

Case report

Intussusception as a rare cause of bowel obstruction in a woman with recurrent ovarian cancer

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1. Introduction

Intussusception is the telescoping of a proximal segment of bowel into a distal segment, usually in the setting of inflammation or another lead-point and is generally a disease of childhood, as only approximately 5% of cases occur in adults.(Marinis et al., 2009) Adult intussusception remains rare, with the majority of cases occurring secondary to other disease processes including inflammatory bowel disease, structural conditions such as diverticula or adhesions, and neoplastic conditions. Colorectal carcinomas make up the majority of cancer-related intussusceptions, representing approximately two-thirds of these cases.(Nagorney et al., 1981) Due to the high incidence of concurrent carcinoma, the standard treatment for adult intussusception is surgical resection, usually without reduction.(Marinis et al., 2009) Children with intussusception classically present with acute onset abdominal pain, vomiting, and passage of blood per rectum. The presentation of intussusception is less distinct in adults, as most cases present more insidiously as bowel obstruction with chief complaints of bowel habit changes, abdominal pain/distention, and nausea/vomiting. Only about 1–5% of all adult bowel obstructions are associated with intussusception; therefore, the diagnosis is typically made on imaging, with computed tomography being the standard modality.(Azar and Berger, 1997) The objective of this case report was to describe a rare case of intussusception associated with recurrent ovarian cancer.

2. Case report

A 71-year-old woman with platinum-resistant recurrent stage IIIC

ovarian/primary peritoneal cancer presented to the emergency department after an incident of bright red blood per rectum in the setting of a three-week history of diffuse, intermittent cramping abdominal pain. She had a personal history of breast cancer and was diagnosed with ovarian cancer in 2014 with ascites, omental caking, and carcinomatosis. Germline testing was negative, and somatic tumor analysis revealed a *BRCA2* mutation. She received neoadjuvant chemotherapy with interval optimal debulking and adjuvant chemotherapy with complete response and normalization of Ca-125. She recurred nine months later and was treated with salvage chemotherapy for four additional lines. At the time of presentation, she had been on olaparib for four months and had an excellent initial response based on Ca-125 and CT scan.

Outside of the bleeding and cramping, she had no other associated bowel symptoms over baseline constipation at presentation. She had normal vital signs. Her physical exam was notable for normal bowel sounds, mild tenderness in the lower abdomen, and a palpable mass in the right lateral abdomen. Rectal exam was notable for known external hemorrhoids that were not bleeding or thrombotic. Abdominal CT scan was obtained which was suggestive of an ileocolic intussusception (Fig. 1). She was admitted to the gynecologic oncology service and taken for emergent exploratory laparotomy. Upon entering the abdomen, carcinomatosis was noted with multiple 1–2 cm tumor nodules throughout the abdominal wall, supracolic omentum, mesentery, and pelvis. Hepatomegaly was noted with palpable intraparenchymal nodules. The terminal ileum was prolapsed into the cecum up to the hepatic flexure, confirming the diagnosis of intussusception. The entire intussuscepted bowel measured approximately 20 × 30 cm and was

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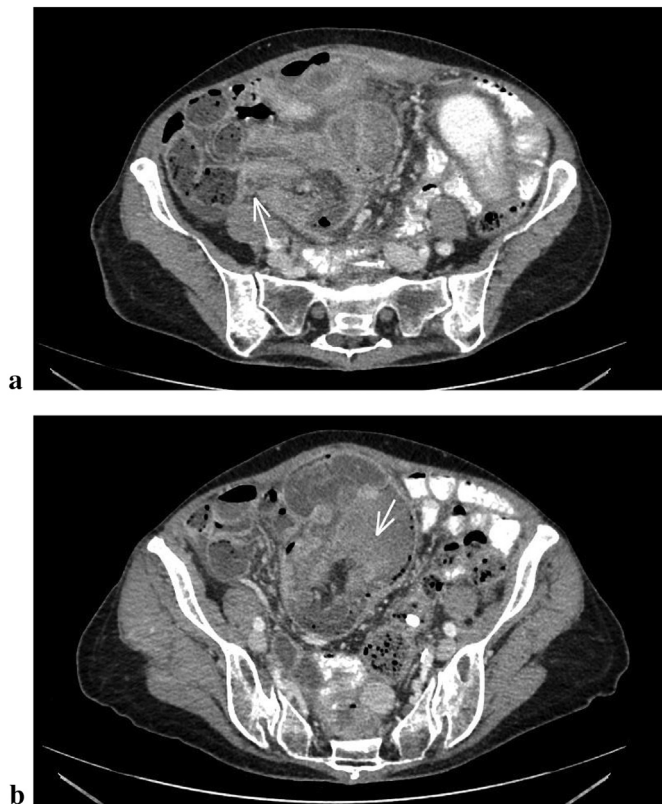


Fig. 1. CT Images Suggestive of Intussusception. (A) Axial CT image of an apparent small bowel transition point in the right lower quadrant (indicated with white arrow). (B) Axial CT image demonstrating abdominal mesenteric fat prolapsing into the cecum (indicated with white arrow). The prolapsed mesenteric fat and colon was seen in subsequent sections extending to the level of the transverse colon.



