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CLINICAL CASE

Symptomatic Meckel's Diverticulum in pediatrics

Divertículo de Meckel sintomático en pediatría

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What do we know about the subject matter of this study?

Meckel's diverticulum is the most frequent gastrointestinal malformation. It is often asymptomatic, and its different forms of presentation will correspond to the possible complications of it, so it is important to also consider atypical manifestations.

What does this study contribute to what is already known?

In relation to the 3 cases, we present different forms of manifestation of Meckel's diverticulum in pediatrics, in order to consider it as a differential diagnosis in abdominal episodes. Its study and current management are discussed.

Abstract

Meckel's diverticulum (MD) is the remnant of the vitelline duct (VD) also called omphalomesenteric duct and it is considered the most frequent gastrointestinal malformation. Most of the cases are asymptomatic and the diagnosis of this type is always a challenge. **Objective:** To describe 3 symptomatic presentations of MD and to discuss its symptoms, signs, and possible diagnostic-therapeutic tools. **Clinical Cases:** Case 1: A six-month-old patient with obstructive bowel syndrome. In exploratory laparotomy, an MD was identified with a mesodiverticular band causing an internal hernia. Case 2: A three-year-old patient presenting with digestive hemorrhage and severe anemia requiring blood transfusion. Upper gastrointestinal endoscopy did not show bleeding origin. Due to persistent melena, the patient required a new blood transfusion. An Abdomen/pelvis tomography scan was performed, showing a suspicious image of MD which was confirmed by laparotomy. Case 3: A newborn with prenatal anencephaly and omphalocele diagnosis. In immediate care of the newborn, meconium evacuation from the umbilical defect was noticed. It was managed as ruptured omphalocele, installing a bowel silo bag. In primary closure, the permeability of the omphalomesenteric duct was confirmed. An intestinal *en bloc* resection and anastomosis were performed in all 3 cases. The last one developed an anastomosis leakage resulting in a terminal ileostomy. **Conclusion:** MD, frequently asymptomatic, is often overlooked as a differential diagnosis of abdominal emergencies in children. When suspecting DM with gastric ectopic mucosa, Tc-99m pertechnetate scintigraphy should be performed as a diagnostic procedure of choice, according to each case.

Keywords:
Meckel's diverticulum;
Omphalomesenteric duct;
Intestinal Obstruction;
Omphalocele

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Introduction

Meckel's diverticulum (MD) is the most frequent congenital digestive malformation in children, with an incidence around 2-4% of the population[1,2], and is twice as frequent in males[3]. It was first described by the German anatomist Johann Friedrich Meckel (1781-1833), who, in 1809, illustrated the persistence of its embryological origin[3,5] due to an incomplete vitelline duct (VC) obliteration between the fifth and seventh week of gestation. It also appears with other less frequent alterations such as fistula, sinus, polyp, cyst, or mesodiverticular band[1], depending on the moment the normal involution stops.

The MD is a true diverticulum located along the small intestine in its antimesenteric border, and it is frequently found in the terminal ileum approximately 60 cm from the ileocecal valve. It is around 2 cm long[3-5] and is also characterized by the presence of heterotopic gastric mucosa in 45 to 80% of the surgical specimens on the histopathological study, mainly gastric and pancreatic mucosa[1,6]. Most of the cases are asymptomatic (about 95.2%)[7] and are usually diagnosed accidentally in abdominal explorations due to other causes[1], whereas its symptomatic presentation depends on the type of the diverticulum's complication, appearing, according to different series, around 2 years of age[4] as gastrointestinal bleeding (30 - 56%), intestinal obstruction (14 - 42%), diverticulitis (6 - 14%), and intussusception[8]. The natural evolution of the MD's complications can be life-threatening, so its early diagnosis and management are crucial[1,3,4].

Unfortunately, its varied and non-specific clinical manifestations such as pain, nausea, vomiting, abdominal distension, gastrointestinal bleeding, among others, often hinder an assertive diagnosis. On the other hand, imaging studies such as abdominal radiography, ultrasound, and CT scan or Tc-99m pertechnetate scintigraphy can be falsely negative and positive. The objective of this report is to describe 3 forms of presentation of symptomatic MD, discuss its symptoms, signs, and possible diagnostic and therapeutic tools.

