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CASE REPORT

Intussusception caused by typhlitis: a rare complication in a patient with acute lymphoblastic leukemia after chemotherapy

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Abstract

Introduction Intussusception, which is common in pediatric patients but rare in adults with leukemia, usually presents with an intralumenal lesion as a lead point in adults.

Case report We herein report the case of a 38-year-old female who developed right lower quadrant abdominal pain and fever on day 16 of chemotherapy. Abdominal computed tomography showed ileocecal intussusception. The patient underwent surgery, and the definitive pathological diagnosis was typhlitis leading to intussusception. Albeit very rare in adults, typhlitis-induced intussusception should be suspected in those with leukemia presenting with abdominal pain.

Keywords Acute lymphoblastic leukemia · Intussusception · Typhlitis

Introduction

In patients with leukemia, the gastrointestinal system can undergo various pathological changes caused by the primary disease or treatments, such as the leukemic intestinal invasion, altered immune state, and toxicity associated with chemotherapy [1]. Intussusception is more common in pediatric patients but rare in adults with leukemia [2]. Few studies reported adult intussusception associated with leukemia accompanied by leukemic intestinal invasion or due to unknown etiology [3, 4]. We herein present an adult patient with acute lymphoblastic leukemia who developed intussusception as a complication of post-chemotherapy neutropenic enterocolitis, i.e., typhlitis. This rare condition should be considered during the differential diagnosis in patients with acute leukemia presenting with abdominal pain.

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Case report

A 38-year-old female was diagnosed with precursor T cell acute lymphoblastic leukemia (ALL), which was confirmed by bone marrow biopsy. Approximately 6 months before admission, she had received chemotherapy including hyper-CVAD (hyperfractionated cyclophosphamide, vincristine, doxorubicin, and prednisolone) and intrathecal methotrexate (MTX) with cytarabine (Ara-C) and had experienced two episodes of neutropenic fever during chemotherapy.

The patient was receiving CVAD-IV when she developed neutropenia on the seventh day of chemotherapy, and granulocyte colony stimulation factor was initiated on day 11 after the completion of chemotherapy. Peripheral blood absolute neutrophil count was above 500/μl since day 15. However, the patient developed intermittent, cramping abdominal pain on day 14 and fever on day 15. She received empirical treatment with cefepime 1 g every 12 h. However, the abdominal pain worsened, and the patient developed abdominal distention with anorexia. The abdominal pain migrated to the right lower quadrant and worsened on day 16. The timeline events were summarized in Fig. 1.

Physical examination revealed tenderness and muscle guarding in the right lower quadrant of the abdomen without a palpable abdominal mass. Abdominal plain X-ray did not show any specific findings. Abdominal computed tomography (CT) to determine the cause of symptom progression revealed cecal wall thickening accompanied with the ileocecal

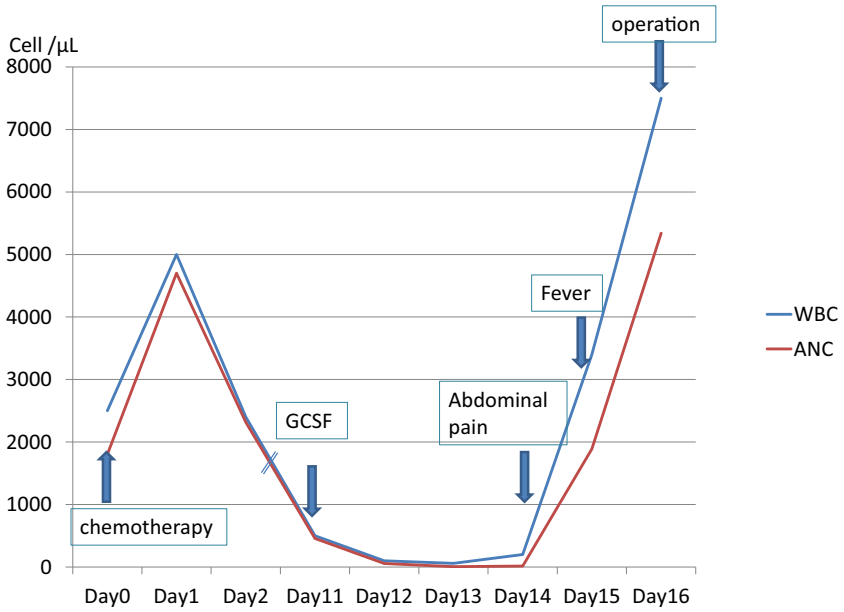

Fig. 1 The events summarized with a timeline. WBC: white blood cell count; ANC: absolute neutrophil count

valve and appendix entering the cecum, and the patient was diagnosed with ileocecal intussusception based on these findings (Fig. 2).

