ADULT ENTERO-ENTERIC INTUSSUSCEPTION CAUSED BY SMALL BOWEL SUBMUCOSAL LIPOMA

Autores: Dr. Karel Borroto Martínez[1], Dra. Raysy Sardiñas Ponce[2], Dra. Yoanna Ramírez Fernández[3].

[1]Especialista de Primer y Segundo Grado en Cirugía General. Profesor Asistente. Investigador Agregado. Centro de Investigaciones Clínicas. Email: karelborroto@infomed.sld.cu

[2] Máster en Cirugía de Mínimo Acceso. Especialista de Primer y Segundo Grado en Cirugía General. Profesora Auxiliar. Investigadora Agregada. Centro de Investigaciones Clínicas. Email: raysyponce@infomed.sld.cu

[3] Especialista de Primer en Cirugía General y medicina General Integral. Profesora Asistente. Investigadora Agregada. Centro de Investigaciones Clínicas. Email: yoannarf@infomed.sld.cu

ABSTRACT

Introduction: Intussusception is the prolapse of one gastrointestinal segment into an adjacent part. Its incidence in adults is less than 10 %.

Aim: This article aims to present a case of adult entero-enteric intussusception caused by small bowel submucosal lipoma.

Clinical case: A 66-year-old male patient with vague clinical manifestation that presented in a computed tomography scan typical invagination image, it showed the classical "target" and "sausage-like" images. He underwent surgery, where the diagnosis of adult entero-enteric intussusception was confirmed and then an en-block resection was performed. Anatomopathological examination revealed a submucosal lipoma of the jejunum. Gastrointestinal lipomas have an incidence of 0,035 % - 4,4 %.

Conclusion: This is a rare case, not only because the intussusception is uncommon in adults but for the lead point found in it.

Key Words: intussusception; adult; lipomas.

INTRODUCTION

Intussusception (IS) is defined as the telescoping or prolapse of one gastrointestinal system segment into the lumen of an adjacent part.[1,2,3,4] The proximal segment is called the instussusceptum and the distal portion is called intussescepiens.[5] This disease was first described in 1674 by Paul Barbette and since that time, it is considered a medical and surgical emergency.[6,7]

This pathology is rare in adults, and the incidence reported is less than 10 % of all cases of IS.[1,4,6,8,9] In terms of cases-per-year, IS stands at two or three cases-per-million-people and it's only responsible for one to five percent of all adult intestinal obstructions.[2,4,5,7,8] In small bowel intussusception a benign lead point is a more frequent cause and when a malignant tumor is found in this cases are often due to a metastatic disease.[2,4,6] Other causes of intussusception in adults are: pancreas divisum, polyps, Meckel diverticulum, mesenteric cysts, abnormal peristalsis, thyroid abnormalities, infections, postoperative adhesions, Chron's granulomas, hernias or intestinal ulcers.[3,4,6,7]

The IS´s production mechanism is unclear.[8] But is generally accepted that any lead point or stimulus that alters normal peristaltic motion may initiate the advancement of the proximal intestine (or intraluminal mass) forward and into the lumen of a fixed distal intestinal segment.[1,6,8] The typical examples of this category include pedunculated tumors, such as lipomas or adenomatous polyps.[1] If this pathology is not detected timely may produce permanent bowel obstruction and even evolve into intestinal necrosis and it can be life-threating.[1,4]

In 1956 Dean et al presented their classification system for adult´s IS and they proposed four distinct anatomic variants, based on location.[5,6] The first one is called entero-enteric in which case only the small intestine is affected.[4,6,9] The second one is the colo-colic and occurs when the intussusception is limited to the colon and rectum (no anal protrusion).[4,6,7,9] The third one is named ileocecal and happens when the ileocecal valve is the

lead point plus this portion of the small bowel invaginates into the ascending colon.[4,5,6,7] And the fourth one is known as ileocolic and occurs when the terminal ileum prolapses to the ascending colon through the ileocecal valve, but the appendix does not invaginate.[4,5,6] Another classification system categorizes cases based on the etiology of the intussusception, benign or malignant.[6]

