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Original Article

4 years institutional experience in the management of Trichobezoar: About 5 Cases

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Abstract

Background: *Trichobezoar is an uncommon clinical entity in which ingested hair mass accumulates within the digestive tract. Trichobezoars are usually located in the stomach, but may extend through the pylorus into the duodenum and small bowel (Rapunzel syndrome). This entity is usually associated with trichotillomania and trichophagia or other psychiatric disorders. It is generally observed in children and young females with psychological disorders. Untreated cases may lead to grave complications. We present our experience with five patients and provide a review of the recent literature.*

Material and Methods: *We retrospectively analysed the clinical data of five patients treated for trichobezoar in pediatric surgery department during 4-year-period between 2018 and 2022. Their medical charts were reviewed with respect to symptoms, diagnostic procedures including any Imaging (abdominal ultrasonography and abdominal computed tomography) or upper GI endoscopy and treatment.*

Results: *Our study involved 5 girls aged 7 to 12 years. All cases presented with symptoms of epigastric pain associated with vomiting of recently ingested food and weight loss in three cases. Physical examination revealed a hard epigastric mass in all cases. The trichobezoar was confined to the stomach in 4 cases. An extension into the jejunum was observed in 1 case. All patients were surgically treated. In one case, the attempt of endoscopic extraction failed and patient was later operated. On surgical exploration gastrotomy was done in all the patients to extract the hair bezoar even those with jejunal extension. All patients proved to suffer from trichophagia and referred to a child psychiatrist or psychologist for regular follow-up. No recurrences have been reported.*

Conclusion: *According to our experience and in line with the published results, conventional laparotomy is still the treatment of choice. The literature provides no evidence of superiority of endoscopy or laparoscopy. After successful treatment, psychiatric consultation is imperative to prevent recurrence and improve long term prognosis.*

Keywords: *Rapunzel syndrome, Trichobezoar, Trichotillomania, Gastrotomy, Endoscopy.*

Introduction

Trichobezoar consists of a ball of ingested hairs in the proximal gastrointestinal tract, is a rare condition essentially observed in teenage girls that have behavioural disorders[1-5]. Human hair is resistant to digestion and it accumulates between the mucosal folds of the stomach. Over a period of time, continuous ingestion of hair leads to the impaction of hair together with mucus and food, causing the formation of a trichobezoar. In most cases, the bezoar is confined to the stomach.Rapunzel syndrome is a rare form of gastric trichobezoar that develops through bezoar extension from the stomach to the intestine[6,7]. In addition, incidentally, parts of the tail can break off and migrate to the small intestine, causing intestinal obstruction [8,9,10]. In the early stage, most trichobezoars may not be recognized due to their nonspecific presentation or even lack of symptoms in the early stages. The diagnosis is established either endoscopically or radiologically. In the current article, we report our experience with management of trichobezoar in five patients who were treated in our department during the 4-years period.

Patients and Methods

We retrospectively reviewed the clinical records of all patients treated for trichobezoar in paediatric surgery department during the 4-year period between January 2018 and January 2022. During this period, five patients were treated for trichobezoar in our center. Their medical charts were reviewed with respect to epidemiological data, clinical symptoms, diagnostic findings, treatment and outcomes.

Results

Our study involved 5 girls aged 7 to 12 years. Median age of presentation was 8 years(Range 7-12 years). Median weight at presentation was 16 kg (Range 10-30 Kg). Common presenting features in our series were abdominal pain, vomiting, constipation and weight loss.Symptoms of epigastric pain associated with vomiting of recently ingested food, early satiety and constipation were present in all cases while weight loss was present in three cases. One patients had acute presentation with upper abdominal distension. After reviewing the records carefully, two patientswere found to have trichophagia and one had trichotillomania. Physical examination revealed a hard epigastric mass in all patients and two patients were found anaemic.**[Table.1]** Xray abdomen revealed haziness in upper abdomen in all patients but none revealed any air fluid level. The computed tomography (CT) was the main diagnostic modality. It underlined a gastric trichobezoar in four cases and an extension to the jejunum in one case which defined Rapunzel syndrome. In addition, the upper GI endoscopy was performed in one case that revealed an intraluminal gastric mass made of hair. However, the attempt at endoscopic extraction failed because of the large size of the mass. Surgical extraction was performed by gastrotomy in all cases. The bezoar was successfully extracted in one piece including those with jejunal extension andno evidence for detached parts of bezoar distally within the intestine, so no additional enterotomy was done. **[Figures 1 and 2]**. The gastrotomy was closed in two layers and the abdomen was closed with drainage. Clinical presentation and treatment is summarised in **Table.2**

