Trichobezoars are a rare cause of intestinal obstruction seen mainly in young females having some form of psychiatric illness. They usually present as hair balls found inside the stomach but sometimes a tail of these hair extends into the intestine which is called Rapunzel’s Syndrome. We had a female child with a history of trichotillomania and trichophagia who presented with intestinal obstruction. The hair tail had broken and got stuck in the jejunum hence the patient presented with intestinal obstruction which is in contrast to the more common presentation of trichobezoars i.e. gastric outlet obstruction.
INTRODUCTION:
Trichobezoars are foreign bodies composed of balls of hair and indigestible fibrous material which are commonly found in the stomach but can extend into the duodenum through pylorus and even up to jejunum, ileum and colon. They are usually associated with a history of some psychiatric or developmental disorder where there is history of trichotillomania and trichophagia. [1] Trichotillomania is included in OCD [2] (Obsessive compulsive Disorder), which is almost exclusively present in girls and younger women. [3] Hair are resistant to digestion. Ingested hair mix with food and mucous present in stomach and lead to trichobezoar formation. Trichobezoars may not be recognized in the early stages because of their non specific presentation or even lack of symptoms. The most common presenting symptom of trichobezoar present in GI (Gastrointestinal) tract is palpable abdominal mass, vomiting, gastric outlet obstruction, intestinal obstruction and noticeable hair loss. It should always be kept as a differential diagnosis in patients with similar symptoms when a girl or a young woman presents with history of trichotillomania and trichophagia. [1] Continuous ingestion of hair leads to enlargement of trichobezoar with possible complications which include gastric erosion, perforation of stomach and small intestine, intussusception, protein losing enteropathy, intestinal obstruction, pancreatitis and even associated with a lethal outcome. [4,5] Following the introduction of minimally invasive surgery and endoscopy with mechanical and laser fragmentation techniques, some authors have questioned the necessity of laparotomy. [6,7,8]

Still, in few selected cases with complications secondary to these trichobezoars, laparotomy is the only choice left. Rapunzel syndrome is an uncommon presentation of trichobezoar, involving strands of ingested hair extending as a tail, beyond the stomach into the small intestine. [9] We hereby present a case where a young girl presented with features of acute intestinal obstruction secondary to a jejunal trichobezoar in addition to gastric trichobezoar, but the gastric bezoar was not causing any symptoms.

CASE REPORT
A 12 year old female, presented to the Emergency Department (ED) with history of colicky pain abdomen, mild abdominal distension, two episodes of vomiting and non passage of flatus and stools for 2 days. She was referred from a local primary health centre. There was history of Appendicectomy 2 years back but no history of any psychiatric or developmental disorder and child was fair at studies as well. On examination, Pulse Rate (PR) was 96 beats per minute and Blood Pressure (BP) was 100/60 Millimetre of mercury (mm of Hg). There was mild fullness in epigastrium but grossly there was no distension in rest of the abdomen. There was no visible peristalsis. A lump of size 4X4 cm (Centimetre) was present in right iliac fossa region, which was firm, non tender and freely mobile in all directions. There was no other lump or organomegaly present. Laboratory findings revealed hypochromic anaemia with haemoglobin levels of 8.6 g/ dl (grams per decilitre). There was no electrolyte disturbance and both, liver and kidney function tests were within normal limits. Plain erect abdominal radiograph showed few air filled dilated jejunal loops and a suspicious radio-opacity in the dilated stomach (Figure 1). Abdominal USG (Ultrasoundogram) showed thickened proximal bowel loops with minimal interloop fluid. CECT (Contrast Enhanced Computerized Tomography) scan revealed intraluminal mottled appearance with debris in stomach and small bowel, suggestive of bezoars with dilated stomach and duodenum with narrowing at mid jejunal region and minimal free fluid in abdomen (Figure 2). On taking a repeat history, child admitted trichophagia but there was no history of trichotillomania. So, on the basis of history and radiological examination, a diagnosis of acute intestinal obstruction secondary to trichobezoar was made and the patient was taken up for explorative laparotomy. On opening the abdominal cavity through an upper midline incision, about 200 ml (millilitre) of serous fluid was present. Jejunum was moderately dilated up to about 60 cm from duodenojejunal junction. There was an area of hyperaemia at that point underneath which a firm FB (Foreign Body) could be felt. It was not possible to move this FB distally but it could be moved in proximal direction. It was moved proximally for about 30 cm in an apparently healthy bowel where after taking stay sutures, jejunum was opened. A trichobezoar of size 6X3 cm was retrieved, which was black and oval shaped. There was no tail present (Figure 3). After thoroughly washing the area with normal saline, opening in the jejunum was closed transversally. Stomach was palpated for other trichobezoar. It was not present in distal body, antrum or pyloric region. On palpation more proximally, it could be felt higher up in proximal body and GE (Gastro-esophageal) junction. After taking stay sutures, an incision of about 5 cm was given in anterior gastric wall in body region, along its long axis. A trichobezoar was retrieved which was about 14X6 cm with a tail (Figure 3). Meticulous search was done for other free bezoars in rest of the gut. Opening in the stomach was closed in 2 layers with absorbable suture. The patient had an uneventful recovery.
Psychiatric consultation was taken and she was discharged on 7th post operative day. The patient is still in follow up.