The clinical diagnosis of adult IS is difficult due to it manifests with nonspecific symptoms.[1,3,6] The classical triad of abdominal pain, palpable mass, and bleeding per rectum (red currant jelly stools) is rarely seen in adults, leading to frequent misdiagnosis.[5] This entity often onsets as an intermittent cramping abdominal pain associated with signs of bowel obstruction.[4] In about 90 % of cases, sudden-onset abdominal pain is the most common symptom, but in some patients, this pain can relax and remit.[6,7] Other symptoms also found in this disease are: pain, rectal bleeding, nausea, vomiting, changes in bowel habits, distension, and the late manifestations are signs of peritoneal irritation and intestinal obstruction.[6,8,10]

Physical examination shows that about 10 % of cases had an abdominal mass; they can also have distended abdomen with decreased or absent bowel sounds, with variable tenderness to palpation.[6,8] But it may even exhibit signs of shock mostly due to peritonitis or bowel ischemia.[6,8] Concerning to laboratorial evaluation, elevations of C-reactive protein and leukocytosis are frequent but non-significant unless ischemia or perforation have already developed.[6]

Imaging evaluation takes a high importance in these patients due to the unspecific of the clinical manifestations and in 36 % of cases, the correct diagnosis can be made by ultrasound; by plain radiographs in 60 %, by barium enema in 36 %, and by abdominal computed tomography (CT) in 72 % of cases.[1,6] This last one has an accuracy of 58-100 % and is the most sensitive imaging evaluation for IS and is considered the "gold-standard" imaging modality for this entity.[6,10] Some typical features of IS observed in CT scans and abdominal ultrasound may include: a "target" sign, a "sausage-

like" lesion, a reniform lump that gives a pseudo kidney appearance, and a double intestinal canal sign.[1,6,8,10]

Regarding the treatment of IS, to date, an unanimous agreement on which is the best conduct does not exist.[6,8] But it is vastly recommended an en-bloc surgical resection of the invaginated part due to the high risk of an underlying malignancy.[6,8,10] This approach can be made for laparotomy, laparoscopy or a combination of these two, and it will depend on the experience of the surgeon.[6,8] In some other cases with small bowel intussusceptions, no signs of ischemia, or in which a short gut syndrome is anticipated after resection, is recommended a conservative treatment.[6]

The aim of this article is to present a case of adult entero-enteric intussusception caused by small bowel submucosal lipoma.

CLINICAL CASE

A 66-year-old male patient with a history of high blood pressure was brought to the Emergency room complaining of abdominal pain in the periumbilical zone. This pain was like a moderate cramp and it was associated with frequent vomiting. The patient referred the expulsion of feces and gas for the rectum. On physical examination he only presented mild pain in the upper abdomen without peritoneal reaction or abdominal mass. An abdominal x-ray was taken and it showed a few hydro-aerial levels in the upper abdomen with gas in the rectum. Those manifestations were interpreted as an incomplete mechanical bowel obstruction. Therefore, a medical treatment based on decompressive measures was implemented, and the symptoms disappeared in the first 24 hours, so the patient was discharged.

Seven days after, the patient was re-admitted because he was complaining of similar signs and symptoms, but this time the medical treatment was not enough. He was suffering from intense periumbilical pain with an asymmetric upper abdominal distention. On physical examination was found peritoneal reaction and increased hydro-aerial sounds on the upper abdomen. A CT scan

(**Fig. 1**) of the abdomen was made and it showed signs of bowel obstruction with the typical "target" and "sausage-like" images.

