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Gastrointestinal trichobezoar: An experience with 17 cases☆,☆☆

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ABSTRACT

Background: Trichobezoar is an exceedingly rare entity in children and mimics other chronic ailments such as abdominal tuberculosis or malignancy. Delayed diagnosis and management result in various complications. The study was conducted to describe our experience with 17 consecutive cases of gastrointestinal tract (GIT) trichobezoars.

Materials and methods: We reviewed medical records of 17 consecutive cases of GIT trichobezoar managed in our department between January 2005 and December 2018.

Results: There were 3 males and 14 females. The median age of presentation was 7 years. 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). At operation, 9 (54%) patients had Rapunzel syndrome, 6 (35%) had gastric trichobezoar, and 2 (12%) had small bowel trichobezoars.

One patient presented with massive bleeding per rectum and gastric perforation, succumbed postoperatively. One patient developed a recurrent trichobezoar.

Conclusion: GIT trichobezoar is rare in children and simulates chronic gastrointestinal ailments. Trichobezoars may reside in the alimentary tract, remain unnoticed for years, and become overt with the onset of complications. The majority of trichobezoars had a tail in our series. Life threatening complications can occur with delayed presentations.

Type of study: Case series.

Level of evidence: Level IV.

Bezoar is derived from Persian meaning 'antidote' historically being used to adsorb poisons from the gastrointestinal tract (GIT). These are indigestible, nonfood items, and classified based on their composition such as phytobezoar: collection of indigestible plant material [1] and trichobezoar: the collection of hair in GIT [2]. Trichobezoars usually reside in the stomach (gastric trichobezoar) [1] and rarely may pass to the small bowel (isolated small bowel trichobezoar) [2–4]. A trichobezoar may elongate to acquire a long tail, the Rapunzel syndrome, named after the novel character 'Rapunzel' with long tresses [5,6]. Vaughan et al. reported the first case of Rapunzel syndrome in 1968 [5].

Trichophagia is universal to these patients and a great deal of them also exhibit trichotillomania, a condition of irresistible desire to pull out one's own hair [7]. Trichobezoars may remain undiagnosed for years which can lead to severe malnutrition as a ramification of frequent emesis and early satiety [8]. Although well reported, its publications are limited to merely case reports and small case series [9]. We are presenting our 14-year experience of managing patients with GIT trichobezoars. We have managed some unusual presentations and complications which may help other pediatric surgeons while dealing with similar clinical scenarios. A management algorithm is also devised based on the pattern of clinical presentation.

1. Materials and methods

The medical records of 17 consecutive cases of GIT trichobezoars managed in our department between Jan 2005 and Dec 2018 were reviewed after taking permission from the Hospital Institutional Review Board. The data were reviewed for demography, clinical features, diagnostic workup, management, complications, and outcomes, and presented as median, frequencies, and proportions. One case was previously reported by our department in 2011 [8].

2. Results

Demography: Of 17 patients, 3 were males and 14 females (M:F 1:4.7). Median age of presentation was 7 years (IQR 6.5 years; range

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Table 1
Clinical features of study population.

Clinical features	Frequency (%)
Abdominal pain and vomiting	15 (88%)
Abdominal distension	8 (47%)
Palpable epigastric mass	7 (41%)
Intestinal obstruction	4 (23%)
Acute peritonitis	2 (12%)
Severe malnutrition	4 (23%)
Trichophagia	7 (41%)
Trichotillomania	6 (35%)
Hair in feces	7 (41%)
Bleeding PR	1 (6%)
Hypertension	1 (6%)
Generalized tonic clonic fits	1 (6%)

4–17 years). Median weight at presentation was 16 kg (IQR 5 kg, range 10–30 kg).

Presentation: Common presenting features in our series were abdominal pain, abdominal distension, and vomiting (Table 1). Six (35%) patients had acute presentations secondary to complications (intestinal obstruction in 4 patients and peritonitis in 2). Six patients (35%) had associated trichotillomania. In 7 patients (41%), a history of trichophagia was given. Median duration of trichophagia was 2 years, while 4 (23%) patients presented after several years of stopping trichophagia. In 7 patients (41%), passage of hair in feces was documented.

