

Where Suspicion Bore Fruit- An Unusual Cause Of Abdominal Pain In A 46-Year-Old Man

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Citation

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Abstract

Abdominal pain is a common presenting complaint among adult patients presenting to the emergency department. Triage to medical versus surgical management is crucial for improved outcome. Here, we present the case of a 46-year-old man who presented with subacute onset of migratory abdominal pain, nausea, vomiting and hematochezia. Abdominal x-ray was normal but a CT scan revealed intussusception of the ileum into the transverse colon. He was treated with an emergency right hemicolectomy with side-to-side ileocolic anastomosis. Histopathology of the resected bowel revealed transmural inflammation but no cancer. We also review published literature on the epidemiology, manifestations, diagnosis and management of ileocolic intussusception in adults.

CASE

A 46-year-old male presented to the emergency department complaining of severe, deep boring, nearly continuous, abdominal pain for 1 month as well as nausea, vomiting and bloody stools. The onset of the abdominal pain was sudden and occurred while he was at work. It began in his epigastric region and was migratory, radiating sequentially to the right upper, right lower, suprapubic, left lower and left upper quadrant. It was worse after eating and with movement and not relieved with rest or change in posture. He had noticed bright red blood in his stools about 4 days after pain started and continued to have bloody bowel movements, sometimes mixed with stools and sometimes frank blood. A review of systems was positive for a 4-5 day history of lightheadedness, decreased appetite, decreased frequency of urination and dark urine but negative for fever, chills, shortness of breath, or cough. He had no past history of similar pain but did have a tumor removed from the bladder (pathology unknown) and nephrolithiasis. He had never had a colonoscopy. He was on baclofen, oxybutinin, hydrochlorothiazide, lisinopril and potassium citrate. His family history was significant for a malignant tumor of an unknown internal organ in his maternal grandmother. He was a past smoker, denied recent alcohol intake, and admitted to marijuana and cocaine use as a young man. Physical examination revealed stable vital signs and marked tenderness to palpation in the suprapubic region with a lesser degree in the right and left lower quadrants of the abdomen.

Rectal examination revealed no mass or hemorrhoids but was positive for occult blood. On the initial laboratory tests, he had a normal complete blood count (including differential), low glucose (68mg/dl, reference range: 90-110 mg/dl) and an otherwise unremarkable complete metabolic profile. An abdominal x-ray was normal. A CT scan of the abdomen was obtained which is shown in Figure 1.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis of migratory abdominal pain worsened after eating is profound and can be divided into traumatic, infectious, inflammatory, neoplastic, autoimmune, metabolic, anatomic and vascular causes (Table 1). Of these, the conditions which are associated with a lower GI bleed include IBD, vascular anomalies (angiodysplasia, ischemia, internal hemorrhoids or brisk upper GI bleeding), neoplastic (colon cancer and polyps), congenital (Meckel's diverticulum) and autoimmune (vasculitis) conditions [1,2].

CLINICAL COURSE

The general surgery team was consulted and the patient underwent an exploratory laparotomy on the day of admission which revealed intussusception from the ileum to the mid transverse colon. A right hemicolectomy was performed followed by a side-to-side stapled anastomosis between the ileum and transverse colon. The patient had an uneventful post-operative recovery and was discharged home 5 days post-operatively. At his 1 week follow-up 6

days post-discharge he was doing well. Gross pathological examination confirmed intussusception of the ileum into a portion of transverse colon. The colonic and ileal wall in the area of intussusception were edematous. Microscopic examination revealed mucosal ulceration, and transmural acute and chronic inflammation. There was no evidence of malignancy (Figure 2).

Figure 1

Figure 1: Contrast-enhanced CT scan of the abdomen demonstrating (a) the typical “target sign” or “pseudo kidney” sign of an ileocolic intussusception. The intussusceptum (black arrowhead) with the mesenteric blood vessels and fat (black arrow) is surrounded by the thick walled intussuscipiens (white arrowhead) (b) another view showing the leading edge (white arrowhead) with surrounding mesentery, blood vessels and fat (white arrow) and the thick intussuscipiens (black arrow)

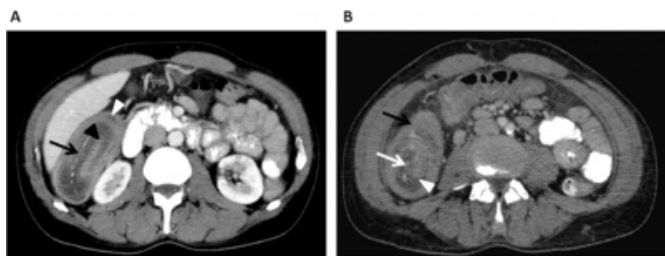


Figure 2

Figure 2: H&E-stained section of the colonic wall showing transmural inflammation (original magnification 20x)

