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CASE REPORT

Trichobezoar: Presenting as Primary Intestinal Obstruction

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ABSTRACT

Rapunzel syndrome is a rare type of trichobezoar with an extension of the hair into the small bowel. Clinical presentation is deceptive and vague ranging from abdominal mass to gastrointestinal symptoms.

Bezoars are usually confined to the stomach which is seen in individuals with psychiatric illness, like trichotillomania, trichophagia and gastric dysmotility. Long standing bezoars may extend into the small intestine leading to a condition known as Rapunzel syndrome. Trichobezoars presenting primarily as intestinal obstruction is very rare. Diagnosis can be established by endoscopy, ultrasonography and computed tomography scan. Treatment includes improvement of general condition and removal of bezoar by laparoscopic approach or laparotomy. Psychiatric consultation is necessary to treat and prevent relapse. We present the case of a 6 years old girl, where the trichobezoar was not suspected at all, especially with negative history of trichophagia and trichotillomania. This girl presented with history of intermittent pain abdomen since 3 months and recent onset severe pain abdomen with abdominal distension and vomiting. Skiagram of abdomen revealed dilated small bowel loops with multiple air fluid levels suggestive of intestinal obstruction. Even ultrasonogram of abdomen was also inconclusive for cause of obstruction. At laparotomy, dilated small bowel loops with multiple interbowel adhesions were seen. A hard mass measuring 5 × 6 cm was seen at distal jejunum causing distal obstruction with pressure necrosis over bowel wall. Enterotomy was done which revealed trichobezoar which was removed *en masse*. Resection and anastomosis of necrosed jejunum was done. Patient recovered completely after the procedure.

Keywords: Intestinal perforation, Primary small intestinal obstruction, Rapunzel's syndrome, Trichobezoars, Trichophagia.

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INTRODUCTION

Trichobezoar is a condition wherein swallowed hair starts accumulating in the stomach over a period of time to form a concretion which presents later with features of malnutrition and intestinal obstruction.

This syndrome is named after the girl with the long tresses in the fairy tale written by the Grimm Brothers in 1812. When the bezoar extends into the small intestine, it is called as Rapunzel syndrome which was first reported in 1968 by Vaughan et al.[1] This syndrome is usually found among teenage girls with psychological disorders, and it is manifested as follows: abdominal discomfort or nausea and vomiting, anorexia, weight loss, and habit of eating hair (intentional alopecia may be a frequent clue for the diagnosis).

The formation of trichobezoar starts with the intake of small bits of hair that do not progress to the gastrointestinal tube, getting accumulated and gathered.[2] This condition can evolve to the following complications: ulceration, partial obstruction, total obstruction, necrosis, intestinal perforation or peritonitis. The dimensions of the bezoar seem to have a relation with the morbidity and mortality evaluation, but not with the evolution of the clinical status and its complications. Therefore, the pathological findings of the complications are not related to the size of the trichobezoar. It can even present with complications, like intestinal obstruction, perforation and peritonitis. Here, we present a rare case of trichobezoars in distal jejunum causing small bowel obstruction with impeding perforation in an adolescent girl.

CASE REPORT

A 16-year-old female presented to our casualty with complaints of pain abdomen with abdominal distension and vomiting of 2 days. History of intermittent pain abdomen and vomiting since 3 months was present. The patient had delayed developmental milestones and poor performance at school. On examination, the patient was conscious, cooperative, pale and hemodynamically stable. Abdominal examination revealed distended abdomen with diffuse tenderness with sluggish bowel sounds. Laboratory investigations revealed hemoglobin of 7.8 gm/dl, serum total protein of 4.5 gm/dl and serum

albumin of 2.5 gm/dl. Skiagram of the abdomen showed dilated small bowel and multiple air fluid levels (Fig. 1). Ultrasound examination of the abdomen was also inconclusive for cause of obstruction.

The patient was given packed cell transfusion to correct anemia and was posted for elective laparotomy. On exploration, dilated small bowel loops were seen along with mass measuring 5 × 6 cm in distal jejunum causing distal obstruction with pressure necrosis of overlying bowel wall (Fig. 2). Multiple interloop adhesions were seen. Enterotomy revealed a hairball which was removed *in toto* (Figs 3 and 4). Adhesiolysis was done with resection and anastomosis of necrosed small bowel. Abdomen was closed after thorough examination of entire small bowel and stomach for any other trichobezoars.

Postoperative recovery was uneventful. She was discharged with psychiatric counseling. At the follow-up examination 2 weeks following surgery, the patient was found to be free of abdominal symptoms and there was an improvement in her hemoglobin and serum total proteins when compared to the preoperative levels.

DISCUSSION

The common presentation of trichobezoar is in young females usually with an underlying psychiatric disorder. In our case, the presentation is in an adolescent girl with hairball in small bowel, causing symptoms, which could mimic congenital bands, abdominal tuberculosis and gastrointestinal infestation, especially in endemic areas like ours. The commonly accepted definition of Rapunzel syndrome is that of a gastric trichobezoar with a tail extending to the jejunum, ileum or the ileocecal junction. This condition is common in teenage girls with trichotillomania and trichophagia and patients with gastric motility problem.[2] Debakey and Oschner suggested that the slippery nature of hair and its entrapment within gastric folds could be the reason for bezoar formation.[3] Over a period of time, hair gets matted with each other and with other indigestible materials like cotton fibers and vegetable matter to assume the shape of the stomach. Sometimes, its tail may extend into the small intestine.

