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Pa Final Diag	atient:	Female, 65-year-old	

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Final Diagnosis: Intussusception Symptoms: Diarrhea • fatigue **Medication:** _ **Clinical Procedure:** _ **Specialty:** Surgery **Objective:** Unusual clinical course Intussusception is a rare pathological entity in adults and remains a diagnostic challenge for clinicians, as it **Background:** shares many clinical signs and symptoms with other morbid conditions (including appendicitis, abdominal hernias, colic, volvulus, and Meckel diverticulum). High clinical suspicion and use of appropriate imaging techniques are essential for early diagnosis and treatment of intussusception. Surgical intervention is the treatment of choice in cases of sustained and persistent invagination. **Case Report:** We present the case of a 65-year-old woman with a medical history of Crohn's disease, diabetes mellitus type II, hypertension, and rheumatoid arthritis. She was hospitalized for diarrhea, fatigue, and anemia. Computerized tomography of the abdomen and a colonoscopy revealed telescoping of the ileum, ileocecal valve, and part of the ascending colon inside the terminal segment of the ascending colon. The antegrade ileocolic intussusception was treated by performing a right hemicolectomy. The pathologic examination of the excised intestine showed mucosal lesions compatible with Crohn's disease, an inflammatory fibroid polyp at the terminal section of the ileum, and a low-grade appendiceal mucinous neoplasm. **Conclusions:** Regardless of the etiology, when the normal motility of the intestine is altered, it can lead to invagination. Although intussusception is rare, it must always be part of the differential diagnosis for a patient presenting with constant abdominal pain. **Keywords:** Causality • Colonoscopy • Diagnosis • General Surgery • Intussusception • Signs and Symptoms

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Background

Intussusception is defined as the invagination of a section of the small intestine or large bowel into the lumen of its adjacent segment [1]. The telescoping of the proximal part inside the distal part is known as an antegrade intussusception and occurs in 95% of all cases [1]. In contrast, the term retrograde intussusception describes the introversion of the distal part of the intestine into the lumen of its adjoining proximal edge [2]. Any part of the gastrointestinal tract can be involved in telescoping; however, the most common sites are the small intestine, jejunum and ileum, and the large bowel [2,3]. Intussusception is a rare pathological entity [1-5]. It is estimated that telescoping is responsible for 5% of all cases of bowel obstruction [1-5] with a 1: 3 female to male ratio [5-8].

As intussusception shares many features with appendicitis, abdominal hernias, colic, volvulus, Meckel diverticulum, and ovarian torsion, an accurate diagnosis is difficult [2-5,9-11]. Furthermore, the diagnosis is challenging as most of the patients present with subacute or chronic symptoms [2-5,9-11]. Therefore, the clinical signs and symptoms are insufficient to establish a diagnosis of intussusception [1-5,9-11]. Currently, the main diagnostic modalities for evaluating invagination are ultrasonography (U/S) of the abdomen, barium enema, and computerized tomography (CT) of the abdomen [1-4,10-13]. Preoperative or intraoperative colonoscopy is useful to identify the lead point of the telescoping [2,10,14]. In most cases, the intussusception is transient and resolves spontaneously or with a colonoscopy [2,10,14]; however, sustained and persistent invagination necessitates surgical intervention [1-5,14].

 Table 1. Patient's hospital admission and preoperative blood results.

We report the case of a 65-year old woman whose persistent antegrade intussusception was treated with an urgent right hemicolectomy. A pathologic examination of the extracted section of the intestine, revealed the simultaneous presence of a large inflammatory fibroid polyp, a low-grade appendiceal mucinous neoplasm (LAMN), and mucosal lesions compatible with Crohn's disease. Each of these findings could be a potential cause of the intussusception.

Case Report

We present the case of a 65-year-old woman who was admitted to the hospital with diarrhea and fatigue. At admission, her vitals were body temperature 37° C, heart rate 80 beats/min, respiratory rate 17 breaths/min, and blood pressure 125/80 mmHg. She was obese (body mass index 33 kg/m²) and did not smoke or consume alcohol.