Clinical Cases

Clinical case 1:

A 6-month-old patient with no history of morbidities or previous surgeries consulted in the emergency department due to 2-day history of food refusal, vomiting, progressive deterioration of the general condition, and lack of stool elimination. Blood tests were performed (blood count, metabolic panel, and C-reactive protein (CRP)) all within the normal range, and an abdominal X-ray in decubitus and standing position (Figure 1) which showed distension of the bowel loops at the central level, scarce air at distal level, and some air-fluid levels. The patient was hospitalized, placing a gastric tube, which showed little bilious residuals. Later, a new abdominal X-ray was performed which showed two large dilated bowel loops with obstructive signs (Figure 2).

With the diagnosis of intestinal obstruction, it was decided to perform an exploratory laparotomy where MD was identified with a mesodiverticular band (Figure 3), forming an internal hernia in the distal intestinal loops, which are clearly defuncioned, with no ischemic compromise. Excision of the fibrous band, *en bloc* resection of the MD, and end-to-end ileal anastomosis were performed.

Progressive oral feeding was reincorporated on the 5th postoperative day with good tolerance and the patient evolved satisfactorily. The biopsy report concluded MD with ileal mucosa.

Clinical case 2:

A 3-year-old patient, with no history of morbidities, consulted in the emergency department due to 2-day history of abdominal pain associated with blood-streaked stools. Blood tests showed normocytic normochromic regenerative anemia (hematocrit 18.3%), and platelets and coagulation tests were normal. The patient received a red blood cell transfusion and was hospitalized for study and management.

Upper gastrointestinal endoscopy showed no evidence of bleeding. During the first day of hospitalization, the patient presented frank and abundant melenic stools, requiring a new transfusion of red blood cells. Since there was no quick access to scintigraphy, the study was complemented with abdominal/pelvic CT angiography, which showed a central abdominal image with a marked mucosal impregnation of contrast, suspicious of MD. It was decided to perform an exploratory laparotomy confirming diverticular lesion at the ileal level with characteristics compatible with MD (Figure 4). The diverticulum showed some congestive signs, thickened walls, and multiple adhesions to the mesentery. Release of adhesions, *en bloc* resection of the diverticulum, and end-to-end anastomosis were performed.

The patient evolved favorably and resolved gastrointestinal bleeding, reincorporating oral feeding on the 5th postoperative day. The biopsy report concluded MD with ectopic gastric mucosa.

Clinical case 3:

Newborn of 37 weeks of gestational age with prenatal diagnosis of anencephaly and omphalocele, viable,

