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Multiple Bezoars with Rare Complications: A Case Report

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Abstract: A bezoar is a mass of indigestible material that accumulates in the digestive tract, sometimes causing a blockage. Bezoars usually form in the stomach, sometimes in the small intestine or, rarely, the large intestine. Trichobezoars are the commonest type of bezoars. Rapunzel syndrome, Intestinal obstruction and small bowel perforation all are rare presentation of bezoars and presentation of all of these in a single patient is even rarer. We are reporting a very rare and interesting case of a 12 years old girl presented with three different type of bezoars with all these rare complications Rapunzel syndrome, intestinal obstruction and jejunal perforation.
Keywords: Bezoar; obstruction; perforation; rapunzel syndrome; laparotomy; enterotomy; ileostomy.

INTRODUCTION

Bezoars are collections of indigestible materials found in the gastrointestinal tract[1]. They are named according to the predominant component material for example, hair-trichobezoar, plant substance- phytobezoar and occupational bezoar- cotton bezoar in tailors[2, 3]. Rapunzel syndrome is a rare complication of a gastric trichobezoar in which the mass of hair extends through the pylorus into the small bowel[1]. Intestinal obstruction by a phytobezoar is a rare entity reported in the literature. Small-bowel perforation secondary to phytobezoar is even a rarer clinical entity which is not well documented in the literature[4]. Number of cases had been reported in literature separatively for trichobezoar, phytobezoar and their complications, but we are reporting a very rare case with three different kinds of bezoars with all rare presentations in a single patient.

CASE REPORT

A 12 years old girl referred from primary health care centre to our hospital with complaints of pain abdomen and vomiting. On examination patient had tachycardia, hypotension with sign of dehydration. Abdominal examination revealed generalized tenderness with mobile lump in epigastric region and bowel sound absent. Her investigations showed Hb 7gm%, TLC 12800, blood urea 63mg/dl, serum creatinine 1.3mg/dl, serum potassium 2.8meq/l and all other blood investigations were within normal limit. Computed tomography(CT) abdomen showed dilated stomach with heterogeneous ball measure about 110 x 54mm size [figure 1] with small similar component seen in the second part of duodenum and an another bezoar seen in distal ileum with dilated bowel loops and multiple air fluid level. After resuscitation, informed written consent was taken and she was posted for exploratory laparotomy. Intra-operatively a large trichobezoar with its tail extending into small gut [k/a Rapunzel syndrome] was found in stomach [figure 2]. Tail of gastric trichobezoar was extruding through a perforation in jejunum at mesenteric border around 15-20 cm distal to duodeno-jejunal junction [figure 3]. Gastric part of bezoar consists of hair and tail made up of hair mixed with synthetic fibers. On further tracing of small gut another bezoar made up of cotton fiber intermingled with fecal matter was found in distal ileum causing luminal obstruction [figure 4]. Gastric trichobezoar was removed by longitudinal gastrotomy. The remaining fiber of tail extruding from jejunum removed through perforation site. It was not possible to push and remove ileal bezoar through perforation site proximally; therefore it was removed by enterotomy. Ileal enterotomy site brought out as loop ileostomy and resection anastmosis was done for jejunal perforation. Peritoneal wash was given with normal saline, pelvic drain kept inside and procedure completed by closing the abdomen in layer. Patient kept in intensive care unit and post operative period was uneventful. On retrospective evaluation she was diagnosed for trichobezoar 6 month back but her father was stuck in flood in an another state of India and her treatment

delayed. Both parents are bagger with no history of psychiatric disorder in family. She had no history of any antipsychotic treatment. During follow up period she was counseled by psychiatrist.

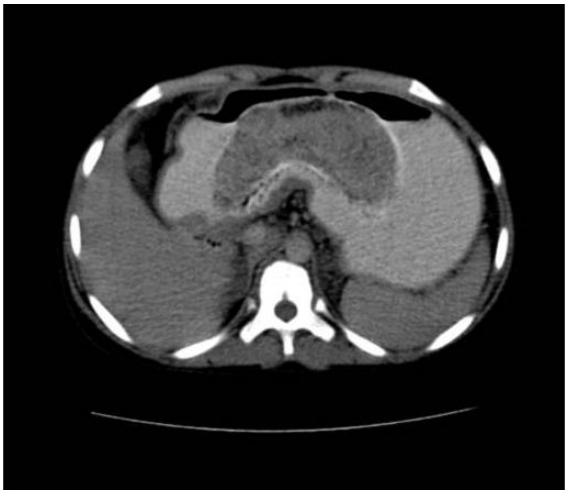

Fig-1: Computed Tomography scan showing a large trichobezoar in stomach.

