Gastric perforation in an extremely low birth weight infant recovered with percutaneous peritoneal drainage

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Neonatal gastric perforation is an uncommon but life-threatening condition, which is mainly encountered in premature infants. Primary surgical repair is the principal mode of the treatment. Gastric perforation in neonates improving with percutaneous peritoneal drainage alone has not been described previously. Therefore, an extremely low birth weight infant is presented herein in order to emphasize that gastric perforation may improve with percutaneous peritoneal drainage alone. Isolated gastric perforations in newborn infants may be improved with percutaneous peritoneal drainage alone without need for primary surgical repair.

Key words: conservative management, extremely low birth weight infant, gastric perforation.

Gastric perforation is an uncommon clinical condition in newborns, and is mainly seen in premature infants and threatens life when diagnosed and treated late[1]. Its incidence is reported to be 1 in 5,000 live births[2]. Gastric perforations constitute 7% of all gastrointestinal perforations[3]. In most cases, the underlying cause could not be precisely determined. Prematurity is a facilitating factor[4]. Some mechanical factors are also considered as risk factors (i.e. the mechanical pressure by nasogastric or orogastric catheters, excessive gastric distension due to positive pressure ventilation)[4,5]. Despite early diagnosis and treatment, the mortality rate is still high due to accompanying problems. Success can be achieved with early recognition of the clinical signs in the patients at risk and rapid interventions for treatment[6].

Although primary surgical repair is the principal mode of treatment, there is an exceptional case report in which perforation improved spontaneously with conservative treatment[7]. Nevertheless, to the best of our knowledge, it has not been described previously that gastric perforation in neonates improved with percutaneous peritoneal drainage alone. Therefore, an extremely low birth weight (ELBW) infant is presented herein in order to emphasize that gastric perforation may improve with percutaneous peritoneal drainage alone.

Case Report

A male baby was born at 28 weeks' gestation by emergent cesarean section, due to oligohydramnios and fetal distress, to a 21-year-old gravida 1, para 1 mother. His birth weight was 1000 g. On prenatal history, although there was a history of early rupture of the membranes lasting about 48 hours, there was no history of preterm labor, abruption of placenta, maternal chorioamnionitis, fever, antibiotic use, or steroid receipt. After delivery, because of insufficient spontaneous respiration, cardiopulmonary resuscitation was attempted at once including endotracheal tube-positive pressure ventilation and chest compression lasting about 30 seconds. The patient responded well to resuscitation by turning pink in color and with an increase in heart rate (>100 beats/min). The Apgar scores were 3 and 7 at one and five minutes, respectively. No esophageal intubation or placement of a nasogastric tube was undertaken in the delivery room. He

also did not receive surfactant or any drug in the delivery room. Following resuscitation, the patient was transferred to our neonatal intensive care unit (NICU). On admission to NICU, blood gas analysis revealed mild hypoxemia with an oxygen partial pressure of 43 mmHg and oxygen saturation of 73%. He was put on mechanical ventilation and then given surfactant because of the severe respiratory distress syndrome. Subsequently, because of the persistence of low oxygen saturation levels and insufficient aeration of the lungs on auscultation and follow-up X-rays, he was given two additional doses of surfactant within the first 24 hours of life. The patient's poor clinical condition did not allow us to perform an echocardiographic study for evaluation of cardiac status until the 10th postnatal day, when echocardiography revealed a hemodynamically insignificant small ductal shunt. Because he was hypotensive, probably due to a hemodynamically significant ductal shunt, inotrope support was given with dopamine. Over the clinical course, the patient needed resuscitation several times. He did not receive postnatal steroid therapy for hypotension resistant to dopamine or any nonsteroidal anti-inflammatory drugs (i.e. indomethacin or ibuprofen) for treatment of a hemodynamically significant ductal shunt. At the 30th hour of the life, his general condition deteriorated suddenly with a distended abdomen and increased respiratory distress. Drainage of the free air, saliva and gastric content was observed from the placed nasogastric tube. To that point, he had not received any feeds. At that time, complete blood count, biochemical analyses and C-reactive protein levels were found within normal limits. Abdominal X-ray demonstrated excessive amount of free air in the peritoneal cavity (massive pneumoperitoneum) with a small stomach shadow (Fig. 1A). However, there was no preceding bowel dilatation or pneumatosis on previous X-rays. These clinical and radiological findings led us to the clinical suspicion of gastric perforation.