The post-operative follow-ups were uneventful. Postoperatively all patients recovered well, allowed orally on an average 5th post-op day, skin stitches were removed on 10th post-op day and all patients discharged from hospital in satisfactory condition. Follow-up in child psychiatry was indicated. After recovery, all patients were referred to the psychiatry department and were diagnosed with trichophagia in all cases. A treatment plan comprising pharmacological and psychotherapeutic interventions and close follow-up was initiated. No case of recurrence was underlined in our series.

Table 1 Clinical features of study population

Clinical Features	Frequency (%)
Abdominal Pain	5 (100%)
Vomiting	5 (100%)
Epigastric Lump	5 (100%)
Constipation	5 (100%)
Early Satiety	5 (100%)
Weight loss	3 (60%)
Trichophagia	2 (40%)
Trichotillomania	1 (20%)
Intestinal Obstruction	0 (0%)
Abdominal Distension	1 (20%)
Bleeding PR	0 (0%)

Table.2 Trichobezoar clinical presentation and management

S.N	Age	Sex	Clinical Presentation	Imaging	Location	Therapeutic Modality
1.	7Y	Female	1,2,3,4,5	CECT, USG	Stomach	Gastrotomy
2.	8Y	Female	1,2,3,4,5	CECT, USG	Stomach	Gastrotomy
3.	8Y	Female	1,2,3,4,5,6	CECT, USG	Stomach	Gastrotomy
4.	11Y	Female	1,2,3,4,5,6,7,9	CECT, USG	Stomach, duodenum, jejunum	Gastrotomy
5.	12Y	Female	1,2,3,4,5,6,7,8	CECT, USG, UGIE	Stomach	Endoscopic removal attempt then gastrotomy

1 Abdominal Pain, 2- Vomiting, 3- Epigastric Lump, 4- Constipation, 5- Early Satiety, 6- Weight loss, 7- Trichophagia, 8- Trichotillomania, 9- Abdominal Distension

