

A case report of successful therapy for neonatal chylothorax with pneumothorax by conservative medical treatment

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Background: Neonatal chylothorax is a rare disease that causes breathing difficulties in newborns and is one of the most common causes of pleural effusion during the neonatal period. Neonatal chylothorax is often caused by lymph leakage into the chest and can be divided into the following 5 types: congenital chylothorax, traumatic chylothorax, postoperative chylothorax, embolic chylothorax, and spontaneous chylothorax. Among them, spontaneous chylothorax is the most common type of neonatal chylothorax and has unknown causes. The mortality rate of neonatal chylothorax is relatively high, but there are still no unified management guidelines or expert consensus on its treatment.

Case Description: In this article, we report the case of a child in whom a large amount of pleural effusion on both sides of the thorax was first found 3 days before delivery. During labor, extrauterine intrapartum treatment (EXIT) was administered to complete the pleural effusion puncture and drainage but was complicated by the right pneumothorax. After delivery, the patient was cured and discharged from the hospital, but required high-frequency oscillating respiratory support, continuous chest drainage, and nutritional management.

Conclusions: The effective control of the pleural effusion suction speed may reduce pneumothorax complications. Infection is the most common complication of neonatal chylothorax. Thus, the multidisciplinary collaborative diagnosis and EXIT may pave a new way for the efficient treatment of neonatal chylothorax. This successful case may serve as a reference for the management of children with congenital chylothorax.

Keywords: Neonatal chylothorax; pneumothorax; high-frequency oscillation; extrauterine intrapartum treatment (EXIT); case report

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Introduction

Neonatal chylothorax is characterized by the accumulation of pleural lymph fluid, which can be caused by the leakage of the lymph fluid into the chest, or by excessive secretion or obstruction (1). Neonatal chylothorax, which is also known as lymph node pectorales, is a rare disease that causes neonatal dyspnea, but it is one of the most common causes of neonatal pleural effusion. Neonatal chylothorax has an incidence of about 1 in 100,000 live births, and has a case fatality rate as high as 20–50% (2). Neonatal chylothorax can be divided into the following 5 categories according to the etiology. (I) Congenital chylothorax: congenital dysplasia of lymphatic system; (II) traumatic chylothorax: excessive central venous pressure resulting from labor

injury or resuscitation may result in hyperdilation and rupture of the thoracic duct; (III) postoperative chylothorax: surgical procedures near the thoracic duct may injure the trunk and branches of the thoracic duct (3); (IV) embolic chylothorax: parenteral nutrition through the central vein can lead to embolization and rupture of the thoracic duct; (V) spontaneous chylothorax: the cause is unknown, the type accounts for 50% of the neonatal chylothorax. At present, the treatment methods of newborn chylothorax: (I) repeated thoracic puncture or closed thoracic drainage; (II) nutrition management: fasting, parenteral nutrition, and pleural effusion were significantly reduced when medium chain triglyceride was fed by milk; (III) drug treatment: somatostatin, erythromycin, propranolol (4); (IV) surgical treatment: medical conservative treatment failed, surgical repair fistula, such as thoracic catheter ligation, pleuropexy. We present the following article in accordance with the CARE reporting checklist (available at https:// tp.amegroups.com/article/view/10.21037/tp-23-49/rc).

Case presentation

The baby patient was gestation 2 production 1 (G2P1), had a gestational age (GA) of 37⁺⁶ weeks, was a singleton, and had a body weight of 3,100 g. The baby patient had an Apgar score of 7 points in 1 min (1 point was deducted for respiration, muscular tension, and laryngeal reflex, respectively), 8 points in 5 min (1 point was deducted for muscular

Highlight box

Key findings

 Before delivery, a large amount of fluid was found in the bilateral pleural cavity of the fetus, and both lungs were obviously compressed and deformed. Right pneumothorax occurred within 1 hour after the pleural effusion puncture, and the pneumothorax was relieved by the second intercostal drainage and the 7/8 intercostal drainage successively. The treatment time was short, and the patient's prognosis was good without sequelae.

What is known and what is new?

- The diagnostic criteria, respiratory support, and thoracic drainage of newborn chylomax are known.
- EXIT and pneumothorax management are new approaches.

