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A Case Report of Ileoileal Intussusception in a Premature Neonate

Benjamin Hopkins, BS; Anabel Goulding, BA; Emily Ong, BA; Kenny Lam, BS

"Intussusception is the telescoping, or invagination, of a part of the intestine into itself; it is the most common abdominal emergency in children under two years of age (1). However, intussusception is extremely rare in the neonatal period, accounting for 1% of cases in children less than three months of age, and is encountered even less frequently in premature neonates (2)."

Keywords: Intussusception, neonate, ileoileal, preterm, premature, peritoneal, spontaneous intestinal perforation

Introduction:

Intussusception is the telescoping, or invagination, of a part of the intestine into itself; it is the most common abdominal emergency in children under two years of age (1). However, intussusception is extremely rare in the neonatal period, accounting for 1% of cases in children less than three months of age, and is encountered even less frequently in premature neonates (2). Establishing intussusception as a diagnosis in a neonate is further complicated due to clinical similarities with Necrotizing Enterocolitis (NEC), a more common disease in premature neonates (3). The clinical similarities between intussusception and NEC and the lack of imaging findings associated with intussusception often lead to a delay in diagnosis, increasing the risk of complications (3). We present a case of a 22.2 week (gestational age) female diagnosed with ileoileal intussusception after exploratory laparotomy on day of life 15. To our knowledge, this is the youngest reported case of intussusception. There have been other intussusception cases where the neonate was born at 23 weeks (3).

Case Presentation:

An extremely preterm, 22.2-week-old female weighing 464 grams was born vaginally to a G2P1 woman who had prenatal care. Pregnancy was complicated by vaginal bleeding and preterm labor. Antenatal steroids were not given. Mother was GBS negative, Rh-positive, and otherwise negative serology.

The newborn (NB) required PPV and intubation at delivery due to respiratory failure. On admission, she had pulmonary insufficiency and was given surfactant and placed on a High-frequency Jet ventilator (HFJV) with 100% FiO2.

Labs at birth revealed elevated WBCs, neutrophils, lymphocytes, monocytes, and bands. The NB was started on Ampicillin and Gentamicin while a blood culture was pending for sepsis. An Umbilical arterial catheter (UAC) was placed to gain central arterial access.

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At 23.3 weeks adjusted gestational age, a KUB showed a perforation in the small bowel. A peritoneal drain was inserted, and a Spontaneous Intestinal Perforation (SIP) diagnosis was made. An additional KUB two days later showed no signs of perforation.

Six days after the peritoneal drain was placed, at 24.3 weeks gestational age, bleeding was noted from the insertion site. The NB was anesthetized, and the peritoneal drain was removed. Exploratory laparotomy was performed, and an ileoileal intussusception was discovered; additional perforations at the site of intussusception and proximal ileum were also noted, confirming the previous diagnosis of SIP. Surgery consisted of segmental distal ileal resection at the intussusception site with primary anastomosis. Additionally, mid-segmental ileal resection with the creation of ileostomy and the mucous fistula was also performed.

The bowel appeared pink without evidence of necrosis, and the NB handled the procedure well. The postoperative period was uneventful, and the patient was transferred to a Level Four NICU, where the patient eventually expired due to further complications.

Discussion:

Most intussusceptions in children are idiopathic, with only 25% caused by a pathological lead point, most commonly lymphoid hyperplasia (4). Intussusception typically occurs between 6 and 36

months of age, with 90% being under 24 months, while only 1% is found in infants younger than three months (5, 6). When intussusception is found outside the typical age range, it is most often associated with a pathological lead point (7). The site of intussusception can be anywhere throughout the bowel and, although rare, can also present at multiple sites throughout the bowel.

"When present in children aged 6 to 36 months, intussusception is most frequently (>90%) identified as involving the ileocecal junction (2). Despite the high prevalence of the ileocolic type of intussusception in children, this is not the case for preterm neonates."

