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Recurrent lymphoma presenting as painless, chronic intussusception: A case report

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Abstract

BACKGROUND

The clinical presentation of acute lymphoblastic lymphoma is highly varied. While prognosis is good, recurrence of disease can occur. Gastrointestinal relapse, including intussusception, is well-described but the absence of abdominal pain in this setting is rare.

CASE SUMMARY

We report a 13-year-old male with B-cell precursor acute lymphoblastic leukemia in remission presenting with anemia and weight loss. Examination was significant for absence of abdominal pain, but a stool sample was positive for occult blood. Pan-endoscopy was performed with colonoscopy revealing a mass filling the colonic lumen. Biopsy of the mass confirmed recurrence of recurrent B-cell lymphoma. Computed tomography scan revealed ileocolic intussusception resulting from the tumor. This case is unusual in that the patient had no abdominal pain despite the presence of intussusception.

CONCLUSION

While intestinal involvement with lymphoma has been well described in the literature, presentation as painless intussusception has not been reported. This case report highlights the wide spectrum of clinical manifestations of recurrent B-cell lymphoma involving the gastrointestinal tract, in particular the near absence of symptoms despite the finding of intussusception.

Key words: B-cell acute lymphoblastic leukemia; Tumor relapse; Intussusception; Case report; Anemia

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Core tip: Acute lymphoblastic leukemia and recurrent B-cell lymphoma are known to present in the gastrointestinal tract, most commonly with abdominal pain, either alone or in association with other symptoms, including rectal bleeding, vomiting, and/or abdominal distention. A common presentation is that of intussusception. We report a patient who was found to have recurrent B-cell lymphoma leading to intussusception during colonoscopy being performed for evaluation of anemia. There were no other clinical features, including abdominal pain, to suggest intussusception prior to colonoscopy. This case should serve as a cautionary tale that serious complications resulting from acute lymphoblastic leukemia may occur despite minimal clinical symptoms.

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INTRODUCTION

Gastrointestinal (GI) involvement by hematologic cancers (leukemia and lymphoma) related to malignant infiltration of the intestinal mucosal-associated lymphoid tissue is well documented. GI complications arising from these cancers include typhlitis, intestinal perforation, hemorrhage, and intussusception. Intussusception has been described both as the initial presentation of the cancer or as a later complication, almost universally presenting with abdominal pain due to bowel obstruction as a major symptom. Various diagnostic imaging studies have been used to diagnose intussusception, however, on rare occasions unsuspected intussusception has been made colonoscopy. Surgical resection or biopsy are the principle modes of documenting lymphoma as the underlying cause of intussusception when present. We report an adolescent with a history of acute lymphoblastic leukemia (ALL) who presented with anemia and was found at colonoscopy to have intussusception due to recurrent lymphoma.

CASE PRESENTATION

Chief complaint

A 13-year-old white male presented with iron-deficiency anemia and weight loss.

History of present illness

The patient had a past medical history of B-cell precursor ALL at 2-years of age. He achieved remission in 28 mo after completing standard risk chemotherapy treatment for ALL with doxorubicin and cyclophosphamide. At 7-years of age he developed cough, shortness of breath, and tiring easily. Echocardiogram was normal. Three months later he presented to the pediatric emergency room due to worsening pulmonary symptoms; a chest-x-ray showed hilar adenopathy and right-sided pleural effusion. Echocardiogram showed a large mass in the right atrium with a pericardial effusion. Computed tomography (CT) scan of the neck, chest, abdomen and pelvis with contrast revealed a 4.5 cm right sided mediastinal mass extending into the right atrium causing obstruction of the superior vena cava, right main pulmonary artery, and distal airway compression with paratracheal and mediastinal adenopathy. Flow cytometry was non-diagnostic for malignancy and bone marrow aspirate showed no evidence of infiltrative disease. Cardiac catheterization and biopsy of the tumor was initially interpreted as inflammatory myofibroblastic tumor, and chemotherapy was initiated with ifosfamide, doxorubicin, and celecoxib which achieved a decrease in tumor size. Following three courses of scheduled chemotherapy, cardiovascular surgery performed tumor resection. Final pathology was consistent with necrotic lymphoma consistent with recurrent B-cell lymphoma. He completed chemotherapy and began maintenance therapy. Eighteen months later he presented with weight loss and anemia.

Approximately 18-mo later, at 13 years of age, he was seen in pediatric gastroenterology clinic for iron-deficiency anemia, decreased oral intake, and weight loss of 5.7 kg. He did not have fatigue, malaise, shortness of breath, palpitations, melena, hematochezia, or abdominal pain.

