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Bleeding Edge Therapy: Ileocolic Intussusception Due to Ileocecal Valve Adenocarcinoma and Its Management in an Adult Patient—Case Report and Literature Review

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Abstract
Adenocarcinoma as the primary cause of bowel intussusception is uncommon. We describe the case of a 86-year-old patient admitted for ileocecal intussusception due to the presence of adenocarcinoma, located in the ileocecal valve and right colon. The etiologies of intussusception, its diagnosis, and conservative or surgical treatments are discussed, with attention placed on the indications for reduction of the invagination prior to surgical resection.

Keywords Adenocarcinoma · Abdominal pain · Bowel intussusception

Abbreviations

SpO_2	O_2 saturation
CT scan	Computed tomography scan
US	Ultrasonography
Hgb	Hemoglobin
WBC	White blood cells
Plt	Platelets
Fe	Iron
CRP	C-reactive protein
Bpm	Beats per minute

Case Report and Evolution

A 86-year-old woman, with severe cognitive impairment associated with a behavioral disorder, was referred to the St. Orsola University Hospital – Emergency Department with complaints of bloody diarrhea and vomiting. On admission her blood pressure was 120/75 mmHg, pulse rate 100 bpm, SpO_2 97%, while breathing room air. Clinical evaluation revealed a Glasgow Coma Scale of 9/15, initial signs of dehydration, a distended and painful abdomen on palpation without signs of peritonitis. Rectal exploration revealed soft, normochromic stools and external hemorrhoids without signs of bleeding. Admission blood tests included Hgb 9.6 g/dl, WBC 11,000/mm^3, plt 565,000/mm^3, Fe 12 µg/dl and CRP 1.35 mg/dl. An abdominal X-ray showed distension of the bowel in the central quadrants with air-fluid levels. The patient was rehydrated with IV fluids and given antiemetics. The day after, the patient was admitted to the Internal Medicine unit where a new episode of vomiting occurred; a bedside US revealed perihepatic and perisplenic ascites and a dilated right colon with thickened walls. Based on these findings, the patient was transferred to the Emergency Surgery unit. An abdominal CT scan with contrast medium was performed reporting an intussusception of the distal tract of the ileal loops, especially at the level of the right abdominal quadrants (Figs. 1, 2). An attempt of non-operative management by rectal administration of the hypertonic radiocontrast dye Gastrografin® was unsuccessful. The patient underwent surgery the following day; after exploration of the entire small bowel, intussusception of the most distal ileal segment into the ascending colon was identified with detection of a firm mass at the ileocecal valve; the length of the involved bowel was ~ 45 cm. The intussusception was manually reduced (Figs. 3, 4), followed by ileocecal resection and a latero-lateral ileo-colic anastomosis. Postoperative course was uneventful with regular intestinal function resumed on day 5; the patient was discharged on postoperative day

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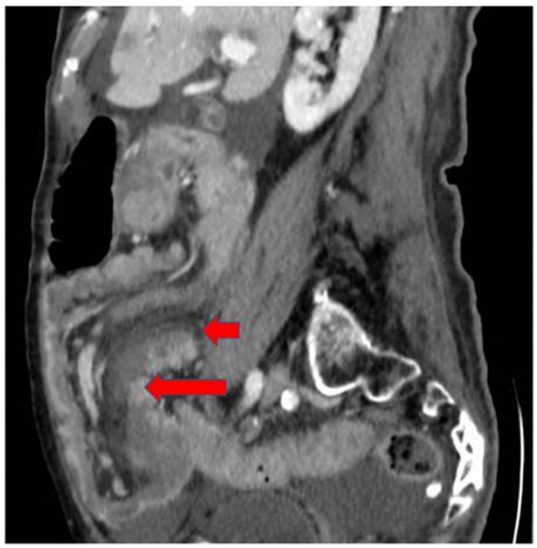

Fig. 1 CT scan showing the presence of ileocecal intussusception in the sagittal plane (red arrows)

