

# Ileocolic Intussusception Due to Endometriosis

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## ABSTRACT

Intussusception is a rare cause of bowel obstruction in adults. Clinical manifestations are not specific, making the preoperative diagnosis difficult to establish. We report a case of acute small-bowel obstruction due to ileocolic intussusception. An emergency explorative laparoscopy was performed and revealed a mass in the right colon proximal to the ileocecal valve. Conversion to open laparotomy allowed us to perform a right hemicolectomy. The pathologic examination of the resected sample revealed endometriosis of the terminal ileus.

**Key Words:** Endometriosis, Intussusception, Small-bowel obstruction.

## INTRODUCTION

Intussusception is the invagination of a proximal segment of bowel (intussusceptum) into the lumen of the adjacent distal segment (intussusciens). Pediatric patients account for 95% of all cases of intussusception,<sup>1</sup> and adult patients account for 1% to 5%.<sup>1</sup> In the pediatric population, intussusception is usually idiopathic or due to a viral illness, whereas in adults an organic lesion is found in 90% of cases.<sup>2</sup> Endometriosis is a disorder characterized by the presence of normal endometrial tissue outside the uterus. Although intestinal endometriosis is quite common, usually affecting the rectum and the sigmoid colon, small-bowel localization causing obstruction is extremely rare with an incidence of 0.15%.<sup>3</sup> We present a case of acute small-bowel obstruction due to intussusception of the terminal ileus because of endometriosis and review the literature on this topic.

## CASE REPORT

A 32-year-old nulliparous Caucasian woman presented to our emergency department reporting abdominal colicky pain and distension in the last 24 hours, with nausea and vomiting. She had constipation and had been unable to defecate for the last 2 days. Her last menstrual period was 2 weeks before and it was unremarkable. She reported a long-standing history of constipation and several episodes of abdominal pain during the last 2 years for some of which she had visited a hospital, but no other diagnosis except that of irritable bowel syndrome was made. The rest of her medical history was insignificant, apart from gastritis for which she had a prescription of omeprazole. She had no operations in the past, and she admitted to smoking and occasional alcohol use. A paternal aunt had died of breast cancer, and a paternal cousin had colon cancer.

Her vital signs included a rectal temperature of 36.8 °C, a blood pressure of 115/75 mm Hg, and a heart rate of 95 pulses per minute. She was in mild distress, and a physical examination revealed abdominal distension with diffuse tenderness. On the auscultation of the abdomen, bowel sounds were hypoactive. There was no involuntary guarding or rebound tenderness; no masses were palpated. The

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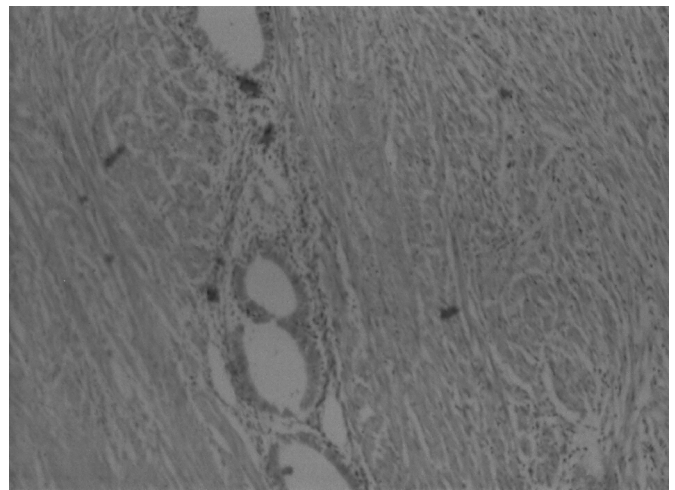
liver and spleen were also not palpable. Pelvic and rectal examinations were unremarkable. Laboratory data revealed an elevation in the white blood cell count to  $19200/\text{mm}^3$ , with 80% neutrophils. The  $\beta$ -hCG test was negative. Abdominal and chest radiographs were ordered. The result was an image of small-bowel obstruction with air-fluid levels (**Figure 1**). Based on that evidence, the patient was admitted to the department of surgery for observation. A nasogastric tube was inserted, and the patient was placed under a prophylactic regimen of antibiotics. Her clinical condition did not improve in the next 24 hours, despite the fact that she had 2 fleet enemas, and her second abdominal radiograph showed that the loops of ileus were still present and the bowel sounds were still hypoactive. Blood tests revealed a 2-fold increase in CA-125 tumor marker values. A decision was then made for a diagnostic laparoscopy. Pneumoperitoneum was obtained with the Hasson technique. A 5-mm subumbilical port was used for the telescope. One 10-mm and one 5-mm infraumbilical midline port were used as the work-



