Available online at www.sciencedirect.com

ScienceDirect

journal homepage: http://Elsevier.com/locate/radcr

Case Report

A hairy situation: trichobezoar presenting with intussusception, and intestinal and biliary perforation in a child

*Akshay D. Baheti MD, Jeffrey P. Otjen MD, Grace S. Phillips MD**

Department of Radiology, Seattle Children's Hospital, University of Washington, 4800 Sand Point Way NE, Seattle, WA 98105, USA

ARTICLE INFO

Article history:
Received 5 July 2016
Accepted 7 July 2016
Available online 3 December 2016

Keywords:
Trichobezoar
Intussusception
Intestinal perforation
Biliary perforation

ABSTRACT

Trichobezoars are an uncommon cause of acute abdominal pain. We present a case of a 12-year-old girl with a history of a trichobezoar who presented to the emergency department with acute abdominal pain. Abdominal sonography was performed which suggested portal venous gas and showed complex peritoneal fluid. Subsequent computed tomography demonstrated both gastric and small bowel bezoars, with a jejunojejunal intussusception, and confirmed portal venous gas and complex ascites. At the time of surgery, there was evidence of intestinal and biliary perforation. Our case illustrates a constellation of complications in association with a long-standing trichobezoar.

Introduction

Trichobezoars are an uncommon entity and are typically seen in girls with a history of trichotillomania and trichophagia. Trichobezoars have been identified as a potentially overlooked cause of abdominal pain in children presenting to the emergency department [1]. We present a case of a 12-year-old girl with a known trichobezoar who presented emergently with complications of intussusception, and intestinal and biliary perforation. Our case highlights the potential for mortality associated with trichobezoars and illustrates a triad of unusual complications.

Case report

A 12-year-old girl presented to the emergency department with progressively worsening abdominal pain, emesis, and constipation, with a medical history significant for Turner mosaicism, trichophagia, and a large gastric bezoar diagnosed 5 months prior which was being treated conservatively. Physical examination findings were notable for tachycardia to 140, and a palpable large, midline epigastric mass. Her abdomen was distended with mild tenderness to palpation in the lower abdominal quadrants. No guarding or rigidity was noted. Abdominal sonography was performed which revealed

Competing Interests: The authors have declared that no competing interests exist.

* *Corresponding author.*
E-mail address: grace.phillips@seattle.childrens.org (G.S. Phillips).

http://dx.doi.org/10.1016/j.radcr.2016.07.003

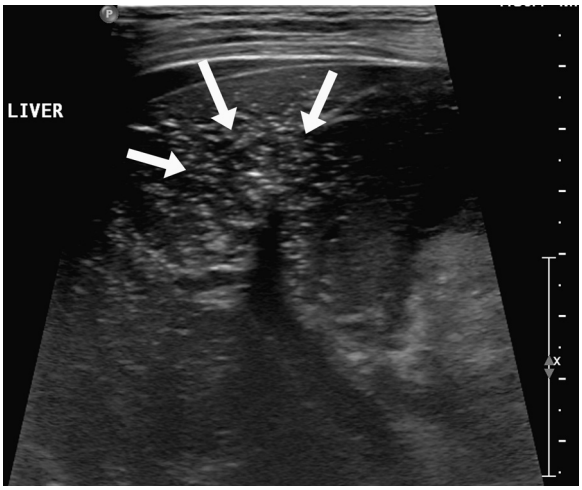

Fig. 1 – Transverse sonogram demonstrates a heterogeneous appearance of the liver with the presence of innumerable hyperechoic specks (arrows), suggestive of possible portal venous gas. Complex fluid collection was also noted (not shown).

Fig. 3 – Surgical specimen shows the resected gastroduodenal and jejunal bezoars.

portal venous gas (Fig. 1) and a large amount of complex peritoneal fluid. Contrast-enhanced computed tomography (CT) showed a large gastroduodenal bezoar and a proximal jejunal bezoar with a large jejunojejunal intussusception (Fig. 2).

Exploratory laparotomy confirmed a large gastroduodenal and jejunal bezoars (Fig. 3). Free abdominal perforation was also found at surgery with bilious peritoneal fluid in the abdomen and gelatinous proteinaceous debris. There was some ecchymosis and bile staining along the right side of the porta hepatis with no obvious bile leak but concerning for pressure necrosis or direct obstructive injury to the bile duct.

The patient's postoperative course was complicated by portal vein thrombosis requiring catheter-directed thrombolysis of the main right and left portal veins and angioplasty of the main portal and right portal veins. Operatively placed drains in the right upper and left lower quadrants drained a large amount of pus postoperatively. In addition, the patient experienced abdominal wound dehiscence requiring incision and drainage, washout, and delayed primary closure of the midline wound. Ultimately, at the time of discharge, there were no clinical signs of ongoing intra-abdominal infection, and she was tolerating adequate enteral feeds.