Diagnostic workup: Seven patients (41%) had the initial suspicion of abdominal tuberculosis or malignancy. Of these, one patient had already completed a 9-month course of antitubercular therapy (ATT), and another patient was started on ATT in a secondary level hospital. Abdominal radiographs (Fig. 1A) suggested an intragastric mass/bezoar in 7 (41%) cases, air fluid levels in 5 (30%) cases, and pneumoperitoneum in 1. Abdominal ultrasound showed a suspected gastric mass/bezoar in 5 cases (30%) and an intussusception in 1. In 4 (23%) patients, an upper GI contrast study or barium meal was performed to confirm a suspected gastric bezoar (Fig. 1B). In 5 cases (30%), upper GIT endoscopy confirmed a trichobezoar. In 4 (23%) patients, CT scan/MRI showed a gastric trichobezoar (performed in other hospitals suspecting abdominal TB/malignancy) (Fig. 2).

Eight (47%) patients were anemic at presentation and one patient had a deranged clotting profile. One patient, with severe pain abdomen, hypertension, and generalized tonic clonic seizures, had an elevated serum amylase and lipase. This patient underwent extensive workup including MRI brain, Doppler ultrasound for renal vessels, and multidisciplinary input from neurology, gastroenterology, and nephrology to ascertain the etiology of the seizures and hypertension, all of which were unremarkable.

Management: In 9 patients (53%), the preoperative diagnosis of trichobezoar was formed based on history, investigations, or endoscopy. The remaining patients were diagnosed intraoperatively. All patients underwent an open operation after clinical optimization, but 6 patients were operated on emergent basis. In two patients, endoscopic retrieval was attempted without any success.

At operation, 9 patients (54%) had Rapunzel syndrome (Fig. 3 A, B, C, D), 6 (35%) had gastric trichobezoar (Fig. 4 A, B, C), and 2 (12%) had isolated small bowel trichobezoars (Table 2). One patient also had ileoileal intussusception with gangrenous bowel secondary to the tail of trichobezoar as the lead point (Fig. 3A). This child was managed with resection of the gangrenous bowel and an ileostomy as the bowel was edematous, and a mucosal ulceration on the mesenteric side of proximal bowel (Fig, 5B) precluded primary anastomosis. One patient, presenting with an acute abdomen and massive bleeding per rectum, had gastric perforation secondary to the gastric trichobezoar (Fig. 4D). Table 2 describes our operative findings, surgical procedures, and outcomes of our patients. The patients operated in the last 3 years had relatively small incision laparotomies.

Complications: Seven (41%) patients presented with complications (intussusception and gangrene in 1, small bowel obstruction in 4, gastric perforation and massive bleeding per rectum in 1, acute transient pancreatitis, seizures, and hypertension in 1). One patient developed a small bowel anastomotic leak on the third postoperative day and underwent a second operation with creation of an ileostomy. This patient also developed a superficial wound infection which was managed with dressings. Moreover, 4 patients had severe malnutrition owing to long standing persistent vomiting prior to their presentation to us.

Outcomes: Median length of stay (LOS) was 6.5 days (IQR4, range 5–15 days). The patients operated electively had less LOS compared to those operated emergently. LOS was 15 days in our patient who had the anastomotic leakage. The patients were discharged and referred to psychiatric department after surgery. One patient (third-degree malnourished, cachexic) presented with massive bleeding per rectum and gastric perforation, succumbed postoperatively. One patient developed a recurrent trichobezoar 12 years after the initial trichobezoar removal. Another cachexic patient with intussusception and ganrenous bowel, managed with resection and ileostomy, gained considerable weight after retrieval of the trichobezoar. Stoma reversal has been perfromed in both patients in whom an ileostomy was fashioned. Five recently managed patients are on follow-up, with the rest of the patients lost to follow-up.

3. Discussion

GIT trichobezoars are exceedingly rare, constituting about 6% of all the bezoars. Commonly encountered in adolescent girls with psychiatric aliments or mental retardation; depression, anxiety, neglect, and sudden emotional event in the family are also not uncommon inciting events [7,9,10]. In our series, we were unable to identify any psychiatric

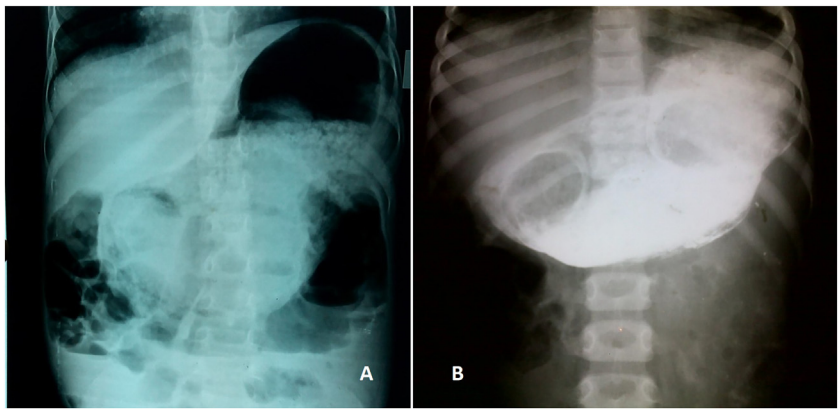

Fig. 1. (A) X-ray abdomen showing a gastric opacity. (B) Barium meal showing filling defect in the stomach indicating a bezoar.