Her medical history included Crohn's disease, diabetes mellitus type II, hypertension, and rheumatoid arthritis. Her medications included linagliptin, perindopril, and prednisolone with hydroxychloroquine for the treatment of diabetes mellitus type II, hypertension, and rheumatoid arthritis, respectively. She was under surveillance for Crohn's disease without any treatment. She had no history of surgical interventions.

The physical examination revealed sensitivity in the right lower quadrant of the abdomen without tenderness and she had hyperactive bowel sounds. There was no blood or palpable mass observed on the digital rectal examination. A clinical examination of the cardiopulmonary and urogenital systems showed no abnormal signs, and except for the diarrhea and fatigue,

Markers	At admission	Preoperative	Normal range
Hemoglobin (g/dL)	7.6	9.6	11.9-14.7
Hematocrit (%)	25.8	30.6	36.8-45.6
White blood cells (per mL)	14,640	14,680	3,800-10,500
Platelets (per µl)	383,000	390,000	150,000-400,000
Urea (mg/dL)	28	29	15-50
Creatinine (mg/dL)	0.81	0.70	0.7-1.3
Serum glutamic oxaloacetic transaminase (U/L)	10	15	10-40
Serum glutamic pyruvic transaminase (U/L)	10	11	10-35
Gamma glutamyl transferase (U/L)	15	17	0-30
Sodium (mmol/L)	139	131	136-145
Potassium (mmol/L)	4.6	4.1	3.5-5.1
C-reactive protein (mg/dL)	4.07	3.71	<0.5

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Figure 1. Abdominal computed tomography scan: The red arrow shows invagination of the ileum, ileocecal valve, and part of the ascending colon inside the terminal section of the ascending colon. (Image processed with MS Paint, edition 6.1, 2009, Microsoft Corporation, Redmond, Washington, USA).



Figure 3. Image of the surgical specimen: The red arrow shows the point of invagination. (Image processed with MS Paint, edition 6.1, 2009, Microsoft Corporation, Redmond, Washington, USA).



Figure 2. Coronal image of the abdominal computed tomography scan: The green arrow shows the parts involved in the intussusception. (Image processed with MS Paint, edition 6.1, 2009, Microsoft Corporation, Redmond, Washington, USA).

there were no other remarkable findings in the physical and neurological examinations.

The blood tests showed an elevation of the inflammatory markers (white blood cells 14.640/mL and C-reactive protein 4.07 mg/dL) and anemia (hemoglobin 7.6 g/dL and hemato-crit 25.8%). The results for all the other markers were normal at hospital admission (Table 1).

Given the findings from the physical examination and the blood tests, a gastroscopy and a colonoscopy were performed 2 days after hospital admission. There were no abnormal pathologic findings in the upper gastrointestinal tract. In contrast, the colonoscopy revealed a large polyp extending from the ileocecal valve to the right colic flexure, giving the impression of ileocecal intussusception. To confirm this diagnosis of intussusception, an abdominal CT scan was performed and the results were in agreement with the endoscopy, demonstrating an invagination of the ileum, ileocecal valve, and part of the ascending colon inside the terminal section of the ascending colon (Figure 1). The coronal images of the CT scan clearly demonstrated the involved parts of the intussusception (Figure 2).

Due to the deterioration of her clinical status (vomiting, exacerbation of abdominal pain, and electrolyte disorders), she underwent an open laparotomy 3 days after her hospital admission. Intraoperatively, the diagnosis of intussusception was confirmed. An extended ileocolic invagination was recognized, with a large palpable mass inside the lumen of the ascending colon. A right hemicolectomy with an extended excision of the terminal ileum and an end-to-side ileotransverse anastomosis were performed. The overall length of the removed intestine was 47 cm. It was composed of 35 cm of the ileum and 12 cm of the cecum and ascending colon (**Figure 3**).

