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Case Report

INTESTINAL DUPLICATION IN THE ADULT: CASE REPORT OF AN ATYPICAL PRESENTATION IN AN UNCOMMON PATHOLOGY

DUPLICACIÓN INTESTINAL EN EL ADULTO: REPORTE DE CASO DE PRESENTACIÓN ATÍPICA EN UNA PATOLOGÍA POCO FRECUENTE

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Abstract

Intestinal duplication is an uncommon congenital abnormality, predominantly occurring in children under two years, these duplications may present as intestinal obstruction, acute abdomen. A 34-year-old male patient who consulted for lower abdominal pain and hematochezia following mild blunt abdominal trauma during sports. Initial endoscopic and abdominal angiotomography studies did not reveal a bleeding site; however, his hemodynamic status progressively deteriorated, requiring vasopressor support and hemoderivatives. An exploratory laparoscopy was performed, revealing an unexpected finding: a segment of intestinal duplication which was resected and confirmed pathologically. Intestinal duplication cases have variable presentations, as discussed in this article, with surgical intervention being the standard management to prevent future complications and malignant transformation.

Resumen

La duplicación intestinal es una anomalía congénita infrecuente, cuya presentación es predominante en niños menores de dos años, debutando con obstrucción intestinal o abdomen agudo. Paciente masculino de 34 años quien consulta por dolor abdominal en hemiabdomen inferior y hematoquecia, posterior a trauma cerrado leve en abdomen durante actividad deportiva. Se realizaron estudios iniciales endoscópico y angiotomografía de abdomen sin evidenciar sitio de sangrado, sin embargo con deterioro progresivo de estado hemodinámico requiriendo soporte vasopresor y hemoderivados. Se realizó una laparoscopia exploratoria con hallazgos de duplicación intestinal, que fue resecada y enviada a patología con resultado confirmatorio. Los casos de duplicación intestinal tienen presentaciones variables como se expone en éste artículo, la intervención quirúrgica representa el manejo estándar, evitando complicaciones futuras y transformación maligna.

Keywords: Congenital abnormality, Gastrointestinal bleeding, Small intestine, Intestinal duplication.

Introduction

Duplications of the gastrointestinal tract are congenital abnormalities previously termed "enteric cysts", "double ileum", "giant diverticulum" and "abnormal Meckel's diverticulum". The vast majority of cases are located in the small intestine, particularly in the ileum (60%), followed by the esophagus, rectum, and colon (6.8%) [1,2]. But it can be located anywhere around the gastrointestinal tract, from the esophagus to the anus [1,2].

The reported incidence in recent years is 1/4500 live birth, without a known association by sex or race [3,4]. The most frequent location is on the mesenteric border of the associated intestine; however, they can be found adjacent to the gastrointestinal wall or as part of the lumen [1,3]. Intestinal duplications vary in shape and size: 80% are cystic, and the remaining 20% are tubular, sharing the same blood supply with the associated intestine [1]. They may be accompanied by other congenital abnormalities, primarily genitourinary and gastrointestinal.

Nearly 80% of cases occur in the pediatric population under 2 years old [1], with the usual clinical manifestation being acute abdominal pain or intestinal obstruction [5]. Asymptomatic patients are diagnosed in adulthood as incidental findings or when presenting complications such as volvulus, gastrointestinal bleeding, perforation, fistula, or malignant transformation, particularly with colonic location [5,6].

Hemorrhagic complications of intestinal duplication are unusual in adults. We present a case of a 34-year-old patient with lower gastrointestinal bleeding secondary to blunt abdominal trauma, without evidence of the bleeding site identified on upper and lower gastrointestinal endoscopy, leading to exploratory laparoscopy and finding of intestinal duplication in the mid intestine, which was surgically repaired and resected.

This case report is presented according to the 2023 Surgical Case Report Guidelines (SCARE)[7].

Presentation of the case

A 34-year-old male patient with no medical history consulted after four days of mild blunt abdominal trauma during soccer practice, and subsequent onset of rectal bleeding and hematochezia. He initially consulted at a primary care hospital without clinical findings of gastrointestinal bleeding and a hemoglobin (Hb) of 8.4 g/dL, he was referred to a secondary-level hospital where underwent an upper endoscopy (EGD) and colonoscopy without evidence

of a bleeding site, and outpatient conservative management was decided.

He consulted again 10 days later with persistent symptoms, dizziness, and syncope, he was admitted with vital signs: Blood Pressure (BP): 85/56 mmHg, heart rate (HR): 106 bpm, respiratory rate (RR): 21 rpm, generalized skin pallor, with admission laboratories showing Hb: 6.2 g/dL, metabolic acidosis with hyperlactatemia, configuring hypovolemic hemorrhagic shock. He was transferred to the Intensive Care Unit (ICU), where new endoscopic studies (EGD and colonoscopy) were performed, without evidence of bleeding site but abundant bloody remnants throughout the path. He received a transfusion of 4 units of packed red blood cells (PRBC), with post-transfusion Hb of 7.3 g/dL, suggestive of active bleeding.

