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Case Report

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Meckel's diverticulum perforation in three preterm neonates: Are early diagnosis and surgical intervention life-saving?

Muhammed Hamidullah Çakmak, Sabri Cansaran, Ayşenur Celayir

Abstract

Meckel's diverticulum (MD) perforation is extremely rare in preterm infants. The diagnosis is usually made during exploratory laparotomy. In this article, we present three premature neonates with early diagnosed MD perforation. Case one was operated in the emergency setting on his second day of life and he is in the second postoperative year without any gastrointestinal problem. Case two had no problems with gastrointestinal system during follow-up; however, he died at the sixth months of his life due to severe cardiac abnormalities with decompensation and respiratory distress related to Trisomy 13. The third case was a premature twin and due to bowel perforation on postnatal Day 3, an abdominal drain was placed and she underwent surgery on postnatal Day 5. In conclusion, MD perforation should be kept in newborns with pneumoperitoneum. The prognosis after surgery in MD perforation is usually favorable for cases diagnosed and treated early.

Keywords: Meckel's diverticulum, newborn, perforation, pneumoperitoneum.

Meckel's diverticulum (MD) is the most common congenital anomaly of the gastrointestinal tract with prevalence of 2% in the population.[1] It is a true diverticulum and located usually on anti-mesenteric side at ileum. It is caused by an incomplete obliteration of the vitelline duct during fetal growth.

Meckel's diverticulum is often clinically silent and discovered incidentally. Complications include gastrointestinal bleeding, bowel obstruction, intussusception and usually occur before the age of two years.[1,2] Intestinal perforation is a rare complication and is seen in 10% of symptomatic MDs.[3-5] Meckel's diverticulum perforation is extremely rare in preterm newborn infants.[6,7] Wedge resection-anastomosis or segmental ileal resection-anastomosis are among the treatment options.[6,8]

In this article, we present three premature neonates with early diagnosed MD perforation to draw attention to early diagnosis and treatment of MD perforation in pediatric cases.

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Correspondence: Muhammed Hamidullah Çakmak, MD.

E-mail: dr.m.hamidullah@hotmail.com

Department of Pediatric Surgery, Health Sciences University, Zeynep Kamil Women and Children's Diseases Training and Research Hospital, İstanbul, Türkiye.

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CASE REPORT

Case 1– A one-day-old male neonate, who was born by urgent cesarean section for placental abruption at 29th gestational week with a birth weight of 1,500 g, was intubated after delivery for respiratory distress and transferred to neonatal intensive care unit (NICU). On the same day, the patient referred to us due to suspected subdiaphragmatic free air on abdominal X-ray (Figure 1a). Intraabdominal free air was confirmed on the control radiograph taken in the NICU (Figure 2a). His abdomen was mild distended on physical examination without discoloration of the abdominal skin.

