

Figure 3. Trichobezoar with a tailed hairy extension

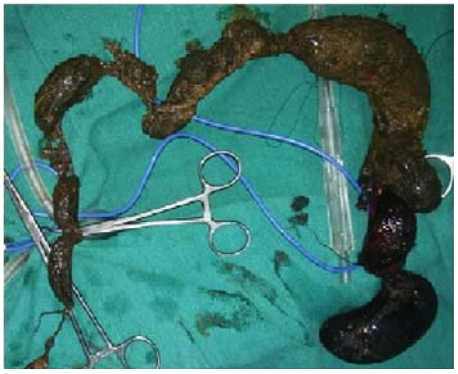

Figure 4. Trichobezoar extending from stomach through pylorus

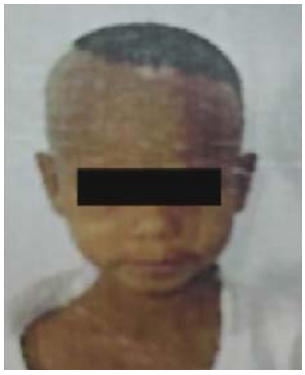

Figure 5. Absent scalp hair

Figure 6. Removed trichobezoar from stomach and jejunum

CASE 3

A seventeen year female presented to outpatient department with chief complaint of pain abdomen for 8 days, which was gradual in onset and progressive in nature, associated with non-passage of stools and flatus for 8 days, associated with one episode of vomiting. There was past history of ingestion of hairs for around 10 years (started at the age of 4 years up to 14 years of age (**Figure 5**). On examination, abdomen was soft to firm with tenderness present in epigastrium, minimally distended. On P/R examination fecal staining was present.

X Ray abdomen (erect and supine) showed multiple air fluid levels. Ultrasound abdomen showed few prominent bowel loops with a maximum caliber of 4.6 cm mimicking Intestinal obstruction. CT scan abdomen showed that stomach was distended. A well circumscribed inhomogenous mass 4.5 x 12.9x 10 cm with mixed air and soft tissue density consisting of mottled gas pattern was seen within the stomach extending up to the pylorus. Another similar mass 4.5x3.1x2.8 cm was seen in small bowel likely jejunum/ proximal ileum with upstream dilatation of proximal small bowel loops. Diagnosis of Trichotillomania was made.

Exploratory laparotomy was done with midline vertical incision. On palpation, the lumen of the stomach was found to be completely occupied by large hard mass and another mass was palpable in jejunum. The stomach was opened on the anterior aspect of the body and its lumen was found to be completely filled with mass made up of hair interspersed with air. Gentle handling of the hair mass resulted in complete evacuation of mass. Stomach was irrigated with normal saline and closure done in layers.

Another mass felt in Jejunum was opened longitudinally and a hard mass made up of hairs extracted and transverse closure done in layers (**Figure 6**).

Discussion

Bezoar is a mixture of swallowed foreign material in gastrointestinal tract, most commonly in stomach. This term 'bezoar' is believed to be arrived from an Arabic word 'badzehr' or Persian word "panzehr" which means antidote. The first case of bezoar in Western countries was reported in 1779 while doing autopsy of a patient who died of gastric perforation and peritonitis.[2,9]

Trichobezoars are formed as a result of accumulation of hair in gastrointestinal tract. Fermentation and decomposition of entrapped food especially fat leads to characteristic rancid odour in patient's breath and bezoar.[5,10] Trichobezoars are most commonly seen in females (approximately 90%) of age group 13–19 years with an unrevealed psychiatric disorder. In our series too one was male and two were females. About 50% of these patients are found to be trichophagic. Around 30% of the trichotillomaniac patients indulge in trichophagia (eat their hair) to the extent that requires surgical interventions in removal of trichobezoar.[11] The site of hair pulling is most commonly from the scalp, but can occur from the eyelashes, eyebrows, and pubic area.

Bezoars were managed laparoscopically.[15] 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.

