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Stercoral perforation of the ileum in a very low birth weight infant: A case report

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ABSTRACT

Stercoral perforation is rare spontaneous rupture of the bowel that is associated with a high risk of mortality. Cases of stercoral perforation have typically been reported in adults, with the colon being the most common site of perforation. There has been no case report of stercoral perforation in infants. In this case report, we describe a stercoral perforation in the distal ileum in a preterm male infant. An 11-day-old male was referred with complaints of vomiting and abdominal distension. Dilatation of bowels with fecal impaction was found on simple abdominal radiographs. Abdominal distension was aggravated with no effect of glycerin enema. Explorative laparotomy was performed and stercoral perforation of the distal ileum was confirmed.

Stercoral perforation is rare spontaneous rupture of the bowel due to fecal impaction and that is associated with a high risk of mortality [1]. Most cases of stercoral perforation have been reported in elderly patients with preceding constipation and the colon was the most common site of perforation [2]. In this case report, we describe the process of diagnosis and treatment of a stercoral perforation of the distal ileum in a very low birth weight male infant born in preterm.

1. Case report

An 11-day-old male in the neonatal intensive care unit was referred to our services because of vomiting and abdominal distension. The patient was born at 28 weeks of gestation and weighed 1270 g. The infant's mother was 34 years old, and the medical history of the pregnancy was unremarkable. The infant's 1-min and 5-min APGAR score was 7 and 8, respectively. Meconium was passed in the first day after birth, with two more bowel movements recorded over the subsequent 4 days. He was being fed with powdered infant formula.

After post-natal day 5, no bowel movement was recorded. A glycerin enema was performed, but it was ineffective. With necrotizing enterocolitis (NEC) being suspected, the infant was treated with antibiotics and placed on nulli per os (NPO) status. On post-natal day 8, abdominal distension developed, accompanied by vomiting. Simple abdominal radiographs confirmed dilation of the bowels, with fecal impaction observed on abdominal ultrasonography. Laboratory findings revealed a leukocytosis and metabolic acidosis, as follows: white blood cell count, 21.81 $\times$ 10^3/mm^3; pH level,7.27; pCO_2, 50 mmHg; pO_2, 41 mmHg; bicarbonate levels (HCO_3), 15.9 mmol/L; and base excess (BE), - 4.8 mmol/L. On post-natal day 11, a radiopaque mass-like lesion was identified in the right lower quadrant (RLQ), in combination with further dilation of the bowels. There was no evidence of a pneumoperitoneum (Fig. 1). An N-acetylcystein enema was performed; this was ineffective as well. On post-natal day 13, the abdomen was further distended and tense, and a mass was palpable in the RLQ. Aggravated low gastrointestinal obstruction was observed on simple abdominal radiographs. On Gastrografin®(Diatrizoate Meglumine and Diatrizoate Sodium) enema, no colonic obstruction was identified except for a small amount of stool in the colon, which was passed following the contrast study (Fig. 2). Progressive metabolic acidosis was identified as follows: pH, 7.17; pCO_2, 21 mmHg; pO_2, 113 mmHg; HCO_3, 73.1 mmol/L; BE, −20.2 mmol/L.

Explorative laparotomy was performed and a dumbbell shaped dilation of the terminal ileum was noted, with a 2-cm bowel perforation on the anti-mesenteric border, 15 cm proximal to the ileocecal valve. There was no fecal spillage into the abdominal cavity, with the perforation site plugged with a fecaloma that was hard and almost dried up. We resected about 7 cm of the bowel

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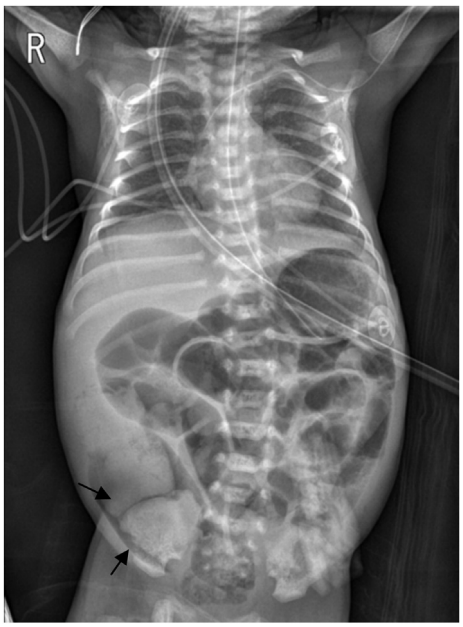

Fig. 1. Abdominal radiograph on post-natal day 11, showing bowel dilation and a radioopaque mass-like lesion in the RLQ(arrow).

segment, including the perforation site, the dilated proximal bowel and the collapsed distal bowel (Fig. 3, Fig. 4), and performed an end-to-end anastomosis. There was no evidence of small bowel atresia or malrotation. Ganglion cells were identified in the whole resected specimen, on pathological examination, with positive immunohistochemistry findings for CD56(NCAM/SCLC), CD117(C-KIT) and S-100. Pressure necrosis and inflammation were noted around the perforation site.

The postoperative clinical course was uneventful. On post-operative day 4, the infant passed a stool that contained bile and feeding was initiated. The amount of feeding was gradually increased, and the infant was in good condition. Seven months has been passed from the operation and the patient is developing well without any gastrointestinal problems.

