

Postoperative Free Air is Not Always Normal - Necrotizing Enterocolitis Leading to Concurrent Colonic and Gastric Perforation

Batool Abdulla Almoallem, MBBCh, BAO, LRCPI, LRCSEd, Hind Zaidan, MBBS** Hussein Ahmed, MBBS, MRCS, FEBPS*** Martin T. Corbally, FRCSI FRCSEd, CCST, FRCS, FEBPS****

We present a case of concurrent gastric and bowel perforation in a preterm neonate associated with necrotizing enterocolitis (NEC). Gastrointestinal perforation of any cause is a surgical emergency with high morbidity and mortality. The most common cause of perforation in neonates is NEC followed by spontaneous ileal perforation.

Early suspicion of gastric perforation in preterm neonates with abdominal distention, and aggressive management with early resuscitation, stabilization and surgical exploration is recommended for better outcome.

Bahrain Med Bull 2020; 42 (1): 55 - 57

Gastrointestinal perforation in neonates is a life-threatening surgical emergency. The most common cause of perforation is necrotizing enterocolitis (NEC). Intestinal perforation is significantly more common than gastric perforation which although possible, is rare^{1,2}. Gastric perforation can be associated with prematurity, low birth-weight, neonatal intensive care unit (NICU) admission and NEC².

The aim of this report is to present a case of concurrent gastric and bowel perforation in a preterm neonate with NEC.

THE CASE

A preterm male infant was delivered by category II emergency cesarean section at 34+6 weeks gestation weighing 2.730 kg. Pregnancy was complicated by preterm labor, premature rupture of membranes and a maternal history of previous myomectomy and several abortions. APGAR scores at birth ranged from 8 and 9 at 1 and 5 minutes, respectively. On day 3 of life, he was transferred from the postnatal ward to the NICU with abdominal distension, poor sucking and bilateral scrotal swelling. A scrotal ultrasound showed bilateral hydroceles



Figure 1: Supine X-ray Showing Gross Pneumoperitoneum

with a right inguinal hernia and a suspected organized scrotal hematoma. He was started on empiric antibiotics (IV Ampicillin and Gentamycin) after blood culture was obtained. Abdominal X-ray revealed extensive pneumoperitoneum, football sign and lucency over the liver, see figure 1.

Emergency laparotomy with resection of the perforated transverse colonic segment and formation of colostomy with mucus fistula were performed. A Penrose drain was positioned in-situ in the right lower quadrant. He was on patient triggered ventilation (PTV) mode with Fraction of Inspired Oxygen (FiO₂) 22% and kept on triple antibiotics (IV Ampicillin, Gentamycin and Metronidazole). He was stable postoperatively and extubated after 2 days. He was fed by total parenteral nutrition via a peripherally inserted central catheter (PICC).

On postoperative day 3, a routine abdominal X-ray showed a persistent pneumoperitoneum, see figure 2. A second laparotomy was performed and a new large gastric perforation at the greater curvature of the stomach was found. A partial gastrectomy was performed and 2 Penrose drains were inserted in the right and left quadrants. He was transferred to the NICU, intubated and ventilated on PTV mode and extubated after one day. Serial imaging showed no recurrence of perforation, but he was maintained for 15 days on antibiotics and 6 days of Fluconazole. A Gastrografin study performed one week after the second laparotomy showed no evidence of contrast leakage or obstruction.



Figure 2: Lateral Decubitus View on Day 3 Postoperative Showing Free Air

* Resident
Department of Pediatrics
Salmaniya Medical Complex
** Registrar
Department of Pediatric Surgery
*** Senior Registrar
Department of Pediatric Surgery
**** Consultant Pediatric Surgeon and Chief of Medical Staff
Department of Pediatric Surgery
King Hamad University Hospital
Kingdom of Bahrain
E-mail: martin.corbally@khuh.org.bh

Enteral feeding was started on day 15 of life (12 days after initial surgery and 9 days after gastrectomy) through a nasogastric tube (NGT) and slowly progressed to oral feeding, which was well tolerated with no additional complications, see figure 3. The histopathology results revealed that both the colonic and gastric perforation occurred as a result of necrotizing enterocolitis.



