A rare case of multiple duodenal perforations in early infancy

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Duodenal perforation in early infancy is an uncommon condition. We describe a case of duodenal perforation from suspected ulcer. A premature boy was born at the gestational age of 26 weeks with a birth weight of 764 g. The Apgar score at 1 min was 3 and at 5 min had decreased to 2. He was given intermittent mandatory ventilation for one month after the birth. Ninety-eight days after birth, the infant's abdomen became distended. A supine and cross-table lateral radiograph of the abdomen revealed massive pneumoperitoneum. An exploratory laparotomy was performed, which revealed two perforations in the anterior wall of the first portion of the duodenum. The operation procedure was direct closure and intra-abdominal drainage. On the postoperative first day, he had central urorrhagia from hematencephalon. The patient's growth after surgery has been normal, with no recurrence of duodenal ulcer.

Key words: duodenal perforation, infant.

Generally, duodenal perforation is a relatively rare disease in infants. Duodenal perforation in low-birth-weight (LBW) infants has been reported to be caused by the following factors: unknown reasons[1], peptic ulcer[2,3], placement of a gastric tube[4], and placement of a face mask and prolonged dependence on a respirator[5]. We treated an infant who had extremely low birth weight and suffered multiple duodenal perforations. In the present study, we report the clinical course with a review of the related literature.

Case Report

A 98-day-old newborn boy (40 weeks and 3 days) presented with the chief complaint of abdominal distension. Review of the medical history revealed that breech presentation, early rupture of membranes and intrauterine infection necessitated the mother's emergency cesarean section at 26 weeks and 2 days gestation. The birth weight was 764 g and Apgar score was 3/2. Surfactant replacement therapy and thyroid hormone replacement therapy were adopted to treat birth respiratory distress syndrome (RDS) and hypothyroidism, respectively. Oral intake was initiated from day 7 and a respirator was needed to control respiration for approximately one month after birth. Body weight increased in a relatively steady manner. The abdomen was suddenly swollen on the 98th day after birth and the scout film of the abdomen revealed abdominal free air. The patient was diagnosed with gastrointestinal perforation and referred to this department.

Physical examination at this presentation revealed weight 1,070 g, blood pressure 60/35 mmHg, regular pulse of 140 beats per minute, regular respiratory rate of 46 per minute and temperature 37.3°C. The whole abdomen was swollen, although no discoloration of the abdominal wall or the scrotum was observed. No symptoms suggestive of cyanosis were recognized.

Blood analysis findings included the following: white cell count 5,500/mm^3, red cell count 4,700,000/mm^3, hemoglobin 14.0 g/dl, platelet count 605,000/mm^3 and C-reactive protein (CRP) 2.78 mg/dl.

Scout film of the abdomen (supine position) (Fig. 1) revealed abdominal free air under the diaphragm. Based on these findings, a diagnosis of gastrointestinal perforation was made and the patient underwent emergency operation.

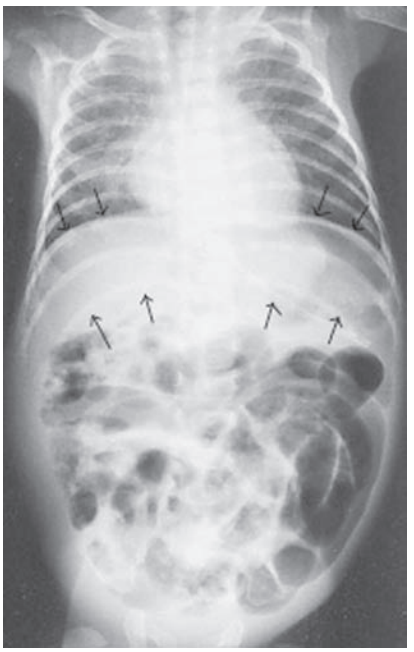

Fig. 1. Scout film of the abdomen (supine position) revealed abdominal free air under the diaphragm (arrowed part).

