

Trichobezoar-Induced Intussusception in a Child Without Rapunzel Syndrome: Case Report

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Abstract

Introduction: Isolated intestinal trichobezoars without gastric involvement are extremely rare and may present with unusual and challenging complications. Among these, intussusception caused by an intestinal trichobezoar in the absence of classical “Rapunzel syndrome” features is particularly uncommon and seldom reported.

Case Presentation: A 5-year-old girl presented with bilious vomiting and was diagnosed with intussusception due to an intestinal trichobezoar, with no gastric involvement. Early imaging and surgical removal resulted in a full recovery.

Conclusion: This case underscores the potential of trichobezoars to cause intussusception without classical “Rapunzel syndrome” features, highlighting the need for a thorough evaluation in pediatric patients.

Keywords: *Intussusception, Trichobezoar, Rapunzel Syndrome, Bowel Obstruction, Case Report*

Introduction

Intussusception, occasionally referred to as ‘telescoping’ action, occurs when a section of the intestine invaginates into a neighboring segment [1,2]. A lead point in the proximal bowel is entrapped and propelled by peristalsis into the distal intestine, resulting in constriction and compression of the mesenteric vasculature [3]. This leads to the characteristic signs and symptoms of colicky abdominal pain, vomiting, ‘red currant jelly’ feces, and a palpable mass. However, the simultaneous presence of all four indicators is uncommon and tends to appear late in the course of the illness, making early diagnosis challenging. If left untreated, intestinal ischemia can result in necrosis, perforation, and peritonitis [3].

Bezoars are conglomerates of ingested extraneous substances. Their nomenclature is determined by their composition: phytobezoars originate from plant material, pharmacobezoars from drugs, and trichobezoars from hair. Although some bezoars may be asymptomatic, others can manifest mass effect symptoms, including abdominal discomfort, early satiety, nausea, and vomiting [4]. Most bezoars develop in the stomach [5], rarely in the small intestine [6], and infrequently in the colon [4]. Intestinal bezoars may present with signs of small-bowel obstruction and can lead to perforation if not promptly identified and treated.

Trichobezoars are the most prevalent bezoars [6,7]. These have been documented to develop secondarily to trichotillomania in up to 38 % of patients, as they are significantly more common among those who pull and ingest their hair.

Trichobezoars are well-documented pathological lead points that cause intussusception in children [4-12]. In most cases, “Rapunzel syndrome” is observed, which is defined as a stomach trichobezoar that develops a tail extending past the pyloric sphincter. A sufficiently large tail can adhere to the intestinal wall and act as a focal point for intussusception, with normal peristalsis propelling the tail and entrapped wall further along the gastrointestinal tract [20]. Trichobezoars can also form primarily in the intestine, leading to intussusception [4,13]. This distinction is clinically significant because the absence of a gastric bezoar may lead to a delayed diagnosis or misinterpretation of symptoms.

Herein, we report the case of a 5-year-old girl who presented with ileocolic intussusception owing to an intestinal trichobezoar, with no associated gastric trichobezoar or ‘Rapunzel syndrome.’ This case highlights the need for increased awareness of intestinal trichobezoars as an independent cause of intussusception and elucidates the diagnostic challenges posed by their atypical presentations. This manuscript was prepared following the CARE guidelines.

Case Presentation

A previously healthy 5-year-old girl presented to our emergency department (ED) with bilious vomiting that began 6 days earlier, accompanied by abdominal pain, decreased appetite, reduced urine output, and constipation. Her parents noticed a significant reduction in her weight over the 6 days, as her regular clothes started to fit more loosely. There were no associated fever, cough, palpitations, chest pain, skin rash, intake of food from outside sources, or similar family episodes. In addition, there was no family history of malignancy, no pets at home, no consumption of unpasteurized milk, and no recent travel history. The patient denied eating hair or having any unusual eating habits and was completely vaccinated.

The patient sought medical advice from several hospitals. At one hospital, she was treated for a presumed urinary tract infection and discharged with oral antibiotics; however, her symptoms did not improve. She then visited another hospital, where she was treated for constipation with a Fleet enema and discharged on a laxative, again with no improvement. Finally, she was transferred to the Emergency Department of our hospital.

On physical examination, the patient appeared unwell, underweight, and dehydrated, with sunken eyes. She weighed 13 kg. Her initial vital signs were as follows: blood pressure, 101/85 mmHg; heart rate, 112 beats/min; respiratory rate, 29 breaths/min; temperature, 36.9°C; oxygen saturation, 99 % in room air; and glucose level, 4 mmol/L. Abdominal examination revealed distension with tenderness in the right lower quadrant and a hard, non-mobile mass, measuring approximately 10 cm, extending from the right upper quadrant to the right lower quadrant, toward the umbilicus. No hepatosplenomegaly or lymph node enlargement was observed. The results of other systemic examinations were unremarkable.

