

Bowel intussusception in adults: a report of three interesting cases and current trends for diagnosis and surgical management

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Abstract

Background: Bowel intussusception in adults remains a rare and constant diagnostic challenge for surgeons. It has an incidence of around 2-3 new cases per million per year, and its primary cause is benign or malignant neoplasms of the small bowel and colon. This report aims to outline the importance of high clinical suspicion regarding intussusception in adults presenting with abdominal pain in the emergency department.

Case report: This is a retrospective review of three cases of adult ileocecal intussusception that were treated in a single surgical department in three years (2015-2018). All patients underwent right hemicolectomy in keeping with the principles of surgical oncology. Each patient had a different clinical presentation, while, in terms of the underlying pathology, the first had an adenocarcinoma of the ascending colon, the second an adenocarcinoma of the ileocecal valve, and the third one an inflammatory fibroid polyp of the ileocecal valve, also known as Vanek's tumor.

Conclusion: Large bowel intussusception in adults is quite an interesting entity, not only for its rarity but for its non-specific and atypical clinical presentation as well. High suspicion from the clinician's part and availability of a computed tomography scan is the key to diagnosis. It is not unusual for imaging modalities to be unable to identify the cause of the intussusception. Thus, surgery is always the preferred method of treatment, as, more often than not, a neoplasm of the small or the large bowel is the underlying pathology. HIPPOKRATIA 2019, 23(1): 37-41.

Keywords: Adult intussusception, ileocecal intussusception, clinical presentation, histology, adenocarcinoma, inflammatory fibroid polyp, Vanek's tumor, computed tomography

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Introduction

Bowel intussusception is defined as the telescoping of a proximal segment of the intestine into a distal one. The proximal part is called the intussusceptum, while the distal part, the intussusciptens. Any parts of the gastrointestinal tract could be involved, but the parts with the highest incidence are the small (jejunum and ileus), and the large bowel. Intussusception is an infrequent entity, especially in adults. It has a higher incidence in the pediatric population, being the second most common cause of acute abdominal pain after acute appendicitis. The incidence in adults is estimated at 2-3 cases/million/year while it accounts for 1-5 % of all bowel obstructions¹⁻³. The male to female ratio appears to be 3 to 1 in both pediatric and adult populations³. Any condition that could alter the peristaltic movement of the intestine could be a reason for intussusception. The etiology of intussusception is idiopathic (more often in children) or due to some kind of a lesion, benign or malignant⁴ (Table 1). Usually, the lesion acts as a "leading point" for the intussusceptum. We report three cases treated in our department, each one with a different clinical presentation and underlying pathology.

Classification

The first one to propose a classification for adult bowel intussusception was Dean et al in 1956, defining four types of intussusception: i) enteric (involving the small bowel), ii) colo-colic (involving the large bowel), iii) ileocecal (the ileocecal valve being the leading point), and iv) ileocolic (the terminal ileum telescopes into the ascending colon through the ileocecal valve)⁵. Another classification system is based on the etiology of intussusception, dividing it into two large groups: benign and malignant⁶. Finally, a radiographic classification discriminates the presence or not of a leading point⁷. Any part of the gastrointestinal tract could be involved in intussusception, but the most common sites are the parts where a more freely moving segment transits into a fixed segment either because it is retroperitoneal or due to adhesions. Up to 90 % of the intussusceptions in adults have a leading point⁶.

Clinical presentation and diagnosis

Identifying intussusception in adults by clinical examination alone is almost an impossible task since there are no pathognomonic symptoms, and they could resemble many other clinical entities. In the pediatric

Table 1: Benign and malignant lesions associated with bowel intussusception.

	Benign		Malignant	
			Primary	Metastatic
Crohn disease	Henoch-Schonlein purpura		Adenocarcinoma	Melanoma
Celiac disease	HIV		GIST	Lung
Lipoma	Postoperative adhesions		Carcinoids	Renal cell cancer
Leiomyoma	Endometriosis		Leiomyosarcomas	Breast
Neurofibromatosis	Meckel's diverticulum		Lymphoma	
Fibro-epithelial polyps				

GIST: Gastrointestinal Stromal Tumor, HIV: human immunodeficiency virus infection.

