

Double Intussusceptions with Necrotizing Enterocolitis Diagnosed in a Premature Infant

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ABSTRACT

Intussusception in neonates, especially in preterm infants, is rare. Common symptoms of intussusception, which include distended abdomen and gastric residuals, lead to misdiagnosis as necrotizing enterocolitis (NEC). Delayed diagnosis of neonatal intussusception delays treatment as well, which can lead to life-threatening complications. The predominant location of intussusception in preterm neonates is the small bowel; therefore, ultrasonography is not indicated. We report the case of an 834g male baby born at 25 weeks 6 days of gestation, who was diagnosed with double intussusceptions with NEC.

Key Words: Intussusception, Premature infants, Ultrasonography

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INTRODUCTION

Intussusception rarely occurs in neonates, especially in preterm infants¹⁻³⁾. The symptoms of intussusception in preterm infants are difficult to differentiate from those of necrotizing enterocolitis (NEC). The common symptoms of intussusception are a distended abdomen and increased gastric residuals, which can lead to misdiagnosis as NEC. This delays proper treatment of the intussusception⁴⁾. Most cases of intussusception in preterm infants are diagnosed upon operation. The diagnostic tool for intussusception in children is abdominal ultrasonography. However, a correct diagnosis in preterm infants is rare because the most common type of intussusception in preterm infants is ileoileal^{1,2)}.

We report a rare case of a premature infant diagnosed with intussusception by using abdominal ultrasonography.

CASE REPORT

A male baby was born at the gestational age (GA) of 25 weeks 6 days via cesarean delivery because of incompetence of the maternal internal orifice of the cervix uteri. His

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birth weight was 834 g and Apgar scores were 4 and 6 at 1 and 5 minutes, respectively. He was intubated in the operation room and received a prophylactic surfactant. On day 5 (GA, 26⁺³ weeks), a large (3.2 mm) patent ductus arteriosus (PDA) was diagnosed. He underwent surgery for PDA ligation on the same day. The next day, he received antibiotics and blood transfusions with packed red blood cells, platelets, and fresh frozen plasma because of disseminated intravascular coagulation syndrome. On day 18 (GA, 28⁺² weeks), he was extubated and nasal continuous positive airway pressure was applied. On day 21 (GA, 28⁺⁵ weeks), feedings were initiated with 1 mL of breast milk three times daily. Feedings were stopped on day 23 (GA, 29⁺⁰ weeks) when bilious residual was detected in the gastric tube and abdominal distention was noted. From day 28 to 33, he was fed with 1 mL of breast milk every 3 hours, with periodic checking for bilious gastric residual. After day 34 (GA, 30⁺⁴ weeks), trophic feeding was started but then withheld multiple times due to decreased bowel sound, abdominal distension, ileus, and increased or bilious gastric residuals. Beginning on day 60 (GA, 34⁺² weeks) of hospitalization, his feeding with breast milk through the gastric tube was gradually increased. He vomited once on day 74 (GA, 36⁺² weeks). A markedly distended abdomen and auscultated hypoactive bowel sound were evident. He became irritable when the abdomen was palpated. His laboratory findings showed the following values: white blood cell (WBC), 7,210/ μ L; segmented neutrophil, 71%; and platelet, 66,000/ μ L. His arterial blood gas analysis revealed a pH of 7.335, pCO₂ of 39.7 mm Hg, and HCO₃ of 25.6 mmol/L. His high-sensitivity C-reactive protein level was elevated to 7.92 mg/dL (reference range: 0–0.1 mg/dL). Methicillin-resistant *Staphylococcus aureus* (MRSA) was isolated from the patient's cerebrospinal fluid and central blood cultures. The diagnosis was MRSA meningitis with sepsis, and a 3-week treatment with vancomycin was started. Feeding was discontinued from day 75 (GA, 36⁺³ weeks). On day 80 of hospitalization (GA, 37⁺¹ weeks), feeding with 1 mL of breast milk at intervals of 3 hours was restarted. Brownish gastric residuals up to 8 mL were observed twice. We examined the simple abdomen and found ileus and progressive abdominal distention but no portal venous gas or pneumatosis intestinalis.

