

Primary Fetal Hydrothorax: Successful Perinatal Outcomes With Single Thoracentesis Just Prior to Delivery

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Abstract

Objective: The aim of this study was to review our experience with isolated primary fetal hydrothorax emphasizing prenatal course, management, and neonatal outcome.

Materials and Methods: We reviewed the prenatal course and outcome of fetuses with primary fetal hydrothorax. A detailed ultrasonography was done for diagnosis of hydrothorax and associated fetal anomaly scanning. All cases were followed-up without intervention and single thoracentesis were performed just prior to delivery when indicated.

Results: Study group included five cases of isolated primary fetal hydrothorax among 22 cases of nonimmune hydrops fetalis. Four cases were diagnosed as bilateral fetal hydrothorax, whereas one of the five cases presented as unilateral hydrothorax. Ultrasonographic and other diagnostic investigations revealed no anomaly other than hydrothorax in all fetuses. Single thoracentesis of 100-200 ml pleural fluid just prior to delivery were performed in three cases. A case of bilateral hydrothorax diagnosed at 23th weeks regressed until delivery, whereas in another case complete resolution of unilateral hydrothorax was identified. Four cases of bilateral hydrothorax were delivered with cesarean section. All fetuses were delivered alive with no sign of pulmonary hypoplasia. At neonatal period two cases needed mechanic ventilation for three days due to respiratory distress. Mean gestational age at diagnosis and delivery and birth weight were 32 weeks (range: 23-38 weeks) and 36 weeks (range: 34-38 weeks) and 3010 g (range: 2880-3130 g) respectively. After breastfeeding, the serous pleural fluid became opalescent and diagnosis of congenital chylothorax were made in four cases.

Conclusions: Our findings suggest that isolated primary fetal hydrothorax diagnosed in late second trimester and third trimester can be managed without intervention and single thoracentesis prior to delivery facilitates neonatal resuscitations and prevent neonatal demise.

Keywords: primary fetal hydrothorax, thoracentesis, neonatal outcome

Özet

Primer Fetal Hidrotoraks: Doğum Öncesi Tek Torakosentez ile Başarılı Perinatal Sonuçlar

Amaç: Bu çalışmada, perinatoloji kliniğinde tanı konulan primer fetal hidrotoraks olguların gözden geçirilmesi, prenatal izlem ve perinatal sonuçların değerlendirilmesi amaçlanmıştır.

Materyal ve Metot: Perinatoloji kliniğinde tanı konulan primer fetal hidrotoraks olguların perinatal izlem, yönetimi ve sonuçları retrospektif gözden geçirildi. Primer fetal hidrotoraks tanısı detaylı bir ultrasonografik inceleme ve anomali taraması sonrasında konuldu. Tüm olgular müdahale edilmeden izlendi ve gerektiğinde doğumdan hemen önce tek torakosentez girişimi uygulandı.

Sonuçlar: Çalışma grubuna 22 "non-immune" hidrops fetalis olgusundan beş olgu dahil edildi. Dört olguya bilateral, bir olguya ise unilateral hidrotoraks tanısı konuldu. Detaylı ultrasonografik inceleme ve diğer tanısız araştırmalar ile tüm olgularda primer hidrotoraks dışında bir özellik tespit edilmedi. Üç olguya doğum öncesi tek torakosentez girişimi uygulandı ve 100-200 ml plevral sıvı boşaltıldı. Yirmi üç gebelik haftasında bilateral primer fetal hidrotoraks tanısı konulan bir olgu ve unilateral hidrotoraks tespit edilen diğer bir olguda kendiliğinden doğuma kadar plevral effüzyon geriledi ve girişime ihtiyaç duyulmadı. Dört bilateral hidrotoraks olgusu sezaryen ile doğurtuldu. Tüm olgular canlı doğurtuldu ve pulmoner hipoplazi bulgusu tespit edilmedi. Yenidoğan dönemde, iki olgu solunum güçlüğü nedeniyle üç gün mekanik ventilasyona ihtiyaç duydu. Tanı ve doğumdaki ortalama gebelik haftası ve ortalama doğum kiloları sırasıyla, 32 hafta (yayılm: 23-38) ve 36 hafta (yayılm: 34-38) ve 3010 g (yayılm: 2880-3130) olarak bulundu. Anne sütü ile beslenme sonrası toraks tüpünden gelen seröz sıvı koyu beyaz renge dönüştü ve dört olguya şilotoraks tanısı konuldu.

Tartışma: Primer fetal hidrotoraks ikinci ve üçüncü trimesterde prenatal dönemde müdahale edilmeden izlem ve gerekirse doğumdan hemen önce tek torasentez ile girişim yenidoğan canlandırılmaya yardımcı olmaktadır.