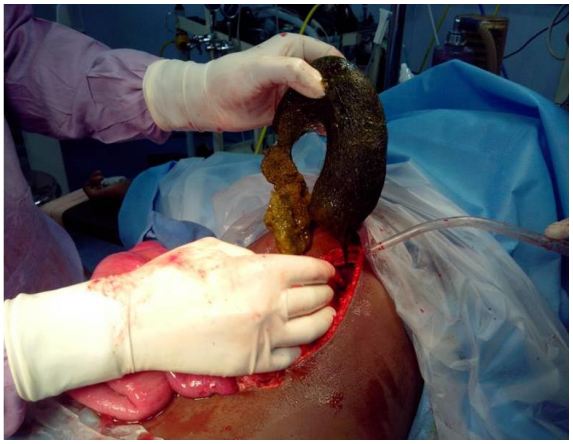

Fig-2: Intra operative photograph showing gastric trichobezoar with its tail extending beyond pylorus [rapunzel syndrome]

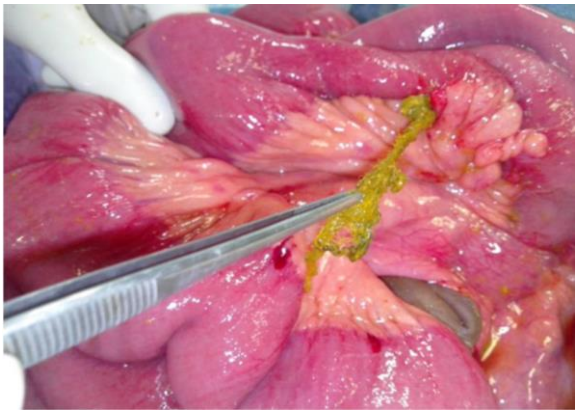

Fig-3: Intra operative photograph showing tail of gastric bezoar extruding through jejunal perforation at mesenteric border.

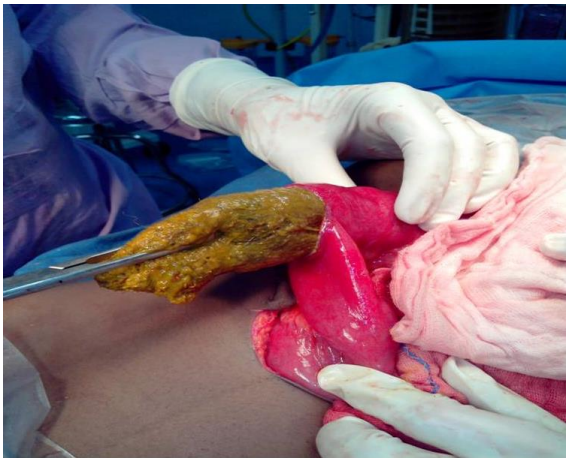

Fig-4: Intra operative photograph showing ileal cotton bezoar removed by enterotomy.

DISCUSSION

Bezoars are concretions of human or vegetable fibers that accumulate in the gastrointestinal tract. In humans, the most common type of bezoar is the trichobezoar, which is mostly made of hair. However, bezoars can also be made of vegetable or fruit fiber (phytobezoar), milk curd (lactobezoar), or any indigestible material[5, 6]. Rapunzel syndrome is a rare form of trichobezoar, and various criteria have been used in its description in the literature. Some define it as a gastric trichobezoar with a tail extending up to the ileocaecal junction; some describe it as a simple trichobezoar with a long tail, which may extend up to the jejunum or beyond[5, 6,]. Our case is rare presentation of Rapunzel syndrome in which gastric part of bezoar made up of human hair and its tail made up of synthetic fiber. Case reports of children with trichobezoar or Rapunzel syndrome are rare and many link the trichophagia to early childhood neglect or abuse[7] psychiatric conditions, mental retardation or bereavement[5]. In our case possible region was childhood neglect. The commonest phytobezoar encountered worldwide is related to the ingestion of the persimmon fruit[8]. Intestinal obstruction by cotton bezoar is unusual and a bezoar leading to perforation also a rare entity reported in literature. Our case presented with both of these complications of bezoars. The most common presenting signs are abdominal pain, nausea and vomiting. Intestinal obstruction and peritonitis are less common[5]. CT is a useful and powerful tool in the detection of small-bowel bezoars and its complications. In our case, CT scan showed gastric bezoar extending into duodenum and another bezoar in ileum with dilated proximal small intestine.

Operative management entails laparotomy and enterotomy with without resection anastmosis and is the main stay of treatment of small bowel bezoars, [2] as was required in our patient. We managed our case by doing exploratory laparotomy, gastrotomy, jejunal resection anastmosis, ileal enterotomy and loop ileostomy.

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