

# Two Positions of Jejunio-jejunal Intussusception in Child with Acute Lymphoblastic Leukemia – A Case Report

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**Abstract:** Acute leukemia is the most common malignant disease in childhood accounting for one third of the cancers among children. Intussusception has been described as a complication of childhood acute lymphoblastic leukemia (ALL). We report here a one year and 11 months old girl with acute lymphoblastic leukemia who developed double jejuno-jejunal intussusception during induction chemotherapy. Intussusception should be considered in any pediatric patient with ALL and acute abdomen.

**Keywords:** intussusception, ALL, chemotherapy, jejuno-jejunal, vincristine.

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## 1. INTRODUCTION

Acute leukemia is the most common malignant disease in childhood accounting for one third of the cancers among children [1]. Gastrointestinal complications are common in patients with acute leukemia. They can be due to an altered immune state, the toxicity of chemotherapy, or leukemic invasion of the bowel [2]. Gastrointestinal lesions have been reported to occur in 20- 25% of patients with leukemia [3]. Intussusception has been described as a complication of childhood acute lymphoblastic leukemia (ALL) [4-11]. Intussusception is the “telescoping of a proximal segment of the gastrointestinal tract within the lumen of the adjacent segment”[11,p 407]. The aim of this article is to increase the awareness of intussusception in acute leukemia patients, to better diagnosis and management of this condition and avoid unnecessary interventions.

## 2. CASE REPORT

A 1-year-11-month old girl was brought to our hospital with a history of fever for one-week duration. Parents also noticed increasing pallor and bruising since one-week for which they did not seek any medical advice. On examination, she had marked pallor, small lymph nodes measuring less than 0.5 cm were palpable over cervical, axillary and inguinal region on both sides. Abdomen was protuberant. Liver (2 cm) and spleen (3 cm) were enlarged. Other systems were within normal limits. The haemoglobin level was 7.4 g/dl, total leukocyte count was 102000 cells/mm<sup>3</sup>, differential showed lymphocytosis. Platelet count was 29000 cells/mm<sup>3</sup>. The blood film showed lymphocytosis with blast. The chest X-ray was normal. Bone marrow aspiration and biopsy confirmed diagnose B-cell ALL. Liver function tests and renal function tests were within normal limits. A diagnosis of high risk ALL was made and the patient was commenced on chemotherapy on 29 of June 2016, (UKALL X ALL protocol) with planned weekly vincristine (1.5 mg/m<sup>2</sup>), 4 weeks of steroids (dexamethasone 6mg/m<sup>2</sup>), weekly daunorubicin (25 mg/m<sup>2</sup>) at the beginning of therapy and one injections of peg-asparaginase (2500 IU/m<sup>2</sup>) starting on day 3. At day 15, bone marrow showed M1(blast less than 5%). On commencing chemotherapy, she had some nausea, vomiting and fever, but quickly recovered. She remained well until day 27 of induction, when following her forth injection of vincristine, she started to complain of central, colicky abdominal pain.

The episodes of pain initially reduced in frequency and intensity but on day 29, her abdominal pain worsened and she developed a fever and abdominal distention. During this time, she continued to pass stools. Abdominal CT with contrast showed two areas of jejuno-jejunal intussusception with the length of intussusception measuring 1.4cm and 2.5 cm respectively. There was mild proximal bowel dilatation, measuring 2.3 cm in diameter. (Figure1-3).

The patient was referred to paediatrics surgeon were decided to treat conservatively, with antibiotics, nasogastric tube decompression with daily follow up for vomiting and abdominal girth. After 7 days, patient was clinically improved, and CT abdomen showed resolved of intussusception. Chemotherapy Augmented consolidation was started 7 days from the last CT. The patient tolerated chemotherapy well, and only one dose of vincristine was reduced to  $1\text{mg/m}^2$ , then she was tolerated full dose.