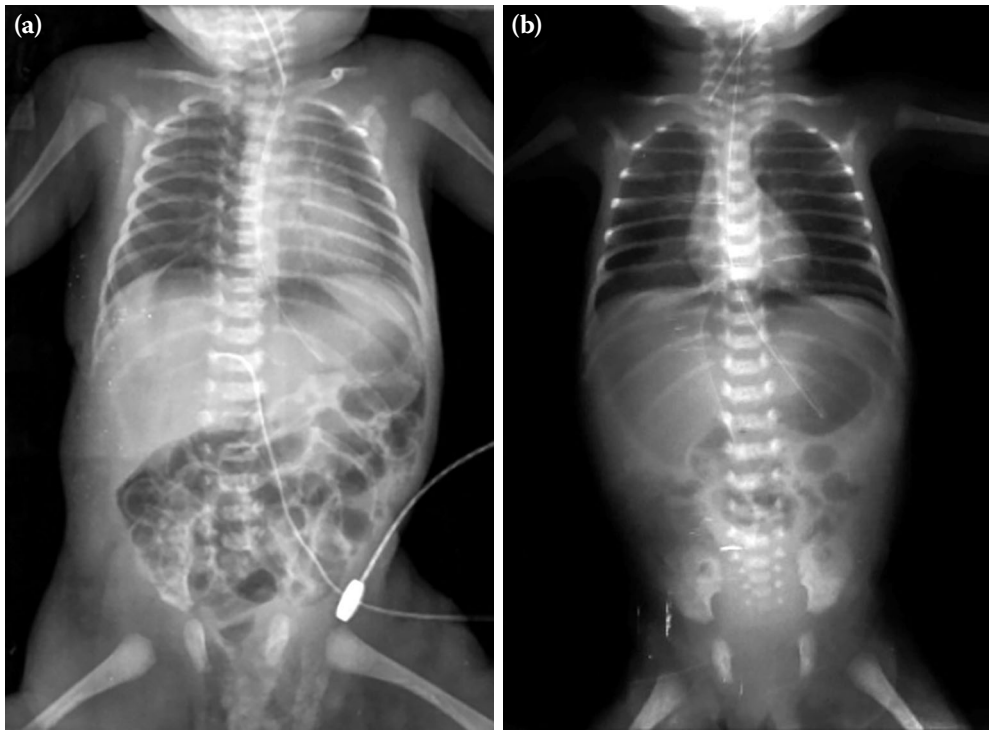

Figure 1. Suspected subdiaphragmatic free air on standing abdominal X-ray. **(a)** First case. **(b)** Third case.

On the second day of his life, emergency laparotomy was performed due to the suspicion of intestinal perforation. Perforated MD with a size of approximately 1 cm was observed on the antimesenteric side of the ileal segment approximately 30 cm proximal to the ileocecal valve (Figure 3a). The appearance of the remaining small intestines, stomach, and colon were normal. Wedge bowel resection and anastomosis with transverse Gambee suturing were performed. Histopathological examination of the resected material was consistent also with MD.

The patient was weaned from ventilatory support and enteral trophic feeding was started via nasogastric tube on postoperative Day 4. The patient was also given treatment for biotinidase

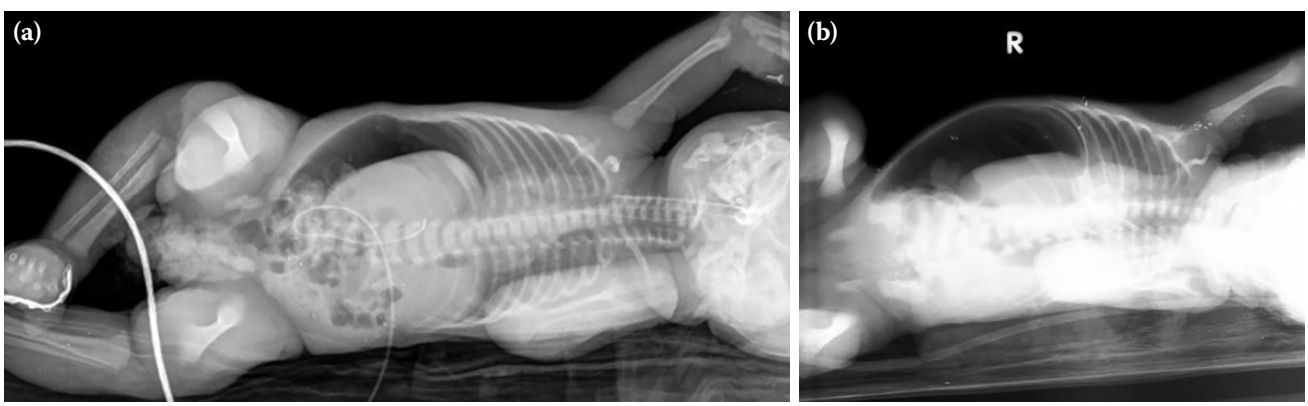

Figure 2. If subdiaphragmatic air is suspected in the standing X-ray, cross table lateral graph taken with the patient lying on the left lateral side, and the air accumulated between the liver and the abdominal wall can be demonstrated in babies with excessively dilated intestines. **(a)** First case. **(b)** Third case.

