Case Report

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Small bowel obstruction secondary to intussusception due to double Meckel's diverticulum in an adult: a case report

Laís L. Menéndez Goti*, Juan P. García Godínez, Nora L. Flores Olmos

Department of General Surgery, "Hospital Regional Dr. Valentin Gomez Farias" ISSSTE, Zapopan, Jalisco, Mexico

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*Correspondence:

Dr. Laís L. Menéndez Goti,

E-mail: lais-menendez@hotmail.com

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ABSTRACT

Meckel's diverticulum (MD) is a normal variant in 2% of the population, it contains all the layers of the small intestine and is considered a true diverticulum. One of the most frequent complications in adults is obstruction secondary to intussusception of MD towards the ileum. We present the case of a 52-year-old man with severe abdominal pain, nausea, and vomiting, which did not respond to conservative management. Imaging studies compatible with small bowel obstruction, identifying the transition zone at the ileum level. An exploratory laparotomy was performed ruling an intussusception due to a diverticulum, an intestinal resection was performed. The histopathological study revealed a double MD. In adults with symptomatic MD, this represents a challenge in early diagnosis and immediate surgical treatment. Intestinal intussusception in adults secondary to MD is rare and should be considered in the differential diagnosis of those with abdominal pain and small bowel obstruction.

Keywords: Intussusception, MD, Bowel obstruction

INTRODUCTION

Intestinal intussusception is defined as the invagination of a segment of the intestine into an immediately adjacent segment. It consists of 2 components; intussusceptum which refers to proximal segment that invaginates into the intussuscipiens, which is distal or receptor segment.¹

This condition is common in children and its clinical presentation is characterized by the triad of cramping abdominal pain, bloody diarrhea, and a palpable tender mass. In adults, intussusception, on the other hand, is considered a rare condition, representing 5% of all cases of intussusception and almost 1-5% of intestinal obstruction.² The prevalence of gender is generally equal between men and women; however, a slight male predominance has been recorded.³

The cases of intussusception among adults between 8 and 20% are primary (idiopathic). Around 90% of cases are secondary to a pathological condition, such as

carcinomas, polyps, Meckel's diverticula, colonic diverticula, strictures, or benign neoplasms, which are usually discovered intraoperatively.² According to several reports, dividing the etiologies by enteric and colonic location, small bowel intussusception is frequently caused by benign lesions. In contrast, colonic intussusception is more likely to have an underlying malignant origin (often a colonic adenocarcinoma). When small bowel intussusception is induced by malignant lesions, it is often metastatic disease (carcinomatosis).⁴

Any pathological lesion of the intestinal wall that disrupts normal peristaltic activity can initiate intussusception of one segment of the intestine into another. Depending on its location, they can be classified into four categories: 1) entero-enteric, confined to the small intestine, 2) coloncolic, which affects only the large intestine, 3) ileocolic, defined as the prolapse of the terminal ileum within the ascending colon, and 4) ileo-cecal, where the ileo-cecal valve is the main point of intussusception and is distinguished with some difficulty from the ileo-colic

variant and this will depend on the junctions between the freely moving segments and the fixed areas, such as the retroperitoneum or through adhesions. 1,2,4

A schematic description of an intussusception could be of an "internal prolapse" of the proximal bowel with its mesenteric fold into the lumen of the adjacent distal bowel due to excessive or impaired peristalsis, further obstructing the free passage of intestinal contents and, seriously, compromises the mesenteric vascular flow of the invaginated segment. The result is intestinal obstruction and inflammatory changes ranging from thickening to ischemia or necrosis of the intestinal wall.^{2,4}

These inflammatory changes clinically manifest as constant crampy abdominal pain, vomiting, and abdominal distension. The patient presents with abdominal tenderness and signs of systemic inflammatory response syndrome, there may be decline or absent bowel sounds, as well as signs of peritoneal irritation, lack of gas passage, abdominal masses, and diarrhea.⁴

As previously mentioned, MD is one cause of intussusception in adults. MD is a normal variant in 2% of the population.⁵ The first report was in 1598 by Fabricius Hildanus, and subsequently named after Johan Friederich Meckel who described the anatomy and embryology of diverticulum in 1809.⁶ Embryologically, the diverticulum is the remnant of the vitelline duct, located on the antimesenteric edge of the ileum within 100 cm of the terminal ileum.^{5,6}