A transverse upper abdominal incision was made and an excessive amount of light yellow and slightly cloudy ascites was observed mainly in the upper abdomen. Two perforations of approximately 4 mm and 6 mm in diameter respectively were observed in the anterior wall of the first portion of the duodenum (Fig. 2). Closure of the two perforations by simple interrupted suture using 5-0 silk required 2 and 3 stitches, respectively. The drainage tube was placed for discharge.

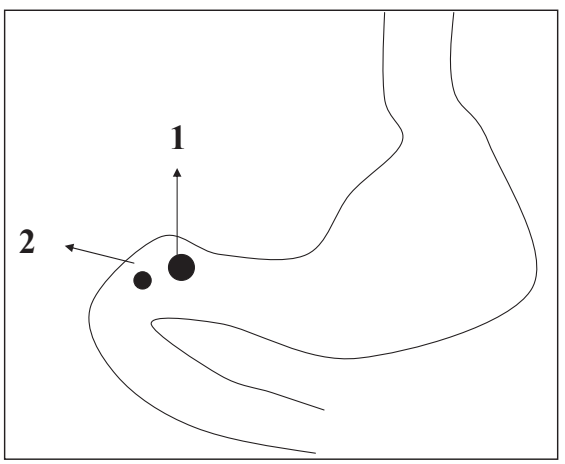

Fig. 2. Two perforations were observed in the anterior wall of the duodenal bulb (1: perforation of 6 mm in diameter; 2: perforation of 4 mm in diameter).

A nasogastric tube was placed for postoperative decompression. H_2 blocker (1 mg/kg/day) and antibiotics (penicillin 240,000 U/day, gentamicin sulfate 3.0 mg/day) were administered. Before the surgery, fresh frozen plasma was concomitantly used for blood transfusion. However, postoperative reduction in platelet count resulted in disseminated intravascular coagulation syndrome (DIC) leading to hemorrhage in the brain parenchyma on the 1st hospital day after the surgery. From the 34th day after the surgery, oral intake was resumed and normal weight gain was confirmed. The postoperative examination of blood showed increased values of *Helicobacter pylori* IgG antibody (positive, 27 U/ml, normal value ≤10 U/ml) and gastrin (480 pg/ml, normal value ≤200 pg/ml). A test on a stool specimen for *Helicobacter pylori* was positive. The parents' medical histories were examined and no history of peptic ulcer was recognized.

Discussion

Several researchers have reported duodenal perforation in children in Japan. Our literature review, however, yielded only four cases of duodenal perforation in neonates and infants[6]. We found only 11 patients with birth weight below 2,500 g (1 in Japan and 10 foreign).

We studied a total of 12 patients with birth weight below 2,500 g (1 previously reported in Japan, 10 foreign, and the present case) and summarized the reported factors of this disease (Table I). The gestational age was less than 30 weeks in seven of 12 cases and the mean gestational age was 30.8±4.3 weeks (mean±SD). Three patients, who had birth weight below 1,000 g, were classified as extremely LBW infants. The mean birth weight of the 12 patients was 1,558.72±535 g (mean±SD). The birth weight of the present patient (764 g) was the lowest among the 12 patients. The gender of six of the 12 patients was reported (4M, 2F), reflecting higher incidence among boys. The symptoms of duodenal perforation included abdominal distension, diarrhea, apnea and cardio-respiratory arrest. Of these, abdominal distension was regarded as the characteristic manifestation of the disease as it was recognized in six cases. Compared with duodenal perforation in ordinary infants, the disease adversely affects the respiratory condition immediately after its onset in

