CASE REPORT

Internal hernia as cause of acute abdomen in a preterm neonate: when necrotizing enterocolitis is not the culprit

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ABSTRACT

Internal hernias in preterm neonates, although rare, can arise due to various anatomical and physiological factors associated with prematurity. We report a case of a preterm infant with symptoms of suspected necrotizing enterocolitis (NEC) that turned out to be an internal hernia during surgical exploration. Given the overlapping symptoms, it is crucial to maintain a high index of suspicion and utilize the appropriate imaging techniques, such as ultrasound or radiographic studies, to aid in the differentiation between NEC and internal hernia, especially when responding to cases that do not improve with standard NEC management or exhibit atypical features. Early recognition and accurate differentiation are crucial for appropriate management and prevention of complications in affected neonates.

KEYWORDS: internal hernia; necrotizing enterocolitis; preterm infant; acute abdomen; bowel ischemia

INTRODUCTION

Internal hernia (IH) is a rare cause of intestinal obstruction which can occur at any age. The mechanism of enteric obstruction is the herniation of viscera, usually a loop of the small intestine, through a peritoneal aperture. Transmesenteric hernia is the most common form of IH, and intestinal herniation is due to a congenital defect in the mesentery. Given the rarity of this entity and the absence of specific radiological findings, the diagnosis is usually delayed, and, in many cases, it is only recognized during surgery.

The clinical presentation of internal hernias in preterm neonates may be subtle and nonspecific and can include abdominal distension, feeding intolerance, and systemic signs of sepsis, which are also commonly observed in necrotizing enterocolitis cases. This similarity in symptoms often leads to misdiagnosis or delayed recognition of the condition. Surgical intervention is often necessary for definitive diagnosis and repair and prompt involvement of pediatric surgeons is essential as the condition is time-critical and is associated with high morbidity and mortality.

CASE PRESENTATION

A 950-gr female neonate was born at 27 weeks and 2 days of gestation from a 30-year-old gravida 2, para 0 woman, by vaginal delivery due to placental abruption. The mother had a free medical history and reported that all the prenatal screening tests were normal. Apgar score was 8 and 9 at 1 and 5 minutes and due to initial respiratory distress, the patient received nasal synchronized intermittent mandatory ventilation for the first six days, followed by high flow nasal cannula. Meconium successfully passed 12 hours after birth, although, enteral feeding was delayed and initiated on the 3rd day of life with no complications. During the following days, the newborn was given surfactant, was full fed with maternal milk, had normal bowel movements and had a stable clinical course until the 17th day of life, when episodes of apnea and fever presented, and C-reactive protein increased. Antibiotic treatment started immediately although there was no clear site of infection and as soon as the urine and blood cultures were negative and inflammatory tests were normal again, it was discontinued.

On the 30th day of life, the infant was clinically disorientated with abdominal distension and tenderness and a mild erythema was noticed in the right lower abdominal quadrant. Based on the clinical suspicion of NEC, enteral feeding stopped, the patient had gastric decompression and broad-spectrum antibiotics initiated. Abdominal x-ray did not show pneumoperitoneum, air-fluid levels or intestinal pneumatosis (Figure 1).

Nevertheless, a common finding of the x-ray and the abdominal ultrasound was a fixed and distended U-shaped intestinal loop in the right abdomen (Figure 2).

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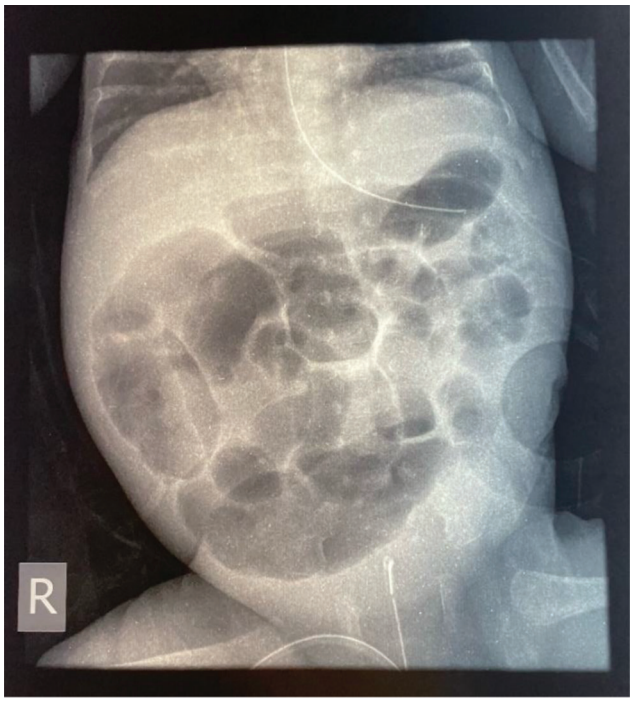

Fig. 1. Abdominal Xray showing a distended U-shaped loop in the right lower abdomen.