What is the implication, and what should change now?

 EXIT provides favorable conditions for lung expansion at birth for neonatal chylothorax. However, more research needs to be conducted on the timing of the delivery, the speed of the chest puncture, and the methods of chest drainage. tension and laryngeal reflex, respectively), 9 points in 10 min (1 point was deducted for muscular tension). A large amount of pleural effusion on both sides of the fetus's thorax (with a maximum range on the left side of 75 mm ×36 mm, a maximum range on the right side of 85 mm ×39 mm, lungs that were obviously deformed under pressure, and too much amniotic fluid) was first found 3 days before the birth. Prenatally, 1 course of dexamethasone was administered to promote lung maturation. The operation plan and rescue process were decided after a hospital consultation, and the baby was delivered at 10:33 on December 24, 2021. However, the baby had a poor response, limp limbs, and no spontaneous breathing.

After the pleural effusion had been located on the operating table by ultrasound, a pediatric surgeon at our hospital completed the left and right pleural puncture, pleural effusion suction, and indwelling drainage tube. About 50 and 115 mL of yellow puncture fluid was pumped out from the left and right thorax, respectively. At 10:41, the umbilical cord was cut immediately after the completion of the bilateral thoracic puncture. After the tracheal intubation (intubation depth: 9 cm, inner diameter: 3.5 mm), air bag pressurization, and oxygen resuscitation, the baby was transferred to the neonatal intensive care unit for further treatment with the assistance of a vehicle-mounted ventilator through a neonatal transfer incubator.

At the time of admission, the baby was cyanotic all over the body, and the percutaneous oxygen saturation was 70%. The inspiratory triple concave sign was visible. As the right respiratory sound was weaker than the left, and more gas was drawn out through the right drainage tube, it was thought that the patient might be complicated by the right pneumothorax. A series of treatments were administered, including assisted ventilation via a highfrequency oscillatory ventilator, bilateral thoracic closed drainage, cephalosporin + ampicillin to prevent infection, fasting, intravenous nutrition, and other treatments. Under the auxiliary ventilation of the high-frequency oscillatory ventilator, the baby still had an inspiratory triple concave sign, but the body skin became ruddy, and the percutaneous oxygen saturation reached 90–92%.

At 6+ h after birth, the transcutaneous oxygen saturation of the baby was 85%, and occasional bubbles in the right water-sealed bottle were observed. After replacing the right draft tube, continuous bubbles emerged in the watersealed bottle, and the transcutaneous oxygen saturation of the child increased to about 90%. At 9+ h after birth, the baby's percutaneous oxygen saturation was 76%, and there

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Table 1 Serial results of the laboratory tests for the newborn

Hydrothorax	3+ h	Day 2	Day 5	Day 12	Day 13	Reference values
Color	Yellow	Yellow	Yellow	Yellow	Milk white	Milk white
S-III-S	-	Negative	-	-	Negative	Positive
Albumin (g/L)	9	-	10.9	-	-	12-41.6
TP (g/L)	32.2	-	24.6	-	-	21–59
Glucose (mmol/L)	11.17	-	5.41	-	-	2.7–11.1
NC	5,575	-	2,280	-	-	×10 ⁶ /L
MC	55.75	-	501.6	-	-	×10 ⁶ /L
Lymphocyte (×10 ⁹ /L)	5.52	-	1.78	-	-	0.4–6.8
Cholesterol (mmol/L)	-	-	0.91	-	1.66	\leq the plasma levels
Triglyceride (mmol/L)	-	-	0.22	-	9.56	> the plasma levels
C/T	-	-	>1	-	<1	<1
HC	Negative	-	Negative	-	-	Negative
PGD	-	Negative	-	-	-	Negative

S-III-S, Sudan III staining; TP, total protein; NC, nucleated cells; MC, multinucleated cells; C/T, cholesterol/triglyceride; HC, hydrothorax culture; PGD, pathogen gene detection.

