CASE REPORTS

Small Bowel Volvulus with a Jejunal Trichobezoar

Ky Young Cho, Dong Ho Shim and *Kyung Tak Yoo

From Departments of Pediatrics and *Surgery, KEPCO Medical Center, Seoul, Republic of Korea.

Correspondence to: Dr Ky Young Cho, Department of Pediatrics, KEPCO Medical Center, 308, Uicheon-ro, Dobong-gu, Seoul, Republic of Korea. choky96@hanmail.net Received: January 29, 2014; Initial review: March 15, 2014; Accepted: April 01, 2014. **Background:** Small bowel volvulus caused by a jejunal trichobezoar is an extremely rare and life-threatening emergency in children. **Case characteristics:** An 8-year-old girl with abdominal pain and persistent bilious vomiting. **Observation:** The abdominal computed tomography scan showed a solitary intraluminal mass and a whirl sign, suggesting the small bowel volvulus. Emergency laparoscopic exploration revealed the rotated segment of small bowel loops by a jejunal trichobezoar. **Outcome:** Satisfactory recovery after surgery. **Message:** Trichobezoars should be considered in the differential diagnosis of abdominal pain and projectile vomiting in children.

Keywords: Intestinal obstruction; Small intestine; Trichobezoar; Volvulus.

richobezoars are concretions of hair in the gastrointestinal tract, and are often associated with underlying psychiatric disorders such as trichotillomania and trichophagia. Trichobezoars are usually present in females during the second decade of life, and are rare in children [1]. Most trichobezoars develop in the stomach; in the small bowel, these are usually located at the ileum, the narrowest part of small bowel [1]. A jejunal trichobezoar without associated gastric trichobezoars is rare, and primary segmental small bowel volvulus caused by a jejunal trichobezoar is also extremely rare in children. We describe acute primary segmental small bowel volvulus caused by a jejunal trichobezoar in a child.

CASE REPORT

An 8-year-old girl presented to our emergency department with a history of intermittent, non-localized abdominal pain and non-bilious vomiting for the preceding 12 hours. She had no history of prior abdominal surgery, chronic abdominal pain, chronic vomiting or ingestion of toxic materials. On physical examination, the patient was in the 50th percentile for weight and height, and was ill-looking and dehydrated. Her hair were long without areas of alopecia. The abdomen was soft, non-distended and diffusely tender, especially on the epigastrium with decreased bowel sounds. Other physical examination, laboratory findings and upright plain films of the abdomen were unremarkable. Abdominal ultrasound showed no evidence of intussusception. She was kept nil-by-mouth and received intravenous fluids but intermittent abdominal pain and non-bilious vomiting continued.

On the second day of admission, the patient developed cramping abdominal pain, bilious vomiting and absolute constipation. Her abdomen was mildly distended with diffuse tenderness and increased bowel sounds. Upright plain films of the abdomen showed some distended small bowel loops and air-fluid levels without any free gas. The abdominal computed tomography (CT) scan showed a whirl sign, a swirl of mesenteric soft-tissue and fat attenuation with adjacent loops of bowel surrounding rotated intestinal vessels (Fig. 1). Distended, U-or C-shaped small bowel loops with transition zones and an intraluminal mottled mass in the small bowel could be seen (Fig. 1). The patient underwent emergency laparoscopic exploration that revealed a segment of small bowel, 20 cm in length, rotated 180 degrees counter-clockwise; there was no gangrene or ischemia. No other intraperitoneal abnormalities or anomalies predisposing the patient to small bowel volvulus were present. After de-torsion, a solitary intraluminal hard mass causing complete obstruction was identified at the level of the proximal jejunum, approximately 260 cm from the ileocecal junction. The dilated, segmental small bowel loop with the intraluminal hard mass was extracted through the single-port, supraumbilical incision site. A longitudinal enterotomy was performed and the 10x4x4-cm hair (trichobezoar) concretion was evacuated. enterotomy was then transversally closed. No residual trichobezoars in the stomach or in the rest of small intestine were found upon manual exploration. The patient had a satisfactory postoperative recovery, and provided a history of swallowing of hairs. The patient was discharged five days after surgery, and was referred to the psychiatrist for further treatment.