The patient underwent emergency surgery. During laparotomy, ileocecal intussusception with one elastic cecal mass as the leading point was observed. A plaque-like lesion, 4.0 × 2.5 cm in size, with an ulcerated surface was located near the ileocecal valve (Figs. 3 and 4). Right hemicolectomy with a dysfunction loop ileostomy was performed without surgical complications. The postoperative course was uneventful. She started oral intake on the first day after the operation. Her intestinal functions recovered, and she had adequate ileostomy output. The patient was discharged on postoperative day 14 without complications and subsequently continued chemotherapy for ALL 1 month after the surgery.

Microscopic examination revealed that the lesion was negative for TdT and CD99 by immunohistochemistry, and there was no evidence of leukemic involvement. Histological analysis revealed only edematous change in the submucosa accompanied with ulcerative mucosa and hemorrhagic necrosis, and microbiological cultures were negative. Based on the pathological features and the clinical history, the patient was diagnosed with typhlitis.

Discussion

Intussusception is defined as the prolapse of a proximal intestinal portion into the lumen of a distal segment [2] and is common in pediatric patients; intussusception is rarely observed in

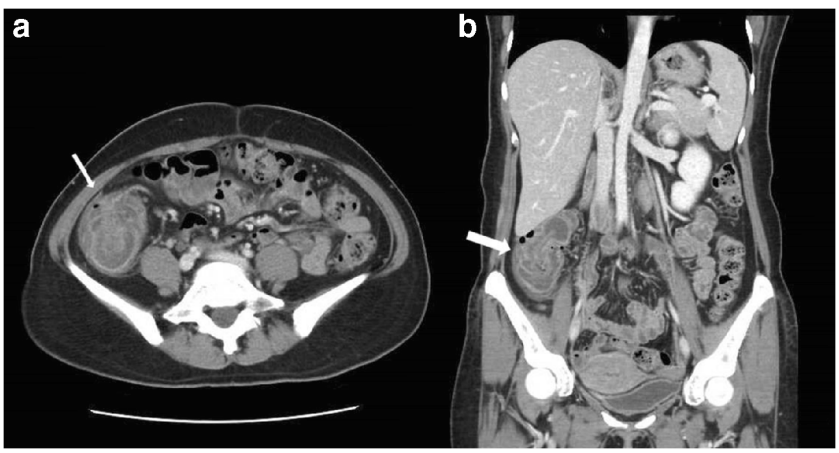

Fig. 2 **a** Axial view of the abdomen and pelvis by contrast-enhanced computed tomography showing the classic target sign (thin white arrow), which suggests intussusception. **b** Coronal reconstruction of the contrast-enhanced abdominal and pelvic computed tomography showing a segment of the ileocolic intussusception (thick white arrow)

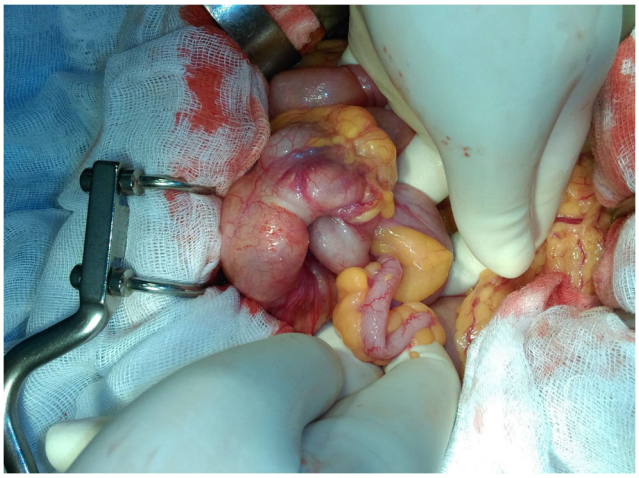

Fig. 3 Ileocolic intussusception found during laparotomy

adults, comprising approximately 5% of all intussusception cases [5]. There are few case reports for intussusception in adult leukemia patients [3, 4, 6–8]. The lead point of intussusception is usually an intraluminal lesion, which can be benign or malignant. The possible pathogenic mechanisms include leukemic infiltrates leading to a tumor mass, granulocytic sarcoma, or chloroma as the extramedullary manifestation of acute myeloid leukemia, enlarged lymph nodes, and intramural hematoma formation caused by coagulopathy [8]. Unlike pediatric cases, surgical intervention is required for adult patients with intussusception because the success rate is low with barium enema.

