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CASE REPORT

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Abdominal wall plasty for a premature infant with congenital diaphragmatic hernia

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Abstract This paper reports a premature infant with a congenital diaphragmatic hernia (CDH) who underwent an abdominal wall plasty to enlarge the abdominal cavity, one of twin infants born at 32 weeks weighing 1,255 g. After stabilization, the herniated viscera were reduced from the pleural cavity and the abdominal wall muscle and skin layers were replaced by a Gore-tex patch without closure of the diaphragmatic defect. Respiratory and circulatory conditions were stable during the perioperative period. Postoperatively, a roentogenogram showed expansion of the lung. However, his condition deteriorated 24 h after surgery, triggered by intratracheal suction, and he died on the 4th day of life despite the use of high-frequency oscillation, catecholamines, and vasodilators. Postmortem examination showed severely hypoplastic lungs. Abdominal wall plasty may be a less invasive initial procedure, however, further studies, such as comparison with the standard method or conservative management, are needed using a large clinical group or animal models to justify the usefulness of this procedure.

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Introduction

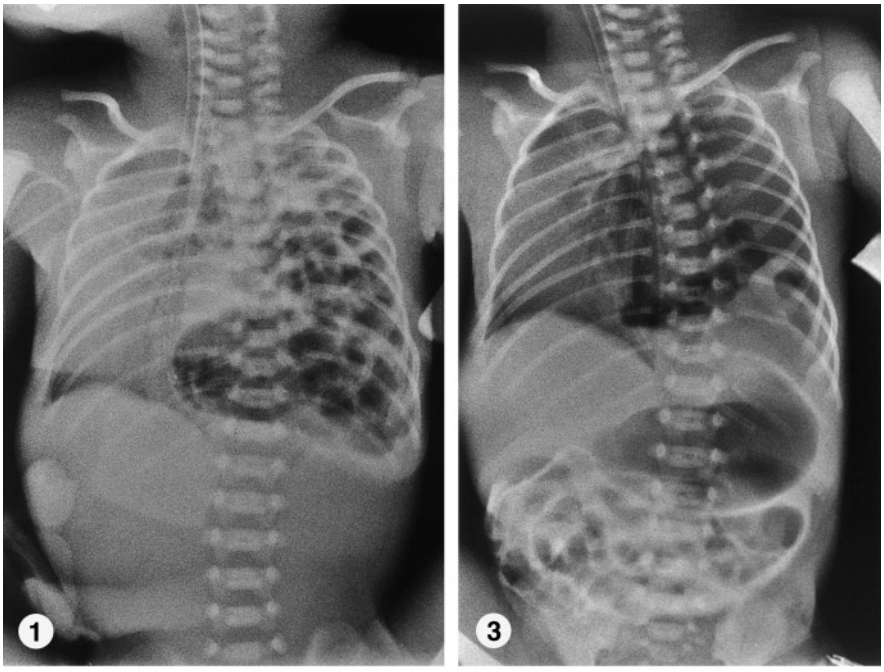
Treatment of congenital diaphragmatic hernia (CDH) is based on induction of the development of the hypoplastic lungs by reducing the herniated abdominal viscera from the thoracic cavity. Newborn infants with severely hypoplastic lungs should undergo stabilization of their general condition using conventional ventilation, paralysis, high-frequency oscillation (HFO), or extracorporeal membrane oxygenation (ECMO), before a surgical procedure is performed [1, 2, 4, 10–12]. In premature infants with severe respiratory distress due to additional lung prematurity, closure of the diaphragmatic defect and abdominal wall may worsen the condition and ECMO is generally contraindicated [4].

This paper reports a premature baby with CDH in whom the less invasive initial procedure of abdominal wall plasty with a Gore-tex patch was performed to enlarge the abdominal cavity without closure of the diaphragmatic defect.

Case report

An 18-year-old primigravida developed premature rupture of the membranes during a twin pregnancy at an estimated 32 gestational weeks. Up to this time she had not been seen by an obstetrician. After a spontaneous delivery, both infants were tracheally intubated because of severe respiratory distress. The first infant, a male weighing 1,255 g, was diagnosed as having a left CDH on a roentogenogram (Fig. 1). A "ground-glass" appearance of the lung fields was seen in the other infant. The initial arterial blood gas data from the CDH patient showed PH 7.087, PaO_2 39.2 mmHg, $PaCO_2$ 73.6 mmHg, and base excess (BE)-9.9 mEq/l on a conventional ventilator with FiO_2 70%, ventilation rate 60/min, peak inspiratory pressure 20 cm H_2O, and end-expiratory pressure 4 cm H_2O. He was given muscle relaxants and deep sedation and treated with intratracheal administration of pulmonary surfactant. After these therapies, a blood gas analysis showed PH 7.398, PaO_2 215.8 mmHg, $PaCO_2$, 41.8 mmHg, and BE + 1.3 mEq/l on the same ventilatory parameters without a significant difference between pre- and postductal arteries; the hemodynamic status was stable. He underwent reduction of the herniated viscera at 12 h of life.

