



Case Report

Adult Intussusception of Appendicular Mucinous Cystadenoma

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Intussusception is a rare cause of adult intestinal obstruction. Clinical manifestations are not specific, which makes a preoperative diagnosis difficult to establish and often causes delay. We report a case of acute intestinal obstruction due to ileocolic intussusception. An emergency laparoscopy was performed revealing an obstructive mass at the ileocecal region resulting from an appendicular tumor. A right hemicolectomy was conducted. The pathologic examination of the resected sample concluded the mass comprised an appendicular mucinous cystadenoma. We discuss the clinical features, preoperative diagnosis, and surgical strategies of adult intussusception, as well as the uncommon finding of an appendicular mucinous tumor as its causative lesion, with a review of the available literature.

Key words: Adult intussusception – Appendiceal mucocele – Intestinal obstruction

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, in whom it is usually idiopathic or the result of a viral process; whereas adult patients account for only 1% to 5%, and an organic lesion is found in up to 90% of the adult cases.^{1,2}

Acute bowel obstruction due to intussusception is rare in adults and accounts for 0.003% to 0.02% of hospital admissions and for only 1% of all intestinal obstructions.¹ Preoperative diagnosis is a real challenge for physicians because of its unspecific and extremely variable presentation and requires a

high index of suspicion. Therefore, the initial diagnosis is usually missed or delayed and may only be established during surgery.^{3–5}

We present a very uncommon case of acute intestinal obstruction resulting from an ileocolic intussusception secondary to a mucinous tumor of the appendix and review the literature about this finding as a lead point.

Case Report

A 57-year-old female presented to the emergency department with a 2- to 3-month history of intermittent and worsening abdominal pain, great-

est in the right lower quadrant, which had increased gradually over the past 3 days. It was associated with dizziness and diarrhea but she denied any history of fever, nausea, vomiting, bloody stools, or weight loss. The patient's medical history included a groin hernia surgery and gastroesophageal reflux on oral treatment.

Initial vital signs were within normal limits. Abdominal examination revealed diffuse tenderness on palpation, above all in the right lower quadrant, but without rebound or voluntary guarding. There was no palpable mass, and the auscultation found struggle bowel sounds. Blood tests were normal as was urine analysis.

Abdominal X-ray exam was not demonstrative of intestinal obstruction. Computed tomography (CT) of the abdomen and pelvis showed a pathologic mass at the ileocolic region, of cystic appearance with parietal calcifications, with edematous pericolic fat stranding suggesting of ileocolic intussusception causing an intestinal obstruction with possible bowel vascular injury (Fig. 1).

These radiologic findings indicated a laparoscopic examination that revealed an intestinal obstruction resulting from a tumor-like mass at the ileocolic region where the intussusception was confirmed, with an appendiceal tumor involved as a lead point (Fig. 2). After laparoscopic mobilization of the right colon, the ileocolic segment was pulled out through a 5- to 6-cm transverse skin incision (Fig. 3). A right hemicolectomy was accomplished and primary side-to-side mechanical ileo-mid-transverse anastomosis was performed extracorporeally. The postoperative course was uneventful, and the patient was discharged on the fourth day.

The specimen pathology revealed an ileocolic intussusception resulting from an appendicular mucinous cystoadenoma.

Discussion

Adult intussusception is a rare entity in contrast to children's intussusception. There is no sex predilection, and the median age of presentation is in the sixth to seventh decade of life.⁶ Adult intussusception represents 0.003% to 0.02% of all hospital admissions.^{1,3}

The exact mechanism remains unknown. However, it is believed that an identifiable lesion within the bowel's lumen interferes with normal peristaltic activity. Then, this difference in motility between 2 intestinal parts produces an area of constriction above the stimulus and relaxation below, thus

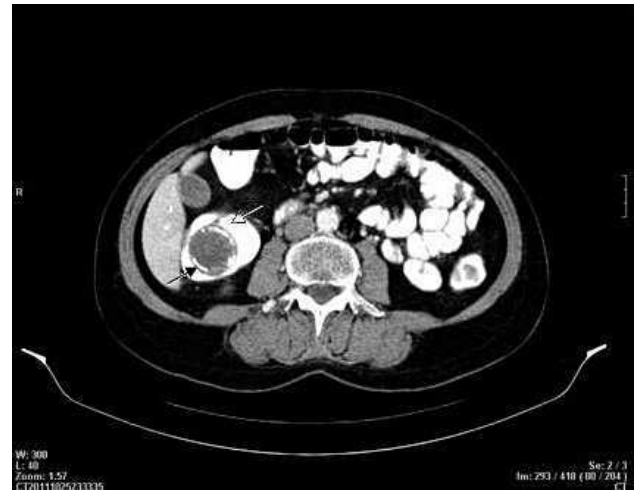


Fig. 1 Abdominal CT (axial contrast image) showing the bowel-within-bowel configuration with the classic “target-shaped mass” in the right abdomen suggesting an ileocolic intussusception. There is a cystic lesion with calcified walls (black arrow) that acts as the lead point of intussusception, enveloped eccentrically by contrasted colon (white arrow).