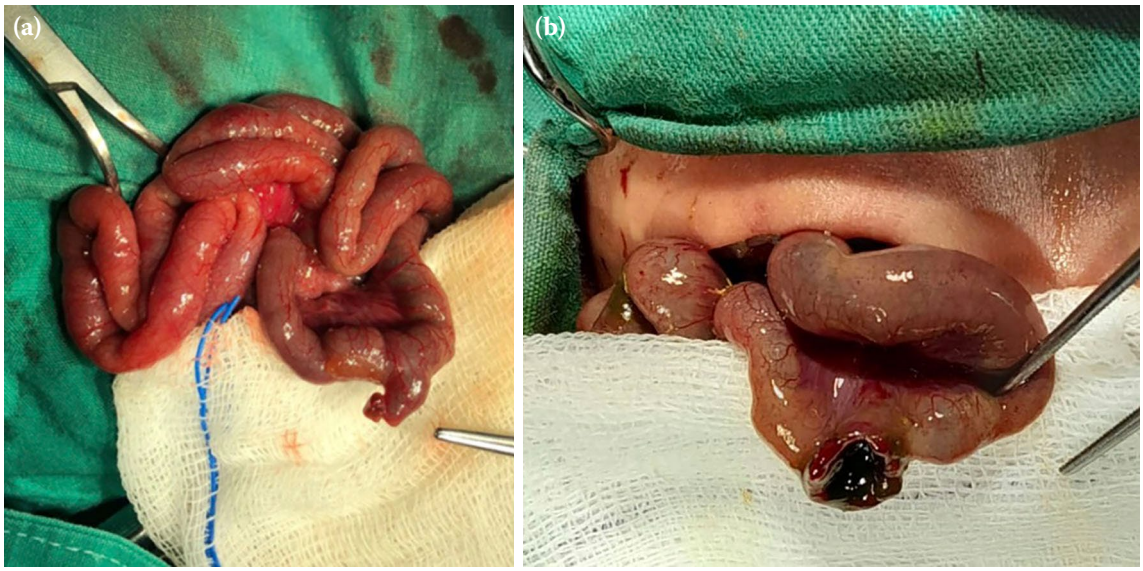

Figure 3. **(a)** Perforated Meckel's diverticulum of first case. **(b)** Perforated Meckel's diverticulum of third case.

deficiency, late neonatal sepsis, intracranial hemorrhage, and seizure. On postoperative Day 23, he was fed full enterally and transferred to the NICU again for follow-up due to prematurity. The patient is still in the second postoperative year and has no gastrointestinal problem. A written informed consent was obtained from the parents of the patient.

Case 2– A two-day-old male neonate, who was born at 34th gestational week with a birth weight of 2,300 g, was transferred to our department with a preliminary diagnosis of bowel perforation due to subdiaphragmatic free air. Postnatal ultrasound examination showed congenital heart anomalies including pulmonary atresia with ventricular septal defect, secondary atrial septal defect and patent ductus arteriosus. He also had thymus hypoplasia, right hydroureteronephrosis, bilateral accessory finger, undescended testicle, and pes equinovarus.

Abundant hemorrhagic fluid drainage was observed at the beginning of laparotomy. During the abdominal exploration, perforation of MD and hemorrhage in the artery supplying the diverticulum due to perforation were detected. Hemostasis was achieved by ligating the artery of diverticulum. The portion of the ileal loop affected by the diverticulum was segmentally resected (1.5 cm) and an ileal end-to-end anastomosis was performed.

The patient was extubated on postoperative Day 1 and enteral trophic feeding was started on postoperative Day 5. He was intubated on postoperative Day 6 due to clinical worsening due to respiratory and cardiac distress, and enteral feeding was stopped on postoperative Day 7 due to his feeding intolerance. Feeding was resumed on postoperative Day 11 again, and full-dose enteral nutrition was achieved on postoperative Day 15. Tracheostomy was also performed in the patient who was referred to another tertiary center for cardiac surgery on postoperative Day 18, as he could not tolerate extubation. The patient who had no problems with gastrointestinal system during the follow-ups died at the sixth months of his life due to severe cardiac abnormalities with decompensation and respiratory distress related to Trisomy 13. Written informed consent was obtained from the parents of the patient.

