

Case Report

Trichobezoar with duodenal perforation: a rare occurrence

Juin Yeen Ooi^{1*}, Kean Leong Koay², Lilius Li Hui Tang¹, Ramamoorthy Velayutham¹

¹Department of General Surgery, Hospital Raja Permaisuri Bainun, Ipoh, Perak, Malaysia

²Department of General Surgery, Hospital Serdang, Selangor, Malaysia

Received: 28 August 2022

Accepted: 30 September 2022

*Correspondence:

Dr. Juin Yeen Ooi,

E-mail: benjamin1193@hotmail.com

Copyright: © the author(s), publisher and licensee Medip Academy. This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

ABSTRACT

Trichobezoar is an intraluminal mass formed from the accumulation of undigested hair. It is a rare condition that is typically seen in young females with trichophagia and trichotillomania. When not recognized, it may present with complications such as obstruction, ulceration and in some rare cases, perforation. While most trichobezoar perforations are located in the stomach, however, duodenal perforation has not been reported before. We report a case of a 13-year-old girl who presented to the emergency with signs and symptoms of peritonitis and perforated viscus. Radiological investigation revealed a large heterogenous intraluminal mass with mottled gas pattern suspicious of bezoar. Oral contrast was seen tracking from the gastric pylorus into the peritoneum suggestive of a perforation. The girl was brought to theatre for emergency laparotomy and a trichobezoar was found extending from the stomach down to the level of D1. There is a perforation in D1 on its antero-superior surface measuring 1×1 cm. The bezoar was removed via a gastrostomy and the perforation was primarily repaired and patched with omentum. Post operatively, her recovery was complicated with wound breakdown and was subsequently discharged after 1.5 months of hospitalization with outpatient referral for psychiatry consults.

Keywords: Trichobezoar, Duodenal, Perforation

INTRODUCTION

Trichobezoar is a rare diagnosis that has been described as early as 1938.¹ It is typically seen in young females with trichophagia and trichotillomania. It results from the accumulation of undigested hair and may present with complications such as obstruction, perforation and in some rare cases; cholangitis and pancreatitis.² When perforation occurs, typically it occurs in the stomach. In this case report, we present a case of trichobezoar with duodenal perforation.

CASE REPORT

A 13-year-old Aboriginal girl with developmental delay presented to the Emergency with general feeling of unwell, reduced appetite, vomiting and abdominal discomfort for 2 weeks. Physical examination reveals a distended, generally tender abdomen with a hard palpable

mass at the left hypochondrium. On further history, the patient reported a habit of ingesting her own hair since childhood, and has not sought medical attention for this. An abdominal X-ray was unremarkable with no dilated bowels seen. A CT scan of the abdomen then revealed a large heterogenous intraluminal mass with mottled gas pattern suspicious of bezoar. There was a suspicious wall defect in the duodenum with pneumoperitoneum suggestive of a perforation.

After resuscitation, the patient was brought to the theatre for an emergency surgery. A midline laparotomy was performed. Intraoperatively, we noted gross contamination in the peritoneum with a D1 perforation on its antero-superior surface measuring 1×1 cm. The bezoar extends from the stomach down to the level of D1. A gastrostomy was made over the anterior wall of the stomach near the greater curvature to remove the

trichobezoar and the perforation was primarily repaired and patched with omentum.

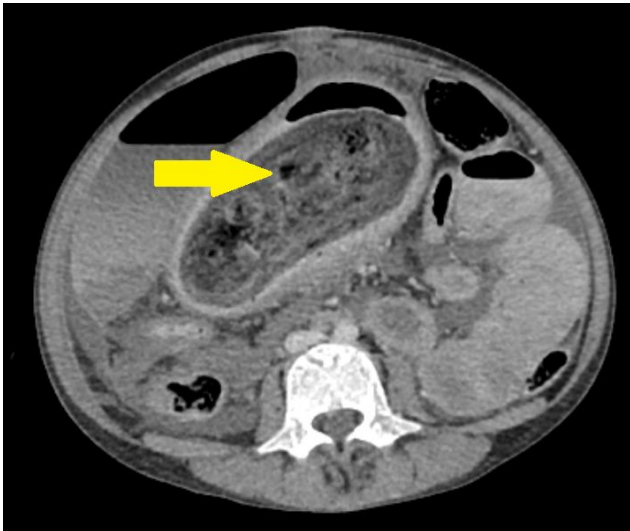


Figure 1: CT scan showing a large intraluminal heterogeneous mass arising in the stomach.

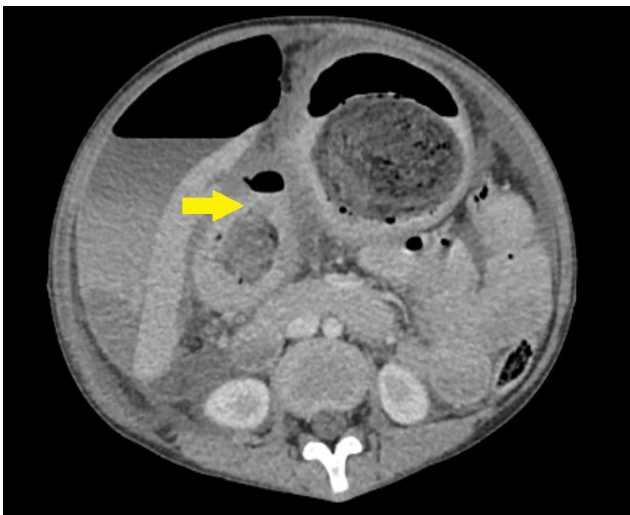


Figure 2: CT scan showing a suspicious wall defect at the duodenum with pneumoperitoneum.