population, it presents with a typical triad of acute symptoms (abdominal pain, bloody diarrhea, palpable mass) leading the doctor to the diagnosis easily, whereas, in adults, symptoms may be subtle or chronic and not specific. Prominent symptoms in adults are abdominal pain, vomiting, diarrhea, melena, or hematochezia, followed by abdominal distention, palpable mass, and poor nutrition⁸. Plain abdominal radiography offers little to the diagnosis. An ultrasound scan may sometimes prove useful, especially in thin adults. The gas contained in dilated bowel loops and the patient's body habitus may distort or obscure the view, resulting in low diagnostic value. Sonography findings suggestive of bowel intussusception are the "target" or "doughnut" sign in the transverse view, the "pseudo-kidney" sign in the oblique view and the "trident" sign on longitudinal view^{3,9}. The most useful tool in the assessment of the patient is the abdominal computed tomography (CT) and its evolution with multi-detectors (MDCT). The diagnostic accuracy of CT scan interestingly varies from 58-100 % with several typical patterns depicted¹⁰. The first pattern is the "target-like" described as a round mass with intraluminal soft tissue and eccentric fat density. The second is the "reniform pattern", depicting a bilobed mass with low central attenuation and higher peripheral density. The third pattern is the "sausage-shaped" which results from alternating areas of low and high attenuation that are related to the bowel wall, intraluminal fluid, mesenteric fat and fluid, contrast material, or air. All three patterns may exist at the same time and show the same lesions because the pattern correlates with the axis of intussusception as "cut" by the CT beam⁷ (i.e., perpendicular, parallel or oblique to the beam). MDCT also has the ability to differentiate between presence and lack of a leading point. Moreover, MDCT can demonstrate the complications of intussusceptions, represented by bowel wall ischemia and perforation, which are mandatory to be referred for surgery promptly⁷.

First case

A 65-year-old female patient presented to the emergency department with dull abdominal pain of sub-acute onset within the preceding months. Her abdomen was soft and non-tender on palpation with normal enteric sounds, and lab tests revealed anemia [hematocrit (Hct): 24 %, hemoglobin (Hb): 7.8 mg/dl]. She reported in her medical history, atrial fibrillation of recent onset treated with rivaroxaban. Colonoscopy revealed an ascending colon tumor,

and biopsies were taken. While inpatient, she developed intense symptoms of acute abdominal pain and blood loss per rectum. On clinical examination, she had a palpable mass with tenderness on her right abdomen; thus, an emergency CT scan was performed and was diagnostic of a large bowel intussusception (Figure 1, Figure 2). The patient was taken to the operating room, and an exploratory laparotomy was carried out, which unveiled a long tract intussusception with part of the terminal ileum and the ascending colon being intussuscepted into the transverse



Figure 1: Axial computed tomography image showing a segment of right sided colon (white arrow) engulfed into the transverse colon with interposed mesenteric fat and vessels (colo-colic intussusception).

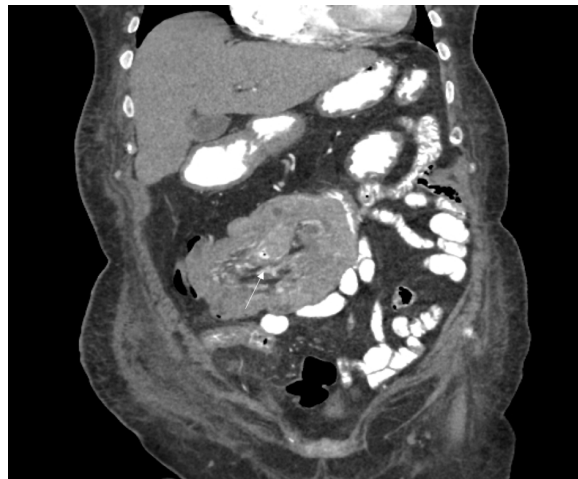


Figure 2: Coronal reformatted computed tomography image demonstrating the colo-colic intussusception (white arrow).

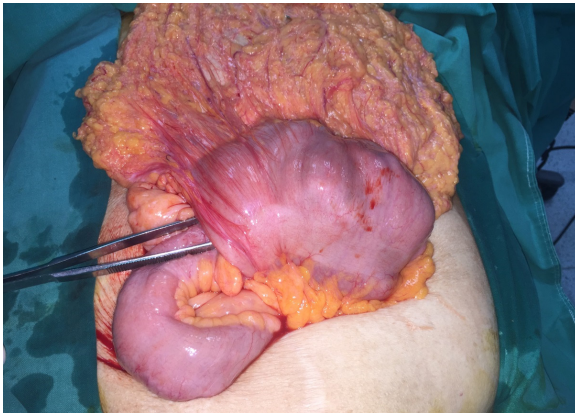


Figure 3: Intraoperative image showing findings consistent with ileocecal intussusception.