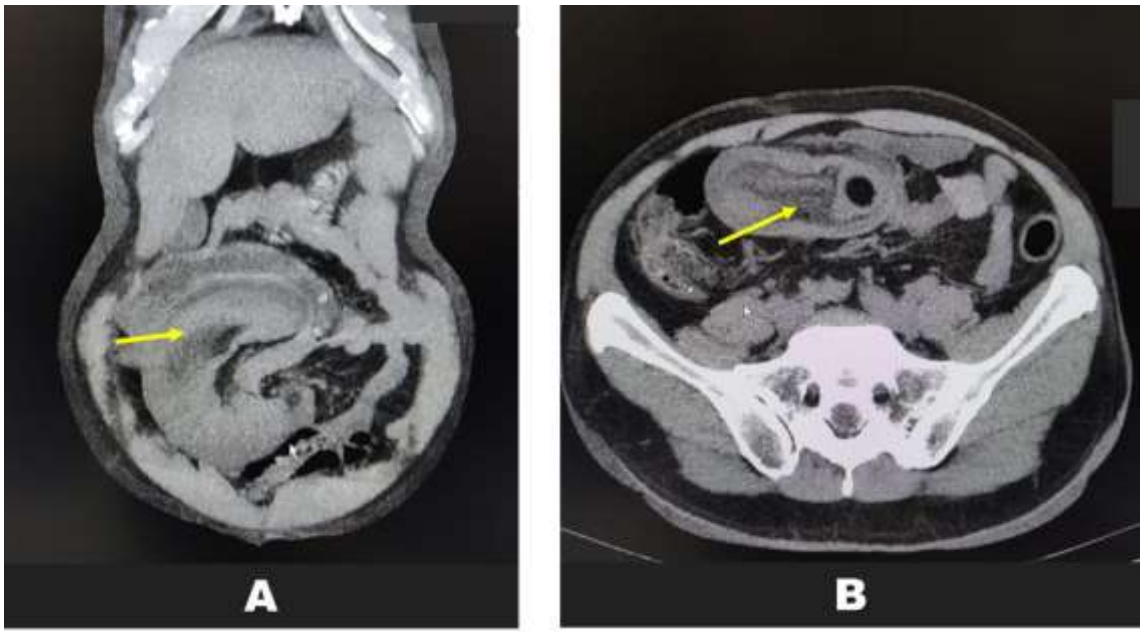

Fig. 1: CT scan. **A**: a typical "sausage-like" image. **B**: a typical "target" image.

The diagnosis of entero-enteric intussusception was made. The patient underwent laparotomic surgery where the clinical suspicion was confirmed (**Fig. 2**). An intestinal (jejunal) resection was performed with a terminus-terminal anastomosis in one continuous seromuscular non-absorbable suture. The anatomopathological examination revealed a submucosal lipoma of the jejunum as a lead point. The patient successfully recovered and was discharged 3 days after surgery without any complications until this moment.

