



Review Article

Surprising Complication of Intussusception after Colonoscopy: A Case Report and A Review of the Literature

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Abstract

This study discusses a case with a history of ileal-caecal intussusception post colonoscopy requiring urgent surgical intervention and a systematic review explores the literature on post-colonoscopy intussusception. A systematic review was conducted according to PRISMA guidelines. Studies reporting entero-enteric, ileo-colic, or colo-colic intussusception after colonoscopy, published in English before June 2023, were included. Data was extracted on patient demographics, indications for colonoscopy, procedural details, clinical symptoms, and management. Overall, 19 cases were identified from 17 studies. The median age was 48 years and 53 % were male. History of abdominal surgery was reported in 42% of cases. Symptoms typically appeared within a week of the procedure. The majority of cases required surgical intervention (63%), while others were managed conservatively (34%). The case report is an 85-year-old man with a history of diabetes, cardiopathy and abdominal surgery presented to the Emergency Department with 2 days of abdominal pain and vomiting after colonoscopy. A Computer Tomography abdomen revealed findings concerning for a small-bowel volvulus. Subsequent laparotomy revealed ileo-caecal intussusception, requiring surgical resection. Histology examination revealed a small-bowel intramucosal adenocarcinoma. The patient recovered and was discharged on postoperative day 7. The etiology of post-colonoscopy intussusception is multifactorial, with potential contributing factors including adhesions, altered bowel motility, and gas insufflation. Patients with a history of abdominal surgery should be considered at increased risk and prompt identification is crucial to reduce morbidity and mortality.

Keywords: Abdominal pain; Colonoscopy; Ileal tumor; Intussusception

Abbreviations: M: Male; F: Female, PPES: Post Polypectomy Electrocoagulation Syndrome

Introduction

Intussusception is defined as the invagination of one segment of the bowel (the proximal tract is the intussusceptum) into an immediately adjacent segment (the distal segment or the intussusciptens). This action of telescoping often obstructs the

passage of food or fluids. Additionally, it stops the blood supply to the affected area of the intestine, leading to possible infection, tissue necrosis, or intestinal perforation [1]. Intussusception is the most common cause of intestinal obstruction in early childhood, whilst in adults is infrequent, being approximately 5% of all intussusception cases, and typically more frequent in the small bowel rather than in the colon [2]. Among adults, intussusception commonly arises from a pathological "lead point" (which can be found intraluminal, mural or extramural), such as malignant lesions, adhesions, benign tumors, inflammatory lesions and Meckel's diverticulum [1]. Intussusception can also occur as a

complication of colonoscopy, although this condition is very rare and poorly described in the literature. Worldwide, colonoscopy is a safe procedure used for diagnosis, screening, surveillance, and treatment. The incidence of complications after colonoscopy is low and varies between diagnostic and operative colonoscopy. Globally, the incidence rate of perforation, post-colonoscopy bleeding, and mortality is reported to be 0.05%, 0.26%, and 0.0029%, respectively [3]. An important distinction within the context of colonoscopy, that is, the differentiation between diagnostic and operative colonoscopies. Diagnostic colonoscopies aim to visualize the colon and rectum to identify abnormalities, obtain biopsies, or perform other non-surgical interventions. In contrast, operative colonoscopies involve therapeutic procedures beyond mere visualization and diagnosis. These may include the removal of polyps or treatment of bleeding lesions. Operative colonoscopies are more invasive and some complications (e.g. bleeding, perforation and intussusception) are direct consequences of resections during operative endoscopic procedures like polypectomies, mucosal resections, and submucosal dissections, whilst other complications are less common and with mechanisms not completely known. Moreover, there's no agreement concerning the temporal definition of a colonoscopy-related complication: some studies consider an event associated with the endoscopic procedure up to 7, 14 or 30 days after colonoscopy [4]. Indeed, the presence of complications up to 30 days after the colonoscopy could have a cause-and-effect relationship with the endoscopic procedure [4].

On the other hand, in patients with subacute or chronic intestinal obstruction, the colonoscopy can be an important diagnostic tool for the diagnosis of ileo-colic or colo-colic intussusception [5]. Furthermore, colonoscopy can also have a role as a potential treatment for intussusception in children. The most common clinical manifestation of intussusception is abdominal pain with associated symptoms like nausea, vomiting, gastrointestinal bleeding, constipation, or bloating. Commonly a computed tomography scan (CT) is recommended for the diagnosis. In contrast to children, abdominal surgery with bowel resection can be a valid treatment to manage intussusception-related obstruction in adults [1]. We report a case of post-colonoscopy ileo-caecal intussusception requiring urgent surgical intervention and a systematic review of current literature about intussusception post-colonoscopy.