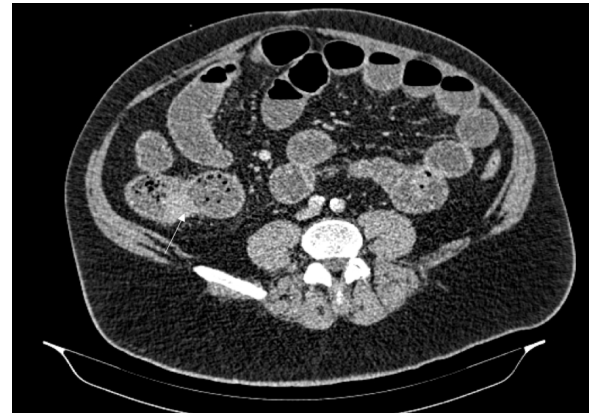


Figure 4: Axial computed tomography image post intravenous contrast media administration showing a hyperdense soft tissue mass (white arrow) at the region of the ileocecum valve as the cause of the ileocecal intussusception.

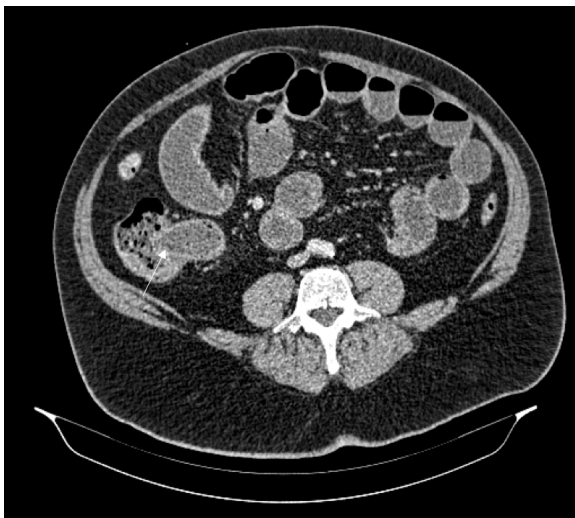


Figure 5: Axial computed tomography image post intravenous contrast media administration showing the region of ileocecal intussusception (white arrow).



Figure 6: Axial computed tomography scan showing a segment of terminal ileum (white arrow) engulfed into the cecum (ileocolic intussusception).



Figure 7: Coronal reformatted computed tomography image visualizing the region of ileocolic intussusception (white arrow).



Figure 8: Specimen of the right hemicolectomy showing a tumor of the ileocecal valve as the leading point of the intussusception.

colon (Figure 3). An extended right hemicolectomy was performed with a side to side anastomosis. Her postoperative course was uneventful, and she was discharged on the eight post-op day. The pathology report showed adenocarcinoma of the ascending colon, and all 18 excised lymph nodes were negative for metastatic disease.

Second case

A 60-year-old man with diffuse abdominal pain of sub-acute onset presented to the Emergency Department due to his persisting symptoms of vague abdominal pain. His abdomen was soft on palpation with mild tenderness, while his lab results revealed an elevated white blood cell count (13,330/ μ l) with 87 % neutrophils. His medical history was significant for diabetes type II. One week earlier, he was subjected to a colonoscopy, which revealed an ulcerative lesion of the cecum. Biopsies taken showed nothing but chronic, non-specific colitis. He was admitted to the Department of Surgery, and a CT scan performed, demonstrated thickening of the wall of the appendix (Figure 4, Figure 5). An exploratory laparotomy was carried out, and an ileocecal intussusception was identified. A right hemicolectomy was performed, and his postoperative course was uneventful. The final pathology reported two different lesions: adenocarcinoma of the ileocecal valve, which is a rare entity, and a submucosal lipoma also located to the ileocecal valve, not being able to discriminate which of the two lesions could have acted as a leading point. The TNM classification was pT3N1M0 (one out of 12 excised lymph nodes infiltrated).

