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Case Report

Inverted Proximal Ileal Loop Prolapse with Ileal Rupture through a Patent Omphalomesenteric Duct: A Rare Case

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Abstract

BACKGROUND: Prolapse of the small intestine through the umbilicus is indeed a rare presentation and is the most significant complication of the patent omphalomesenteric duct which requires pediatric surgical emergency due to its significant increase of mortality. To date, it is less than twenty cases of this presentation have been reported in medical literature. We are reporting a case of the same in an infant presenting with it on 1st week after he was delivered, but was followed by ileal rupture as well.

CASE PRESENTATION: We present a case of a patent omphalomesenteric duct with ileal prolapse and ileal rupture as its complication. It is a case of a 1-year-old infant with a history of unusual bleed-on-touch mass emerging from the anterior abdominal wall with absent umbilicus. Once his condition is stabilised, he underwent a reduction of the prolapsed bowel along with complete excision of the omphalomesenteric duct and restoration of the ileal continuity. Post-operatively he regained normal bowel function and resumed breastfeeding 5 days after surgery.

CONCLUSION: This case is an important addition to the literature about patent omphalomesenteric duct with complications of inverted proximal ileal loop prolapse and ileal rupture.

Introduction

The Omphalomesenteric duct (OMD) (vitellointestinal duct) connects the midgut to the yolk sac and provides nutrition until the placenta is established. It normally attenuates, involutes, and separates gradually from terminal part of ileum during 5th to 9th weeks of gestation [1], [2], [3], [4], [5], [6], [7]. Its persistence after intrauterine life can manifest as different pathologies called *omphalomesenteric duct remnants* (OMDR). *Patent omphalomesenteric duct* (POMD) is total incomplete obliteration of the omphalomesenteric (vitelline) duct [8].

Exact aetiology of POMD remains an enigma. None accurately addresses a direct cause of this anomalies although various teratogenic models are present in the literature, such as carbimazole or amine exposure within the first trimester of pregnancy, yet recent studies show the association between them has largely been anecdotal and still need more robust scientific evidence [9].

Various forms of OMDR is presented in Fig 1 with letter F illustrating our case [2], [7]. The OMDR occurs in approximately 2% of newborns and in 6% of these the duct remains patent, with only 20% of patent omphalomesenteric duct cases being complicated by intussusception of small bowel through the patent duct. Males are thrice more prone to be in this condition, and 73% of this case exhibit symptoms within the first 28 days of life [2], [6], [10], [11], [12], [13], [14].

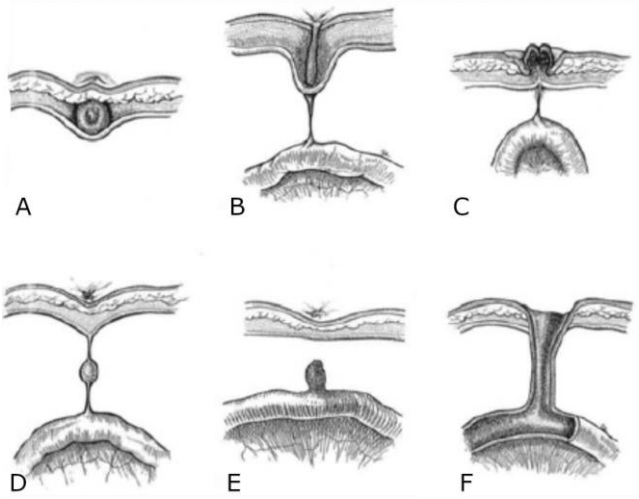

Figure 1: Various Omphalomesenteric duct remnants; A) Containing-intestinal-tissue of an umbilical cyst; B) Umbilical sinus with a band; C) Umbilical polyp covered with intestinal mucosa; D) The fibrous band containing a cyst; E) Meckel's diverticulum; F) Patent omphalomesenteric duct