Materials and Methods

This systematic review was performed according to the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines [6,7]. To assess the association of intussusception after colonoscopy, two electronic databases, MEDLINE through PubMed and Embase, were searched up to June

26, 2023, using the following search query: “((((intussusception) OR (invagination)) AND (after colonoscopy)) OR (post colonoscopy)) AND (complication)”.

Study Selection

Case-report and case-series studies regarding history of entero-enteric, ileo-caecal, ileo-colic or colo-colic post colonoscopy intussusception that have been published before June 26, 2023, were retrospectively included. Studies focusing on colonoscopy-related complication that did not refer to intussusception were excluded. Moreover, studies with unavailable or unextractable data, studies published in languages other than English, letters, comments, studies involving children, and duplicate publications were likewise excluded.

Search Strategy, Data Identification and Extraction

The research was independently conducted by three authors (IL, AR, CS). The initial screening included independent evaluations of titles. After that, the three independent reviewers assessed the abstracts of the selected titles for inclusion. A full-text analysis and data-extraction was finally performed on the remaining studies. In case of disagreement, consensus is reached through discussion among reviewers during each of the previous phases. The reference sections from all selected studies were additionally examined to discover any further relevant paper. The following features were extracted from each study: first author, year of publication, country. For each patients, the following data were collected: age, sex, previous surgical history, indication of colonoscopy, procedural details, clinical symptoms, onset of symptoms, the intussusception type, cause of intussusception, and management.

Results

The search identified a total of 745 articles. Twenty-nine studies were excluded because they were published in languages other than English. The flow chart describing the process of study selection is shown in Figure 1. From a first screening, based on titles, 25 articles were included for abstract and full-text review. Among these studies and from the analysis of reference sections of pertinent systematic reviews [8,9], 17 articles were finally included [8-24]. Eight studies were excluded because they didn't meet the eligibility criteria. Sixteen out of the 17 articles included were case reports [8-10,12-24], and one was a case series [11], with a total of 19 patients. Of these 19 patients, 53% (10/19) were males with a mean age of 48 (range 19-73) years old. Among these patients, a history of abdominal surgery was reported only in 8 patients (42%): three had surgery for colorectal cancer, one for jejunal carcinoid, one for splenectomy, one for cesarean delivery, one for endometriosis and appendicectomy, one for perforated ulcer. As shown in Table 1 the clinical question for colonoscopy

differed between the studies analyzed, being abdominal pain, the most frequent indication (21%) followed by chronic diarrhea (16%) and polyp resection (16%). Other indications were colon cancer follow-up, rectal bleeding, screening, iron deficiency anemia, family history of colon polyps, family history of colon cancer, personal history of colon polyps, and assessment of colostomy closure. In all cases, colonoscopy was conducted to the cecum. Concerning procedures eventually performed during the endoscopic procedures, polypectomies and biopsies were performed in 8 and 5 patients respectively, whilst endoscopic submucosal dissection was performed in one patient, and one patient with a 4 cm polyp of terminal ileum was referred to surgery.

