Intestinal intussusception: a diagnostic dilemma in adults. Two case reports and literature review

ABSTRACT

Background: Intestinal intussusception is a common pathology among children, whereas it is a rare entity in adults. The child/adult ratio is > 20:1. The condition is found in < 1/1300 abdominal surgeries and in 1/100 patients operated on for intestinal obstruction. Clinical manifestations of adult intussusception are nonspecific and patients may present with acute, intermittent or chronic symptoms, predominantly those of intestinal obstruction.

Clinical case: We report two cases of intussusception in adults. The first case, ileo-ileal intussusception, was secondary to hamartoma in the terminal ileum. The second case presented as an ileo-colonic intussusception in which no underlying lesion was identified as a causal factor.

Conclusion: Adult intussusception is a rare entity. Most cases have a precipitating factor and due to its association with a malignant pathology, intestinal resection without reduction is the surgical procedure of choice.

Key words: Adult intussusception, hamartoma, intestinal obstruction.
BACKGROUND

Ileal intussusception is defined as two consecutive segments of the gastrointestinal tract that invaginate into one another. Intussusception is rare in adults representing < 5% of all invagination cases and ~1 to 5% of intestinal obstruction. From a clinical point of view, it manifests with nonspecific symptoms such as abdominal pain, nausea, diarrhea and rectal bleeding. The classic triad of symptoms seen in children is a palpable tumor in “sausage” form, red-colored stool (similar to red currant jelly) and acute abdominal pain. This is less common in adults. Preoperative diagnosis requires a high index of suspicion because of the specific diagnostic symptoms. Two cases of invagination in adults that required surgical exploration are reported.

Case 1

We present the case of a 26-year-old female patient with no clinically significant history with a clinical picture of 5 months of abdominal pain in the right lower quadrant accompanied by occasional diarrhea, with spontaneous relief. One day prior to hospital admission, the pain became more intense and localized in the lower abdomen and was associated with nausea and vomiting. These symptoms led the patient to hospitalization. Physical examination revealed a dehydrated patient with extremely painful abdomen upon light palpation. Bowel sounds were increased with frequency. On rectal examination, the ampulla was empty. Abdominal radiographs showed that the loops were distended with fluid levels. Ultrasound revealed a 17-mm fluid-filled image in the pelvic cavity.

During exploratory surgery, ileo-ileo intussusception of ~15 cm at 20 cm of the ileocecal valve was found. Intestinal resection and anastomosis was performed. Histopathology showed polypoid lesions with vascular proliferation, fibroblasts, some adipocytes and smooth muscle fibers compatible with ileal hamartoma (Figures 1 and 2). At the 12-month follow-up, the patient remained with a favorable clinical status.

Case 2

We present the case of an 18-year-old female with 6 months of disease evolution characterized by diffuse abdominal cramping accompanied by abdominal bloating, alternating periods of diarrheal stools and constipation, with these symptoms all remitting spontaneously about every 3 weeks. During that period, the patient reported a weight loss of 10 kg and, on one occasion, during the acute episodes she reported hematochezia. Physical examination found her

Figures 1 and 2. Microscopic view of polypoid lesions with vascular proliferation, fibroblasts, adipocytes and smooth muscle fibers.
dehydrated with abdominal bloating, pain in lower quadrants of the abdomen and without signs of peritoneal irritation. Abdomen X-rays revealed fluid levels (Figure 3).

During exploratory laparotomy a tumor was found in the transverse colon, corresponding to an ileo-colic intussusception in the terminal ileum. Cecal appendix, cecum and ascending colon were found invaginated in the transverse colon (Figure 4). A right hemicolecctomy was performed without complications and after 18 months the patient remains in good clinical condition. Histopathological study failed to demonstrate the probable cause of the event.

**DISCUSSION**

Abdominal pain causes 5 to 10% of patient visits to the emergency department of a hospital where it remains a diagnostic challenge due to the wide range of differential diagnoses: gastrointestinal, gynecological, genitourinary and cardiopulmonary diseases.5

Intussusception or ileal intussusception refers to the telescopic movement of a proximal segment of the intestine (intussusceptum) in the lumen of the adjacent distal segment (intussuscipiens). It is a common condition among children and rare in adults; 95% of cases of intussusception occur during childhood. Intussusception is responsible for 1 to 2% of adult intestinal obstructions.3 The mean age is 49.57 to 54.4 years and the male to female ratio is 1:1.3.6,7 Our patients are young adults below the age of the indicated average (26 years for the patient reported in case 1 and 18 years for the patient reported in case 2).