The postoperative period was uneventful and 6 days after the surgery, she was discharged in good condition. During her hospital stay (including after surgery), she was intravenously administered 3 g cefoxitin/day for 3 days, 1.5 g metronidazole/



Figure 4. Gross images of the polyps (All the images were processed with Adobe Photoshop CS3, Adobe Inc., San Jose, California, USA): (A) Adjacent to the ileocecal valve. (B) Close to the proximal margin. (C) White fleshy cut surface.

day for 3 days, 4 g paracetamol/day for 4 days, and 200 mg tramadol/day for 3 days. Postoperatively, she was administered intravenous fluids for 3 days and oral feeding from day 4.

Gross pathological examination of the excised colon revealed a large polyp originating from the terminal section of ileum (Figure 4A-4C). Due to its ample length (6 cm), it had projected into the ileocecal valve. The ileal mucosa had a cobblestone appearance, which is a pathognomonic sign of Crohn's disease (Figure 5A, 5B).

A microscopic examination of the surgical specimen indicated that the polypoid lesion arising from the submucosa had the characteristics of an inflammatory fibroid polyp (Figures 6A-6C, 7A-7D). An LAMN was an incidental finding (Figure 8).

After being discharged from the hospital, she had monthly follow-ups. Three months after the surgical intervention, she was in good clinical condition at her last follow-up (March 2021). There were no findings of a pathological entity in her chest and abdominal CT. Her next follow-up appointment is in 6 months.

This case report was conducted in accordance with the Ethical Standards of the Institutional and National Research Committee and the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards. The case report was approved by the Ethics Committee of the Scientific Board of Venizeleio General Hospital of Heraklion (Decision No. 50-3/ Meeting 5/22-04-2021). The patient gave written informed consent for the publication of this case report.

Discussion

We report the case of a rare pathological entity, an intussusception in a 65-year-old woman, who was hospitalized for fatigue, diarrhea, and anemia. The clinical examination revealed sensitivity in the right lower quadrant of the abdomen with hyperactive bowel sounds, and the blood tests revealed an elevation of the inflammatory markers and anemia. The clinical



Figure 5. (A) Inflamed terminal ileum mucosa proximal to the larger polyp (Image processed with Adobe Photoshop CS3, Adobe Inc, San Jose, California, USA). (B) Microscopic image of the terminal ileum mucosa proximal to the larger polyp revealed chronic active inflammation, submucosal involvement, and serositis, as seen in Crohn's disease (hematoxylin and eosin, 20×) (Image processed with Adobe Photoshop CS3, Adobe Inc, San Jose, California, USA).



Figure 6. Microscopic images of the polyps (All the images were processed with Adobe Photoshop CS3, Adobe Inc, San Jose, California, USA): (A) Submucosal mesenchymal lesion (hematoxylin and eosin, 20×). (B) Hypocellular neoplasm composed of spindle and stellate cells embedded in edematous stroma with admixed eosinophils and lymphocytes (hematoxylin and eosin, 40×).
 (C) Hypocellular neoplasm composed of spindle and stellate cells embedded in edematous stroma with admixed eosinophils and lymphocytes (hematoxylin and eosin, 100×).



Figure 7. (A) Neoplastic cells stained positive in CD34 (100×) (Image processed with Adobe Photoshop CS3, Adobe Inc, San Jose, California, USA). (B) Neoplastic cells stained negative in CD117 (400×) (Image processed with Adobe Photoshop CS3, Adobe Inc, San Jose, California, USA). (C) Neoplastic cells stained negative in S-100 (100×) (Image processed with Adobe Photoshop CS3, Adobe Inc, San Jose, California, USA). (D) Neoplastic cells stained negative in α-smooth muscle actin (400×) (Image processed with Adobe Photoshop CS3, Adobe Inc, San Jose, California, USA). (D) Neoplastic cells stained negative in α-smooth muscle actin (400×) (Image processed with Adobe Photoshop CS3, Adobe Inc, San Jose, California, USA).



