□ Case Report □

Primary peritoneal drainage as a treatment for perforated necrotizing enterocolitis with bacterial peritonitis in an extremely low birth weight infant : a case report

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Necrotizing enterocolitis(NEC) is the most common life-threatening surgical emergency in neonates, and remains a major cause of morbidity and mortality. In addition to conventional laparotomy, intraperitoneal drains have been used for the treatment of perforated NEC, especially in extremely low birth weight(ELBW) infants. We report a case of perforated NEC with bacterial peritonitis in an ELBW infant managed with primary peritoneal drainage(PD) without further need for surgery. To our knowledge, this is the first documented Korean case of an ELBW infant where PD was used as primary treatment for perforated NEC. Primary PD is effective and safe in ELBW infants with perforated NEC; although it is not considered a definitive procedure, it should be considered in all cases where infants are too unstable to tolerate anesthesia and surgery. **(Korean J Pediatr 2006;49:800-804)**

Key Words : Necrotizing enterocolitis, Peritonitis, Infant, Low birth weight

Introduction

Necrotizing enterocolitis (NEC) is the most common life-threatening surgical emergency in neonates, and remains a major cause of morbidity and mortality[1]. As a result of the advances in neonatal intensive care, the increased survival of ELBW infants has led to an increased need for surgical intervention. Although many cases of NEC can be managed with medical treatment, approximately 33% to 50 % of patients with NEC will not respond to medical management and will require surgery[2]. Traditionally, the surgical treatment for perforated NEC has been laparotomy, visceral exploration and resection of the involved intestine. In 1977, peritoneal drainage (PD) was initially described by Ein and colleagues; it was originally introduced as a way to stabilize critically ill low birth weight infants with intestinal perforation until a formal laparotomy could be performed[3]. Over time it has been observed that PD alone can result in rapid and effective improvement thereby reducing the need for laparotomy.

We here report the first Korean case of an ELBW infant where PD was used because the infant was extremely unstable to perform a laparotomy. Current concepts of peritoneal drainage are discussed extensively, and a review of perforated NEC is presented.

Case Report

A male infant of 24 weeks' gestation and weighing 855 g was born to a 37-year-old gravida 4, para 2 mother by spontaneous vaginal delivery. Apgar scores were 2 and 5 at 1 and 5 mimutes, respectively. Due to severe respiratory distress, the infant received urgent intubation and cardiopulmonary resuscitation; he was transferred to the neonatal intensive care unit (NICU) where mechanical ventilation was provided. Surfactant replacement was done after radiographic confirmation of severe hyaline membrane disease

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was obtained. In addition a dopamine drip was needed for hypotension. On the third day of life, indomethacin was started due to detection of a hemodynamically significant PDA. On the sixth day of life, abdominal distension developed followed by emesis of bilious and bloody gastric contents, frequent apnea and concomitant bradycardia, decreased urine output, hypotension, and poor perfusion. An abdominal radiograph revealed bowel loops with suspicious pneumatosis intestinalis of the bowel on the left side (Fig. 1). No portal venous gas was observed. Thus, a diagnosis of stage IIa NEC was suspected. The patient's condition was not alleviated by intensive medical treatment and conservative management including nasogastric decompression, bowel rest, fluids, intravenous nutrition and the use of broad-spectrum antibiotics.

On the eighth day of life, plain abdominal radiography revealed free air under the diaphragm and mid-abdomen (Fig. 2). Laboratory analysis of the blood revealed a hemoglobin of 8.7 g/dL, white blood cell count of 22,620/μL, platelet count of 103,000/μL, aPTT of 86.3 seconds, PT of 18.0 seconds (INR 1.76), D-dimer of 885 ng/mL, FDP of 49.5 μg/mL, plasma antithrombin III of 9.6 mg/dL. On physical examination, there was prominent distension of the abdomen with apparent tenderness and muscle guarding on palpation. No periumbilical discoloration was observed. In view of his laboratory analysis and the clinical condition, we felt the patient was too unstable and could not tolerate a laparatomy. Thus, a peritoneal drainage procedure was performed at the bedside in the NICU, under sterile conditions and with the use of local anesthesia. Under ultrasound guidance we punctured the abdomen just below the umbilicus with an 18-gauge IV catheter (Jelco, Medex Medical Ltd, Great Britain). A guidewire was inserted through the IV catheter which was discarded later. Using dilator instruments the abdomen was entered with care. An 8F (2.7 mm diameter) pigtail catheter (APDL drainage catheter, Meditech, Boston Scientific, MA, U.S.A.) was advanced carefully into the peritoneal cavity and positioned below the liver (Fig. 3A, 3B). The catheter was fixed and sutured in place. The end of the catheter was left free in a collection bag. Evacuation and irrigation with normal saline was not done. The characteristic of drained fluid was dark, brown and sticky. About 10 cc of fluid was drained from the catheter. Laboratory analysis of the drained fluid resulted in specific gravity, 1.030, pH, 7.8, white blood cell count, 14,400/μL (neutrophils, 80%; lymphocytes, 10%; eosinophils, 10%), red blood cell count, 480/μL (fresh form, 40%; old form, 60%). The infant had a positive peritoneal culture, Staphylococcus epidermidis. He received antibiotics for 14 days.

