

Unsuspecting Lead Points Causing Nonobstructing Intussusceptions in Pediatric Patients Presenting with Chronic Pain Abdomen

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Abstract

Keywords

► pediatric

nonobstructing intussusception

pathologic lead point

Intussusception is one of the common causes of bowel obstruction in pediatric age group and is of idiopathic variety in most of the cases. Nonobstructing intussusception presenting with nonspecific symptoms (or chronic pain abdomen) is rare and usually clinically misdiagnosed/difficult to diagnose. The authors present a pictorial review of 10 cases of pediatric nonobstructing intussusception presenting with acute/chronic abdominal pain which were not suspected clinically but diagnosed with a pathologic lead point radiologically.

Introduction

Intussusception is one of the most common surgical emergencies encountered in infancy and early childhood.^{1,2} It develops when the proximal segment of bowel (intussusceptum) telescopes into the distal segment (intussuscepiens) causing obstruction.3

Nonobstructing intussusception is frequently misdiagnosed clinically and is also a challenging diagnosis for a radiologist. Knowledge of presenting complaints and imaging features is essential for early diagnosis of intussusception to start timely management, thus preventing patient morbidity and complications.

In this pictorial case series, there are 10 cases in which all the patients presented with pain abdomen, few of them had episodic colicky pain and remaining had nonspecific chronic pain (>Table 1). None of the patients had typical symptoms of intestinal obstruction. Radiological diagnosis was of immense importance in these cases.

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Case 1

A male patient of 12 years had complaints of colicky recurrent abdominal pain for 3 to 4 months, diarrhea, and vomiting on and off for 2 months. Target sign with hypoechoic lesion was seen on ultrasonography (USG; -Fig. 1A, B). Magnetic resonance (MR) enterography was done (Fig. 1C, D). Multiple intussusceptions, namely, jejunojejunal, ileoileal, and ileocolic types with multiple hyperplastic polyps were confirmed on surgery (Fig. 1E, F). On histopathology, it was diagnosed as associated with Peutz-Jegher syndrome.

Case 2

A 3-year-old female patient presented with vomiting on solids and liquids and constipation since 2 months with no relief on medication. After 15 days, she developed bloody stools with colicky abdominal pain and distension. Barium study revealed dilated cecum and ascending colon,

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 Table 1
 Patients' clinical profile, investigations and final diagnosis

	Sex	Age	Presenting com- plaint	Obstruction	Red currant jelly stools	Chronic/ acute	Radiography	USG/CT	Therapeutic reduction	Postoperative find- ing	Lead point
-	Σ	12 years	Colicky recurrent abdominal pain for 3 to 4 months, diarrhea, and vomiting on and off for 2 months	Absent	Absent	Chronic	Not done	USG: target sign with hypoechoic lesion in target	Saline enema reduction not done	Multiple intussusceptions, namely, jejunojejunal, ileoileal and ileocolic types	Multiple hyperplastic polyps
5.	ш	3 years	Vomiting on solids and liquids, constipation for 2 months, no relief from medication, bloody stools with colicky pain, and abdominal distension for 15 days	Absent	Absent	Chronic	Dilated cecum and ascending colon, increased transit time from ascending colon to transverse colon, narrowing of transverse colon	USG: typical target lesion with a hypoechoic lymph node within it	Saline enema reduction done thrice	lleocolic intussusception	Lymph nodal mass, on IHC diagnosed as Burkitt lymphoma
3.	Σ	2 years	Colicky abdominal pain, later loose stools	Absent	Absent	Chronic	Not done	USG: target lesion with a thick-walled cystic lesion and echogenic contents within relation to it	Saline enema reduction done	lleocolic intussusception	Meckel's cyst
4	Σ	12 years	Recurrent on-off colicky abdominal pain for 3 months, nausea and vomiting for 1 month	Absent	Absent	Chronic	Not done	USG and CT findings: target lesion with soft tissue density lesion in left hemiabdomen	Not done	Colocolic intussusception	Non–Hodgkin's lymphoma
5.	Ŀ	2 years	Recurrent pain abdomen which was not relieved by medicines	Absent	Absent	Chronic	Gas filled bowel loops in left half of abdominal cavity	USG: target lesion with compressible cystic lesion CT: target sign, thick walled cystic lesion seen	Saline enema reduction done	lleocolic intussusception	Duplication cyst
9	ш	16 years	Chronic on and off pain abdomen since 2 months	Absent	Absent	Chronic	Not done	USG; target lesion in left hemiabdomen, suspicious echogenic lesion supposed to be a lead point. CT: target lesion containing a round intraluminal fat density mass within it	Not done	Colocolic intussusception	Lipoma
7.	Σ	3 years	On-off colicky abdominal pain for 1 month	Absent	Absent	Chronic	Not done	USG: target lesion with a thick-walled cystic lesion filled with echogenic contents seen in relation to it	Water enema reduction done	lleocolic intussusception	Meckel's cyst
%	Σ	8 years	Recurrent abdominal pain since 1 month	Absent	Absent	Chronic	Not done	USG: typical target lesion with a large hypoechoic mass within it	Not done	lleocolic intussusception	Non–Hodgkin' lymphoma
6	Σ	6 months	Incessant crying with leg pulling, fever, blood in stools since 2–3 days.	Absent	Absent	Acute	Not done	USG: S-shaped target lesion with thickened hypoechoic bowel wall	Not done	Long segment ileocolic intussusception with gangrenous bowel	Polyp
10.	Σ	3 years	Nonspecific abdominal pain for 15–20 days	Absent	Absent	Chronic	Not done	USG: target lesion with thick walled cyst filled with echogenic fluid	Not done	lleocolic intussusception	Duplication cyst