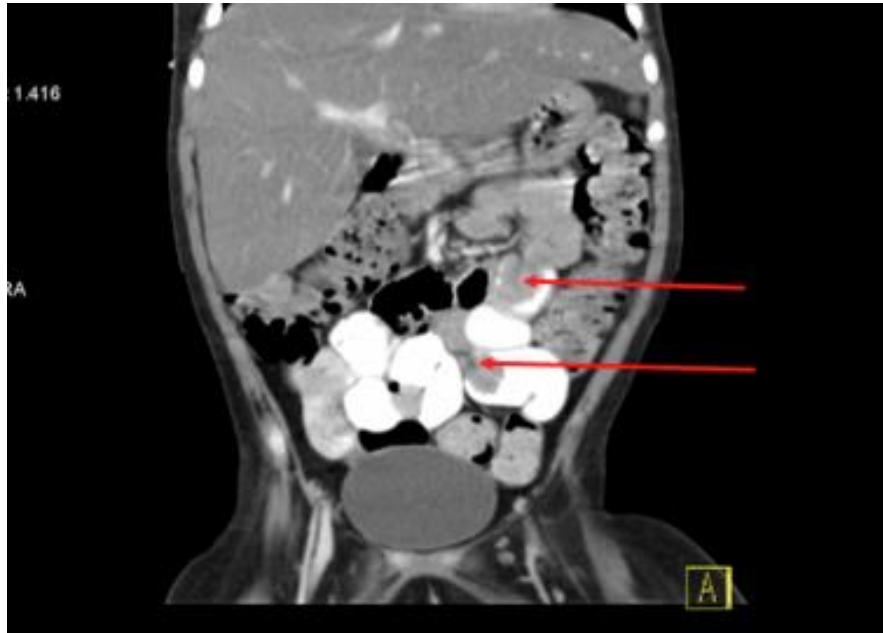


Fig.1.: Abdominal CT coronal view shows two position of jejuno-jejunal intussusception.

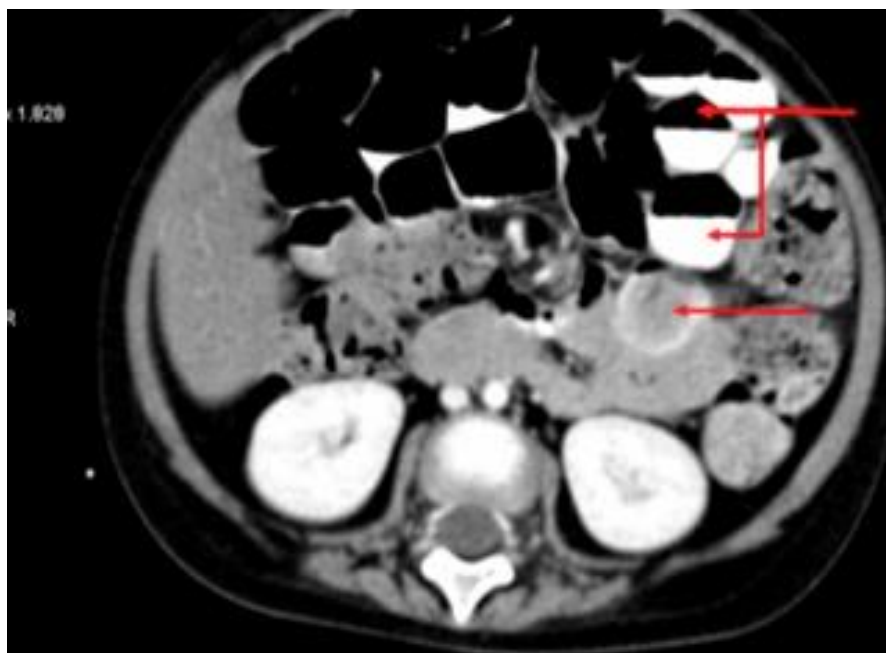
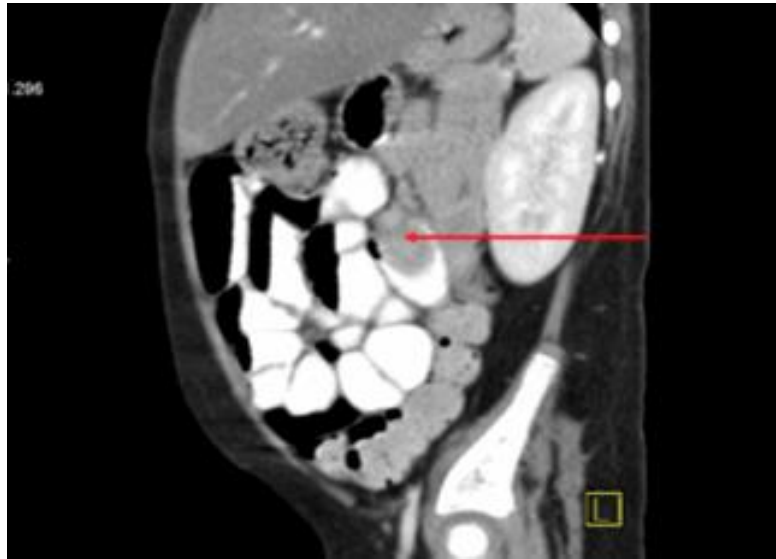


Fig.2.: Abdominal CT axial view shows intussusception with proximal bowel dilatation and air fluid level.