Fig. 1. Abdominal radiograph revealed a dilated segment of bowel with pneumatosis intestinalis (arrows).

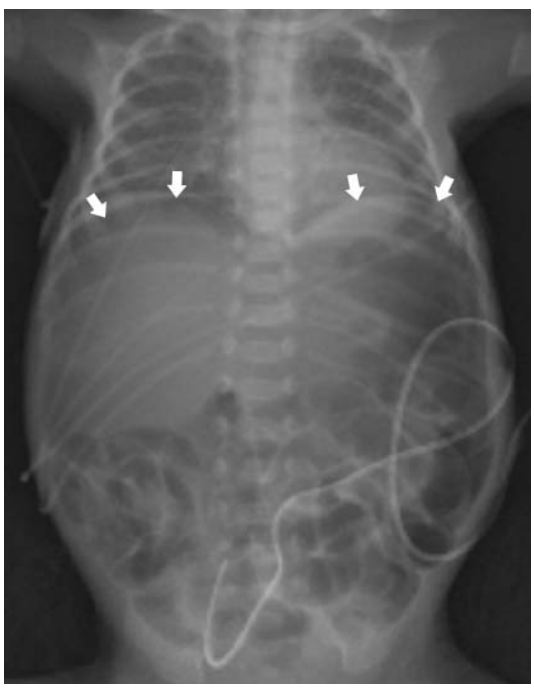

Fig. 2. Pneumoperitoneum was diagnosed by the presence of free air under the diaphragm bilaterally (arrows).

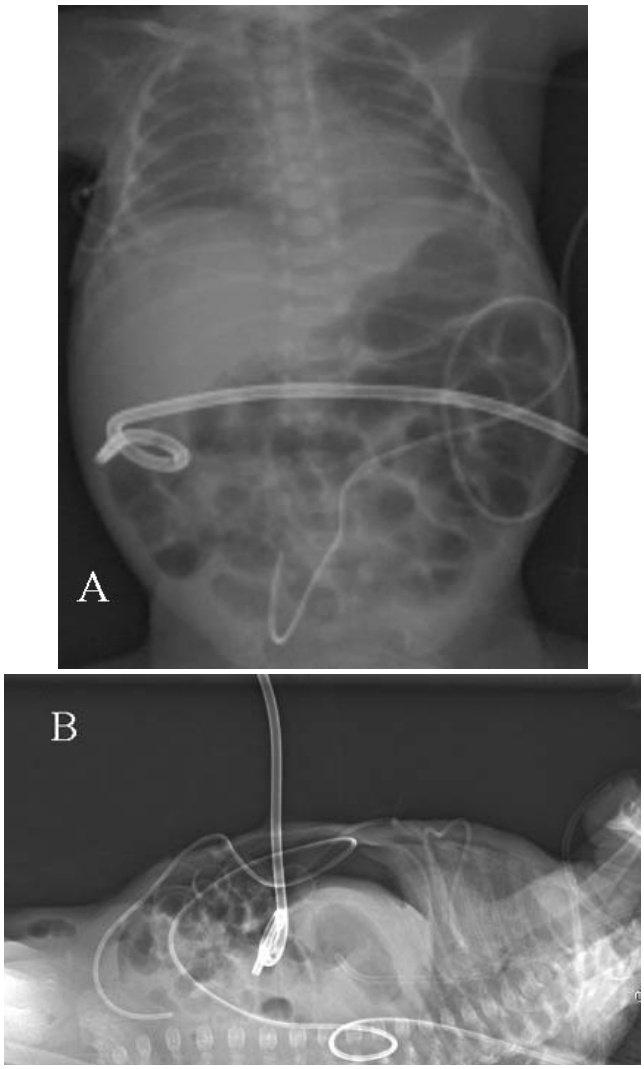

Fig. 3. Plain anterioposterior **(A)** and lateral **(B)** radiographs of the abdomen revealed that the pigtail drainage catheter was inserted into the peritoneal cavity and located below the liver showing free air under the non-dependent portion (age of 8 day).

The volume and nature of drained fluid was recorded daily. The clinical and radiological improvement was impressive starting from the following day. The clinical features were improved : a decrease of abdominal distension, hemodynamic stability, and better oxygenation were noted, and the abnormal laboratory findings stabilized gradually. Radiologically, the pneumoperitonium was observed to improve soon after peritoneal drainage was started along with a normal gas distribution. The drain was removed after 7 days, when there was no more drainage and the intestine appeared continuous on radiographs. There was no recurrent pneumoperitoneum. The infant then tolerated enteral feeding with no abdominal complications. Thus, neither contrast study nor laparotomy was needed for further therapy. The patient was discharged at 88 days of age with normal intestinal function. At the time of discharge his body weight was 2,250 g.