In the neonatal intensive care unit (NICU), an abdominal wall patch plasty was performed through a transverse supraumbilical incision (Fig. 2). The stomach, spleen, and small and large intestine, which were herniated into the left pleural cavity through a 3-cm posterolateral diaphragmatic defect with a defective posterior ridge, were reduced into the abdominal cavity. The diaphragmatic defect was not closed. A thick Gore-tex patch was sutured to the edges of the incision to enlarge the abdominal cavity and keep the reduced organs in the abdomen. The operation lasted 75 min. Postoperatively, a roentogenogram showed slightly expanded lungs with the abdominal organs remaining in the abdomen (Fig. 3). The arterial blood gas data and hemodynamics were stable during the perioperative period. However, 24 h after surgery the blood gas parameters



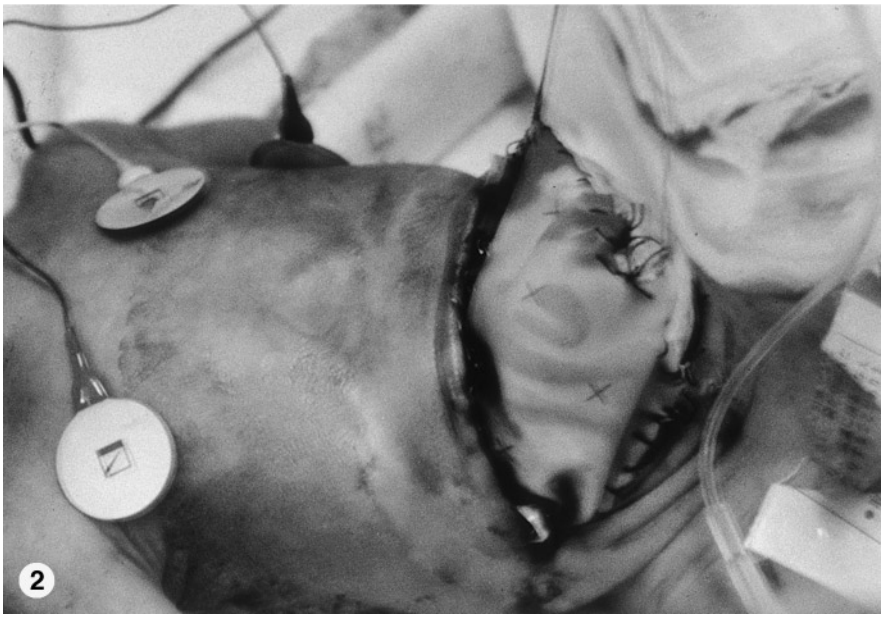


Fig. 1 Postnatal roentogenogram showed abdominal organs herniated into left pleural cavity, both lungs severely compressed and opaque

Fig. 2 Abdominal wall patch plasty performed through supraumbilical transverse incision

Fig. 3 Postoperative roentogenogram showing abdominal organs remaining in enlarged abdominal cavity, trachea shifted to center, right lung more translucent, and left pleural cavity containing air and effusion

worsened, triggered by intratracheal suction as the patient started to move. His condition progressively deteriorated despite the use of HFO and administration of catecholamines and vasodilators, with subsequent death on the 4th day of life. Postmortem examination showed severely hypoplastic lungs with a lung-body weight ratio [13] of 0.0078.

Discussion

Surfactant deficiency has been demonstrated to be associated with CDH from experimental studies and clinical results [6, 7, 13]. In addition, premature delivery itself usually produces hyaline membrane disease (HMD) caused by inadequate secretion of surfactant. Our patient was born at around 32 weeks of gestation with a body weight of 1,255 g, and his twin clinically showed severe HMD. In addition, the arterial blood gas data improved after intratracheal administration of pulmonary surfactant as well as other agents. Thus, respiratory failure in the present case may have been enhanced by HMD from the prematurity in addition to the hypoplastic lungs.