Small intestinal diverticula contain only mucosal and submucosal layers (and, variably, a thin serous layer) that herniate through muscular layer and therefore it classifies as false diverticula, unlike MD which it contains all the layers of small intestine and categorizes as true diverticulum. The lining epithelium is predominant of the intestinal type; however, it may present gastric, colonic, duodenal epithelium or even pancreatic tissue.⁷

Complications of MD often occur in childhood and adolescence and less often in adults. Although hemorrhage is the most common presenting symptom in children, one of the most common complications in adults is obstruction secondary to intussusception of MD into the ileum. Which manifests with abdominal pain, bloating, nausea and vomiting, with characteristic findings on physical examination such as bloating, abdominal tenderness, decreased or absent peristalsis, and even signs of peritoneal irritation.

CASE REPORT

A 52-year-old male presents to the emergency room with 24 hours history of generalized abdominal pain, cramping, VAS (visual analogue scale) 10/10, disabling, associated with nausea, and vomiting on four occasions. Past medical history of type 2 DM, long-standing smoking, and alcoholism. Regarding his surgical history,

he refers bilateral inguinal tension repair in 2005. Vital signs: BP 115/70 mmHg, HR 89 bpm, RR 18 rpm, temperature 36.8°C. On physical examination, the abdomen was globose, distended, with peristalsis present, with pain on superficial and deep palpation, generalized VAS 8/10, negative rebound sign. Laboratory tests with hemoglobin 18.1 g/dl, hematocrit 47.4%, platelets 290×10³/ul, leukocytes 8.34×10³/ul, neutrophils 77.9%, glucose 408 mg/dl, urea 46.08 mg/dl, creatinine 1.04 mg/dl, amylase 15 U/l, lipase 20 U/l. Anteroposterior projection abdominal X-ray is requested where it is observed dilation of intestinal loops, predominantly topographic in jejunum, associated with air-fluid levels in relation to small bowel obstruction, no free air or intestinal perforation data was observed (Figure 1).



Figure 1: Anteroposterior projection abdominal X-ray where it is observed dilation of intestinal loops, predominantly topographic in the jejunum, associated with air-fluid levels in relation to small bowel obstruction.

It was decided to start management with fasting, intravenous fluids as well as gastric decompression with a nasogastric tube. No improvement with conservative management was achieved. The patient is reassessed in the following shift where the physical examination calls attention to absent bowel sounds, with pain on superficial and deep palpation VAS 9/10, with muscular resistance and positive Blumberg's sign. An abdominal-pelvic

tomography with oral contrast is requested, where contrast passage to the ileum is observed, without leaks, identifying diffuse dilatation of intestinal loops with airfluid levels without inflammatory data in its wall, locating the transition zone in the ileum topography and saccular image in the wall (Figure 2-4).

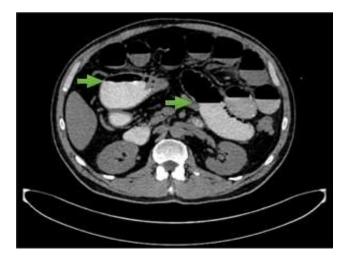


Figure 2: Abdominal-pelvic tomography with oral contrast of contrast passage to ileum is observed.



Figure 3: Abdominal-pelvic tomography with oral contrast identifying diffuse dilatation of intestinal loops with air-fluid levels (green arrow) with pneumatosis intestinalis (orange arrow).

Due to clinical deterioration and tomographic study findings, it was decided to proceed to the operating room for exploratory laparotomy, finding abundant free serohematic fluid, an extensive review of the abdominal cavity was performed, finding intussusception at 105 cm from the ileocecal valve (Figure 5), releasing invaginated intestinal portion, finding a diverticulum at 110 cm from the ileocecal valve, with 20 cm of devitalized intestinal wall tissue. A second diverticulum with no signs of complication is found at 120 cm (Figure 6), therefore 60 cm resection of small bowel is performed at 100 cm from the ileocecal valve. Subsequently, manual end-to-end

anastomosis in 2 planes, it is reinforced with Tisseel, a Blake type drain is placed, and the wall is closed with the reinforced tension line technique with no. 1 Prolene, with a total of 300 cc of bleeding.

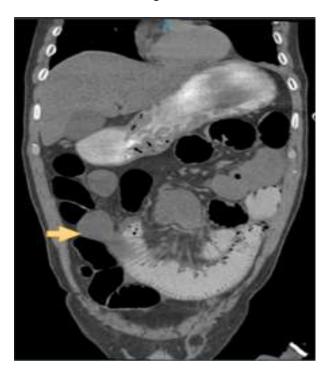


Figure 4: Abdominal-pelvic tomography with oral contrast demonstrating transition zone in the ileum topography and saccular image in the wall (yellow arrow).