Initially, she was kept nil per mouth and received an intravenous bolus of normal saline (20 ml/kg). Basic laboratory testing showed mild leukocytosis (13.60 cells/ μ L) and low hemoglobin levels (10.40 g/dl). However, normal renal function, amylase, lipase, and uric acid levels, and peripheral smears were also observed. Bedside ultrasonography of the right iliac fossa demonstrated a target-like view consistent with intussusception and preserved vascularity (**Fig. 1**).



Fig 1. Target ultrasound image of the abdomen at the right inferior fossa showing target sign.

Enhanced computed tomography (CT) of the abdomen and pelvis with intravenous contrast was performed to rule out other differential diagnoses, such as malignant lymphoma. Abdominal CT revealed a high-grade bowel obstruction owing to ileocolic intussusception spanning approximately 13 cm and an upstream small-bowel obstruction with a diameter of up to 3.3 cm. A large fecaloma measuring 14 × 3.6 cm was observed, potentially contributing to intussusception. At least one smaller intussusception was identified immediately distal to the obstruction site. No free air or fluid was observed (**Fig. 2**).

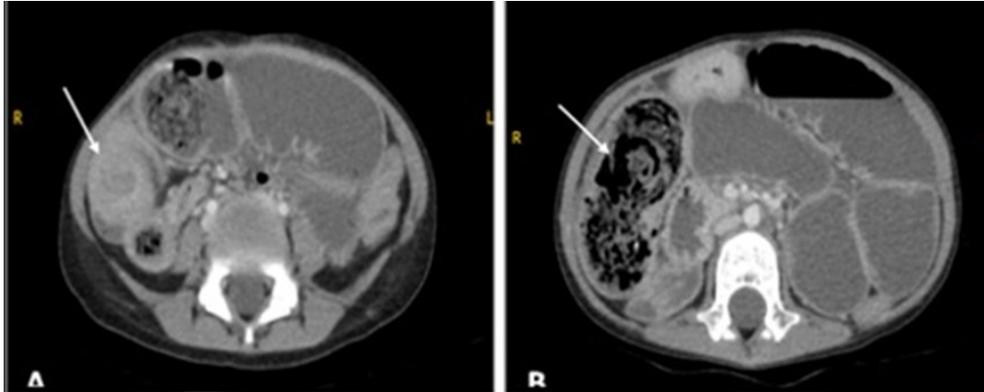


Fig 2. Computed tomography of the abdomen with contrast. *A: Axial image showing dilated bowel loops and whirlpool sign (arrow) indicating the presence of intussusception. B: Axial image showing dilated bowel loops and fecaloma (arrow).*

The patient was started on broad-spectrum antibiotics (cefazoline), analgesics, and antiemetics and was taken to the operating room immediately. Under general anesthesia in the supine position, deep abdominal palpation before the incision revealed an oblong, firm mass in the right hypochondrium, which was mobile in all directions but restricted along the long axis. A right transverse upper abdominal incision was made, and exploration of the peritoneal cavity revealed an “intestinal trichobezoar” along with fecal matter in the mid-ileum, which was delivered through the wound with no evidence of a gastric trichobezoar or “Rapunzel syndrome”. An enterotomy was performed, and the trichobezoar mass and fecal matter was removed and fully delivered from the enterotomy site. The enterotomy and abdominal wall were closed in layers. (**Fig. 3**)



Fig 3. An Intraoperative finding of the mass. *A: A mass within the intussuscepted bowel directly visualized. B: A Dilated bowel with a mass inside the intestine. C: An enterotomy showing the foreign body inside intestine. D: The extracted trichobezoar.*

The patient’s postoperative course was unremarkable. Enteral feeding was initiated on postoperative day 3. She was discharged after 4 days in a stable condition, with complete recovery and resolution of symptoms. At the one-month follow-up with both the psychology and pediatric surgery teams, she reported no active complaints, and her wound had properly healed.

Discussion

Intussusception associated with trichobezoars is often linked to “Rapunzel syndrome”; however, having “Rapunzel syndrome” is not a prerequisite for intussusception. Male patients accounted for only 4 of 24 such cases, yet “Rapunzel syndrome” was present in most of these males, with only one case not exhibiting the condition [4,13–15]. The lower incidence of intussusception attributable to trichobezoars in males may stem from the generally shorter length of their hair [13]. Primary intestinal bezoars are less likely to induce intussusception because there is no anchored gastric mass or tail, creating a mechanically unfavorable environment for the lead point. Although all reported cases described symptoms such as abdominal discomfort, nausea, and vomiting, the patient presentations exhibited considerable variability. The timeline for abdominal pain ranged from 12 months to the day before admission. Obstructive symptoms, including constipation, obstipation, and distention, may have been observed in certain instances [9,12–13,15–17], whereas diarrhea may also be a presenting symptom [4]. Physical examination can help localize a trichobezoar, as individuals with “Rapunzel syndrome” frequently present with a palpable abdominal mass, whereas those without the syndrome may not present with this finding.

