

Spontaneous Intestinal Perforation in a Full Term Neonate: A Case Report

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ABSTRACT

Spontaneous intestinal perforation suggests a perforation of the gastrointestinal tract of no demonstrable cause. Only few such cases have been described in full term newborns. Although some authors have suggested ischemia and fetal or neonatal hypoxia as the most likely cause, the etiology and pathogenesis of this disease are still widely unknown. Here, we present a case of Spontaneous intestinal perforation in a full term neonate who was born to a mother with chronic hypertension. Radiological images revealed pneumoperitoneum on a three day old baby with feeding intolerance and abdominal distention. There was no clinical evidence of Hirschsprung's disease, necrotizing enterocolitis, anorectal malformations or bowel obstruction. An emergency laparotomy revealed a localized perforation in distal ileum. Histopathological examination failed to reveal any etiology for the perforation. The neonate recovered rapidly following a double barrel ileostomy, with no immediate gastrointestinal complications.

Keywords: Bowel perforation; intestinal; neonate; spontaneous I

INTRODUCTION

Neonatal bowel perforation is commonly secondary to necrotizing enterocolitis (NEC) or mechanical obstruction, and rarely spontaneous. Spontaneous intestinal perforation (SIP) suggests a perforation, typically found in the terminal ileum with no demonstrable cause. ¹ Although seen more frequently in preterm, low birth weight newborns, only a few cases have been described in full-term newborns.¹⁻³ Although conditions associated with fetal or neonatal hypoxia have been proposed as important antecedent causes, the etiology and pathogenesis of the disease is still unknown.¹ It is a separate clinical entity from NEC and this differentiation is important because of management and outcome considerations.

CASE REPORT

A 2870 gram full term male infant was admitted to our Neonatal Intensive Care Unit (NICU) with a diagnosis of infant of diabetic mother and suspected transient tachypnea of newborn. Baby was born to a 28 year old primigravida mother following in-vitro fertilization. Mother also had chronic hypertension, under treatment with labetalol and amlodipine, and subclinical hypothyroidism. Caesarean section was performed for borderline oligohydramnios. Baby cried immediately. APGAR score was 8/10 and 9/10 at 1 and 5 minutes respectively. Few minutes after birth, baby developed grunting and was transferred to NICU. Baby

was supported with bubble CPAP. He was fed via orogastric tube. He passed stool within 24 hours after birth. At 24 hours of life, he developed 2 episodes of vomiting. Septic screening as well as chest and abdominal x rays done at the time were normal. In view of suspected early onset neonatal sepsis, he was treated with intravenous antibiotics and was kept nil by mouth.

At 46 hours of life, there was abdominal distension without abdominal wall erythema, tenderness or muscle guarding on palpation. In view of the stable general condition and no clinical evidence of peritonitis, conservative management was continued. At 50 hours of life abdominal distension significantly increased. Repeat abdominal x-ray revealed signs of pneumoperitoneum. [Figure 1(a), 1(b)]. Clinical diagnosis of perforation was made. Immediate exploratory laparotomy was done and double barrel ileostomy with peritoneal lavage and peritoneal drainage was performed. Intraoperative findings showed a single perforation of terminal ileum approximately 20 cm proximal to ileocolic junction, measuring 0.5 x 0.5 cm². The intestinal mesentery was oedematous and inflamed with petechial bleeding and

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inflammatory fibrin flakes. [Figure 2(a), (b)] There was no clinical evidence of intestinal necrosis. Biopsy was taken from the perforation site for histopathological examination. Stoma was functional within 12 hours of surgery. Trophic feeding was started on second post-operative day and full enteral feeding was achieved by fifth post-operative day. He was discharged on eighth post-operative day.

The histopathological examination was consistent with inflammatory changes secondary to perforation. Although mature ganglion cells were not identified in immunohistochemistry, presence of granular staining by calretinin of myenteric nerve plexus and lack of hypertrophic nerve bundles excluded the possibility of Hirschsprung's disease. (HD)

DISCUSSION

Pneumoperitoneum is a rare condition with high morbidity and mortality. It is usually an indication of perforated intestine and requires prompt surgical intervention.^{4, 5} Neonatal bowel perforations have varied etiologies and NEC is the leading cause.¹ Besides NEC, there are numerous other causes of bowel perforation including intestinal obstruction, regional hypoperfusion due to stress, hypoxia or shock, Hirschsprung's disease, meconium ileus and SIP.⁶

In HD, intestinal perforation is seen in 3.2% to 4.4% of the cases. Areas of perforation are most often observed in the colon and appendix. Our baby had passed stool within 24 hours of life and perforation was noted in terminal ileum which is not in keeping with HD. Although mature ganglion cells were not identified in immunohistochemistry, presence of granular staining by calretinin of myenteric nerve plexus and lack of hypertrophic nerve bundles excluded the possibility of HD.

