# Surgery Section

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# Primary Trichobezoar in Small Intestine: A Case Report

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# ABSTRACT

Trichobezoar is a rare surgical condition that mostly occurs in the stomach and often causes a surgical emergency when left untreated. It is often predisposed by psychiatric conditions, mental retardation, previous gastric surgery, or abnormal anatomy. Trichobezoar progresses extremely slowly and is diagnosed through imaging such as a barium series, endoscopy, or computed tomography. Surgical removal is typically required. The patient in this case was a 12-year-old girl who presented with symptoms of abdominal pain, vomiting, nausea and constipation. She was found to be anaemic and underweight. Contrast-enhanced Computed Tomography (CECT) diagnosed acute intussusception and small bowel obstruction, which were treated by emergency laparotomy. During the operation, a large primary ileal trichobezoar was found in the distal part of the ileum with hair strands extending up to the transverse colon. The absence of masses elsewhere made the diagnosis even more peculiar, as primary intestinal trichobezoar is highly uncommon. Primary closure of the enterotomy was performed as it was viable, and the postoperative period was uneventful. A history of trichophagia was elicited, and a psychiatric assessment of the patient was conducted. Behavioural therapy was initiated for the trichophagia. In conclusion, trichobezoar is a rare surgical entity with modifiable predisposing factors that require a multidisciplinary management approach.

Keywords: Intestinal obstruction, Intussusception, Rapunzel syndrome

# **CASE REPORT**

The present case report discusses the case of a 12-year-old girl who presented to the Surgical Emergency Ward with colicky abdominal pain for a duration of five days, bilious vomiting and constipation for a period of two days. The patient was initially managed conservatively in a local hospital and was referred for definitive treatment to the present Institution. The patient had similar episodes in the recent past, thrice during a period of the past six months, each time being managed symptomatically. On eliciting a detailed history, it was found that the patient had not undergone any surgeries in the past. The patient was yet to attain menarche. She belonged to a middle socio-economic class family that consumes a mixed diet. No significant family or personal history relevant to the diagnosis was noted.

The patient was observed to be thin-built, and on bedside examination, she was found to be underweight for her age and undernourished according to the Indian Academy of Paediatrics (IAP) growth chart. Her Body Mass Index (BMI) was 18.3 kg/m<sup>2</sup>, with a weight of 31 kilograms and a height of 130 cm. Pallor was noted, and the girl was found to be afebrile. She was mildly dehydrated with no tachypnoea or tachycardia. She was normotensive, and other systemic examinations were found to be normal.

Examination of the abdomen revealed a mildly distended abdomen, mild tenderness over the right lumbar and right iliac regions, and absent bowel sounds. However, the abdomen was soft with no guarding or rigidity, and no crepitation was elicited. Examination of the external genitalia was normal. Digital rectal examination revealed normal tone of the external sphincter and the presence of fecal staining.

The routine blood investigations were within normal range except for moderate anaemia (haemoglobin of 9.7 g/dL). The abdominal radiograph showed multiple air-fluid levels without any air under the diaphragm, as depicted in the pictures below [Table/Fig-1,2], indicating intestinal obstruction, possibly of the small intestine,



[Table/Fig-1]: X-ray abdomen erect showing multiple air fluid levels in the abdomen (white arrow). [Table/Fig-2]: X-ray chest Posterioanterior (PA) view shows absence of air fluid levels under diaphragm (orange arrow). (Images from left to right)

without any intestinal perforation. Ultrasonography of the abdomen demonstrated free fluid in the pelvic cavity, with the presence of mucosal thickening at the distal ileum with sluggish peristalsis. The appendix was not visualised in the ultrasonogram. In the CECT of the abdomen, a bowel-within-bowel appearance near the ileocaecal junction was demonstrated, and proximal small bowel loops appeared dilated and filled with fluid with a maximum diameter of 3.3 cm. Thus, the CECT of the abdomen diagnosed ileocaecal intussusception with small bowel obstruction [Table/Fig-3].



[Table/Fig-3]: CECT abdomen showing bowel in bowel appearance and dilated small bowels (yellow arrows).