Figure 3: Post-partial Gastrectomy Contrast Study (Gastrograffin)

DISCUSSION

Gastrointestinal perforation is a surgical emergency. NEC, multiple perforations, prematurity, and delayed presentation are the mortality risk factors¹². Our patient had three risk factors, which together constituted a major threat to survival. The etiology may differ as it depends on the site of perforation, intestinal or gastric. Most perforations occur in the intestines, mainly due to NEC followed by spontaneous perforation¹².

Gastric perforation is a rare entity in neonates with a mortality rate of 45% to 58%¹⁵. It is associated with prematurity, low-birth weight, neonatal intensive care unit (NICU) admission and NEC. It presents mainly during the first week of life with sudden abdominal distension². The exact etiology is unknown; it could be either iatrogenic (tube perforation, TEF fistula) spontaneous or secondary to NEC¹. Spontaneous perforation is rare (1:2900 live births)¹⁶. It could be associated with extreme resuscitation, use of nasal CPAP and perinatal stress, perinatal hypoxia-ischemia and distal obstruction. In addition, an ischemic perforation is described in the case of necrotizing enterocolitis^{1,15}.

NEC could be seen in the first 2 weeks of life, but it might be seen at 2 months of age in low birth-weight children⁸. The incidence rate of NEC in the United States varies from <1 to 5 cases per 1,000 live births¹³. The average annual incidence of NEC is 0.72 per 1,000 live births¹⁴. NEC is either primary or secondary. Primary NEC is more commonly seen in the first week of life of preterm infants with no trigger. Secondary NEC occurs in preterm or term infants associated with perinatal asphyxia, polycythemia, respiratory distress and congenital anomalies, myelomeningocele and congenital heart disease^{4,5}.

Studies have found that preterm infants have immature intestinal motility and an undeveloped intestinal barrier with insufficient colonization by commensal flora compared to term infants⁵. The main risk factors for NEC are prematurity, low

birth weight and enteral feeds^{4,5,7,13}. Formula milk increases the risk of NEC and gastrointestinal perforation¹². NEC is 6 to 10 times more common in infants with exclusive formula feeding; it is 3 times more common in infants who are fed both formula and breast milk compared to infants on exclusively breast feeding⁷. There is no association between gender and the rate of NEC⁵. There is a high mortality and morbidity rate, and 20%-40% require surgical intervention^{5,7,13}.

Bell's staging is used to classify neonates with NEC: stage 1 (suspected), stage 2 (definite) and stage 3 (advanced)^{5,7}. Treatment options are divided into medical and surgical. Medical treatment is conservative by discontinuing oral feeds, insertion of a nasogastric tube, IV fluids/TPN with antibiotics and close monitoring of vital signs^{6,8}. Surgical options are either insertion of primary peritoneal drainage or laparotomy. Laparotomy may involve resection with primary anastomosis or resection with enterostomy and mucus fistula formation. If the remaining bowel is healthy, primary anastomosis is the procedure of choice⁸. Pneumoperitoneum on simple radiography is a diagnostic sign of gastrointestinal perforation; therefore, it is a definite indication for surgery^{2,5,10,12}.

It is not unusual to find residual pneumoperitoneum after laparotomy. Persistent free air suggests an unrecognized perforation from the initial laparotomy, progression of the disease and further perforation or perforation from another site as in our case. Significant free air always indicates the need for further exploration because gastric perforation requires very aggressive treatment with wide resection/debridement and closure.

We have found no previous similar reports in the literature that describes a case of simultaneous gastrointestinal perforations in a neonate caused by NEC. We propose that the cause was a translocation of NEC due to the close proximity of the transverse colon with the gastric curvature. We would like to underline the importance of considering causes other than intestinal perforation in a neonate with or without NEC.

