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A right colonic volvulus requiring extensive colectomy in an infant with trisomy 13

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ABSTRACT

Colonic volvulus is a rare surgical emergency condition in children. Only approximately 40 children with cecal volvulus have been reported in English literature in the past 50 years. Among these, a right colonic volvulus involving the long segment from the ileal end to the transverse colon, as in our case, is limited to a few reports. Neurodevelopmental delay and a history of chronic constipation have been reported as common associated disorders. This is the first report about a case of right colonic volvulus in an infant with trisomy 13 who required extensive colectomy during an emergency laparotomy.

Colonic volvulus is an unusual cause of intestinal obstruction, and it is responsible for approximately 3–5% of all colonic obstructions in adults. In the pediatric population, colonic volvulus has a lower incidence and is extremely rare [1–6]. However, common disorders associated with colonic volvulus include neurodevelopmental delay, a history of chronic constipation, and aerophagia [1,2,4–8]. The purpose of this report was to describe the unusual occurrence of right colonic volvulus in a patient with trisomy 13, and consider the precipitating factors.

1. Case report

A 1-year-old female infant with trisomy 13 had sudden onset of abdominal distension and continuous vomiting. On admission to our hospital for close evaluation, it was difficult to assess whether she presented with an acute abdomen because of lack of adequate communication. She presented with a high fever of 39 °C, neutrophilia, and signs of mild dehydration, with no elevation of the C-reactive protein level or white blood cell count. The initial abdominal radiograph showed a dilated left bowel (Fig. 1). After conservative treatment for a diagnosis of chronic constipation, aerophagia, and bacterial enteritis, her condition worsened. An enhanced computed tomography (CT) scan showed a dilated bowel from the transverse to the sigmoid colon, indicating a left colonic obstruction. An internal hernia and intussusception were ruled out (Fig. 2a and b). Emergency laparotomy was required, with suspicion of transverse colonic volvulus; a contrast enema demonstrated poor flow of gastrografin into the transverse colon (Fig. 3), and blood testing suggested intestinal necrotic changes with elevated myogenic enzymes (Table 1).

During surgery, a transverse abdominal incision was made. The cecum and ascending colon were surprisingly located on the left side in the abdominal cavity, with fecal impaction throughout (Fig. 4a). Necrotic changes from the ileal end to the transverse colon, due to right colonic volvulus with a nonrotation form of malrotation, were found (Fig. 4b). The necrotic, dilated bowel was resected, and primary anastomosis was performed after removing the volvulus (Fig. 5). A midgut volvulus was ruled out because the small intestine was intact. Histopathological examination showed total necrosis of the resected bowel, with no evidence of chronic inflammation. The patient is presently doing well and is asymptomatic 4 months postoperatively. No complication was noted at this follow-up.

2. Discussion

With malformation of the mesentery said to be the primary etiology of colonic volvulus [9], the detailed factors are as follows.

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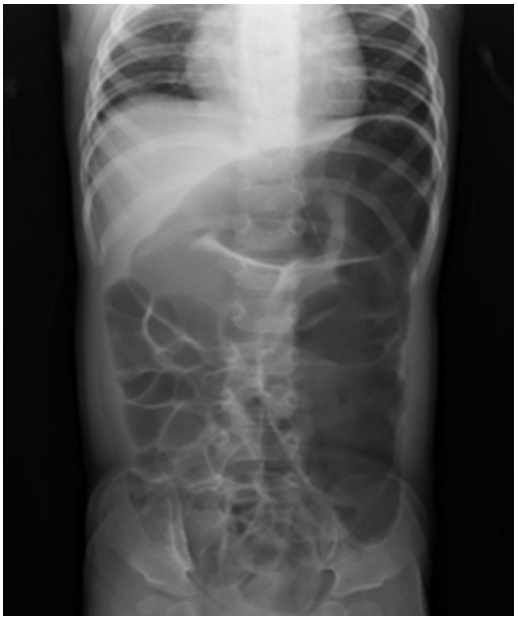

Fig. 1. An abdominal radiograph showing an extensively dilated left bowel; gas in the rectum is indistinct.