An abdominal angiography with contrast was performed without being able to identify the bleeding site. A mesenteric arteriography was performed but was negative, ruling out the possibility of percutaneous management. He had another episode of rectal bleeding and melena, associated with hemodynamic instability (BP: 104/58 mmHg, HR: 130 bpm, RR: 20 rpm), worsening despite transfusion support. Given persistent bleeding without an evident cause, urgent diagnostic laparoscopy was decided, revealing at 170 cm from the ileocecal valve, a diverticular-like prolongation of the small intestine with mesentery, 2x7 cm in size, with a 2 cm pedicle and containing abundant clots inside (Image 1-2).

Bowel exteriorization was performed through umbilical port extension using an Alexis device, followed by intestinal section containing the surgical piece and an antiperistaltic side-to-side anastomosis with a 75 mm linear cutting stapler and reinforcement with simple invaginating PDS 3-0 invaginating sutures.

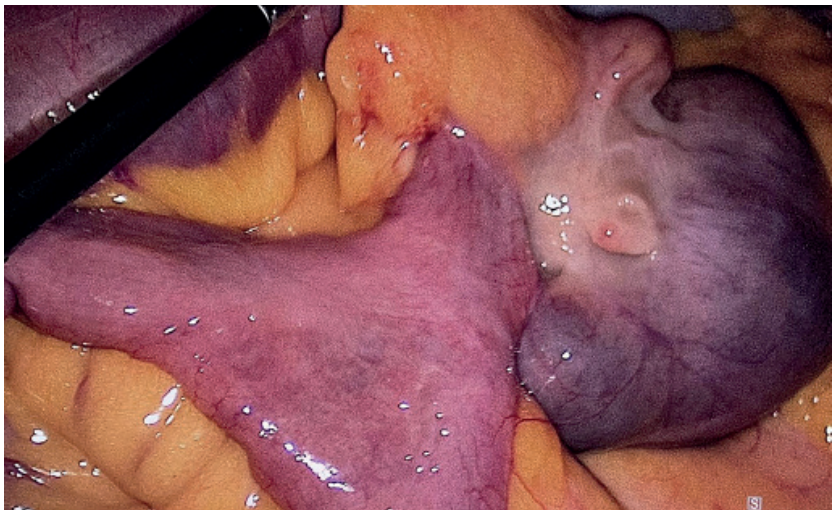

Image 1. Laparoscopic view of the surgical finding. Source: authors.

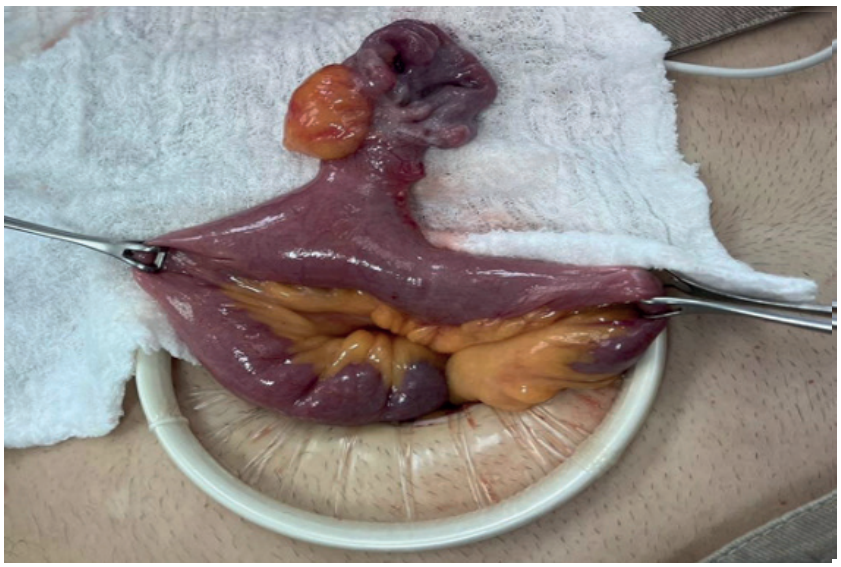

Image 2. Exteriorized surgical finding. Source: authors.

During the procedure, the patient required 5 units of cryoprecipitate, 4 units of PRBC, and 6 units of plasma. Postoperatively, the patient was monitored in the ICU for 72 hours with adequate clinical evolution, hemodynamic stability, and no new episodes of gastrointestinal bleeding. He was transferred to the general ward with an adequate postoperative course and discharged after 4 days of hospital stay. At a 10-day follow-up, the pathology report was reviewed confirming a structure with usual intestinal wall histology and viable resection margins, compatible with intestinal duplication.