Personal and family history

The patient was a student in good standing. Family history was negative for inflammatory bowel disease, polyp syndromes, and bleeding dyscrasias.

Physical examination

On physical examination he had no tachycardia or cardiac murmur, his lungs were clear to auscultation, his abdomen was soft, non-tender, without hepatosplenomegaly, there was no bruising, digital rectal exam revealed brown stool that was positive for fecal occult blood.

Laboratory examinations

Complete blood count revealed a normal white blood cell count ($9200/\text{mm}^3$), microcytic anemia (hemoglobin = 9.0 g/dL, hematocrit = 30.9%, mean corpuscular volume = 61 fL), and thrombocytosis (platelets = $726000/\text{mm}^3$). Other laboratory values were normal including a liver panel, renal panel, lactate dehydrogenase and uric acid.

Imaging examinations

The patient underwent esophagogastroduodenoscopy (EGD) and colonoscopy. EGD results were unremarkable; however, a large, firm mass in the mid-ascending colon was discovered on colonoscopy and subsequently biopsied (Figure 1). The biopsy confirmed recurrent B-cell ALL. CT scan showed ileo-colonic intussusception (Figure 2) with the mass acting as the lead points. Pathology results showed a recurrent B-cell lymphoma in the ileum causing chronic intussusception.

FINAL DIAGNOSES

The final diagnosis was ileocolic intussusception due to intestinal relapse of B-cell ALL.

TREATMENT

The patient was admitted and started on chemotherapy including intrathecal methotrexate, cytarabine, and clofarabine.

OUTCOME AND FOLLOW-UP

His hospital course was complicated by febrile neutropenia, thrombocytopenia, anemia, and clostridium difficile infection. The patient was minimally responsive to chemotherapy, and palliative right hemicolectomy with primary anastomosis was performed 3 mo after diagnosis. This procedure was complicated by anastomotic leak, so the patient underwent bowel resection with ileostomy. The patient and family initially planned to proceed with bone marrow transplant. However, the patient no longer desired to pursue chemotherapy or transplant and was transferred to hospice care where he passed away 5 mo after relapse.

DISCUSSION

We report our patient to alert physicians to the subtlety to which intussusception can present, as well as to highlight the rare presentation and unusual diagnostic method utilized. Our patient was in remission and developed anemia, but was otherwise asymptomatic. He was found to have occult blood in his bowel movements. He underwent EGD and colonoscopy as part of the investigation and was found to have a luminal mass, biopsy proved to be a lymphoma. Colonoscopy as a modality for the diagnosis of intestinal lymphoma has only been reported once to date^[1]. After making the diagnosis of relapsed lymphoma in the small intestine as the cause of intussusception, we performed a literature search and found three large retrospective reviews and a number of case reports/series describing intussusception related to



Figure 1 Findings at colonoscopy. A large, friable, lobulated mass was identified in the mid-ascending colon.

hematologic cancers in children. We review and report on the clinical features of intussusception related to hematologic cancers.

Intussusception resulting from GI involvement in hematologic cancers (leukemia and lymphoma) is a well-described, yet occurs rarely. In a retrospective review of 378 children identified with intussusception over 31-years, only 1.6% were found to have lymphosarcoma as a lead point^[2]. In two large retrospective reviews of children with Burkitt's lymphoma, the prevalence of developing intussusception was 6.7% in one study and 17.5% in the other^[3,4]. In a smaller study of children with primary abdominal non-Hodgkin's lymphoma, 25% developed intussusception^[5]. Intussusception has been reported less frequently in children with leukemia. In a study of 364 Taiwanese children with leukemia only 1 (0.3%) developed intussusception^[6]. In a study from the United Kingdom no intussusception was found in 800 children^[7].

Combining the data from the retrospective reviews and case reports (Table 1) there were 74 patients with intussusception and a hematologic malignancy for review^[8-22]. The majority of patients were male (80.3%). The average age (8.0 ± 3.8 years) and median age (7.5 years) were higher than the typical age (< 2 years) of children with intussusception. Only two children were less than two years of age. The type of cancer was more commonly lymphoma (85.3%) than leukemia (14.3%), however, the data may be skewed as two large retrospective studies were done to assess for intussusception in children with lymphoma^[3,4].

Intussusception as the initial presentation of hematologic cancer occurred in 47.6% of the patients (Table 2). Intussusception was seen as a complication of known cancer in the remaining patients; occurring more commonly during maintenance (19.0%) or at relapse of disease (19.0%), then during induction (14.3%). Where documented, most patients (89.7%) had an identifiable lead point; the most common lead point was the cancer (80.8%), followed by mucosal necrosis (7.7%), typhlitis (3.8%), hematoma (3.8%), and lymph node (3.8%). The majority of intussusceptions were ileo-colic (86.7%), followed by ileo-ileal (9.3%), ileo-ileo-colic (2.7%) and colo-colic (1.3%). Intussusception was acute in 60% of children and chronic in 40%.