Weinberg et al. in 1989 suggested a vascular theory where localized vascular accident over the anti-mesenteric wall of the intestine might cause ischaemic necrosis leading to perforation. But in our case, there were no features of NEC and no localized ischaemia.⁷

The most common abdominal cause of the pneumoperitoneum in premature infants is pneumatosis intestinalis.⁸ Complication of mechanical ventilation such as pneumothorax and pneumomediastinum can lead to pneumoperitoneum due to extension of air along vascular planes through the normal diaphragmatic openings into the abdomen.⁶ Our case had no such complications and laparotomy revealed the presence of bowel perforation.

In our case, the mother had chronic hypertension which was difficult to manage during pregnancy. This might have theoretically caused gut hypoxia in the fetus. This is by far the only possible etiology for SIP in this baby.

SIP without clinical or histological evidence of NEC is a distinct entity in neonates. Our case was a full term neonate without features of NEC, and hence, was presumed to have SIP, a localized condition. The clinical experience and intra-operative findings in SIP suggest a self-limiting character of the disease, but there are a few reports of recurrent or multiple perforations as well.⁶ It is debated whether SIP can seal spontaneously restoring the integrity and patency of the small bowel. However, there is a small subset of patients with even mild abdominal distension and minimal peritoneal signs, who still require surgical intervention. Therefore, the neonates with suspected pneumoperitoneum require a proper clinical and radiographic correlation to establish the etiology of perforation and the clinical picture should guide the therapy.

Although spontaneous healing of gut perforations in neonates has been documented and an initial conservative management for intestinal perforation have been suggested by some authors, early surgical intervention remains to be the cornerstone in the treatment of SIP.⁶

Definitive surgical treatment in SIP includes primary closure if possible, resection and re-anastomosis and ileostomy formation.^{9,10} Due to abdominal contamination with bile and intestinal material, primary closure or resection and re-anastomosis were found to be hazardous and so a loop double barrel ileostomy at the ileal perforation site was performed in our case. Although there is no general consensus concerning timing of stoma closure in neonates, early ileostomy closure is recommended by some of the authors.⁶

CONCLUSION

Bowel perforation in a neonate is an emergency with a grave prognosis if it is not diagnosed and treated early. The most common cause remains NEC, although SIP is also seen rarely in neonates with pneumoperitoneum. The exact aetiology of SIP is still debatable and multiple theories have been proposed. Despite varied etiologies, definitive treatment for pneumoperitoneum is surgical and has a better outcome if early intervention is performed. The problem of delay in getting adequate medical care among developing countries may attribute to more morbidity and mortality rate among neonates being admitted to NICUs with a diagnosis of pneumoperitoneum.

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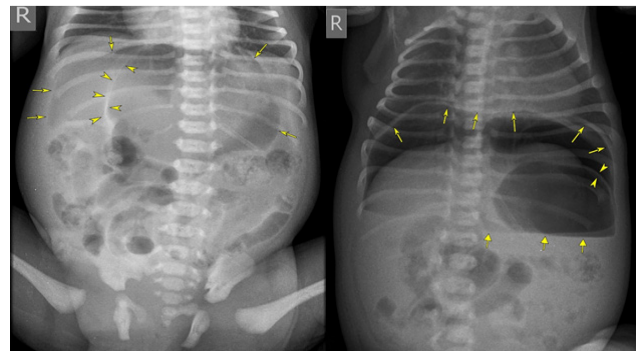


Fig 1a: Supine abdominal radiograph demonstrates large oval lucency (arrows) in upper abdomen divided by falciform ligament (arrow heads) giving the appearance of “football sign” and “falciform ligament” sign due to presence of large amount of air in peritoneal cavity.

Fig 1b: Radiograph of chest and abdomen (erect) revealed air under the domes of diaphragm with continuous visualization of lower margin of diaphragm across the midline “continuous diaphragm” sign (arrows). Both walls of body of stomach (arrow heads) seen in superolateral aspect (“Rigler double wall sign”). Air fluid level also seen (thick arrows) suggesting fluid in peritoneal cavity Air fluid level also seen (thick arrows) suggesting fluid in peritoneal cavity

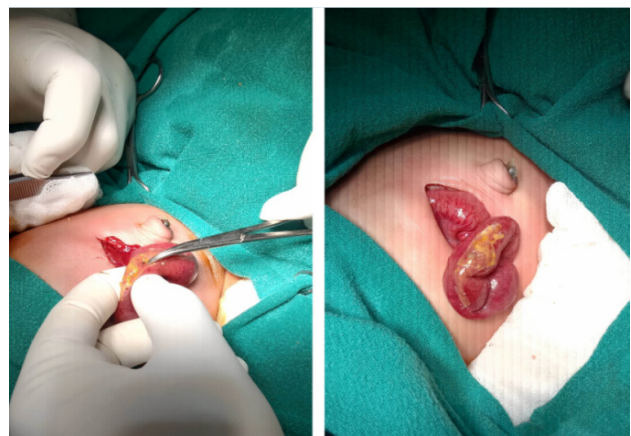


Figure 2a

Fogure 2b

Fig 2(a): A localized perforation in ileum.

Fig 2(b): Inflamed and edematous intestinal mesentery with petechial bleeding and inflammatory fibrin flakes.