FIG. 1 Abdominal computed tomography scan showing whirl sign (white arrow head) suggesting small bowel volvulus and distended small bowel loops with intraluminal mottled mass (white arrow).

DISCUSSION

Primary small bowel volvulus is a rare, but lifethreatening surgical emergency. It can affect the entire small bowel and its mesentery or only a segment, and may occur without any underlying causes [2]. Most cases of primary small bowel volvulus in adults are related to sudden ingestion of a large volume of indigestible food after long periods of fasting causing the loop to descend to the pelvis because of the increased weight and displacing empty small bowel loops upwards, initiating the rotation of the mesentery leading to volvulus [3]. This mechanism can also be applied to the relatively heavy trichobezoar and an empty bowel. An abdominal CT scan is the most useful diagnostic tool for small bowel volvulus [4]. Timely diagnosis, surgical detorsion and mesenteric decompression are important to avoid mesenteric ischemia and gangrene [4].

The majority of trichobezoars are confined to the stomach, though they rarely extend into jejunum, ileum and colon as a tail, which is called Rapunzel syndrome [5]. Accumulation of slippery hair strands between the mucosal folds of the stomach prevents their propulsion by peristalsis [5]. Small bowel trichobezoars without any associated gastric trichobezoars have been described, but most trichobezoars are impacted in the narrowest locations of the small bowel, such as the ileum, jejunal diverticulum or at a postoperative stenosis [6,7]. The

presentation of small bowel volvulus by a jejunal trichobezoar is extremely rare in children. The diagnosis of pediatric trichobezoars is difficult until the patients develop symptoms of small bowel obstruction caused by a hair ball of sufficient size [8]. Eliciting a history of trichophagia or trichotillomania is also difficult in a child. Several treatments have been proposed, including removal by conventional laparotomy, laparoscopy and endoscopy [9]. After evacuation of trichobezoars, exploration of the stomach and the rest of small intestine is necessary to prevent secondary intestinal obstruction by satellite trichobezoars. If available, postoperative esophago-gastroduodenal endoscopy and small bowel series are preferred. Parental counseling, long-term psychotherapy and behavioral modification are essential to prevent recurrences.

In conclusion, primary segmental small bowel volvulus can be rarely caused by a jejunal trichobezoar. A high level of suspicion is required to make a diagnosis, and to provide timely and adequate treatment.

Contributors: All authors were involved in management of patient, manuscript writing and its final approval. Funding: None; Competing interests: None stated.

REFERENCES

- 1. Gorter RR, Kneepkens CM, Mattens EC, Aronson DC, Heij HA. Management of trichobezoar: case report and literature review. Pediatr Surg Int. 2010;26:457-63.
- Birnbaum DJ, Grègoire E, Campan P, Hardwigsen J, Le Treut YP. Primary small bowel volvulus in adult. J Emerg Med. 2013;44:e329-30.
- Papadimitriou G, Marinis A, Papakonstantinou A. Primary midgut volvulus in adults: report of two cases and review of the literature. J Gastrointest Surg. 2011;15:1889-92.
- Duda JB, Bhatt S, Dogra VS. Utility of CT whirl sign in guiding management of small-bowel obstruction. Am J Roentgenol. 2008;191:743-7.
- Chogle A, Bonilla S, Browne M, Madonna MB, Parsons W, Donaldson J, et al. Rapunzel syndrome: a rare cause of biliary obstruction. J Pediatr Gastroenterol Nutr. 2010;51:522-3.
- Sharma RD, Kotwal S, Chintamani, Bhatnagar D. Trichobezoar obstructing the terminal ileum. Trop Doct. 2002;32:99-100.
- Fagenholz PJ, de Moya MA. Laparoscopic treatment of bowel obstruction due to a bezoar in a Meckel's diverticulum. JSLS. 2011;15:562-4.
- 8. Khattala K, Boujraf S, Rami M, Elmadi A, Afifi A, Sbai H, et al. Trichobezoar with small bowel obstruction in children: two cases report. Afr J Paediatr Surg. 2008; 5:48-51.
- 9. 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;48:830-4.