**Figure 1.** Plain abdominal radiograph showing air-fluid levels and the absence of gas throughout the colon.

ing ports. A phlegmon in the right flank was discovered, which involved the terminal ileum, the cecum, the appendix, and the right fallopian tube. There were adhesions indicating past inflammation. Free fluid was present in the peritoneal cavity. The loops of small bowel were dilated. A cyst of dark brown (chocolate) color could be seen on top of the mass, but the cecum and the appendix could not be visualized. The terminal ileum was indurated and bulbous. We performed adhesiolysis with the use of ultrasound scissors, but despite extensive attempts to mobilize the terminal ileum, the cecum and the appendix remained inaccessible. Given these difficulties along with the peril of perforation of the small bowel, we decided to proceed to an open laparotomy with a subumbilical midline incision. Under direct observation, we identified the intussusception of the distal ileum through the ileocecal valve and into the cecum. The appendix was found in the retrocecal region. With the use of the ultrasound scissors, we managed to free the right fallopian tube, and we decided to excise the right colon with the cecum, appendix, and terminal ileum en block. An end-to-end anastomosis was performed in 2 layers of absorbable stitches. The postoperative outcome was uneventful. Two days after the operation, the patient passed gas, and the day after she passed stool. Six days after the operation, she left the hospital in good health.

The histological examination macroscopically showed a parietal tumor at the terminal ileum with a diameter  $>3$  cm. It was intussuscepting through the ileocecal valve into the cecum. The microscopic image showed that this was a tumor composed of endometriosis (**Figure 2**).



**Figure 2.** Endometriotic tissue in the resected specimen (hematoxylin and eosin stain x 40).

The patient, after the results of the histological examination, was advised to seek a gynecologist for the identification and treatment of any residual foci of endometriosis.

**DISCUSSION**

Adult intussusception is a rare condition, and it is agreed that a general surgeon may encounter only a few cases in his career. Begos et al<sup>1</sup> in their review of 1048 cases considered that 1 or 2 cases of adult intussusception occur in each institution every year. There is no sex predilection, and the median age of presentation is in the sixth to seventh decade of life.<sup>4</sup> Adult intussusception represents 0.003% to 0.02% of all hospital admissions.<sup>2</sup>

As far as the mechanism is concerned, it is believed that an identifiable lesion within the bowel's lumen interferes with its peristaltic activity. Subsequent peristalsis of the bowel produces an area of constriction above the stimulus and relaxation below, thus telescoping the lead point (intussusceptum) through the distal intestinal part (intussusciptens). The intussusceptum is propelled onwards by peristalsis with mesentery and vessels becoming involved leading to the impediment of venous flow and afterwards of arterial supply.<sup>5</sup>

According to its location, intussusception can be classified as enteric, colonic, ileocolic, and ileocecal. Enteric and colonic intussusceptions are confined to the small and large bowel, respectively. Ileocolic intussusceptions occur when the ileum prolapses through the ileocecal valve into the colon. Lastly, an intussusception is characterized as ileocecal when the lead point is found on the ileocecal valve.<sup>2</sup>

Clinical presentation of intussusception varies, and the symptoms are wide ranging and often nonspecific. Fewer than 20% of cases present acutely with complete bowel obstruction, and usually the presentation is either sub-acute or chronic, consistent with partial obstruction.<sup>6</sup> Our patient developed acute symptoms, although she reported many episodes of abdominal pain the preceding 2 years. The classic triad consisting of abdominal pain, palpable mass, and heme-positive stools is rarely present. The most common presenting symptoms are intermittent abdominal pain (75% to 78%), nausea and vomiting (68% to 78%), and physical examination may reveal abdominal tenderness (60%), distention (45%), and an abdominal mass (7% to 50%).<sup>2,7</sup> Azar et al<sup>2</sup> reported that patients with benign enteric lesions tend to more frequently experience abdominal pain, nausea, and vomiting, whereas patients with colonic lesions present more often with melena or guaiac stool.

**Table 1** summarizes the findings of the biggest series concerning the location of intussusception and its cause. Of 1206 cases of adult intussusception, 35% were colonic and 65% involved the small intestine. Of the 780 small bowel intussusceptions, 482 (62%) were due to a benign lesion, whereas a malignant lesion was found in 18% of cases. In contrast, large bowel intussusceptions were attributed to a benign lesion in 30% of cases, and malignant lesions caused 245 (58%) cases. This is consistent with the fact that malignancy is much more common in the large bowel. In addition, it is evident that an underlying cause was present in nearly 83% of cases of adult intussusception.

**Table 1.**  
Literature Review of 1206 Cases of Adult Intussusception (Location and Cause)

	No. of Patients	Small Intestine	Large Intestine	Small Intestine			Large Intestine		
				Benign	Malignant	Idiopathic	Benign	Malignant	Idiopathic
Begos <sup>1</sup> , 1997	1048	671	377	423	94	154	109	219	49
Azar <sup>2</sup> , 1997	58	44	14	23	21	0	6	6	2
Eisen <sup>4</sup> , 1999	27	22	5	10	8	4	1	4	0
Tan <sup>29</sup> , 2003	9	0	9	0	0	0	4	5	0
Takeuchi <sup>30</sup> , 2003	7	4	3	2	1	1	0	2	1
Toso <sup>31</sup> , 2005	10	9	1	4	5	0	1	0	0
Erkan <sup>32</sup> , 2005	13	10	3	6	4	0	1	1	1
Barussaud <sup>21</sup> , 2005	44	29	15	18	11	0	7	8	0
TOTAL	1216	789 (65%)	427 (35%)	486 (62%)	144 (18%)	159 (20%)	129 (30%)	245 (57%)	53 (13%)

