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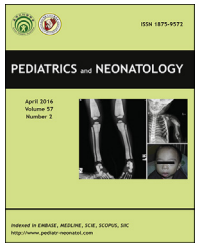

ORIGINAL ARTICLE

Prenatal Therapy Improves the Survival of Premature Infants with Congenital Chylothorax

Chia-Jung Lee [a], Po-Nien Tsao [b], Chien-Yi Chen [b], Wu-Shiun Hsieh [b], Jyun-you Liou [b], Hung-Chieh Chou [b,*]

[a] *Department of Pediatrics, Far Eastern Memorial Hospital, New Taipei City, Taiwan*
[b] *Department of Pediatrics, National Taiwan University Hospital, National Taiwan University, Medical College, Taipei, Taiwan*

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Key Words
congenital chylothorax;
hydrops;
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Background: Chylothorax is a rare condition among neonates, although it is considered clinically significant, as it is difficult to manage in these patients. In addition, the course of chylothorax varies widely. Therefore, we aimed to elucidate the clinical features and effect of prenatal therapy on the prognosis of congenital chylothorax in neonates.
Methods: We retrospectively reviewed the medical records of all infants with congenital chylothorax who were admitted to National Taiwan University Hospital, Taipei, Taiwan between January 2000 and December 2012. Their demographic characteristics, as well as their antenatal, perinatal, and postnatal information, were collected for our analysis of the mortality risk.
Results: We found 29 infants who were diagnosed with congenital chylothorax during the study period. The median gestational age at birth was 34 weeks (range, 28—41 weeks), and 71% of the infants presented with hydrops fetalis. Most cases of congenital chylothorax were bilateral (bilateral: 86.2%, unilateral: 13.79%), and the overall survival rate was 59.6%. Among the cases with a prenatal diagnosis at $\leq$ 34 weeks of gestation, infants who received prenatal therapy had a significantly higher survival rate, compared to infants who did not receive prenatal therapy (76.9% vs. 11%, respectively; $p = 0.008$).
Conclusion: We found that infants whose chylothorax was diagnosed $\leq$ 34 weeks of gestation, and who subsequently received prenatal therapy, experienced a better perinatal condition and exhibited improved postnatal outcomes.

* Corresponding author. Department of Pediatrics, National Taiwan University Hospital, Number 8, Chung-Shan South Road, Taipei 100, Taiwan.
E-mail address: hcchou@ntu.edu.tw (H.-C. Chou).

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1. Introduction

Pleural effusion is the accumulation of fluid in the pleural cavity, and can be caused by chylothorax, hemothorax, empyema, hydrothorax, or leakage of central-line fluid. Although pleural effusion is rare in the neonatal period, chylothorax (which is characterized by chyle collection) is the most common etiology.[1–4] These cases can be acquired as postoperative or iatrogenic complications, as isolated congenital causes, or in association with other congenital abnormalities, and subsequently result in chyle accumulation in the pleural space.[5–7] The severity of the clinical manifestation depends on the amount of chyle accumulated. Some infants remain asymptomatic or experience only mild respiratory distress, while others present as potentially life-threatening conditions that require urgent drainage and other resuscitative management immediately after birth.[6–8] Unfortunately, prolonged severe fetal pleural effusion can compromise normal lung maturation and progress to fetal hydrops, ultimately resulting in premature birth and pulmonary hypoplasia, with a high rate of intrauterine death and perinatal mortality.

No consensus has been reached regarding the management of congenital chylothorax, although it is commonly accepted that respiratory and life support, diet restriction, and pharmacological and surgical interventions are needed. However, the specific methods or extent of the management differ among neonatal care units. In this setting, prenatal therapies for congenital chylothorax, such as transabdominal thoracocentesis, *in utero* pleurodesis, or the placement of a thoracoamniotic shunt, may alter the natural course and improve the perinatal outcome.[8–11] Therefore, we conducted the present study to clarify whether prenatal therapy could improve the survival outcomes of infants with congenital chylothorax.

2. Methods

This retrospective study was performed in the neonatal intensive care unit (NICU) at the National Taiwan University Hospital, which is a tertiary perinatal and neonatal unit in Northern Taiwan. The study design was reviewed and approved by the Research Ethics Board Committee of National Taiwan University Hospital.

We retrospectively evaluated all neonates who were admitted to the NICU between January 2000 and December 2012 with a diagnosis of any-cause congenital pleural effusion or hydrops fetalis. We also evaluated cases that had undergone thoracocentesis or the insertion of an intercostal catheter for fluid drainage during hospitalization. From these cases, we included only neonatal cases with a confirmed diagnosis of congenital chylothorax (Figure 1). We then extracted various clinical data from the patients' medical records, including sex, birth body weight, gestational age, time of the diagnosis, etiology of the pleural effusion, underlying disease, fetal therapy, perinatal or postnatal management, respiratory condition, site and duration of the pleural effusion, morbidity, and mortality.

The diagnosis of pleural effusion was made antenatally via ultrasonography or postnatally via chest radiography or ultrasonography. Congenital pleural effusion was defined as pleural effusion that was diagnosed antenatally or at birth. The antenatal therapies included transabdominal thoracocentesis, *in utero* pleurodesis, and the placement of a thoracoamniotic shunt. The attending obstetrician made a personal decision to determine which patients received antenatal therapy. Hydrops fetalis was defined as a fetus with two or more sonographic findings of excess fluid accumulated in the form of ascites, pleural or pericardial effusions, skin edema, placental edema, or polyhydramnios.[12] The presence of chylothorax was confirmed based on the criteria that were proposed by Büttiker et al[13]: triglyceride levels of $>$ 110 mg/dL, or a total white blood cell count $>$ 1000/μL with a predominance of lymphocytes. We followed up these cases until they discharged and the mortality cases referred to the cases who did not survive at discharge.

2.1. Statistical analysis

Statistical comparisons were performed using the Mann-Whitney *U* test for continuous variables and Fisher's exact test for categorical variables. Multivariate logistic regression analysis was used to calculate the adjusted odds ratios for mortality. All statistical tests were two-tailed and

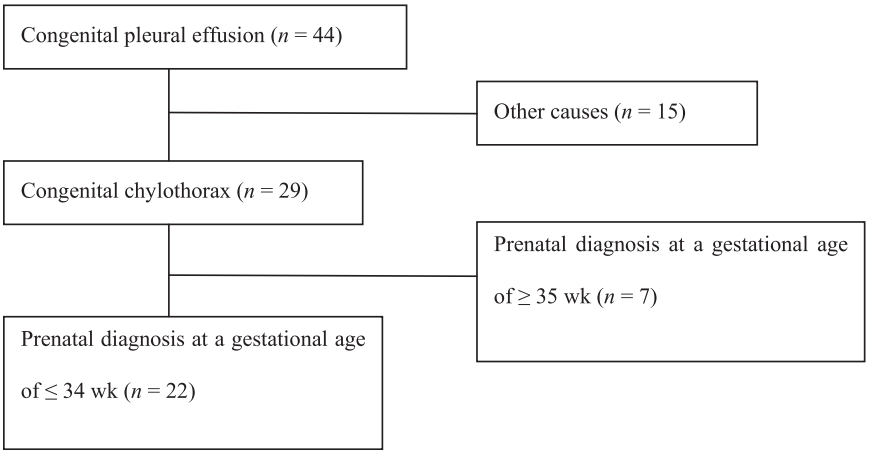

Figure 1 Flow chart of the enrollment process. We identified 44 neonates with congenital pleural effusion. Most infants had congenital chylothorax and were diagnosed before 34 weeks of gestation.