Our findings emphasize that while “Rapunzel syndrome” remains the most widely recognized cause of trichobezoar-related intussusception, it is not the only mechanism responsible. This distinction is crucial for accurate diagnosis and timely intervention. In “Rapunzel syndrome,” imaging plays a key role in diagnosis, as a large gastric trichobezoar typically forms before the tail grows long enough to cause intussusception. Therefore, CT may reveal a heterogenous mass in a distended stomach, a “beehive pattern” described by Bolivar-Rodriguez et al. [16]. Although abdominal radiography and ultrasound have been used to effectively identify “Rapunzel syndrome,” CT is preferred owing to its greater specificity [18].

Diagnosing intussusception caused by a trichobezoar without “Rapunzel syndrome” is more challenging because endoscopy may fail to detect the source of obstruction, and smaller intestinal trichobezoars may be mistaken for fecal matter [4]. Laparotomy is commonly used to extract trichobezoars and reduce intussusception; however, a laparoscopic approach has also been successful in managing “Rapunzel syndrome” in the absence of intussusception [19].

Other pediatric cases of intussusception caused by a trichobezoar but lacking “Rapunzel syndrome,” share key similarities with our case. Won et al. [4] reported the case of a previously healthy 8-year-old boy with a preliminary diagnosis of appendicitis who presented with abdominal pain and vomiting. Imaging revealed a bowel obstruction related to small-bowel intussusception. Diagnostic laparoscopy revealed ileoileal intussusception due to an ileal bezoar, and the patient recovered without any postoperative complications. [4]. The size and extent of the trichobezoar suggested a possible non-gastric “Rapunzel syndrome,” with a primary ileoileal mass causing high-grade bowel obstruction [4]. Au et al. [13] described a similar case involving a 5-year-old child with a 3-day history of abdominal discomfort and non-bilious vomiting accompanied by obstipation. This patient’s distinct medical history included failure to thrive and fecal impaction a year earlier; the obstructing bezoar was located in the ileum [13].

Trichobezoars do not always have a homogeneous composition. Two reported cases did not involve trichotillomania: Raghu et al. [20] noted a decade-long history of pica, specifically involving threads from clothes, and Won et al. [4] did not document a significant behavioral history. Therefore, our study further supports the notion that the absence of trichotillomania does not rule out trichobezoar formation, which can result from the ingestion of hair, threads, or fibers from the environment [6].

Conclusion

This case report highlights the importance of recognizing trichobezoars as an uncommon but significant cause of intussusception, particularly in pediatric patients.

Although trichobezoars are typically associated with “Rapunzel syndrome,” their occurrence in the absence of gastric components should not be overlooked. This case also highlights the diagnostic challenges associated with this condition, as the patient presented with nonspecific symptoms and was initially misdiagnosed several times. Early imaging, particularly CT, plays a crucial role in identifying obstructions and guiding surgical intervention. In this case, successful laparotomy and removal of the bezoar led to full recovery, emphasizing the need for timely and accurate diagnosis and intervention. This report contributes to the growing awareness of bezoars in pediatric gastrointestinal pathology and reinforces the necessity of a high index of suspicion for similar presentations.

Glossary

CT- Computed tomography

ED- Emergency department

Statements

Informed Consent: Informed consent was obtained from the patient or guardian.

Authorship: All authors attest that they meet the current ICMJE criteria for Authorship

Ethics Approval and Consent to Participate

This study was approved by the Institutional Research Ethics Committee in King Fahad Medical City, Riyadh, Saudi Arabia.

Consent for Publication

A written informed consent was taken from the parents to write this case report.

Availability of Data and Materials

Data that support the findings in the current study are available from the corresponding author on reasonable request.

Competing Interests

None of the authors have any competing interests to declare.

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Authors' Contributions

N.A. contributed to the conceptualization, case identification, initial draft preparation, and critical revision of the manuscript. A.A. conducted the literature review, manuscript drafting, and critical revision for intellectual content. S.N. was responsible for clinical case management, data collection, and manuscript editing. M.K. performed the surgical intervention, contributed to the case discussion, and provided critical revision of the surgical aspects of the manuscript. Y.A. was involved in radiological interpretation, imaging analysis, and manuscript review. All authors reviewed and approved the final version of the manuscript and agreed to be accountable for its content.

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