

Case Report

Spontaneous Ileal Perforation in a Case of Neutropenic Enterocolitis Managed Laparoscopically

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Abstract

Neutropenic enterocolitis (NE) or 'typhlitis' is a serious condition. It is commonly associated with malignancy, intensive chemotherapy and severe neutropenia. The decision for surgery remains a clinical dilemma. We present a case of NE with ileal perforation that underwent laparoscopic small bowel resection and ileostomy. An 11 years old boy diagnosed with acute lymphoblastic leukemia, was receiving induction chemotherapy. He developed neutropenic fever which was treated with antibiotics. He subsequently experienced lower abdominal pain predominately at the right iliac fossa radiating to the suprapubic region. Abdomen examination revealed tenderness over the right iliac fossa with localised guarding. Chest X-ray erect showed no air under diaphragm. Ultrasound abdomen showed presence of minimal free fluid in the abdomen. A computed tomography abdomen was performed, which revealed evidence of pneumoperitoneum predominantly at the dependent areas of the abdomen. There was a focal defect seen at the anterior wall of distal ileum with connection with a fluid locule, suspicious of ileal perforation. The patient subsequently underwent an urgent laparoscopic small bowel resection and double barrel ileostomy. Intraoperatively, there was a 1 x 1 cm perforation at the distal ileum about 3 cm from ileocaecal junction with localised contamination. Postoperatively, the patient gradually recovered and was discharged. NE is a potentially life-threatening condition associated with high mortality. Contrary to belief, surgery in neutropenic patients is not deleterious, especially in complicated NE. Prompt and accurate decision-making holds an important role in improving patient outcomes.

Keywords: Acute lymphoblastic leukaemia; intestinal perforation, laparoscopy; necrotising enterocolitis, neutropenic enterocolitis; typhlitis

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Introduction

Neutropenic enterocolitis (NE), commonly known as 'typhlitis,' is a severe condition, most commonly causing inflammation and thickening of the caecum and ileum up to ascending colon (A). The incidence varies from 0.8-26% but has a high mortality rate of up to 50% (1,2). Typically, the presentation is abdominal

pain with fever and neutropenia two weeks after chemotherapy. The immunocompromised patient is at risk for various reasons, such as malignancy, intensive chemotherapy, organ transplant, acquired immunodeficiency syndrome (AIDS) and multiple others (1,3-5). Despite being rare (2%), bowel perforation is a known complication of NE, underscoring the severity of the cases (2). Though the

majority of NE can be managed conservatively, surgery can be an option in case of progressive disease or complications such as perforation of the bowel. Herein, we reported a case of NE with ileal perforation that underwent laparoscopic small bowel resection and ileostomy.

Case report

We reported a case of an 11 years old boy whom was diagnosed to have acute lymphoblastic leukaemia. He was started on induction chemotherapy which involved oral dexamethasone, intravenous (IV) vincristine 1.5 mg/m², IV daunorubicin 25 mg/m², intramuscular (IM) pegaspargase 1000 iu/m² and intrathecal methotrexate 12 mg. He subsequently developed neutropenic fever on day 16 of admission, where he had a near zero total white cell (0.19 x 10⁹/L) as well as absolute neutrophil count (0.03 x 10⁹/L). He was initially started on IV cefepime and IV amikacin, which was then escalated to IV meropenem in view of persistent temperature spikes. His temperature finally resolved after completion of 1 week of IV meropenem. On day 29 of admission, patient complained of lower abdominal pain which was acute in onset. The pain predominately occurred at the right iliac fossa and radiated to the suprapubic region. He had no fever. His vital signs were within the normal range. Abdomen examination revealed tenderness over the right iliac fossa with localised guarding. Chest X-ray erect showed no air under diaphragm. Ultrasound abdomen was arranged, where there was presence of minimal free fluid in the abdomen, however, appendix was not visualised in the scan. A computed tomography abdomen was subsequently performed, which revealed evidence of pneumoperitoneum predominantly at the dependent areas of the abdomen. There was a focal defect seen at the anterior wall of distal ileum with connection with a fluid locule (Fig. 1). There was mild bowel wall thickening over the caecum. Patient subsequently underwent an urgent laparoscopic small bowel resection and double barrel ileostomy. Intraoperatively, there was a 1 x 1 cm perforation at the distal ileum about 3 cm from ileocaecal junction with localised contamination (Fig. 2). Decision against anastomosis was made. The perforation site was brought out and fashioned as a loop ileostomy. Post operatively, the patient gradually recovered without severe complications. Histopathology results of the ileal perforation site showed acute serositis changes and was negative for malignancy. He was planned for reversal of stoma after completion of chemotherapy.