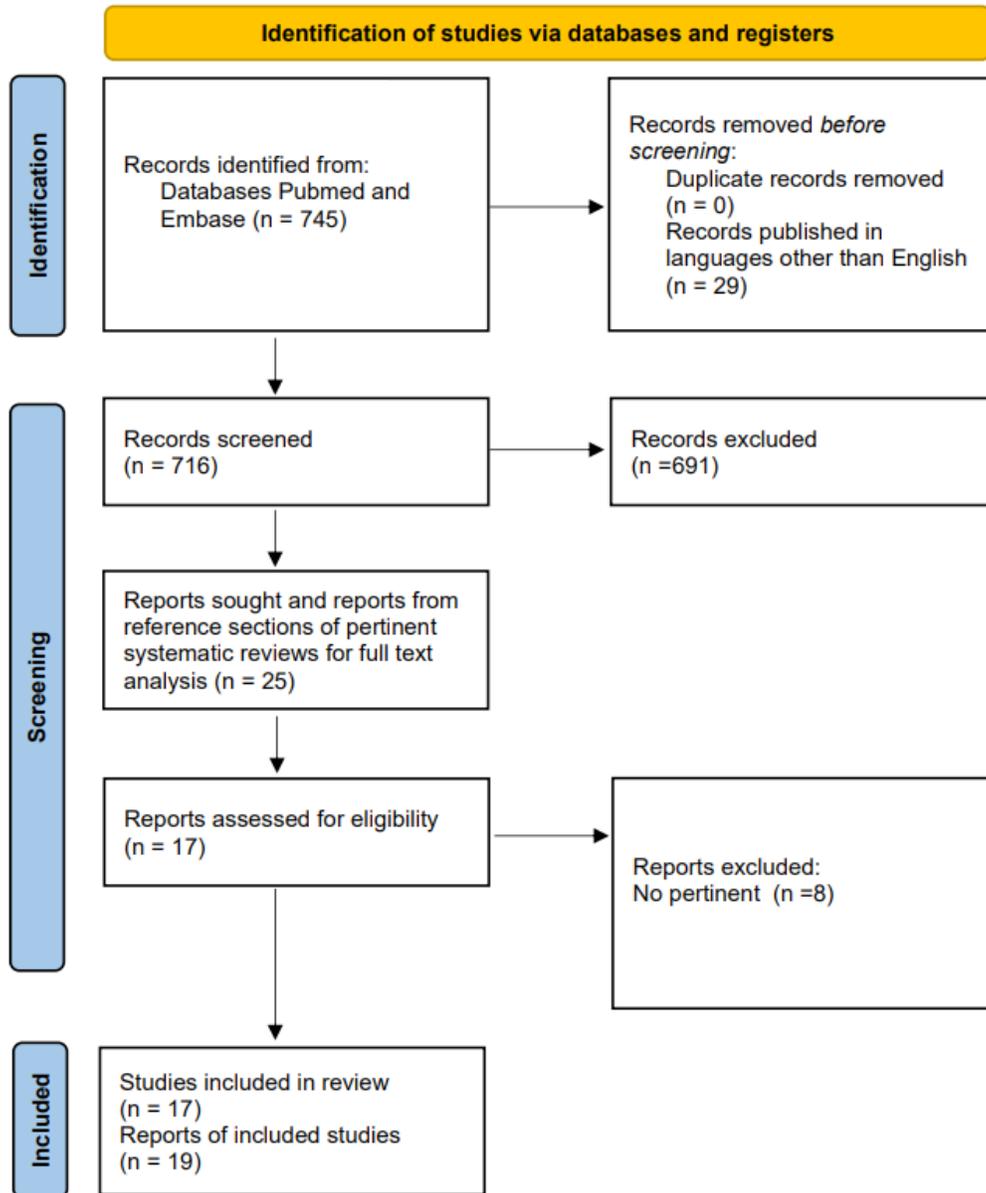


Figure 1: Flowchart of included studies.