In children, 90% of the cases are idiopathic in contrast to 90% of adults in whom the cause is identified.8,9 Most causes in the small intestine are due to benign diseases: Meckel’s diverticulum, appendicitis and adhesions. This is in
accordance to what was reported by Soria-Cés-pedes et al. in a 19-year-old male patient with an ileal intussusception secondary to Meckel’s diverticulum. Malignant causes account for 25% of invaginations in the small intestine, whereas in the large intestine they increase to 50% as in the case reported by Franco-Herrera et al. of an 81-year-old male who suffered a conditional jejunal jejunal intussusception caused by a metastatic tumor of a malignant melanoma.

According to Alexander et al., African Americans are 2.5 times more likely to have a malignant cause of ileal intussusception. Similarly, in their study comprising 1,178 cases, they compared those who had intussusception of the small intestine with those of the large intestine with a similar distribution in terms of number of cases, age and gender. Cases of malignant etiology were more frequent for large bowel intussusception.

According to Nuño et al. in 1940, Clarke used the term “myoepithelial hamartoma” to describe a type of gastrointestinal submucosal tumor composed of glandular elements limited by epithelial cells and smooth muscle. Only a few cases have been reported of ileal intussusception secondary to a solitary hamartoma with the majority being in the pediatric population. In our patients we only identified the cause of the intussusception in one patient and this was due to a hamartoma. This report adds a new case of a poorly described ileal intussusception in the world literature.

In adults, cases may be either acute or chronic, and abdominal pain is the most common symptom (71 to 100%) followed by nausea and vomiting in 40 to 60% of the cases. Rectal bleeding may be present in 4 to 33% of the cases. This wide range of symptoms is generally based on the location of the intussusception. Acute abdominal pain with muscle guarding coexists in only 50% of the cases. Abdominal tumors are palpable in < 10% of patients. The main symptoms are those of a partial intestinal obstruction where the most outstanding characteristic is periodic, intermittent and colicky abdominal pain. In the cases we report, the clinical course was chronic, insidious, and characterized by symptoms of intestinal obstruction. In none of these cases was the preoperative tentative diagnosis that of intussusception. Cakir et al. published their experience and in 72% of the cases there was an acute intestinal obstruction and 2% had rectal bleeding. This is in contrast to the series published by Guíllén Paredes et al. in which 28.5% of the cases presented with a clinical scenario of intestinal obstruction. Invaginations are classified according to their location. The four types are ileo-colic, ileo-ileo-colic, colo-colic and small intestine intussusception (jejunum-jejunal and ileo-ileo).

Correct preoperative diagnosis is reported in 30 to 70% of the cases primarily due to the nonspecific clinical presentation. Because obstructive symptoms predominate in most cases, plain radiographs of the abdomen are the primary method of diagnosis. Signs of intestinal obstruction such as dilated asas and air-fluid levels are evident and can lead to the site of the obstruction. Ultrasonography is a useful diagnostic method where the classic sign is the image in “target” in a cross-sectional view and “pseudo-kidney” in a longitudinal view. However, obesity, air in the distended bowel and operator skill can limit the accuracy of the study. Currently, CT of the abdomen is considered the most sensitive radiological method for diagnosis of intussusception. Characteristics of the tomographic image are those of a target image or a homogeneous tumor in the shape of a “layered sausage”. Diagnostic accuracy of tomography ranges from 58 to 100% in the most recent reports.

For adults with intussusception, surgical exploration remains critical. However, controversy persists with the optimal strategy. The principle
of resection without reduction is well established.\textsuperscript{11} Recent reports recommend the initial reduction of viable bowel externally prior to resection.\textsuperscript{7,13} It is necessary to take into account various situations before carrying out a reduction: frequency of the condition associated with the event, malignant potential as a cause of ileal intussusception, anatomic site and extent of the intussusception, and degree of inflammation and ischemia of the affected bowel segment. The high probability of malignancy in colonic intussusception justifies resection without reduction. In small bowel intussusception it appears much more feasible to follow a more selective approach. If information exists of a previously confirmed benign lesion, then resection is not performed.\textsuperscript{20} The extent of this remains a subject of debate.\textsuperscript{21}

Although ileal intussusception itself has a good prognosis, it is the nature of the culprit lesion that provides the decisive factor in terms of the evolution. Mortality due to intussusception in adults increases from 8.7\% for benign lesions to 52.4\% for the malignant variety.\textsuperscript{3}

To summarize, a rare case of intestinal obstruction was reported in one adult patient secondary to hamartoma as a triggering cause of an ileal intussusception. In the second case, the trigger of the intussusception was not identified.

In conclusion, intussusception in adults is a rare disease but is also well recognized and requires high clinical suspicion due to the nonspecific symptoms at the time of patient contact. Most cases have a precipitating cause and due to its high frequency of malignancies, bowel resection without reduction is the surgical procedure of choice.

REFERENCES