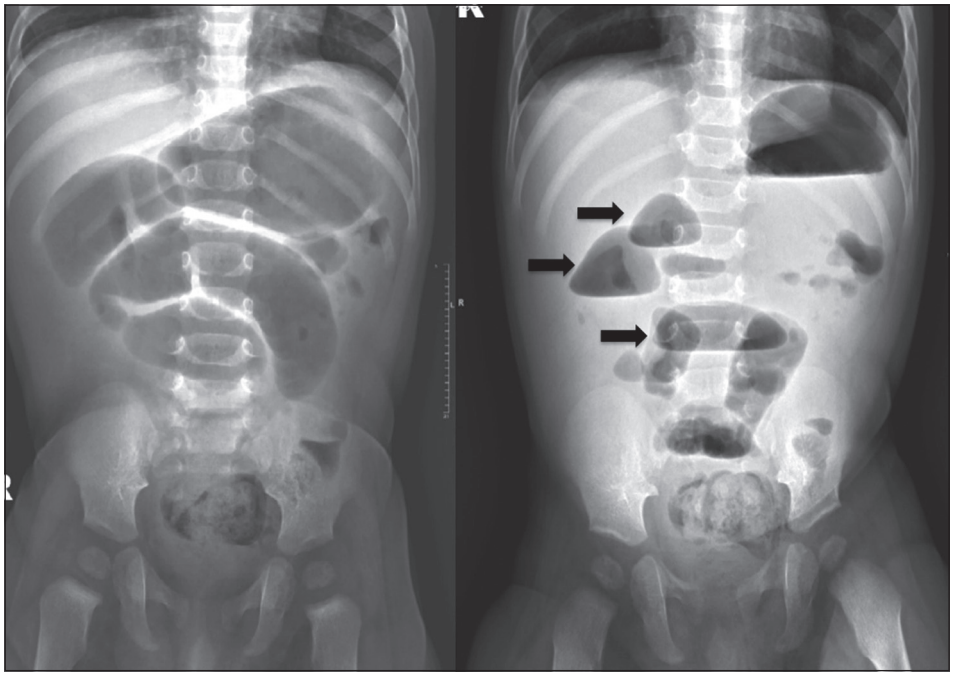

Figure 1. Case 1. Anteroposterior supine Abdominal X-ray (left) and erect position (right). Dilatated bowel loops and air-fluid levels (arrows).

and born by cesarean section due to breech presentation. Immediate care showed an omphalocele with herniated bowel loops and meconium coming out of one of its edges. A silo with a transfusion bag was placed.

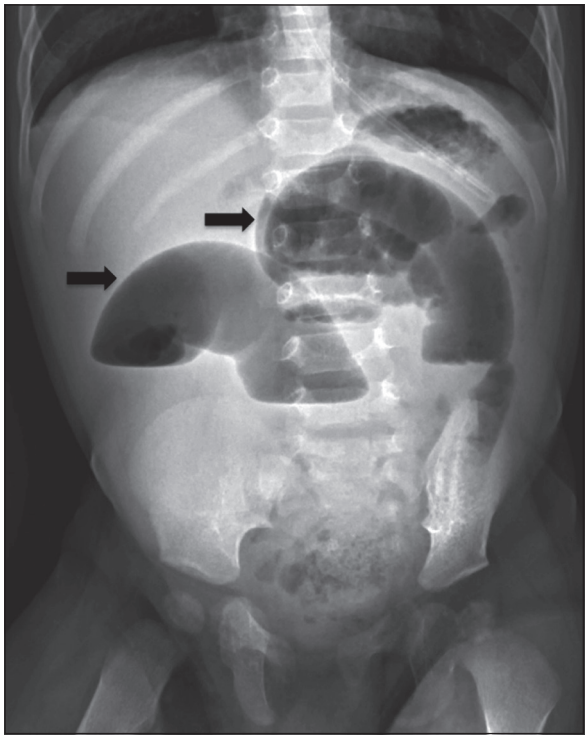

Figure 2. Case 2. Anteroposterior erect Abdominal X-ray. Dilatated small bowel loops with air-fluid levels (arrows) and scarce distal gas.

The same day, the patient was admitted to the operating room with a diagnosis of ruptured omphalocele for primary closure of the defect. Persistent vitelline duct was identified (Figure 5) which was permeable and drained meconium. Ileal resection was performed including the persistence, end-to-end anastomosis, and closure of the abdominal wall defect without incident.

The biopsy report describes a segment of the terminal ileum with open diverticular dilatation located in

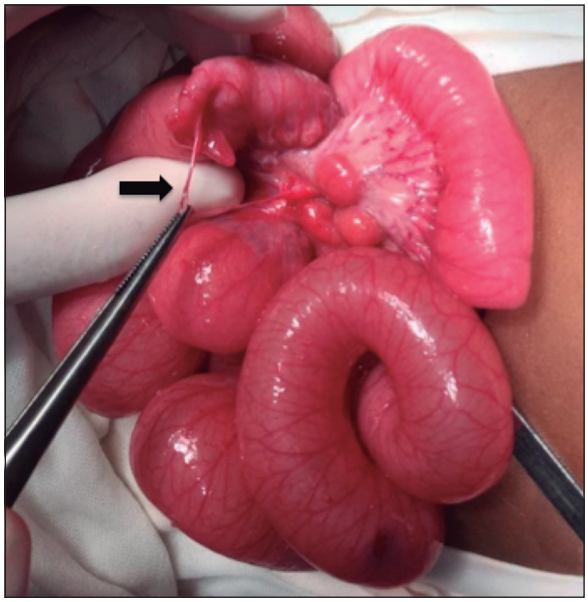

Figure 3. Case 1. Mesodiverticular band causing an internal hernia (arrow).

the antimesenteric border suggestive of MD, without ectopic mucosa. On the fifth postoperative day, the patient started progressive enteral stimulation with breastfeeding, tolerated regularly until achieving a volume of 120 cc/kg/day.

