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Aggressive gastrointestinal food allergy in neonates and its possible relationship to necrotizing enterocolitis

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ABSTRACT

INTRODUCTION: The incidence of gastrointestinal food allergy (FA) in neonates is increasing. Despite this, cases of patients with gastrointestinal FA who develop necrotizing enterocolitis (NEC) requiring laparotomy are extremely rare.

PRESENTATION OF CASE: We describe two cases that presented with bloody stool with a probable diagnosis of FA as eosinophils were positive in the stool at onset. Both cases failed conservative treatment. Jejunostomy and ileostomy were performed in both cases due to secondary NEC with underlying acute FA. Post-surgery, raised peripheral blood eosinophil count, presence of cow's milk-specific IgE antibody and positive allergen-specific lymphocyte stimulation test were found. Stoma closure were performed 3 and 1 months later in both cases. Postoperative recovery was uneventful.

DISCUSSION: A few reports have not identified risk factors for NEC secondary to FA. Thrombocytopenia and rise in C-reactive protein (CRP) levels 2 days after the development of FA may be suggestive of FA with NEC. Methicillin-resistant *Staphylococcus aureus* (MRSA) was detected in the fecal culture of both patients at the time of the onset of NEC. The toxic antigen produced by MRSA may cause activation of milk-protein-primed T cells and exacerbate FA.

CONCLUSION: The decrease of platelet levels and rise in CRP may indicate the development of secondary NEC in patients with FA. Additionally, MRSA detected in the fecal culture also may be a risk factor for NEC through the activation of cellular immunity reaction pathways.

1. Introduction

Gastrointestinal food allergy (FA) in children tends to occur in the first 1–6 months of their life [1–3]. The incidence of FA in neonates is increasing; although, it is still a relatively rare condition. Progressive FA in the neonatal period may need to be clinically differentiated, especially in those with mechanical bowel obstruction [2]. Reports of FA in neonates who subsequently develop necrotizing enterocolitis (NEC) is extremely rare [1,3–5]. Furthermore, common risk factors for the development of NEC secondary to FA are unknown. Targeted treatment for FA in patients cannot be started until a definite diagnosis is made, with repeated detailed clinical inspection and conservative treatment being a reasonable initial strategy. However, a decision should be made for laparotomy if a patient develops NEC. Therefore, a combination of both conditions presents difficult clinical problem. We report 2 neonates with FA who required laparotomy due to secondary NEC. We reviewed previous reports (in English) on patients with this condition. Here, we attempt to determine risk factors for the development of NEC in patients diagnosed with FA based on the clinical features of our 2 patients. Our report is in accordance with the SCARE criteria [6].

2. Presentation of case

2.1. Case 1

A preterm boy born at 28th week of gestation with a birth weight of 1133 g suddenly presented with abdominal distension and bloody stool on day 25 at the neonatal intensive care unit (NICU) while being fed on both breast milk and infant formula. The development of FA was suspected as stool examination was positive for eosinophilic inflitration. However, abdominal X-ray imaging showed pneumatosis intestinalis soon after the development of FA. Abdominal distention worsened progressively and another abdominal X-ray image showed diffusely dilated intestine (Fig. 1a) within a couple of hours after the initial X-ray. An emergent laparotomy was performed based on the diagnosis of NEC. During surgery, the colon was noted to be discolored, dark red and spotty, with the presence of pneumatosis intestinalis,

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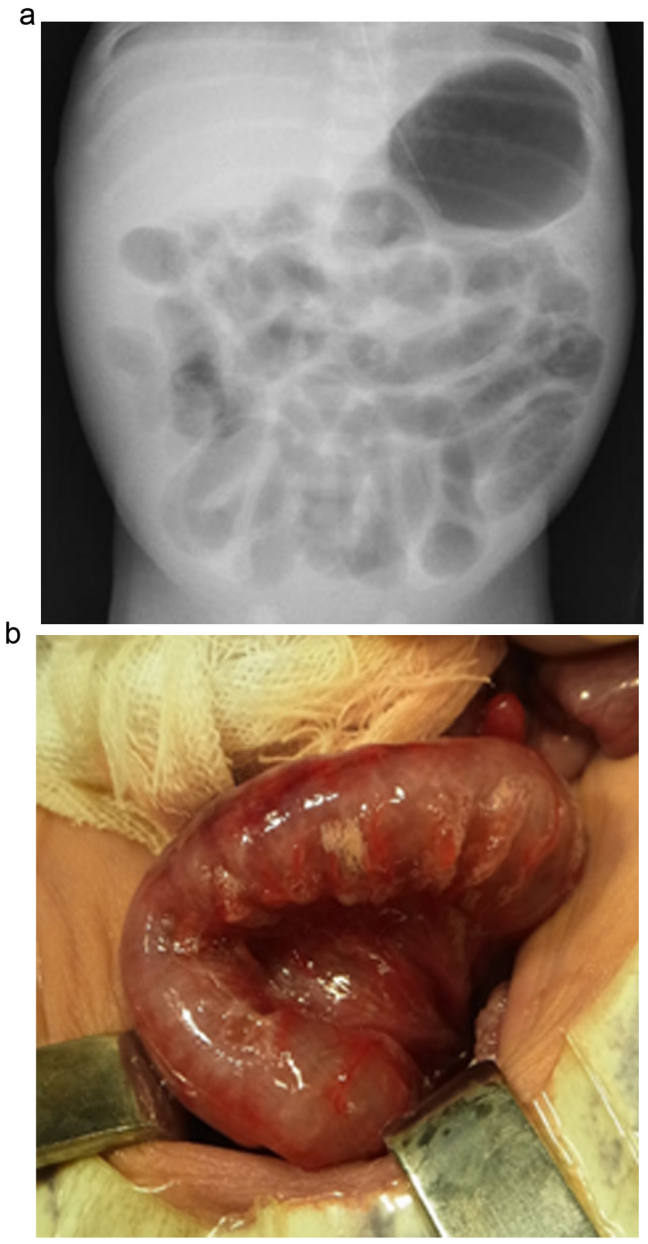