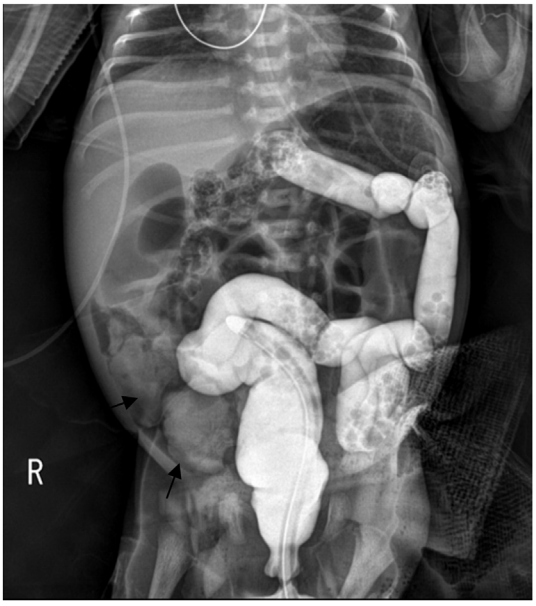

Fig. 2. Gastrografin enema performed on post-natal day 13, showing fecal impaction in the RLQ (arrow), with no signs of obstruction in the colon.

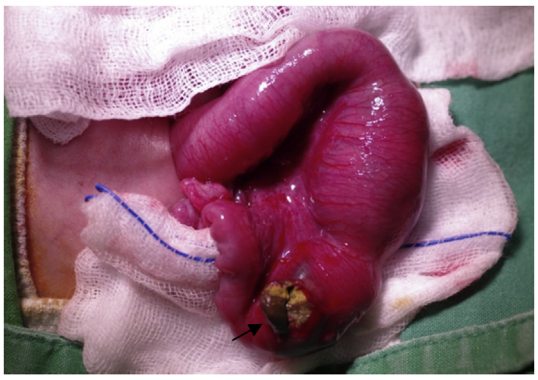

Fig. 3. Surgical findings, showing the perforation site (arrow) plugged with fecaloma, with a dumbbell-shaped dilation of the proximal bowel filled with hard feces.

2. Discussion

Stercoral perforation is defined as a spontaneous perforation of the bowel due to feces. The first stercoral perforation was reported in 1984 [1]. Most reported cases of stercoral perforation have been reported in elderly patients, with chronic constipation typically preceding the perforation [2]. The possible causes of chronic constipation were various: pregnancy, anticholingergic agents, nonsteroidal antinflammatory drugs, opioids, bed ridden state, elderly patients, and all conditions which predispose to constipation [1]. Stercoral perforation during pregnancy has also been reported [3,4].The most common site of perforation is the sigmoid colon [2]. A case of stercoral perforation of the sigmoid colon was previously reported in a 2-year-old female after use of high dose of a non-steroidal anti-inflammatory drug (NSAID) over a short period [5]. However, our review of the literature did not identify a prior case of stercoral ileal perforation in a neonate. Here, we have described a case in a very low birth weight 13-day-old infant, with the perforation occurring in the distal ileum.

In adults, stercoral perforation is diagnosed based on clinical signs and symptoms, supplemented by computed tomography imaging [6]. Maurer et al. [7] suggested three diagnostic criteria for stercoral perforation of the colon, which were present in our case: the perforation site was localized on the antimesenteric border and

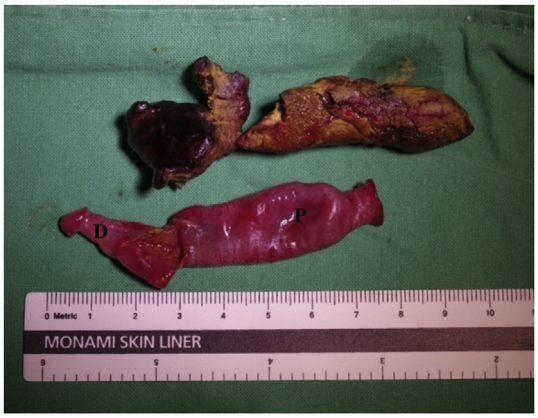

Fig. 4. The resected ileum and fecaloma. The specimen included both the dilated proximal bowel and the collapsed distal bowel. P: dilated proximal bowel, D: collapsed distal bowel.

was round, with its 2 cm diameter exceeding the 1 cm criterion for diagnosis; the intestinal lumen was plugged by a fecaloma which projected through the perforation site; and pressure necrosis and inflammation at the site of perforation were confirmed on pathological findings.

The clinical features and pathological findings of stercoral perforation in this neonate were different from NEC and spontaneous intestinal perforation (SIP) [8,9]. NEC, which is a common cause of intestinal perforation in preterm infants, usually occurs at 2–6 weeks postnatally, while SIP is characterized by the absence of villus necrosis on histology. Therefore, for our patient, the age at onset and pathological findings were different from NEC and SIP. Furthermore, serial abdominal radiographs did not show evidence of a pneumoperitoneum, which is the most common diagnostic finding of SIP. The passing of meconium within 24 h of birth eliminated a differential diagnosis of meconium ileus or Hirschsprung's disease. Hirschsrpung's disease was clinically ruled out in our case report; the infant passed meconium within 24 h after birth and several bowel movements were recorded following post-natal 4 days. Normal intestinal gas patterns were found on simple abdominal radiographs and bowel movements were recognized during the postoperative follow up periods.

Performing a Hartmann's closure is the treatment of choice recommended for stercoral perforation of colon [4]. In our case, as there was no spillage into the abdominal cavity, we opted to perform segmental resection of the small bowel, with end-to-end anastomosis.

3. Conclusion

We describe the clinical presentation and treatment of a stercoral perforation of the ileum in a very low birth weight neonate. Based on our experience, stercoral perforation could be considered as differential diagnosis of NEC, spontaneous intestinal perforation and meconium ileus in the neonatal period. Stercoral perforation should be suspected in the presence of a long standing fixed fecaloma on simple abdominal radiographs. More studies regarding the development of fecal impaction after normal excretion of meconium in very low birth weight neonates are needed.

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