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Journal of Pediatric Surgery CASE REPORTS

journal homepage: www.jpascasereports.com

Segmental volvulus of the ileum without malrotation in an infant: A case report



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ARTICLE INFO

Article history:

Received 4 June 2015
Received in revised form
7 August 2015
Accepted 8 August 2015

Key words:

Volvulus
Ileum
Infant

ABSTRACT

Intestinal volvulus usually occur secondary to malrotation, and primary segmental volvulus has rarely been reported. A 12-month-old female infant presented with a 3-day history of excessive vomiting. An ultrasonography revealed a “whirlpool sign” in the right upper abdomen, suggesting small bowel volvulus with obstruction. Laparotomy revealed a twisted, viable loop of small bowel in the right upper abdomen, and abnormal adhesions were noted between the distal and mid ileum, with resulting mesenteric narrowing. Attempted mesenteric widening by dissection of the peritoneum overlying the adhesions failed, because of abnormal, taut mesenteric vessels. Subsequent resection of the involved segment cured the patient. Recurrent obstructive symptoms in an infant can be an atypical presentation of segmental volvulus, and segmental volvulus should be included in the differential diagnosis of such cases.

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Most cases of intestinal volvulus are associated with malrotation; segmental volvulus without malrotation is rare. The usual presentation of segmental volvulus in a neonate is acute onset of rapidly progressive bowel ischemia with obstruction. We report herein a case of segmental volvulus of the ileum in a 1-year-old infant with long history of vomiting, which turned out to be the result of segmental narrowing of the mesentery.

1. Case report

A 12-month-old female infant presented with vomiting that had been aggravated for 3 days before presentation. Vomiting occurred about 30 min to 1 h after a meal, and the vomitus contained bile. The patient did not have a history of prior abdominal surgery, but had had recurrent episodes of vomiting without a definitive diagnosis. Her body weight was 6 kg, which was below the 3rd percentile. On physical examination, the patient was lethargic with decreased skin turgor. Her abdomen was distended, but there was no tenderness or rebound tenderness. An infantogram showed marked small bowel dilatation (Fig. 1A); and abdominal ultrasonography demonstrated a “whirlpool sign” in the right upper abdomen just below the liver, suggesting small

bowel volvulus with obstruction (Fig. 1B). The axis of the superior mesenteric artery and vein was in normal orientation; and the cecum and appendix were located in the right lower quadrant in their normal anatomic position, which thereby excluded malrotation. On laparotomy, the duodenal C loop was normally configured, and a segment of small bowel was twisted and adherent to the right upper abdominal wall and gallbladder without evidence of strangulation (Fig. 2A). Adhesions between the distal ileum (5 cm proximal to the ileocecal valve) and mid ileum (35 cm proximal to the ileocecal valve) were noted, resulting in mesenteric narrowing (Fig. 2B). Mesenteric widening was attempted by releasing the peritoneum overlying the adhesions, but failed because of taut mesenteric vessels (Fig. 2C). The involved segment was resected, and end-to-end anastomosis was performed. The infant's postoperative course was uneventful, and she was discharged 7 days after surgery.

2. Discussion

Most cases of intestinal volvulus in children are associated with malrotation, and the traditional teaching has been that segmental volvulus without malrotation is extremely rare. However, Kitano et al. [1] and Lister et al. [2] reported a respective prevalence of segmental volvulus without malrotation of 26.3% and 22.9%, suggesting that the prevalence and clinical implications of this entity have long been underestimated.

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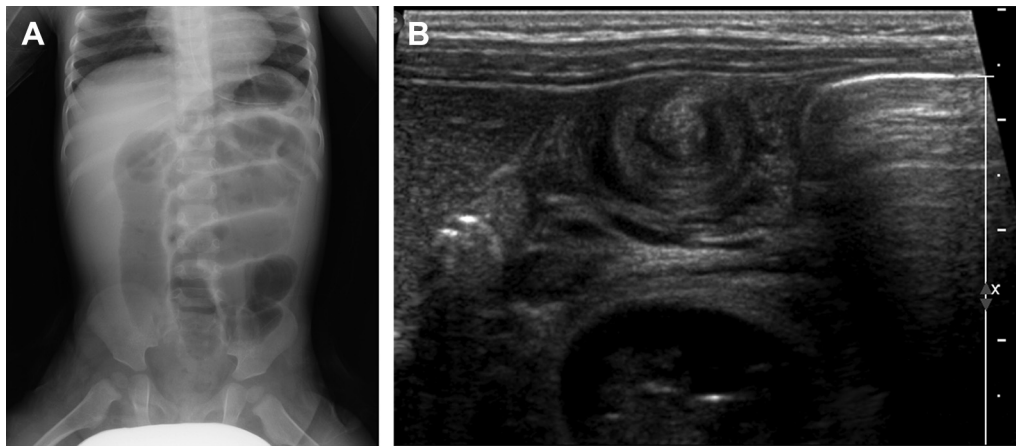


Fig. 1. (A) Markedly dilated bowel loops are seen on an infantogram. (B) Whirlpool sign shown in right upper abdomen on an ultrasonographic scan.