Usually, trichobezoar is confined to the stomach, but rarely it may extend from the stomach to the small intestine (even colon). This is an unusual form called Rapunzel syndrome.[14] Less than 40 cases of trichobezoar with Rapunzel syndrome have been reported in medical literature.[5] Bezoars can also be found distally in the gastrointestinal tract without continuity with the stomach due to breakage and distal propulsion. It has been described as being as strong as a dandelion and as strong as a dog leash.[6,7]

The Rapunzel syndrome was first reported in the literature by Vaughan et al in 1968.[8] Very few reports of Rapunzel syndrome are documented in psychiatric literature. An explanation for such disparity is due to the fact that most cases of trichotillomania are referred early to psychiatrist before the development of Rapunzel syndrome.[12,13] Clinical manifestations depend on the bezoar's location and size. Affected patients mostly remain asymptomatic for many

years, till the bezoar increases in size to the extent which is responsible to produce clinical symptoms. It is commonly presented as abdominal pain, nausea, vomiting, decreased appetite, early satiety and symptoms due to intestinal obstruction and peritonitis can also be there.[5] Clinical presentation of these patients may be confusing as often they are not forthcoming with a history of trichophagia either due to embarrassment or the unintentional nature of the problem. Trichobezoars should be considered as a differential diagnosis in a young female patient with a mobile epigastric mass. The common complications are anaemia, haemetemesis, gastric mucosal erosion, ulceration, and perforation of the stomach or the small intestine, gastric outlet obstruction, intussusception, obstructive jaundice, protein-losing enteropathy, acute pancreatitis, and death.

The gold standard for diagnosis though is upper gastrointestinal endoscopy.[15,16] Successful management and treatment of a trichobezoar demands removal of the mass and prevention of relapse. In the early stages endoscopic removal is possible, if bezoar is limited to small area and is of small size. The hair appears black (despite the normal hair colour) due to denaturing of the hair protein by the acid. Management almost always requires surgical removal. It can be removed by using endoscopic techniques or surgery (laparoscopic approach or open surgery). Phytobezoars and lactobezoars are easy to remove using endoscopic techniques because of small size. It is less effective in trichobezoar removal as they are usually of large size. Gorter et al, in a retrospective review of 108 cases of trichobezoar, evaluated the available management options.[14] It was noted that whereas 5% of attempted endoscopic removals were successful, 75% of attempted laparoscopies were successful. However, laparotomy was 100% successful and thus favoured as their management of choice.[14]

All of our patients were managed surgically because of delayed presentation in our set up as psychiatric illness is still a taboo in a developing country like India where majority of the population resides in villages. So, open surgical intervention in the form of laparotomy and gastrotomy with retrieval of the mass was done.

During the last few years, trichobezoar cases have attracted debate about the application of minimally invasive techniques, such as endoscopy and laparoscopy,[10-14] rather than laparotomy, as well as medical treatment and enzymatic degradation, which are attractive because of their non-invasiveness but have been reported to be ineffective.[15,16]

Endoscopic removal, if successful, would be considered the most attractive treatment option. The first report of the successful endoscopic removal of a trichobezoar was for one that was relatively small, weighing only 55g.[17] Reports of successful endoscopic removal of trichobezoars in children are remarkably scarce - they are vastly outnumbered by case reports documenting unsuccessful attempts of endoscopic removal with or without fragmentation.[8,14] In our patient, two big masses were found, and endoscopic removal would have been challenging and not safe.

An analysis of the published case reports revealed that out of 40 cases in which endoscopic removal had been tried, only two (5%) were successful.[8] In one of these, a trichobezoar was successfully removed whole from the distal esophagus.[8] In a series of 15 patients with bezoars, a 15-year-old girl underwent fragmentation of a large trichobezoar by means of a modified needle-knife and mono-polar coagulation current. In most case reports, however, fragmentation was considered impossible because of the size, density and hardness of the mass, and endoscopy was not considered a viable therapeutic option.[14] Moreover, because the removal of all fragments requires repeated introduction of the endoscope, pressure ulceration, esophagitis and even esophageal perforation may occur.[9] Also, fragments of a large trichobezoar might migrate through the pylorus after fragmentation or repeated manipulation, causing intestinal obstruction. Careful examination of the intestine for satellites cannot be performed by endoscopy, and the removal of those fragments is impossible. Although not a therapeutic option, endoscopy may prove to be extremely valuable as a diagnostic modality in patients in whom the nature of the gastric mass is unclear. It enables the differentiation between trichobezoars and foreign bodies that can be fragmented and removed using endoscopy.[18]

