CASE REPORT

Trichobezoar: A Rare Cause of Chronic Abdominal Pain in Childhood

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ABSTRACT

A 12.5-year-old girl presented to the Pediatric emergency department in hypotensive shock; with complaints of abdominal pain and vomiting for 15 days, which had worsened over the last 2 days. She had marked abdominal distension along with tenderness. The child was resuscitated immediately with intravascular volume replacement. X-ray abdomen revealed massive air under diaphragm, which was almost occupying, whole of the abdomen. CRP was markedly elevated i.e., 259 mg/L. After initial stabilization with IV rehydration and antibiotics, patient underwent emergency laparotomy. One liter of purulent fluid was suctioned out. A hard mass of entangled hair was found in stomach with proximal stomach wall perforation. After mass removal and gut repair, patient remained in ICU for two weeks for completion of treatment with antibiotics and she had uneventful recovery later. On follow up, the abdominal complaints were settled, and patient remained under care of psychiatry department for management of behavioral disorder and trichophagia.

Key Words: *Trichobezoar, Abdominal pain, Childhood*

INTRODUCTION

A bezoar is an accumulation of indigestible material in the gut, in the form of a mass, that may result in intestinal obstruction especially in pediatric age group.[1] Among these bezoars, trichobezoar is an extremely rare condition with the prevalence around 90% in females, and 80% of these occur in patients under 30 years of age. Bezoars are mainly related to anxiety behaviors and psychiatric disorders. [2]

Many types of bezoars have been described in literature depending upon their composition, including phytobezoars (fibers from, vegetables, plants, and fruits), trichobezoars (hair), lactobezoars (milk protein), and pharmacobezoars composed of medications.[3] Gastrointestinal trichobezoar that accounts for less than 6% of all bezoars, is a compact aggregate mass of swallowed hair.[4] GI trichobezoars mostly affects young females with habits of trichotillomania (hair-pulling) and trichophagia (hair-ingesting: one's own hair and of others including the family members' or toy dolls'). It is commonly associated with certain psychiatric and behavioral disorders.[3]

Human hair has somewhat slippery surface and hence they tend to accumulate in mucosal folds of the stomach. As it is resistant to digestion, the continued ingestion of hair, results in the formation of a ball, comprising of entangled hair as well as food and mucous particles. Later, it slowly resumes the shape of stomach. Bezoar becomes darker in color with the passage of time, as the acidic environment of the stomach results in protein denaturation of hair.[5] Trichobezoars mostly reside in stomach but may extend downward into the colon. Rapunzel syndrome is a rare variety of gastric trichobezoar, in which there is a tail-like extension of gastric trichobezoar, that

extends beyond the pylorus, approaching the small intestine or even colon.[5]

Trichobezoar requiring surgical intervention is a rare phenomenon in the context of early childhood, with an incidence of <1% of patients who ultimately underwent surgery. The first reported case of trichobezoar was in 1968, and afterwards around 48 cases of combined trichotillomania and trichophagia leading to trichobezoar in both genders have been reported.[6] The patient we report here was having trichobezoar and presented with acute abdomen in emergency that is not among the frequently documented presentation according to the literature. This adds to the further rarity and complexity of this case requiring engagement of various disciplines towards management of the case in pediatric age group. So, this case report is aimed at contribution to the limited literature regarding this rare clinical condition in pediatric age group.

CASE REPORT

A 12.5-year-old girl, presented in the Pediatric emergency, Madinah Teaching Hospital, Faisalabad, with complaints of abdominal pain and vomiting for the past 15 days. The patient's condition worsened over the last two days and on the day of presentation, she was drowsy and unable to bear weight on her own. Examination revealed the patient was in hypotensive shock, on the basis of clinical findings, such as the altered state of consciousness, thready pulses, blood pressure 70/40mmHg, cold clammy extremities, capillary refill time >4 seconds. Abdominal distension was very marked along with tenderness and hyper-resonant percussion note. The child was resuscitated immediately according to the WHO guidelines of management of hypotensive shock, with intravenous pushes of normal saline 20 ml/kg given twice, followed by the rehydrating solution Ringer lactate 100 ml/kg. x-ray abdomen was performed immediately afterwards, that revealed the signs of perforation, in the form of massive air under diaphragm which involved the whole abdomen. Nasogastric tube and Foley's catheter was passed to decompress the gut and to monitor intake and output. Meanwhile the department of surgery was taken on board. The relevant investigations at the time of admission revealed Hb 15 g/dl (normal 12-16 g/dL). TLC 6000/cmm (normal 5000-10,000/cmm), platelet count 491,000/cmm (150,000 to 400,000/cmm), CRP was 259 mg/L (normal <10 mg/L), blood urea 25 mg/dl (5-18 mg/dL), creatinine 0.31 mg/dl (0.5-1 mg/dL). Electrolytes were reported as serum Na level 136 mmol/L (normal 135-145 mEq/L), serum K 3.68 mmol/L (3.4-4.7 mmol/L) and serum chloride 107.20 mmol/L (normal 90-110 mmol/L). Ultrasonography abdomen showed collapsed bowel loops with gaseous shadows; moderate fluid was seen in the peritoneal cavity. After initial stabilization with IV rehydration and antibiotics, patient underwent emergency laparotomy.