At 15 days of life, the patient presented sudden abdominal distension, in addition to an increase in blood inflammatory parameters with leukocytes 15.35 mil/mm^3, Bacilliform 20%, Segmented 39.4%, Lymphocytes 16.9%, and CRP 26.8 mg/dL. An abdominal X-ray showed obstructive signs. Exploratory laparotomy was performed showing dehiscence of the previous anastomosis associated with an adhesive obstructive process at this level and fibroadhesive meconium peritonitis. Careful release of adhesions, *en bloc* resection of the compromised ileal segment was performed as well as an end ileostomy.

The patient evolved favorably with parenteral nutritional support initially, receiving progressive oral enteral stimulation on the 5th postoperative day, with good tolerance, and then was discharged receiving complete oral feeding. Currently is awaiting transit reconstitution.

Discussion

MD is a well-known entity in pediatric surgery, of low overall incidence, and frequently asymptomatic. However, when symptomatic, it presents a wide range of clinical manifestations and possible complications including severe forms. Among its presentations are gastrointestinal bleeding, inflammation (diverticulitis), and intestinal obstruction; either by adhesions, intussusception, or midgut volvulus[1,5,9].

This series shows three different forms of presentation, each at different age of onset. The first case presented an intestinal obstruction secondary to an omphalomesenteric adhesion. Intestinal obstruction in all its variants is one of the most frequent symptoms, accounting for approximately 35-47% in the different series[1,3,5]. These symptoms generally occur at school age, although intestinal obstruction secondary to MD is less frequent in adults. The omphalomesenteric adhesion is a rare cause of intestinal obstruction and the main cause is intestinal intussusception, with the diverticulum serve as a lead point. Despite its infrequency, cases of bowel incarceration and necrosis of intestinal segments secondary to obstructive symptoms due to this adhesion have been reported[1]. It should be mention that it is a cause of intestinal obstruction that, despite being unusual, should be considered in a pediatric population with no surgical history, as was reported in the first case.

The second case presented gastrointestinal bleeding, which is the classic symptom of MD, corresponding to 25-35% of the symptomatic forms[5,10]. In 50-65% of cases, MD presents heterotopic mucosa, where gastric and pancreatic mucosa are the most frequent, and even a combination of both is described in up to 2% of cases. Gastrointestinal bleeding occurs mainly in cases of ectopic gastric mucosa due to irritation and even peptic ulceration of the adjacent intestinal mucosa, secondary to the secretion produced by the ectopic one. Classically, it appears in preschool age as the case described, however, cases in adult patients have also been described.

Although intestinal bleeding is described as hematochezia, in clinical practice it can appear in many

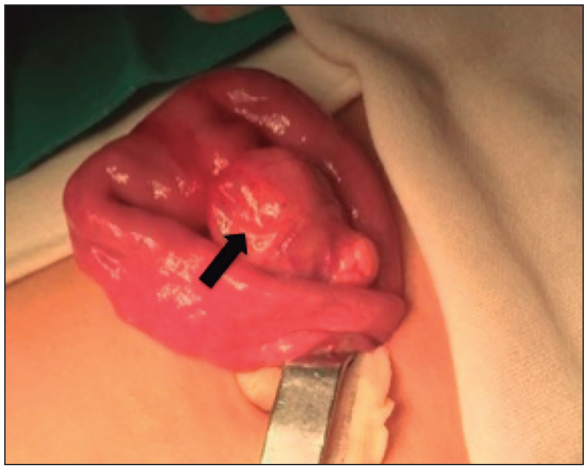

Figure 4. Case 2. Distal ileal diverticular lesion compatible with Meckel's Diverticulum (arrow).

