

Management of Trichobezoar: About 6 Cases

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Abstract

Background: Trichobezoar is an uncommon clinical entity in which ingested hair mass accumulates within the digestive tract. It is generally observed in children and young females with psychological disorders. It can either be found as an isolated mass in the stomach or may extend into the intestine. Untreated cases may lead to grave complications. **Material and Methods:** We retrospectively analyzed the clinical data of six patients treated for trichobezoar in Monastir pediatric surgery department during 16-year-period between 2004 and 2019. Imaging (abdominal computed tomography and upper gastroduodenal opacification) and gastroduodenal endoscopy were tools of diagnosis. **Results:** Our study involved 6 girls aged 4 to 12. Symptoms were epigastric pain associated with vomiting of recently ingested food in 3 cases and weight loss in one case. Physical examination found 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 2 cases. Surgery was indicated in all patients. In two cases, the attempt of endoscopic extraction failed and patients were then operated on. All patients had gastrotomy to extract the whole bezoar even those with jejunal extension. Psychiatric follow-up was indicated in all cases. The six girls have evolved well and did not present any recurrence. **Conclusion:** open surgery still plays a crucial role in Trichobezoar management. After successful treatment, psychiatric consultation is imperative to prevent recurrence and improve long term prognosis

Keywords: Pediatric surgery, rapunzel syndrome, trichobezoar

INTRODUCTION

Trichobezoar consists of ingested hair accumulating in the gastric mucosa folds instead of being digested.^[1,2] It is essentially observed in teenage girls that have behavioural disorders such as trichotillomania and trichophagia.^[3,4] In most cases, the bezoar is confined to the stomach.^[5] Rapunzel syndrome is a rare form of gastric trichobezoar that develops through bezoar extension from the stomach to the intestine.^[6] The diagnosis is established either endoscopically or radiologically. In the current article, we report our experience with management of trichobezoar. Six sample patients were treated in our unit during the 16-year- study period.

The purpose of the present study is to discuss the diagnosis, imaging and therapy of trichobezoars.

PATIENTS AND METHODS

We retrospectively reviewed the clinical records of all patients treated for trichobezoar in Monastir paediatric surgery

department during the 16-year period between January 2004 and May 2019. Epidemiological data, clinical symptoms, diagnostic findings, treatment and outcomes were analysed.

RESULTS

There were six girls aged 4 to 12 years. They were hospitalised for epigastric pain associated with food vomiting in three cases and weight loss in two cases. Two girls had trichophagia and one trichotillomania. Physical examination revealed a hard epigastric mass in all patients with partial alopecia in three cases. The upper gastrointestinal opacification performed in two patients showed an aspect in favour of a gastric trichobezoar. The computed tomography (CT) was the main diagnostic modality. It underlined a gastric trichobezoar in five cases

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and an extension to the jejunum in two cases which defined Rapunzel syndrome. In addition, the upper digestive fibroscopy performed in two cases highlighted an intraluminal gastric mass made of hair. However, the attempt at endoscopic extraction failed because of the large size of the mass. Bezoar surgical extraction was performed by gastrotomy in all cases. The bezoar was successfully extracted in one piece including those with jejunal extension [Figures 1 and 2]. Intraoperative findings revealed no evidence for detached parts of bezoar distally within the intestine, so no additional enterotomy was done. The post-operative follow-ups were uneventful. Follow-up in child psychiatry was indicated. After recovery, all patients were referred to the psychiatry department and were diagnosed with trichotillomania and trichophagia in all cases. Only one patient had been previously treated for trichotillomania. A treatment plan comprising pharmacological and psychotherapeutic interventions was initiated. All children were successfully managed with disappearance of the alopecia in three cases and progressive improvement of trichotillomania and trichophagia in all cases. No case of recurrence was underlined in our series.

Table 1 summarises the trichobezoar presentation and management in our series.

