

Spontaneous Intestinal Perforation in Prematurity: A Case Report and Review of Literature

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Introduction

Spontaneous Intestinal Perforation (SIP) refers to a perforation in the gastrointestinal tract of a newborn with no demonstrable cause. This clinical entity is frequently seen in preterm newborns with Very Low Birth Weight (VLBW) and Extremely Low Birth Weight (ELBW) and typically found in the terminal ileum [1-4]. In this report management of a SIP in a preterm infant with ELBW is presented and discussed under the light of relevant literature.

Case

A 800-g pre-term female infant at 28 weeks gestational age was admitted to our Neonatal Intensive Care Unit (NICU) with a diagnosis of prematurity and respiratory distress syndrome. Antenatal history revealed an oligohydramnios and amniocentesis was performed. During stay in the NICU, mechanic ventilation for days and surfactant therapy was administered for hyaline membrane disease. A right pneumothorax was detected on 7th day and a tube thoracostomy under water seal was performed. After resolution of pneumothorax abdominal distention and a bluish discoloration was observed on the 26th day. Abdominal x-ray revealed a pneumoperitoneum (Figure 1). Laboratory investigations revealed neutropenia, thrombocytopenia and metabolic acidosis compatible with sepsis. Initial peritoneal drainage (PD) was performed for temporary stabilization and recovery. After a stabilization period of 6 days, exploratory laparotomy was performed. Intraoperatively a single isolated ileal perforation on the antimesenteric side of the bowel was found. Whole abdominal cavity was contaminated with bile and intestinal material. Biopsy was taken from the site of perforation and incidental appendectomy was performed. A loop ileostomy including perforation site was performed (Figure 2). Early gastrointestinal feeding was started on the first postoperative day. Histopathological examination of the excised specimens did not reveal Hirschsprung's disease or NEC. Integrity and patency of the distal bowel was confirmed with distal ileostography taken on the 28th postoperative day and the ostomy was closed 5 weeks

after the ileostomy procedure (Figure 3). Nasogastric feeds were started 5 days after surgical intervention. With an uneventful follow-up the patient is well and has a steady increase in body weight.



Figure 1: Abdominal graphy showing free air under diaphragm

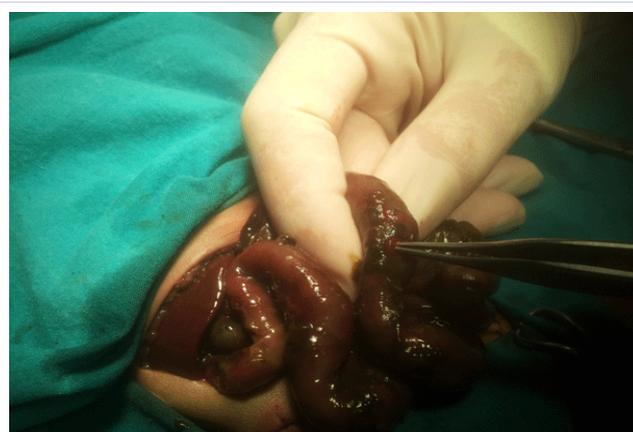


Figure 2: Operative view. Note there is a single perforation in the ileum and serosal surfaces of the bowels were stained with bile



Figure 3: Distal ileostography showing integrity and patency of bowel distal to ileostomy before stoma closure

Discussion

Despite improvements in anesthesia and neonatal intensive care, gastrointestinal perforation in neonates and pretermes presents a great challenge. Reported rates of mortality range from 15% to 70% and even more in critically ill premature infants [5-11]. Neonatal bowel perforations have varied etiologies and Necrotising Enterocolitis (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, mechanical injury during gavage feedings, rectal thermometers, resuscitation with oxygen under high pressure and SIP [12-15]. Although the etiology of SIP remains unknown, it has been postulated that transient intestinal ischemia results in SIP [16]. SIP and NEC have also been regarded as different manifestations of the same pathogenic process. Whatever the initiating cause, intestinal perforation is commonly seen on the antimesenteric region of ileum in SIP without evidence of NEC. But isolated perforations resembling SIP have also been reported in the transverse and descending colon [16]. Apart from a single perforation located in the distal ileum, the abdominal cavity and serosal surfaces of the abdominal viscerae were found to be contaminated with bile and intestinal material in our patient and there was no evidence of NEC such as pneumatosis intestinalis or necrosis in the remaining bowel.

Pneumoperitoneum is usually an indication of perforated intestine and requires prompt surgical intervention [17,18]. In addition to bluish discoloration of the abdominal wall, a gasless abdomen and absence of pneumatosis intestinalis have also been reported as further significant markers in infants with SIP [4]. All these findings were observed in our patient. Other radiological findings of intestinal perforation other than pneumoperitoneum include clear visualisation of the outer and the inner wall of bowel loops (Rigler's sign), triangular gas collections between the intestinal loops (sign of triangle) and gasless abdomen [2,16,19]. Only gasless abdomen on x-ray was observed in our patient preceding pneumoperitoneum.

Although spontaneous healing of gut perforations in neonates have been documented and an initial conservative management for intestinal perforation have been suggested by some

authors, early surgical intervention remains to be cornerstone in the treatment of SIP. Primary peritoneal drainage (PD) have been suggested as a primary or definitive procedure [20-25]. It allows acute improvement, systemic recovery but most of these infants require a subsequent laparotomy. Initially PD was performed in our patient because general anesthesia and laparotomy were regarded as risky and it provided time for stabilization of the baby. Definitive surgical treatment in SIP include primary closure if possible, resection and re-anastomosis and ileostomy formation [16,26]. Due to abdominal contamination with bile and intestinal material, primary closure or resection and re-anastomosis were found to be hazardous and a loop ileostomy at the ileal perforation site was performed.

Although there is no general consensus concerning timing of stoma closure in neonates and pretermes, early ileostomy closure can be safely done and should not be delayed [27-29]. Ileostomy complications occur as the time passes by and include stomal prolapse, skin excoriation, stricture etc. It has been reported that patients with ileostomy face with more stomal complications if ostomy closure was performed 2 months or later after ileostomy procedure [26]. Although stoma closure was performed on the 5th postoperative week in our patient, stomal prolapse was observed during her stay in NICU. Nevertheless, as soon as the patient gains weight and after performing a distal ileostography confirming the patency and integrity of the bowel distal to ostomy, there should not be a delay for stomal closure in these patients.

SIP is a distinct clinical entity in neonates and pretermes. Apart from PD, ileostomy formation especially critically ill pretermes may be life saving procedure. SIP seems to have a good prognosis even in VLBW infants if diagnosed and treated promptly.

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