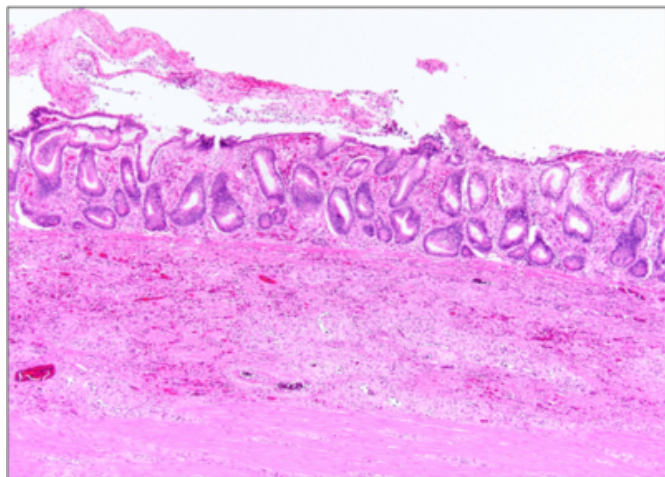


Figure 3

Table 1: Differential diagnosis of migratory abdominal pain aggravated by eating

Category	Diseases
Traumatic	Bowel contusion Gastric perforation
Infections	Gastroenteritis Intestinal parasites Ileocecal tuberculosis Whipple's disease Cholecystitis Typhilitis
Inflammatory	Acute pancreatitis Primary sclerosing cholangitis
Neoplastic	Cancer involving the small bowel, stomach, pancreas Intestinal lymphoma
Autoimmune	Inflammatory bowel disease (IBD)
Metabolic	Lactose intolerance
Anatomic	Intestinal obstruction Intestinal perforation Cholelithiasis Cholecystitis
Vascular	Ischemic bowel disease Venous thrombosis

DISCUSSION

Intussusception is defined as the telescoping of one segment of the gastrointestinal tract into an adjacent segment. This can either be anterograde (when a proximal part of the bowel telescopes into a distal portion) or retrograde (when the distal segment telescopes into the lumen of the adjacent proximal segment). The part of the bowel that is telescoping is called the intussusceptum while the part of the bowel into which telescoping occurs is called the intussuscipiens. The major complication of intussusception is intestinal obstruction. Further, blood vessels and soft tissue can get trapped, followed by edema (due to obstruction to vascular outflow) and subsequently necrosis and perforation. Unlike in children, where it accounts for 95% of cases of intestinal obstruction, intussusception is rare in adults (ratio of pediatric to adult cases 7:1 in one study), accounting for only about 1% of all cases (annual incidence of intussusception in adults estimated to be 1-2 per 1,000,000) [3-5]. Further, unlike in children, where this condition is common and often without any pathology, an identifiable pathological lesion accompanies intussusception in about 90% of adults. The characteristic symptoms of abdominal pain, nausea and hematochezia, and physical examination findings of a sausage-shaped palpable abdominal mass seen in children may not always be present in adults. Even if present, they may mimic other more common etiologies [6,7]. Hence, it is important for clinicians, particularly internists and family physicians to keep this condition in mind when evaluating a patient with abdominal pain. Rapid surgical consultation and

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surgery can be lifesaving in these patients.

We conducted a literature search for published case reports and case series of ileocolic intussusception in adults. The PubMed database was searched with the term “ileocolic intussusception adult”. For case reports for which a full text was available, the patient and treatment characteristics were tabulated (Table 2).