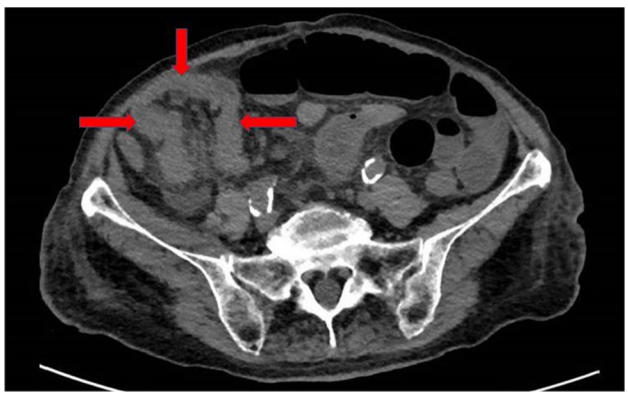

Fig. 2 CT scan showing the formation of target sign in right iliac fossa (red arrows)

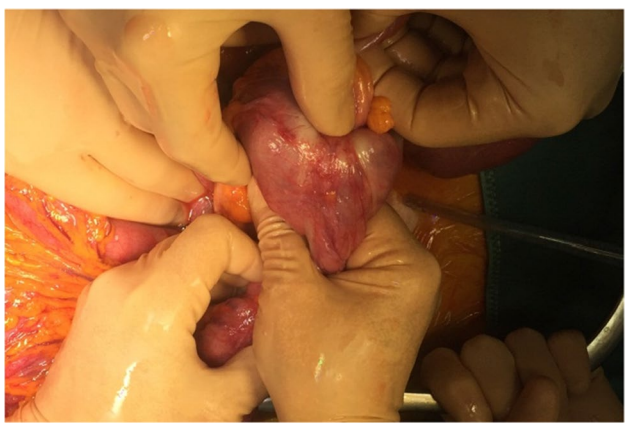

Fig. 3 Manual reduction of the intussusceptions

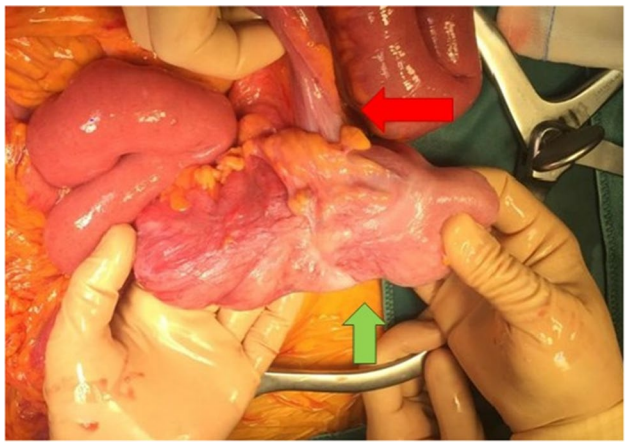

Fig. 4 Healthy portion of invaginated ileum (red arrow) in cecum (green arrow)

12 in good conditions. Histo-pathologically, the mass was diagnosed as an ulcerated, moderately differentiated (G2) intestinal adenocarcinoma with focal mucinous aspects and full-thickness infiltration of ileocecal valve and colon wall with initial extension to the perivisceral adipose tissue (pathological stage: pT3N0M0.)

Discussion

Intussusception of the bowel is defined as the telescoping of a proximal segment of the intestinal tract (intussusceptum) within the lumen of the adjacent distal segment (intussuscipiens). This condition was initially described in 1674 by Barbette from Amsterdam and presented in 1789 by John Hunter as "*introssusception*," a rare form of bowel obstruction [1]. Intussusception has traditionally been described as a childhood condition and a rarity in adults, accounting for 1% of all bowel obstructions with only 5% of all intussusceptions occurring in adults [2–4]. In contrast to childhood intussusception, 70–90% of adult intussusception has a demonstrable etiology [5–9]. Invagination in adults is usually secondary to an anatomic lesion including benign or malignant tumor (80–90% of cases), inflammatory lesions and foreign bodies of the small intestine. Though enteric, ileocolic and ileocecal invaginations are usually due to benign lesions, in contrast colo-colic invaginations are usually caused by malignancies [2–5, 10, 11]. Adenocarcinoma is the most common malignant cause in the colon, whereas metastases and lymphomas are the leading malignant etiologies in the small intestine. Benign tumors are frequently lipomas [12]. Intussusception occurs most frequently between the fourth and seventh decades of life; the mean age for intussusception due to secondary malignancy is in the 6th decade, whereas for intussusception associated due to benign

