Review

DOI: 10.5582/irdr.2017.01028

Fetal pleural effusion and Down syndrome

Li Cao[1], Yan Du[2,*], Ling Wang[3,4,5,*]

[1] *Ultrasound Department, Obstetrics and Gynecology Hospital of Fudan University, Shanghai, China;*
[2] *Office of Clinical Epidemiology, Obstetrics and Gynecology Hospital of Fudan University, Shanghai, China;*
[3] *Laboratory for Reproductive Immunology, Hospital & Institute of Obstetrics and Gynecology, IBS, Fudan University Shanghai Medical College, Shanghai, China;*
[4] *The Academy of Integrative Medicine of Fudan University, Shanghai, China;*
[5] *Shanghai Key Laboratory of Female Reproductive Endocrine-related Diseases, Shanghai, China.*

Summary **Fetal pleural effusion is a rare abnormality that results from accumulation of fluid in the chest cavity. It can be classified as primary fetal hydrothorax and secondary fetal hydrothorax. The underlying causes of pleural effusion are still unknown, and the current treatment strategies are mainly based on symptoms. The prognosis of fetal pleural effusion varies significantly, ranging from spontaneous resolution to perinatal death. Recent advances in prenatal diagnostic methods and treatment such as thoracoamniotic shunting have significantly improved the survival rates for patients with or without hydrops.**

Keywords: Pleural effusion, Down syndrome, etiology, diagnosis, treatment, prognosis

1. Introduction

Fetal pleural effusion is an abnormality resulting from accumulation of fluid in the chest cavity, and the condition was first described by Carroll in 1977 (*1*). Fetal pleural effusion is a rare condition, with a reported incidence ranging from 1/10,000 to 1/15,000 (*2-4*). The incidence of fetal pleural effusion in newborns ranges from 2.2 to 5.5 per 1,000 births (*5*). The underlying causes of fetal pleural effusion are still unclear; it can occur as an initial symptom of hydrops fetalis, but it can also occur in isolation (*6*).

2. Causes and classifications

Fetal pleural effusion can be classified as primary fetal hydrothorax and secondary fetal hydrothorax. Primary fetal hydrothorax, also known as congenital chylothorax, can result from multiple lymphatic vessel anomalies or thoracic cavity defects caused by external force, a tumor, or cardiovascular diseases (*7*). It can occur unilaterally or bilaterally and affects males more than females at a ratio of 2:1; primary fetal hydrothorax has a perinatal mortality rate of 22% to 53% (*8,9*). Secondary fetal hydrothorax is a feature of immune and non-immune hydrops. Autoimmune conditions include Rh or ABO blood type incompatibility; non-immune factors include chromosomal abnormalities, genetic disorders, infections, congenital cardiac anomalies, congenital lung anomalies, hematologic diseases, metabolic diseases, and noncardiac anomalies (*10*). Hydrops is usually bilateral, and is also often associated with ascites, pericardial effusion, subcutaneous edema, hydramnios, and placental thickening. The most common causes of non-immune hydrops are chromosomal anomalies such as Down syndrome and Turner syndrome, which can also be present with additional structural abnormalities (*1,4*).

3. Diagnosis

The treatment of fetal hydrothorax and patient prognosis are closely associated with its specific causes (*11-15*); therefore, a detailed prenatal examination and evaluation must be performed. Steps include identification of maternal blood type and screening serum specific antibodies to exclude immune hydrops, detection

Released online in J-STAGE as advance publication July 24, 2017.

*Address correspondence to:
Dr. Ling Wang, Hospital and Institute of Obstetrics and Gynecology, IBS, Fudan University, 413 Zhaozhou Road, Shanghai 200011, China.
E-mail: Dr.wangling@fudan.edu.cn

Dr. Yan Du, Office of Clinical Epidemiology, Obstetrics and Gynecology Hospital of Fudan University, No. 419 Fangxie Road, Shanghai 200011, China.
E-mail: sophiedu_61@163.com

Table 1. Characteristics and outcomes of Down syndrome cases with fetal or neonatal hydrothorax