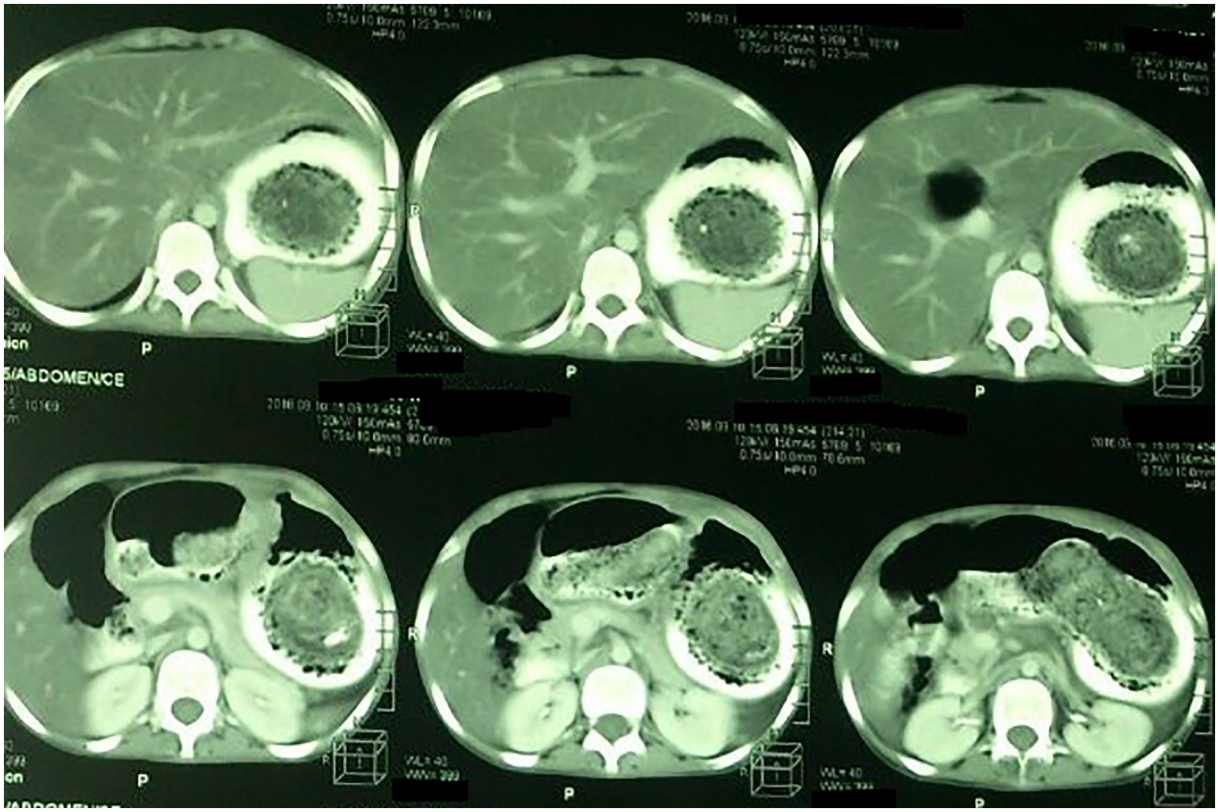

Fig. 2. CT scan abdomen showing intragastric bezoar.

Fig. 3. (A) Operative figure of patient with Rapunzel syndrome and intussusception, showing long trichobezoar and gangrenous small bowel (white arrow), after reduction of intussusception. (B) Operative figure showing a case of Rapunzel syndrome being retrieved from gastrotomy. The surgical wound around the stomach has been packed with povidone–iodine solution soaked gauzes to prevent spillage and wound infection. (C, D): Long trichobezoars after retrieval in other cases of Rapunzel syndrome.