were no bubbles in the right water-sealed bottle. After adjusting the thoracic catheter, the baby's percutaneous oxygen saturation increased to about 90%. At 20 h after birth, the baby's percutaneous oxygen saturation was 79%, and no bubbles emerged from the right water-sealed bottle. After adjusting the position of the right thoracic catheter, the percutaneous oxygen saturation of the baby increased to about 90%. At 29 h after birth, the baby's percutaneous oxygen saturation was 87%, and still no bubbles emerged from the right water-sealed bottle. A chest catheter (with a diameter of 4 mm) was placed at the junction of the middle clavicle and the second intercostal space on the right side of the baby at a depth of 6 cm. A water-sealed bottle was connected, and continuous bubbles were observed. The skin color of the child turned ruddy. At 50 h after birth, the baby's transcutaneous oxygen saturation was 76-80%, and there were no bubbles emerging from the 7/8th intercostal water-sealed bottle on the right side. Review chest radiography showed that there was a large amount of pneumothorax on the right side (about 90% of the lung tissue was compressed), so the drainage tube was pulled out, and a chest catheter (with a diameter of 4 mm) was replaced at a depth of 6 cm. A continuous emerging bubble was observed when connecting the water-sealed bottle. The baby's body skin gradually turned ruddy, and the baby's transcutaneous oxygen saturation was about 90%.

At 72 h after birth, with ventilator assisted ventilation, the baby's percutaneous oxygen saturation reached 90-93%. There was no polypnea and a negative triple concave sign. Additionally, no bubbles emerged from the 2 right thoracic catheters, and the respiratory sounds were symmetrical of both sides. A large number of moist rales were heard in both lungs. The antibiotic was upgraded to linezolid combined with ceftazidime. At 4+ days after birth, the right intercostal thoracic catheter was clamped. At 6+ days after birth, the treatment was changed to synchronized intermittent mandatory ventilation (SIMV) mode, and the right second intercostal thoracic catheter was removed. On the 7th day after birth, nasal continuous positive airway pressure (NCPAP)-assisted ventilation was used. At 10+ days after birth, the baby was changed to oxygen inhalation in the box, and the left and right thoracic drainage volume decreased significantly, and the baby began to drink milk. On the 13th day after birth, the pleural effusion turned into milky white. On the 14th day after birth, oxygen inhalation in the box was stopped. On the 16th day after birth, the left drainage tube was removed. On the 21st day after birth, the last thoracic catheter was removed. On the 27th day after birth, the child was discharged from hospital with a good mental reaction, stable breathing, and complete enteral nutrition through oral feeding. For further details, see Tables 1-4.

All procedures performed in this study were in

 Table 2 Serial results of the laboratory tests for the newborn

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Hematologic	2+ h	Day 4	Day 7	Day 22	Reference values
White blood cell (×10 ⁹ /L)	13.96	10.77	7.40	8.84	12–20
Neutrophil (×10 ⁹ /L)	9.39	8.09	4.26	3.28	3.6-8.0
Lymphocyte (×10 ⁹ /L)	3.18	1.12	1.52	4.04	4.8–12
Red blood cell (×10 ¹² /L)	4.54	3.92	4.01	3.38	5.20-6.40
Hemoglobin (g/L)	165	144	141	113	180–190
Platelets (×10 ⁹ /L)	387	295	311	490	100–300
Procalcitonin (ng/mL)	-	1.338	0.136	_	0.000-0.100
C-reactive protein (mg/L)	-	4.32	1.44	_	0.00-8.00
ALT (U/L)	14	-	9	20	7–40
AST (U/L)	181	-	24	31	13–35
Total protein (g/L)	55.9	-	36.8	52.2	65.0–85.0
Albumin (g/L)	27.6	-	22.2	34.3	40.0–55.0
Globulin (g/L)	28.3	-	14.6	17.90	20.0-40.0
Cholesterol (mmol/L)	-	-	-	3.02	3.10–5.72
Triglyceride (mmol/L)	-	-	-	0.96	0.50–1.80
Urea (mmol/L)	2.69	-	-	2.41	2.60-7.50
Uric acid (µmol/L)	317.3	-	-	140.5	150.0–370.0
Creatinine (µmol/L)	42.4	-	-	17.6	41.0-73.0
Blood culture ①	Negative	-	Negative	-	Negative
Blood culture 2	Negative	-	Negative	-	Negative
CMV-DNA (copies/mL)	-	-	<4.000E+2	_	<4.0E+02
PGD	-	Negative		-	Negative
WES	-	-	Negative	-	Negative