Figure 5: Intussusception at 105 cm from the ileocecal valve.

The resected piece was sent to histopathological study, an intestinal portion with hemorrhagic necrosis and chronic inflammation was identified. Diverticulum with inflammation in the wall and lymphoid hyperplasia.

Gastric ectopic mucosa with oxyntic cells were detected in both diverticula, reporting two Meckel's diverticula.



Figure 6: Diverticulum at 110 cm from the ileocecal valve, with 20 cm of devitalized intestinal wall tissue. A second diverticulum with no signs of complication is found at 120 cm from the valve.

DISCUSSION

Distinct from the pediatric variety, the clinical presentation of intussusception in adults is mostly variable and nonspecific. Adult patients often present with intermittent symptoms or chronic symptoms that indicate partial bowel obstruction. The classic triad of abdominal pain, rectal bleeding, and a palpable tender mass, is no often present in all cases. Needless to say, many patients present with intermittent abdominal pain and nausea/vomiting, which is consistent with other findings. Given its rare prevalence and confusing clinical picture, the diagnosis of intussusception in adults is a challenge and can go undetected without imaging studies.³

Currently, abdominal computed tomography is the most effective imaging tool to detect intussusception, with diagnostic accuracy ranging from 58 to 100%. The typical image is composed by a thickened intestinal wall and mesentery within the lumen giving the pathognomonic "target" or "sausage" sign appearance depending on the axial projection. CT scan can identify the exact location of the intussusception and reveal any threatening signs of intestinal in viability.³

Jejunoileal diverticula usually arise at the mesenteric border of the small intestine, in places of muscle weakness, usually at the entry sites of the vasa recta, although location on the antimesenteric border has been also described. Due to their location at the mesenteric border, jejunoileal diverticula can often be located between the sheets of the mesentery, which can complicate radiographic visualization and identification.

Jejunal diverticula vary in size, usually from 1 to 4 cm, but can be as large as 8 to 9 cm. Ileal diverticula are generally smaller, ranging from 2 to 15 mm. 8,9

Diverticulosis of the small intestine is widely considered to be a rare condition. The cited incidence ranges from 0.2 to 4.5% in autopsy studies and 0.5 to 2.3% in contrast studies. Studies of ERCP and USG have cited an incidence ranging from 0.16 to 27% on ERCP and 7.5% on USG. This condition can be seen in individuals of all ages, but it is most common in adults over the age of 50. Diverticulosis is more common in the duodenum compared to the jejunum or ileum; Among the diverticula distal to the duodenum, approximately 80% were in the jejunum. The higher prevalence in the jejunum compared to the ileum is thought to be due to the presence of larger penetrating blood vessels in the proximal jejunum.

While small bowel diverticulosis can be solitary, most patients have other diverticula in other parts of the digestive tract, usually the colon. There is an approximately 2:1 male predominance of jejunoileal diverticulosis.⁸

Currently, the diagnosis is usually made based on a CT scan or an abdominal MRI. Common CT scan findings include discrete round or ovoid small bowel outpouchings outside the small bowel lumen and no recognizable small bowel folds, which may contain air, fluid, or contrast. Recently studies have suggested that MR enterography is an effective modality in diagnosis.⁹

Most patients with small bowel diverticulosis are asymptomatic and are often identified on imaging studies directed at other conditions. Jejunoileal diverticulosis is more likely than duodenal diverticulosis to be associated with chronic symptoms as well as acute complications. Bleeding is believed to be the most common complication of duodenal diverticula, while diverticulitis has been cited as the most common complication of jejunal and ileal diverticulosis. Jejunal diverticula are thought to be more prone to complications than ileal diverticula.⁸

MD is the remnant of the vitelline duct, located on the antimesenteric edge of the ileum within about 100 cm of the terminal ileum. Other medical problems resulting from MD include intussusception, which often presents first as obstruction, inflammation or diverticulitis, perforation, hemorrhage, neoplasia, and fistula.⁵