**Fig.3.: Abdominal CT sagittal view shows intussusception**

### 3. DISCUSSION

The gastrointestinal tract was reported to be infiltrated with leukemia cells in 13-25% of patients with ALL at autopsy [12]. Intussusception may occur at sites of benign or malignant lesions, or may be idiopathic [13]. A metastatic intestinal mass may act as the leading point of the intussusception [9]. Leukemia infiltrates which can become necrotic following chemotherapy; such lesions may be responsible for intestinal obstruction and intussusception [14]. Enlarged lymph nodes may also provide a leading point for intussusception in particular the lymphoblastic type of acute leukemia [9]. An intramural hematoma can be the leading point of intussusception especially leukemic patient with thrombocytopenia [16]. Chemotherapy may directly produce necrosis and weakened areas in the bowel wall while destroying the underlying malignant cells [14]. Typhlitis, as a cause of intussusception, has been suggested by other authors [4]. Other causes of intussusception as pneumatosis [17], Henoch Schonlein purpura petechia [18] should be ruled out. Intussusception in children with ALL is infrequently reported as complication of either the disease itself or the chemotherapy [6]. The first cases of intussusception in patients with ALL were reported by Hoffman et al. in 1905 and Sinclair et al. in 1920 [19]. A retrospective analysis of 364 patients with leukemia, treated at the National Taiwan University Hospital, found that 11 patients (3%) developed acute abdominal complications, including intussusception [7]. Exelby et al. reported in a study of 286 children with acute leukemia, gastrointestinal complications in 16 patients (5.6%), of whom only 1 (0.3%) had intussusception [20]. Micallef-Eynaud et al. study of 800 patients entered into the UK ALL VIII trial, there were no reports of intussusceptions [4]. Dudoon et al. found this complication in only 3 of their patients with ALL over a 17-year study period [9].

**TABLE 1: shows the features of previously reported cases of intussusception in acute leukemia in the literature and our present case.**

Author	Year	Diagnosis	Age/Gender	Treatment stage	Anatomical location of intussusception	Management
Thompson et al [21]	1930	Presumptive diagnosis of ALL	4 year old Female	Initial presentation	Enteric (Jejunojejunum)	Failure of soap enema reduction. Laparotomy and resection with side to side anastomosis
Feldman et al [16]	1963	ALL	5 year old Female	On 6-mercaptopurine, folic acid antagonist (methotrexate)	Four areas of intussuscepted ileum	Gastric suction, antibiotic therapy, supportive measures. Patient died.

				and intermittent steroids		
Dudgeon et al [9]	1972	ALL	7 year old Male	Rotational courses of prednisone, methotrexate, mercaptopurine, cyclophosphamide, vincristine sulfate.	Ileoileal	No treatment (condition diagnosed at autopsy)
Dudgeon et al [9]	1972	ALL	4 year old Male	Mercaptopurine and prednisone for six weeks followed by daunorubicin	Ileoileal	At laparotomy, necrotic ileum was resected. A primary anastomosis and an appendicostomy was performed.
Dudgeon et al [9]	1972	ALL	14 year old Male	Vincristine, prednisone, daunorubicin, cyclophosphamide, mercaptopurine, cytarabine, asparaginase, methotrexate	Colocolic	Contrast study of the colon demonstrated an intussusception in the mid descending colon. This was reduced to the ileocecal valve. But not to the ileum. At Laparotomy necrotic ileum and cecum were resected primary ileocolic anastomosis, and appendectomy were performed.
Karakoussis et al [8]	1974	AML	11 year old Male	Not reported	Ileocolic	Supportive treatment (nasogastric decompression).
Karakoussis et al [8]	1974	ALL	4 year old Female	Not reported	Ileoileal	Chemotherapy
Micallef et al [4]	1990	ALL	7 year old Female	Day 13 of Induction chemotherapy	Caeco-colic	Failure of water-soluble contrast enema reduction. Operative reduction was done.
Seckl et al [38]	1991	ALL	13 year old Female	Vincristine, daunorubicin, prednisolone, L asparaginase, intrathecal methotrexate and cranial irradiation (the therapy was discontinued 10	Double intussusception involving the ileo-ileal and ileo-caecal regions	Surgical excision of the involved bowel and a 9 cm segment of ileum, a small mesenteric draining lymph node and the appendix