Anahtar sözcükler: primer fetal hidrotoraks, torakosentez, perinatal sonuçlar

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Introduction

Hydrothorax in fetal life are non-specific collections of fluid within the pleural cavity which can be identified by ultrasound. Primary fetal hydrothorax presents a wide spectrum of severity ranging from small, harmless effusions, to life-threatening thoracic compression. It may occur in isolation or in association with other thoracic and fetal anomalies. Possible causes include fistula or atresia of the thoracic duct (1), pulmonary sequestration (2), infections (3), fetal goitre (4), congenital pulmonary lymphangiectasis (5), familial (6) and aneuploidy, particularly trisomy 21 (7).

The natural history of primary fetal hydrothorax remains unclear. So far a few series have been published. The timing of diagnosis can vary from late first trimester to term. Polyhydramnios complicates approximately 42-55% of pregnancies with hydrothorax (8,9). Spontaneous resolution does occur and has been reported (8,9,10) in up to 10% of cases, with one series (9) reporting resolution in six of 11 cases (55%). In addition, resolution has been followed by recurrence (12). Early diagnosis is associated with a poor prognosis, and mortality rate, as high as 36-53%, has been reported (9,10).

Prenatal management of primary fetal hydrothorax is still controversial. Once all the diagnostic investigations are completed, management options include drainage with thoracentesis, thoraco-amniotic shunting or conservative approach. The role of intervention in isolated primary fetal hydrothorax is less clear and decision-making in management is still problem for physicians and parents. In this paper we reviewed the prenatal course and neonatal outcome of five fetuses with isolated fetal hydrothorax managed conservatively and three cases with single thoracentesis just prior to delivery.

Materials and Methods

Istanbul SSK Bakırköy Women and Children Hospital is a resident training and referral tertiary center and has approximately 22 000 delivery a year. Demographic and pregnancy details and ultrasonographic findings of all pregnancies admitted or referred to maternal and fetal unit have been prospectively recorded in medical charts and computer database between January 15, 2002 and August 31, 2003. At first admission, a careful ultrasound examination was performed to diagnose or confirm the fetal anomaly. Isolated or primary fetal hydrothorax was defined as marked unilateral or bilateral pleural effusion and/or subcutaneous edema >5 mm at occiput, and/or polyhydramnios >25 cm four quadrant amniotic fluid index (AFI), and/or placental thickness >6 cm but absence of ascites, pericardial effusion and no other associated fetal anomaly by ultrasonographic scan. Other diagnostic investigations included fetal echocardiography, maternal blood group, Rhesus status, indirect Coombs test, syphilis serology, intrauterine infection screening of toxoplasmosis, rubella, cytomegalovirus, and fetal blood sampling for assessment of the hematological status and karyotyping.

After diagnostic investigations and counselling with the pa-



Figure 1. Bilateral hydrothorax diagnosed as chylothorax at the neonatal period.

rents, serial sonography was done to monitor the cases without intervention until delivery unless indicated. When labour begun or attainment of 37 weeks' gestation, if indicated, single thoracentesis for drainage of hydrothorax just prior to delivery were performed to facilitate neonatal resuscitation. The following data were compiled: gestational age at the time of diagnosis and delivery, location of hydrothorax, fluid withdrawn and postnatal course including diagnostic studies, need for thoracic tube and respiratory support, and outcome.

Results

The study group included five cases of isolated fetal hydrothorax among 22 cases of nonimmune hydrops fetalis out of 2900 admission to maternal and fetal unit during the study period. Prenatal and neonatal findings of the cases are summarised in Table 1 and Table 2. Four cases were diagnosed as bilateral isolated fetal hydrothorax (Figure 1), whereas one of the five cases presented as unilateral hydrothorax. We identified two cases with polyhydramnios, AFI >25 cm, and three cases with increased subcutaneous edema >5 mm at occiput and placental thickness >6 cm. Ultrasonographic and echocardiographic scan, and postnatal examination revealed no anomaly other than hydrothorax in all fetuses. We performed cordocentesis for blood sampling in three cases at 23th, 32th and 32th weeks, all with normal karyotyping and hemogram. Other diagnostic investigations, indirect Coombs test, intrauterine infection screening were found as negative.

All cases were followed-up without intervention until delivery. We performed single thoracentesis to right pleural cavity in three cases for drainage of severe bilateral hydrothorax, resulting in expansion of lungs just prior to delivery. Bilateral hydrothorax of the case 4 diagnosed at 23th weeks regressed until delivery, whereas in the case 5 complete resolution of unilateral hydrothorax was identified. Four cases of bilateral hydrothorax were delivered with cesarean section. Thoracic tube drainage of hydrothorax were performed in four cases.