Figure 4

Table 2: Summary of adult ileocolic intussusception case reports

First author	Age	Gender	Presenting complaints	Location of abdominal pain	Duration of symptoms	Surgery	Histopathology	Outcome
Aydede [38]	52	Male	Abdominal pain, weight loss	RLQ (palpable mass)	N/A	Right hemicolectomy	Primary intestinal B-cell non-Hodgkin lymphoma	Full recovery with post-operative chemotherapy (last fu date N/A)
Balamoun [21]	65	Male	Abdominal pain, vomiting	RLQ	3 days	Right hemicolectomy with ileo-transverse colon anastomosis	Submucosal lipoma (1.2cm) in terminal ileum	Full recovery (last fu date not mentioned)
Butt [4]	28	Male	Abdominal pain and bloody diarrhea	LLQ	3 days	Reduction and partial resection of ileum with end-to-end anastomosis*	Inflammatory pseudopolyp	Full recovery (3 months)
Bravo [15]	75	Male	Abdominal pain, vomiting, diarrhea	N/A	Pain - days	Right hemicolectomy with ileo-transverse colon anastomosis	Submucosal ileal lipoma (5.5cm) Ovarian appendix	Full recovery (fu date not mentioned)
Bui [49]	39	Male	Abdominal pain, nausea, vomiting	Epigastric	5 days	Right hemicolectomy	Lipoma	Full recovery (last fu date 7 days post surgery)
Chughthai [25]	58	Male	Abdominal pain, vomiting	RLQ	6 months	Right hemicolectomy	Tubulovillous adenoma of ileocecal valve	Full recovery (last fu date 5 days post-operatively)
Chen [28]	45	Male	Abdominal pain, watery diarrhea	RLQ	2 months	Right hemicolectomy	Cecal lymphangioma	Full recovery (last fu date not mentioned)
Cheung [32]	72	Male	Abdominal pain	Pentambilical	1 week	Right hemicolectomy, partial ileostomy, ileocolostomy	Cecal adenocarcinoma	N/A
Fanney [56]	41	Male	Abdominal pain, vomiting, hematochezia, h/o AIDS	RLQ	N/A	Right hemicolectomy	Lymphoma and CMV colitis	N/A

Figure 5

Watanabe [47]	59	Male	Abdominal pain	Lower	4 days	Manual release of intussusception and resection of affected ileum	Submucosal lipoma	N/A
Hirasaki [55]	47	Male	Abdominal pain	N/A	Several months	Ileocecal resection	Schwannoma of ileum	Full recovery (last fu date not mentioned)
Ibrahim [23]	66	Male	Abdominal pain, nausea, vomiting, diarrhea	Suprapubic	4 days	Right hemicolectomy with ileo-transverse anastomosis	Adenocarcinoma of the ascending colon (Dukes C)	Full recovery (last fu date not mentioned)
Khan [66]	74	Male	Abdominal pain, altered bowel habits, weight loss	RUQ	4 months	Right hemicolectomy with end-to-end ileocolic anastomosis	Submucosal lipoma of the cecum	Full recovery (last fu date 7 days post surgery)
Khanum [67]	81	Male	Abdominal pain	RUQ	5 days	Right hemicolectomy	Tubulovillous adenoma with severe dysplasia	N/A
Kozishi [41]	73	Male	Severe anorexia	N/A	N/A	Surgical resection of tumor	Large ileal lipoma (5.5cm x 5cm)	Full recovery (last fu date N/A)
Lague-de-Launay [22]	32	Male	N/A	N/A	N/A	Surgery N/A	Hodgkin lymphoma	N/A
Mahajna [3]	21	Male	Abdominal pain, nausea	RLQ	3 days	Right hemicolectomy	Cecal fecolith	Full recovery (last fu date 8 days postoperative)
Meisler [11]	75	Male	Abdominal pain, distention, constipation	Diffuse	8 days	Right hemicolectomy with end-to-side ileocolic anastomosis	Pedunculated lipoma	Full recovery (last fu date 4 days post surgery)
Namikawa [16]	68	Male	Abdominal pain	RUQ	6 month	Laparoscopy-assisted ileocecal resection of tumor with LN dissection and 1 anastomosis	Well differentiated tubular adenocarcinoma of cecum	Full recovery (5 years follow-up)
Nestorovic [17]	26	Male	Abdominal pain	N/A	N/A	Right hemicolectomy with end-to-end ileo-transverse anastomosis	Lymphoma	Full recovery
Shaba [30]	33	Male	Abdominal pain	Epigastric	3 weeks	Partial resection of ileum	Submucosal small bowel lipoma	Full recovery (last fu date 2 years post surgery)
Ho [56]	55	Male	Abdominal	RLQ	2 days	Ileocecal	Inverted	Full recovery