Two blood cultures were collected at the same time. AST, aspartate transferase; ALT, alanine aminotransferase; CMV-DNA, cytomegalovirus deoxyribonucleic acid; PGD, pathogen gene detection; WES, whole exome sequencing.

accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was taken from the patient's legal guardians for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

In the delivery rescue process for this child, a thoracic puncture was first performed, followed by a tracheal intubation. The pleural effusion on the right side was significantly more than that on the left side, after rapid drainage during pleural puncture. It took less than 30 s to aspirate the left pleural diffusion and less than 1 min to aspirate the right pleural effusion. After a large amount of pleural effusion on the right side was aspirated, the negative pressure on the chest increased significantly, and the "T" trachea intubation and combination treatment were then conducted for anabiosis. The collapsed heterogeneous alveoli were over expanded and ruptured, resulting in pneumothorax of the right side (5). The left side had less pleural effusion than the right side, and the

Table 3 Changes of bilateral pleural effusion drainage volume.

Age	Left thoracic drainage	Right 7th/8th intercostal thoracic drainage	Total (mL)
Day 1	65	115	180
Day 2	15	20	35
Day 3	3	30	33
Day 4	20	50	70
Day 5	21	89	110
Day 6	10	17	27
Day 7	10	18	28
Day 8	15	23	38
Day 9	8	13	21
Day 10	1	5	6
Day 11	3	3	6
Day 12	0	4	4
Day 13	0	5	5
Day 14	0	2	2
Day 15	0	1	1
Day 16	Removed	0	0
Day 17	-	0	0
Day 18	-	0	0
Day 19	-	0	0
Day 20	-	0	0
Day 21	-	Removed	0

negative pressure on the chest was higher on the left side than the right side, and thus the occurrence of left-side pneumothorax was avoided. In the subsequent delivery rescues of similar patients, we recommend that the rate of pleural effusion suction be controlled according to the pleural effusion volume to reduce pneumothorax complications.

At 72 h after birth, a large number of moist rales were heard in both lungs of the baby. As the baby had lost a large amount of albumin through pleural fluid, the immunity of the body was poor. Additionally, as the baby had a long history of multiple thoracic drainage, required an invasive ventilator and other invasive operations, and had positive no specific blood infection indicators and chest X-ray changes, the diagnosis was consistent with septicemia. The upgrading of the antibiotic treatment proved to be timely and effective in improving the baby's condition. Neonatal chylothorax with large pleural effusion are more likely to be infected, so active anti-infection treatment is need.

Due to the unclear pleural effusion and the pneumothorax in early childhood, the blood oxygen of the child fluctuated repeatedly. With the clear diagnosis of pleural effusion and the following adjustment of the pleural drainage plan, the pneumothorax of the child was controlled, and the child's breathing improved significantly 2 days after birth. The ventilator was successfully withdrawn on the 7th day after birth, and the child achieved freedom of breathing on the 14th day after birth without respiratory system sequelae. The treatment of respiratory failure and pneumothorax of neonatal chylothorax with high-frequency oscillation-assisted ventilation is in line with evidence-based medicine (6), and the improvement of the baby's condition also shows that our respiratory management was effective.

A large amount of bilateral pleural effusion was found for the first time during an examination of the fetus 3 days before delivery at our hospital. The Obstetrics Department at our hospital attached great importance to it. A multidisciplinary diagnosis and treatment model was quickly implemented whereby members of the Neonatal, Pediatric Surgery, Cardiothoracic Surgery, Anesthesiology, and Ultrasound Departments were invited consult. The first intrapartum extrauterine treatment was then successfully carried out at our hospital after discussing the detailed operation plan and rescue process. The respiratory failure of the baby was quickly alleviated through active respiratory support. Neonatal chylothorax was quickly diagnosed by routine and biochemical examinations of pleural effusion 24 h after the birth, indicating the direction for subsequent thoracic drainage, infection prevention, and nutrition management. The pleural effusion of the child turned milky white 2+ days after the child began drinking milk. Following 2 pleural effusion examinations, the diagnosis of congenital chylothorax was further confirmed. The left and right thoracic drainage tubes were pulled out successively on the 16th and 21st days after birth with no recurrence. During hospitalization, no obvious abnormalities were observed based on the head ultrasound monitoring. Multidisciplinary diagnosis and treatment model provides a rapid, effective and reasonable delivery mode for congenital neonatal chylothorax. EXIT provides favorable conditions for lung expansion at birth for neonatal chylothorax, and it is effective.

