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CLINICAL CASE

Neonatal intestinal obstruction due to transmesenteric hernia with jejunoileal atresia: an unusual etiology

Obstrucción intestinal neonatal por hernia transmesentérica con atresia yeyunoileal: una etiología inusual

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

Transmesenteric hernia is a sacless internal hernia through a congenital defect of the mesentery and is a rare cause of intestinal atresia. Its presentation encompasses several pictures within the spectrum of partial or complete intestinal obstruction syndrome.

What does this study contribute to what is already known?

This clinical case reinforces the concept that in the presence of neonatal intestinal obstruction, an adequate differential diagnosis should be made excluding the most common causes of intestinal occlusion, without excluding unusual but potentially serious conditions such as transmesenteric hernia, whether or not associated with intestinal atresia. The clinical and radiological non-specificity of this malformation, and meconium expulsion, could lead to diagnostic-therapeutic delays and increase morbidity and mortality.

Abstract

Transmesenteric hernia is an internal hernia without a sac caused by a congenital defect of the mesentery. It is a rare cause of intestinal atresia, usually diagnosed intraoperatively, therefore, its prognosis is variable and may be associated with high morbidity and mortality. **Objective**: To report a case of transmesenteric hernia with multiple intestinal atresia of late diagnosis. **Clinical Case**: Male newborn, born at term, referred due to vomiting, scanty bowel movements, and abdominal distention. At 8 days of age and after excluding various causes of abdominal distention, the patient underwent exploratory laparotomy, identifying a transmesenteric hernia and two sites of intestinal atresia. Resection of the atretic segment and primary anastomosis were performed, with good evolution. **Con-**

Keywords:
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clusions: In the presence of neonatal intestinal obstruction, an appropriate differential diagnosis should be made, excluding the most frequent causes of intestinal obstruction, without leaving aside those unusual but potentially serious conditions, such as transmesenteric hernia associated or not with intestinal atresia.

Introduction

Congenital internal hernias can present with various postnatal clinical pictures within the spectrum of partial or complete intestinal obstruction syndrome and are classified according to their location as paraduodenal (with or without intestinal malrotation), pericecal, intersigmoid, transmesocolic, transhiatal (Winslow's hiatus), and transmesenteric[1]. This last form occurs in 0.6-5.8% of all cases of pediatric lower intestinal obstruction[2]. The objective of this work is to report the case of a newborn with symptoms suggestive of lower intestinal obstruction in whom the surgical diagnosis of transmesenteric congenital internal hernia was established as a cause of multiple intestinal atresias.

Clinical Case

Male infant born at 38 weeks of gestation and without prenatal obstetric check-ups, born from an uncomplicated eutocic delivery, weighing 2678 grams, and Apgar score 7 and 8 at one minute and five minutes of life, respectively; referred due to suspected necrotizing enterocolitis at seven days of life. At birth, in his hospital of origin, external congenital malformations were ruled out and an orogastric tube was placed to verify esophageal patency, obtaining 200 mL of amniotic fluid. Subsequently, he started oral feeding but due to weak suction and bilious vomiting, he was admitted to the neonatal intensive care unit. At 4 hours of life, abdominal radiography was performed and an atypical double bubble image was observed (figure 1A), for which a radiological control was performed 48 hours later, identifying apparently distal intestinal gas, and he presented meconiorrhexis at 50 hours of life (figure 1B). Two days later, the patient restarted oral feeding, after which he presented again bilious vomiting and abdominal distension that improved with fasting and orogastric tube; therefore, necrotizing enterocolitis was suspected and absolute diet, total parenteral nutrition, and antibiotics therapy with ampicillin and amikacin were indicated. After seven days of life and due to the persistence of symptoms despite the treatment administered, he was referred to our center. On admission, he presented abdominal distension, without bowel sounds or signs of peritoneal irritation, so an intestinal transit test was performed without distal progression and opaque enema which showed microcolon, all suggestive of jejunoileal atresia (figure 1C, D).

