CASE REPORT

Intussusception: a rare complication in a patient with acute leukaemia after consolidation chemotherapy

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SUMMARY

Intussusception is telescoping of one segment of the gastrointestinal tract into an adjacent one. It is more common in children than adults. When it occurs in adults, it is usually associated with a lead point. Intussusception is very rare in acute leukaemia and has only been reported in few cases. We present a case of an adult woman who presented with intussusception after a cycle of consolidation chemotherapy with high-dose cytarabine for acute myeloid leukaemia. Other causes of acute abdominal pain were excluded, and the diagnosis was established by CT scan of the abdomen and barium enema. No pathological lead points were found intraoperatively. She underwent a right-sided hemicolectomy with complete recovery. To the best of our knowledge, this is only the fourth case of intussusception that has been reported in an adult patient with acute myeloid leukaemia.

BACKGROUND

Abdominal pain is not uncommon in acute leukaemia with a wide differential diagnosis. This includes many medical and surgical problems, such as neutropaenic enterocolitis, appendicitis, cholecystitis, megacolon, pseudomembranous colitis and haemorrhagic colitis.¹ However, intussusception has rarely been reported in adult patients with acute leukaemia; its occurrence has been limited to few case reports mainly involving children.^{2–7} There are only three reported cases of intussusception in adult patients with acute myeloid leukaemia (AML).^{3–4–7} In this report, we present a case of intussusception in a patient who has been receiving consolidation chemotherapy for AML. We have reviewed the literature and attempted to highlight the pathogenic mechanisms responsible for occurrence of intussusception in patients with leukaemia.

CASE PRESENTATION

A 55-year-old woman with a medical history of AML presented to our institution with fever, nausea, vomiting, abdominal pain and an episode of bloody diarrhoea on day 13 of the third cycle of consolidation chemotherapy with high-dose cytarabine.

She was diagnosed with AML with minimal maturation (WHO classification M1) 4 months prior to presentation. A bone marrow biopsy showed total replacement by myeloblasts. Induction

chemotherapy with cytarabine and idarubicin (7+3 regimen) was given. However, the follow-up bone marrow biopsy showed non-remission with reduction of blasts to 15%. Therefore, reinduction chemotherapy with cytarabine and idarubicin (5+2 regimen) was attempted. She subsequently achieved complete remission. Cytogenetics was normal with a negative FLT-3 mutation. Therefore, she was scheduled for consolidation therapy with high-dose cytarabine (3 g/m²) given twice daily on alternative days for six doses. Her first two cycles of consolidation chemotherapy were uneventful except for an episode of febrile neutropaenia after the second cycle; in which she was treated with antibiotics and antifungals. She was discharged on oral voriconazole and ciprofloxacin. After recovery of the bone marrow, she was given the third cycle of consolidation chemotherapy.

On day 13 of the cycle, she presented with the above described symptoms. Her abdominal pain was located in the lower abdomen, constant, sharp in nature and without radiation. Her abdominal examination was unremarkable, and the other findings from the physical examination did not reveal a focus of infection.

Initial laboratory workup showed an absolute neutrophil count (ANC) of 175 cells/μL (normal >1500 cells/μL), haemoglobin of 7.7 g/dL and platelet count of 16 000/cmm. Routine chemistry values were within normal ranges. Chest radiography showed only minimal vascular congestion. Urine analysis was remarkable for +4 haematuria, and stool analysis showed 2–3 red blood cells/hpf and 10–15 leukocytes/hpf. Blood, stool and urine cultures were sent for analysis, and the patient was started on intravenous piperacillin-tazobactam and vancomycin for febrile neutropaenia. Her voriconazole was continued, and granulocyte colony-stimulating factor was given at a dose of 5 μg/kg once daily.

Seventy-two hours after admission, she still had many fever spikes, and her diarrhoea and abdominal pain persisted. Her examination was remarkable for right lower quadrant tenderness with rebound tenderness. A preliminary diagnosis of neutropaenic enterocolitis was made. Therefore, metronidazole was added. CT scan of the abdomen and pelvis was performed, which indicated ileocolic intussusception (figure 1). No evidence of acute appendicitis or cholecystitis or signs of bowel perforation were noted.

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Figure 1 CT scan of the abdomen, (A) sagittal section and (B) coronal section showing ileocolic intussusception (red arrow).

TREATMENT

A barium enema was performed, which confirmed the diagnosis of intussusception but failed to reduce it (figure 2). She was referred to surgery where laparoscopic right hemicolectomy was performed. No lead point for the intussusception was seen on abdominal imaging or by careful intraoperative exploration. Pathological examination of the resected tissue was also non-revealing.

OUTCOME AND FOLLOW-UP

Postoperatively, she started to improve symptomatically and remained afebrile. Her counts started to progressively increase and all cultures were still negative. She was discharged on the fifth postoperative day.