DISCUSSION

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.^[7] They may remain asymptomatic or may present several digestive symptoms.^[8] Patients can present with abdominal pain, vomiting and constipation.^[9] Early diagnosis is essential since obstructive bezoars may cause serious problems, including gastrointestinal (GI) ulceration, visceral perforation, bleeding and pressure necrosis.^[10]

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.

Trichobezoar diagnosis is based on imaging and often upper GI endoscopy.^[11] On CT, small intestinal bezoars classically appear as a well-defined intraluminal mass containing mottled gas. Intestinal loops are dilated proximally and collapsed distally.^[12] Direct visualisation of the bezoar through upper GI endoscopy is the gold standard for imaging. It is used for both diagnostic and therapeutic purposes.^[13,14] Gastric bezoar management mostly focuses on the dissolution or elimination of the mass.^[13] It can be achieved either medicinally, endoscopic, or surgically. With different rates of success and frequently multiple failed attempts, several studies have reported lavage and aspirate using large gastric tubes, hydrolytic solutions, or mechanical fragmentation with lithotripsy or electrosurgical knife.^[15] Chemical dissolution is an economical and non-invasive procedure, using agents that destroy bezoars, such as Coca-Cola® and acetylcysteine.^[16,17] The Coca-Cola® action may be due to its low pH, mucolytic effect of its high sodium bicarbonate concentration and carbon dioxide bubbles that improve dissolution.^[16] None of our series



Figure 1: Intraoperative view showing the extraction of the trichobezoar



Figure 2: The trichobezoar after being extracted: Rapunzel syndrome

Table 1: Trichobezoar clinical presentation and management

Age	Sex	Clinical presentation	Imaging-enoscopy	Location	Therapeutic modality	
1	4	Female	Ap, Am, vomiting constipation	CT	Stomach, duodenum, jejunum	Gastrotomy
2	6	Female	Ap, Am	CT, gastroduodenoscopy	Stomach	Endoscopic removal attempt then gastrotomy
3	8	Female	Ap, vomiting	CT, UGIO	Stomach, duodenum, jejunum	Gastrotomy
4	8	Female	Ap, Am	CT	Stomach	Gastrotomy
5	12	Female	Ap, Am	CT	Stomach	Gastrotomy
6	12	Female	Ap, weight loss	UGIO gastroduodenoscopy	Stomach	Endoscopic removal attempt then gastrotomy

Ap=Abdominal pain, Am=Abdominal mass, CT=Computed tomography, UGIO=Upper gastrointestinal opacification

underwent chemical dissolution because of the large size of the masses. The patients under study could not get better without surgery; attempted endoscopic extraction failed. A simple longitudinal gastrotomy was performed to remove the gastric mass. In addition, we managed to extract the trichobezoar in the two cases of Rapunzel syndrome using the same gastric incision. 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.^[18] The surgery can also be achieved by employing the hand-assisted laparoscopic technique. Endoscopy frequently fails to remove the trichobezoar, except if small in size, while successful extraction can be achieved by mechanical and laser hair fragmentation.^[19,20] 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.^[19]

Psychiatric evaluation and treatment as well as regular follow-ups is imperative to prevent trichophagia and recurrence.^[21]

CONCLUSION

The diagnosis of trichobezoar should be suspected in young girls with digestive symptoms associated with alopecia. While endoscopic extraction or laparoscopic surgical approach may be useful, open surgery still plays a crucial role. 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.

REFERENCES

1. Hamidi H, Mohammadi M, Saberi B, Sarwari MA. A rare clinic entity: Huge trichobezoar. *Int J Surg Case Rep* 2016;28:127-30.