Figure 6

						resection	Mechel's diverticulum	(fu not mentioned)
			pain and vomiting					Rectovaginal fistula treated with temporary colostomy closed successfully 3 months later
Inducolo [25]	36	Female	Fluctuating abdominal pain, diarrhea, distention	RLQ	1 month	Laparoscopic debulking of pelvic endometriosis followed by right hemicolectomy	Intestinal endometriosis	Full recovery (last fu date 3 days post operative)
Ibrahim [24]	21	Female	Abdominal pain, distention, diarrhea	RLQ	7 hours	Ileocecal resection with primary repair	Prior appendectomy scar leading to thickened cecum telescoping into itself	Full recovery (last fu date 5 days postoperative)
Kim [10]	30	Female	Abdominal pain	RLQ	1 day	Ileocecal resection	Cecal lymphangioma	Full recovery (last fu date 5 days postoperative)
Kostasouralis [57]	32	Female	Abdominal pain, distention, constipation	Diffuse	24 hours	En-bloc resection of the cecum, right colon, appendix and terminal ileum	Endometriosis	Full recovery (last fu date 6 days post surgery)
Murden [12]	69	Female	Abdominal pain and lump	LUQ	N/A	Laparoscopic partial resection followed by right hemicolectomy	Mucinous adenocarcinoma of cecum (T3N0 with margins free of tumor)	Full recovery (last fu date 3 days postoperative)
Yoshino [18]	39	Female	Abdominal pain, nausea, vomiting coinciding with menses	RLQ	Months	Ileocecal resection	Widespread submucosal endometriosis of the intestine	Full recovery (last fu date 7 days post surgery)

*This patient also had incidentally diagnosed situs inversus totalis; N/A: Not available, RLQ (right lower quadrant), RUQ (Right upper quadrant), LLQ (left lower quadrant)

EPIDEMIOLOGY

There are four main types of intussusception depending on the intussusceptum and intussusciptens - a) enteric (confined to the small intestine), b) ileocecal (the ileocecal valve is the lead point), c) ileocolic (terminal ileum telescopes into the colon) and d) colocolic (confined to the colon). The relative

incidence of ileocolic intussusception (as in our patient) is variable depending on the study, but its relative rarity is a matter of agreement. In a retrospective review of nearly 381,000 CT reports, Rea and colleagues found that the incidence of adult intussusception was 0.04% (n=170) with a mean age of 41 years. Nearly 88% of these cases were enteroenteric, 4.7% ileocecal, 6% colonocolic and 1.8% gastroenteric. About 7% of patients had evidence of intestinal obstruction on CT [8]. Another study from Winnipeg noted that 2 (9%) of 22 adult intussusceptions were ileocolic [7] and a slightly lower figure of 5% was noted in a retrospective study of 20 adult intussusception cases from France [9]. It has been suggested that African countries may have a higher incidence of ileocolic intussusception due to higher incidence of abdominal lymphadenopathy, chiefly due to infections like tuberculosis [10,11].

Causes of ileocolic intussusception in adults reported in literature include congenital anomalies (inverted Meckel's diverticulum), inflammation (pseudopolyp, endometriosis, surgical scar tissue, colitis and lymphadenopathy), mechanical obstruction (fecolith), benign (lipoma, lymphangioma, appendiceal mucocele, hamartoma, neurofibroma, schwannoma and giant mucocele of the appendix) and malignant (lymphoma, cecal adenocarcinoma, leiomyosarcoma and metastatic melanoma) neoplasms and iatrogenic (post-surgical) [3,8,12-48,48-57]. Two cases have been reported of patients with AIDS who developed an ileocolic intussusception secondary to B-cell lymphoma in the small intestine and cecum, respectively [36;51]. Hyperplastic lymphoid tissue was noted to be the cause of intussusception in another patient with AIDS [58]. In another case, a 29-year-old woman presented with symptoms of intussusception due to an inflammatory myofibroblastic tumor, a neoplasm usually occurring in the lungs [59]. There is an increasing tendency for the intussusciens to be a malignant neoplasm with advancing age of the patient. Occasionally, no pathology is found except for edema of the bowel wall. In these cases, altered peristalsis (for instance due to medications) is suggested to contribute to the disease process [27]. The incidence of these idiopathic cases varies, being nearly 23% in one report [10]. Sometimes, tumors may metastasize to the colon and cause intussusception [60]. Often the tumor telescopes into an adjacent bowel segment by virtue of its size and pulls the rest of the bowel owing to persisting peristalsis [61]. In one study from China comprising 15 patients with adult ileocecal intussusception (comprising 36% of all intussusceptions in that cohort), the

most common cause of intussusception was a benign tumor (33%) including small-intestine lipoma, hamartoma and adenoma. Thirty-three percent of the cases were due to inflammation (suppurative appendicitis, cecal inflammation, mesenteric adenitis and non-specific necrosis), 20% due to a malignant tumor (gastrointestinal stromal tumor and malignant B-cell lymphoma) and one case each was due to scar tissue from a prior appendectomy and a Meckel's diverticulum [62]. In another study from Nepal about 50% of ileocecal intussusceptions were malignant (compared to 38% of enteric and 70% of colocolic intussusceptions)[63]. A retrospective review of 28 cases of adult intussusception from Turkey found that only 2 (7%) were of ileocolic type. The mean age of patients with intussusception was 38.6 years [64]. Further evaluation of this study could not be carried out due to lack of an English language version of the article. Ahn and co-workers, in a study from Korea, reported that ileocolic intussusception accounted for about 9.5% of adult intussusceptions (n=4/42) with no malignancy in any patient (versus 9% in enteric, 50% in ileocecal and 50% in colocolic intussusception patients with an underlying malignancy) [65].