Patient with patent omphalomesenteric duct can present with the anomaly itself or due to its complications secondary to the anomaly, including progressive prolapse of the omphalomesenteric duct, leading to a rupture of ileum as the compensation of high intraluminal pressure [6], [10]. In our case, the complications are ileal prolapse and intestinal perforation (rupture). This anomaly, which is known as a rare case, needs to be managed urgently for fear of gangrene of the prolapsed bowel and high risk of sepsis [3], [10], [13]. The appropriate treatment and timing of the surgery remain controversial. The principle surgical management of POMD is a reduction of the prolapsed bowel, complete excision of the omphalomesenteric duct, restoration of the ileal continuity, and umbilical reconstruction [11], [13].

At this moment we present a case report of a 1-year-old boy with patent omphalomesenteric duct (POMD) with proximal ileal prolapse and ileal perforation.

Case Report

A 1-year-old male infant was admitted in an emergency room with the chief complaint of bright red, polypoid-shaped mass emerging from the anterior abdominal wall with absent umbilicus. He was born at full term by normal spontaneous vaginal delivery to a porous 3 mother without antenatal care, helped by a midwife. The mother denied that the infant was born with the unusual abdominal mass, yet she stated convincingly the mass appeared since 1 week after delivery, with size initially 3 mm and three-looped shape.

She also noted peri-umbilical erythema, blood-mucus-containing umbilical drainage since 1 week after the infant was delivered. She also admitted the infant had recurrent fever and vomitus, yet he still passed gas and normal stool per rectally for the last one year. One day before admission, the infant was suffering from cough leading to protrusion of the red-coloured mass from the umbilicus. The mass which was initially small grew over a size of 3 cm within five hours. Five hours before admission, the infant presented feculent umbilical drainage with an absence of passing gas and faeces.

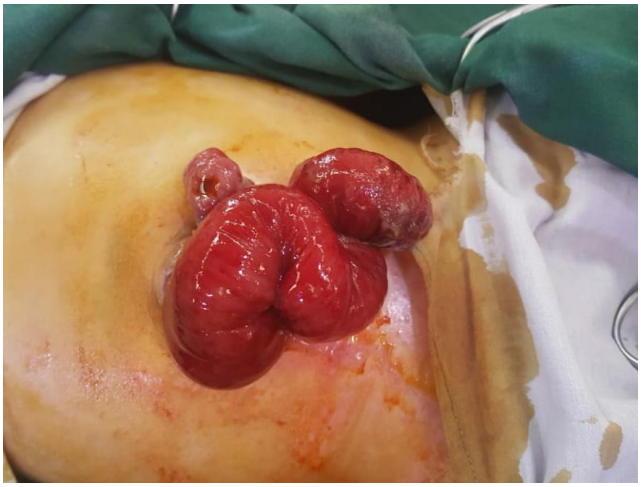

Figure 2: A Polypoid-shaped loop of intestine protruding from the umbilicus revealed prolapsed small bowel mucosa

On careful examination in the ER, the infant was pale but still comfortable and alert. Abdominal examination revealed a bright red polypoid-shaped loop of intestine protruding on the anterior abdomen (Figure 2). One of the tips of the mass was discharging feculent fluids. The mass was irreducible and bled on touch suggestive of mucosal surface. Bowel sound was normal. Anal opening was normally placed and patent.