A chylous jejunal cyst, mesenteric mass such as lymphatic malformation, pseudocyst, and lipoblastoma have been identified as pathologies associated with segmental volvulus in the absence of malrotation [3–6]; but in most cases, the etiology of primary volvulus has been undetermined. In our case, a section of the ileum and associated mesentery were anomalous, in that the distal 30 cm of ileum was supplied by a single, dominant pedicle, which resulted in narrowed mesentery associated with the involved bowel segment. In the case series reported by Kitano et al., a long, narrow, band-like mesentery was seen in 3 cases, but the authors could not clarify whether this was the result or the cause of the volvulus [1]. We believe that the anomalous anatomy of the mesentery and related vessels could have been congenital and the cause of volvulus in our case, because the branches from the dominant pedicle precluded mesenteric widening; and such an anomaly could not be generated secondarily from an acquired volvulus.

The typical clinical picture of primary segmental volvulus is acute intestinal strangulation manifesting early in life, and such conditions have even been reported in fetuses [7,8]. Compared to volvulus with malrotation, the ischemic changes in the twisted bowel without malrotation are thought to progress rapidly because the colon does not act as a cushion [9]. We speculate that our case manifested a chronic form of segmental volvulus; her long history of excessive vomiting led to growth retardation, the twisted bowel with adhesions to the surrounding organs were probably a result of

chronic inflammation, and the involved bowel remained viable. Because the general clinical spectrum of segmental volvulus does not include the presentation of our patient, the correct diagnosis, which was readily achieved by ultrasonography, was delayed. Segmental volvulus should be included in the differential diagnosis of infants with recurrent vomiting, abdominal distension, and no discernible causative factors.

Volvulus is not synonymous with bowel necrosis or strangulation, and the treatment of volvulus should consist of detorsion and removal of causative factors, if any. However, in most cases of primary segmental volvulus, bowel resection has been required because of the presence of strangulation [1,10]. For our case, mesenteric widening was unsuccessfully attempted to avoid unnecessary resection; however, because of taut mesenteric vessels, segmental resection was inevitable. Careful evaluation of the anatomy of this condition, in addition to determination of its causative pathology, should aid in identifying those patients who do and do not require bowel resection. In summary, we successfully treated a segmental volvulus of the ileum without malrotation in a 1-year-old infant with chronic vomiting. A single ultrasonographic examination was sufficient for arriving at the correct diagnosis, and laparotomy found that an anomalously narrowed mesentery was the cause of the volvulus. Mesenteric widening had failed because of taut mesenteric vessels, and segmental resection cured the patient. Although unusual in infants, segmental volvulus should be

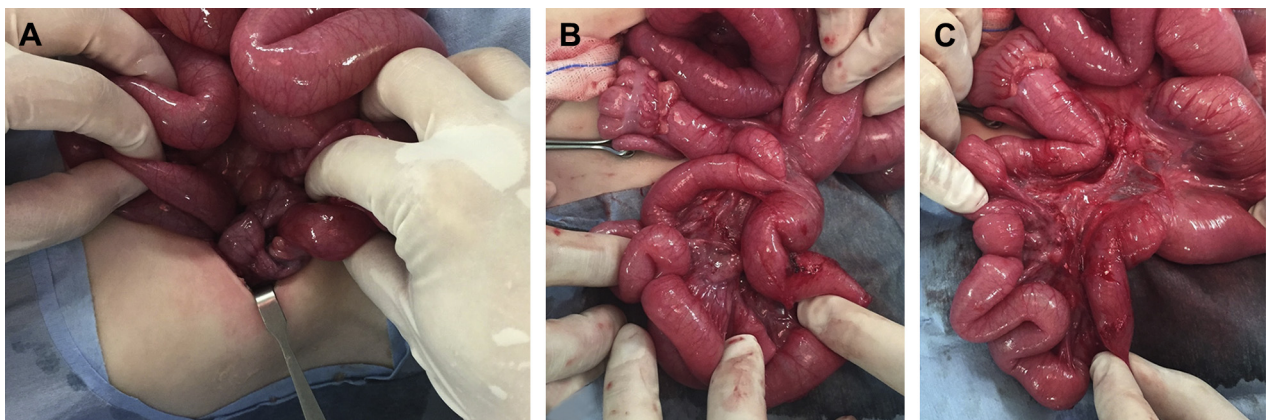


Fig. 2. (A) Segmental volvulus without strangulation. (B) Note the abnormal adhesions between ileal loops. (C) Tension in the side branches of mesenteric vessels precluded complete widening of the mesentery.

considered in those patients with recurrent signs of bowel obstruction. Treatment should be tailored according to the anatomy and bowel status of the patient.

Conflict of interest

There were no conflicts of interest.

Source of funding

There were no sources of funding.

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