Table I. Duodenal Perforation in Low-Birth-Weight Infants

Author	Gestational age (wk)	Birth weight (g)	Sex	Symptoms	Etiology	Day of diagnosis	Site of perforation	Surgery	HP	Ventilator	Outcome	No. of patients
Freeark et al[3], 1961	1: unknown 2: unknown	1: 1,953.75 2: 2,463.45	1: M 2: M	1: diarrhea, abdominal distension 2: diarrhea, abdominal distension	1: ulcer 2: ulcer	1: 6 wks 2: 7 wks	1: first portion 2: first portion	closed with omental graft	unknown	unknown	1 survived	2
Boros et al[4], 1974	30	937	M	abdominal distension, Apnea	internal tube feeding	4 days	third portion	unknown	unknown	unknown	unknown	1
Steves et al[2], 1987	mean 30	mean 1,450	unknown	unknown	acute ulcer	mean 66 days 30~158 days	unknown	primary closure	unknown	3: low pressure	1 died of non-correctable congenital heart disease	4
Prabhakar et al[7], 1991	unknown	unknown	unknown	unknown	spontaneous	unknown	unknown	unknown	unknown	unknown	unknown	2
Miller et al[1], 1990	30	1,343	F	apnea, CRA, abdominal distension	spontaneous	9 hrs after birth	third portion 1.5 cm	primary closure	unknown	peak inspiratory pressure 14 cmH2O	unknown	1
Kohtani et al[9], 1998	41	2,290	F	vomiting, abdominal distension	ulcer	2 days	first portion 8 mm	primary closed with omental graft	unknown	–	survived 7 years	1
This case 2003	26	764	M	abdominal distension	ulcer	98 days	first portion 2 holes 4 mm, 6 mm	primary closure	+	–	survived 6 months	1

the LBW infants. By promptly introducing appropriate therapeutic interventions, we can avoid the risk of further aggravation leading to serious conditions. The time from birth to development of duodenal perforation ranged from nine hours to 158 days (50.0±34 days, mean±SD). In the present case, the disease developed on the 98th day after birth and the interval was rather long. According to Zamir et al.[8], gastrointestinal perforation develops in neonates most frequently within one week after birth. Therefore, most of the patients suffer the disease within two weeks after birth. On the other hand, the number of cases of duodenal perforation that developed within two weeks after birth was limited and only two neonates suffered the disease during that period. Of the 12 patients, eight including our patient suffered duodenal perforation resulting from ulcer. Four suffered spontaneous duodenal perforation and one suffered the disease associated with placement of a gastric tube. Although ulcer seems to be a critical factor in duodenal perforation, no findings supporting the diagnosis of ulcer were found in the reports other than of our case. We considered this a case of duodenal perforation caused by ulcer based on results of the stool specimen test and the *Helicobacter pylori* IgG antibody values. According to the scout films of the abdomen taken some weeks before the surgery, the tip of the gastric tube was located in the upper part of the stomach rather than the duodenum. Therefore, it is not likely that placement of the gastric tube caused duodenal perforation. Besides these 12 patients with duodenal perforation, those with gastrointestinal perforation resulting from various causes were reported by Garland et al.[5]. These patients suffered gastrointestinal perforation associated with placement of face masks or air pressure created by long-term placement of a respirator. Of the 12 infants whose data were used for our statistical analysis, five were on a respirator before the surgery and the airway pressure was adjusted to a low level. Because respiration was not mechanically controlled in the present case, duodenal perforation developed as a result of some event other than placement of a respirator. Clear description of the site of perforation was found in some reports. According to the data obtained, perforation was detected in the first portion of duodenum in four cases and in the third portion in two cases. In view of the fact that ulcer frequently caused perforation, we speculated that the common site was the first portion of the duodenum. Only our patient suffered two duodenal perforations. Ulcer was considered as the cause of multiple perforations.

We examined the reports that included descriptions of surgical procedures and found that primary closure was selected as the therapeutic intervention in these cases. Prognosis should be guarded since it depends largely on the preoperative condition. The survival rate was low and only six patients including the present patient survived. Because peritonitis in neonates and LBW infants often develops into sepsis or DIC, early detection and early treatment are essential to assure better survival.

We encountered an infant with a rare disease. The patient had extremely LBW and suffered multiple duodenal perforations at the age of three months. Generally, the patient with upper gastrointestinal perforation has a good prognosis for survival. However, neonates or LBW infants often progress to a serious situation immediately after development of gastrointestinal perforation. Early detection of perforation and early introduction of appropriate therapeutic interventions are indispensable for preventing further progression of the disease.

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