				monthes before the presentation)		
Gavan et al [22]	1994	ALL	3 year old Male	Day 11 of Induction chemotherapy	Ileocolic	Failed air enema redaction. Operative redaction.
Gavan et al [22]	1994	ALL	9 year old Female	Day 8 of Induction chemotherapy	caeco-colic	Failure of water-soluble enema reduction. Operative reduction was done.
Kumari et al [5]	1997	ALL	8 month old Female baby	Initial presentation of ALL	Ileoileocolic	Failed barium enema reduction followed by operative reduction.
Manglani et al [6]	1998	ALL	7 month old Male baby	Day 4 of Induction chemotherapy	Ileocecal	Failed contrast enema reduction. Leading point resected and temporary ileostomy performed.
Arestis et al [23]	2005	ALL	7 year old Female	Induction day 8	Not reported	Failed contrast enema reduction followed by operative reduction.
Arestis et al [23]	2005	ALL	7 year old Female	Delayed intensification dexamethasone, methotrexate, doxorubicin and vincristine, asparaginase (8 months post diagnosis)	Ileocolic	Failed contrast enema reduction. Right hemicolectomy for irreducible intussusceptions.
Shah et al [24]	2006	ALL	8 year old Female	Week 9 of consolidation chemotherapy	Caeco-colic	Successfully reduced with a barium enema
Shah et al [24]	2006	ALL	15 year old Male	At week 14 of consolidation chemotherapy	Ileocolic mild wall thickening of the cecum	Barium enema resulted in successful reduction of the intussusception
Akahane et al [25]	2007	ALL	14-year-old Male	After the third administration of vincristine (VCR)	Jejunojejunal	Induction chemotherapy was immediately discontinued, and he was kept fasted under intravenous hyperalimentation and administration of antibiotics, H2-blocker, and gabexate mesilate. Surgical treatment was not performed

						(supportive treatment)
M.S. Shalaby et al [26]	2014	ALL	2 year old Male	A Four-drug induction regime of intrathecal methotrexate, vincristine, asparaginase and dexamethasone. Three days prior to completion of induction chemotherapy	Left sided colocolic	Operative redaction
Present case	2016	ALL	1 year and 11 months old Female	Day 27 of induction chemotherapy	Two areas of jejunojejunal	Supportive management

Overall, 95% of intussusceptions in children are ileocolic [27], double intussusception is extremely rare. Only few paediatric cases [28-34,38] have been reported. Causative factor for intussusception could be linked to the intensity of a four drug induction[4].

Some chemotherapeutic agents produce a direct neurotoxic effect on the gut, especially vincristine [35, 36]. The majority of cases occurred during induction chemotherapy as in our patient. We suspected the double intussusception in our case to be related to chemotherapy or leukemia infiltration as no bleeding, haematoma or lymph node involvement were reported in the CT of the abdomen. 80-90% of idiopathic intussusceptions are successfully reduced by enema [37]. Indications for surgery in leukaemic patients are the same as in non-leukaemics [14]. Early diagnosis and treatment are essential for survival in the leukemia-related gastrointestinal diseases [14]. We treated our patient with conservative management, we kept patient NPO with IV hydration, antibiotic, blood and platelet transfusion and a nasogastric tube was inserted. After one week patient returned to normal and chemotherapy resumed again with one dose of vincristine reduced.

#### 4. CONCLUSION

Intussusception should be suspected in any paediatric patient with ALL who has symptoms suggestive of this diagnosis, particularly during the induction phase of chemotherapy. Double intussusception is infrequent in children with acute lymphoblastic leukemia, especially jejuno-jejunal.

#### *Consent*

Written informed consent was obtained from the patient for publication of this case report and accompanying images. Consent is available upon request.

#### *Conflicts of interest*

None

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