Discussion

Bezoar is an uncommon but potentially serious cause of abdominal pain in children. Several types of bezoars exist, including phytobezoars (plant material), lactobezoars (formula or milk, seen exclusively in infants) [2], pharmacobezoars (medications), and trichobezoars (hair). Bezoars are most commonly seen in the stomach, although may also involve the small bowel, particularly in the case of

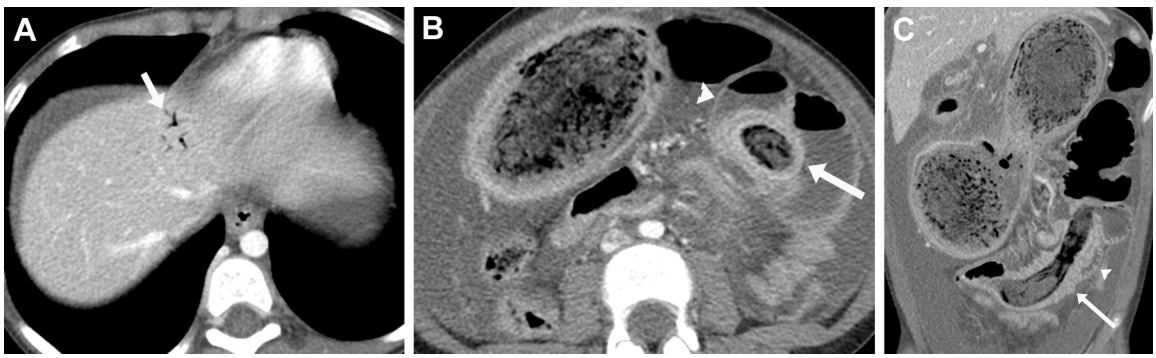

Fig. 2 – (A) Axial contrast-enhanced CT image confirms the presence of portal venous gas (arrow). (B, C) Axial and coronal contrast-enhanced CT images demonstrate the large trichobezoar causing small bowel–small bowel intussusception (arrows pointing at the intussusceptum and arrowheads pointing at the intussuscipiens). Note the bowel wall thinning involving the intussuscipiens, the best appreciable on the axial image.

trichobezoars. "Rapunzel syndrome" is the name given to trichobezoars that extend into the small bowel, potentially traversing the ileocecal valve, with associated with obstructive symptoms [3]. While some phytobezoars and lactobezoars may be managed conservatively with enzyme-based therapy or observation, respectively, trichobezoars typically require surgical intervention in the form of laparotomy [4].

Trichobezoars overwhelmingly tend to occur in females over males, with predisposing factors of trichotillomania and trichophagia. A recent case series of 7 patients with trichobezoars, all of whom were girls, found a mean age at presentation of 11.5 years [5]. Common presenting signs and symptoms of trichobezoars include chronic gastrointestinal complaints, a palpable abdominal mass, and small bowel obstruction [5]. Imaging plays an important role in diagnosis. On abdominal radiography, the diagnosis may be suspected when there is abundant mottled material admixed with gas distending the stomach. On CT, bezoars present as a heterogeneous, intraluminal mass typically interspersed with gas, distending but not adherent to the stomach or small bowel [6]. CT is helpful in delineating small bowel extension including more distal satellite lesions [6].

Trichobezoars may undergo complications, particularly when they extend into the small bowel. Our patient presented emergently both with intussusception as well as intestinal and biliary perforation. Prior case reports have described trichobezoars complicated by intussusception and peritonitis in separate patients [3]. Trichobezoars are a described potential cause of atypical intussusception in children [7]. In our patient, we speculate that she was at an increased risk for complications due to the chronicity of her symptoms. It was theorized that there was pressure necrosis of the bile ducts due to mass effect from the duodenal portion of the bezoar. Such a phenomenon has been reported with other mass lesions, such as with an ampullary tumor causing biliary duct perforation in an adult [8]. Biliary perforation in children is rare but can be seen in the setting of trauma, choledochal cysts, and less commonly idiopathically [9].

In summary, our case highlights trichobezoar as an uncommon, yet important potential cause of abdominal pain in girls presenting with acute abdominal pain. The diagnosis of trichobezoar may be suspected in female patients with a history of trichotillomania, trichophagia, gastrointestinal symptoms, and a palpable abdominal mass. However, imaging is a mainstay for accurate diagnosis, particularly of potential complications.

Acknowledgment

The authors would like to acknowledge Dr. Erin Rudzinski, Seattle Children's Hospital, Department of Pathology, for the surgical pathology specimen photograph displayed in Figure 3.

REFERENCES

[1] Lynch KA, Feola PG, Guenther E. Gastric trichobezoar: an important cause of abdominal pain presenting to the pediatric emergency department. Pediatr Emerg Care 2003;19(5):343–7.
[2] Otjen JP, Iyer RS, Phillips GS, Parisi MT. Usual and unusual causes of pediatric gastric outlet obstruction. Pediatr Radiol 2012;42(6):728–37.
[3] Naik S, Gupta V, Naik S, Rangole A, Chaudhary AK, Jain P, et al. Rapunzel syndrome reviewed and redefined. Dig Surg 2007;24(3):157–61.
[4] Gorter RR, Kneepkens CM, Mattens EC, Aronson DC, Heij HA. Management of trichobezoar: case report and literature review. Pediatr Surg Int 2010;26(5):457–63.
[5] Fallon SC, Slater BJ, Larimer EL, Brandt ML, Lopez ME. The surgical management of Rapunzel syndrome: a case series and literature review. J Pediatr Surg 2013;48(4):830–4.
[6] 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(4):228–32.
[7] Dalshaug GB, Wainer S, Hollaar GL. The Rapunzel syndrome (trichobezoar) causing atypical intussusception in a child: a case report. J Pediatr Surg 1999;34(3):479–80.
[8] Kaplan M. A case report of an ampullary tumor presenting with spontaneous perforation of an aberrant bile duct and treated with total laparoscopic pancreaticoduodenectomy. World J Surg Oncol 2012;10:142.
[9] Kanojia RP, Sinha SK, Rawat J, Wakhlu A, Kureel S, Tandon R. Spontaneous biliary perforation in infancy and childhood: clues to diagnosis. Indian J Pediatr 2007;74(5):509–10.