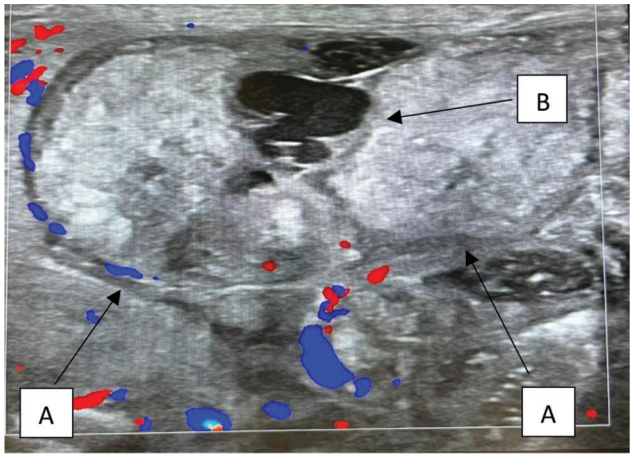

Fig. 2. Abdominal ultrasound showing two distended intestinal loops (A) with increased blood flow and fluid (B) between them.

Over the following two days the abdomen was further distended and firm and the orogastric output increased, which prompted an exploratory laparotomy on the 35th day of life. The peritoneal cavity was full of fecal content. A transmesenteric hernia and a gangrenous small bowel with three sites of rupture were identified (Figure 3).

The hernia reduced and the necrotic loop, approximately 35 cm in length, was completely resected. An ileostomy was created, 2 cm proximal to the ileocecal valve and the mesenteric defect was repaired. The remaining small bowel was 55 cm, measured from the ligament of Treitz and the ileocecal valve was intact. The postoperative course was uneventful, and she slowly returned to full enteral feeds. Ostomy closure carried out 18 weeks later and a side- to -side

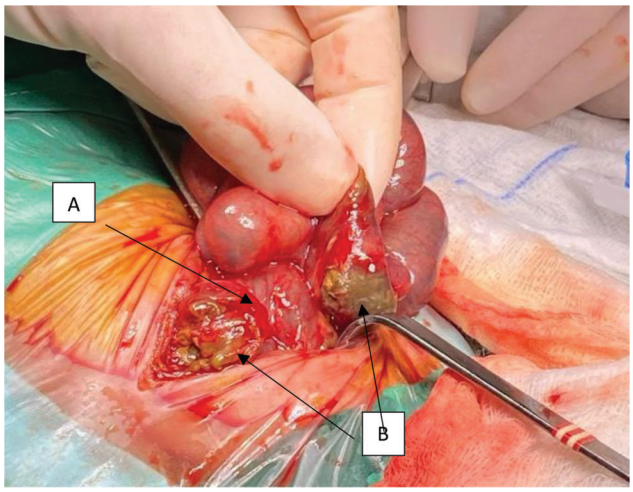

Fig. 3. Transmesenteric hernia (A) and the two out of three sites of rupture (B).

stapled anastomosis was performed. Due to the proximity of the stoma to the valve, it had to be removed. Since then, the patient has normal growth and development, has achieved full enteral feeding and does not present gastrointestinal symptoms.

DISCUSSION

In preterm infants, there are few abdominal emergencies that need immediate surgical exploration. These emergencies are intestinal obstruction or intestinal perforation, and the causes are, predominantly NEC and spontaneous intestinal perforation (SIP). The differential diagnosis can also include intestinal atresia, meconium ileus, malrotation, Hirschsprung's disease, intussusception, or incarcerated hernia but they are not so common in this age group [1,2]. Internal hernia is a rare condition causing bowel obstruction and can occur at any age. Nevertheless, there are, to our knowledge, three cases in the literature describing internal herniation as the cause of acute intestinal obstruction in extreme preterm patients [3,4] (Table 1).