All of the reported patients presented with abdominal pain and various combinations of abdominal distension, vomiting, and hematochezia (Table 1). The patient described herein was asymptomatic regarding GI complaints. He underwent colonoscopy as part of the evaluation for anemia and was found to have a large mass filling the colon lumen; biopsy of the mass showed recurrent lymphoma.

A number of children had a contrast enema performed in an attempt to reduce intussusception, the majority (85.7%) were not successful resulting in the need for surgery (Table 2). Overall, surgical intervention occurred in the vast majority (95.7%) of the children. A tissue diagnosis was most often obtained by surgical biopsy (77.8%); diagnosis was made by colonoscopic biopsies on 2 occasions (11.1%), by PET scan (5.6%) and at autopsy (5.6%) once each.

CONCLUSION

We report a young man with ALL relapse presenting with asymptomatic intussusception with the diagnosis being made with colonoscopic biopsies. This case should serve as a cautionary tale that serious complications resulting from ALL may occur despite minimal clinical symptoms.

Table 1 Characteristics of patients presenting with intussusception due to hematological malignancy

Characteristics	Value
Gender	Male 80.3%
	Female 19.7%
Age	Average 8.0 ± 3.8 yr
	Median 7.5 yr
Type of malignancy	Lymphoma 85.3%
	Leukemia 14.7%
Presenting symptom(s)	Anemia 6.3%
	Abdominal pain 25.0%
	Abdominal pain, distension 31.3%
	Abdominal pain, vomiting 12.5%
	Abdominal pain, hematochezia 12.5%
	Abdominal pain, vomiting, hematochezia 12.5%

Table 2 Characteristics of the intussusceptions

Characteristics	Percentage
Timing of intussusception in relation to malignancy	At diagnosis 47.6%
	Induction 14.3%
	Maintenance 19.0%
	Relapse 19.0%
Location of intussusception	Ileo-colic 86.7%
	Ileo-ileal 9.3%
	Ileo-ileo-colic 2.7%
	Colo-colic 1.3%
Chronicity of intussusception	Acute 40.0%
	Chronic 60.0%
Presence of a lead point	Yes 89.7%
	No 10.3%
Cause of lead point	Malignancy 80.8%
	Necrosis 7.7%
	Hematoma 3.8%
	Lymph node 3.8%
	Typhlitis 3.8%
Barium enema reduction	Successful 14.3%
	Unsuccessful 85.7%
Surgical intervention required	Yes 95.7%
	No 4.3%
Diagnostic mode for cancer	Surgery 77.8%
	Colonoscopy 11.1%
	Imaging 5.6%
	Autopsy 5.6%

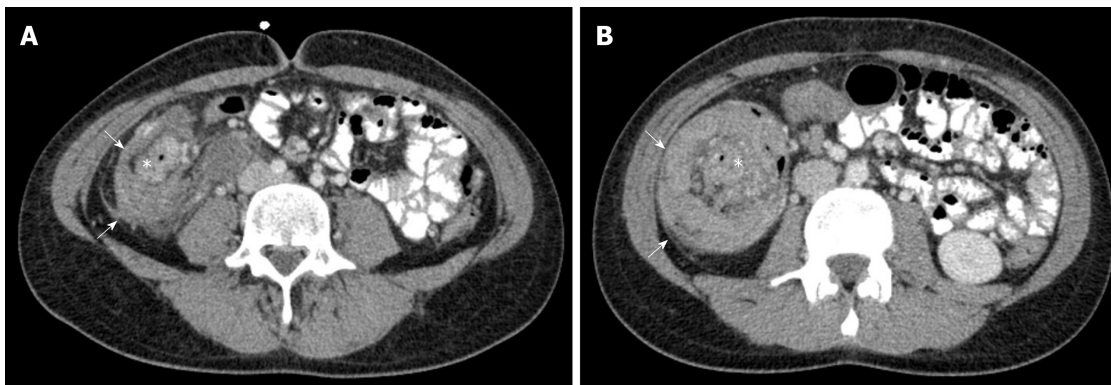


Figure 2 Abdominal and pelvic computed tomography scan. A, B: The computed tomography scan showed an ileocolic intussusception (arrows) with an intraluminal mass lesion (*) acting as a lead point in the right colon.

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