Author	Case number	Time of detection	Unilateral or bilateral	Hydramnios (Yes/No)	Structural abnormalities (Yes/No)	Hydrops (Yes/No)	Intervention (Yes/No)	Karyotype	Outcome
Yoss *et al*. (1977) (*1*)	1	Neonatal period	Unilateral (right)	No	No	No	No	47,XY,+21	Alive
	1	Neonatal period	Unilateral (left)	Yes	AVSD	No	No	46,XY,-22,+t 21q:22q	Alive
Foote *et al*. (1986) (*22*)	1	34 weeks of gestational age	Unilateral (right)	Yes	No	No	No	46,XY,t(21:21)	Unknown
	1	28 weeks of gestational age	Unilateral (left)	Yes	AVSD	No	No	47,XY,+21	Unknown
Modi *et al*. (1987) (*23*)	1	Neonatal period	Unilateral (right)	Unknown	No	No	No	47,XX,+21	Alive
Rodeck *et al*. (1988) (*24*)	1	32 weeks of gestational age	Unilateral (right)	Yes	AVSD	No	No	47,XX,+21	Alive
Blott *et al*. (1988) (*25*)	1	32 weeks of gestational age	Unilateral (right)	Unknown	ASD	No	Thoracoamniotic shunting	Trisomy 21	Alive
Petrikovsky *et al*. (1991) (*26*)	1	34 weeks of gestational age	Bilateral	No	No	No	No	Trisomy 21	Alive
	1	18 weeks of gestational age	Bilateral	No	Fifth digit clinodactyly	No	No	47,XX,+21	TOP
Ilagan *et al*. (1992) (*27*)	1	32 weeks of gestational age	Bilateral	Yes	No	Yes	No	47,XX,+21	Neonatal death after 3 days of birth
Hamada *et al*. (1992) (*28*)	1	36 weeks of gestational age	Bilateral	No	No	Yes	No	47,XY,+21	Alive
Sherer *et al*. (1993) (*29*)	1	18 weeks of gestational age (resolved after 1 week)	Unilateral (left)	No	Fifth digit clinodactyly	No	No	47,XY,+21	TOP
Achiron *et al*. (1995) (*30*)	1	18 weeks of gestational age	Unilateral	No	No	No	No	47,XX,+21	Alive
	1	28 weeks of gestational age	Bilateral	No	No	No	No	47,XY,+21	IUD
Yamamoto *et al*. (1996) (*31*)	1	32 weeks of gestational age	Bilateral	No	No	Yes	No	47,XX,+21	Alive
Rotmensch *et al*. (1997) (*32*)	1	24-28 weeks of gestational age	Unknown	Unknown	Pyelectasis	Yes	No	Trisomy 21	Unknown
Puddy *et al*. (1999) (*33*)	1	29 weeks of gestational age	Bilateral	Yes	No	Yes	Thoracocentesis (29 weeks of gestational age); Thoracoamniotic shunting (29 weeks of gestational age)	46,XX/47XX, +21 mosaicism	Alive
Turan *et al*. (2001) (*34*)	1	Neonatal period	Bilateral	Unknown	No	No	No	47,XY,+21	Alive
Hwang *et al*. (2003) (*35*)	1	Neonatal period	Unilateral (left)	Unknown	No	No	No	47,XY,+21	Alive
Picone *et al*. (2005) (*36*)	1	Pregnancy period	Unknown	Unknown	Unknown	Unknown	Thoracoamniotic shunting	Trisomy 21	TOP
	1	Pregnancy period	Unknown	Unknown	Unknown	Unknown	Thoracoamniotic shunting	Trisomy 21	IUD
Kallanagowdar *et al*. (2006) (*37*)	1	Pregnancy period	Unilateral (left)	Unknown	AVSD, fifth digit clinodactyly	No	No	47,XX,+21	Alive
Rocha *et al*. (2006) (*38*)	1	Neonatal period	Unilateral (left)	Unknown	Unknown	Unknown	No	Trisomy 21	Alive
	1	Neonatal period	Bilateral	Unknown	Unknown	Unknown	No	Trisomy 21	Neonatal death
Albano *et al*. (2007) (*39*)	1	32 weeks of gestational age	Unilateral (right)	Yes	Hepatic hemangioma	Yes	No	47,XX,+21	Alive
Kim *et al*. (2008) (*40*)	1	32 weeks of gestational age	Bilateral	Yes	Posterior urethral valve	No	Repeated amniocentesis, then thoracoamniotic shunting and vesicoamniotic shunting	47,XY,+21	Alive
Ergaz *et al*. (2009) (*41*)	1	Neonatal period	Right	Unknown	AVSD, TOF	No	No	47,XY,+21	Alive
	1	Neonatal period	Bilateral	Unknown	VSD	Yes	No	47,XY,+21	Alive