The diagnosis of intussusception is difficult because of the variability in the clinical presentation. The most common symptom is abdominal pain, followed by vomiting or gastrointestinal tract bleeding [9]. Plain abdominal X-ray may demonstrate features of bowel obstruction. Ultrasonography can reveal the presence of the target sign and is useful in pediatric patients; however, its diagnostic accuracy is dependent on the experience of the radiologist.

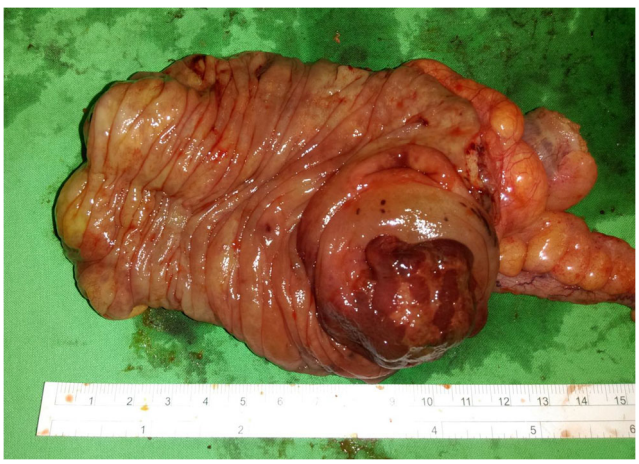

Fig. 4 The specimen of an elastic, brown-red mass near the ileocecal valve. Note the debris coating the mass

Ultrasonography may be affected by obesity or the presence of massive air in the distended bowel loops, which hinders its diagnostic performance in adults [10]. Abdominal CT remains the most useful diagnostic modality for assessing the lesion and its characteristics as well as its relationship with the surrounding tissues. Abdominal CT is also useful for distinguishing intussusception from enterocolitis.

Another important etiology of acute right lower quadrant abdominal pain in leukemia patients is typhlitis, which is characterized by necrosis involving the ascending colon, cecum, and the terminal ileum. A peripheral neutrophil count of less than 500/μl is associated with an increased risk for typhlitis [11], and approximately 2.6% of patients with acute leukemia were reported to develop typhlitis [12]. Conservative management recommended for typhlitis includes bowel rest, aggressive fluid resuscitation, broad-spectrum antibiotics, correction of thrombocytopenia, and treatment with granulocyte colony stimulation factors. Surgical indications in typhlitis are bowel perforation, uncontrolled bleeding, and abscess. However, primary anastomosis is not recommended due to impaired healing and immunosuppression [11].

The current patient had abdominal pain with fever accompanied with a neutrophil count of less than 500/μl. Despite conservative treatment, her clinical symptoms worsened. Based on the abdominal CT, surgical, and pathological findings, she was initially considered to have typhlitis. Unfortunately, the infected intestinal section could not be recovered and subsequently became the lead point for the intussusception.

This presentation is a rare complication in adult patients with leukemia. A study in a pediatric cohort reported that bacterial enteritis can be associated with intussusception [13]. Although the current patient underwent primary anastomosis, dysfunction ileostomy was also performed due to the poor immune status of the patient. The patient did not develop any postoperative complications.

This first report of ileocolic intussusception caused by typhlitis in an adult patient with acute lymphoblastic leukemia illustrates that, albeit rare in adults, the differential diagnosis of abdominal pain in patients with leukemia should include typhlitis-induced intussusception and that CT is a successful diagnostic tool. During surgical intervention, intestinal anastomosis should be considered carefully due to the impaired healing ability and immunosuppression in these patients.

Authors' contributions RH Chan and YP Chen contributed equally to the study design, collection of data, and the writing of the manuscript.

Compliance with ethical standards

Competing interests The authors declare that they have no competing interests.

Ethical approval Ethical approval was obtained and available upon request.

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