Fig. 1. (a) The abdominal X-ray shows pneumatosis intestinalis and diffusely dilated intestine. (b) Pneumatosis intestinalis is seen in the transverse colon at laparotomy.

a characteristic feature of NEC (Fig. 1b). Thankfully, no intestinal perforation was found. Ileostomy was performed. After 2 days from the onset of FA, C-reactive protein (CRP) levels were noted to have risen from 0.57 mg/dL to 12.22 mg/dL, and the platelet count dropped from $35 \times 10^4/\mu L$ to $15 \times 10^4/\mu L$. Post-operatively, peripheral blood eosinophil count reached 22%, from 4% preoperativelyand cow's milk-specific IgE antibody and allergen-specific lymphocyte stimulation test (ALST) to lactoferrin were positive. This supports the diagnosis of FA complicated by NEC. In addition, methicillin-resistant *Staphylococcus aureus* (MRSA) was detected in the fecal culture at the time of the onset. The patient recovered without MRSA sensitive antibiotics. Ileostomy closure was performed 3 months later with concurrent sigmoidectomy and descending colectomy due to severe stenosis. Postoperative recovery was uneventful.

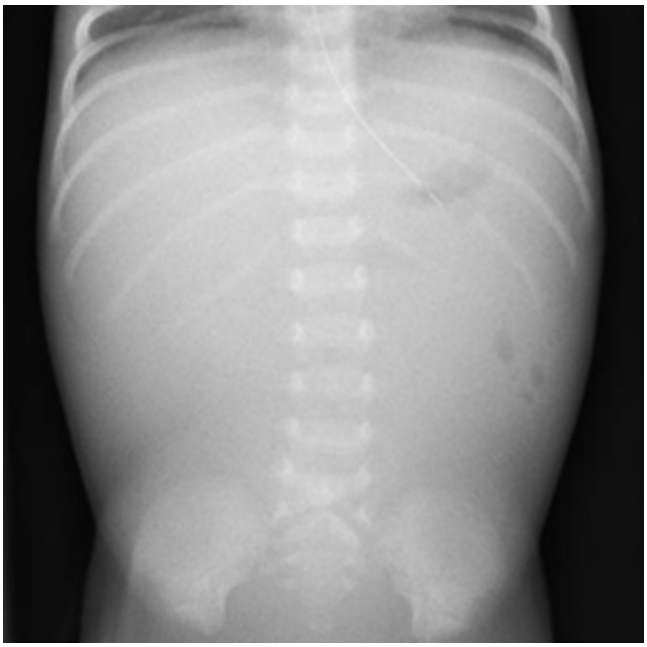

Fig. 2. Gasless abdomen is seen on abdominal X-ray.

2.2. Case 2

A full-term boy born at 38th week of gestation with a birth weight of 1980 g was transferred to NICU because of the sudden presentation of bilious vomiting and bloody stool on day 6 while being fed breast milk. Stool examination revealed eosinophilic infiltration. Development of FA was suspected. Conservative treatment was continued as mechanical intestinal obstruction was ruled out with abdominal ultrasonography and upper gastrointestinal series. However, he soon became unresponsive. An abdominal X-ray image showed gasless abdomen 2 days after the onset of symptoms (Fig. 2), and ascites was detected on abdominal ultrasonography. Two days after the onset of symptoms, CRP levels rose from 0.11 mg/dL to 14.0 mg/dL, and the platelet count decreased from $23 \times 10^4/\mu L$ to $4.0 \times 10^4/\mu L$. He was diagnosed with NEC and an emergent laparotomy was performed as conservative treatment was unlikely to succeed. Proximal jejunostomy was performed since almost the whole intestine and some parts of the colon showed extensive and severe inflammation, evidenced by marked swelling and a pale appearance. MRSA was detected in the fecal culture at the time of the onset. However, the patient also recovered without an MRSA sensitive antibiotics. The intestinal biopsy specimen taken during surgery showed irreversible necrotic change (Fig. 3a) and more than 20 eosinophils per field at x400 magnification (Fig. 3b). Phagocytosed MRSA were not identified in the biopsy specimen. After the surgery, peripheral blood eosinophil count reached 16% with positive ALST to lactoferrin. These findings support the diagnosis of FA complicated by NEC. Jejunostomy closure was performed a month later. Postoperative recovery was uneventful.

3. Discussion

Reports on neonates with FA who develop secondary NEC are extremely rare [1,3–5]. It is necessary to be cognizant of this condition as the incidence of FA in neonates is increasing. Therefore, it is critical to investigate the difference between patients with FA who recover with conservative treatment and those who require laparotomy. In this series, we performed laparotomy to decompress the distended intestine with resection of necrotic intestine as res-