Abbreviations: ASD, atrial septal defect; ABSD, atrioventricular septal defect; IUD, intrauterine death; TOP, termination of pregnancy; VSD, ventricular septal defect.

Table 1. Characteristics and outcomes of Down syndrome cases with fetal or neonatal hydrothorax (continued)

Author	Case number	Time of detection	Unilateral or bilateral	Hydramnios (Yes/No)	Structural abnormalities (Yes/No)	Hydrops (Yes/No)	Intervention (Yes/No)	Karyotype	Outcome
Yinon *et al.* (2010) (*42*)	3	Pregnancy period	Unknown	Unknown	Unknown	Unknown	Unknown	Trisomy 21	Unknown
Ugras *et al.* (2010) (*43*)	1	Neonatal period	Left	Unknown	Anal atresia, VSD	No	No	47,XY,+21	Alive
Caserio *et al.* (2010) (*44*)	3	Pregnancy period	Unknown	Unknown	Unknown	Unknown	Unknown	Trisomy 21	Unknown
Ruano *et al.* (2011) (*45*)	5	Pregnancy period	Unknown	Unknown	Ventriculomegaly; complex heart defect; absence of nasal bones	Unknown	Unknown	Trisomy 21	Unknown
Karagol *et al.* (2012) (*46*)	1	30 weeks of gestational age	Unilateral (right)	Yes	Fifth-finger clinodactyly	No	No	47, XY, +21	Alive
Bialkowski *et al.* (2015) (*47*)	1	Newborn	Unknown	Unknown	Unknown	Unknown	Unknown	Trisomy 21	Alive
Mallmann *et al.* (2016) (*48*)	14	Pregnancy period	Unknown	Unknown	Unknown	Unknown	Thoracoamniotic shunting	Trisomy 21	Unknown
Li *et al.* (1998) (*49*)	1	Pregnancy period	Bilateral	No	No	No	No	46,XY,+21-3	Neonatal death
Guan *et al.* (1998) (*50*)	1	Pregnancy period	Unilateral (right)	No	Congenital hypothyroidism	No	No	47, XY, +21	Neonatal death
Wei *et al.* (2005) (*51*)	1	Neonatal period	Unilateral (left)	Unknown	Known	No	No	47, XX, +21	Alive
Hu *et al.* (2007) (*52*)	1	Pregnancy period	Unilateral (right)	No	ASD	No	No	46,XY,-14,+t(21;14)	Alive
Li *et al.* (2009) (*53*)	1	Pregnancy period	Unilateral (left)	No	No	No	No	47, XX, +21	Alive
Zhang *et al.* (2011) (*54*)	1	Neonatal period	Bilateral	Yes	No	Yes	No	Trisomy 21	Neonatal death
Zhang *et al.* (2011) (*55*)	1	Neonatal period	Left	Unknown	No	No	No	47, XY, +21	Alive
Jiang *et al.* (2012) (*56*)	1	Pregnancy period	Unknown	Unknown	Pulmonary defect	Unknown	No	Trisomy 21	TOP
La *et al.* (2014) (*57*)	1	23 weeks of gestational age	Unknown	Unknown	Unknown	Unknown	No	Trisomy 21	Neonatal death
Li *et al.* (2015) (*58*)	1	Neonatal period	Unknown	Unknown	Unknown	Unknown	No	Trisomy 21	Neonatal death

Abbreviations: ASD, atrial septal defect; ABSD, atrioventricular septal defect; IUD, intrauterine death; TOP, termination of pregnancy; VSD, ventricular septal defect.

of infectious factors including TORCH, syphilis, and parvovirusB19, and performing a K-B test to exclude fetal maternal transfusion syndrome. A careful ultrasound examination should also be performed to observe the placenta, amniotic fluid, and fetal structure (and especially the fetal heart), and pulse Doppler should be used to detect the blood flow spectrum of the umbilical artery, middle cerebral artery, and venous system (*16,17*). Karyotyping or genetic testing is also routinely performed, especially in fetuses in which early pleural effusion has been detected (*18-21*).