Case 3– A three-day-old twin female baby was born by cesarean section at 29th gestational weeks, weighing 1,200 g, from a 35-year-old mother with gestational diabetes mellitus. A drain was placed due to intraabdominal perforation on postnatal third day (Figure 2b), and exploratory laparotomy was done on postnatal fifth day. During exploration, meconium was cleaned from the abdomen. A 5 to 10-mm serosa defect, 50 cm distal to the Treitz, was repaired. There was a

1-cm diameter wide-based MD perforation area 45 cm proximal to the terminal ileum, and a resection anastomosis was performed (Figure 3b). The patient started to be fed on postoperative Day 5 and was discharged on 72nd day of his life. Written informed consent was obtained from the parents of the patient.

DISCUSSION

Meckel's diverticulum is a congenital true diverticulum containing all layers of the normal bowel wall and located usually on the antimesenteric side of ileum. It is mainly caused by an incomplete obliteration of the vitelline duct during fetal growth by the fifth week of gestation.[3]

Meckel's diverticulum is a common anomaly with prevalence of 2% in the general population.[1] The male-to-female ratio is about 2:1.[4] In a recent systematic review, it was reported that MD perforations in the neonatal patient group mostly occurred in the first week of life (84.7%), and they showed a significant male predominance with a ratio of approximately 6:1.[8] In this report, our patients were one day, two days, and three days old, respectively; the first two cases were male and the last case was female.

The majority of patients are asymptomatic. Complication rates vary between 4 and 34% and it is common before two years of age.[1,5] Intestinal perforation is a rare complication and is seen in 10% of symptomatic MDs.[3,4] Complications such as gastrointestinal bleeding, bowel obstruction, Littre's hernia, fistula and intussusception are usually seen in patients with ectopic tissue.[1,2] The histopathological findings of our patients were compatible with MD, and none of them had ectopic mucosa.

Symptomatic neonatal MD is extremely rare and has been reported to account for less than one-fifth of all pediatric cases.[9] The most common forms of presentation in neonates include bowel obstruction due to inflammation or ileal volvulus, pneumoperitoneum, and intussusception.[10,11] Our first case was asymptomatic except for mild abdominal distension and was diagnosed after an abdominal X-ray taken after the suspicion present in the routine posterior-anterior chest X-ray. This finding highlights the importance of timely evaluation of patients with appropriate imaging methods, particularly in the presence of clinical doubt.

Pneumoperitoneum is an urgent condition which may occur due to serious reasons in newborns. The differential diagnoses of pneumoperitoneum include commonly necrotizing enterocolitis, perforated appendicitis, meconium ileus in cystic fibrosis, intestinal atresia, extensive aganglionosis, intestinal volvulus or idiopathic intestinal perforation.[6,12] Perforated MD is uncommon in neonates.[6] Risk factors predisposing to MD perforation include antenatal or postnatal steroid therapy, poor intrauterine blood flow, hypoxia, congenital absence of the muscles in the gastrointestinal wall and exchange transfusion for hemolytic disease.[13] None of these factors were detected in our patients.

In the literature, several methods of treatment for MD have been described including wedge resection and transverse suturing to decrease the risk of ileal obstruction, segmental resection of the ileum and end-to-end anastomosis, and rarely ileostomy.[13,14] Dehiscence after a primary anastomosis has been reported about 5%.[14] Wedge resection with transverse suturing was performed to our first patient; and segmental resection with ileal end-to-end anastomosis to the second and third patients. None of our patients experienced surgical complications. Taken together, we can speculate that early surgery performed in both cases may have reduced the increased morbidity risk of the patients and positively affected the surgical recovery process.

In conclusion, in newborns with feeding intolerance, it is possible to diagnose intestinal perforation with appropriate physical examination and abdominal X-rays. Although it is a rare condition, MD perforation should be kept in mind in all newborns who develop intestinal perforation postnatally. Postoperative prognosis may be more favorable with early diagnosis and appropriate emergency surgical intervention.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Idea/concept, design, analysis and/or interpretation, writing the article: M.H.Ç., S.C.; Control/supervision: S.C., A.C.; Data collection and/or processing, literature review: M.H.Ç.; Critical review: A.C.

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