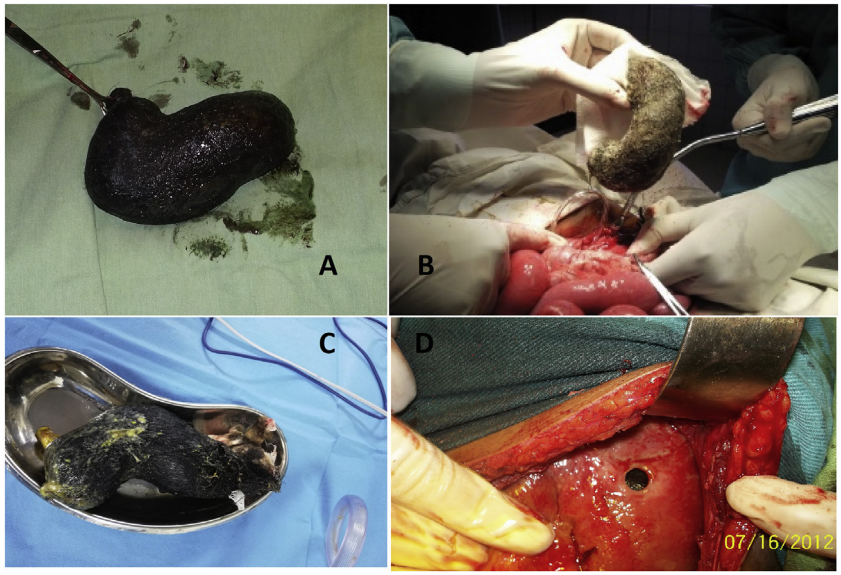

Fig. 4. (A–C) Gastric trichobezoar in various patients. (D) Showing gastric perforation owing to trichobezoar that can be seen through the rounded perforation.

ailments in our cases; nevertheless, 35% of patients had trichotillomania to the extent of alopecia. One parent kept their child bald for more than a year to try to break this habit. In our country, it is known that parents with a rural background are used to inserting hair in small holes and cracks in the walls of their houses. This practice facilitates their children finding another person's hair to eat, in addition to their own hair. Many of the children in our series also ate fabric fibers, paper napkins, plastic shopping bag pieces, and cloth pieces, etc.

Being indigestible, smooth, and slippery, hair can remain in the folds of stomach, intermixing with food and other fibers to form a gastric-shaped bezoar. The propulsive movements of the stomach can extend the part of trichobezoar to the small bowel to form the Rapunzel syndrome [6]. To classify for Rapunzel syndrome, the tail should be long enough to traverse at least a few loops of the bowel, although the exact length has not been objectively defined. Nevertheless, a few of our gastric trichobezoars had an inconspicuous tail, but we did not label those patients as Rapunzel syndrome. The development of a long tail signifies a long duration of trichophagia and the trichobezoar in the GIT. In our series, we encountered 9 patients (53%) with Rapunzel syndrome, and 6 patients (35%) with just a gastric bezoar.

On occasion, a part of Rapunzel syndrome or gastric trichobezoar may get detached from the main tail and form a satellite trichobezoar [11,12]. Only 1 patient (6%) in our series had a satellite small bowel trichobezoar in addition to the gastric trichobezoar. This patient presented with signs of small bowel obstruction, the cause of which was an intraluminal mass in the terminal ileum. The enterotomy divulged it as small bowel trichobezoar. On palpating the stomach, a gastric trichobezoar was also found. Thus, we recommend that whenever a trichobezoar is encountered in the small or large bowel, the surgeon should look for a gastric trichobezoar.

The bezoar tail of Rapunzel syndrome may remain asymptomatic for years but may produce complications such as intussusception, gangrene, volvulus, perforation peritonitis, and rarely transient pancreatitis [8,13–15]. Similarly, small bowel trichobezoars and satellite trichobezoars may produce small bowel obstruction [2]. In a review by Naik et al., about 25% of the patients presented with intestinal obstruction and 18% with peritonitis [9]. Similarly, in our series 24% of the patients presented with intestinal obstruction and 12% with peritonitis. In our series, many of the known complications were found. Additionally, we have observed mucosal ulceration on the mesenteric side of bowel on account of sustained pressure of trichobezoar. In one patient, the mucosal ulceration presumably extended to the outer bowel layers, causing a part of the jejunum to adhere on its mesenteric aspects, forming a U-shaped loop (Fig. 5A).