4. Hydrothorax and chromosomal anomalies

Studies have shown that chromosomal anomalies are associated with fetal and/or neonatal hydrothorax. Table 1 summarizes the literature regarding characteristics and outcomes of Down syndrome cases involving patients with fetal or neonatal hydrothorax.

5. Treatment and prognosis

The prognosis for fetal pleural effusion is highly variable and difficult to predict, ranging from spontaneous resolution to progression to fetal hydrops and eventual perinatal death (*59-64*). The current strategy for treatment of fetal pleural effusion is based more on symptoms rather than underlying causes. Primary hydrothorax with small volumes of pleural fluid and no hydrops is more likely to resolve or remain stable, so more conservative treatment can be provided. Aubard *et al*. reported that the survival rate for conservative treatment of primary hydrothorax was 24% when hydrops was present and 75% when it was not (*60*), and Rustico *et al*. noted similar survival rates of 35% and 73%, respectively (*4*). Survival rates have improved significantly in recent years, and Wada *et al*. (*21*) reported survival rates of 58% and 97.8%, respectively, that can be largely attributed to improved methods of neonatal treatment. Thoracentesis is easy to perform and can reduce distress and improve fetal pulmonary development, but the procedure must be repeated after 24-48 hours in many patients, so thoracoamniotic shunting is usually recommended for fetuses with hydrops (*65*). Recent studies have indicated that the survival rate for congenital hydrothorax with hydrops is around 60% for patients treated with thoracoamniotic shunting, approximately 50% for those treated with thoracentesis, and from 35% to about 60% for those receiving conservative treatment (*4,21*).

Acknowledgements

This work was supported by the National Natural Science Foundation of China (Grant No. 31571196 to Ling Wang), the 2015 Program to Guide Medicine ("Yixueyindao") of the Shanghai Municipal Science and

Technology Commission (Grant No. 15401932200 to Ling Wang), the FY2008 JSPS Postdoctoral Fellowship for Foreign Researchers (P08471, Ling Wang), the National Natural Science Foundation of China (Grant No. 30801502 to Ling Wang), the Shanghai Pujiang Program (Project No. 11PJ1401900, Ling Wang), and the Shanghai Program for Support of Leading Disciplines-Integrated Chinese and Western Medicine (Project No. 20150407).