Discussion

NEC is almost exclusively a disease of prematurity, with >90% of all cases occurring in premature infants and 90% of those infants weighing less than 2,000 g[1, 4, 5]. ELBW infants have the greatest morbidity and highest mortality with perforated NEC[6]. Although the mortality rates are not fully stratified below 1,000 g[7-9], several case series have reported that mortality associated with intestinal perforation caused by NEC, and treated by laparotomy and bowel resection, remains at about 40% in premature infants weighing less than 1,000 g, and about 50% in those under 750 g [9-11]. Because such babies weighing less than 1,000 g generally have underlying cardiopulmonary disease, adrenal insufficiency and other disorders of immaturity, the risks associated with laparotomy are significant. In addition, the combined effects of general anesthesia and major abdominal surgery increase the risk of hemodynamic instability caused by hypotension, transfusion requirements, third spacing of fluids, and hypothermia[10]. Therefore, PD has some advantages. PD is a procedure performed at the bedside; it is technically easy and inexpensive. The infant does not have to be transported to an operating room, no general anesthesia is required, and the infant may not require a stoma or a second operation to reverse the stomas. Thus the morbidity and mortality associated with surgery may be potentially avoided. However, evaluation of the disease process and extent of involved intestine is limited by this approach, and bleeding cannot be controlled. Drainage alone will fail in infants with intestinal perforation secondary to circumferential necrosis of the intestine[12].

It remains unclear why peritoneal drainage can effectively drain the peritoneal cavity of a premature newborn but not an adult with bowel rupture. Perhaps factors such as the thin abdominal wall, a less well developed omentum and intraabdominal fat, small size of the peritoneal cavity relative to the drain, and lower intestinal bacteria counts contribute to its success in infants. In addition, the premature infant has a different host inflammatory response to injury for tissue repair compared to that of older children

and adults; the "scarless" healing process of the bowel wall may be demonstrated in these affected neonates[13, 14]. When PD was initially describes by Ein and colleagues[3], it was originally introduced as a method that could be used to stabilize critically ill low birth weight infants with intestinal perforation from NEC until the infant was stable and a laparotomy could be performed. That is, the drainage of air and stool would relieve the symptoms of the abdominal compartment syndrome and sepsis and allow the infant to better tolerate subsequent laparotomy. Subsequently, it was observed that many of these infants that had the PD did not require further treatment. Following this report and criticism for this technique, they reported additional experience with PD in 15 infants with perforated NEC. About 40% of the infants treated with PD had rapid clinical improvement and laparotomy was not performed[15]. Several additional reports noted that PD is a definitive treatment for some infants[9, 10, 16]. Morgan, et al[9] reported that drainage alone provided definitive surgical intervention in 74% of the infants with complicated NEC. Lessin, et al[10] recommend PD for the initial management of all ELBW infants with perforated NEC.

However, this does not mean that PD is superior to laparotomy for the treatment of perforated NEC. There is no clear consensus on the optimal surgical management. Some studies have demonstrated that although primary PD remained useful in the initial resuscitative management of perforated NEC, most infants ultimately require laparotomy for complications or deterioration[16, 17]. To determine the procedure of choice for perforated NEC, Moss, et al[18] performed a meta-analysis of 10 published studies comparing PD with laparotomy. The combined probability of survival did not show a significant difference when the two procedures were compared an advantage for PD or laparotomy (55% for PD vs 67% for laparotomy, P=0.27).

A recent study reported by Blakely, et al[19], compared the frequency of postoperative complications in initial drainage versus initial laparotomy subgroups, and found no significant statistical difference between the two subgroups. Some studies have concluded that the total number of comorbidities affects outcome rather than therapeutic options [12, 20]. Therefore, further prospective, controlled, and randomized trials that compare laparotomy versus drainage are needed to determine the best guidelines for treating perforated NEC.

In summary, primary PD is effective and safe in ELBW infants with perforated NEC; although it is not considered a definitive procedure, it should be considered in all cases where infants are too unstable to tolerate anesthesia and surgery.

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한 글 요 약

세균성 복막염이 동반된 천공성 괴사성 장염을 일차적 복강 배액술로 완치한 초극소 저출생 체중아 1례

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괴사성 장염은 미숙아에서 외과적 응급 수술을 요하는 가장 흔한 질환이며 여전히 높은 이환율과 사망률을 차지하고 있다. 괴사성 장염의 치료로는 전통적으로 천공성 괴사성 장염인 경우 개복술이 시행되어 왔으나 미숙아에서 일차적 복강 배액술이 시도된 이후 초극소 저출생 체중아에서도 양호한 결과를 보인 사례들이 보고되고 있다. 저출생 체중아에서 천공성 괴사성 장염 치료시 일차적 복강 배액술과 개복술 중 어느 시술이 더 우수한 결과를 보이는지에 대한 연구는 아직 진행 중에 있으나 일차적 복강 배액술은 전신 마취나 수술을 시행하기에는 불안정한 환자 상태인 경우에 시행을 고려할 수 있다. 저자들은 초극소 저출생 체중아에서 천공성 괴사성 장염으로 인한 세균성 복막염 치료로 환아 상태상 전신 마취 및 수술을 바로 시행하기에는 어려워 일차적 복강 배액술을 시행 후 호전된 1례를 경험하였기에 문헌 고찰과 함께 보고하는 바이다.

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