Discussion

Gastrointestinal duplications are rare congenital abnormalities with various theories proposed to establish their etiology, such as vascular lesions, defects in embryonic development, and abnormal recanalization [2,8]. They can vary in size, location, and symptoms, predominantly presenting in the pediatric population; however, some patients manifest symptoms or complications in adulthood [4].

These congenital anomalies have distinctive characteristics: 1) an epithelial layer of the gastrointestinal tract, 2) a well-defined smooth muscle wall, 3) close proximity to a gastrointestinal tract structure, sharing the wall, and 4) may or may not have communication with the adjacent gastrointestinal structure [9].

Intestinal duplications can be diagnosed prenatally when the malformation generates intestinal obstruction and consequently polyhydramnios [10]. However, only 20% - 30% of cases are diagnosed at this stage and if diagnosed it occurs between the second and third trimesters of pregnancy [10]. In the pediatric population, typical symptoms include vomiting from intestinal obstruction, abdominal masses, rectal bleeding, and peritonitis [8].

In adults, most cases are asymptomatic and incidental findings. Clinical manifestations vary and depend on the duplication's characteristics. Symptomatic patients may present with acute abdomen, volvulus, gastrointestinal bleeding, or intestinal obstruction [1]. Abdominal pain is non-specific, associated with nausea, vomiting, signs of intestinal obstruction, and may have gastrointestinal bleeding [1].

In the reported patient, after blunt abdominal trauma, he developed as a complication lower gastrointestinal bleeding in the mid intestine developed, with no endoscopic diagnosis or imaging identification possible. Thus, diagnostic approaches in adults can be challenging due to nonspecific clinical presentation, low suspicion of these anomalies, and diagnostic imaging sensitivity depending on the duplication site.

Regarding imaging approaches, ultrasound can be useful, with the "double wall" sign described, which is formed by the echogenic inner muscular layer and hypoechoic outer layer, with high sensitivity for this pathology [11]. Similarly, computed tomography and magnetic resonance imaging can show a non-calcified cystic structure displacing or not other organs or signs mimicking an intussusception, their bigger benefit is providing information around the location and the possible complications [12]. In 2013, the 99 m Tc - pertechnetate scintigraphy was described as an option for the diagnosis, showing an uptake at the level of the duplication, related to gastric mucosa [13]. However it is not a spot uptake. It seems that images are a tool to provide additional information around the diagnosis but they are not the key.

In the attempt of getting an earlier and more accurate diagnosis there have been described some endoscopic alternatives. Taking into account the more frequent locations, the double balloon enteroscopy can be considered the first diagnosis choice. Even more than the capsule endoscopy, considering the retention risk, higher in these patients, the easier detection and characterization of the intestinal duplication [14].

On the other hand, endoscopic and colonic studies in cases of gastrointestinal bleeding allow direct visualization of the mucosa, taking of samples for histopathological analysis, and exclusion of communications with the colonic wall [12]. Oncological pathologies should be considered as differential diagnoses, since adenocarcinomas, squamous cell tumors, neuroendocrine tumors, and even pseudomyxoma have been documented within the duplicated segmenta [6].

The treatment of choice is surgical resection of the duplicated segment, ideally early, to prevent complications such as intestinal perforation, bleeding, obstruction, and/or malignant transformation [1]. The extension of the resection can be determined using a double balloon enteroscopy [14]. In the case presented, the endoscopic approach was inconclusive, necessitating exploratory laparoscopy for diagnosis and treatment.

At the moment of the surgical approach, there are two alternatives, an enucleation and a resection with anastomosis. Enucleation was described first for locations where a resection implies a big morbidity, for example in esophagus, duodenum or ileocecal junction. In 2024 Laplanche et al published a retrospective monocentric study comparing these techniques. Finding that the length of stay was shorter in resection and anastomosis group, without differences for postoperative complications [15]. There is another surgical technique, described in 1963 by Wrenn, that consist in a dissection on the submucosal layer, removing all the mucosa of the intestinal duplication, in order to avoid the short bowel syndrome, specially in cases that involve long tubular intestinal duplications [16].

Conclusion

Gastrointestinal duplications are congenital abnormalities that can affect any site in the gastrointestinal tract. They are rare and usually diagnosed in the pediatric population, with some cases presenting in adulthood with nonspecific acute abdominal pain. The diagnosis is challenging because of the non-specificity of the presentation, making diagnostic and therapeutic exploration through surgery the best decision. The early diagnosis is a key point in the clinical approach, due to the high index of complications in advanced stages. There is a wide range of complications depending on the location in the gastrointestinal tract. But all these morbidity, including gastrointestinal bleeding, peritonitis and malignant transformation, associated with intestinal duplications, can be prevent promoting a promptly diagnosis and treatment. Finally the best treatment decision must take into account all around the duplication features and patients clinical status.

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