References

1. Yoss BS, Lipsitz PJ. Chylothorax in two mongoloid infants. Clin Genet. 1977; 12:357-360.
2. Ibrahim H, Asamoah A, Krouskop RW, Lewis D, Webster P, Pramanik AK. Congenital chylothorax in neonatal thyrotoxicosis. J Perinatol. 1999; 19:68-71.
3. Wilson RD, Pawel B, Bebbington M, Johnson MP, Lim FY, Stamilio D, Silber A, Zakii E, Flake AW. Congenital pulmonary lymphangiectasis sequence: A rare, heterogeneous, and lethal etiology for prenatal pleural effusion. Prenat Diagn. 2006; 26:1058-1061.
4. Rustico MA, Lanna M, Coviello D, Smoleniec J, Nicolini U. Fetal pleural effusion. Prenat Diagn. 2007; 27:793-799.
5. Shih YT, Su PH, Chen JY, Lee IC, Hu JM, Chang HP. Common etiologies of neonatal pleural effusion. Pediatr Neonatol.2011; 52:251-255.
6. Hagay Z, Reece A, Roberts A, Hobbins JC. Isolated fetal pleural effusion: A prenatal management dilemma. Obstet Gynecol. 1993; 81:147-52.
7. van Straaten HL, Gerards LJ, Krediet TG. Chylothorax in the neonatal period. Eur J Pediatr. 1993; 152:2-5.
8. Derderian SC, Trivedi S, Farrell J, Keller RL, Rand L, Goldstein R, Feldstein VA, Hirose S, MacKenzie TC. Outcomes of fetal intervention for primary hydrothorax.J Pediatr Surg. 2014; 49:900-903; discussion 903-904.
9. Rocha G. Pleural effusions in the neonate. Curr Opin PulmMed. 2007; 13:305-311.
10. Carlson DE, Platt LD, Medearis AL, Horenstein J. Prognostic indicators of the resolution of nonimmune hydrops fetalis and survival of the fetus. Am J Obstet Gynecol. 1990; 163:1785-1787.
11. Soto-Martinez M, Massie J. Chylothorax: Diagnosis and Management in Children. Paediatr Respir Rev. 2009; 10:199-207.
12. Tutor JD. Chylothorax in infants and children. Pediatrics. 2014; 133:722-733.
13. McGrath EE, Blades Z, Anderson PB. Chylothorax: Aetiology, diagnosis and therapeutic options. Respir Med. 2010; 104:1-8.
14. Nair SK, Petko M, Hayward MP. Aetiology and management of chylothorax in adults. Eur J Cardiothorac Surg. 2007; 32:362-369.
15. Fairfax AJ, McNabb WR, Spiro SG. Chylothorax: A review of 18 cases. Thorax 1986; 41:880-885.
16. Simpson JH, McDevitt H, Young D, Cameron AD. Severity of non-immune hydrops fetalis at birth continues to predict survival despite advances in perinatal care. Fetal Diagn Ther. 2006; 21:380-382.
17. Bellini C, Donarini G, Paladini D, Calevo MG, Bellini T, Ramenghi LA, Hennekam RC. Etiology of non-immune hydrops fetalis: An update. Am J Med Genet A. 2015; 167A:1082-1088.
18. Cadkin A, Pergament E. Bilateral pleural effusion at 8.5 weeks' gestation with Down syndrome and Turner syndrome. Prenat Diagn. 1993; 13:659-660.
19. Shimizu T, Hashimoto K, Shimizu M, Ozaki M, Murata Y. Bilateral pleural effusion in the first trimester: A predictor of chromosomal abnormality and embryonic death. Am J Obstet Gynecol. 1997; 177:470-471.