telescoping the lead point (intussusceptum) through the distal segment (intussusciens). The lead point is propelled onwards by peristalsis with mesentery and vessels becoming involved in the bowel vascular supply.^{1-3,7,8} The most common locations are at the junctions between freely moving segments and retroperitoneally or adhesioneally fixed segments.⁴ There are 4 categories of intussusception according to its location: (1) enteroenteric, confined to the small bowel; (2) colocolic, involving the large bowel only; (3) ileocolic, when the terminal ileum prolapses itself within the ascending colon, and (4) ileocecal, where the ileocecal valve is the lead point of the intussusception.⁹ It is very difficult to distinguish between ileocecal and ileocolic intussusceptions.¹

Clinical presentation varies with a wide range of symptoms, which are often nonspecific. Usually, the presentation is either subacute or chronic. The classic triad of abdominal pain, palpable mass, and heme-positive stools is rarely present. In fact, some patients have no previous symptoms, and two-thirds of the patients present with chronic, recurrent, colicky pain, which may reflect transient intussusceptions and may be missed by routine examinations.^{1,3} Thus, patients may be discharged from the hospital as having a functional disorder, tending to delay treatment of an underlying malignant tumor.³ The most common presenting

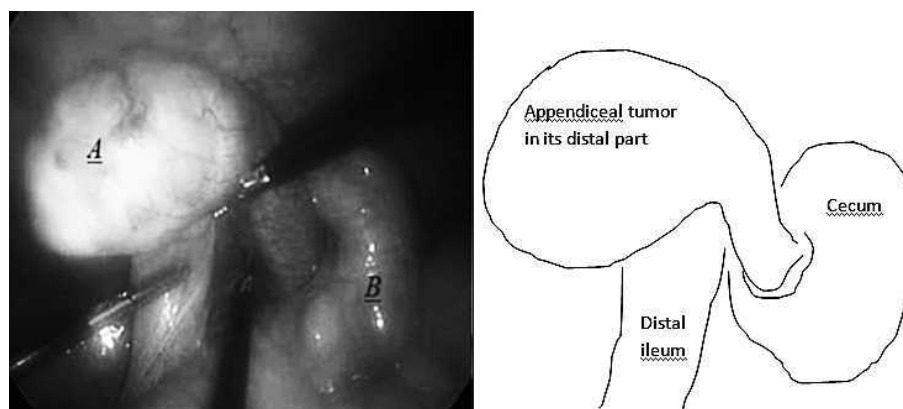


Fig. 2 Laparoscopic image showing (A) the appendiceal tumor (intussusceptum) from (B) the cecal “wrapping” (intussusciens).

symptoms are intermittent abdominal pain (75%–78%) and nausea and vomiting (68%–78%). Physical examination may reveal abdominal tenderness (60%), distention (45%), and abdominal mass (7%–50%).^{1,10}

Preoperative diagnosis of adult intussusception is difficult to establish, ranging from only 32% to 51%, becoming a real challenge for the physician.^{5,11} The absence of a pathognomonic clinical manifestation leaves the burden of diagnosis to radiologic methods. Plain X-rays may reveal the site of intestinal obstruction, but they lack sensibility, and false-negative results usually occur. Although contrast studies like barium enema may locate the intussusception, giving a “coiled spring” sign, they are not routinely recommended because of the perforation risk and common false-positive results. The presence of air in the bowel always becomes a significant

constraint for abdominal ultrasound, giving common false-negative results. CT is the method of choice for evaluating adult intussusception, its leading point, and associated pathology, with diagnostic accuracy of 58% to 100%.^{1,12–14} The “target” or “doughnut” sign and the sausage-shaped appearance are 2 typical images seen on CT.^{15,16} The appearance of bowel-within-bowel configuration with or without mesenteric fat and vessels compressed between the walls of small bowel is considered pathognomonic. The intussusceptum is the center, and the edematous intussusciens forms the external ring.^{1,17} Furthermore, CT can identify metastases, lymphadenopathy, free liquid, or proximal bowel occlusion. However, detection of the casual lesion remains difficult.³