After performing imaging studies and with the suspicion of intestinal atresia, an exploratory laparotomy was performed through a transverse supraumbilical approach, observing hepato-ileal adhesions, an internal hernia through a mesenteric defect with incarceration of a jejunal loop and type IV jejunoileal atresia (figure 2A). Two sites of atresia were identified 56 cm from the angle of Treitz and 36 cm from the ileocecal valve, with a 10 cm interatresic segment with intraluminal meconium and color changes with a violaceous appearance but without perforation; the difference in proximal to distal intestinal diameters had a 3:1 ratio (figure 2B). It was decided to dissect the mesentery and resect the blind loop (10 cm) to perform a jejunoileal end-to-oblique anastomosis, corroborating distal patency with intraluminal catheter and administration of physiological solution until he presented rectal evacuation (figure 2 C, D).

The histopathology report confirmed a type IV jejunoileal atresia with mild chronic enteritis. The patient evolved favorably and, on the 5th postoperative day, he started oral feeding with adequate progressive tolerance and gradual decrease of total parenteral nutrition with withdrawal on the 8th postoperative day. On the 6th postoperative day, he presented spontaneous stools, and 20 days after surgery he was discharged without complications. Twelve months later the patient continues to be asymptomatic with adequate weight gain and tolerance to complementary feeding with normal daily stools.

Discussion

Transmesenteric internal hernia was first described by Rokitanski in 1836[3]. It occurs in approximately 12% of all cases of congenital intra-abdominal herniation. The most frequent location is the mesentery of the terminal ileum, and it was Sir Frederik Treves who, in 1885, called attention to the defects located in the terminal mesentery and proposed its congenital origin[4]. It has been more commonly observed in males and about half of the cases consequently suffer ischemic intesti-

nal necrosis. Congenital mesenteric defects may be associated with intestinal atresia in up to 5.5% of cases, malrotation, intestinal duplication, Hirschsprung's disease, and cystic fibrosis in the rest, suggesting a genetic etiology[5].

In the newborn, hernias through mesenteric defects have clinical signs and symptoms similar to those of middle volvulus secondary to malrotation. These signs may be indistinguishable from intestinal atresia and, as in our case, potentially be the conditioning factors for it. Intestinal mesenteric malformations as a cause of intestinal atresia, are unified under the same etiology that causes intestinal atresia based on a vascular accident that produces a mesenteric defect where internal herniation occurs, subsequently there is scarring of the defect over the herniated intestinal loop generating atretic sites[6,7]. However, it is not possible to establish the pathophysiological chronology with which the malformations develop or if the ischemic event has originated both the intestinal atresia and the mesenteric gap and subsequently the internal herniation occurs, or if the atresia is a consequence of the loop trapped in the mesenteric defect.

Internal transmesenteric hernias are a rare cause of intestinal atresia and their diagnosis is usually intraoperative. It is not known how the hole through which the intestine is introduced is formed; however, a phenomenon that determines very selective ischemia during the differentiation stage could be the reason[8,9]. We can suggest that they are late mesenteric vascular accidents, probably from the second trimester of gestation, after the recanalization of the intestinal lumen and the initiation of meconium production with subsequent herniation of the already formed intestinal loop, as shown by the surgical findings of our patient, where intraluminal meconium was identified in the atretic segment, entrapment of an intestinal segment of similar caliber to the proximal segment, which is extremely infrequent since the distal atretic segment is usually hypotrophic and above all because there was