Fig. 2. Surgical Specimen of Resected Intussuscepted Bowel. The involved bowel was resected en-bloc, necessitating resection of the terminal ileum and right hemicolectomy through the proximal segment of transverse colon. The mass measured approximately 20 × 30 cm at the time of resection.

resected en-bloc without reduction with a right hemicolectomy and terminal ileum resection (Fig. 2). A stapled side-to-side anastomosis was performed between the proximal ileum and transverse colon in the usual fashion.

She recovered well from surgery and was discharged on post-operative day three. She restarted olaparib one month following surgery

but progressed two months later. She continues on other treatment.

3. Discussion

The differential for patients with ovarian cancer presenting with bowel complaints is broad. Many cases of ovarian cancer are diagnosed in the setting of a primary bowel complaint in both the primary and recurrent setting. Malignant bowel obstruction due to ovarian cancer is extraordinarily common, with some studies reporting that up to 50% of women with ovarian cancer experience bowel obstruction. (Chen et al., 2014) Ovarian cancer-related bowel obstruction can be secondary to primary disease burden or secondary to treatment (i.e. surgical adhesions).

Patients with malignant bowel obstruction are typically treated conservatively initially with bowel decompression (nasogastric tube) and bowel rest, and surgery is avoided if possible in a chemotherapy-resistant setting. This patient had a more acute onset and lacked some of the hallmark signs of bowel obstruction, such as cessation of flatus/bowel movements and nausea/vomiting. Surgical management was initiated once intussusception was identified on imaging, as this issue must be solved surgically whereas malignant bowel obstruction in the setting of recurrent cancer would initially be managed conservatively and surgical intervention only pursued if there was reasonable expectation of improved survival as recurrent malignant bowel obstruction is the usual terminal event.

This case also highlights operative technique regarding adult intussusception. In children, less invasive means of reduction (i.e. air enemas) are usually employed. Surgery remains the standard of care in adults. The nature and extent of the surgery do vary with the location of the intussusception, as was relevant in this case. Colon intussusceptions are presumed to be secondary to a neoplastic process, as approximately two-thirds of these cases are related to an underlying cancer. (Haas et al., 2003) Small bowel intussusceptions are less likely to be secondary to an underlying cancer, as only one-third of these cases result from a neoplastic process. (Haas et al., 2003) The majority of these cancer-related cases are metastatic to the small bowel, often in the setting of metastatic melanoma. (Karakousis et al., 1974) Of particular relevance to gynecologic surgeons, endometriosis is a known benign cause of intussusception, generally because of cecal involvement. (Indraccolo et al., 2010; Koutsourelakis et al., 2007)

Because small bowel intussusception is more likely to be secondary to benign causes, reduction may be attempted to avoid the morbidity of extensive bowel resection. Conversely, in cases involving the colon, reduction is not recommended throughout the literature due to the concern for seeding of the cavity. This was relevant in this patient's operative management because her intussusception spanned the small bowel and colon (ileocolic). Her history of marked peritoneal disease/carcinomatosis was suspicious as a potential lead point for invagination via bowel to peritoneal tethering, allowing normal peristaltic activity to result in intussusception.

This case differs from previously reported cases of cancer-related intussusception in both primary cancer site and metastatic status. Cancer-related intussusceptions most often occur with primary colorectal cancers (approximately two-thirds of all cases). (Nagorney et al., 1981) Other cancer-related cases of intussusception are generally from lead-points created by gastrointestinal metastases. This case represents a primary ovarian malignancy with secondary intussusception, notably without evidence of intraluminal metastasis to the gastrointestinal tract. Two other cases of ovarian cancer-related intussusception have been reported; however, both of these cases were in the setting of an intraluminal gastrointestinal metastasis of primary ovarian cancer. Kohyama et al. reported a case of ovarian cancer-related intussusception due to metastasis to both the ascending colon and sigmoid colon six years after original surgical intervention for papillary serous adenocarcinoma. (Kohyama et al., 2005) Langner et al. described a case of intussusception due to rectosigmoid metastasis of a recurrent ovarian

fibrosarcoma.(Langner et al., 2009) Both of these cases were treated with surgery in a similar fashion to this case.

This case highlights how broad differentials must be for complaints of abdominal pain, constipation, and nausea/vomiting. While intussusception is a rare cause of these complaints in the setting of adult bowel obstruction, its frequent association with an underlying cancer makes it a dangerous diagnosis to miss. Furthermore, although the majority of cancers resulting in intussusception are either primary or metastatic gastrointestinal lesions, treating physicians must keep other malignancies in mind.

Conflicts of interest

The authors report no conflicts of interest.

Author contributions

Susan C. Modesitt and Mackenzie W. Sullivan both contributed fully to the planning, writing, and submission of this article.

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