CONCLUSION

Gastrointestinal perforation in neonates is a life-threatening surgical emergency. The most common cause of perforation in neonates is necrotizing enterocolitis (NEC). Early suspicion of gastrointestinal perforation needs early resuscitation and intervention for a better outcome. Free air is always subnormal if it is still present three days post-laparotomy and should raise suspicion of another perforation.

Author Contribution: All authors share equal effort contribution towards (1) substantial contributions to conception and design, analysis and interpretation of data; (2) drafting the article and revising it critically for important intellectual content; and (3) final approval of the manuscript version to be published. Yes.

Potential Conflicts of Interest: None.

Competing Interest: None.

Sponsor Ship: None.

Acceptance Date: 7 September 2019.

Ethical Approval: Approved by the Research and Ethics Committee, King Hamad University Hospital, Bahrain.

REFERENCES

1. Prashant SP, Abhaya G, Paras LK, et al. Gastric Perforation in Two Neonates: Spontaneous? Secondary to Feeding Tube: A Case Report. *SM J Pediatr Surg* 2016; 2(1): 1009.
2. Lee D, Shim S, Cho S, et al. Comparison of Gastric and Other Bowel Perforations in Preterm Infants: A Review of 20 Years' Experience in a Single Institution. *Korean Journal of Pediatrics* 2015; 58(8): 288-293.
3. Parasuraman D, Roy R, Mitchell S. Iatrogenic Gastric Perforation in a Preterm Infant: Clinical Presentation and Radiological Indicators. *IJPN* 2006; 7(1): 1-4.
4. Cerone J, Munshi U, Clark D. Case Reports: Necrotizing Enterocolitis. *Case Reports in Clinical Pathology* 2014; 2(2): 27-33.
5. Lin P, Stoll B. Necrotising Enterocolitis. *Lancet* 2006; 368(9543): 1271-1283.
6. Wu S, Caplan M, Lin H. Necrotizing Enterocolitis: Old Problem with New Hope. *Pediatrics and Neonatology* 2012; 53(3): 158-163.
7. Patel B, Shah J. Necrotizing Enterocolitis in Very Low Birth Weight Infants: A Systemic Review. *ISRN Gastroenterol* 2012; 2012: 562594.
8. Eltayeb A, Mostafa M, Ibrahim N, et al. The Role of Surgery in Management of Necrotizing Enterocolitis. *International Journal of Surgery* 2010; 8(6): 458-461.
9. Moss R, Dimmitt R, Barnhart D, et al. Laparotomy versus Peritoneal Drainage for Necrotizing Enterocolitis and Perforation. *The New England Journal of Medicine* 2006; 354(21): 2225-2234.
10. Thakkar H, Lakhoo K. The Surgical Management of Necrotising Enterocolitis (NEC). *Early Human Development* 2016; 9725-28.
11. Rees C, Hall N, Eaton S, et al. Surgical Strategies for Necrotising Enterocolitis: A Survey of Practice in the United Kingdom. *Arch Dis Child Fetal Neonatal Ed* 2005; 90(2): F152-F155.
12. Hyginus E, Jideoffor U, Victor M, et al. Gastrointestinal Perforation in Neonates: Aetiology and Risk Factors. *Journal of Neonatal Surgery* 2013; 2(3): 30.
13. Holman R, Stoll B, Curns A, et al. Necrotising Enterocolitis Hospitalisations among Neonates in the United States. *Paediatric and Perinatal Epidemiology* 2006; 20(6): 498-506.
14. Llanos A, Moss M, Pinzón M, et al. Epidemiology of Neonatal Necrotising Enterocolitis: A Population-Based Study. *Paediatric and Perinatal Epidemiology* 2002; 16(4): 342-349.
15. Kshirsagar A, Vasisth G, Ahire M, et al. Acute Spontaneous Gastric Perforation in Neonates: A Report of Three Cases. *African Journal of Paediatric Surgery: AJPS* 2011; 8(1): 79-81.
16. Morsi A, Omar H, Osama A, et al. Clinical Spectrum of Neonates Presenting with Pneumoperitoneum: A Retrospective Study. *African Journal of Paediatric Surgery: AJPS* 2016; 13(3): 120-124.