Author	Country	Year	Sex, Age (yrs)	Prior surgical history	Indication to colonoscopy	Procedural details	Onset and symptoms	Cause of intussusception	Site and type of intussusception	Management
Keefe	USA	1985 (10)	M, 36	subtotal colectomy	Colon cancer follow-up	Colonoscopy	4 days after abdominal pain, nausea and vomiting	Adhesions from previous surgery and/or distension from air insufflation	Small bowel	Laparotomy with resection
Malki	Australia	2001 (11)	M, 75	Small intestine resection	Constipation	Ileo-colonoscopy and polypectomy	2 days after abdominal pain, vomiting and constipation.	Not specified	Small bowel	Laparotomy with resection
Malki	Australia	2001 (11)	M, 73	Colon resection	Colon cancer follow-up	Colonoscopy	3 days after nausea, vomiting, abdominal pain, and distension.	Not specified	Small bowel	Conservative
Malki	Australia	2001 (11)	M, 51	Colon resection	Colon cancer follow-up	Colonoscopy and polypectomy	1 day after abdominal pain and vomiting	Not specified	Small bowel	Conservative with nasogastric tube decompression
Zanati	Canada	2005 (12)	M, 47	Splenectomy	diarrhea and abdominal pain	Colonoscopy and random biopsies	Suddenly after colonoscopy abdominal pain	sympathetic overstimulation, excessive air insufflation and opioid effects	Small bowel	Conservative
Theodoropoulou	Greece	2009 (13)	M, 19	No previous surgical history	Abdominal pain	Ileo-colonoscopy and enteroscopy	7 hrs after abdominal pain	Hyperperistalsis	Ileo-colic	Laparotomy with right hemicolectomy
Ho	USA	2010 (14)	M, 32	No previous surgical history	Family history of colon polyps	Colonoscopy and polypectomy with cold snare	1 day after abdominal pain and vomiting	mucosal edema postpolypectomy	Colo-colic intussusception ascending colon	Laparoscopic reduction
Nachnani	USA	2012 (15)	F, 73	Not reported	Chronic diarrhea	Colonoscopy and random biopsies	1 day after abdominal pain	Hyperperistalsis or some complex loops	Colo-colic intussusception hepatic flexure	Laparoscopic reduction
Lasithiotakis	Greece	2012 (16)	F, 58	Not reported	Rectal bleeding	Colonoscopy (4 cm polyp of ileum send to surgery)	8 hrs after abdominal pain, nausea, vomiting and bloody diarrhea	Aspiration of insufflated air, peristalsis of small intestine, relaxation of ileocecal valve	Ileo-caecal intussusception	Right hemicolectomy
Lee	Korea	2013 (17)	M, 47	Not reported	Polypectomy	Colonoscopy and hot snare polypectomy	12 hrs after abdominal pain	Bowel edema secondary to polipectomy	Caeco-colic intussusception	Right hemicolectomy
Araki	Japan	2017 (18)	F, 43	Not reported	Polypectomy	ESD	24 hrs after bloody stool	Not specified	Caeco-colic intussusception	Colonoscopic reduction
Min	USA	2017 (19)	F, 31	Appendicectomy and low anterior resection	Abdominal pain and constipation	Colonoscopy and random biopsies	1 day after abdominal pain	Intestinal dysmotility and previous surgical history	Ileo-colic	Ileo-colic resection
Hassan W	Australia	2018 (8)	M, 28	No previous surgical history	Iron deficiency anemia	Ileo-colonoscopy and random biopsies	2 hrs after abdominal pain, nausea, vomiting, and diarrhea, bloody stool	Aspiration of gas during withdrawal of the colonoscope created a vacuum effect	Colo-colic (ascending colon to the hepatic flexure)	Conservative
Papastergiou	Greece	2018 (9)	F, 42	Two cesarean section deliveries	Family history of colon cancer	Colonoscopy	6 hrs after abdominal pain and nausea	Closed-loop strangulation of the terminal ileum by a postoperative adhesive band	terminal ileum	Laparotomy with resection
Ahmed	USA	2020 (20)	F,42	No previous surgical history	Diarrhea	Colonoscopy and random biopsies and polypectomies	Few hrs after abdominal pain	Not specified	colo-colic intussusception	Conservative (antibiotic therapy)
Moon	Korea	2022 (21)	F, 58	No previous surgical history	Polypectomy	Colonoscopy and polypectomies	1 day after abdominal pain and fever	Colon wall thickening due to PPES	Colo-colic intussusception (right colon)	Conservative (antibiotic therapy)
Luciano	USA	2022 (22)	M,72	Rectosigmoidectomy colostomy	Evaluation of colostomy reversal	Colonoscopy	1 week after swelling and tenderness of the stoma	Hyperperistaltic state and vacuum effect induced by aspiration of gas	Colo-colic intussusception of the proximal colon into colostomy	Surgical reduction, colostomy reversal and coloproctostomy
Vadakkenchery	Australia	2022 (23)	F,36	No previous surgical history	Abdominal pain	Colonoscopy	1 day after abdominal pain, nausea and vomiting, diarrhea	Secondary to mucosal edema	Colo-colic near the distal transverse colon and splenic flexure	Laparoscopic reduction
Jastaniah	Canada	2023 (24)	F,50	No previous surgical history	Personal history of polyps	Colonoscopy and polypectomy	2 hrs after abdominal pain	Bowel edema secondary to polipectomy	Ileo-colic intussusception	Right hemicolectomy

Table 1: the main characteristics and results of included studies in the systematic review.

