

## Case Report

# Intestinal intussusception due to “Rapunzel syndrome” in an adult

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## ABSTRACT

The bezoars are collections of foreign bodies or ingested material that accumulate over time and are often located in the stomach or small intestine, but can be found throughout the digestive tract. Most bezoars are of indigestible organic matter such as trichobezoars (hair); or vegetable and fruit-phytobezoars; or a combination of both but other rare substances have also been described in literature. In some cases, the trichobezoar extends through the pylorus into duodenum, jejunum or even into the colon. Rapunzel syndrome is a rare manifestation of a trichobezoar, which occurs when strands of swallowed hair extend beyond the pylorus of the stomach, into the intestine as a tail. It was first described by Vaughan et al in 1968. Trichobezoars can block the intestinal tract and, if left untreated, can be a life-threatening emergency requiring surgery. Patients are often women, under the age of 20 and have insidious non-specific abdominal symptoms and staggered presentations over months to years. The diagnosis of this type of pathology is usually a challenge for the attending physician, so we present a case report of a 38-year-old female with gastric-intestinal occlusion secondary to trichobezoar.

**Keywords:** Intestinal intussusception, Trichobezoar, Rapunzel syndrome

## INTRODUCTION

Trichobezoars form in patients with trichotillomania and trichophagia. They begin as retained hairs between the gastric folds; the hair is then denatured by gastric acid, becomes black due to oxidation, and combines with food to form an enmeshed mass.<sup>1</sup> Trichobezoars subsequently become colonized by bacteria resulting in halitosis.<sup>2</sup> Once formed, bezoars grow by the continuing ingestion of food rich in cellulose and other indigestible materials, matted together by protein, mucus, and pectin.<sup>3</sup>

On the other hand, intussusception of the bowel is defined as the telescoping of a proximal segment of the gastrointestinal tract within the lumen of the adjacent segment. This condition is frequent in children and presents with the classic triad of cramping abdominal pain, bloody diarrhea and a palpable tender mass.<sup>4</sup> However, bowel intussusception in adults is considered a

rare condition, accounting for 5% of all cases of intussusceptions and almost 1%-5% of bowel obstruction. Eight to twenty percent of cases are idiopathic, without a lead point lesion. Secondary intussusception is caused by organic lesions.<sup>5</sup>

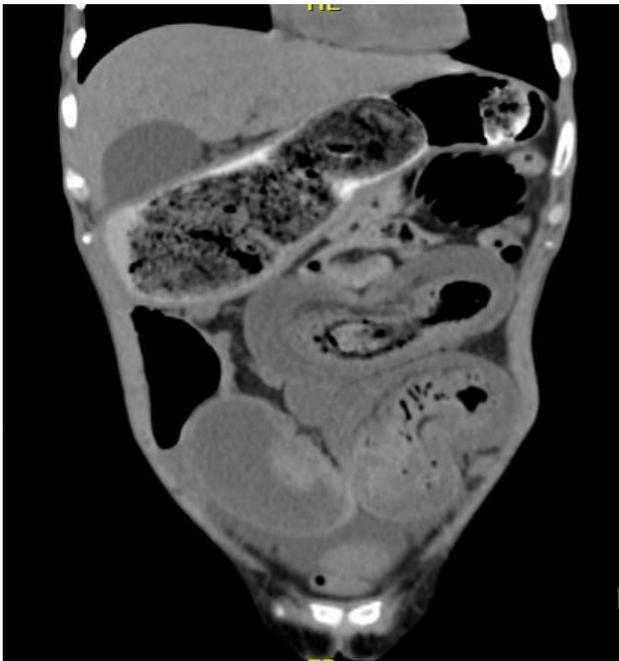
## CASE REPORT

A 38-year-old female patient is admitted, according to interrogation she has no pathological history of importance. Surgical intervention by exploratory laparotomy 20 years ago secondary to trichobezoar, no further details are available. The patient started 1 week ago with diffuse abdominal pain, predominantly in the left hypochondrium, associated with nausea and vomiting. On examination, the patient was oriented, cooperative, ectomorphic constitution. Sclerae, skin and oral mucosa with dehydration, pale color. Abdomen distended, peristalsis decreased in intensity, localized

abdominal pain on palpation of the left hypochondrium and mesogastrium, with no evidence of peritoneal irritation at the time.

Laboratory and imaging studies were performed with the following results: albumin 1.43, total bilirubin 0.2 gr/dl, Indirect bilirubin 0.14 gr/dl, direct bilirubin 0.114, glucose 80.1 mg/dl, hemoglobin 6.7 gr/dl, platelets 187,000/UI, leukocytes 3,900/mm<sup>3</sup>.

An abdominal x-ray was performed showing gastric and intestinal loops distension. Subsequently, an abdominal tomography was performed with a finding of trichobezoar that caused intestinal occlusion, as shown in the images (Figure 1).