His complete blood cell count revealed the following values: WBC, 5,050/ μ L; segmented neutrophil, 53%; platelet, 83,000/ μ L; and high-sensitivity C-reactive protein (CRP), 5.14 mg/dL. His arterial blood gas analysis revealed a pH of 7.231, pCO₂ of

16.1 mm Hg, HCO₃ of 6.5 mmol/L, and metabolic acidosis. Upon suspicion of NEC, a rectal tube was inserted for decompression. During insertion of the rectal tube through the anus, bloody stool was discovered. Abdominal ultrasonography revealed a target sign suggesting ileocolic intussusception (Figure 1). Gastrografin reduction was performed with Gastrografin diluted in water at a ratio of 1:3, which was recommended for neonates by the manufacturer. During the reduction, Gastrografin leaked, indicating bowel perforation. He underwent immediate operation for manual reduction. Upon opening the peritoneum, brownish ascites was discovered. The intussusception was of the ileoileal type at the upper side, 20 cm from the ileocecal (IC) valve (Figure 2). Ileocolic intussusception was not observed because it had been reduced during the Gastrografin enema. Up to 8 cm of the terminal ileum was necrotized, and

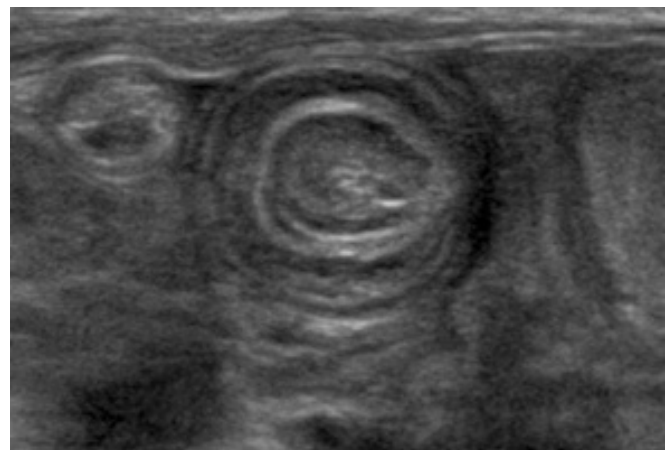


Figure 1. Abdominal ultrasonography showing target sign at the ileocolic area.

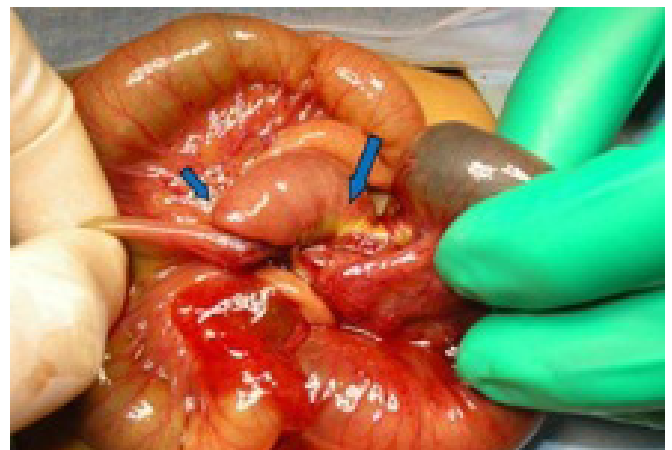


Figure 2. An intraoperative view of the ileo-ileal intussusception (short arrow) with dilated proximal bowel and intussusceptum, which was a necrotic bowel wall (long arrow).



Figure 3. The resected terminal ileum with hemorrhagic necrosis and perforation about 8 cm in length.

some parts of ileum was perforated or ulcerative (Figure 3). A segment of the ileum was resected, and then double barrel ileostomy was performed. The intraoperative tissue pathological findings showed an ischemic portion with a length of 7.5 cm, and transmural hemorrhagic necrosis with mucosal atrophy and loss of villi was observed.

