Case Report

Trichobezoars: a hairy cause of intestinal obstruction

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ABSTRACT

Trichobezoars are an infrequent form of bezoar found in the stomach or intestine, created from ingested hair. Their most frequent location is in the stomach but they may extend through the pylorus into the small bowel. This condition is known as Rapunzel syndrome. Trichobezoar with Rapunzel syndrome is an uncommon diagnosis in children. Authors report 3 cases of Rapunzel syndrome with a large bezoar in paediatric age group that presented with features of acute intestinal obstruction. They were managed with surgical exploration.

Keywords: Bezoar, Rapunzel, Trichobezoar, Trichophagy

INTRODUCTION

Bezoars are conglomerates of food and fiber in the gastrointestinal tracts. These are encountered after gastric surgeries in adults, due to impaired gastric emptying and decreased acid production, phytobezoars being the commonest. But, in children, trichobezoars predominate & are associated with pica, mental retardation and co-existent psychiatric disorders. Trichobezoars are rare conditions that consist of hair bundles in the stomach or small bowel. Though they can be found anywhere in the GI tract but stomach is the commonest site. A rare type of trichobezoar is the Rapunzel syndrome in which the bezoar extends in the small intestine. The prevalence rate varies from 0.06% to 4%.¹ The bezoars although rare, when undiagnosed can lead to complications such as ulcers, gastric bleeding or perforation and obstruction.²³ Though a rarity, watchfulness is to be exhibited while managing patients, especially children with acute intestinal obstruction.

Authors describe three cases of paediatric age group presenting with acute intestinal obstruction secondary to Rapunzel syndrome.

CASE REPORT

Authors came across 3 cases of intestinal obstruction at our hospital in Bihar, which presented with typical clinical features like abdominal pain, epigastric discomfort, vomiting, constipation and obstipation. With a palpable mass on per abdominal examination. All our 3 cases were in the paediatric age group with 2 girls aged 11 yrs and 8 yrs, and a boy aged 6 yrs. None of them had history of any surgical procedures performed in the past, medical history or any significant drug history. One of the girls aged 8 years had history of psychiatric disorder and her mother reported that she had a habit of hair pulling and chewing, but without any evidence of alopecia.

On general examination, vital signs were normal, higher mental functions were normal and they did not show any signs of depression, anxiety or mental disorder on admission. Physical examination revealed a palpable mass in the epigastric region in 2 of the cases. Abdominal distension, diffuse tenderness with hypoactive bowel sounds were present in all.
A plain erect abdominal radiograph was done, which showed multiple air fluid levels suggesting intestinal obstruction. Ultrasound of the abdomen showed an intraluminal mass with interspersed air in the first case and a hypoechoic small bowel mass, with no confirmed origin, in the other two cases.

Barium meal follow through was done in all the cases since computed tomography was unavailable due to the rural setup of the hospital. Barium meal showed Intraluminal mottled mass outlined by gas, distended stomach/bowel loops and filling defects without bowel wall attachment.

Based on the clinical features, laboratory findings and radiographic evidence, decision for emergency exploratory laparotomy was taken. The intraoperative findings were consistent with trichobezoar with huge clumps of hair interspersed with mucus & fecal matter, retrieved on enterotomy from ileum in two cases and jejunum in one. Primary closure was done in all the three cases. The patients were discharged on the 10th post-operative day after a psychiatric consultation and counselling. There were no post-operative complications. They were followed up for 2 years in the outpatient department of our hospital with no recurrence of abdominal pain, vomiting or constipation.