On the 35th day after birth, a whole exon gene sequencing report for the baby and his parents revealed no

Age	Bedside chest X-ray changes
3 h	Massive pneumothorax on the right side; a mediastinal shift to the left; no lung texture transparent area on the right chest; the left lung texture was not clearly displayed
7 h	A large amount of pneumothorax on the right side; the change was not obvious compared to the old film from 13:31 on December 24
10 h	A large amount of air was accumulated in the right chest; a little air was accumulated subcutaneously in the right chest wall; the mediastinum moved to the left; the transmittance of both lungs was the same as before. Compared to the old film at 15:42 on December 24, the air in the right thorax was slightly reduced
24 h	A large amount of air was accumulated in the right chest; a little air was accumulated subcutaneously in the right chest wall; the mediastinum moved to the left; the transmittance of both lungs was the same as before. Compared to the old DR films at 20:26 from December 24, the air in the right thorax was slightly reduced
29 h	(I) Right pneumothorax (about 20% lung tissue compression) was significantly reduced compared to the old DR film from 11:26 on December 25; a little subcutaneous gas was observed in the right chest wall; (II) the proximal end of the drainage tube was located at the 11th–12th thoracic intervertebral space plane
50 h	(I) Massive right pneumothorax (about 90% lung tissue compression) with mediastinal hernia; (II) the proximal end of the peripherally inserted central catheter, PICC tube was located at the right cervical root; (III) the right thoracic drainage tube was located in the 6/7th intercostal space of the right chest wall; there was gas in the soft tissue of the right chest wall
72 h	(I) Compared to the old DR film from December 26, there was no obvious sign of air accumulation in the right chest cavity, and the right lung was basically re-expanded; (II) gas accumulation in the soft tissue of the right chest wall; (III) the right lung may have been slightly inflamed. Follow-up was recommended; (IV) the proximal end of the PICC tube was located at the upper edge plane of the right 1st frame
144 h	The right lung re-expanded: the right chest wall gas was absorbed: the exudative lesions of both lungs were aggravated

Table 4 Chest X-ray changes in the child

DR, digital radiography.

pathogenic or suspected pathogenic mutation consistent with the baby's phenotype. The father of the infant was contacted by telephone and informed that the growth and development of the infant at the age of 1 month was consistent with that of other infants at the same age. Thus, the baby was diagnosed with spontaneous chylothorax but had a good prognosis after treatment.

Conclusions

Congenital chylothorax is the most common cause of pleural effusion in neonates, and has a prevalence rate of about 1/10,000–1/24,000, and an overall survival rate of about 30–70% (7). Severe congenital chylothorax cases require respiratory support, which prolongs the disease course, and have significant mortality rates. Lymphatic research on newborn infants has been limited, and lymphatic examinations are not common. In the fetal period, pleural effusion increases the intrathoracic pressure, which leads to a decrease in fetal swallowing of amniotic fluid, which in turn leads to excessive amniotic fluid and premature delivery. Fetal chylothorax may also increase the risk of death and complications caused by pleural lymph accumulation, which might damage lung development, lung and cardiovascular functions, and lead to complications caused by the loss of lymphatic excretion. Prenatal interventions have been shown to improve the survival rates of children with congenital chylothorax (8-11). The majority of congenital chylothorax cases improved after conservative treatment (12,13), including pleural effusion drainage, mechanical ventilation, albumin supplementation, infection prevention, and diet adjustment. Somatostatin has been shown to be effective in some cases, but conservative medical treatment was shown to be ineffective in a few children, and surgical treatment was required (14,15). There have been many reports on the treatment of congenital chylothorax in recent years (16); however, there are still no unified treatment guidelines or expert consensus. The case report in this study could provide a reference for the management of more children with congenital chylothorax.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://tp.amegroups.com/article/view/10.21037/tp-23-49/rc

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