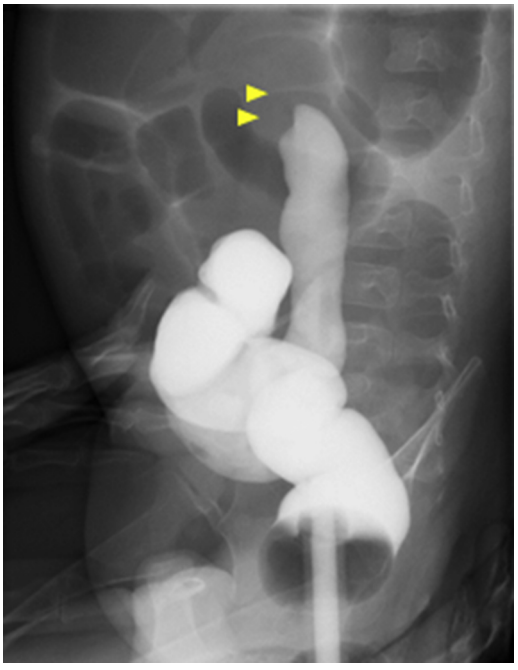

Fig. 3. Contrast enema showing poor flow of gastrografin into the transverse colon (arrowheads).

The possible congenital factors are a redundant and mobile colon, narrow base of the mesenteric root, elongation of the mesentery with a freely mobile bowel, and absent ligamental attachments at the splenic or hepatic flexures, according to some reports [2,3,5,9]. In contrast, precipitating acquired factors are reported to be neurodevelopmental delay, severe chronic constipation, and aerophagia [1,2,4–8]. The coexistence of these factors causes chronic bowel distension, and inadequate treatment increases the incidence of colonic volvulus. Folaranmi et al. [1] hypothesized that these acquired factors may promote stretching of the ligaments responsible for fixation. By the time the mesentery of the colon becomes longer, a volvulus can occur.

In our case, several acquired risk factors for colonic volvulus, including neurodevelopmental delay, were also present. In addition to providing conservative treatment for a diagnosis of chronic constipation, aerophagia, and bacterial enteritis, midgut volvulus was suspected as the cause of an acute abdomen because malrotation is presumed to be a major complication in children with trisomy 13. However, this was excluded by imaging. At one point, the influence of a nonrotation form of malrotation on causing a colonic volvulus was presumed. Nonrotation may be a risk factor of colonic volvulus because the cecum and ascending colon were shifted to the left side in the abdominal cavity in our case. In addition, the lack of broad colonic fixation to the retroperitoneum and the absence of ligamental attachments at the hepatic and splenic flexures may be factors that influence the range of the strangulated intestinal segment. Berger et al. [10] also reported a case of volvulus of the ascending colon as an unusual complication of nonrotation of the midgut. Thus, some

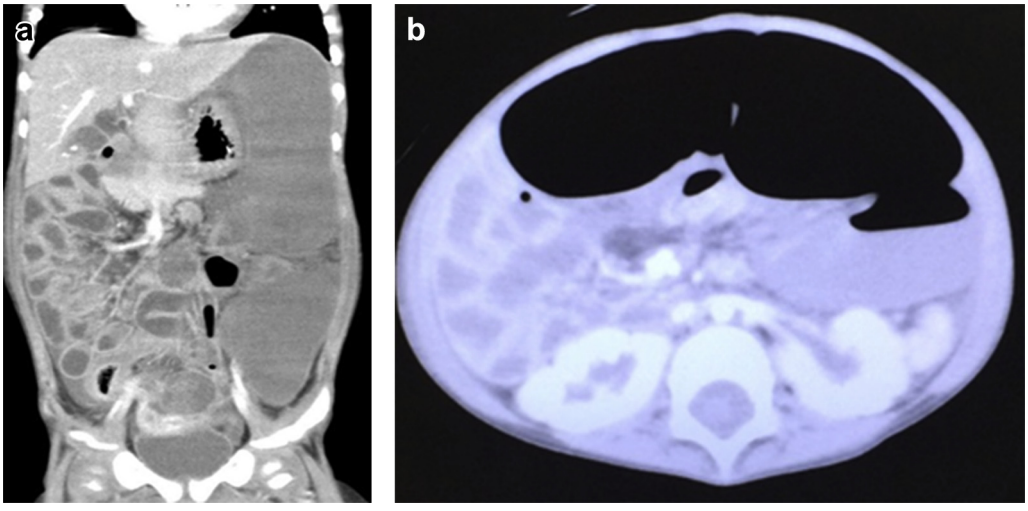

Fig. 2. a and b) An enhanced computed tomography image showing a dilated transverse and descending colon, and dilated intestinal tract wall. The small intestine is not dilated.