Third case

A 42-year-old lady complaining of diffuse abdominal discomfort for the preceding seven days accompanied by vomiting and diarrhea presented to the Internal Medicine department. On examination, no findings were found other than a mild tenderness on deep palpation of the abdomen, while her labs showed anemia (Hct: 26.5 %, Hb: 8.3 mg/dl). Her medical history was significant for pyoderma gangrenosum (under treatment with methylprednisolone) and depression. A CT scan performed, identified a lesion located in the terminal ileum that was causing an ileocolic intussusception (Figure 6, Figure 7). She underwent an exploratory laparotomy that confirmed the intussusception, and a right hemicolectomy was performed (Figure 8). Final histology showed the rare finding of an inflammatory fibroid polyp of the ileocecal valve, also called Vanek's tumor¹¹. Her postoperative course was uneventful, and she was discharged on the seventh post-op day.

Discussion

The reported cases are solid proof that a clinical diagnosis of intussusception is almost impossible due to the non-specific symptomatology. Each of the cases poses different diagnostic problems and has something unique to consider. In the first case, the diagnosed cecal mass caused mild only symptoms with no signs of intussusception. While scheduled for urgent surgery, she developed

acute symptoms due to ileocolic intussusception, raising a clinical emergency that required exploratory laparotomy. Nevertheless, surgery performed followed the oncological principles yielding the desired results. Another issue that poses particular interest, in this case, was the great length of the intussusception, which is relatively rare¹². In the second case, there was a diagnostic issue as the endoscopic result reported an ulcerative lesion of the cecum with no pathologically proven malignancy, while imaging was suggestive of appendiceal pathology. Laparotomy, being the only choice, revealed the previously mentioned results. Another issue of note regarding this case is the co-existence of two lesions in the ileocecal valve; adenocarcinoma, and submucosal lipoma, both of them rare¹². Finally, in the third reported case, based on her history of pyoderma gangrenosum and the presence of severe anemia, inflammatory bowel disease was suspected, and a benign condition was expected to be found. On laparotomy, there was a mass in the terminal ileum causing the intussusception, so we performed an oncologic resection. The final pathology reported a non-malignant condition, setting the diagnosis of an extremely rare lesion not only histologically but in terms of location as well^{11,13}.

Adult intussusception is a rare condition, accounting only for 1-5 % of all bowel obstruction in adults^{1,8}. About 90 % of intussusceptions are caused by tumor-like lesions, of which approximately 65 % are malignant¹⁰. Symptoms may be subtle, thus making the diagnosis difficult. The onset is usually chronic, whereas acute symptoms arise only in 20 %⁸. The most challenging in the management of intussusception is making an accurate diagnosis rather than treating the disease itself. The non-specific symptoms, the chronic or sub-acute onset, the delay of seeking medical attention, and its rarity render the diagnosis difficult. Thus, emergency physicians, and especially surgeons, should be highly suspicious of this clinical condition since one could encounter this pathology only a few times throughout their medical career.

Bowel intussusception in adults will, in the majority of cases, present as an acute condition in the emergency department; thus, the initial assessment should be followed by fluid resuscitation and correction of the electrolyte imbalance. Regarding the definitive treatment, controversy still exists on whether it should be attempted to reduce the intussusception. There is a theoretical danger of tumor spread during the effort to reduce the intussusception via direct seeding or venous emboli. This hypothesis seems logical but has yet to be proved. There is a consensus that the resection should comply with the principles of surgical oncology when large bowel is involved due to the higher incidence of underlying malignancy. On the other hand, when a considerable length of the small bowel is involved, or there is an accurate preoperative diagnosis excluding malignancy, the reduction could be attempted to avoid short bowel syndrome^{8,14}. The operative approach could be either open or laparoscopic, depending on the surgeon's experience, the pa-

tient's clinical condition, and the suspected underlying pathology^{14,15}.

In conclusion, intussusception in the adult population is a rare entity. Non-specific symptoms and the absence of a characteristic clinical presentation pose a challenge for the surgeon, especially on an emergency basis. That is why it is so imperative that the clinician should be highly suspicious of this pathology. The use of CT scan is of great importance for the diagnosis and a valuable tool for the planning of the optimal treatment. Because in the adult population, the majority of lesions causing intussusception are malignant, surgical treatment should adhere to oncological principles, whether laparotomy or laparoscopy is chosen.

Conflict of interest

The authors declare no conflict of interest regarding this paper.

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