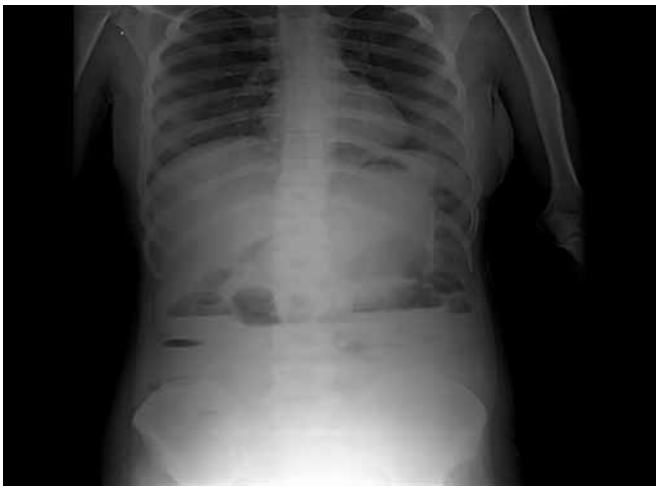

Fig. 1: Erect abdomen X-ray showing multiple air fluid level

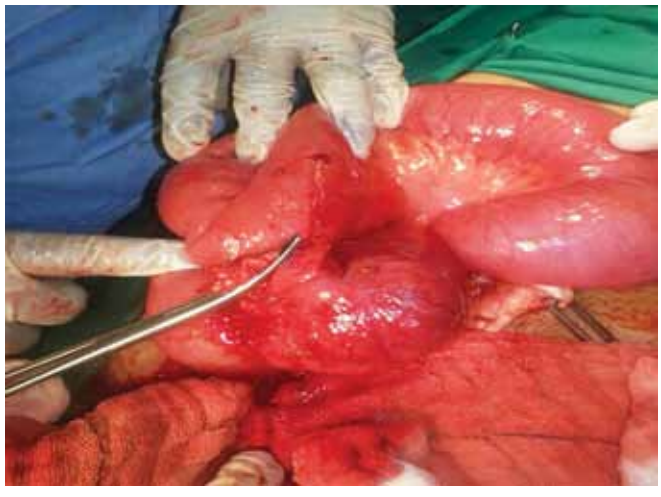

Fig. 2: Peroperative showing interloop bowel adhesions and mass formation

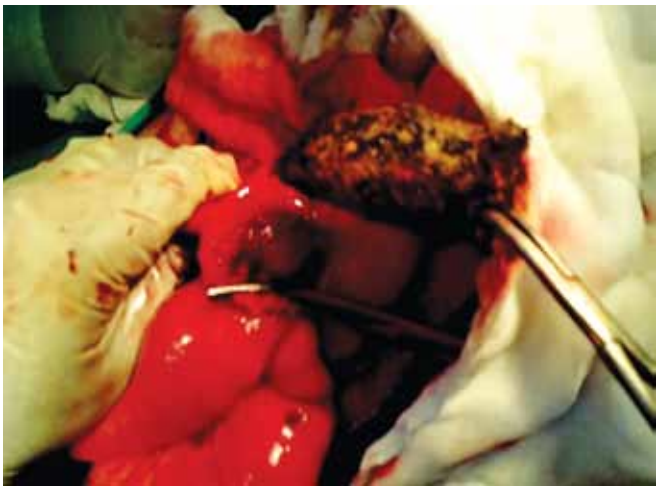

Fig. 3: Enterotomy and retrieval of trichobezoar

Fig. 4: Trichobezoar—hair ball

Majority of cases of trichobezoar present late, due to the low index of suspicion by the physician. Of 131 collected cases of trichobezoar, a palpable abdominal mass was present in (87.7%), abdominal pain (70.2%), nausea and vomiting (64.9%), weakness and weight loss (38.1%), constipation or diarrhea (32%) and hematemesis (6.1%). The laboratory investigations revealed low hemoglobin in about 62% (average).[4]

The complications of Rapunzel syndrome ranges from attacks of incomplete pyloric obstruction to complete obstruction of the bowel to perforation to peritonitis and mortality.[5]

Patient with trichotillomania (a psychological condition that involves strong urges to pull hair), around 30% will engage in trichophagia and of, these, only 1% will go on to eat their hair to the extent requiring surgical removal.[6] Less than half of the patients give a history of trichophagia. There have been few cases of recurrence following successful surgery. The early detection of trichophagia and trichobezoar depends on an effective screening for trichotillomania and related behaviors, in order to prevent a possibly life-threatening condition with important medical and surgical morbidity. Such effort must include a better collaboration between medical and surgical specialties, dealing with particular aspects of therapeutic relationship regarding shame and guilt as well as considering that trichophagia may be more often present than the majority of clinicians, psychiatrists in particular would expect.[7]

A trichobezoar represents a serious surgical condition. It is important to consider such diagnosis in face of suggestive symptoms, even if signs of trichotillomania are not present. Unless psychiatric counseling and follow-up is done, this condition can recur.

CONCLUSION

A trichobezoar represents a serious surgical condition. It is important to consider such diagnosis in face of suggestive symptoms, even if signs of trichotillomania are not present. The complication of trichobezoars ranges from attacks of incomplete pyloric obstruction to complete obstruction of the bowel which can lead to bowel perforation, peritonitis and mortality. All patients with Trichobezoar should be referred for psychiatric evaluation after surgery to avoid recurrence.

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