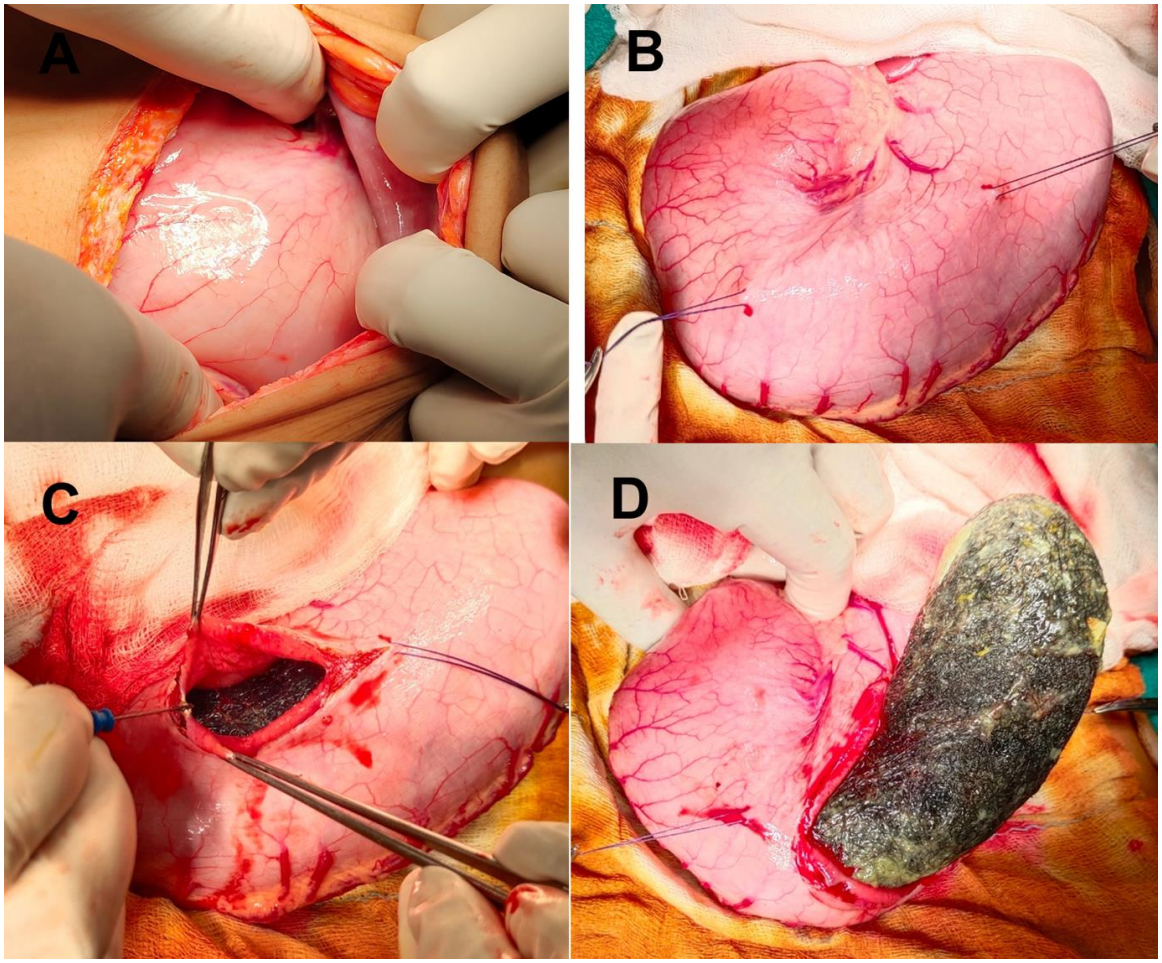

Figure.1 Intraoperative view showing the extraction of the trichobezoar. A-C. Gastrotomy through anterior wall of the stomach, D. Extraction of trichobezoar

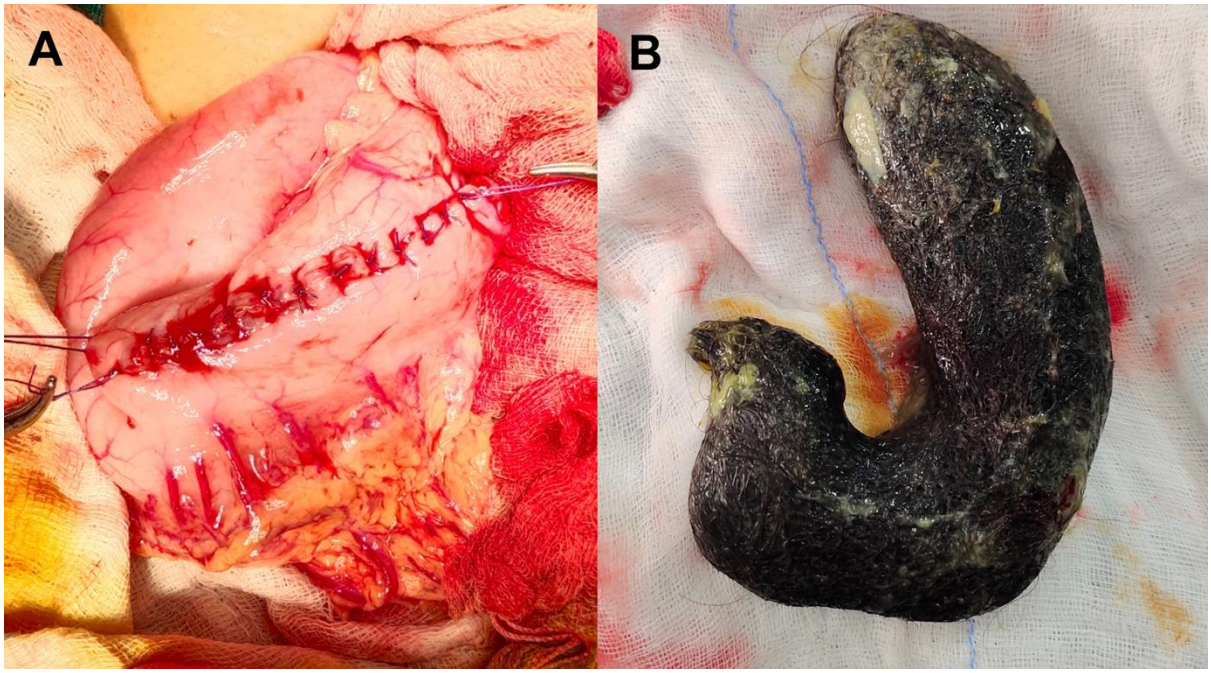

Figure.2 A.double layer closure of gastrotomy, B. The trichobezoar after being extracted