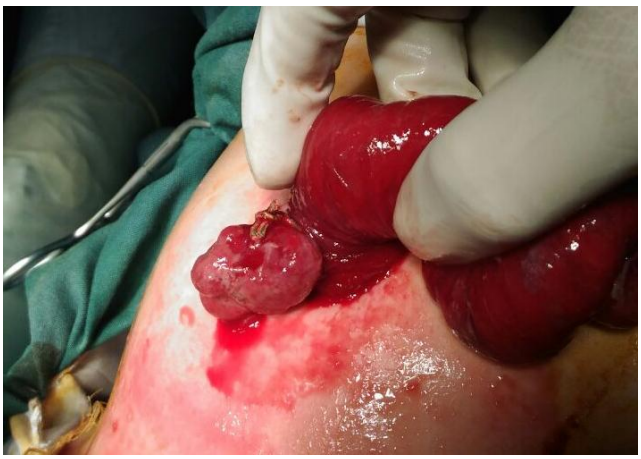

Figure 3: Prolapsed of ileum seen protruding from the umbilicus

The abdomen was neither distended nor

tender. He had passed clear urine twice, and the bladder was not palpable. Rest of the systemic examination was normal. A provisional diagnosis of the patent omphalomesenteric duct was made on clinical signs for the appearance of 5 cm duct connecting from bowel to the umbilicus and obvious feculent discharge.

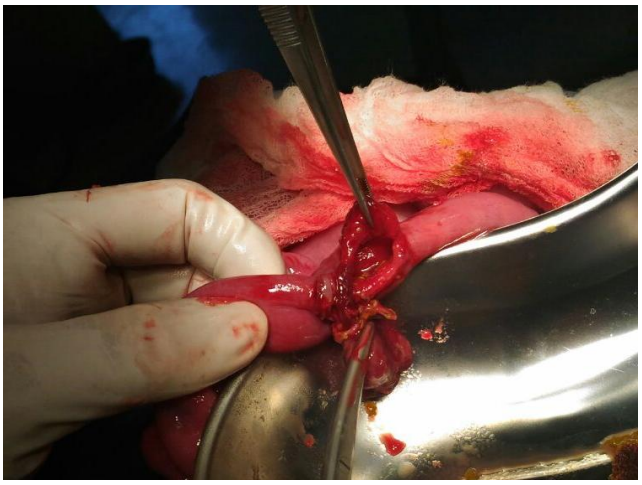

Figure 4: Intraoperative picture of the patent omphalomesenteric duct to the umbilicus with fistula seen in the picture

Blood investigations showed severe anaemia (Hb: 7.7 mg/dL) and leukocytosis (11.800/µL). To maintain the stability of the infant's condition and to prevent further complications, we performed emergency laparotomy surgery without further X-ray investigation nor ultrasound examination. The baby was initially managed and stabilised with the administration of intravenous fluid for rehydration, transfusion of 100 cc packed red cell, and intravenous broad-spectrum antibiotics cefotaxime (50 mg/kg/day).

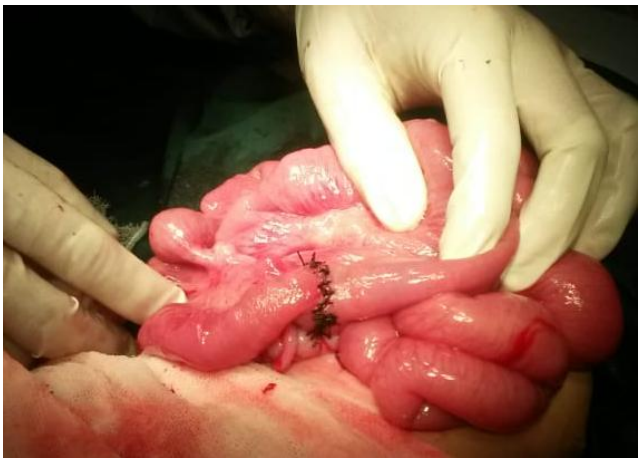

Figure 5: Intraoperative picture after resection and ileoileal end-to-end anastomosis at the site of prolapsed patent omphalomesenteric duct

A midline incision up to the abdominal cavity was made, and a 5 cm length of the proximal ileal loop that had prolapsed through a patent omphalomesenteric duct was separated from the abdominal wall by fine dissection and was meticulously reduced using milking method (Figure 3).

After a complete reduction, a defect of 5 x 5 cm rupture was found in the small intestine at the point of adherence with the abdominal wall suggesting its patency with the external environment through the umbilicus (Figure 4). The duct was released from the umbilicus. A patent omphalomesenteric duct with prolapse of ileum and ileal rupture was diagnosed.