Abbreviations: CT, computed tomography; F, female; M, male; USG, ultrasonography.

Fig. 1 A 12-year-old boy presented with colicky recurrent abdominal pain for 3 to 4 months, diarrhea, and vomiting on and off for 2 months. Ultrasonography images (A) show pseudo kidney sign of intussusception in longitudinal view. (B) Target sign and a hypoechoic lesion (arrow) within it. Magnetic resonance enterography axial section (C and D) shows bowel within bowel appearance of intussusception (arrow) and hypointense lesion arising from wall protruding into lumen seen. (E) Resected part of the bowel with multiple polyps (thin arrow). (F) Single polyp specimen. On histopathological examination it was diagnosed as hyper plastic Peutz-Jegher polyp.

increased transit time from ascending colon to transverse colon, and narrowing of transverse colon (►Fig. 2A-D). On USG, typical target lesion with a hypoechoic lymph nodal mass showing vascularity on color Doppler was seen within it (Fig. 2E, F). Findings were suggestive of ileocolic intussusception. Saline enema reduction was done for therapeutic purpose. The findings were confirmed surgically and histopathologically lymph nodal mass was proven to be as Burkitt lymphoma.

Case 3

A 2-year-old male child presented to OPD with colicky abdominal pain for 5 to 6 days and loose stools for 3 days. On USG, a target lesion was seen in right lower abdomen. A thick walled cystic lesion filled with echogenic contents was noted in close relation to this which was the probable lead point (Fig. 3A-C). It was provisionally diagnosed as ileocolic intussusception later proven surgically. The cyst was histopathologically identified as Meckel's cyst.

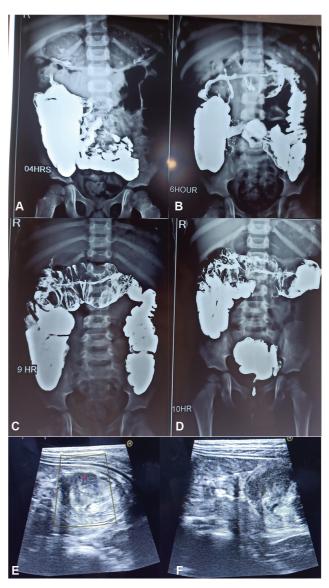


Fig. 2 A 3-year-old girl had recurrent vomiting, constipation for 2 months, bloody stools after 15 days with colicky pain, and abdominal distension. Delayed images of barium meal follow through study reveals dilated cecum and ascending colon (A), delayed transit time of contrast from ascending colon to transverse colon at 4 hours (B), beaking appearance with narrowing of lumen in the transverse colon (C and D). Ultrasonography image of the patient (E and F) shows typical target sign of ileocolic intussusception. A hypoechoic lesion with internal vascularity (arrow) is clearly seen in it. The lesion was diagnosed as Burkitt lymphoma on immunohistochemistry study.