In the small-bowel adult intussusception, primary lesions are found in up to 80% of the cases. They are most often benign, including lipoma, leiomyoma, Meckel’s diverticulum, adenoma, and inflammatory fibrous polyps, although some rare malignant lesions are possible (over 30% of the cases) such as lymphoma and metastases. In contrast, in the colon, malignant underlying lesions are the most involved (over 66% of the cases), including adenocarcinoma.^{2,3,7,18} However, the surgical patient’s history should be taken into account because there are also postoperative intussusceptions caused by the mechanical disturbance of peristalsis induced by lesions such as a suture line, bowel edema, or adhesions. Postoperative ileus itself may also favor intussusception.³

Regarding our case, the involvement of the cecal appendix as a cause of intussusception was first described in 1858.^{19,20} It’s an uncommon condition with an incidence rate of 0.01% in an autopsy series.²¹ Several etiologies have been noted: ana-

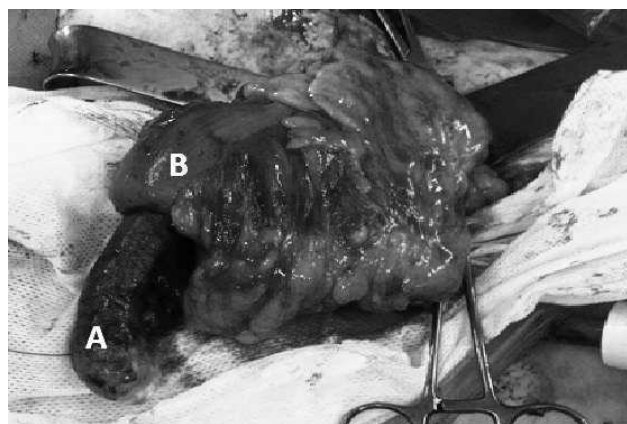


Fig. 3 Operative image of the ileocolic segment pulled out of abdominal cavity that will be resected, showing (A) the reduced appendiceal mucinous tumor (intussusceptum) from (B) the intussusciens (the cecum).

tomic variations of the appendix, or pathologic conditions such as tumors [polyps, mucinous cystoadenoma or cystoadenocarcinoma, carcinoid tumor, and gastrointestinal stromal tumors (GIST)], endometriosis, parasitism, cystic fibrosis, abnormal appendicular peristalsis, fecaliths, and foreign bodies.^{20,22–25} Intussusception of the resultant appendiceal stump after inversion-ligation appendectomy has even been described.²² Nevertheless, appendiceal intussusception may develop without any underlying abnormality. In all these cases in which the cecal appendix is involved as the lead point of intussusception, the clinical presentation may mimic an acute appendicitis.^{20,22,23}

There are differences neither in symptomatology nor in clinical features between intussusception caused by malignant or benign lesions. In most of cases, the clinical findings are unspecific but, when there is high suspicion of appendiceal mucinous tumor, prompt surgical intervention is advocated not only to solve the intestinal occlusion itself but also to prevent development of pseudomyxoma peritonei.²⁶

In our case, we found a benign mucocele of the appendix as a lead point of intussusception. The term mucocele refers to an enlarged appendix and to mucoid content, and it includes both benign and malignant tumors (cystoadenoma or adenocarcinoma).²⁷ Their classification has been controversial, and terminology used inconsistently. Misraji *et al* reviewed 107 appendiceal mucinous neoplasms and concluded that they can be classified as either low-grade mucinous neoplasms or mucinous adenocarcinomas, based on their architectural and cytologic features.²⁸

Appendicular mucocele is a very uncommon disease, estimated to be found in 0.2% to 0.3% of all appendectomies and in up to 3.3% of adult intussusception.^{29,30} It may even appear as an incidental finding in imaging tests carried out for other reasons or during surgery. Few series have been reported on this type of appendiceal tumor; one of the broadest was published in 2003 at the Mayo Clinic with 132 patients,³¹ and another was published in 2007 with 35 cases in a 21 year period,³² but in them no case of appendicular mucocele as a lead point of intussusception was found.²⁹ We could only find a few cases of appendicular mucocele as a lead point of intestinal adult intussusception published in the medical literature. For instance, in 2004, Yamaguchi *et al* noted that only 13 cases of appendicular mucocele as a lead point have been listed among 400 cases of adult intussusception over

the past 10 years from Japanese domestic report.³⁰ Table 1 shows a list of available literature review over the past 20 years of mucinous cystoadenoma of the appendix as a cause of adult intussusception.^{25,26,30–44}