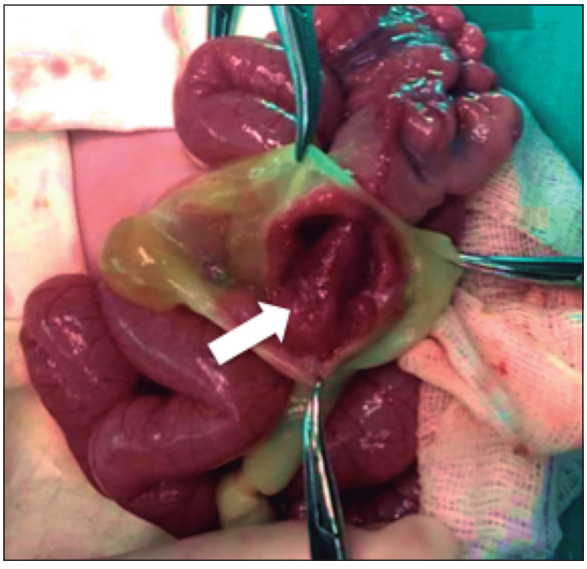

Figure 5. Case 3. Omphaloenteric duct persistence and it's permeability (arrow).

different ways, including melena, rectal bleeding in different amounts, and acute, subacute, or chronic forms, so MD should be suspected in all pediatric patients with gastrointestinal bleeding of unspecified origin[11].

Finally, the third case reports an omphalocele associated with a permeable omphalomesenteric remnant, which is an atypical presentation. In 16% of patients with omphaloceles, it is associated with MD, more frequently in small omphaloceles, and only in 0.9% as a permeable persistence, as in our case[12]. Although this is unusual, it should be considered in the management of these patients at birth.

There are different tests for diagnostic confirmation of MD. Simple abdominal radiography has poor performance unless the complication is intestinal obstruction. Ultrasonography is also of little use except in the case of intestinal intussusception. The test of choice in patients with intestinal bleeding is Tc-99m pertechnetate scintigraphy, commonly known as "Meckel's scan", where the radioisotope is preferentially captured by the mucus-secreting cells of the gastric mucosa and heterotopic gastric tissue in the diverticulum[11]. Given this, its diagnostic usefulness is limited only for those cases in which ectopic gastric mucosa is present, with a sensitivity and specificity of 94% and 97%, respectively[14]. The results can be improved using premedication with histamine H2 receptor blockers (ranitidine, cimetidine, famotidine), proton-pump inhibitors, and glucagon[15]. In the cases described, both due to the limited availability and the general condition of the patients, it was not possible to perform this test. Abdominal tomography is usually of low performance since it is difficult to identify MD in the absence of complications[13]. In our second case, it was the impregnation with a radioisotope that made it possible to identify the complicated diverticular structure.

Once the diagnosis has been established, the surgical alternatives are divided into categories; a complicated MD must be explored surgically, either open or laparoscopically. Surgery consists of resection of the diverticulum in isolation (diverticulectomy) or associated with the adjacent intestinal portion. The decision will depend on the intestinal compromise associated with the diverticulum at the time of surgery. *En bloc* resection is considered safer than isolated or wedge diverticulectomy since the latter can leave diverticulum margins with ectopic mucosa. The case of asymptomatic MD is different, in which the surgical approach is not so clear.

It is subject of discussion whether an MD incidentally identified at laparotomy should be removed or not. Some trends support its removal in order to avoid future complications, which have been reported in 4.2%[6]. In contrast, another trend recommends expectant management if the laparotomy was due to another clear cause since resection will result in greater perioperative morbidity and the complication rate of asymptomatic MD is very low.

Conclusion

MD is the most frequent congenital digestive malformation and, due to its wide clinical variety, it is important to know its spectrum of presentation and to have a high index of suspicion, in order to start an early study that allows adequate and timely management to minimize the risk of morbidity and mortality.

Ethical Responsibilities

Human Beings and animals protection: Disclosure the authors state that the procedures were followed according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

Data confidentiality: The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.

Rights to privacy and informed consent: The authors have obtained the informed consent of the patients and/or subjects referred to in the article. This document is in the possession of the correspondence author.

Conflicts of Interest

Authors declare no conflict of interest regarding the present study.

Financial Disclosure

Authors state that no economic support has been associated with the present study.

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