Figure 8. Low-grade appendiceal mucinous neoplasm: Neoplasm composed of mucinous filiform/villous epithelium with low-grade cytology (hematoxylin and eosin, 100×) (Image processed with Adobe Photoshop CS3, Adobe Inc., San Jose, California, USA). Table 2. Most common causes of intussusception in adults.

Benign causes [references]	Malignant causes [references]
Appendiceal neoplasm [2,3,5,9-11,15-17]	Adenocarcinoma [2,3,5,9-11,15-17]
Celiac disease [2,3,5,9-11,15-17]	Carcinoids [2,3,5,10,11,15-17]
Crohn's disease [2,3,5,9-11,15-17]	Gastrointestinal stromal tumor [2,3,5,9-11,15-17]
Endometriosis [2,3,5,10,11,16,17]	Leiomyosarcoma [2,3,5,10,11,16]
Fibroepithelial polyps [2,3,5,9-11,15-17]	Lymphoma [2,3,5,10,11,15-17]
Henoch-Schönlein purpura [2,3,5,10,11,16,17]	
Leiomyoma [2,3,5,10,11,15-17]	
Lipoma [2-5,9-11,15-17]	
Meckel's diverticulum [2,3,5,9-11,15-17]	
Neurofibromatosis [2,3,5,9-11,16,17]	
Postoperative adhesions [2,3,5,9-11,16,17]	

Table 3. Common symptoms of intussusception.

Symptoms	References
Abdominal distension	[2,3,10,11,15,17]
Abdominal mass	[2-5,9-11,15,17]
Abdominal pain (most common)	[2-5,9-11,15,17]
Black stools	[2,3,5,10,11,15,17]
Change of bowel habits	[2,3,5,10,11,15,17]
Diarrhea	[2,3,5,9-11,15,17]
Fatigue	[2,3,10,11,15,17]
Nausea	[2,3,5,9-11,15,17]
Poor diet	[2,3,5,10,11,15,17]
Respiratory discomfort	[2,3,10,11,15,17]
Vomiting	[2-5,9-11,15,17]
Weight loss	[2,3,10,11,15,17]

signs and symptoms were not specific and insufficient to establish a diagnosis; therefore, a colonoscopy and an abdominal CT were performed. Both these investigations revealed an invagination of the ileum, ileocecal valve, and part of the ascending colon inside the terminal section of the ascending colon. Due to the deterioration in her clinical status, she underwent an open laparotomy, where a right hemicolectomy and an end-to-side ileotransverse anastomosis were performed. The pathologic examination of the surgical specimen simultaneously revealed the presence of a large inflammatory fibroid polyp and an LAMN. In addition to the presence of the polyp and LAMN, she had a history of Crohn's disease (under surveillance without treatment), which could be another etiologic factor that provoked the intussusception. It is extremely rare to have the synchronous presence of 3 possible etiologic factors for an intussusception.

Any part of the gastrointestinal tract can be involved in telescoping [2,3]. Intussusception is subdivided into 4 categories according to the topographic distribution of the engaged segments [3,4]. Intussusception is characterized as enteric, when only the jejunum or the ileum is involved; ileocolic, when the ileum and colon participate; colocolonic, when the colon is the only participating section; and sigmoidorectal, when the engaged sections are the sigmoid colon and rectum [3,4].