Case 4

A 12-year-old boy had complaints of multiple episodes of on and off colicky abdominal pain for 3 months and nausea and vomiting for 1 month. USG showed a target lesion in left side of abdominal cavity with a hypoechoic mass within it (**>Fig. 4A**). Non-contrast computed tomography (NCCT) with oral contrast was done which showed intussusception with a soft tissue density lesion in descending colon (Fig. 4B, C). Postoperatively, it was confirmed as colocolic



Fig. 3 A 2-year-old boy had colicky abdominal pain, later loose stools. Ultrasonography images showing typical pseudokidney sign of intussusception in longitudinal view (A) and well defined cystic lesion (arrow) in the intussusception (B and C) which was proven post operatively Meckel's cyst acting as a lead point.

intussusception with non-Hodgkin's lymphoma as the lead point (**►Fig. 4D, E**).

Case 5

A 2-year-old female patient presented with recurrent pain abdomen which was not relieved on medication. No other clinically relevant history was present. X-ray abdomen showed gas filled bowel loops in left half of abdominal cavity. On USG, target lesion with a compressible cystic lesion was seen (>Fig. 5A, B). On contrast-enhanced computed tomography (CECT), ileocolic intussusception with a thick walled cystic lesion was identified in relation to it (>Fig. 5C). Saline enema reduction done to relieve the symptoms. Later surgery was done which confirmed ileocolic intussusception with a duplication cyst as lead point (**►Fig. 5D**).

Case 6

A 16-year-old female patient had chronic abdominal pain without any other significant complaints. On USG, a target lesion with a round echogenic lesion within it was noted in left of abdominal cavity (>Fig. 6A). NCCT was done to confirm USG findings. CT findings were similar to that of USG findings and the round lesion picked up on USG was a fat density lesion suggestive of lipoma (Fig. 6B, C). The findings were confirmed surgically as colocolic intussusception and the lead point was proven to be a lipoma on histopathological examination (\succ Fig. 6D-F).

Case 7

A 3-year-old male child had recurrent chronic pain abdomen and vomiting on and off for 1 month. On USG, bowel within bowel appearance was seen as a target sign and a cyst filled with fluid having internal echoes was noted adjacent to it (Fig.7A, B). Therapeutic saline enema reduction was done multiple times. Postoperatively, it was

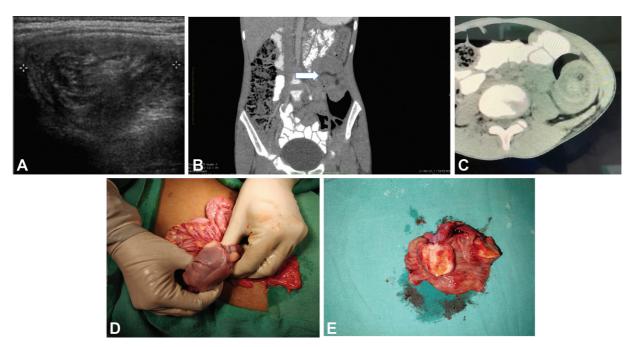


Fig. 4 A 12-year-old boy had complaints of recurrent on-off colicky abdominal pain for 3 months, nausea and vomiting for 1 month. Ultrasonography image (A) shows typical target lesion (star). Coronal (B) and axial (C) contrast-enhanced computed tomography images showing target lesion (arrow) involving colon seen in left lower quadrant with thick soft tissue density mass (thin arrow) in intussusceptum. (D and E) shows intraoperative and postoperative specimen of the resected bowel showing mass in wall of involved bowel loop. On histopathological examination it was proven as lymphoma.

Fig. 5 A 2-year-old female presented with acute pain abdomen which was not relieved on medication. Ultrasonography abdomen image (A and B) shows telescoping bowel loops in transverse section with a compressible cystic lesion (arrow) with echogenic contents within. Axial contrast-enhanced computed tomography (C) showing thick walled cystic lesion (thin arrow) with wall enhancement filled with fluid density contents and target sign of ileocolic intussusception (arrow). Intraoperative image (D) shows cystic lesion (star) in relation to ileocolic junction with proximally dilated ileal loops (arrow). The cyst was diagnosed as duplication cyst on histopathology.

confirmed to be an ileocolic intussusception with Meckel's cyst (►Fig. 7C).

Case 8

An 8-year-old male patient had complaints of nonspecific chronic pain abdomen since 1 month. USG was done as screening test to find out any cause of pain. Typical target lesion with hyperechoic mass with internal vascularity on color Doppler was detected in right side of abdomen (Fig. 8A, B). In CECT, abdomen with intravenous (IV) and rectal contrast, ileocolic intussusception with soft tissue density mass was noted (>Fig. 8C). After surgery and histopathological examination, it was proven as non-Hodgkin's lymphoma with ileocolic intussusception.