After the operation, the patient experienced acute renal failure, which necessitated peritoneal dialysis. For 2 days after the operation (GA, 37⁺³ weeks), the blood urea nitrogen (BUN) and creatinine levels decreased and urination was acceptable. However, starting at postoperative day 3 (GA, 38⁺⁴ weeks), anuria persisted and the BUN and creatinine levels were elevated. The patient did not recover from the sepsis and disseminated intravascular coagulation. He died of sepsis and renal failure on day 94 of hospitalization and postoperative day 13 (GA, 38⁺⁶ weeks).

DISCUSSION

Intussusception is most commonly found in children aged between 3 months and 6 years¹⁾. The cause of intussusception in preterm neonates is idiopathic, unlike in full-term neonates^{3,5-7)}. The leading points of intussusception in full-term neonates are Meckel's diverticulum, congenital duplication cyst, polypoid lesion, hematoma, or meconium⁷⁻⁹⁾. In children and infants, the intussusception is ileocolic, while the predominant location in preterm neonates is the small bowel. Ileoileal intussusception makes contrast enema less valuable and diagnosis using abdominal ultrasonography less useful^{1,2,10)}. Only one of 10 cases are

preoperatively diagnosed as intussusception^{1,10)}. Most cases of intussusception are diagnosed by using laparotomy instead of ultrasonography¹¹⁻¹³⁾. In this case, ileocolic intussusception, clearly involving the ileocecal valve, was confirmed by performing ultrasonography. Diagnosis of ileoileal intussusception is sometimes missed on sonography. Surgical finding of ileoileal intussusception, in the present case, was located 20 cm above the IC valve and therefore regarded as an intussusception in a different location.

Neonatologists usually suspect NEC in preterm infants when the abdomen is distended due to the high prevalence of NEC at this age. The incidence of NEC is 1-5% of infants in the neonatal intensive care units¹⁴⁾. Symptoms of NEC are abdominal distention, gastric residuals, bilious vomiting, and bloody stool, which are similar to the symptoms of intussusceptions in neonates^{4,15)}. Differential diagnosis between neonatal intussusception and NEC can be confusing⁷⁾. The prevalence of intussusception is only 0.3% in neonates and is extremely rare in preterm infants^{1,3)}. Therefore, if symptoms of intussusception occur, clinicians could misdiagnose it as NEC, which is more common than intussusception in preterm infants. Delayed diagnosis of neonatal intussusception leads to delayed treatment and possible life-threatening complications¹⁻⁴⁾.

Distinguishing intussusception from NEC in the neonatal intensive care unit is not easy clinically and radiologically¹⁶⁾. Ultrasonography in diagnosing intussusception has about an 88% sensitivity, and simple abdominal test is preferable as the first diagnostic test for intussusception in infants¹⁷⁻¹⁹⁾. Even though intussusception in the small bowel is difficult to diagnose by using ultrasonography, the technique is useful in differentiating from NEC¹⁾.

Some cases of NEC with intussusception have been described^{5,6)}. Intussusception could be the cause of NEC but is rare. However, whether NEC precedes intussusception is unclear because the enteric vascular ischemia is the cause in both of cases²⁰⁾. Moreover, in the present case, whether NEC preceded intussusception, or vice versa, is unknown.

In this case, interesting points are that intussusception occurred at a later period than that in other reported cases diagnosed in preterm infants, as well as the fact that double intussusception occurred. In addition, we report this case because ileocolic intussusception, which is rare in preterm infants, is usually discovered upon explorative laparotomy. However, in this particular case, this rare type of intussusception in preterm infants was

discovered via ultrasonography. In conclusion, when the abdomen of a premature infant is erythematous or the infant displays gastric residuals at a later period than when NEC generally occurs, clinicians should also consider the possibility of intussusception and conduct screening via ultrasonography. Furthermore, the diagnosis of intussusception early in the disease course could help lessen complications due to the delayed treatment.

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