**Figure 1: Coronal section of abdominal tomography with oral contrast showing an occupying mass compatible with trichobezoar, oral contrast medium surrounds the heterogeneous images.**

During her initial management, she was started with fluid therapy, placement of a nasogastric tube with more than 1000cc of intestinal output, antibiotic therapy based on ceftriaxone and metronidazole. She had a torpid evolution because she was anxious and tachycardic, so it was decided to admit her to the emergency surgical room with prior perform exploratory laparotomy, gastrotomy and extraction of trichobezoar, intestinal reduction due to the presence of intussusception of jejunum, intestinal resection was performed due to the presence of intestinal necrosis at 30 centimeters from the Treitz ligament also a terminal end anastomosis (Figure 2-4).

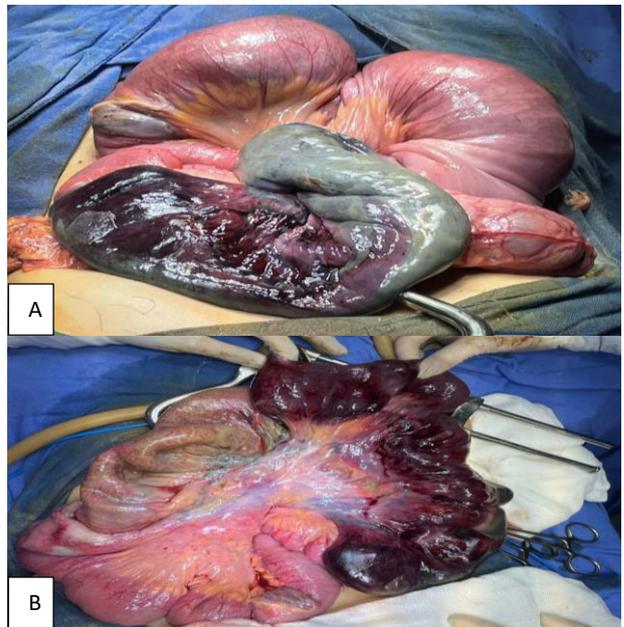
In her post-surgical follow-up, a consultation with the psychiatry service was performed, which diagnosed generalized anxiety disorder and trichophagia, the treatment with quetiapine was started.



**Figure 2: Exploratory laparotomy, trichobezoar is observed in the gastric cavity.**



**Figure 3: Trichobezoar being delivered.**



**Figure 4 (A and B): Intestinal ischemia due to intestinal intussusception. Exploratory laparotomy. Jejunum intussusception, intestinal ischemia was observed 30 cm from the Treitz ligament in figure, in the general hospital of zone 1A of the IMSS.**

## DISCUSSION

Secondary intussusception is believed to initiate from any pathologic lesion of the bowel wall or irritant within the lumen that alters normal peristaltic activity and serves as a lead point, which is able to initiate an invagination of one segment of the bowel into the others.<sup>6</sup> The result is bowel obstruction and inflammatory changes ranging from thickening to ischemia of the bowel wall as in our patient occurred.<sup>7</sup>

Common presenting signs and symptoms of trichobezoars include chronic gastrointestinal complaints, a palpable abdominal mass, and small bowel obstruction.<sup>8</sup> Current evidence supports a 6-month timeframe for the development of the symptomatic trichobezoar, although different time courses have been noted.<sup>9</sup>

Imaging studies for this type of pathology are important, abdominal radiographs usually have a non-specific mass appearance, and upper gastrointestinal series may demonstrate a filling defect in the stomach. The computed tomographic (CT) scan distinguishes trichobezoars from other possible etiologies of an epigastric mass (pseudocyst, duplication cyst, tumor) and should be diagnostic with the appropriate history.<sup>10</sup>

Small trichobezoars may be extracted by endoscopic fragmentation, vigorous lavage enzymatic therapy or a combination of these approaches. Endoscopic removal after fragmentation using water pick, laser, extracorporeal shock-wave lithotripsy or by enzymatically dissolving the mass is possible in small-sized bezoars. Another option for removing small bezoars is with laparoscopy through a small incision. Large bezoars (> 20 cm) usually require removal through open surgery, as none of the above techniques are effective.<sup>11</sup>

There are few reported cases of recurrence of trichobezoar, in this case corresponds to one, however we do not have the details of the previous surgery. The patient was managed as detailed in the literature. Likewise, cases of intussusception due to this type of pathology are few reported and most of them in children. Adult intussusception represents 5% of all cases of intussusception and accounts for only 1%-5% of intestinal obstructions in adults.

## CONCLUSION

Trichobezoars are an infrequent pathology, it is important to know in detail the approach given to patients for a good evolution. Up to 20 percent of patients showed recurrent bezoars. In order to prevent recurrence, remember that the treatment is not purely surgical, patients should be encouraged to increase water intake,

modify their diet (egg, avoid persimmons, stringy vegetables, and high-fiber foods), chew their food carefully, in addition seek psychiatric evaluation in favor of improve the health of those who suffer from it.

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