In one study, laparoscopy was used for the initial procedure but then converted into a laparotomy when difficulties were encountered as a result of a large intragastric mass. In some centers, laparoscopy is considered inferior to laparotomy for the treatment of a trichobezoar. Nirasawa et al[10] were the first to report on laparoscopic removal of a trichobezoar. Since then, only six other reports of attempted laparoscopic removal have been published.[11-13] The lack of reports on endoscopic treatment might partly be explained by the rarity of trichobezoars, but it could also indicate that laparoscopy is not an attractive treatment modality for trichobezoar. Of the six case reports, two reported failure to remove the trichobezoar, which was attributed to the large size of the trichobezoar and to the presence of satellite trichobezoars in other locations of the gastrointestinal tract (as in our patient).[13-14] In one study, endoscopic and laparoscopic approaches were combined; because endoscopic fragmentation of the bezoar was not possible, a laparoscopic approach was used to fragment the trichobezoar, then endoscopy used to remove the fragments.[19] Successful laparoscopic removal, however, requires a significantly longer operation time compared to conventional laparotomy, mostly due to the complexity of the operation. Careful examination of the entire digestive system (stomach and intestine) is necessary to avoid secondary intestinal obstruction due to satellites. With laparoscopy, this procedure is far more challenging; the risk of spilling contaminated hair fragments into the abdominal cavity makes the laparoscopic approach even less desirable. In addition, the rarity of trichobezoars makes it difficult to achieve a good technique for laparoscopic removal and

inspection of the entire intestine. However, the laparoscopic removal of trichobezoars with intestinal obstruction has advantages compared to laparotomy, including better cosmetic outcome, fewer postoperative complications and reduced admittance time.[20] Though several reports stress the excellent cosmetic result of the laparoscopic approach, they also report the frequent need to extend the initial port wounds, sometimes by up to 4cm.[10]

Laparotomy was successful in most cases of trichobezoars, including our case. In the literature, the cases of 100 patients who underwent successful conventional laparotomy were identified. However, 12% had one or more complications, including perforation of the intestine during removal of the trichobezoar,[21,22] minor wound infection,[23] pneumonia, paralytic ileus,[24] and ileal trichobezoar and fecal leakage through the lower part of the laparotomy wound.[25] Due to the high success rate, and the relatively low complication rate, the low complexity, and the ability to carefully examine the entire gastrointestinal tract for satellites in a short period of time, laparotomy is still considered the treatment of choice in our center.

The literature provides no evidence of superiority of endoscopy or laparoscopy over laparotomy. The lack of invasiveness of these techniques does not seem to outweigh the disadvantages and the complexity of these procedures.

It is emphasized that the majority of these patients have an underlying psychiatric or social disorder. So, the treatment of co-existing psychiatric illness is of utmost importance and regular follow up is strongly recommended for psychiatric evaluation. All of our patients had psychiatric illness which was diagnosed only at the time of index admission after finding the presence of trichobezoar incidently for their vague symptoms that too after extensive inquest. A multidisciplinary approach is essential to prevent recurrence of the problem.

Conclusion

A diagnosis of gastric bezoar should be suspected in any child with symptoms of gastric outlet obstruction, and surgical removal is usually indicated for large trichobezoar. Trichobezoar has to be considered in the differential diagnosis of abdominal pain and a non-tender abdominal mass even in young children. Small trichobezoars may be extracted by endoscopic fragmentation, enzymatic therapy or combinations of these approaches. Rapunzel syndrome, on the other hand, need surgical removal. Counseling by a psychiatrist is an important part of management to prevent recurrence.

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