Figure 3: Gastrostomy revealing a large trichobezoar.



Figure 4: Trichobezoar removed through the gastrostomy.

Post operatively, her recovery was stormy with multiple issues including sepsis, surgical site infection and wound breakdown and difficulty establishing feeding. After 1.5 months of hospitalisation, she was discharged and subsequently received psychotherapy consultations in the psychiatry outpatient clinic.

DISCUSSION

Trichobezoars are defined as foreign body from the accumulation of ingested hair and is most commonly found as a hard mass in the stomach. The true incidence is unknown due to its rarity, but typically, it is found in female patients with trichotillomania and trichophagia.³ They form as retained hairs in between the folds of the gastric rugae and then combines with food content to become an enmeshed mass. Patients with trichobezoars are usually asymptomatic for many years but may present with abdominal discomfort, vomiting and symptoms of gastric outlet obstruction. Physical examination is usually unremarkable as well except for a vague abdominal mass and halitosis. When a trichobezoar extends from the stomach to the small bowel (at least the jejunum), it is known as Rapunzel syndrome.⁴⁻⁶

Treatment modalities for uncomplicated trichobezoars include chemical dissolution with Coca-Cola, cellulase or papain. Failure of which, an endoscopic removal may be indicated by fragmenting the bezoar and direct endoscopic extraction.⁷ As the last resort, laparotomy may be used to remove the trichobezoar.

On the other hand, trichobezoar perforation is a very rare entity, and unique to this case report, based on our limited case report reviews, no cases of duodenal perforation have been reported in the literature. In the rare event of perforation, all cases reported were associated with stomach perforation.⁸ Based on the trichobezoar's size, density and hardness, it reduces blood supply to the mucosa of the stomach, resulting in ulceration and

leading to perforation.⁹ However similar explanation can also be used to explain the duodenal perforation in our patient, as she had post pylorus extension of bezoar to first part of duodenum, resulting in distension. The distension, with the force of propulsion from a chronically hypertrophied stomach with an incompetent pylorus, resulted in the duodenum taking the brunt of the force. This perforation would have been probably avoided if she developed Rapunzel syndrome instead.

The gold standard of treatment for trichobezoar with perforation is laparotomy.¹⁰ Laparotomy is widely regarded as the treatment of choice because of its high success rate, relatively low complication rate and low complexity along with the ability to carefully examine the entire gastrointestinal tract for satellites. In our patient however, her recovery was complicated with surgical site infection and wound breakdown. This is likely due to the nature of the surgery itself, which is contaminated.

A laparoscopic approach for removal of trichobezoars has been described as successful by a few authors.^{11,12} Whilst being superior in terms of cosmesis and shorter hospital stay, laparotomy was the only suitable treatment in our case as there was evidence of perforation. The ability to examine other parts of the GI tract to prevent secondary sites of bezoar obstruction also makes laparotomy a more favourable choice of treatment.

CONCLUSION

In conclusion, gastric trichobezoar should be considered in the diagnosis of perforated bowel, especially in those with history of trichophagia and trichotillomania. Trichobezoar with duodenal perforation is a rare entity and has not been reported to the best of our knowledge. Treatment modality of choice remains to be laparotomy in the presence of complications (i.e., perforation). Following the surgical removal of trichobezoar and repair of the perforation, long term follow-up with psychiatric consultation is important to prevent recurrence.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

REFERENCES

1. DeBakey M, Ochsner A. Bezoars and concretions. *Surgery.* 1939;4:934-63.
2. Nirasawa Y, Mori T, Ito Y, Tanaka H, Seki N, Atomi Y. Laparoscopic removal of a large gastric trichobezoar. *J Pediatr Surg.* 1998;33:663-5.
3. Coufal NG, Kansagra AP, Doucet J, Lee J. Gastric trichobezoar causing intermittent small bowel obstruction: report of a case and review of the literature. *Case Rep Med.* 2011;217570.
4. Marginean CO, Melit LE, Sasaran MO. Rapunzel Syndrome-An Extremely Rare Cause of Digestive Symptoms in Children: A Case Report and a Review of the Literature. *Front Pediatr.* 2021;9:684379.
5. Vaughan ED, Sawyers JL, Scott HW. The Rapunzel syndrome. An unusual complication of intestinal bezoar. *Surgery.* 1968;63(2):339-43.
6. Naik S, Gupta V, Naik S, Rangole AK, Jain P, Sharma AK. Rapunzel Syndrome Reviewed and Redefined. *Dig Surg.* 2007;24(3):157-61.
7. Kim SC, Kim SH, Kim SJ. Large Trichobezoar Causing Rapunzel Syndrome. *Medicine.* 2016; 95(22):e3745.
8. Tayyem R, Ilyas I, Smith I, Pickford I. Rapunzel syndrome and gastric perforation. *Ann R Coll Surg Engl.* 2010;92(1):e27-8.
9. Gorter RR, Kneepkens CMF, Mattens ECJL, Aronson DC, Heij HA. Management of trichobezoar: case report and literature review. *Pediatr Surg Int.* 2010;26(5):457-63.

Cite this article as: Ooi JY, Koay KL, Tang LLH, Velayutham R. Trichobezoar with duodenal perforation: a rare occurrence. *Int Surg J* 2022;9:1874-6.