In terms of clinical manifestation, symptoms started from 2 hours to 7 days after the endoscopic procedure. Specifically, 9 patients complained of nausea, vomiting and abdominal pain (47%), 7 patients complained of abdominal pain solely (37%), two patients presented abdominal pain associated with rectal bleeding (10%), and one patient presented a decreased colostomy output (5%). Different hypotheses about the etiology of intussusception were reported by the authors: an alteration of bowel motility due to air insufflation “hyperperistalsis” (six cases, 32%), mucosal edema secondary to polypectomy (three cases, 16%) or secondary to localized ischemia (one case, 4%), or adhesions from previous surgeries (three cases, 16%). Finally, for 6 cases (32%) the authors didn’t provide any hypothesis. The site of intussusception was more frequently colo-colic (seven out of the nineteen patients, 37%), while it was ileal in 6 patients (31%), ileo-caecal in four patients (21%), and caeco-colic in two patients (10%). Nine cases (47%) were managed with abdominal surgery. Among these four had a right hemicolectomy, three had ileum resection, one had ileocolic resection, and one had a surgical reduction. Other cases were treated with conservative management (31%) and laparoscopic reduction (16%). One patient was successfully treated with endoscopic reduction (4%). None of the reported cases died from this complication.

Case Report

The patient is an 85-year-old man that presented to our department for a scheduled colonoscopy. The endoscopy procedure was performed to investigate a right colonic Positron Emission Tomography (PET) uptake for oncological follow-up. Past medical history included previous distal splenic-pancreatectomy and Radiotherapy (RT) for pancreatic adenocarcinoma, ischemic cardiopathy, diabetes, surgical intervention for squamocellular hard palate tumor and installation of a medullary stimulator for neuropathy. Colonoscopy was conducted until the caecum with a sufficient bowel preparations (Boston scale 6: 2+2+2) and showed some diverticula and hemorrhoidal congestion (Figure 2). The procedure proved to be technically demanding due to the severe stiffness of the sigmoid. A few hours after the colonoscopy, the patient presented again to our department with diffuse abdominal pain associated with nausea. Initially, on abdominal examination, he had diffuse mild pain on superficial and deep palpation and a negative Blumberg sign, so the patient was discharged. After two days, he had persisting diffuse abdominal pain and vomiting so he went to the Emergency Room of our hospital. On examination, he had 54 beats/min and a normal temperature (36°C). He had right abdominal tenderness, negative Blumberg sign and a mass of increased consistency was noted in the hypogastrium and right iliac fossa. Laboratory findings showed leukocytosis of $12.7 \times 10^9/L$ (normal range, $4.3-10.8 \times 10^9/L$) and a C-reactive protein level of 2.05 mg/dL (normal range, 0.0-0.5 mg/dL). Abdominal CT revealed rotation of some ileal loops associated with thickening of intestinal walls, a convoluted appearance of mesenteric vessels and intestinal pneumatosis, to be referred to volvulus. CT revealed distension of proximal jejunoileal loops and no evidence of pathological distension of colic loops. No obvious mass lesion was appreciable on CT (Figure 3). He was managed with urgent surgery. Laparotomy revealed ileo-caecal intussusception, manually not reducible, and adherences that were cut. The last ileal loop and the ascending colon were removed with the construction of a lateral-lateral ileocolic anastomosis (Figure 4). Macroscopically, the pathologist confirmed ileo-caecal intussusception, describing a 2 cm exophytic formation proximally to the invaginated ileal tract. Histology showed small-bowel tubular adenoma with high-grade dysplasia/intramucosal adenocarcinoma (pTis). Invaginated ileum was necrotic. Caecum walls were characterized by mucosal chronic inflammation, cryptic microabscesses and distortion of

glandular architecture. Margins of surgical resection were free from pathological alterations (R0). The patient continued to improve with normalizing inflammatory markers, he fed in the absence of complications and was discharged home 7 days later.

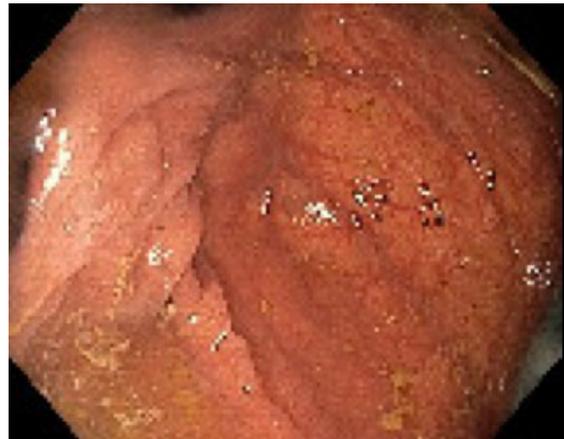


Figure 2: Caecum picture obtained during colonoscopy showed a sufficient intestinal cleansing with aspirable liquid residues and normal mucosal and vascular pattern.