The incidence of ileocolic intussusception appears to be higher in males. In one case series by Wang and colleagues, 53% of intussusception patients were males and 47% females [66]. From Table 2, we infer that 78% of case reports of ileocolic intussusception have involved males. It can affect adults of nearly any age (21 years to 81 years, Table 2) and all races. In one series, the mean age of 15 adult ileocolic intussusception patients was 37 years (± 14 years) with a range from 18 to 64 years [66].

CLINICAL FEATURES

The most common symptoms of intestinal intussusception in order of occurrence are abdominal pain (48-50%), symptoms of intestinal obstruction (19-31%), a palpable mass (14%), hematochezia (9%), and diarrhea or constipation (5%). Interestingly, in about 5% of the patients, there was no complaint at presentation [67]. The time from onset of symptoms to presentation can be quite variable, ranging from a few hours to 6 months (Table 2). This likely reflects the size of the intussusciens, the rapidity of intussusception, the degree of obstruction and severity of associated symptoms. Abdominal pain, when present, most commonly occurs in the right lower quadrant, although virtually any part of the abdomen may be painful and diffuse pain is not uncommon.

DIAGNOSIS

From the two large institutional studies, it can be concluded that CT scan is the most accurate imaging modality for the diagnosis of ileocecal intussusception (80-90% accurate), followed by ultrasonography (50-60%), contrast enema (50%) and colonoscopy (40%) [62;63]. The classic description of intussusception on CT scan is the “target lesion” or “pseudokidney image” which represents the outer intussusciens and the inner intussusceptum. The ultrasound finding in intussusception is characteristically termed as the “donut sign” in a transverse view and “pseudo-kidney sign” on a longitudinal view [68]. In some studies, finding of a “coiled spring” or “spiral sheath”, “mushroom-like” or “crescent shaped” appearance on barium enema has been suggested to be diagnostic [38].

MANAGEMENT

Surgery is the mainstay of treatment for ileocolic intussusception due to a higher incidence of an underlying neoplasm being the lead point and resulting bowel-wall edema, ischemia, and risk of gangrene and perforation [69]. In one study from China [62], 100% of patients with ileocolic intussusception underwent surgery. Nearly 67% underwent a right hemicolectomy, 27% a segmental small-bowel resection and one an appendectomy. In a Nepalese study, 58% of ileocecal intussusceptions were reduced successfully while 42% required resection. Colonoscopy was the most accurate diagnostic test (100%) followed by CT scan (90%) and ultrasound (70%) [70]. A single center experience in Spain reported 15 cases of adult intussusception over a 14-year period (age range 17-77 years). More than 50% of these were ileocecal. Malignant neoplasms were responsible for 50% of ileocecal intussusceptions. In a retrospective analysis from Virginia, about 18% of the 170 patients diagnosed with intestinal intussusception (including 3 with ileocolic disease) underwent surgery [8]. Patients who were found to have intussusception at surgical exploration had a longer length of intussusception (9.6cm vs. 3.8cm), a wider diameter of the involved segment (4.8cm vs. 3.2cm) and more commonly showed the presence of a lead point (53% vs. 30%) [8]. In a 1996 article, Steinwald and colleagues recommended that ileocecal (and ileocolic) intussusceptions should be managed either by an attempt at gentle reduction (if the cecum is in its normal position) or a right hemicolectomy (if the cecum is invaginated upon itself or is observed to have migrated distally), based on the relatively higher risk of an underlying malignancy driving the intussusception in the latter [71].

CONCLUSION

Internists and family physicians are often the first to be called to evaluate a case of abdominal pain. Ileocolic intussusception is a rare cause of abdominal pain in adults which can often be missed primarily due to the frequent lack of peritoneal signs, lack of significant toxicity and a normal abdominal radiograph. A high index of suspicion needs to be maintained and awareness that CT scan is the most accurate diagnostic modality is important for early diagnosis and surgical referral of patients for a favorable outcome.

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