Intussusception in preterm infants

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Abstract
Two cases of intussusception in infants born at 26 and 30 weeks' gestation are described. The two infants presented in the neonatal period with abdominal distension, intolerance of feeds, and rectal bleeding. An initial diagnosis of necrotising enterocolitis was made and the infants were treated medically. This led to a delay in the diagnosis of the intussusception.

Published work on neonatal intussusception is reviewed and attention is drawn to the fact that the presenting signs and symptoms can be similar to those of necrotising enterocolitis. A diagnosis of intussusception should therefore be considered in any preterm infant with suspected necrotising enterocolitis.

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Abdominal distension, intolerance of feeds, and rectal bleeding in infants receiving intensive care usually leads to a diagnosis of necrotising enterocolitis. Intussusception can mimic this disorder so that there may be a delay in making the diagnosis. We report two cases of intussusception in infants born at 26 and 30 weeks' gestation.

Case reports

PATIENT 1
A boy was born by elective caesarean section at 26 weeks' gestation, the third of sextuplets to a 28-year-old primigravida. His birth weight was 765 g. He was ventilated for hyaline membrane disease from birth until day 8, after which he was weaned onto continuous positive airways pressure via a nasal prong. The first four days were complicated by episodes of hypotension which were treated with infusions of colloids and a dopamine infusion. A cerebral ultrasound scan on the fourth day showed bilateral grade 3 periventricular haemorrhages.

Initial feeding was parenteral; enteral feeds were introduced, by nasogastric tube, on day 3. These were discontinued on day 15 because of bile stained aspirates, but examination of the abdomen was normal. From day 30 nasogastric feeds were reintroduced, but not well tolerated, so on day 38 a transpyloric tube was passed and its position was confirmed radiographically to be in the proximal duodenum. Feeding was started again using a half strength preterm formula, but on day 48 large gastric aspirates occurred, abdominal examination showed distension, and several masses were palpable in the left iliac fossa. Normal stools had been passed 48 hours earlier.

In view of the possibility of constipation, a glycerine enema was given and resulted in large amounts of stool and fresh blood. Treatment for necrotising enterocolitis, namely stopping feeds by mouth, total parenteral nutrition, and intravenous antibiotics, was begun. Stool and blood cultures taken at this time did not grow pathogenic organisms.

Abdominal radiographs on day 48 showed a dilated bowel loop and fluid levels indicating the presence of intestinal obstruction. This persisted over the next five days. A laparotomy on day 54 showed an intussusception in the mid-jejunum and three perforations in the necrotic segment of the bowel distal to the intussusception. Reduction of the intussusception and resection of the necrotic segment with end to end anastomosis were performed. Progress after the operation was uneventful and feeds by mouth were well tolerated.

PATIENT 2
A boy, birth weight 1240 g, at 30 weeks' gestation, was the first of twins and was born by assisted breech delivery. He developed respiratory distress secondary to congenital pneumonia, which was treated with antibiotics. Blood cultures and secretions from the endotracheal tube, taken before treatment with antibiotics was started, were sterile. He required intermittent positive pressure ventilation from the age of 5 hours, at which time he had a severe metabolic acidosis and haematological evidence of disseminated intravascular coagulation. At 15 hours he received a two volume exchange transfusion of fresh heparinised blood for hyperbilirubinaemia (228 µmol/l) via a posterior tibial artery catheter and a peripheral vein. The exchange transfusion was repeated via the same routes 16 hours later. His condition improved and at 34 hours of age he was extubated and in room air. He remained well until 79 hours of age when he became lethargic, then rapidly developed apnoea, shock, and acidosis. A diagnosis of septicaemia was made, the antibiotics were changed and a further two volume exchange transfusion was performed via an umbilical venous catheter. At 86 hours his abdomen became distended and he passed fresh blood and mucus through the rectum. Abdominal radiographs showed a dilated small bowel with fluid levels. A diagnosis of necrotising enterocolitis was made and he was treated medically for 45 hours, but then his condition...
deteriorated and radiographs showed free air, indicating the presence of perforation. At laparotomy a distal jejunojejunal intussusception was found with a segment of necrotic bowel proximally; there was no evidence of necrotising enterocolitis. A double barrelled stoma was used. He made a gradual recovery and his ileostomy was closed on day 37.