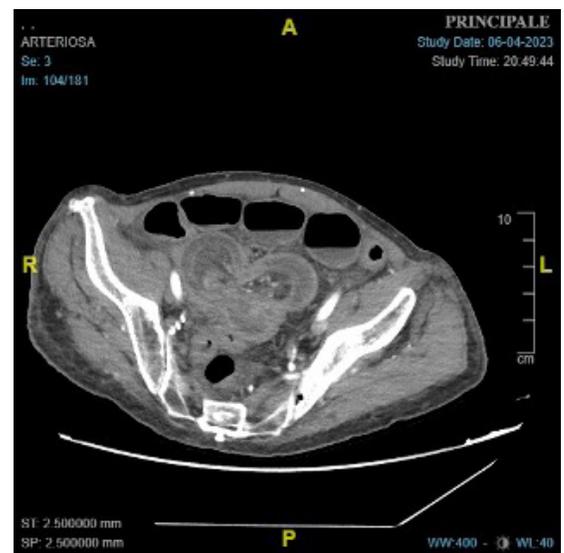


Figure 3: CT scan showing swelling in the proximal jejunoileal loops and no signs of abnormal distension in the colic loops.



Figure 4: Operative specimen. An ileocecal resection was performed for ileocolic intussusception.

Discussion

Post-colonoscopy intussusceptions can be classified into two primary categories: secondary and idiopathic. Secondary intussusceptions are intussusceptions that result from a specific leading point, as previously described. Identifying the leading point is crucial in understanding the etiology and management of secondary intussusceptions, as in our case report. On the other hand, idiopathic intussusceptions have no clear identifiable cause other than the fact that they occur after the colonoscopy procedure. These cases pose a diagnostic challenge as the underlying mechanisms are less apparent. To our best knowledge, this is the first case of a patient with ileocolic intussusception after colonoscopy for an ileal adenocarcinoma. We conducted a systematic review of the literature to highlight the current hypothesis that may explain the etiology/nature of this rare condition.

The other case reported in literature showed different hypotheses to explain the occurrence of intussusception after colonoscopy. One hypothesis suggested that mucosal edema or ischemia resulting from polypectomy may cause a lead point for intussusception [14,17,21,23,24]. However, due to the merely diagnostic nature of our colonoscopy, this hypothesis is not reliable to explain our case. Another hypothesis proposed that the lead point could arise following post-surgery adhesions [9,10,19]. This hypothesis fits well with most of the examined cases, that, similarly to our case, were associated with a prior history of abdominal or pelvic surgery. Given this evidence, the occurrence of adhesions following surgical procedures seems to be the main cause of intussusception. Moreover, an alternative theory suggested that colon over insufflation during colonoscopy

may induce an alteration of bowel motility with a hyperperistaltic state which is known to be a trigger factor for intussusception [10,13,15,16,19,22,23]. Furthermore, the act of aspirating gas during colonoscope removal could potentially create a vacuum effect, which might also contribute to the onset of intussusception [8,16,22]. Other factors associated with colonoscopy factors that could similarly play a minor role in intussusception development, include procedure-related sympathetic system stimulation, and the use of sedatives with opioid properties [12]. Finally, minor predisposing factors concerning the anatomy and pathophysiology of the small intestine and colon are the peristalsis of the small intestine and the relaxation of the ileocecal valve [16].

In our case report, only the histological examination revealed the presence of a 2 cm intestinal adenocarcinoma in the ileum, which thus may have been the lead point causing the ileocolic intussusception together with the presence of post pancreas surgery abdominal adhesions. It is noteworthy that the PET scan had shown an uptake in the right colon, which was eventually found to be correct, albeit of a lesion at the ileal site.

Conclusion

Intussusception after colonoscopy is an extremely rare complication, little described/reported in current literature. The etiology is poorly known and it is usually activated by a combination of mechanisms and often associated with an organic pathology called “pathological lead point”. It can often present with abdominal pain and signs of bowel obstruction (i.e., vomiting). As regards its management, since bowel necrosis needs a timely surgical management, an intussusception early identification would allow to decrease related morbidity and mortality. For these reasons, in agreement with our results, patients with a history of intraabdominal or pelvic surgery, including gynecologic procedures, should be considered at increased risk of developing ileocolic intussusception after colonoscopy. However, to the best of our knowledge, our patient represents the first case of post colonoscopy ileocolic intussusception that revealed an ileal adenocarcinoma that have been successfully managed by surgery.

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