CASE REPORT

Simplified technique for retrieval of large trichobezoars in children

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Accepted 6 May 2015

SUMMARY

Trichobezoars are concentrations of indigestible hair or hair-like fibres within the proximal intestinal tract. In children, delayed presentation with large bezoar masses is not unusual as bezoar formation is an indolent process that takes many months or years before becoming symptomatic. Surgical management is challenging and becomes inevitable once a trichobezoar becomes more established. The standard approach involves a sizeable transverse or midline laparotomy. We describe a less invasive technique for extraction of large gastric trichobezoars via a mini-laparotomy. The key aspect to this technique involves insertion of an Alexis O Wound Protector/Retractor (Applied Medical, Rancho Santa Margarita, California, USA) into the stomach following creation of a secure temporary gastrostomy by hitching gastrostomy edges to the abdominal wall. This simplified approach has advantages of (1) secure and excellent direct intragastric access, (2) shorter operating time and (3) reliable protection of both the wound edges and peritoneal cavity from bezoar contamination.

CASE PRESENTATION

A 5-year-old girl presented with chronic upper abdominal pain that had worsened over a period of weeks. Associated symptoms included an increasingly poor appetite. There was a long-standing history of trichotillomania, trichophagia and passage of hair in faecal matter. There had been one previous episode of vomiting a large bolus of matted hair. The child was systemically well with a weight that was on the 50th centile for her age. Clinical examination identified a large, firm, smooth, non-tender mass in the left upper quadrant and epigastrium. Blood tests confirmed low circulating iron (5 µmol/L) with normal iron stores (ferritin 54 µg/L). Haemoglobin (132 g/L), mean corpuscular volume (80 fL) and mean corpuscular haemoglobin (28 pg) were normal.

INVESTIGATIONS

There was one presentation to the emergency department with symptoms suggestive of partial gastric outlet obstruction. A plain abdominal X-ray identified a distended stomach that had the unusual appearance of a solid soft tissue organ. This finding prompted additional abdominal ultrasound and contrast swallow study imaging. Ultrasound demonstrated a 5.9 cm echogenic lesion in the stomach, with a linear echogenic tail that extended into the distal duodenum. Sonographic appearances were consistent with Rapunzel syndrome. A contrast swallow study identified a distended stomach with

Figure 1 Contrast swallow study image identifying a large intragastric lesion resulting in filling defect within a distended stomach. The bezoar is seen to form a cast appearance of the entire stomach, with the exception of the fundus that is occupied by an air bubble.

a large intragastric lesion resulting in a filling defect (figure 1).

TREATMENT

The parents were counselled regarding the need for surgical extraction of the large intragastric lesion, presumed to be a trichobezoar. The patient proceeded to undergo a mini-laparotomy via a 3.5 cm upper midline incision. Opposing stay sutures were inserted in the greater curve of the stomach to facilitate a transverse gastrostomy. The medial margin of the gastrostomy extended to the antrum. A 'small' size (2.5–6 cm) Alexis O Wound Protector/Retractor (Applied Medical, Rancho Santa Margarita, California, USA) was inserted into the stomach following creation of a secure temporary gastrostomy by hitching gastrostomy edges to the abdominal wall with interrupted polydioxanone sutures (figure 2). A 125 g gastric trichobezoar was observed that extended beyond the pylorus (figure 3). Following extraction of the specimen in its entirety, the stomach was irrigated with normal saline and gastrostomy was closed with a double-layered hand-sewn anastomosis. Total operating time was 121 min. A nasogastric tube was not required. The patient started oral intake on the first postoperative day; she was discharged on the second postoperative day after an uneventful recovery. Longer term

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To cite: Cundy TP, Brownlee EM, Goh DW, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2015-210472

Figure 2 Alexis O Wound Protector/Retractor in situ with (A) trichobezoar on view prior to (B) enmass extraction.

management included behavioural therapy to mitigate the risk of recurrence.¹

DISCUSSION

Trichobezoars are concentrations of indigestible hair or hair-like fibres within the proximal intestinal tract. Ingested hair is somewhat impervious to peristaltic propulsion and has a tendency to accumulate with other gastric content to form an impacted and enmeshed mass. The condition is rare, with less than 120 cases reported in the literature.² The demographic of patients is almost exclusively young females.² The condition is invariably associated with underlying behavioural or psychiatric

disturbance resulting in trichotillomania and trichophagia.^{1 2} Delayed presentation with large bezoar masses is not unusual as bezoar formation is an indolent process that takes many months or years before becoming symptomatic. The condition usually does not present as a surgical emergency, but has been known to become problematic due to gastric ulceration, perforation, acute bowel obstruction, intussusception, pancreatitis and obstructive jaundice.² Rapunzel syndrome is a more advanced variation of trichobezoar that involves post-pyloric extension of a hair tail.

There are numerous described techniques for management of trichobezoars. These include gastric lavage, dissolution therapy (eg, administration of pancreatic lipase, cola, cellulose, acetylcysteine), specialised benzotome and benzotriptor devices, endoscopic extraction and surgical retrieval.³ Surgical management becomes inevitable once a trichobezoar becomes more established.² Fragmentation of trichobezoars is often not possible due to the impenetrable nature of the dense enmeshed hair. There have been a small number of laparoscopic, laparoscopic-assisted and hybrid laparoendoscopic procedures reported.^{2 4-7} Minimally invasive approaches for large trichobezoars are limited by the need for a suitably sized specimen extraction incision, substantial operating times and greater potential for peritoneal spillage.^{2 5} For these reasons, laparotomy remains the standard surgical approach for this challenging pathology.² The use of the Alexis device for this indication has been previously described by Tudor and Clark⁵ and Son *et al*⁸ in laparoscopic-assisted approaches. We found that this device is ideal to complement and simplify 'minimally invasive' mini-laparotomy approaches that have been reported.⁹ Advantages of our described technique include (1) secure and excellent direct intragastric access, (2) shorter operating time and (3) reliable protection of both the wound edges and peritoneal cavity from bezoar contamination.

Learning points

- ▶ Trichobezoars in children are a challenge to manage as these insidiously accumulate and therefore, typically present late and are large in size.
- ▶ Surgical extraction becomes a necessary treatment option once trichobezoar become more established.
- ▶ Options for minimally invasive surgery are limited by the need for a suitably sized specimen extraction incision, substantial operating times and the greater potential for peritoneal spillage.
- ▶ A mini-laparotomy approach is feasible with the aid of an Alexis O Wound Protector/Retractor device.
- ▶ This simplified approach has advantages of (1) secure and excellent direct intragastric access, (2) shorter operating time and (3) reliable protection of both the wound edges and peritoneal cavity from bezoar contamination.

Figure 3 Trichobezoar specimen demonstrating a dense cast-mold impression of the stomach.

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Acknowledgements The authors would like to acknowledge Dr Paul Hammond (Department of Gastroenterology, Women's and Children's Hospital, South Australia).

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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