When the baby's clinical status worsened, insertion of a Penrose drain was done under local anesthesia without performing a preceding percutaneous peritoneal paracentesis. Radiocontrast imaging of the stomach prior to the laparotomy was not considered. An excessive amount of free air and some amount of gastric content, but no intestinal content or bile, were drained from the abdominal cavity when the Penrose drain was first placed (Fig. 1B). Afterward, surgical repair was planned when the patient's general status would permit it. On the 10th day of the hospitalization, with recovery in the general status of the patient, follow-up abdominal X-ray demonstrated complete resolution of the free air with presence of gastric gas shadow (Fig. 1C). While the drain was in place, drainage of gastric contents and saliva was noted, but there was no drainage of intestinal content. Overall, the patient was mechanically ventilated for 15 days. Feeding with mother's milk was started on the 24th postnatal day via a nasogastric tube. Without requirement for primary surgical repair, percutaneous drainage was terminated on the 27th postnatal day. After tolerating oral feeding and showing an increase in weight, the patient was discharged from the hospital on the 72nd postnatal day. He remains well after eight months of follow-up.

Discussion

Although the most common cause of gastrointestinal perforation in premature infants is necrotizing enterocolitis, perforation in the present case was thought to be gastric in origin based on his clinical and radiological findings. The etiopathogenesis of gastric perforation in newborns is still not completely understood. It is encountered mainly in premature infants and male babies. The first opinion on the pathogenesis is that it develops spontaneously due to congenital gastric muscular agenesis, with no accompanying gastrointestinal problem[8]. It is also suggested that gastric acidity reaches its highest level on the second day of life and the perforations are likely related to the high level[9]. It has been reported that ischemic necrosis due to hypoxia, perinatal stress and shock[10] and extensive resuscitation performed postnatally[5] could also cause gastric perforation. Furthermore, some pathologies like duodenal obstruction and tracheoesophageal fistula may increase the risk of gastric perforation by causing excessive gastric distension[11]. It is also known that dexamethasone and indomethacin may cause gastric perforation in newborns[11], but these were not administered to our patient. There are some cases (20%) not related to any known underlying factors that are accepted