**Table 1: Summary of the cases presented with trichobezoar.**

<table>
<thead>
<tr>
<th>Feature</th>
<th>Case 1</th>
<th>Case 2</th>
<th>Case 3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age</td>
<td>11</td>
<td>8</td>
<td>6</td>
</tr>
<tr>
<td>Sex</td>
<td>F</td>
<td>F</td>
<td>M</td>
</tr>
<tr>
<td>Features of intestinal obstruction</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>Palpable mass</td>
<td>Yes</td>
<td>No</td>
<td>Yes</td>
</tr>
<tr>
<td>Known psychiatric illness</td>
<td>No</td>
<td>Yes</td>
<td>No</td>
</tr>
<tr>
<td>X-ray abdomen</td>
<td>Multiple air fluid levels</td>
<td>Multiple air fluid levels</td>
<td>Multiple air fluid levels</td>
</tr>
<tr>
<td>Usg Abdomen</td>
<td>Intraluminal mass</td>
<td>Hypoechoic mass</td>
<td>Hypoechoic mass</td>
</tr>
<tr>
<td>Intraoperative finding</td>
<td>TBZ in ileum</td>
<td>TBZ in jejunum</td>
<td>TBZ in ileum</td>
</tr>
<tr>
<td>Post operative course</td>
<td>Uneventful</td>
<td>Uneventful</td>
<td>Uneventful</td>
</tr>
</tbody>
</table>
DISCUSSION

Trichobezoar is a conglomeration of swallowed hair. This rare condition is usually located in the stomach but it may extend through the pylorus into the small bowel and colon. Rapunzel syndrome is an unusual and rare form of trichobezoar extending into the small intestine. The name “Rapunzel” syndrome comes from the Grimm Brothers’ fairy tale of a 12-year-old princess who was shut into a tower with neither stairs nor doors by an enchantress who climbed up the tower’s walls with the help of Rapunzel’s long tresses.  

The first reference to a bezoar in a human was in 1779 during an autopsy of a patient who died from gastric perforation and peritonitis.  

Trichobezoars are most commonly found in children and adolescents, but all age groups may be affected. Naik et al. reported a mean age of 10.8 years, and 96% were female patients. In our study the mean age is 8.3 years, 66.66% being females. Trichobezoars are typically seen in women in their 20s and are often associated with psychiatric disorders. The most common symptoms include abdominal pain, nausea, vomiting, early satiety, anorexia, and weight loss. More severe manifestations of this entity depend on trichobezoar location. Intestinal obstruction in the terminal ileum is uncommon and may cause ischemia and perforation. Overall, gastrointestinal obstruction has been documented in 26% of the patients, and perforation in 18%.  

During the last few years, trichobezoar cases have attracted debate about the application of minimally invasive techniques, such as endoscopy and laparoscopy, rather than laparotomy, as well as medical treatment and enzymatic degradation, which are attractive because of their non-invasiveness but have been reported to be ineffective. Open surgery or laparotomy has been the treatment of choice for large trichobezoars. Unfortunately, surgery may have postoperative complications, such as perforation, pneumonia, bleeding, intussusception, wound infections, or unsightly scarring. Laparoscopic-assisted techniques have been suggested to reduce the complication rate. Due to unavailability of CT scan and endoscopy at our centre, we relied on clinical diagnosis for the management of our patients. We performed an explorative laparotomy as an emergency operation to save our patient’s life. However, despite laparotomy in all our three cases, the post-operative period was uneventful.

Gorter et al, in a retrospective review of 108 cases of trichobezoar, evaluated the available management options it was noted that whereas 5% of attempted endoscopic removals were successful, 75% of attempted laparoscopies were successful. However, laparotomy was 100% successful and thus favoured as their management of choice. Due to the high success rate, low complication rate, and the ability to carefully examine the entire gastrointestinal tract, laparotomy is still considered the treatment of choice in our centre.

After trichobezoar removal, prognosis is good if there is prevention of recurrence. To prevent recurrence patients should be encouraged to seek psychiatric opinion, modify their diet, chew adequately and drink plenty of fluid.

CONCLUSION

Due to rarity of Trichobezoars, it gets overlooked frequently. Hence, there should be a high index of suspicion while dealing with age group. Large trichobezoars including Rapunzel syndrome are removed surgically.

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REFERENCES