The patient was initially diagnosed with subacute intestinal obstruction with a differential diagnosis of subacute appendicitis, intestinal tuberculosis, or intestinal bands or strictures. Upon

admission to the Surgical Ward, she initially passed flatus and showed no signs of perforation. Therefore, patient was initially treated medically with intravenous fluids, antiemetics and proton pump inhibitors. She was followed-up and further evaluated while being managed with nasogastric decompression using a nasogastric tube, and the patient was restricted from oral intake. Subsequently, the patient developed symptoms of acute intestinal obstruction with progressing abdominal distension, vomiting and obstipation, and was given the final provisional diagnosis of acute intussusception with small bowel obstruction. Consequently, emergency laparotomy under general anaesthesia was planned, and the patient was evaluated for the surgery.

The laparotomy proceeded with a subumbilical vertical lower midline incision. Upon performing the laparotomy, free fluid was observed in the peritoneum, and a few enlarged mesenteric nodes were noted. During the initial assessment, the jejunum and proximal ileum were found to be distended. Distal ileal loops, located 10-11 cm from the ileocaecal junction, were found to be collapsed, as shown in the figure below [Table/Fig-4]. An obstructing thin, firm mass of 15 cm in length was noted in the distal part of the ileum, starting 11 cm from the ileocaecal junction. A small incision was made over the antimesenteric border to reveal an entangled mass of a hairball shown in [Table/Fig-5], of which the distal part was removed through the same incision. The incision was further extended, and



[Table/Fig-4]: Peroperative findings of dilated proximal small intestine (green arrow), dilated distal ileal loops (red circle), mass in the distal most ileum (blue ellipse).

the removal of the trichobezoar was performed with countertraction to prevent mesenteric tear.

An intestinal trichobezoar measuring 20×5×4 cm in size [Table/ Fig-5] was entirely removed along with the evacuation of the contents [Table/Fig-6]. A thin string of hair extending beyond the ileocaecal junction to the transverse colon was also found by palpating the mass up to the transverse colon and removed. After the removal of the obstruction, the bowel was found to be completely viable, and hence it was decided to perform primary closure of the enterotomy. Primary ileal closure [Table/Fig-7] was sutured with a two-layer technique, where the inner layer seromucosa was closed with interrupted 3-0 Vicryl sutures, and the outer seroserosa layer was closed with interrupted 3-0 silk sutures. An abdominal drain was placed, and the procedure was completed.

The patient's postoperative period was uneventful with a smooth recovery. Intravenous fluids, antibiotics and analgesics were administered. The patient passed stools on day 3 and was started on a solid diet on day 5. The drain was removed on day 6, and the patient was discharged on day 7. Before discharge, a psychiatric evaluation was conducted, and trichophagia and pica were diagnosed postsurgery. The patient had a history of trichophagia for the past four years. The patient was advised to undergo behavioural therapy postdischarge. Surgical follow-up was performed to assess wound healing and recovery after 14 days and 30 days postdischarge, respectively. Furthermore, the patient was instructed to attend the Outpatient Department in case of any complaints.

# DISCUSSION

A bezoar is a compacted, indigestible ingested foreign material that causes impaction in the gastrointestinal tract, leading to its obstruction [1,2]. There are many types of bezoars, depending on their constitution, namely phytobezoar, trichobezoar, lactobezoar, pharmacobezoar, polybezoar and biliary bezoar [3,4]. Phytobezoar is the most common form among bezoars, whereas trichobezoars are very rare, amounting to only 6% of all bezoars [5]. The pathogenesis is depicted in [Table/Fig-8].



[Table/Fig-5]: Incision in the distal ileum and the intestinal trichobezoar. [Table/Fig-6]: Removal of trichobezoar and evacuation of contents with presence of viable ileum [Table/Fig-7]: Primary ileal closure. (Images from left to right)



Hair, when ingested, remains undigested and escapes peristalsis due to its slippery nature, becoming trapped in the mucosal folds of the stomach. Over time, it accumulates with the help of pyloric resistance, taking on a ball shape as discussed by Singh H et al., [6]. As years pass, the ball entangles more and more hair, which is partially digested by the acidic secretions of the gastric mucosa and coated with mucus [7]. Khalifa MB et al., observed that trichobezoar most commonly appears in children with mental retardation [5]. This rare disorder is also frequently documented among young females, with the majority of cases occurring between the ages of 13 years and 20 years, as noted by Ahmed MM et al., [8]. The patient in the present case is also a young female nearing adolescence.