The clinical presentation depends upon the size, site, and duration of stay of the trichobezoar in GIT which, in turn, leads to the development of complications [8]. Three patterns of presentation were observed in our series: acute presentation with complications; insidious presentation with stable lesions; and incidentally found lesions in children misdiagnosed as other chronic maladies such as abdominal TB/malignancy. The majority of trichobezoars are destined to remain in the stomach. Thus they produce symptoms of a space-occupying gastric lesion, such as early satiety, nausea, and vomiting [9]. A prolonged stay in stomach may produce complications such as failure to thrive and malnutrition, gastric ulcerations and hemorrhages, and occasionally gastric perforation [16]. In our series, one adolescent girl presented with massive bleeding per rectum and signs of peritonitis, had a gastric perforation secondary to a gastric trichobezoar.

Table 2
Operative findings, surgical procedure done, and outcome.

Operative findings	Surgery performed	Outcome
Isolated gastric trichobezoar (4 pts)	Gastrotomy and removal	Alive
[a]Gastric trichobezoar with small bowel satellite trichobezoar (1 pt)	Gastrotomy and removal, milking out of small bowel trichobezoar through rectum	Alive
[b]Gastric trichobezoar with gastric perforation (1 pt)	Gastrotomy and removal with repair of gastric perforation	Expired
[c]Rapunzel syndrome (Gastric, small bowel) (7 pts)	Gastrotomy and removal	Alive
[a,d]Rapunzel syndrome (Terminal ileum, ascending colon) (1 pt)	Enterotomy and removal. Had anastomotic leak thus reoperated and ileostomy was formed	Alive
[b]Rapunzel syndrome with Intussusception (1 pt)	Gastrotomy and removal, resection of gangrenous bowel and ileostomy	Alive
[a]Isolated small bowel trichobezoar (2 pts)	Enterotomy and removal	Alive

[a] Presented with intestinal obstruction.
[b] Presented with peritonitis.
[c] One patient developed recurrent Rapunzel syndrome.
[d] Had anastomotic leak; thus, redo laparotomy was done and ileostomy formed.

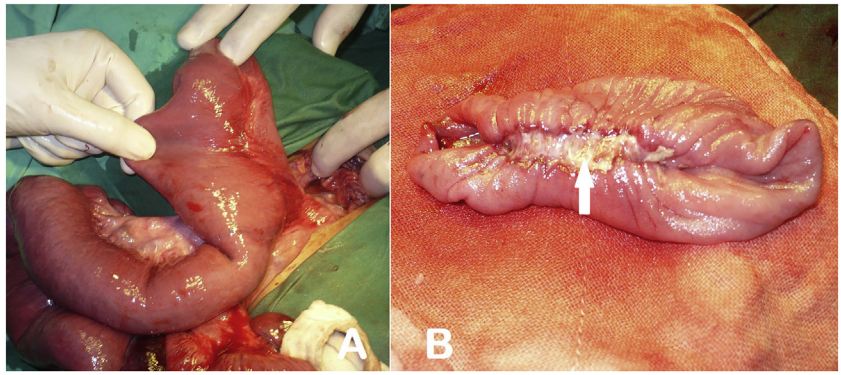

Fig. 5. (A) Operative figure showing U-shaped configuration of proximal jejunum in a case of Rapunzel syndrome with intussusception. (B) Showing mucosal ulceration on mesenteric aspect in the same patient. The part of bowel is resected at the level of midileum.

The preoperative diagnosis can be suspected based on a history of trichophagia, trichotillomania, passage of hair in feces, and a palpable mass in the epigastrium [17]. We were able to diagnose the trichobezoar as the cause of the symptoms in 53% of our patients. However, the lack of complications and a proper history may preclude making the preoperative diagnosis [18]. This was also true in our series, especially in patients with an acute presentation. The parents did not describe a history of hair ingestion because their children had already given up the habit of ingesting their hair for a considerable time before presentation and others were not considering hair ingestion as a cause of the clinical condition.

The diagnosis can be confirmed on radiological investigations. Plain radiographs often show a gastric shaped partly opacified area in context of a patient with trichophagia and a palpable mass. An ultrasound may help demonstrate an intragastric, heterogenous lesion consistent with a gastric bezoar. Contrast meal shows a faintly contrast laden filling defect. CT scan is a superior investigation as it not only identifies a heterogenous bezoar but can also define its extension [19,20]. Endoscopy is diagnostic in cases of gastric trichobezoar but its therapeutic value is still not established [21,22]. Not every patient requires all these diagnostic evaluations. Fig. 6 shows a diagnostic and treatment algorithm which can help select an appropriate diagnostic workup based on the pattern of clinical presentation.

