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Isolated Non-chylous Pleural Effusion in Two Neonates

INTRODUCTION

Congenital isolated pleural effusion is a rare condition, with an incidence at about 1 in 12,000 to 1 in 15,000 pregnancies (1). The content of the effusion is usually chylous, however, a minority of cases is not chylous (2, 3). The incidence of non-chylous, isolated pleural effusion in neonate and its association of chromosome anomaly have not been exactly unknown. Only one case report has been described about congenital non-chylous pleural effusion with Down syndrome (4). We experienced 2 cases of non-chylous pleural effusion, one of which was associated with Down syndrome.

CASE REPORT

Case 1

A newborn, whose mother had not received antenatal care, was transferred with respiratory distress. The initial Apgar scores were 2 at 1 min and 6 at 5 min. He had no cardiac problem. He was not febrile. Chest radiograph showed haziness of the left hemithorax. The endotracheal tube was pushed rightward, indicating mediastinal shifting (Fig. 1A). Ultrasonogram showed pleural fluid in the left hemithorax with the collapsed left lung (Fig. 1B). A chest tube was inserted for drainage of the pleural fluid which was clear. Analysis of the fluid was shown protein 34 g/L, glucose 88 mg/dL, Cl- 100 mM/L, LDH 953 I/U/L, WBC 2,000/μL and RBC 1,000/μL. Bacteriologic examination demonstrated no microorganism. Subsequent analysis of the pleural fluid following total parenteral nutrition revealed triglyceride (TG) 3 mg/dL, indicative of non-chylous pleural effusion.

His karyotype was 47,XY,+21, so he was diagnosed as Down syndrome. Follow-up chest radiograph showed well expanded both lungs without residual pleural effusion (Fig. 1C).

Case 2

A newborn at 36 weeks of gestational age was delivered by cesarean section without respiratory distress. He had been diagnosed as fetal hydrothorax by fetal ultrasonogram at 32 weeks of gestational age (not shown). The initial chest radiograph showed diffuse increased opacity of the right hemithorax with widening of pleural space (Fig. 2A). Ultrasonogram showed anechoic fluid collection in right hemithorax (Fig 2B). He underwent sono-guided thoracentesis. Analysis of pleural fluid showed protein 32 g/L, glucose 85 mg/dL, Cl- 103 mM/L, LDH 281 I/U/L, WBC 2,500/μL and RBC 220/μL. The level of TG in pleural fluid after milk feeding was 4 mg/dL. No chromosomal anomaly was detected. He had neither cardiac problem. He had neither congenital mass nor infectious disease. Follow-up chest radiograph showed clear resorption of pleural effusion (Fig. 2C). He did well until he was 4 yr old.

DISCUSSION

Neonatal pleural effusion may be congenital, inflammatory, iatrogenic following line placement, or secondary to congenital heart failure (5). The effusion is mostly unilateral and about 60% of cases has been found in the right hemithorax (2).
Among the neonatal pleural effusions, isolated pleural effusion may be diagnosed when there are no other findings of hydrops fetalis nor any inflammatory, iatrogenic, and cardiac problem (5).

The content of the isolated pleural effusion is mostly chylous, resulting from a malformation or tear in the fetal thoracic duct. Chylous pleural effusion may be initially serous and turns into chylous only after milk feeding (2). Distinguishing features of chylous effusion from serous effusion are milky-white or yellow bloody color, more than 110 mg/dL (>1.24 mMol/L) of TG level and lymphocytosis. However, lymphocytosis can be also discovered in other conditions such as tuberculosis or viral infection. Our two cases proved to non-chylous effusion after milk feeding and total parenteral nutrition containing intralipid with medium-chain TG (6).

In a minority of the cases, the content of the effusion is serous. Some authors reported that non-chylous or serous congenital pleural effusion may be associated with underlying thoracic cause such as primary lymphangiectasia, congenital cystic adenomatoid malformation, bronchopulmonary dysplasia, diaphragmatic hernia, chest wall hematoma, and pulmonary vein atresia (3, 7). Like our case, isolated non-chylous pleural effusion is rare. Several cases have been reported about congenital or fetal pleural effusion with chromosomal anomaly such as Down syndrome and Turner syndrome (8-11). Most of these pleural effusion were chylothorax or associated hydrops fetalis (6, 9). Hence, karyotyping is indicated in a fetus or newborn with isolated pleural effusion for the evaluation of associated chromosomal anomaly (8, 11). However, association of Down syndrome with isolated non-chylous pleural effusion has been rarely reported (4). One of our cases was diagnosed as Down syndrome.

Imaging diagnosis was done by plain chest radiograph and ultrasonogram with thoracentesis (5). Chest radiograph usually demonstrates homogeneous haziness in the involved hemithorax. It may be sometimes confusing. Diaphragmatic hernia filled with fluid contents, congenital chest mass, such as congenital cystic adenomatoid malformation or pulmonary sequestration and total atelectasis should be included in differential diagnosis. Chest ultrasonogram readily and noninvasively distinguish pleural effusion from other conditions (5).

The clinical course of the isolated pleural effusion is variable. Congenital pleural effusion causing pulmonary hypoplasia or maternal polyhydramnios by extrinsic compression of the fetal esophagus may be associated with high mortality (1). However, it is reported that some cases showed spontaneous resolution in utero or fared well with residual small amount of pleural effusion (7, 11).
REFERENCES