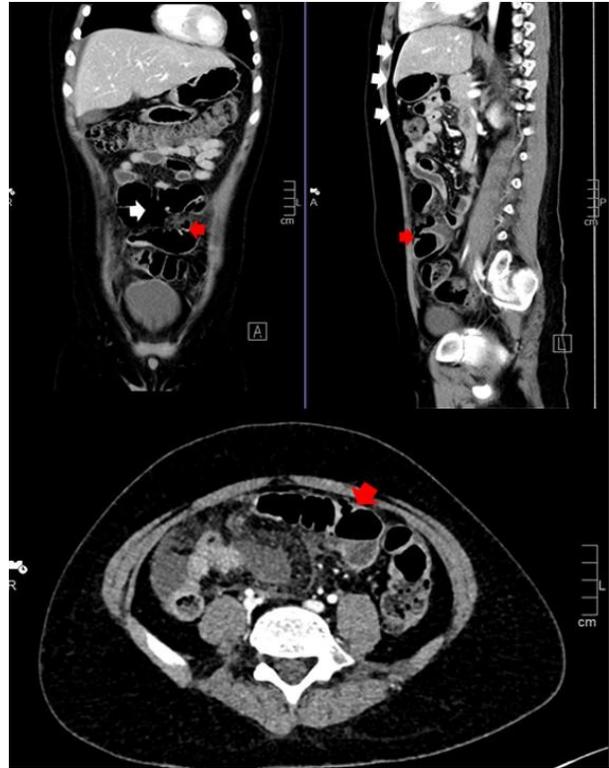


FIGURE 1: Computed tomography abdomen images of ileum perforation site (red arrow) with pneumoperitoneum (white arrow)



FIGURE 2: Laparoscopic image of perforation (black arrow) at ileum (white arrow)

Discussion

NE is a potentially life-threatening necrotising inflammation of the caecum and the colon that develops in patients with severe neutropenia (6). The term thyphlitis (when localised in the caecum), necrotising enterocolitis and ileocecal syndrome have been used interchangeably by authors, which suggests bowel involvement may not be strictly limited to only the caecum (7-9).

The true incidence of NE is unknown. Hadar et al. reported an incidence of 5% among 843 patients with 76% of the patients being treated for haematological malignancies (10). In contrast, a post-mortem review identified an incidence of 24% of NE cases for patients with leukaemia (7). This figure is based on autopsy data from deceased patients, it likely overestimates true prevalence and instead highlights the potential contribution of NE to mortality in leukaemia patients. The wide range of results may represent under detection of NE among patients. Reports on incidence of bowel perforation among NE patients are scarce. Abu-Sbeih et al. reported of only 2% of patients developed bowel perforation (11). Overall, the estimated mortality among patients with NE was reported to be 42.2% (12).

NE is hypothesised to be associated with exposure to chemotherapy. This induces mucositis and mucosal wall interruption. Colonising gut flora then enters the damaged gut wall without neutrophil clearance which causes further mucosal damage (8,9). The multiagent nature of chemotherapy regimes makes it difficult to definitively determine the contribution of a single agent or drug pair in assessing the risk of NE (6).

The decision for surgery remains a clinical dilemma. 90% of paediatric NE cases can be handled conservatively (13,14). Surgery on the other hand may be challenging due to friable bowel, adjacent inflammation, poor healing and bleeding diatheses (8-10,14). These factors may lead to a reluctance in managing these patients surgically. In contrast, a systematic review suggested that surgery was not associated with an increase in mortality. The study identified that surgery was not deleterious, regardless of underlying malignancy time prior and presence of neutropenia at the time of surgery (12). Surgical intervention is generally reserved for complicated NE, characterised by perforation, bowel necrosis, uncontrolled sepsis or failure of conservative management (12-14). The presence of pneumoperitoneum and confirmed ileal perforation in our patient warranted urgent operative management. Hence, in our case, we promptly brought the patient

for surgery once evidence of perforation was confirmed. Intraoperatively, primary repair or anastomosis was avoided in view of patient's neutropenia and ongoing chemotherapy-induced immunosuppression. Severe neutropenia is associated with impaired inflammatory response, reduced collagen deposition and delayed wound healing, thereby increased in risk of anastomosis leak (8,14). Therefore, loop ileostomy was deemed the safer option to achieve source control while minimising the risk of catastrophic anastomotic leak. This staged approach also facilitates recovery and continuation of chemotherapy before definitive reconstruction. Post operatively, the patient slowly recovered without any severe complications despite being in a neutropenic state.

Conclusion

NE is a potentially life-threatening condition associated with high mortality. Contrary of belief, surgery in neutropenic patients is not deleterious especially in complicated NE. Prompt and accurate decision making holds an important role in improving patient outcomes.

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Conflict of interest: The authors declare that they have no competing interests

Consent for publication: Written informed consent from the patient's father were obtained for the publication of this case report, any accompanying data and images.

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