Numerous pathological entities, which modify the normal peristaltic movement of the intestine have been incriminated as the etiologic factors for intussusception [2-5,9-11]. Any reason, which alters the normal motility of the intestine can lead to invagination [2-5,9-11,15-17] The majority of cases of invagination in adults occur between the ages of 45 years and 55 years, with a 1: 3 (female to male) ratio. The causes that provoke intussusception can be benign or malignant. Neoplasms are responsible for 65% of all cases. Malignant neoplasms account for 66% of colonic intussusceptions and 30% of small intestine cases. Adenocarcinoma is the most common malignant lead point in the colon (Table 2). The pathophysiological explanation for an invagination is not yet fully understood; however, it is believed to be due to an imbalance in the longitudinal forces along the intestinal wall [8,9]. In many cases, the cause of this process is a malignant or benign mass acting as a lead point and disorganizing the pattern of peristalsis [8,9]. In the present case, both the fibroid polyp and LAMN could have been a lead point for the invagination. Our patient also had Crohn's disease, which is another potential etiologic factor for intussusception. The correlation between Crohn's disease and motility disorders has been established, although not fully explored, rendering this pathological entity as a third potential cause of intussusception. Wide experimental evidence shows that inflammation affects the motor and perceptive functions of the intestine [18]. At least part of this abnormal motility is due to

inflammatory cytokine trafficking and neuromuscular changes [19]. These effects are observed after resolution of the inflammatory phenomena [18]. This could be related to the fact that a complete mucosal healing is seldom achieved in patients with Crohn's disease and the inflammation is not limited to the mucosa, although it involves neuroenteric circuitry [18,20]. The synchronous presence of 3 etiologic factors is extremely rare. To our knowledge, this is the first time that the combination of these factors is simultaneously reported in a patient diagnosed with ileocolic intussusception.

Invagination is an uncommon pathological entity [1-5]. The majority of patients with invagination present with subacute or chronic symptoms, which are also seen in other diseases, and therefore, telescoping lacks specific clinical signs and symptoms [2-5,9-11,15,17] (Table 3). The most frequent clinical symptoms are abdominal pain, nausea, vomiting, diarrhea, fatigue, black stools, and weight loss [2-5,9-11,15,17]. In the case of our patient, 3 of these clinical symptoms were present.

Clinical examination alone is insufficient to establish the diagnosis of intussusception [1-5,9-11]. Therefore, abdominal CT and U/S, barium enema, and colonoscopy are essential investigations for the recognition of invagination [1-4,10-13]. U/S has a 60% diagnostic accuracy for bowel obstruction, and it increases significantly in the presence of a palpable mass [2,21,22]. A barium enema was used as a diagnostic tool in several studies, with a diagnostic accuracy of 25% to 45% [3]. CT has a diagnostic accuracy of 58% to 100% and is the criterion standard imaging technique [1,2,4,12,13]. A CT of the abdomen shows an intussusception as a soft tissue mass with an eccentrically located area of low density [1,2,4,12,13]. A CT is superior because it demonstrates the exact location and type of the telescoping, presence of a lead point, existence of complete bowel obstruction, and the presence of ischemic signs [1,2,4,12,13]. Additional information about possible

References:

- 1. Zubaidi A, Al-Saif F, Silverman R. Adult intussusception: A retrospective review. Dis Colon Rectum. 2006;49(10):1546-51
- 2. Wang N, Cui XY, Liu Y, et al. Adult intussusception: A retrospective review of 41 cases. World J Gastroenterol. 2009;15(26):3303-8
- Honjo H, Mike M, Kusanagi H, Kano N. Adult intussusception: A retrospective review. World J Surg. 2015;39(1):134-38
- 4. McKay R. Ileocecal intussusception in an adult: The laparoscopic approach. JSLS. 2006;10(2):250-53
- Paramythiotis D, Goulas P, Moysidis M, et al. Bowel intussusception in adults: A report of three interesting cases and current trends for diagnosis and surgical management. Hippokratia. 2019;23(1):37-41
- 6. Charles T, Penninga L, Reurings JC, Berry MCJ. Intussusception in children: A clinical review. Acta Chir Belg. 2015;115(5):327-33
- Lehnert T, Sorge I, Till H, Rolle U. Intussusception in children-clinical presentation, diagnosis and management. Int J Colorectal Dis. 2009;24(10):1187-92
- Potts J, Al Samaraee A, El-Hakeem A. Small bowel intussusception in adults. Ann R Coll of Surg Engl. 2014;96(1):11-14

lymphadenopathy and metastatic disease can be obtained from a CT scan [1,2,12,4,13]. Colonoscopy is a useful tool in cases of antegrade intussusception, as it can identify the leading point of invagination [2,10,14].