tumors usually occurs in the 4th decade. Incidence is similar between men and women [13]. Chronic symptoms and colonic-type intussusception are predictive factors for malignancy [14]. Though the exact mechanism is still unknown, it is believed that any lesion in the bowel wall or within the lumen modifying normal peristaltic activity can initiate the process of intussusception. Invaginations have been classified into four categories according to their location: entero-enteric, colo-colic, ileo-colic and ileo-cecal [8]. From a clinical perspective, the classic pediatric triad of abdominal pain, palpable abdominal mass and bloody stool is rarely seen among adults, since in most cases intussusceptions manifest with chronic intermittent cramping abdominal pain associated with nonspecific signs of bowel obstruction such as nausea, vomiting, constipation, or abdominal distention, with consequent delay of diagnosis [5, 8, 15–18]. Preoperative diagnosis requires standard imaging: ultrasonography is considered a useful method with key findings including the "target" or "doughnut" signs in the transverse view and the "pseudo-kidney" or "hay-fork" sign in the longitudinal view. Computed tomography is the most sensitive diagnostic method with imaging characterized by a "target" or "sausage shaped" soft mass with a layering effect [12, 19–26]. CT scans also define the extent of the occlusion, informing the clinician on the degree of surgical urgency and providing information regarding the existence of intestinal ischemia or perforation [16, 22]. Colonoscopy is a resource to be considered in cases of CT-diagnosed colonic and colo-rectal intussusception, in order to obtain a preoperative diagnosis such as malignancy, that will guide the surgical approach. Generally, colonoscopy is performed for both diagnostic and therapeutic indications. First of all, it is the gold standard for colonic cancer screening and surveillance: the age at which screening starts depends upon the patient's medical and family history and, if polyps are found during the procedure, they should be removed endoscopically if possible. Colonoscopy is also indicated in case of patients with recent hematochezia, positive fecal occult blood, or melena causing bleeding when an upper gastrointestinal source has been excluded. In addition, unexplained iron deficiency anemia should be evaluated with colonoscopy, as colon cancer is an important cause of iron deficiency anemia in adults and it should be performed in patients with chronic, clinically significant diarrhea without an explanation to rule out microscopic colitis. Furthermore it is indicated to evaluate abnormalities on imaging studies, the presence of synchronous or metachronous malignancy in patients with colon cancer and the extent and/or severity of inflammatory bowel disease. Finally, therapeutic indications for colonoscopy include foreign body removal, decompression of sigmoid volvulus or colonic pseudo-obstruction, balloon dilation of strictures, palliative treatment of bleeding or stenosed neoplasms, and percutaneous endoscopic cecostomy tube placement [27–29].Therapeutic strategy depends on several variables [18, 30]. Asymptomatic intussusception does not require treatment. Although the vast majority of cases require operative treatment, initial non-operative therapy with Gastrografin® enema can be attempted in subjects with ileo-colonic or colonic-type intussusception if no malignancy is suspected or in high-risk patients with high-grade obstruction [31]. The enema is generally used for diagnostic / therapeutic purposes in volvolus, intussusceptions and meconium ileus and can use gastrografin, barium sulfate and tap water as agents; in this case Gastrografin® was used as it has a greater hygroscopic action and is safer than barium in a pre and postoperative context [32, 33]. Within the category "non operative surgery" there are also (a) the use of water-soluble contrast agent, an effective treatment for the adhesive small bowel obstruction [34], and (b) colonic stenting, performed as a bridge to surgery in patients with acute mechanical obstruction by providing preoperative decompression or as a palliation in those with advanced disease [35, 36]. Laparotomy is required for the treatment of most cases among adult symptomatic intussusceptions since malignant lesions are present in a substantial proportion of patients [2, 3, 6, 7, 12, 16, 31, 37–40]. Intussusception associated with bowel obstruction frequently requires an urgent surgical solution; if there are signs of intestinal ischemia, a resection of the affected segment should be performed [17] although if there is absence of vascular impairment, the surgeon should try to reduce the invagination, then performing the selective resection of the causal element in order to avoid a larger intestinal resection. The latter is necessary when the primary pathology requires a resection, as in the case of malignant tumors. Nevertheless, most authors advise to avoid reduction of the intussusception if the presence of a neoplasm cannot be excluded; in this case it is preferable to carry out an intestinal resection, including the intussusception, according to the oncologic criteria used for adenocarcinomas [15, 41]. The main issue is that in most cases preoperative endoscopic examination is not able to define the cause of invagination. In the case of malignant tumor, few authors feel that excessive tumor manipulation could disseminate the malignancy [17, 39, 42–44]. Ileo-colic intussusception in adults is rarely caused by adenocarcinoma, with only a few cases described in the recent literature. In our review, we identified eleven cases (Table 1) of ileocolic intussusception due cecal or ileocecal valve malignancy, comparing them with the one reported in the present case. Our case is unusual for two reasons: (1) the clinical presentation of our adult patient was characterized by blood-tinged diarrhea and vomiting, elements more commonly found in pediatric population: in this case the presence of blood in the stool may be due to the suffering of the intestinal wall with subsequent ischemia and detachment of the mucosa (as it happens in the "red jolly" stools of children [45]) or to ulcerated adenocarcinoma; and