20. Hashimoto K, Shimizu T, Fukuda M, Ozaki M, Shimoya K, Koyama M, Murata Y. Pregnancy outcome of embryonic/fetal pleural effusion in the first trimester. J Ultrasound Med. 2003; 22:501-505.
21. Wada S, Jwa SC, Yumoto Y, Takahashi Y, Ishii K, Usui N, Sago H. The prognostic factors and outcomes of primary fetal hydrothorax with the effects of fetal intervention. Prenat Diagn. 2017; 37:184-192.
22. Foote KD, Vickers DW. Congenital pleural effusion in Down's syndrome. Br J Radiol. 1986; 59:609-610.
23. Modi N, Cooke RW. Congenital non-chylous pleural effusion with Down's syndrome. J Med Genet. 1987; 24:567-568.
24. Rodeck CH, Fisk NM, Fraser DI, Nicolini U. Long-term in utero drainage of fetal hydrothorax. N Engl J Med. 1988; 319:1135-1138.
25. Blott M, Nicolaides KH, Greenough A. Pleuroamniotic shunting for decompression of fetal pleural effusions. Obstet Gynecol. 1988; 71:798-800.
26. Petrikovsky BM, Shmoys SM, Baker DA, Monheit AG. Pleural effusion in aneuploidy. Am J Perinatol. 1991; 8:214-216.
27. Ilagan NB, Liang KC, Delaney-Black V, Perrin E. Hydrops fetalis, polyhydramnios, pulmonary hypoplasia, and Down syndrome. Am J Perinatol. 1992; 9:9-10.
28. Hamada H, Fujita K, Kubo T, Iwasaki H. Congenital chylothorax in a trisomy 21 newborn. Arch Gynecol Obstet. 1992; 252:55-58.
29. Sherer DM, Abramowicz JS, Sanko SR, Woods JR Jr. Trisomy 21 presented as a transient unilateral pleural effusion at 18 weeks' gestation. Am J Perinatol. 1993; 10:12-13.
30. Achiron R, Weissman A, Lipitz S, Mashiach S, Goldman B. Fetal pleural effusion: The risk of fetal trisomy. Gynecol Obstet Invest. 1995; 39:153-156.
31. Yamamoto T, Koeda T, Tamura A, Sawada H, Nagata I, Nagata N, Ito T, Mio Y. Congenital chylothorax in a patient with 21 trisomy syndrome. Acta Paediatr Jpn. 1996; 38:689-691.
32. Rotmensch S, Liberati M, Bronshtein M, Schoenfeld-Dimaio M, Shalev J, Ben-Rafael Z, Copel JA. Prenatal sonographic findings in 187 fetuses with Down syndrome. Prenat Diagn. 1997; 17:1001-1009.
33. Puddy V, Lam BC, Tang M, Wong KY, Lam YH, Wong K, Yeung CY. Variable levels of mosaicism for trisomy 21 in a non-immune hydropic infant with chylothorax. Prenat Diagn. 1999; 19:764-766.
34. Turan O, Canter B, Ergenekon E, Koç E, Atalay Y. Chylothorax and respiratory distress in a newborn with trisomy 21. Eur J Pediatr. 2001; 160:744-745.
35. Hwang JY, Yoo JH, Suh JS, Rhee CS. Isolated non-chylous pleural effusion in two neonates. J Korean Med Sci. 2003; 18:603-605.
36. Picone O, Benachi A, Mandelbrot L, Ruano R, Dumez Y, Dommergues M. Thoracoamniotic shunting for fetal pleural effusions with hydrops. Am J Obstet Gynecol. 2004; 191:2047-2050.

