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Pediatric Radiology

Enteric fistula formation secondary to necrotizing enterocolitis

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Abstract. We report two cases of enterocolonic fistula formation following necrotizing enterocolitis, and a review of the six previously published cases. Ischemic mechanism is the most likely cause. The fistulas were diagnosed by upper gastrointestinal series and contrast enema.

Although stricture formation is a known sequela of necrotizing enterocolitis (NEC) in infants, the development of a fistula is rare. We present two cases of enterocolonic fistula developing as a complication of NEC and review six cases previously described in the literature.

Case reports

Case 1

A female infant, the 820 g product of a 30 week gestation, was admitted to the neonatal intensive care unit for respiratory distress. The infant was treated for hyaline membrane disease and at the age of 6 days developed NEC. This resolved following medical therapy. At age 30 weeks, the infant developed abdominal distention and diminished bowel sounds. Plain films showed mildly distended bowel loops (Fig. 1 a). A water soluble enema demonstrated a fistula between the splenic flexure and jejunum (Fig. 1 b), which was confirmed on upper G. I. series (Fig. 1 c and 1 d). The patient did poorly and died one week later. At autopsy, a jejuno-colic fistula was present.

Case 2

A male infant was the product of a 30 week gestation and weighed 1190 g at birth. The patient developed abdominal distention at age 22 days with plain film findings consistent with NEC (Fig. 2 a). He was treated medically and did well until 43 days of age when abdominal distention developed. Plain films showed multiple dilated bowel loops consistent with obstruction (Fig. 2 b). A water soluble enema demonstrated a fistula between

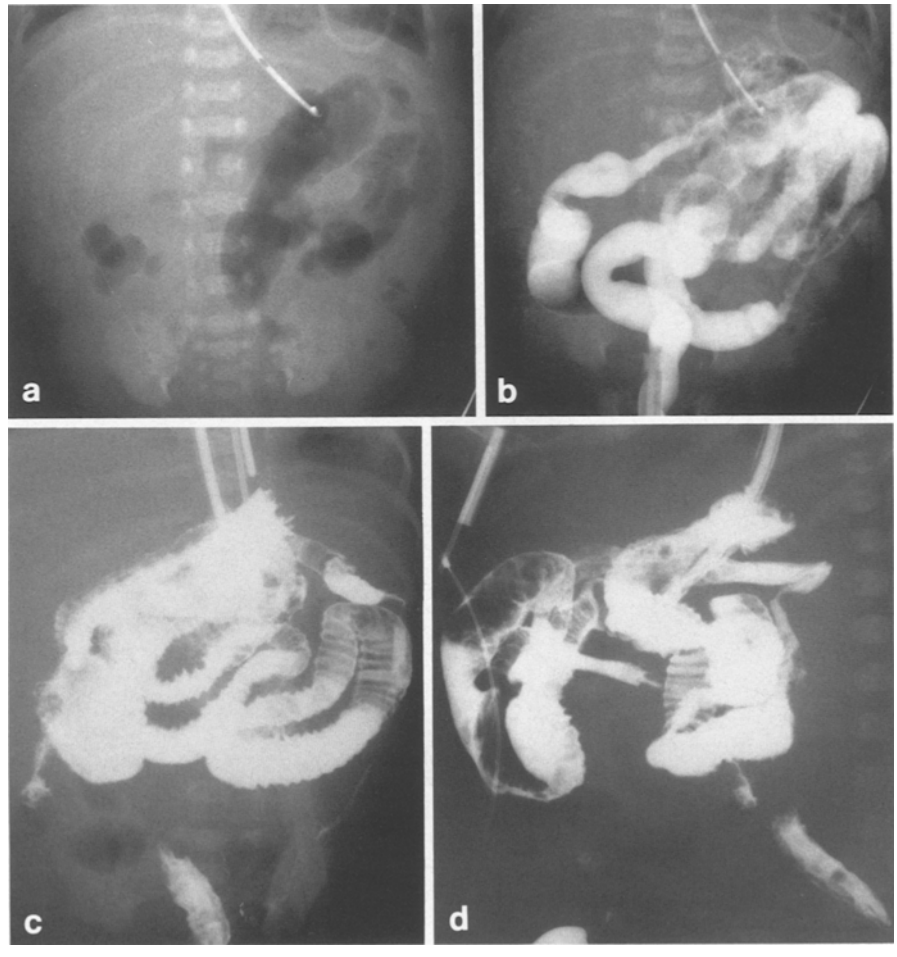

Fig. 1. **a** Supine abdominal film, two weeks after an episode of NEC, demonstrates mild distention of several bowel loops. **b** Water soluble enema. There is filling of several small bowel loops in the left upper quadrant from the splenic flexure. **c, d** Frontal (**c**) and lateral (**d**) views from a barium upper G. I. series the following day demonstrate early filling of the colon and rectum from the proximal small bowel