Surgical intervention is mandatory in all cases of intussusception in adults. The treatment consists of surgical resection of the intestinal segment affected, with primary anastomosis if possible. The debate is focused around the risks of reduction before resection. The associated risks are (1) intraluminal seeding or venous embolization in regions of ulcerated mucosa, (2) perforation during manipulation, and (3) the increased risk for the patient owing to anastomotic complications in the setting of edematous or weakened bowel. It is agreed that reduction should not be attempted when signs are present of ischemia or inflammation, and also in colocolic intussusception because of the high risk of underlying malignant lesion. If the colon is involved or the lesion appears malignant, a surgical resection without prior reduction intent is strongly recommended.^{9,45–49} Of note, it may be added that there is no way to distinguish between a colonic intussusception that harbors a benign lesion and one with a malignant lesion.¹ Regarding the small bowel, the principle of reduction without resection is chosen more often, especially when a malignancy is not suspected.⁵⁰ With respect to the surgical technique, a laparoscopic reduction of intussusception is feasible if a benign underlying lesion is highly suspicious.^{25,30} Furthermore, in such cases as Meckel's diverticulum, benign polyps, Peutz-Jeghers syndrome, adhesions, terminal ileitis, and postoperative and posttraumatic intussusception, when no other cause can be found, resection may not be needed. Nevertheless, the patient's age, radiologic findings, and the site of intussusception must be taken into account.^{1–3,10,15}

Conclusion

Intussusception is a rare cause of intestinal obstruction in adults, and appendicular mucinous tumor as underlying lead point lesion is even more uncommon. A high degree of clinical suspicion is mandatory, taking into account the nonspecific clinical presentation. The physician should keep this condition in mind in order to allow earlier diagnosis and to perform without delay a surgical resection, as there is an underlying malignant lesion in almost half of both colonic and enteric intussusceptions.

Table 1 Literature review of mucinous cystoadenoma of the appendix acting as a lead point of adult intussusception over the past 20 years^a

Reference	Publication year	Institution	Country	No. of Cases
Turrin <i>et al</i> ³³	1994	Servizio di Radiologia, Ospedale Generale di Zona Umberto, Bellano, Como	Italy	1
Jones <i>et al</i> ³⁴	1997	University of California, Davis Medical Center, Sacramento	USA	1
Heithold <i>et al</i> ²⁵	1997	Surgery Department, Georgia Baptist Medical Center, Atlanta	USA	1
Gockel <i>et al</i> ³⁵	1998	Chirurgische Klinik, Städtische Kliniken, Wiesbaden	Germany	1
Rudek <i>et al</i> ³⁶	2001	Surgery Department, University of Heidelberg	Germany	1
Coulier <i>et al</i> ³⁷	2002	Department of Diagnostic Imaging, Cliniques St. Luc, Bouge	Belgium	1
Yamaguchi <i>et al</i> ³⁰	2004	Surgery Department, Yokohama City University School of Medicine	Japan	14 ^a
Waseem <i>et al</i> ³⁸	2007	Surgery Department, Ittefaq Hospital Trust, Lahore	Pakistan	1
Butte <i>et al</i> ³⁹	2007	Pontificia Universidad Católica de Chile	Chile	1
Blondiaux <i>et al</i> ⁴⁰	2007	Radiology Department, Rouen University Hospital Charles Nicolle, Rouen	France	1
Siddiqi <i>et al</i> ⁴¹	2007	Radiology Department, Northwestern, Memorial Hospital, Feinberg School of Medicine, Chicago	USA	1
Okuda <i>et al</i> ⁴²	2008	Department of Diagnostic Radiology, Toranomon Hospital, Tokyo	Japan	1
Wei-Ming <i>et al</i> ⁴³	2010	Department of Diagnostic Radiology, Chang-Gung Memorial Hospital, Chiayi	Taiwan	1
Fernández-Rey <i>et al</i> ⁴⁴	2010	Servicio de Radiodiagnóstico, Hospital Universitario Central de Asturias, Oviedo	Spain	1
Ashrafi <i>et al</i> ²⁶	2011	Surgery Department, University Hospital of South Manchester NHS Trust, Manchester	UK	1
Total				28

^aOne case added by the author and the other 13 listed in Japanese reports from 1994 to 2004.

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