2. Nettikadan A, Ravi MJ, Shivaprasad M. Recurrent Rapunzel syndrome – A rare tale of a hairy tail. *Int J Surg Case Rep* 2018;45:83-6.
3. Baheti AD, Otjen JP, Phillips GS. A hairy situation: Trichobezoar presenting with intussusception, and intestinal and biliary perforation in a child. *Radiol Case Rep* 2017;12:42-4.
4. Al-Janabi IS, Al-Sharbaty MA, Al-Sharbaty MM, Al-Sharifi LA, Ouhtit A. Unusual trichobezoar of the stomach and the intestine: A case report. *J Med Case Rep* 2014;8:79.
5. Hall JD, Shami VM. Rapunzel's syndrome: Gastric bezoars and endoscopic management. *Gastrointest Endosc Clin N Am* 2006;16:111-9.
6. Appak YÇ, Ertan D, Karakoyun M, Özyurt G, Özdemir T, Baran M. The cause of abdominal mass in a child with celiac disease: Rapunzel syndrome. A case report. *Sao Paulo Med J* 2019;137:292-4.
7. Soon YQ, Low HM, Huey CW, Wansaicheong GK. Clinics in diagnostic imaging (198). Small bowel obstruction secondary to a bezoar. *Singapore Med J* 2019;60:397-402.
8. Paschos KA, Chatzigeorgiadis A. Pathophysiological and clinical aspects of the diagnosis and treatment of bezoars. *Ann Gastroenterol* 2019;32:224-32.
9. Erzurumlu K, Malazgirt Z, Bektas A, Dervisoglu A, Polat C, Senyurek G, *et al.* Gastrointestinal bezoars: A retrospective analysis of 34 cases. *World J Gastroenterol* 2005;11:1813-7.
10. Abou Azar S, Wehbe MR, Jamali S, Hallal A. Small bowel obstruction secondary to a metamucil bezoar: Case report and review of the literature. *Case Rep Surg* 2017;2017:2702896.
11. Gayer G, Jonas T, Apter S, Zissin R, Katz M, Katz R, *et al.* Bezoars in the stomach and small bowel – CT appearance. *Clin Radiol* 1999;54:228-32.
12. Zissin R, Osadchy A, Gutman V, Rathaus V, Shapiro-Feinberg M, Gayer G. CT findings in patients with small bowel obstruction due to phytobezoar. *Emerg Radiol* 2004;10:197-200.
13. Khan S, Khan IA, Ullah K, Khan S, Wang X, Zhu LP, *et al.* Etiological aspects of intragastric bezoars and its associations to the gastric function implications: A case report and a literature review. *Medicine (Baltimore)* 2018;97:e11320.
14. Katsinelos P, Pilpilidis I, Chatzimavroudis G, Katsinelos T, Lazaraki G, Fasoulas K, *et al.* Huge gastric bezoar caused by honeycomb, an unusual complication of health faddism: A case report. *Cases J* 2009;2:7077.
15. Paschos KA, Chatzigeorgiadis A. Surgical and endoscopic treatment of a double phytobezoar causing ileus and jaundice: A case report. *Iran J Med Sci* 2019;44:70-3.
16. Ladas SD, Kamberoglou D, Karamanolis G, Vlachogiannakos J, Zouboulis-Vafiadis I. Systematic review: Coca-Cola can effectively dissolve gastric phytobezoars as a first-line treatment. *Aliment Pharmacol Ther* 2013;37:169-73.
17. Schlang HA. Acetylcysteine in removal of bezoar. *JAMA* 1970;214:1329.
18. Blejc Novak A, Zupančič Ž, Plut D, Gvardijančič D, Homan M. Rapunzel syndrome: A rare form of trichobezoar in the stomach with some extension into the small intestine. *Acta Dermatovenerol Alp Pannonica Adriat* 2018;27:155-7.
19. Gorter R, Kneepkens C, Mattens E, Aronson D, Heij H. Management of trichobezoar: Case report and literature review. *Pediatr Surg Int* 2010;26:457-63.
20. Palanivelu C, Rangarajan M, Senthilkumar R, Madankumar MV. Trichobezoars in the stomach and ileum and their laparoscopy-assisted removal: A bizarre case. *Singapore Med J* 2007;48:e37-9.
21. Bashir EA, Samiullah, Sadiq MA, Yusuf O, Karim K. Rapunzel syndrom. *J Ayub Med Coll Abbottabad* 2010;22:218-20.