Figure 1: Showing radio-opacity inside the stomach and dilated jejunal loops

Figure 2: Showing foreign bodies within the dilated stomach and the jejunum.

DISCUSSION
Bezoars are composed of hair, animal or plant fibres, minerals and medicaments etc, which are accumulated in the GI tract, mostly in the stomach and rarely in the small intestine. Bezoars composed of hair or hair like fibres are called trichobezoars. These, unlike other bezoars are not associated with alterations in GI motility. They are almost exclusively seen in women with only isolated cases reported in men. They commonly occur between the age group of 13-20 years. Trichobezoars usually occur in patients with psychiatric disorders who usually have a history of trichotillomania and trichophagia. Trichotillomania is a behavioural disorder which implies compulsive hair pulling. Hair is not only pulled out from the scalp, but also from eyelashes, eyebrows, pubic region and other body parts as well. It can lead to alopecia. Approximately 5%-18% of patients who suffer from trichotillomania develop trichophagia (swallowing hair), which leads to potentially serious complication, trichobezoar formation. Approximately 37.5% of patients suffering from trichophagia will ultimately form trichobezoar. Rapunzel syndrome is a rare form of trichobezoar formation, which was first described by Vaughan in 1968, as a tail like extension of gastric trichobezoar into the small intestine. The symptoms and physical signs that characterize Rapunzel syndrome depend on the size and location of trichobezoar as well as presence of complications. Most common symptoms and signs associated with this disease are abdominal pain, nausea and vomiting, obstruction and peritonitis. Uncommonly, patients have also presented with weight loss, anorexia, haematemesis and intussusceptions. Less than 40 cases of Rapunzel Syndrome have been reported in the literature.
The most common complication is intestinal obstruction, whereas perforation and peritonitis result in death in approximately 30% of cases. An abdominal mass is also a common sign which is usually felt in epigastrium and umbilical region, but in this case mass was located near right iliac fossa. Laboratory findings confirm hypoproteinemia and anaemia as the most common manifestation of Malabsorption Syndrome. Various imaging modalities can be used for diagnosis of trichobezoars. Plain abdominal radiograph is helpful as it can pick up radio opaque foreign bodies as well as features suggestive of intestinal obstruction. The presence of multiple acoustic interfaces created by trapped air and food, limits the use of USG for trichobezoars. Upper GI Endoscopy is a method of choice for diagnosing trichobezoars. CECT abdomen is also very useful in diagnosing a trichobezoar. It is seen as ovoid, intraluminal heterogeneous mass occupying almost entire lumen. A typical CT finding shows air bubbles within the mass at the same time it can rule out the other intra-abdominal pathologies. Small trichobezoars can be removed through endoscopy after fragmentation. However, in most cases it is not possible due to the size and its content. Also, when patient presents with more than one trichobezoar which includes the one that have gone beyond the duodenum after it is detached from the main mass, poses challenge for treatment which was evident in this case. In such cases, laparotomy is the procedure of choice, which allows careful examination of whole GI tract. most common site of foreign bodies getting stuck inside the small gut is terminal portion of ileum, usually about 2 feet proximal to ileocecal junction (ICJ). In this case the trichobezoar was stuck in jejunum which is quite unusual. The patient presented with symptoms of intestinal obstruction rather than gastric outlet obstruction, and the broken Rapunzel’s tail was found in the jejunum rather than terminal part of ileum.

CONCLUSION

Rapunzel syndrome is a rare entity. Trichobezoar should always be kept as a differential diagnosis in all young women who present with abdominal pain, nausea and vomiting, palpable abdominal mass and psychiatric disorder. Treatment modalities depend on size, site and any possible complications related to trichobezoars, but most of the cases demand surgical treatment. The treatment is not complete without psychiatric evaluation of the patient.

CONSENT

Written informed consent was obtained from the patient. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

REFERENCES


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