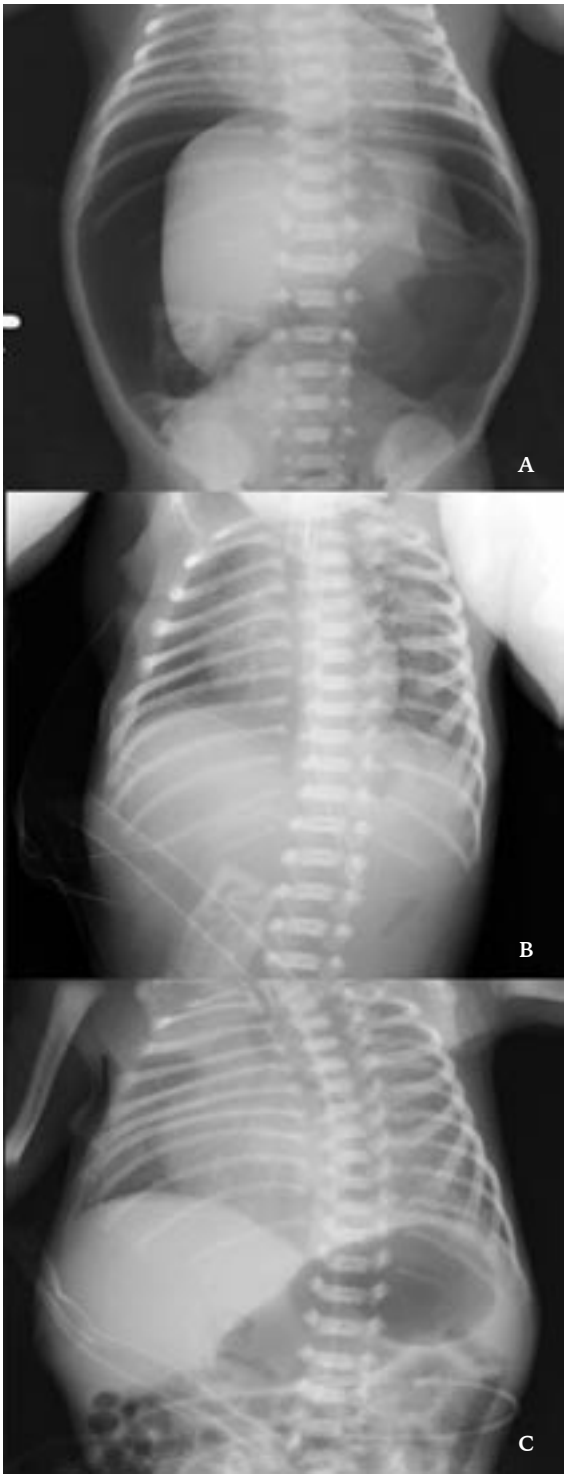

Figure 1: Abdominal radiographs of the patient. **A)** note the excessive amount of free air in the peritoneal cavity (massive pneumoperitoneum) with a small stomach shadow; **B)** note the lower tip of the orogastric/nasogastric catheter and absence of gas shadows in the abdomen following insertion of the drain; **C)** complete resolution of the free air with presence of gastric gas shadow on follow-up abdominal radiograph.

as spontaneous/idiopathic. Recent studies suggested that spontaneous gastric perforation may be caused by the lack of intestinal Cajal cells[12]. In present case, it is considered that, on the basis of prematurity, perinatal stress and hypoxia together with gastric distension due to positive pressure ventilation might have played a role in the etiopathogenesis of gastric perforation.

As in this case, abdominal distension with sudden onset and rapid progress and tachypnea with respiratory distress are the most common clinical findings. Other clinical findings include feeding intolerance, temperature instability, acidosis, and shock[1,10]. Gastric perforation is the most common cause of pneumoperitoneum in the first week of life. On the abdominal X-ray, as with our patient's radiological findings, an oval gas shadow was reported in which the lower and upper margins were constituted by the pelvic floor structures and the diaphragm with an opacity related to the falciform ligament laying on the medial axis at the midline of this shadow[13]. As seen in the present case, free air under the diaphragm and the absence of gastric air together with the displacement of intraabdominal organs are the other important radiological findings that support the diagnosis[14].

Idiopathic spontaneous intestinal perforation is a currently described novel condition, which does not show clinical or diagnostic features of necrotizing enterocolitis. Neonatal stress consequent to preterm birth is a determining factor in the etiopathogenesis. Infants under 28 weeks of gestational age and with LBW show a particular predisposition to this entity[15]. In the present case, massive pneumoperitoneum with a small stomach shadow on the abdominal X-ray, drainage of an excessive amount of free air and some gastric contents but of no intestinal content or bile when the drain was first placed, and drainage of gastric contents and saliva but of no intestinal content while the drain remained in place excluded the diagnosis of intestinal perforation.

Due to the fact that gastric perforation is a rapidly progressing pathological condition, early diagnosis and treatment are very important. Urgent primary surgical repair together with medical approaches including reconstitution of liquid-electrolyte balance, correction of

acidosis and use of wide spectrum antibiotics are the appropriate treatment modalities. These patients usually need intubation and mechanical ventilation support because of respiratory distress, which is caused by the excessive amount of free air in the peritoneal cavity that leads to diaphragmatic pressure. With percutaneous peritoneal paracentesis in the preoperative period, respiratory distress and abdominal compartment syndrome developing secondary to abdominal distension caused by free air can be reduced[7,8].