Immediately after the initial incision, free air was detected in the abdomen. One liter of purulent fluid was suctioned out of the peritoneal cavity. A hard mass of entangled hair was found in stomach with proximal perforation of the stomach wall **(fig. 1 and 2).** Trichobezoar was removed from the stomach, perforation was repaired and patient was shifted to the ICU for post-operative monitoring and management. On routine post-operative care, serial electrolytes were monitored on daily basis and levels were maintained with intravascular fluid and electrolytes replacement. She was encouraged to gradually resume oral intake and the course of injectable antibiotics was completed for next two weeks, including vancomycin and meropenem. The serial CRP monitoring was done that showed progressive improvement in response to the antibiotics. She recovered uneventfully within a week, with restoration of bowel movements, normalization of CRP, and stable body temperature. The patient was also being evaluated from the psychiatry department to address the unusual eating behavioral disorder i.e., trichophagia in order to prevent the relapse. Further inquiry revealed the chronic history of recurrent abdominal pain for which she used to take medication for symptomatic relief. Parents had also taken her to some hospital one year back for complaint of thinning of hair. The actual cause of thinning of hair and abdominal pain, that was trichophagia, remained undiagnosed and ultimately resulted in formation of massive trichobezoar **(fig 3).** She was discharged from MTH after completing the course of antibiotics, and when oral intake was normalized. On further follow up for next two

months, the abdominal complaints were settled, and patient was on regular follow up with the psychiatry department.

Fig 1: Perforation visible in the stomach wall

Fig 2: Trichobezoar (a ball of entangled hair and food particles, typically black colour)

Fig 3: Trichobezoar (ball like mass of entangled hair with long tapering tail)

DISCUSSION

Trichotillomania (compulsive hair pulling) and trichophagia (ingestion of pulled hair), are psychiatric conditions, that can lead to the formation of trichobezoars when present simultaneously in the same patient.[7] Trichobezoars are more common in females than in males, due to the predisposing factors of trichotillomania and trichophagia, and their prevalence tends to increase during adolescence and early adulthood. The early literature suggested that while childhood onset is possible, it is relatively uncommon.[8] A case series was conducted at Baylor College of Medicine, Houston, comprising of seven patients with trichobezoars, all of whom were girls, found a mean age at presentation of 11.5 years.[9] Our patient, 12.5 years, is comparable to this documented case series.

Most of the time the patient with a gastric trichobezoar presents with nonspecific symptoms, including abdominal pain (70%), nausea and vomiting (64%), gastrointestinal bleeding (61%), epigastric pain or discomfort, early satiety, indigestion, weight loss (38%), diarrhea or constipation (32%)[10]. Rapunzel Syndrome is a rare presentation where trichobezoars cross the pylorus to enter the duodenum, ileum and colon.[11] However, a retrospective study by Erzurumlu et al, which included 34 cases, documented that abdominal pain was the most common symptom of a gastrointestinal bezoar in all cases, followed by mild to severe nausea and vomiting in 33 cases.[12] Therefore, trichobezoar might be considered as a differential diagnosis in patients presenting with chronic symptoms of abdominal pain, vomiting, indigestion and constipation.

A case series of 17 patients of trichobezoars, conducted by Mirza *et al.* from 2005-2018, at the Children Hospital and Institute of Child Health, Lahore, Pakistan, documented that Fifteen patients (88%) presented with abdominal pain and vomiting, while 8 (47%) had abdominal distension. Seven (41%) patients developed complications secondary to the GIT trichobezoar (intussusception and gangrene in 1, small bowel obstruction in 4, gastric perforation and massive bleeding per rectum in 1, acute transient pancreatitis and hypertension in 1).[4]

Our patient had the past history of consultation in dermatology outpatient for the concern of alopecia. She had the milder symptoms of abdominal discomfort for one year that remained unnoticed and due to low index of suspicion, the trichobezoar was not considered in the differential diagnosis of abdominal pain and alopecia. It ultimately resulted in bowel perforation and she presented in a critical state, with hypovolemic

shock. This is comparable to a case of an 18-year-old girl with alopecia, reported from Iraq. She was misdiagnosed for many years and ultimately presented with intestinal obstruction.[13]

Trichotillomania and trichophagia often have their roots in underlying psychosocial stress disorders. Children may adapt the hair-pulling habit as a coping mechanism, in response to family related or environmental stressors. Hence the understanding of psychosocial stressors is mandatory for effective management.[14] The early detection of trichophagia and trichobezoar depends on effective screening for trichotillomania and related behaviors, in order to prevent a possibly life threatening condition with important medical and surgical morbidity.

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Authors' contribution

AS: Identification and management of the case, principal author, literature review and referencing, approval of the final version to be published.
TA: Management of the case, contributed in write up of discussion component, critical review, approval of final version.
EI: Management of the case, Literature review & referencing, corresponding author, case report writing and corrections as per journal requirements.
AN: Management of the case, contributed in write up of introduction component, critical review, literature review.

All the authors have approved the final manuscript draft and accept the responsibility of research integrity.