A wide range of underlying lesions causing intussusception has been reported, but there is only one case of endometriosis of the small bowel resulting in intussusception.<sup>8</sup>

Endometriosis is a common entity, affecting around 15% of childbearing women and was first described by von Rokitansky in 1860.<sup>9</sup> Various theories have been proposed to explain its development, and the most widely accepted is that of retrograde migration of endometrial tissue from the uterus to fallopian tubes and then to the peritoneal cavity.<sup>10</sup> Two reviews comprising almost 7200 cases of endometriosis each showed that small-bowel involvement has a frequency of less than 1%.<sup>3,11</sup> Ileal endometriosis is usually located within the last 10cm before the ileocaecal valve.<sup>12</sup>

The complications of intestinal endometriosis include intestinal obstruction, perforation, hemorrhagic ascites, protein-losing enteropathy, anasarca, and intussusception, which is mainly related to the vermiform appendix.<sup>13–18</sup>

Protein CA-125 is generally found elevated in endometriosis, as was the case in our patient. Nonetheless, its diagnostic accuracy has been shown to be limited in several series (sensitivity 24% to 94%, specificity 83% to 93%).<sup>19</sup>

In our patient, the intestinal damage was not recent. The inflammation, fibrosis, and hyperplasia of the intestinal smooth muscle led to the formation of adhesions. This has been described in other cases, the proposed mechanism being an endometrioma that responds to hormonal influence.<sup>20</sup>

Preoperative diagnosis of adult intussusception is difficult to establish, varying from 32% to 51%.<sup>2,21</sup> The absence of a pathognomic clinical manifestation leaves the burden of diagnosis to radiologic methods. Plain radiographs show intestinal obstruction and may indicate the site of obstruction, but they lack sensitivity and false-negative results may occur. Contrast studies like barium enema or upper gastrointestinal series can locate the intussusception, giving a “coiled spring” sign and in some cases can be therapeutic, reducing it spontaneously.<sup>22</sup> Because of the perforation risk and common false-positive results, it is not routinely recommended.<sup>23</sup> Abdominal ultrasound is useful, but the presence of air in the intestine can always give false-negative results. Lim et al<sup>24</sup> and Sofia et al<sup>25</sup> using sonography managed to diagnose 11 and 3 cases, respectively, of adult intussusception. Computed tomography (CT) and magnetic resonance imaging (MRI) are the diagnostic methods of choice for evaluating adult intussusception, its leading point, and possible associated pa-

thology.<sup>23</sup> The “target sign” and the sausage-shaped appearance are 2 typical images seen on CT. In addition, the appearance of bowel-within-bowel configuration with or without mesenteric fat and vessels compressed between the walls of small bowel is considered pathognomic.<sup>26</sup> Magnetic resonance with ultrafast multiplanar techniques permits adequate and rapid assessment, especially of cases of small-bowel obstructions.<sup>27</sup> Colonoscopy and flexible sigmoidoscopy has also been used in cases of large-bowel lesions to not only diagnose but also to correct intussusception.<sup>1</sup>

The treatment of intussusception caused by ileal endometriosis consists of surgical resection of the affected intestinal part with termino-terminal anastomosis, which is often done on an emergency basis, as was the case with our patient. The debate is focused around the risks of reduction before resection. The associated risks are (a) intraluminal seeding or venous embolization in regions of ulcerated mucosa, (b) possible perforation during manipulation, and (c) subjecting the patient to increased risk of anastomotic complications in the setting of edematous or weakened bowel.<sup>4</sup> It is generally agreed that reduction should not be attempted when signs are present of ischemia or inflammation.<sup>1</sup> Also it should never be attempted in the large bowel because of the aforementioned high frequency of malignancy. Regarding the small bowel, some authors advocate the principle of resection without reduction except for cases where there is a danger of short-gut syndrome.<sup>28</sup> In recent series, a selective approach without bowel resection is more widely adopted, especially when a malignancy is not suspected. The age of the patient, radiologic findings, and the location of the intussusception must always be taken into account.<sup>4</sup> Furthermore, in some cases, such as Meckel’s diverticulum, benign polyps, Peutz-Jeghers syndrome, adhesions, terminal ileitis, and postoperative and posttraumatic intussusception, when no other cause can be found in the bowel, resection may not be needed.<sup>1</sup> The experience of the surgeon and his ability to determine the possibility of a malignancy should be the determining factor when reduction is contemplated.

## CONCLUSION

Intussusception is a rare cause of bowel obstruction in adults, and endometriosis as its underlying lesion is even more uncommon. It is important to maintain a high index of clinical suspicion taking into account the nonspecific clinical presentation and the findings of radiologic studies to establish the correct diagnosis and proceed without delay to the resection of the intussuscepted bowel.



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