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Management of Internal Hernia in Neonates with Multiple Heart Diseases: A Case Report

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Abstract

Internal hernia is a rare clinical entity. In all cases, less than 1% found as the cause of intestinal obstruction. However, delayed treatment of internal hernia may lead to necrosis of the intestines. Neonates with congenital heart disease may be at increased risk of morbidity and mortality than another concurrent disease.
A male infant was delivered with a Cesarean section due to a congenital disorder of umbilical hernia, ventricular septal defect, and aortic transposition. The infant was born full-term with a 2,515 g birth weight and 3, 5, 7 APGAR score, positive ventilation, and intubation proceeded. Prostaglandin E1 10 mcg/kg/minute, packed blood cell, and thrombocyte concentrate was administered preoperatively. The patient underwent laparotomy for intestinal resection on day-3; the necrotic intestine was found starting 70 cm from ligament of Treitz to midsection of the transverse colon. Postoperatively, the stoma was vital, and we noted its production. The infant died one day14 due to respiratory failure caused by hospital-acquired pneumonia. Infants with an internal hernia and multiple congenital heart diseases require prompt management to prevent intestinal necrosis and other respiratory-related complications.

Key words: congenital heart diseases, internal hernia, laparotomy, intestinal resection, necrosis

Introduction

According to autopsy studies, internal hernia occurs in less than one percent of the population studied; however, it accounted for approximately 0.5 to 5.8% of all cases of intestinal obstruction.[1] Internal hernia occurred in less than 0.01% of infants with jejunoileal atresia or stenosis, with an incidence rate of 1.3 – 2.25 cases per 10,000 live births in Spain, Latina America, and France. In other states in North America, incidence rates may increase up to 2.9 cases per 10,000 live births. From previous studies, infants with congenital heart disease are at higher risk of morbidity and mortality compared to the healthy infants.[2,3,4]

Other case reports had described intestinal necrosis on internal hernia cases incidentally found during abdominal surgery; thus, requiring additional resection during the same period. Felizes et al. had reported a neonate with internal hernia diagnosed intraoperatively; resection of the ileum (21 cm long) was performed due to ileal necrosis.[3] Saka et al. had reported five patients requiring ileal resection due to intestinal necrosis associated with an internal hernia (all of the cases were diagnosed intraoperatively) with varying lengths of small intestines preserved, ranging from 150 to 200 cm.[5] Additionally, Galazka et al. had performed ileal and liver resection on neonates with internal hernia due to liver penetration of the hernia.[6] Diagnosis of multiple congenital anomalies, particularly of the intestine and heart, are extremely rare; we present a case report of a male infant diagnosed with congenital umbilical cord hernia with multiple congenital heart defects.

Case illustration

A male infant was delivered on cesarean section due to congenital umbilical cord hernia, ventricular septal defect, and aortic transposition in dr. Cipto Mangunkusumo General Hospital. The infant was born full-term at 38th gestational week with 2,515 g birth weight. The infant was found to be cyanotic with poor respiratory effort; the APGAR scores were 3, 5, and 7. He received positive pressure ventilation for five minutes but did not show improvement. Intubation was performed with FiO2 45% and oxygen saturation down to 60%. The ileal loop had invaginated the umbilical defect with the enlarged left and right scrotum. The skin color of the scrotum was within normal ranges.

In the first three hours of life, the infant was diagnosed with the umbilical cord and bilateral scrotal hernia and referred to pediatric surgery. On the 1st day of surgical care, the meconium passed out, and inserted orogastric tube produce reddish fluid. The umbilical hernia had reddish fluid content. The tissue viability of invaginated ileal loop was hard to assess.

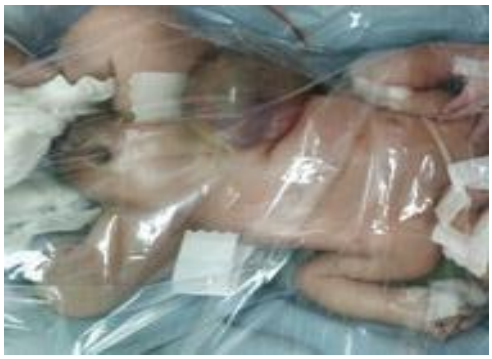

Figure 1. The first presentation. Ileal loop invaginated the umbilical defect, enlarged right and left scrotum, no abnormal scrotum skin color.

To this finding, he has planned a surgical procedure. The ventilator was set to SIMV mode, with PEEP 5, FiO_2 21%, and 69–70% oxygen

saturation. Laboratory findings were significant for an abnormally decreased hemoglobin content of 6.9 mg/dL and a platelet count of 84,000 /mcL. An Echocardiography showed severe aortic coarctation, atrial and ventricular septal defects, and persistent ductus arteriosus. To these findings, from the cardiologist's perspective, there was no contraindication for the operation. Prostaglandin E1 10 mcg/kg BW/min was suggested if the oxygen saturation dropped below 70%. The surgery was delayed due to packed blood cell and thrombocyte concentrate transfusion. During the preoperative examination, the skin around the umbilical defect and bilateral scrotum became inflamed (Figure 2), orogastric tube fluid was red–brownish, and the patient had bloody stool.

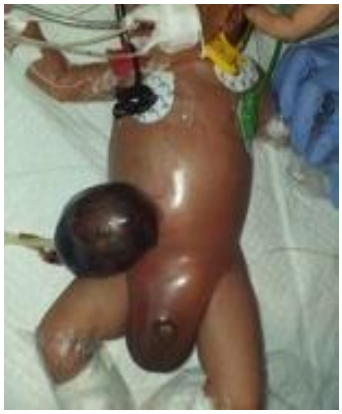

Figure 2. Clinical presentation on day 3. The reddish colored on the defect of the umbilicus and bilateral scrotum.