Lamanna A et al., observed that trichotillomania, an irresistible urge to pull body hairs, is a rare psychological condition frequently associated with trichophagia, the impulse to eat hair (found in approximately one out of five cases) [1]. Although the prevalence of both conditions is less than 1%, Dutta P et al., and Dong ZH et al., presented that one-third of patients with trichophagia develop trichobezoar, as seen in the current patient [7,9]. Rapunzel syndrome is the term given to the condition where a large trichobezoar is found in the stomach, with long hair strands extending beyond the pylorus into the small bowel, leading to complications [9,10].

The stomach is the most common site for trichobezoar, with almost all cases occurring there. Primary small intestine trichobezoar in the absence of a gastric bezoar is very uncommon [11]. In the present patient, a gastric bezoar was ruled out by CECT scan. Small bowel obstruction or intussusception is usually due to the extension of a gastric bezoar into the small bowel [12]. However, the present case patient had a rarer form of this rare condition, with a primary ileal trichobezoar causing intussusception and intestinal obstruction.

The clinical presentation of trichobezoar is non specific and often deceptive, making the diagnosis extremely challenging, as most cases are asymptomatic for years [5,13]. Common symptoms that lead patients with bezoar to the hospital include an abdominal mass, especially in the epigastric region (~75%), abdominal pain (~70%), vomiting (~65%), gastrointestinal bleeding (~60%), early satiety, weight loss (~40%), and constipation (~30%). Most symptoms result from complications of the condition, such as obstruction of the pylorus or small bowel in most cases, or erosion and perforation of the gastric mucosa, pancreatitis and intussusception in rare instances [13,14]. Many patients are also noted to be anaemic [11]. The patient also exhibited symptoms of undernourishment, such as being underweight and anaemic. The patient only had symptoms of intestinal obstruction, including abdominal pain, constipation, nausea and vomiting. Similar cases of trichobezoar are presented in [Table/Fig-9] [1,3,5,8,13-15].

Author	Gender	Age (years)	Trichobezoar location	Complications
Khalifa MB et al., [5]	Female	17	Stomach, lleum	Intestinal obstruction
Ahmed MM et al., [8]	Female	18	Stomach	Jejunojejunal intussusception
Al Wadan AH et al., [13]	Female	7	Stomach	Nil
Kim SC et al., [3]	Female	8	Stomach	Nil
Lamanna A et al., [1]	Female	20	Stomach	Nil
Wang CK et al., [15]	Female	16	Stomach	Nil
Gorter RR et al., [14]	Female	9	Stomach	Intusussception
	Female	14	Stomach	Peritonitis with reactive pancreatitis
	Female	15	Stomach	Gastric ulcer
	Female	7	Stomach	Gastric ulcer
[Table/Fig-9]: Similar cases of trichobezoars presented in the literature [1,3,5,8, 13-15]				

A definitive diagnosis of the condition through clinical examination is almost impossible, and diagnosis is best done using imaging techniques such as upper gastrointestinal endoscopy, contrast X-rays like barium series, ultrasonography and CECT scan. Endoscopy provides direct visualisation of the mass and is considered the gold standard for diagnosis, with additional therapeutic implications. Computed tomography is a standard diagnostic tool frequently used, as demonstrated in the patient where it detected the complications of the condition.

The patient exhibited symptoms of acute obstruction during the hospital stay, necessitating laparotomy and removal of the trichobezoar through enterotomy [11]. Although laparotomy is the traditional treatment modality, it is still considered the cornerstone in cases of advanced conditions with expected complications [7,13]. Other minimally invasive modalities, such as endoscopic removal, laser-induced mini-explosive methods and laparoscopic removal, are attempted in early-diagnosed patients [13]. Postoperative care should always include a psychiatric assessment of the patient to reveal trichotillomania, trichophagia, depression, anxiety, or any other obsessive-compulsive disorder, as was the case in the present patient where the postassessment revealed trichophagia. The stigma associated with the condition attributes to the late diagnosis of the condition, thereby causing complications [8]. Hence, a multidisciplinary approach is required for such patients.

# CONCLUSION(S)

Given its insidious course and extremely non specific presentation, a primary ileal trichobezoar causing intussusception and intestinal obstruction in this patient proved to be a diagnostic conundrum. Laparotomy is the treatment of choice, involving the removal of the obstructing mass and repair of the gastrointestinal tract as required. Surgical treatment must always be complemented with a psychiatric assessment of the patient, and any underlying issues like trichophagia must be addressed. A multidisciplinary approach is required to improve the patient's condition and prevent further similar instances.

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