Management goals are directed at removing the trichobezoar and preventing further trichophagia. Interventions in the form of open, laparoscopic, or endoscopic procedures have been described [8,21,23,24]. Open surgical removal is the most used technique for ease of removal and investigation for a possible small bowel tail or satellite trichobezoars. Small incision laparotomy has also been described [23]. Few authors have described laparoscopic and endoscopic retrieval with success [21,24]. Complicated cases seem better treated with an open approach [8]. In our series, we employed the open surgical technique for its retrieval. Endoscopic retrieval was attempted by our gastroenterologists but was not successful. The success rate of endoscopic retrieval is not promising to date. Over the last few years of practice, we have been using small incision laparotomies for retrieval of the trichobezoar, and the surgical incision is protected with transparent adhesive film and the incision margins are covered with povidone–iodine impregnated gauzes. The incidence of wound infection in our series was only 6%. Interestingly, on literature search, wound infection did not appear to be a major problem [25]. In the future, we plan to use laparoscopy for the management of uncomplicated bezoars (Fig. 6).

For prevention of recurrence, psychiatric evaluation and counseling should be done as a few cases of recurrent trichobezoar have been reported in literature [7,26]. We also had a case of recurrent trichobezoar in our series. Finally, the mortality is relatively low (less than 8%)

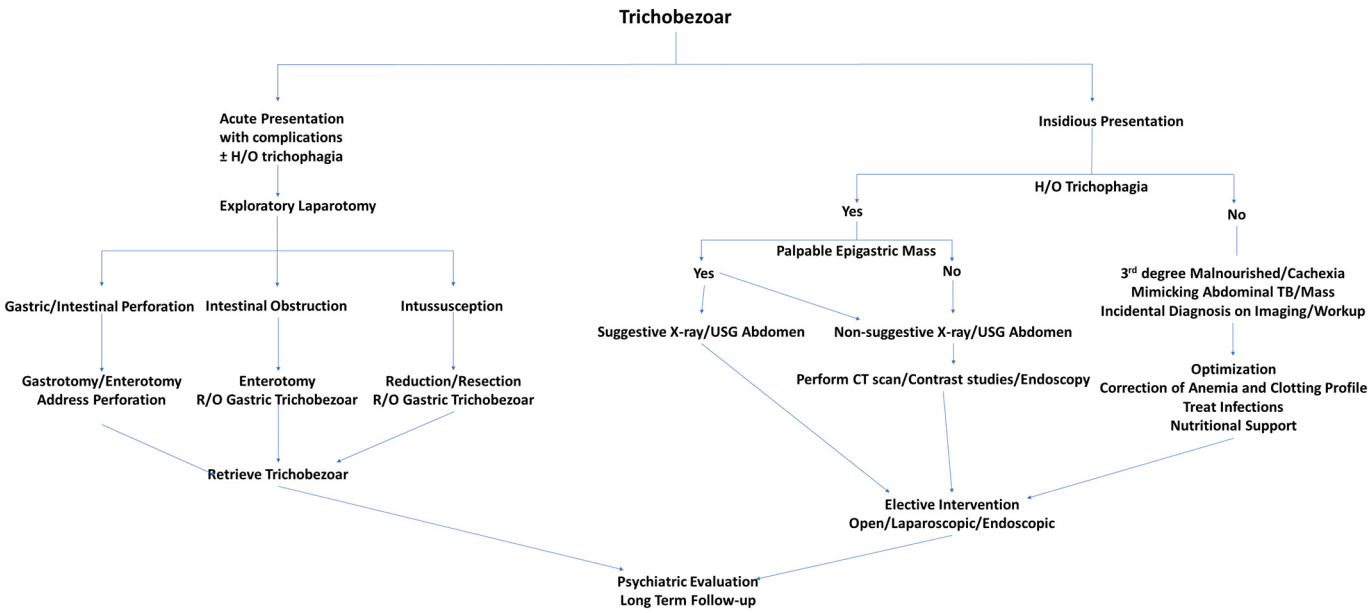

Fig. 6. Diagnosis and treatment algorithm of patients with trichobezoar. (H/O: history of; R/O: rule out)

and attributed to complications and septicemia [9]. We also had one mortality (6%) in our series.

4. Conclusion

GIT trichobezoars are rare bezoars. Common presentation is abdominal pain and vomiting. Complications are also common. The majority of the patients had the Rapunzel syndrome in our series. Early diagnosis and prompt surgical intervention after rapid clinical optimization help ensure a good outcome. Psychiatric evaluation and management are key to prevent further trichophagia and trichobezoars.

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