Discussion

Bezoars are collections of nondigestible matter that usually accumulates in stomach and can extend to small bowel. Different types of bezoars are Phytobezoar (vegetable origin), Trichobezoar (composed mainly of hair), Lactobezoar (concentrated milk formula), Pharmacobezoar (mixed medicine bezoars), and food bolus bezoars. Bezoars typically develop in the stomach and the small intestine. While gastric bezoars are more common, intestinal bezoars are more likely to be revealed by bowel obstruction.[6,7] Trichobezoars are usually found in young psychiatric females, with the habit of eating their own hair (trichophagy). In some cases, however, the trichobezoar extends through the pylorus into jejunum, ileum or even colon. This condition, called Rapunzel syndrome, was first described by Vaughan et al., in 1968[6]. Bezoars may present with abdominal pain, nausea/vomiting, early satiety, weight loss, intestinal obstruction, ulceration leading to bleeding and/or perforation. Rarely intussusception can also happen[12,13]. They may remain asymptomatic or may present several digestive symptoms. Patients can present with abdominal pain, vomiting and constipation.[6,7,10] In our series abdominal pain, vomiting, early satiety, constipation and weight loss were the common symptoms.Early diagnosis is essential since obstructive bezoars may cause serious problems, including gastrointestinal (GI) ulceration, visceral perforation, bleeding and pressure necrosis.[12] The patients under study showed gastric trichobezoar and complained essentially about abdominal pain and vomiting. Furthermore, we did not have any case of bowel obstruction. An upper abdominal mass remains the commonest presenting sign[5]. In our series also all patients had palpable intraabdominal lump in epigastric region moving well with respiration and hard in consistency with curved (convex) lower border.

Diagnostic modalities include US, CT scan and upper endoscopy. CT scan has a high accuracy rate. The diagnosis is made easily at endoscopy. On CT, small intestinal bezoars classically appear as a well-defined intraluminal mass containing mottled gas.[11,14,15] We also diagnosed and confirmed our cases with US, CT and plain radiograph. Direct visualisation of the bezoar through upper GI endoscopy is the gold standard for imaging. It is used for both diagnostic and therapeutic purposes.[14,15] Gastric bezoar management mostly focuses on the dissolution or

elimination of the mass. It can be achieved either by endoscopically, or surgically. All the patients under study underwent surgical exploration while an attempt of endoscopic extraction was failed in one case. Gorter et al., in a retrospective review of 108 cases of trichobezoar, evaluated the variable treatments tried in these cases; it was noted that whereas 5% of attempted endoscopic removals were successful, 75% of attempted laparoscopic surgical extractions were successful. However, laparotomy followed by gastrotomy was 99% successful and thus favoured as their management of choice.[17] The majority of cases in the literature have been managed with surgical removal of the hair mass by laparotomy. The small bowel can be explored to look for detached bezoars. Hence, trichobezoar extensions may be extracted and intestinal segments which show extensive ulcerations or gangrene may be resected.[16] Endoscopy frequently fails to remove the trichobezoar, except if small in size, while successful extraction can be achieved by mechanical and laser hair fragmentation.[17,18] The conventional open surgery is still the preferred treatment method due to the very high success rate, shorter operative time, less complications and possibility to explore the whole GI tract.[17]

Treatment should also focus on prevention of recurrence, elimination of contributory condition including psychiatric evaluation and treatment as well as regular follow-ups.

Conclusion

The diagnosis of trichobezoar should be suspected in young girls with behavioural disorders and digestive symptoms. As far as treatment is concerned, however, we consider conventional laparotomy to be the treatment of choice in children with trichobezoar and to be the only valid treatment in children with Rapunzel syndrome. The literature provides no evidence of superiority of endoscopy or laparoscopy. After successful treatment, psychiatric consultation and treatment is imperative to prevent reoccurrence and improve long-term prognosis.

Declaration of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of Interest: There are no conflicts of interest.

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