Internal hernias can be congenital or acquired and are classified based on their location as transmesenteric, paraduodenal, intersigmoid, foramen of Winslow, transomental, pericecal, supra or perivesical, and omental [5]. Transmesenteric hernias were first reported by Rokitansky in 1836 and are the most common type in children [6]. They are related to a mesenteric defect of the small bowel and are located either near the ligament of Treitz or proximal to the ileocecal valve, in an area called Treves' field, as in our case [5]. The exact pathogenesis of mesenteric defects is still not clear but there are several theories. A widely accepted hypothesis is an incident of mesenteric ischemia in a hypovascular area during fetal life, leading to a hiatus that can cause herniation.

Clinical presentation widely varies as the intestinal loops passing through the defect can cause episodes of obstruction or subobstruction. The majority of patients present with acute abdominal pain and distention, accompanied by bilious or non-bilious vomiting. Symptoms of shock can appear as the obstruction leads to bowel ischemia and gangrene [2]. Nevertheless, there are cases with vague and non-specific symptoms such as intermittent abdominal pain

Table 1. Existent data of intestinal herniation in extremely premature infants.

Authors	Hirata et al.	Hirata et al.	Kylat RI
Patient's Sex	Male	Male	Female
Gestational Age at Birth	22^{+5} weeks	28^{+4} weeks	24 weeks
Weight	548 gr	1084 gr	625 gr
Age at intervention	9 days	31 days	19 days
Type of intervention & Intraoperative findings	Urgent drainage	• Emergency laparotomy • Trans-mesenteric IH with a mesenteric defect close to the ileocecal valve • Ileostomy	• Emergency laparotomy. • Trans-mesenteric IH with a segment of gangrenous bowel. • Enterectomy and proximal ostomy
Outcome	• Death by septic shock on day 22 of age • Autopsy revealed herniation of most of the small intestine through a defect of the mesentery with ileal perforation	Death by liver dysfunction and recurrent infection on day 127 of age.	Optimal growth parameters at 2 years age follow-up.

or vomiting that can be investigated for a long time before the diagnosis is confirmed [7]. During the first days of life there are some specific clinical modalities that appear with bowel obstruction such as atresias, meconium plug or sepsis-related ileus. Although, in preterm neonates who initially tolerated enteral feeding and suddenly deteriorated, NEC and SIP are the foremost diagnoses [4]. Malrotation can also be rarely considered such as the diagnosis of internal hernia.

Abdominal X-rays and ultrasonography (US), as the first imaging choices, can rarely help with the diagnosis of IH and an upper gastrointestinal study (UGS) should be performed in all patients if their condition is not critical. As Tang et al. noticed the radiological confirmation of the diagnosis in neonates was not feasible prospectively. In the other hand, almost all the older children were diagnosed preoperatively by either UGS or Computed Tomography [7]. In cases of NEC, there are several sonographic findings that can both establish the diagnosis and help with the sequential monitoring of these patients. Yet, the use of US in the evaluation of NEC patients is not broadly accepted by the pediatric surgery community as plain radiographs and laboratory markers are considered as more helpful tools [8].

Immediate intervention is critical for the prognosis of the infant as the rapid progression can lead to lethal complications. If the intestinal tissue seems viable, the hernia can be repaired after the entrapped bowel is released. In cases of severe ischemia or necrosis, a bowel part should be resected and the surgeon should decide if anastomosis can be conducted immediately or a temporary stoma should be created in order for the bowel to rest and heal. In all cases, the surgeon should keep in mind that the length of the intestinal resection in such young patients should be seriously considered, as anatomic loss of bowel absorptive capacity can lead to intestinal failure.

In most cases referred in the literature, neonates with IH had additional surgical findings including malrotation, small bowel atresia or volvulus. In our case, surgical exploration did not reveal any other pathology, which can also explain the late onset of symptoms and the clinical stability of the infant.

CONCLUSION

Internal hernias in children end especially in neonates represent a rare but significant surgical challenge as the mortality rates are high, if left untreated. Condition may remain elusive preoperatively despite the advances in medical imaging. Early recognition and surgical intervention, through laparotomy, are essential to prevent bowel ischemia and obstruction. Optimal outcomes accrue from the collaboration of different specialties as neonatologists, pediatric surgeons and radiologists.

Informed Consent

Written informed consent was obtained from the patient's family for publication of this case report. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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There are no competing financial interests.

Conflict of interest

The Authors declare that there is no conflict of interest.

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