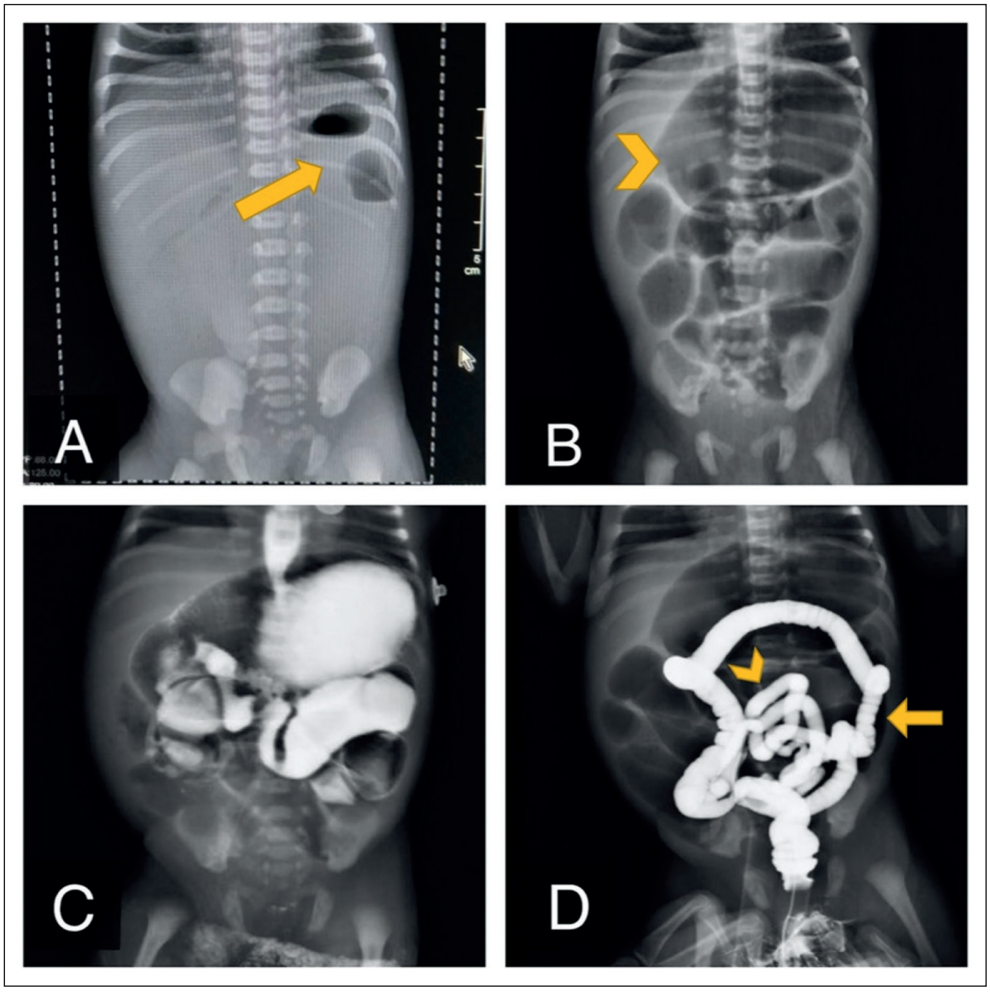

Figure 1. Radiographic Series. **A**. Plain abdominal radiograph at 4 hours of life showing an "atypical" double bubble (arrow) and absence of distal air. **B**. Plain radiograph at 48 hours of life displaying gastromegaly (arrowhead) and intestinal dilation. **C**. Intestinal transit study showing poor distribution of distal contrast. **D**. Opaque enema revealing microcolon (arrow) with reflux into the distal ileum (arrowhead).