Table 1. Clinical findings and neonatal outcome of the cases with primary fetal hydrothorax

Case No	Hydrothorax	Amniotic Fluid Volume	Gestational age at diagnosis (wk)	Gestational age at delivery (wk)	Intervention at delivery	Birth Weight (g)	Neonatal outcome
1	Bilateral	P	35	37	200 ml thoracentesis	2900	Live birth
2	Bilateral	P	32	34	100 ml thoracentesis	3100	Live birth
3	Bilateral	N	38	38	150 ml thoracentesis	3130	Live birth
4	Bilateral	N	23	37	-	3040	Live birth
5	Unilateral	N	32	36	-	2880	Live birth

P:polyhydramnios; N:normal

Table 2. Laboratory findings of pleural fluid in the cases of fetal hydrothorax

Case No.	Neonatal intervention	Pleural fluid	Glucose (mg/dl)	Protein (g/dl)	LDH (IU/L)	Cell		Bacterial culture
						n/mm ³	Lymphocyte (%)	
1	Thoracic tube	Clear	75	2.8	195	1200	91	Sterile
2	Thoracic tube	Clear	77	4.5	143	4920	85	Sterile
3	Thoracic tube	Clear	80	3.5	210	1600	89	Sterile
4	Thoracic tube	Clear	88	3.9	91	2400	90	Sterile
5	No intervention	-	-	-	-	-	-	-

LDH: lactate dehydrogenase

All fetuses were delivered alive with no sign of pulmonary hypoplasia. At neonatal period two cases needed mechanic ventilation for three days. Mean gestational age at diagnosis and delivery and birth weight were 32 weeks (range: 23-38 weeks) and 36 weeks (range: 34-38 weeks) and 3010 g (range: 2880-3130 g) respectively. Biochemistry and cell count of pleural fluids were found as consistent with chylothorax (Table 2). After beginning breast feeding, the serous pleural fluid became opalescent and diagnosis of congenital chylothorax were made to four cases. Feeding was changed to medium-chain triglyceride feeding and the production of plural effusion disappeared gradually. All infants were discharged as healthy but hydrothorax of the case 3 recurred at postnatal three-month-old.

Discussion

It is difficult to draw firm conclusions from the small number of patients, because of the wide range of outcomes found for the sonographic variables described and different prenatal management. However the results of this study show that isolated fetal hydrothorax diagnosed in late second and third trimester, may regress or have complete resolution. Neonatal outcomes suggest that isolated fetal hydrothorax may be followed-up without intervention until delivery and may not cause pulmonary hypoplasia. This may be due to that hydrothorax in the study group developed after 32 weeks of gestation except case 4 in which regression of hydrothorax was observed at follow-up. As it is known, congenital intrathoracic lesions are of importance in that space occupying lesions present in the fetal chest may restrict normal lung growth with the possibility of resultant pulmonary hypoplasia, the most devastating consequence of fetal thoracic lesions. If ve-

nous cardiac return is embarrassed, hydrops fetalis may occur. Presence of hydrops, gestational age less than 35 weeks at delivery and bilateral effusions were reported as associated with poor prognosis (10). Possibilities of these severe complications are main reasons for prenatal interventions.

The drainage of pleural effusion with serial thoracentesis, thoraco-amniotic shunting or conservative approach are available management options. But there is no randomised study comparing the results of interventions and conservative management. Case series (9,10) regarding natural outcome of isolated fetal hydrothorax were reported as 36-53% of overall mortality rate. Successful (13) and unsuccessful (14) fetal outcome with thoracentesis remote from delivery were presented but it is well known that reaccumulation of pleural fluid in a few days is main problem (15). Long term drainage with thoracoamniotic shunting is another intervention, with which favorable neonatal outcomes were reported by several authors (15,16,17,18,19). But intervention with thoracoamniotic shunting has some procedure related complications such as unsuccessful shunting (14,19), dislocation and occlusion of the shunt (19,20), severe hypoproteinemia (20,21) and worsening hydrothorax (21), fetal and neonatal demise (19,21,22). Bernaschek et al (19), regarding the experience of fetio-amniotic shunting for different indications from four European centres, reported six percent of difficulties in insertion, 29% of dislocation or occlusion and mortality rate as high as eight percent.

Literature review point out that intervention with serial thoracentesis or thoracoamniotic shunt is not harmless and is associated with severe complications (19-22). Follow-up and delivery without intervention may cause neonatal death, be-

cause fetal hydrothorax, particularly bilateral effusions, restrict expansion of lungs at delivery. Thoracentesis just prior to cesarean delivery is minimal invasive procedure and may improve neonatal outcomes. In addition the findings of this study implicates that isolated fetal hydrothorax diagnosed in late second trimester or third trimester may not result in pulmonary hypoplasia.

In summary this report shows a tendency of complete resolution of isolated unilateral or regression of bilateral hydrothorax diagnosed in late second and third trimester, with good outcome. Single thoracentesis prior to cesarean section avoids complications of serial thoracentesis and thoracoamniotic shunt placement, and facilitate expansion of lungs, hence neonatal resuscitations and prevent neonatal demise.

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