Case 9

A 6-month-old male infant came with incessant crying and pulling of legs towards abdomen, fever, and bloody stools. USG examination showed S-shaped target lesion with thickened and hypoechoic wall, without vascularity on color Doppler (►Fig. 9A, B). No lead point was picked up on USG. Postoperatively, it was confirmed as ileocolic intussusception with necrotic bowel wall. Polyp was identified as the causative lead point.

Case 10

A 3-year-old male patient came with chronic pain abdomen. On USG, a target lesion with thick-walled cyst in relation to it was noted in right side of abdomen, diagnosis given as

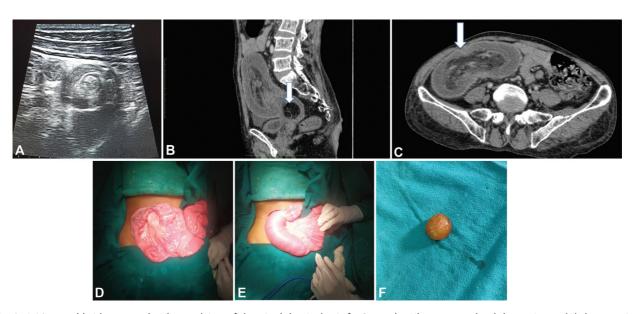


Fig. 6 A 16-year-old girl presented with complaints of chronic abdominal pain for 2 months. Ultrasonography abdomen image (A) shows typical target sign of telescoping bowel loops in transverse section. Contrast-enhanced computed tomography abdomen images (B) sagittal view and (C) axial view showing intussusception and fat density lesion of lipoma (arrow) as lead point. Intraoperative images (D) showing telescoping bowel loops (thin arrow) and proximal dilated bowel loops (arrow; E). (F) Specimen which was proven as lipoma on histopathological examination.

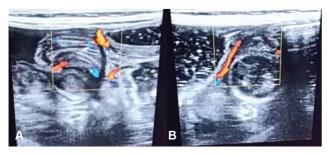
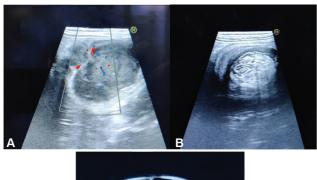


Fig. 7 A 3-year-old male child presented with on-off colicky abdominal pain for 1 month, not relieved on medication. Ultrasonography images (A) showing intussusception in longitudinal view with cystic lesion in relation to it. (B) The cystic lesion. (C) Intraoperative image of dilated bowel loops (arrow) of ileocolic intussusception with Meckel's cyst as lead point which was detected on histopathological examination.



Fig. 9 A 6-month-old male infant had incessant crying with leg pulling, fever and bloody stools since 2 to 3 days. Ultrasonography (USG) images (A and B) shows S-shaped target sign with hypoechoic, thickened bowel wall suggesting possibility of ischemic changes which was surgically proven as long segment ileocolic intussusception with complication of gangrenous bowel. A polyp was identified as lead point which could not be picked up on USG.



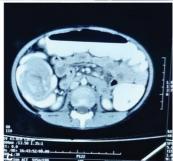


Fig. 8 A male patient of 8 years had complaints of recurrent abdominal pain. Ultrasonography images (A and B) showing target sign of intussusception (arrow) with hyperechoic mass with internal vascularity (star) suspected to be a lead point seen in target. (C) Axial contrast-enhanced computed tomography image with rectal contrast showing intussuscepiens (black arrow) filled with contrast. A large soft tissue density mass (curved arrow) seen in intussusceptum. Thin rim of contrast also filling it (thin arrow).

ileocolic intussusception. Postoperatively duplication cyst with ileocolic intussusception was confirmed.

Results and Discussion

Intussusception typically occurs in children aged 6 months to 4 years. In neonates and children older than 2 years, intussusception have high incidence of associated bowel abnormality with a lead point.^{5,6} Similar results were observed in our study. Out of 10 cases of intussusception with pathologic lead point, 9 cases included patients aged 2 years and/or above and one case was of 6 months of age. In our study, incidence was more in male population as compared with female population. Out of 10 cases, 7 were male and 3 were female.