Primary surgical repair is the principal mode of treatment, and partial gastric resection may sometimes be required. However, there is an exceptional case report in which perforation improved spontaneously with conservative treatment[7]. In the present case, however, gastric perforation improved with percutaneous peritoneal drainage alone. This approach can be considered especially in patients in whom their clinical condition does not permit surgical repair. However, if persistence of free air, ongoing acidosis and presence of peritonitis findings exist, surgical exploration should be performed.

The mortality rates vary between 27% and 83%[5,6]. Despite the decrease in mortality rates in recent years with early diagnosis and appropriate surgical intervention, mortality is still high due to accompanying pathologies that cause multi-organ failure. The degree of prematurity, the duration of peritoneal contamination and asphyxia are the most important factors increasing the mortality rate[1,5,6]. Problems like sepsis and respiratory distress are frequently accompanying clinical conditions in premature infants, which increase the mortality. With early diagnosis and appropriate treatment, the prognosis of idiopathic perforations is reported to be better than with gastric perforations secondary to an underlying cause[5,11,16].

In conclusion, the gastric perforation in our patient was thought to have developed due to gastric distension as a result of positive pressure ventilation on the basis of prematurity and perinatal stress. It has been observed that isolated gastric perforations in newborn infants may be improved with percutaneous peritoneal drainage alone without need for primary surgical repair.

REFERENCES

1. Lin CM, Lee HC, Kao HA, et al. Neonatal gastric perforation: report of 15 cases and review of the literature. Pediatr Neonatol 2008; 49: 65-70.
2. Jawad AJ, Al-Rabie A, Hadi A, et al. Spontaneous neonatal gastric perforation. Pediatr Surg Int 2002; 18: 396-399.
3. Attridge JT, Clark R, Walker MW, Gordon PV. New insights into spontaneous intestinal perforation using a national data set: (2) two populations of patients with perforations. J Perinatol 2006; 26: 185-188.
4. Duran R, Inan M, Vatansever U, Aladag N, Acunas B. Etiology of neonatal gastric perforations: review of 10 years' experience. Pediatr Int 2007; 49: 626-630.
5. Rosser SB, Clark CH, Elechi EN. Spontaneous neonatal gastric perforation. J Pediatr Surg 1982; 17: 390-394.
6. Leone RJ Jr, Krasna IH. 'Spontaneous' neonatal gastric perforation: is it really spontaneous? J Pediatr Surg 2000; 35: 1066-1069.
7. Guha DK, Rashmi A, Khanduja PC, Kochar M. Neonatal gastric perforation. Report of 3 cases with one survival. Indian J Pediatr 1969; 36: 404-406.
8. Herbut PA. Congenital defect in the musculature of the stomach with rupture in the newborn infant. Arch Pathol 1943; 36: 191-194.
9. Miller FA. Neonatal gastrointestinal tract perforations. Lancet 1957; 77: 439-442.
10. Kuremu RT, Hadley GP, Wiersma R. Neonatal gastric perforation. East Afr Med J 2004; 81: 56-58.
11. Grosfeld JL, Molinari F, Chaet M, et al. Gastrointestinal perforation and peritonitis in infants and children: experience with 179 cases over ten years. Surgery 1996; 120: 650-656.
12. Ohshiro K, Yamataka A, Kobayashi H, et al. Idiopathic gastric perforation in neonates and abnormal distribution of intestinal pacemaker cells. J Pediatr Surg 2000; 35: 675-676.
13. Rampton JW. The football sign. Radiology 2004; 231: 81-82.
14. Durham MM, Ricketts RR. Neonatal gastric perforation and necrosis with Hunt-Lawrence pouch reconstruction. J Pediatr Surg 1999; 34: 649-651.
15. Messina M, Molinaro F, Ferrara F, Messina G, Di Maggio G. Idiopathic spontaneous intestinal perforation: a distinct pathological entity in the preterm infant. Minerva Pediatr 2009; 61: 355-360.
16. Dillon PW, Cilley RE. Lesions of the stomach. In: Ashcraft KW, Murphy PJ, Sharp RJ, Sigalet DL, Snyder CL (eds). Pediatric Surgery (3rd ed). Philadelphia: WB Saunders; 2000: 391-402.