Since there was no bowel oedema and the mucosa was healthy, ileostomy was not performed. Thus resection of the patent duct along with ileoileal end-to-end anastomoses was done (Figure 5). The infant started a small number of feeds on the 5th postoperative day. He was followed up for 3 weeks. Postoperative period was uneventful.

Discussion

The incidence of the patent omphalomesenteric duct is reported to be 1 in 5000 to 8000 newborns (approximately 2% of the population) and may begin at birth or occur within 1 to 2 weeks after delivery [6], [12]. This is suitable in our case, in which the mother admitted the appearance of unusual-red-coloured mass from the umbilicus on the first week after vaginal delivery [13]. POMD may remain silent throughout life or may present incidentally sometimes with an intrabdominal complication [5], [6].

The duct remains patent in 60% of OMDR cases and can present discharging umbilical sinus, umbilical nodule or polyp, and bleeding from the intestinal mucosa, with umbilical faecal drainage as the most symptomatic presentation of omphalomesenteric duct anomalies in developing countries [7], [14]. Another significant complication is either intussusception of the small bowel through the patent duct, which happened in 20% of the incidence of POMD or progressive prolapse of the omphalomesenteric duct, leading to polypoid-shaped bowel protrusion on the anterior abdomen which is seen in our case [5], [11].

Intussusception, volvulus, internal hernia (closed-loop obstruction) from the POMD, and a fibrous connection between umbilicus and ileum are the mechanisms of POMD causing small bowel obstruction [3], [4], [7], [10]. Meanwhile, the mechanisms of ileal prolapse through umbilicus is hypothesized by these two reasons, such as the wide mouth of the patent duct and the short distance between the patent duct and ileocecal valve in infants leading to high intraluminal pressure [2], [4], [5], [10], [12]. Besides, these conditions are exacerbated by the increased intraabdominal pressure, such as cry or cough [12]. In our case, rupture of ileum happened as

the compensation of increased intraluminal pressure caused by the end of the POMD pathogenesis, such as the small bowel obstruction. It explains the reason for infant's absence passing gas and faeces, which happened for 5 days before admission [1], [13].

Understanding the aetiology of small bowel obstruction caused by POMD without diagnostic laparotomy or laparoscopy is difficult. Abdominal plain radiographs and ultrasonography are non-specific for it. Although computerized abdominal tomography may be useful to show the band originating from the umbilicus and continuing between the small bowel loops, we did not perform them due to the lack of facilities and resources. Investigations like fistulogram were not performed as well since not only there is no need to differentiate POMD from patent urachus, but it also would not change the surgical decision in our case. In conclusion, we provisionally made the diagnosis based on history type of discharge (faecal) from the umbilicus along with clinical signs and confirmed it during laparotomy [6], [11].

Management options may include reduction of prolapsed bowel, definitive surgery such as laparoscopy or open laparotomy, wedge resection in a viable bowel, and intestinal resection in a non-viable bowel with complications of strangulation, gangrene, and perforation [4], [5], [6], [7], [10], [12], [15]. In our case, indications for emergency laparotomy are perforation and obstruction caused by ileal entrapment of the duct [15], [16]. In consideration that bacterial translocation may occur or as prophylaxis for resection, we administered broad-spectrum antibiotics although there are no controlled data about the antibiotherapy [16].

We believe this is an emergency case which must be dealt urgently due to the associated intestinal rupture caused by the prolapsed intestinal loop as any delay can lead to catastrophic consequences. In our case, despite the late referral of the patient to the hospital, we were able to stabilise the patient, performed prolapsed bowel reduction, resection of patent omphalomesenteric duct, and ileoileal end-to-end anastomoses. The infant was followed up for 3 weeks. Postoperative period was uneventful.

In conclusion, due to its rareness, this case is an important addition to the literature about patent omphalomesenteric duct with complications of inverted proximal ileal loop prolapse and ileal rupture.

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