The classical triad of symptoms of colicky abdominal pain, vomiting, and bloody stools occur in <25% of patients.⁷ A wide spectrum of symptoms may range from painless intussusception to constipation, dehydration, diarrhea, rectal prolapse, rectal bleeding, sepsis, shock, syncope, vomiting, and altered mental status (lethargy and irritability).8,9

Patients in our study presented with colicky pain abdomen (acute and chronic type) with nausea, vomiting, loose stool, constipation, and bloody diarrhea. None of them had classical triad of symptoms of intussusception or symptoms typical of intestinal obstruction.

Ileocolic type of intussusception is common and generally idiopathic. 10,11 Other variety of intussusceptions include jejunojejunal, ileoileal, and colocolic. These are far less common than ileocolic type. In our study, ileocolic intussusception was commoner than other types. Out of 10 cases, 7 were of ileocolic and 2 were of colocolic types. One case was of multiple intussusceptions including jejunojejunal, ileoileal, and ileocecal.

Colonic intussusception usually have a malignant lead point 12 but small intestinal intussusception most of the time have benign lead point. 7,13 We noticed similar results in our

Meckel's cyst, duplication cyst, multiple polyps of nonfamilial polyposis syndrome, Burkitt lymphoma, and non-Hodgkin's lymphoma were present in two, two, one, one, and one cases of ileocolic intussusception, respectively. In jejunojejunal intussusception familial polyposis syndrome (Peutz-Jegher syndrome) was the lead point. In colocolic intussusception cases, the lead point was non-Hodgkin's lymphoma and lipoma in one case each.

Lead point causes alteration in normal peristaltic movements which leads to propulsion of bowel distal to it, predisposing to intussusception.

Plain radiography is the first imaging modality but it lacks sensitivity. 14,15 USG remains the standard modality. Its

All 10 cases in our study were diagnosed on USG and confirmed postoperatively as intussusception with lead points, however, was not clinically suspected.

Lead point was picked in 9 out of 10 cases on USG, identified as cyst or suspicious lesion in close relation to the intussusception. In one case where the lead point was not picked up sonographically, it was identified intraoperatively.

In another case of ileocolic intussusception, thickened and hypoechoic wall of invaginating bowel segment suggesting possibility of necrosis/ischemia was identified on USG which was later confirmed intraoperatively.

Intussusception is seen on USG as target/bull's eye/doughnut sign in the form of alternate concentric hypoechoic and hyperechoic rings on transverse view.¹⁷ In longitudinal view, peripheral hypoechoic and central echogenic area is seen, termed as pseudokidney sign.¹⁸ Central echogenecity of pseudokidney sign is contributed by mesentery due to its fat content. Color Doppler study has an important role in detection of bowel viability.

The invaginating segment or intussuscepiens undergoes ischemia due to compression of mesenteric vessels which are pulled along with its mesentery. It can complicate into necrosis of bowel wall resulting in perforation, peritonitis, and shock.¹⁹ Superimposed infection may deteriorate the condition. Such patients can develop fever. Sloughing of mucosa presents as red currant jelly stools.²⁰

Although CT is gold-standard investigation for diagnosing intussusception but it is usually done only in cases of doubtful diagnosis or to confirm suspected lead point, since it involves radiation and frequently necessitates sedation of children. In our study, it was done in two cases of colocolic type and one case of ileocecal intussusception with non-Hodgkin's lymphoma. MR enterography was done in one case of multiple intussusceptions with Peutz–Jegher syndrome to confirm USG diagnosis.

In our study, saline enema reduction was attempted in 5 out of 10 cases; however, it was not a sonographically guided hydrostatic reduction. Both hydrostatic reduction and air insufflation reduction are safe without any complications during or after procedure and can be attempted before surgery.^{21,22} These have high success rate and are perfect method for the nonoperative treatment of pediatric intussusception and can be widely used as routine therapy.^{23,24} The main benefits of USG-guided hydrostatic reduction apart from avoidance of exposure to radiation include less patient discomfort, shorter hospital stay, and less morbidity and mortality compared with surgical modality of treatment.²⁵ Knowledge of optimum pressure needed for successful reduction is the main prerequisite of the procedure. Successful reduction of intussusception eliminates the need of surgery in many cases.

Conclusion

Pain abdomen (acute and recurrent types) can be the only presenting symptom/feature of pediatric intussusception.

Pathologic lead point can be a causative factor for intussusception in children. It should be kept as one of the less common differential in pain abdomen patients of pediatric age group.

USG remains gold standard in diagnosis. Early diagnosis and, thereby, early treatment helps in reducing mortality and morbidity.

Conflict of Interest None declared.

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