Table 1. Cases of enteric fistulas due to necrotizing enterocolitis

Author	Diagnosis of NEC (age)	Diagnosis of fistula (age)	Birth weight	Fistula location	Colonic stricture	Clinical presentation	Follow up
Pein [2]	7 days	35 days	?	Gastrocolic	No	Vomiting, Diarrhea	Died post surgery
Firor [3]	?	28 days	?	Gastrojejunocolic	No	Vomiting, Diarrhea	Died post surgery
Beck [4]	5 days	18 weeks	2610 g	Ileocolic	Yes	Distention, Constipation	Surgery, did well
Kosloske [5]	?	18 weeks	?	Enterocolic	No	?	Surgery, did well
Paley [6]	7 days	63 days	3800 kg	Jejunocolic	Yes	Distention, Vomiting	Surgery, did well
Kiely [7]	5 days	32 days	3600 g	Jejunoileocolic proximal	Yes	Vomiting, Diarrhea	Surgery
Case 1	6 days	25 days	820 g	Jejunocolic	No	Distention	Died prior to surgery
Case 2	22 days	43 days	1190 g	Jejunoileocolic (Terminal ileum abscess)	No	Distention	Surgery, did well

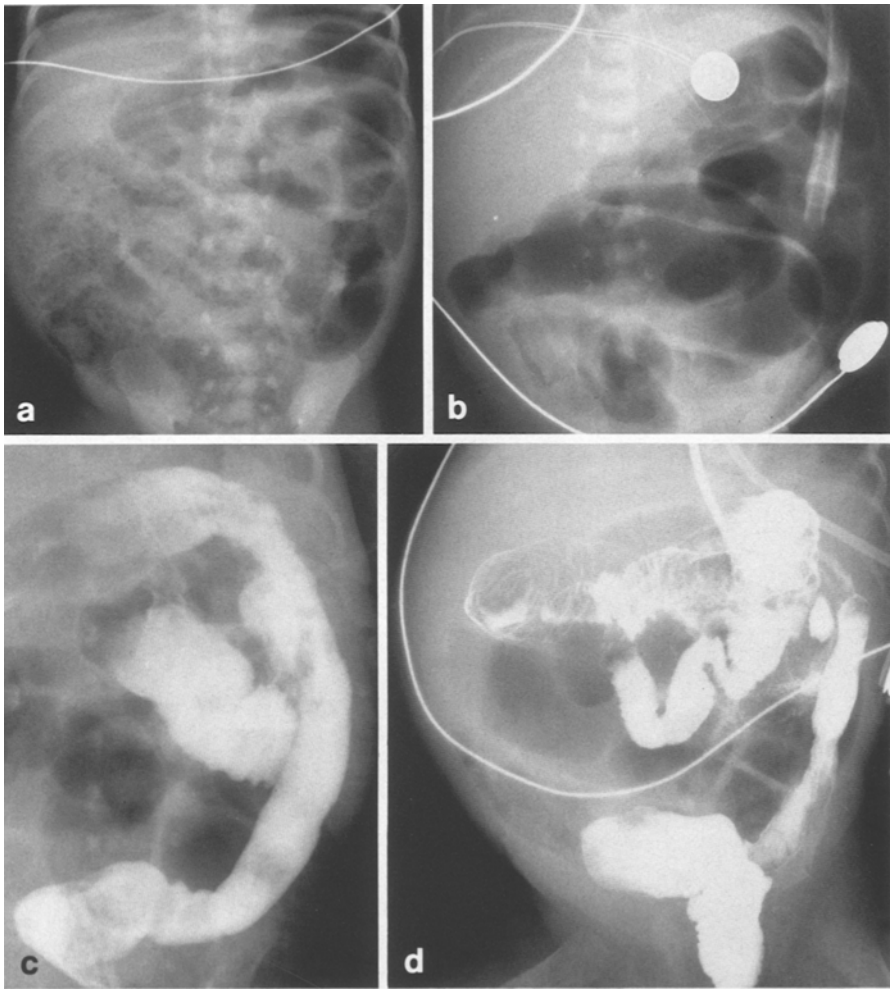

Fig. 2. **a** Supine abdominal film showing multiple dilated bowel loops and extensive pneumatosis intestinalis. **b** Supine abdominal film several weeks later showing multiple distended bowel loops. **c** Water soluble enema demonstrates filling of a single small bowel loop from the descending colon. **d** Barium upper G. I. series. There is filling of the descending colon from a proximal small bowel loop

the small bowel and the proximal descending colon (Fig. 2c) which was confirmed by an upper G. I. series (Fig. 2d). Surgery demonstrated a large fistula between the jejunum and descending colon, as well as fistulae between various loops of mid small bowel, requiring intestinal resection and diversion. In addition, an abscess in the right lower quadrant resulting from a local perforation of the proximal ileum was resected.

Discussion

Strictures complicating NEC occur in 20% of patients and usually involve the colon [1]. In contrast, fistula formation complicating NEC is rare and may occur with or without an associated stricture. Clinical data on our 2 patients and the 6 previously described in the literature are summarized in Table 1.

The plain film findings in the presence of a fistula are nonspecific. Clinically, our patients and the previously reported cases presented with diarrhea and/or abdominal distention (Table 1). These symptoms, in an infant several weeks following the resolution of an episode of NEC, and in the presence of a nonspecific bowel gas pattern on plain film, should raise the possibility of an existing fistula. The obstructive bowel gas pattern seen in our second case of fistula formation, although more suggestive of a stricture complicating NEC, can be explained by the presence of an abscess at the terminal ileum, acting as an obstructing lesion.

Several mechanisms may be proposed for the development of enterocolonic fistulae following NEC. Severe colonic ischemia and subsequent bowel necrosis may develop over time, inciting an inflammatory response rather than resulting in frank bowel perforation. Continuous inflammation may result in adherence of the affected segment of colon to adjacent bowel and eventual fistulization. Alternatively, a subacute perforation may be walled off by adjacent viscera, resulting in fistula formation.

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