Table 1 Eleven cases of ileocolic intussusception due to adenocarcinoma of the cecum or of the ileocecal valve described in the English medical literature (NA = not available)

References	Age and gender M = male F = female	Symptoms	Radiology	Treatment	Extension of intussusception	Cause
Shioda et al. [26]	27-year F (Pregnant)	Abdominal pain, nausea, vomiting	Abdominal US	Right hemicolectomy	NA	Adenocarcinoma
Erbil et al. [10]	NA (adult)	Abdominal pain	CT scan, colonoscopy	Right hemicolectomy	NA	Adenocarcinoma
Takenoue et al. [46]	73-year F	Constipation, anorexia	Abdominal US, CT scan, barium enema	NA	NA	Adenocarcinoma
Cheung et al. [16]	72-year F	Abdominal pain	CT scan	Right hemicolectomy, partial ileal resection	NA	Adenocarcinoma
Marsden et al. [9]	69-year F	Abdominal pain	X-rays, CT scan	Laparoscopic right hemicolectomy	Extending to splenic flexure	Mucinous Adenocarcinoma
Verre et al. [39]	74-year F	Abdominal pain, nausea, vomiting	CT scan	NA	Extending to ascending colon	Adenocarcinoma
Mrak [17]	40 year M	Abdominal pain, nausea, vomiting	X-rays, abdominal US, CT scan	Extended right hemicolectomy	Extending to transverse colon	Adenocarcinoma
de Clerck et al. [18]	56-year M	Abdominal pain	Abdominal US, CT scan	Right hemicolectomy	NA	Adenocarcinoma
Elm'hadi et al. [42]	30-year F	Abdominal pain, nausea, vomiting	Abdominal US, CT scan	Laparoscopic right hemicolectomy	NA	Adenocarcinoma
Gruenberger et al. [40]	61-year M	Abdominal pain, distension, anorexia, vomiting	CT scan	Right hemicolectomy, Ileal resection (15 cm)	Extending to splenic flexure	Mucinous Adenocarcinoma
De Mesquita et al. [11]	44-year M	Abdominal pain, dyspeptic symptoms	CT scan	Laparoscopic right hemicolectomy	NA	Mucinous Adenocarcinoma

(2) the intussusception had a length of 45 cm, reaching the mid transverse colon, a distinctly unusual finding [9, 17, 40, 46]. As a result, due to this condition, it was decided to proceed with a manual reduction of intussusception in an attempt to prevent the development of a postoperative short bowel syndrome in an elderly patient, despite the possibility of dissemination of malignant cells during this attempt. Our case differs from the majority of those described in the English literature due to the attempt to reduce the invagination preoperatively despite the lack of a preoperative diagnosis of neoplasia. Sarr et al. from the Mayo clinic [30] questioned the accepted notion that preoperative reduction of intussusception was not recommended due to its association with malignant spread, since tumor cells are always shed from primary lesions, and the reduction of intussusception produces little damage to the intestinal mucosa. They believe that an attempt to solve the invagination with conservative therapy may be useful in order to avoid emergency surgery without upgrading the tumor. If unsuccessful, intraoperative reduction of the invagination is a useful maneuver in order to reduce the extent of the resected bowel without affecting the oncologic stage [17, 18, 30].

Key Messages

- Intussusception is a rare condition and should be considered in the differential diagnosis of bowel obstruction even in an adult patient.
- In case of colonic or ileo-colic intussusception, an attempt at reduction with a Gastrografin® enema should be considered, in particular in high-risk patients.
- Preoperative colonoscopy should be considered for CT-diagnosed colonic and colo-rectal intussusception in order to assist surgical planning with a preoperative diagnosis
- Intraoperative manual reduction of intussusception in selected cases can reduce the length of resection, reducing the occurrence of short-bowel and related syndromes.

Compliance with Ethical Standards

Conflict of interest None of the authors have any conflicts of interest to disclose pertaining to the study.

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