Simple radiographs are not usually helpful in making the diagnosis of MD, only being able to identify small bowel obstruction on plain abdominal radiographs. On CT, it remains a challenge. In a report of CT findings in 11 patients with Meckel's diverticulitis, the presence of gangrene or secondary small bowel obstruction was associated with poorer diagnostic acuity. Administration of both intravenous and oral contrast material can help

establish the diagnosis and should be given whenever possible. 10

Ultimately, laparoscopy, as a diagnostic tool in cases of symptomatic MD, has also been reported. Delay in the diagnosis of complicated MD can lead to significant morbidity and high mortality.¹¹

In adult patients, MD has a complication rate of 0.03% per year. Therefore, MD is usually missed in the differential of an acute abdomen in adults. In asymptomatic patients with incident MD on imaging, resection is generally not indicated. Elective surgery, however, is recommended in a selective approach when MD is found intraoperatively. Surgical complications, including deaths, have been reported. Incidental removal of MD has been reported to have morbidity and mortality rates of 3.5% and 0.2%, respectively.⁵

In symptomatic cases, morbidity increases to 5.25% and patients have a mortality risk of 1.5%. As a result, great consideration must be given when deciding the appropriate management in young and healthy patients. Factors such as male gender, age <40 years, anesthetic risks, type of surgery, size, location, length (>2 cm) and morphology (narrow implantation base) of diverticulum and the presence of a palpable mass may indicate a higher risk of complications later in life and therefore favor surgical removal of an asymptomatic MD.⁵

The treatment of MD in cases that present as symptomatic is surgical, and in patients who are asymptomatic, the therapeutic approach is controversial; it is suggested not to resect them because it increases surgical morbidity (9 to 12%) compared to 4.2 to 6.4% morbidity throughout the life of patients if it is decided not to perform resection. controversy increases in the case of adults.¹²

Cullen et al suggested eliminating incidental MD at any age up to 80 years unless additional conditions such as generalized peritonitis required surgical intervention. 12 They found that prophylactic resection postoperative complications are low, with morbidity and mortality generally related to the primary operation or the general condition of the patient and not to diverticulectomy. They also demonstrated that incidental diverticulectomy carries lower rates of operative mortality and morbidity than diverticulectomy for symptomatic DM. They believe that the complication rate due to incidental diverticulectomy is low, even with near-zero mortality, and others agree with this concept. 6

Park et al recently recommended a selective approach, advising resection of incidentally detected MD in the case of 1) patients under 50 years of age; 2) male patients; 3) MD length greater than 2 cm; 4) detection of an abnormal feature within the diverticulum. However, all the above recommendations come from the personal experience of the authors or from single center series.¹³

Three types of surgical management have been described, including resection and segmental anastomosis, wedge resection, and tangential stapling. These last two options are difficult to imagine in cases of perforation, inflammation, or hemorrhagic ulceration, which must be treated by intestinal resection.¹⁴

Whenever we find macroscopic involvement of the base of the diverticulum, the only possible laparoscopic solution is a T-shaped segmental resection with intracorporeal anastomosis. In other cases, where the diverticulum is not involved, diverticular resection with linear stapling or wedge resection seems acceptable, even if there is a small risk of leaving residual heterotopic tissues. In this setting, the mucosa of intestinal segments adjacent to the diverticulectomy should be visualized to ensure that all macroscopically visible diseased mucosa has been removed.¹⁴

The two most frequently used surgical techniques are: simple diverticulectomy or ileal resection of the affected segment with enteroenterostomy. Recently it has been considered that minimally invasive surgery for diagnosis and treatment with linear staplers has become the therapeutic option for being safe, for having good results and low morbidity and mortality rates in children as well as in adults.¹²

CONCLUSION

Small bowel obstruction in an adult patient secondary to intussusception is rare. Likewise, the finding of two Meckel's diverticula in the same patient is exceptional and there are few cases reported in the literature. Intussusception in adults, unlike in children, causes intermittent symptoms of partial bowel obstruction. MD is a remnant of the vitelline duct, located on the antimesenteric edge of the ileum within about 100 cm of the terminal ileum. Unlike the jejuno-ileal pseudoverticula that only have mucosal and submucosal layers.

In adults with symptomatic MD, it represents a challenge in early diagnosis and immediate surgical treatment. Due to the rare occurrence of these cases, their suspicion is difficult, as is their differential diagnosis, since the clinical presentation is variable. In cases of adult patients presenting with small bowel obstruction, the diagnosis is rarely made before surgery. To achieve a more accurate diagnosis, a computed tomography with oral and intravenous contrast is recommended, to observe the characteristic signs of this pathology.

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