When present in children aged 6 to 36 months, intussusception is most frequently (>90%) identified as involving the ileocecal junction (2). Despite the high prevalence of the ileocolic type of intussusception in children, this is not the case for preterm neonates. A 2021 review of 52 cases of intussusception in premature neonates found that the site involved most frequently in this age group was ileoileal intussusception (61%), while ileocolic intussusception was only found to be the third most common site for intussusception (8%) in a premature neonate (3). Also discussed in this review was the frequency of presenting symptoms for premature neonates that were diagnosed with intussusception. Abdominal distension was reported in 85% of cases, gastric residue in 77% of cases, bloody stool in 44%, and an abdominal mass in 16% (3). Only a few aspects of our case are consistent with the most common presentation outlined in this review paper. Abdominal distension and ileoileal intussusception were both found in the presenting case; however, gastric residue, bloody stool, and an abdominal mass were absent. Furthermore, the co-occurrence of SIP and intussusception further complicated the case. The lack of clinical manifestations in this case and the rarity of this condition contributed to the difficulty of diagnosing an intussusception prior to exploratory laparotomy.

Intussusception and NEC present similar symptoms: abdominal distention, bilious vomiting, feeding difficulties, and bloody stool (8). When these similar symptoms are presented in a premature infant, NEC would be the first differential due to the prevalence in this population and the need for rapid diagnosis. Following suspicion of NEC, serial abdominal x-rays would be performed. Radiologic findings of pneumatosis intestinalis and portal venous gas are found in NEC, while free abdominal air is the usual finding in neonates with intussusception (8). Spontaneous intestinal perforation (SIP) is also common in extremely premature infants (9). SIP is usually located in the terminal ileum, with the rest of the bowel showing no abnormalities (8). The diagnosis of SIP was made intraoperatively in this patient at eight days of life. While SIP and NEC are common diagnoses in premature infants, SIP presents in the first two weeks of life, whereas NEC usually occurs from the second to the third week of life (8).

The exact etiology of neonatal intussusception is still unknown. A possible explanation includes an intestinal stricture caused by hypoperfusion acting as a lead point for the intussusception (10). Meconium was found during the explorative laparotomy in our patient. Dysmotility of the intestine could be due to a combination of abdominal ganglia immaturity and ischemia with reperfusion causing meconium ileus (11). The congealed nature of meconium can subsequently act as a lead point for intussusception (12).

Conclusion:

Intussusception is exceedingly rare in a premature neonate. Moreover, this case presents the youngest diagnosed incidence of intussusception. The common causes of intussusception in infants include hyperplasia of lymphoid tissue or congenital anomalies; however, the exact etiology of this case has yet to be identified. Infants with intussusception can present variably from the symptoms of abdominal distention, vomiting, and occult blood in the stool to just lethargy. The rarity of intussusception in premature neonates and the difficulties of differentiating it from more common bowel disorders, such as NEC, presents a challenge for rapid diagnosis and prompt management of intussusception.

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Furthermore, a premature infant may present with both disorders simultaneously. A high degree of clinical suspicion is often needed to make this diagnosis early to prevent treatment delay and subsequent complications of perforation and bowel resection. Further research of intussusception in conjunction with other abdominal disorders like NEC and SIP in extremely premature neonates is warranted to improve the understanding of GI abnormalities in this age group.

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Corresponding Author

Benjamin Hopkins, BS
Western University of Health Sciences,
College of Osteopathic Medicine of the Pacific,
Pomona, CA, USA

Anabel Goulding, BA
Western University of Health Sciences,
College of Osteopathic Medicine of the Pacific,
Pomona, CA, USA

Emily Ong, BA;
Western University of Health Sciences,
College of Osteopathic Medicine of the Pacific,
Pomona, CA, USA

Kenny Lam, BS
Western University of Health Sciences,
College of Osteopathic Medicine of the Pacific,
Pomona, CA, USA