37. Kallanagowdar C, Craver RD. Neonatal pleural effusion. Spontaneous chylothorax in a newborn with trisomy 21. Arch Pathol Lab Med. 2006; 130:e22-23.
38. Rocha G, Fernandes P, Rocha P, Quintas C, Martins T, Proença E. Pleural effusions in the neonate. Acta Paediatr. 2006; 95:791-798.
39. Albano G, Pugliese A, Stabile M, Sirimarco F, Arsieri R. Hydrops foetalis caused by hepatic haemangioma. Acta Paediatr. 1998; 87:1307-1309.
40. Kim A, Park IY, Young L, Shin JC. A rare case of posterior urethral valve and pleural effusion in Down syndrome with successful intrauterine shunt. Fetal Diagn Ther. 2008; 24:372-375.
41. Ergaz Z, Bar-Oz B, Yatsiv I, Arad I. Congenital chylothorax: Clinical course and prognostic significance. Pediatr Pulmonol. 2009; 44:806-811.
42. Yinon Y, Grisaru-Granovsky S, Chaddha V, Windrim R, Seaward PG, Kelly EN, Beresovska O, Ryan G. Perinatal outcome following fetal chest shunt insertion for pleural effusion. Ultrasound Obstet Gynecol. 2010; 36:58-64.
43. Ugras M, Yakinci C, Ozgor B. Somatostatin for the treatment of chylothorax in a premature baby with Down syndrome. Pediatr Int. 2010; 52:e141-143.
44. Caserío S, Gallego C, Martin P, Moral MT, Pallás CR, Galindo A. Congenital chylothorax: From foetal life to adolescence. Acta Paediatr. 2010; 99:1571-1577.
45. Ruano R, Ramalho AS, Cardoso AK, Moise K Jr, Zugaib M. Prenatal diagnosis and natural history of fetuses presenting with pleural effusion. Prenat Diagn. 2011; 31:496-499.
46. Karagöl BS, Okumuş N, Karadağ N, Zenciroğlu A. Isolated congenital pleural effusion in two neonates. Tuberk Toraks. 2012; 60:52-55.
47. Bialkowski A, Poets CF, Franz AR. Congenital chylothorax: A prospective nationwide epidemiological study in Germany. Arch Dis Child Fetal Neonatal Ed. 2015; 100:F169-72.
48. Mallmann MR, Graham V, Rösing B, Gottschalk I, Müller A, Gembruch U, Geipel A, Berg C. Thoracoamniotic Shunting for Fetal Hydrothorax: Predictors of Intrauterine Course and Postnatal Outcome. Fetal Diagn Ther. 2017; 41:58-65.
49. Li H. A case of congenital bilateral pleural effusion. Journal of clinical radiology.1998; 17:331. (in Chinese)
50. Guan L, Xie J, Du J. Neonatal 21-trisomy syndrome with congenital hypothyroidism in 1 case. Sichuan medicine.1998; 19:540. (in Chinese)
51. Wei F, Zhu J, Yan J. 21-trisomy syndrome and chylothorax in 1 case. J Appl Clin Pediatr. 2005; 20:236-238. (in Chinese)
52. Hu J, Mei G, Huang C. One case of congenital pleural effusion with 21-trisomy syndrome. Chin J Pediatr.2007; 20:236-237. (in Chinese)
53. Li Z. A case of neonatal congenital pleural effusion with mongolism. Journal of Clinical Pulmonary Medicine.2009; 14:1138. (in Chinese)
54. Zhang M, Wei D, Tang J. One case of neonatal chylothorax combined with 21-trisomy syndrome. Journal of Practical Medicine.2011; 27:3670. (in Chinese)
55. Zhang H, Zhang X, Li X, Wang Y, Feng Q. A case of 21-trisomy syndrome with congenital pleural effusion as the main manifestation and literature review. Chin J Clinicians. 2011; 5:4889-4890. (in Chinese)
56. Jiang X, Deng X, Ling C, Tang Y, Liang H. Ultrasonographic diagnosis and clinical prognosis of fetal congenital diseases of the lung. Chin J Med Ultrasound. 2012; 9:602-607. (in Chinese)
57. La R, Hong Kang H. Diagnosis and treatment of neonatal congenital chylothorax: A report of 11 cases. Journal of Qinghai Medicine.2014; l44:20-22. (in Chinese)
58. Li B, Zhang Q, Chen X. One case of fetal and neonatal congenital pleural effusion: A report of 6 cases and literature review. Chin J Appl Clin Pediatr. 2015; 30:1090-1092. (in Chinese)
59. Longaker MT, Laberge JM, Dansereau J, Langer JC, Crombleholme TM, Callen PW, Golbus MS, Harrison MR. Primary fetal hydrothorax: Natural history and management. J Pediatr Surg. 1989; 24:573-576.
60. Aubard Y, Derouineau I, Aubard V, Chalifour V, Preux PM. Primary fetal hydrothorax: A literature review and proposed antenatal clinical strategy. Fetal Diagn Ther. 1998; 13:325-333.
61. Randenberg AL. Nonimmune hydrops fetalis part II: Does etiology influence mortality? Neonatal Netw. 2010; 29:367-380.
62. Petersen S, Kaur R, Thomas JT, Cincotta R, Gardener G. The outcome of isolated primary fetal hydrothorax: A 10-year review from a tertiary center. Fetal Diagn Ther. 2013; 34:69-76.
63. Kyeong KS, Won HS, Lee MY, Shim JY, Lee PR, Kim A. Clinical outcomes of prenatally diagnosed cases of isolated and nonisolated pericardial effusion. Fetal Diagn Ther. 2014; 36:320-325.
64. Ota S, Sahara J, Mabuchi A, Yamamoto R, Ishii K, Mitsuda N. Perinatal and one-year outcomes of non-immune hydrops fetalis by etiology and age at diagnosis. J Obstet Gynaecol Res. 2016; 42:385-391.
65. Bianchi S, Lista G, Castoldi F, Rustico M. Congenital primary hydrothorax: Effect of thoracoamniotic shunting on neonatal clinical outcome. J Matern Fetal Neonatal Med. 2010; 23:1225-1229.

(Received May 10, 2017; Revised May 22, 2017; Accepted May 27, 2017)