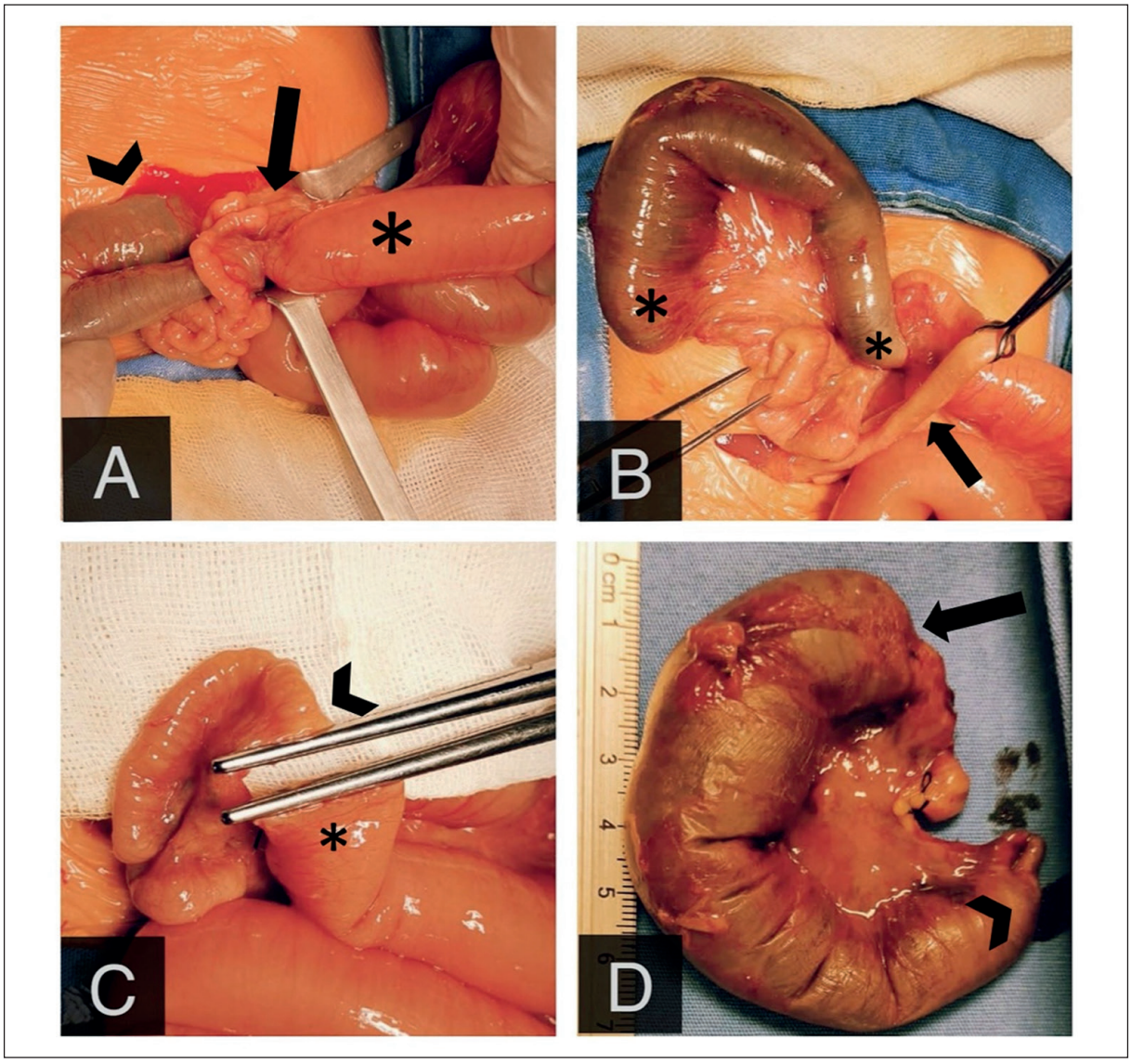

Figure 2. A. Congenital transmesenteric internal hernia (arrow) with trapped intestinal loop (arrowhead). Note the proximal intestinal dilation (*). **B**. Release of the intestinal loop revealing two atretic ends (*) and a patent distal intestinal loop causing the internal hernia (arrow). **C**. Difference in intestinal diameters with a 3:1 ratio, proximal (asterisk) and distal (arrowhead). **D**. Surgical specimen showing the intestinal loop with blind ends, proximal (arrow) and distal (arrowhead).

distal meconium that was evacuated at 50 hours of life, which could have influenced the diagnostic-therapeutic delay.

As in most of the reported cases, the diagnosis of intestinal atresia secondary to transmesenteric hernia was intraoperative[10,11]. Preoperative diagnosis is difficult due to the absence of specific radiological or laboratory signs. In the plain abdominal X-rays of our case, a significant proximal bowel dilatation was observed that displaced the loops to the pelvis, generating a false impression of distal air; however, upon complementing the study with contrast in the colon, the classic image of microcolon suggestive of intestinal atresia was identified, in addition to ruling out intestinal malrotation and Hirschsprung's disease[2,12].

In our case, the diagnostic delay was because the symptoms were initially subtle and inconclusive, such as mild abdominal distension, and gastric and biliary vomiting but presenting meconium evacuations, partial improvement of the distension with fasting and the suspicion of enterocolitis, which diverted attention from the obstructive process for a week. Surgical treatment of these patients includes mesenteric dissection and intestinal resection of the herniated blind loop with subsequent primary anastomosis, as in our case, or intestinal bypass in the presence of wide dispropor-

tion of the proximal and distal segment (greater than 3:1), with the closure of the mesenteric defect to avoid new internal hernias[13].

Pictures suggestive of intestinal obstruction in the newborn should be evaluated by a medical-surgical team with experience in neonatal pathology, in order to make an adequate differential diagnosis excluding the most frequent causes of intestinal occlusion, without leaving aside those unusual but potentially serious pictures, such as transmesenteric hernia associated or not to intestinal atresia.

Conclusion

The diagnosis of intestinal atresia secondary to congenital transmesenteric hernia can be challenging due to its infrequency and un-specificity and variability of clinical presentation as well as meconium expulsion in some cases. In neonates with abdominal distention and vomiting, the diagnostic approach should include imaging studies that rule out congenital lower intestinal obstruction, such as opaque enema, without delaying surgical treatment.

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

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