Treatment of premature infants with CDH should depend on the patient's condition. Groner et al. [8] reported a 960-g infant with CDH who survived after conventional ventilation and transabdominal closure of the diaphragmatic defect shortly after birth. The lung hypoplasia in Groner's patient [8], however, appeared milder than that in the present case on the postnatal roentogenogram. Treatment for more compromised infants with CDH should be initiated by satisfactory stabilization of pulmonary hemodynamics with intubation, mechanical ventilation, vasodilators, catecholamines, HFO, or ECMO before surgical reduction of the herniated viscera [1, 2, 4, 10–12]. In the present case, both the arterial blood gas data and hemodynamic condition stabilized after surfactant replacement, muscle relaxants, and deep sedation. Although muscle paralysis has been recommended for initial stabilization by many authors [1, 2, 4, 10–12], Stolar's group recently recommended maintenance of spontaneous respiration without using muscle relaxants [14].

The generally recommended transabdominal operation for CDH is direct or patch closure of the diaphragmatic defect with closure of the abdominal incision or formation of a ventral hernia [3]. However, these procedures usually require transfer to the operating room, which sometimes causes problems, and the operations take a long time. When the abdomen is closed after reduction of the herniated viscera into the abdominal cavity and clo-

sure of the diaphragmatic defect, the intra-abdominal pressure increases and may result in decreased venous return and compression of the pleural cavity, with decreasing respiratory compliance [10, 11]. Subsequently, hypotension and decreased ventilatory volume worsen gas exchange and pulmonary hemodynamics, resulting in increased pulmonary vascular resistance and persistent fetal circulation [2, 10, 11]. In severely compromised infants with prematurity, cardiac anomalies, neonatal distress, etc., deterioration may easily occur even after satisfactory stabilization.

Although a full repair of the diaphragmatic defect could have been accomplished in the present case even in the NICU, we selected the much less invasive procedure of abdominal wall plasty without closure of the diaphragmatic defect as an initial surgical procedure because the 3-cm defect with a defective posterior ridge seemed to require a patch closure in this 1,255-g infant. The present procedure is basically the same as the lateral abdominal patch used as a part of intrauterine correction of CDH reported by Harrison et al. [9] and of ventral hernia as reported by De Lorimier [3].

The total operation time of 75 min may be acceptable, but can be shortened. There was no concern about problems associated with transfer to the operating room. This procedure does not reduce the ventilatory volume or disturb venous return. The patient had no deterioration due to surgical stress during the perioperative period. This procedure may induce development of the hypoplastic lungs by keeping the herniated viscera within the enlarged abdominal cavity. The postoperative roentogenogram of our patient showed slightly expanded lungs. This procedure may be indicated for CDH infants with more complicated problems such as prematurity or associated severe anomalies, for whom ECMO or closure of the diaphragmatic defect would be difficult. A disadvantage of the procedure may be infection, especially when persisting more than 1 week; renewal of the patch or closure of the diaphragmatic defect should be considered before infection occurs.

Our patient died despite the procedure. Although the cause of death was not clear, the lung-body weight ratio [13] was very low. As mentioned above, the respiratory failure may have been enhanced by lung prematurity. It is regrettable that the CDH was not diagnosed prenatally, because his 18-year-old mother had not been seen by an obstetrician until the occurrence of premature rupture of the membranes. Prolongation of the pregnancy to gain maturity could have resulted in a better outcome by appropriate prenatal management.

Another problem may be the timing of the operation. Conservative therapy may reduce the volume of the herniated viscera in the chest by decompression of the gastrointestinal tract [1, 2, 4, 10–12]. It is recommended to continue it for more than 24 h until the respiratory and circulatory conditions become stabilized [1, 2, 4, 10–12]. This patient underwent the procedure 12 h after birth, when the arterial blood gas data were satisfactory without a persistent fetal circulation and the circulatory status was stable. For 24 h after the operation he had been doing well, with good respiratory and circulatory parameters. Deterioration was encountered 36 h after birth and was triggered by intratracheal suction, probably because of spasms in the pulmonary microcirculation, when he began to move. Even if the patient had been managed conservatively for several more days, the deterioration might have occurred, and the effect of mediastinal shift to distend the lungs during conservative management could not have been greater after another procedure. Therefore, further study is required to compare the effectiveness of this procedure and conservative management.

Abdominal wall plasty may thus be a less invasive initial procedure. However, further studies such as comparison with the standard method or conservative management using large clinical groups or animal models are needed to determine its usefulness.

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