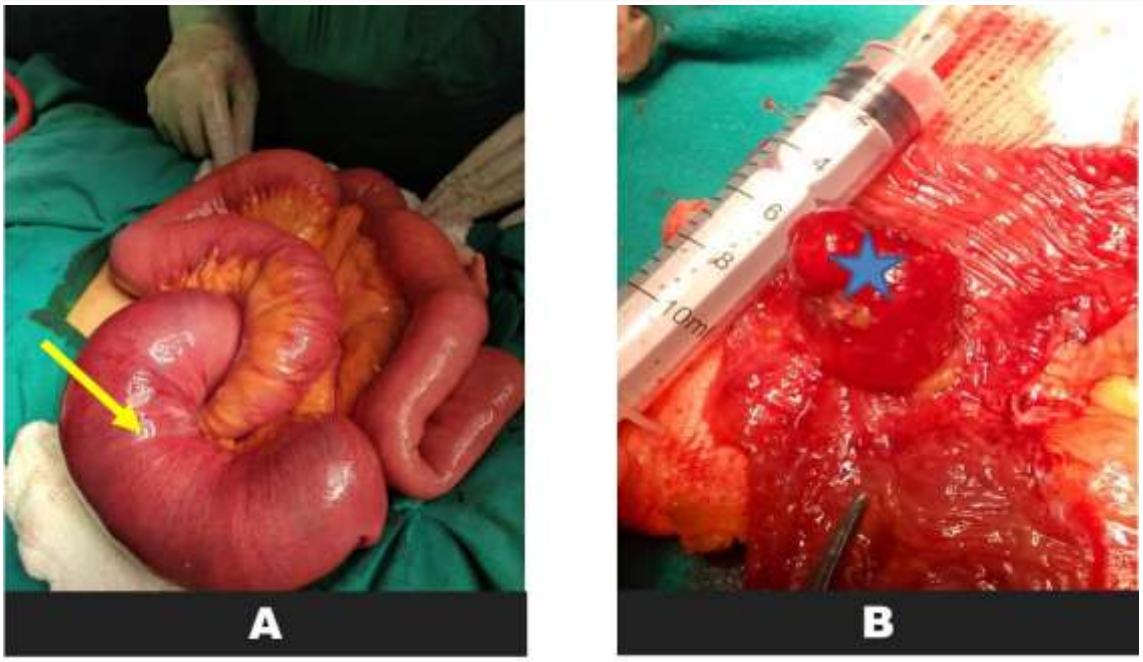

Fig.2: Laparotomic surgery. **A**: entero-enteric (jejunal) intussusception. **B**: Submucosal lipoma.

COMMENTS

Lipomas are rare, benign, slow growing tumors of mesenchymal cells and can be found wherever normal fat cells preside.[5,7,9,10] Gastrointestinal lipomas (GL) have a reported incidence of 0,035 % up to 4,4 %.[2,7,10,11] Of these almost 75 % are located in the large intestine and 30 % in the small intestine.[1,2,5,7] Small intestinal lipomas account for 2,6% of all benign gastrointestinal tumors and generally are solitary and more commonly located in the ileum (50 %) while jejunum and duodenum are the least common location.[1,2,8] A review of the literature shows only 50 cases of bowel intussusception due to lipomas reported in the first decade of the millennium, with around 21 being solitary ileal lipomas.[5]

The GL and specifically the small bowel ones are classified into three types.[7,10] The submucosal ones are the most frequent of all with an incidence of 90 % to 95 %.[1,7] This last one in junction with the subserosal commonly function as a lead point in the intussusceptum, but a large subserosal lipoma is prone to causing intestinal compression and volvulus.[1,7,10] The intramuscular ones are less common than the others types.[7,10]

GL usually presents in adults between 50 to 70 years, with no difference among males and females.[2,4,11] Those <1 cm are considered incapable of producing symptoms and in some cases are detected incidentally.[2,7,11] However, 75 % of those greater than 4 cm are symptomatic due to obstruction, hemorrhage or intussusception by acting as a lead point.[2,7,9,11] But only 32–50 % of cases are diagnosed preoperatively.[2]

The diagnosis of GL mainly depends on imaging, and barium gastrointestinal imaging can show round, ovoid, or lobulated filling defects in the lumen of the small intestine with a smooth border, no tip, variable morphology under pressure, and normal local intestinal peristalsis and mucosa.[8] In the CT scan, GL appears as a rounded low-density shadow in the intestinal lumen with CT values of −70 to 120 Hounsfield units (HU) and no enhancement after enhanced abdominal CT.[8] Taking under consideration the location of GL in the gastrointestinal tract there will be possible an endoscopic visualization by conventional, balloon, or capsule endoscopy; some endoscopic characteristics of these tumors are: a smooth, yellowish surface with pedunculated, or sessile base, "cushion sign" (the tumor indents on pressure application by biopsy forceps) and "naked fat sign" (multiple biopsies cause protrusion of yellowish fat).[5]

In the specific case of GL discussions continues as to whether or not a surgical approach is better than a medical reduction or an endoscopic resection.[2] So far, when a patient presents a large and/or symptomatic lipoma the surgical resection offers an excellent prognosis and is the standard method of treatment.[2,7] Regarding the surgical approach, it is reported that laparoscopic intervention can be used in adults with intussusception with satisfactory safety and efficacy outcomes, and the conversion rate to an open procedure ranged from 0 to 16,7 %.[2,9] Endoscopic mucosal resection appears to have a limited role in the management of small bowel lipomas.[11]

This clinical case is a rare one, not only because the intussusception is uncommon in adult patients but for the lead point found in it: a submucosal lipoma.

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