The condition worsened on day two. Oxygen saturation dropped to 50 – 70%, so prostaglandin E1 25 mcg/minute was administered, and oxygen saturation increased to 70–80%. After transfusion of 87 mL of packed red cell and 75 mL of platelet concentrate, the surgical procedure proceeded on day 3. Intraoperatively, we found an ileal loop to invaginate through the defect on the mesentery in the ileocecal region. The loop was pinched and had entered the umbilicus and right scrotum; the cecum was not fixated. Intestinal necrosis was found, 70 cm from the ligament of Treitz to the midpart of the transverse colon. After the surgery, the stoma had remained viable. Production was noted, and the patient was able to feed. The infant, however, died on the day-14 postoperative due to respiratory failure from hospital-acquired pneumonia.

Discussion

Hirata et al. described a similar case with our reports; a premature infant with a low birth weight of 1,084 g and multiple heart disease (ventricular septal defect and aorta coarctation). Most of the loops attached to the mesentery defect were still vital in the assessment, but the necrotic ascending colon was noted. In this case, resection of ascending colon followed by stoma. Necrosis on ascending colon was caused by ischemia of the tissue and compromised vascularization due to increased pressure from ileal loops pressing on the arteries to the colon. The patient died at the age of 127 days due to recurrent infection and feeding intolerance. Like our case, the poor prognosis was also noted in infants with intestinal hernia with intraoperative findings of congenital heart disease. Multiple congenital heart disease is a risk factor for increased morbidity and mortality in other pediatric surgery cases; however, there is still limited published study reporting the treatment and outcome of hernia on infants with congenital heart diseases.

Several hypotheses about the pathogenesis of mesentery defects have been suggested, such as regression of dorsal mesentery, increased area of hypervascularity on the mesentery, rapid elongation of the mesentery, and increased pressure from colon towards the mesentery during the herniation of fetal midgut to the yolk sac. The subject of this study underwent laparotomy surgery on the third day after the diagnosis of intestinal obstruction.

The surgical procedure proceeded following the transfusion of packed red cells and platelet concentrate. The case series by Hirata et al. had also reported delayed surgical procedures.

Similar age of diagnosis and management had been given in comparison to the study. Prostaglandin E1 and pulmonary artery banding had been performed to manage the existing congenital cardiac anomaly before the intestinal management on the 10th day of life.

In comparison, Hirata et al. had performed the surgery as early as possible. We have delayed the surgery due to the patient's age; as a comparison, the infant managed by the other case report was older. Thus, the infant was eligible for emergency operation.

Laparotomic intestinal resection was performed in this study. It was performed 70 cm from the ligament of Treitz to the midsection of the transverse colon and stoma. The surgical procedure was like the cases treated by Hirata et al., laparotomic resection of ascending colon and stoma.

Necrosis of the intestine, in this case, was caused probably due to increased pressure on the vascular system of the mesentery (from internal hernia entering the mesentery defect) that supplies the ascending colon. The intestinal loop entering through the mesentery defect was pinched out. Despite the increased pressure, the tissue in Hirata's case had remained viable. In contrast, the ileal loop was necrotized in our study due to prolonged strangulation from delayed surgery. Prompt surgical management for intestinal obstruction in infants with multiple heart disease was essential. Delayed surgery may eventually lead to the spreading of necrosis to other intestinal tissue.

Multiple factors should be of one's consideration before deciding on intestinal anastomosis, including local and systemic factors. Local factors consist of vascularization, surgical technique, and risk of bacterial contamination. Systemic factors include nutritional status, tissue perfusion, and oxygenation.[8]

We create the stoma as the systemic condition was unsuitable due to inadequate perfusion and oxygenation of the intestinal tissue. The ventilator fitted on PC-AC mode during the surgery, PEEP 5, FiO_2 30%, and 70–85% oxygen saturation. In the Hirata reports, the stoma was created due to the exact reason in this case. During the surgery, the infant showed increased C-reactive protein levels (1.04 x 105 mcg/L0 and metabolic acidosis (pH 6.982 with base excess of 19.9).

Postoperatively, the stoma vital and produces, and the patients had been given an oral diet. However, the infant died after the 14th postoperative day due to respiratory failure from hospital-acquired pneumonia. In contrast, the case reported by Hirata et al. had died on the 96th postoperative day due to extensive fasting and recurrent infection. Neonates with congenital heart disease may experience recurrent respiratory tract infection due to increased blood flow to the lungs, thus adversely affecting the lungs' immune system.9

This study is the second study that had reported a case of intraoperative diagnosis of internal hernia on a neonate with multiple heart disease and the first study that had reported on a neonate with normal birth weight (2,515 g).

Internal hernia is an uncommon disease. The author only found one case report as a reference (level of evidence IV according to criteria from Center of Evidence-Based Medicine University of Oxford 2010).

Conclusions

Internal hernias in neonates with multiple heart diseases are extremely rare. Neonates with multiple heart diseases may often have unstable

clinical parameters that lead to inadequate tissue perfusion and oxygenation; thus, intestinal resection and stoma creation must be performed immediately to prevent necrosis and additional stress on the cardiovascular system. Delayed surgery in our case might be associated extending necrosis and subsequent worsening patients' condition.

Disclosure

Authors declare no conflict of interest

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