Discussion
The classical presentation of necrotising enterocolitis is with the triad of abdominal distension, rectal bleeding, and intolerance of feeds. The diagnosis is confirmed by the radiographic presence of intestinal, intramural gas (pneumatosis intestinalis). There is a broad spectrum of presentation, however, and necrotising enterocolitis is often diagnosed in the absence of these classical features. Rabinowitz and Siegel highlighted this in their review of 40 patients with necrotising enterocolitis, many of whom had atypical features.1

Although well described in older children, intussusception is uncommon in the neonatal period. Rachedel et al found that of almost 6000 published cases, only 28 had occurred in the neonatal period.2 Talwalker reviewed the 26 cases of neonatal intussusception described up to 1960,2 and a further review was made by Patriquin et al 17 years later.3 In most of their patients, which included only four preterm infants, vomiting and rectal bleeding were the presenting features; an abdominal mass and abdominal pain were much less commonly seen, though intussusception in the first 48 hours of life was often associated with a leading mass. These and several other papers have also described the occurrence of intrauterine intussusception leading to small bowel atresia.5–7

Thus, intussusception is a rare event in preterm infants. Smith and Giacoia summarised the characteristics of five previously reported cases and described two of their own.8 Of these, six presented with feeding intolerance, an abdominal mass was palpable in two, and rectal bleeding in four; abdominal distension was an almost universal finding. The onset of symptoms occurred at a mean of 11 days of age (range 1–30 days) and the diagnosis was made at a mean of 28 days (range 3–71 days). In one of these infants an initial diagnosis of necrotising enterocolitis was made.9 More recently, a case of intussusception in association with meconium plugs has been described and yet again diagnosis was delayed by an initial diagnosis of necrotising enterocolitis.10 Moreover, two cases described by Smith and Giacoia occurred in association with necrotising enterocolitis, thus further complicating the issue. It appears that there are no definite clinical features that allow a neonatologist to differentiate intussusception from necrotising enterocolitis.

There has been no previous report of the radiological features of intussusception in preterm infants. Several reports, however, have stressed the variety of radiological signs found in necrotising enterocolitis.11–12 Pneumatosis intestinalis, which is regarded by many as diagnostic of necrotising enterocolitis, may not occur simultaneously with clinical deterioration and has been observed in some patients before any clinical manifestation of the disorder. In a previous case report, intramural gas was seen in only 40% of the patients with necrotising enterocolitis described by Pochachevsky and Kassner11 and in only 12 of the 34 patients presenting to the Alder Hey Children’s Hospital in a four year period.12

At present, the clinical and radiological differentiation between necrotising enterocolitis and intussusception is not easy. It is important for those working in neonatal intensive care to be aware of the possibility of intussusception in association with necrotising enterocolitis or as an alternative diagnosis, particularly when there is a poor response to medical treatment. In the small, sick infant with bowel perforation, the diagnosis of necrotising enterocolitis, conservative management has been advocated, using simple peritoneal drainage. This technique may jeopardise the viability of the bowel if the infant has intussusception and not necrotising enterocolitis.13–14

Ultrasonography has proved to be useful in the diagnosis of intussusception in older children. Using strict diagnostic criteria, an ultrasound scan diagnosed intestinal intussusception in 100% of 145 consecutive patients described by Pracros et al.19 There were no false negative results among the 241 children in whom intussusception was excluded by ultrasound. The youngest child examined in this series was 3 weeks of age. Although this technique has not yet been described in preterm infants it may prove to be a valuable aid in distinguishing intussusception from necrotising enterocolitis.