In the majority of cases, telescoping is transient and resolves spontaneously [2,10,14]. Studies show that colonoscopy can be used as a tool for the resolution of invagination in cases of benign lesions and extended intussusceptions [2,10,14]. However, surgery remains the most reliable intervention in case of consistent and persistent intussusception, as it offers the opportunity for definite and radical elimination of the cause, which provoked the introversion [1-5,14]. A pathologic examination of the surgical specimen can distinguish between benign and malignant lesions [3].

Conclusions

We report the rare case of an antegrade intussusception in a woman with 3 possible etiologic factors for invagination and she was treated with an urgent laparotomy. Although rare, this disorder should be recognized, examined, and treated appropriately in order to avoid fatal complications.

Department and Institution Where Work Was Done

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Declaration of Figures Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

- 9. Weilbaecher D, Bolin JA, Hearn D, Ogden W. Intussusception in adults. Review of 160 cases. Am J Surg. 1971;121(5):531-35
- 10. Shenoy S. Adult intussusception: A case series and review. World J Gastrointest Endosc. 2017;9(5):220-27
- 11. Marinis A, Yiallourou A, Samanides L, et al. Intussusception of the bowel in adults: A review. World J Gastroenterol. 2009;15(4):407-11
- 12. Ozan E, Atac GK, Akincioglu E, et al. lleocaecal intussusception with a lead point: Unusual MDCT findings of active Crohn's disease involving the appendix. Case Rep Radiol. 2015;2015:856483
- 13. Kim YH, Blake MA, Harisinghani MG, et al. Adult intestinal intussusception: CT appearances and identification of a causative lead point. Radiographics. 2006;26(3):733-44
- 14. Omori H, Asahi H, Inoue Y, et al. Intussusception in adults: A 21-year experience in the university-affiliated emergency center and indication for nonoperative reduction. Dig Surg. 2003;20(5):433-39
- Yakan S, Caliskan C, Makay O, et al. Intussusception in adults: Clinical characteristics, diagnosis and operative strategies. World J Gastroenterol. 2009;15(16):1985-89

- Ayyanar P, Behera G, Mishra TS, et al. The clinico-histopathological spectrum of tumors and tumor-like lesions in adult intussusception. J Gastrointest Cancer. 2021 [Online ahead of print]
- Ahn JH, Choi SC, Lee KJ, Jung YS. A clinical overview of a retrospective study about adult intussusceptions: Focusing on discrepancies among previous studies. Dig Dis Sci. 2009;54(12):2643-49
- Bassotti G, Antonelli A, Villanacci V, et al. Gastrointestinal motility disorders in inflammatory bowel diseases. World J Gastroenterol. 2014;20(1):37-44
- Choi K, Chen J, Mitra S, Sarna SK. Impaired integrity of DNA after recovery from inflammation causes persistent dysfunction of colonic smooth muscle. Gastroenterology. 2011;141(4):1293-301
- Villanacci V, Antonelli E, Geboes K, et al. Histological healing in inflammatory bowel disease: A still unfulfilled promise. World J Gastroenterol. 2013;19:968-78
- Park BL, Rabiner JE, Tsung JW. Point-of-care ultrasound diagnosis of small bowel-small bowel vs ileocolic intussusception. Am J Emerg Med. 2019;37(9):1746-50
- 22. Greco S, Giambelluca D, Pecoraro G, Salvaggio G. The pseudokidney sign in intussusception. Abdom Radiol (NY). 2020;45(1):241-42