DISCUSSION

Intussusception is a process in which there is prolapse of a proximal bowel segment into a distal one.⁸ Adult intussusception is a rare occurrence since ~95% of the cases occur in children.^{9 10} Among all cases of bowel obstruction occurring in adults, intussusception is responsible for 1–5% of all cases.^{8 10 11}

Figure 2 Barium enema showing ileocolic intussusception.

The pathogenesis that leads to invagination is poorly understood, but generally in 90% of adult cases, there is an intraluminal or rarely an extraluminal lesion.¹¹ This is commonly called the 'lead point' and can be either a benign or malignant lesion. Benign lead points include polyps, and Meckel's and colonic diverticula.⁸ Malignant lead points include but are not limited to carcinomas, lymphomas, sarcomas and metastatic tumours such as melanoma.^{8 12–14} In addition, a large study in a paediatric population showed that bacterial enteritis can be associated with intussusception.¹⁵

There are multiple pathogenic mechanisms that can potentially explain the occurrence of intussusception in patients with leukaemia. Leukaemic infiltrates leading to a tumour mass is the first mechanism.⁴ Granulocytic sarcoma (GS) or chloroma is the extramedullary manifestation of AML. It rarely involves the gastrointestinal (GI) tract.¹⁶ The submucosal infiltration by immature myeloid precursors can lead to a tumour mass and work essentially as a lead point that can lead to intussusception.^{3 17–19} GS usually occurs in association with AML but can rarely occur as an isolated tumour mass without a leukaemic phase which is termed non-leukaemic GS.¹⁹ This might be misleading as GS can be confused with other types of malignancies.^{3 16 19 20} In a mechanism similar to chloroma, chronic lymphocytic leukaemia can cause intussusception by mucosal leukaemic invasion leading to a tumour mass.^{21–23} Enlarged lymph nodes can also act as a lead point, particularly in patients with acute lymphoblastic leukaemia (ALL).^{2 24} Acute leukaemia and subsequent treatment can lead to thrombocytopenia and coagulopathy. This can lead to intramural extravasation of blood with haematoma formation, which can act as lead point for intussusception.⁵ Another postulated mechanism is chemotherapy toxicity. Vincristine-induced ileus was suggested as a potential cause of intussusception in a child with ALL.²⁴ In addition to vincristine, Shah *et al*²⁵ suggested that high doses of methotrexate might be related to intussusception occurrence in two children with ALL.

Preoperative diagnosis of intussusception is very challenging and difficult due to the variability in the clinical presentation.⁸ Adults usually present with abdominal pain or features of intestinal obstruction, which sometimes includes rectal bleeding.⁴ Neutropaenic enterocolitis is an important differential diagnosis of intussusception. The two are difficult to differentiate and may coexist.²⁴ CT scan is the preferred modality of investigation and often demonstrates the pathological lead point and the intussusception.²⁶ Accurate diagnosis is very essential because intussusception warrants immediate surgical attention.⁸

Finally, cytarabine has been associated with multiple GI complications, including oral and anal inflammation or ulceration, nausea, vomiting, and diarrhoea. Bowel necrosis, necrotising enterocolitis and pneumatosis cystoides intestinalis leading to peritonitis have also been described.^{27 28} Jones and Abramson²⁹ studied 14 patients with acute leukaemia who developed acute abdominal complications, including an acute surgical abdomen, diarrhoea, GI bleeding and a paralytic ileus. Cytarabine was administered to 13 of the 14 patients.²⁹ Camera *et al*³⁰ also studied the occurrence of GI complications in 169 adult patients with AML after induction chemotherapy with cytarabine-based regimens. GI complications occurred in 41% of the patients, and neutropaenic enterocolitis was diagnosed in 5.9% of them.³⁰ Our patient received a high dose of cytarabine, and we do not have an explanation for her intussusception. There is no known association between high doses of cytarabine and intussusception. We postulate that cytarabine might alter bowel motility, which may lead to intussusception. However, this cannot be proven due to the rarity of intussusception in acute

leukaemia and the presence of other explanations in some of the reported cases.

To the best of our knowledge, this is only the fourth case of intussusception that has been reported in an adult with AML.^{3 4 7} In our case, whether cytarabine is responsible or not is unknown. Probably, the underlying disease or chemotherapy altered small bowel mobility. This served as the possible aetiology for ileal prolapse through the relatively stiffer ileocecal valve. However, intussusception should be considered in the differential diagnosis of a patient with acute leukaemia who presents with abdominal pain whether the patient is on chemotherapy or not.

Learning points

- Intussusception is a rare cause of acute abdomen in adults with acute myeloid leukaemia
- CT scan of the abdomen is very essential in patients with acute leukaemia who present with neutropaenic fever and acute abdomen.
- High-dose cytarabine can be associated with various gastrointestinal complications. Some of them are severe enough to warrant surgical intervention.

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