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- Experimental Study
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- Letter to the Editor

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- Review Article
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Technical notes aims to present a newly diagnostic or therapeutic method. They should not exceed 2000 words (max. 8 pages) and include a maximum of 10 references.

Letters to the Editor should be no more than 500 words long (max. 2 pages) and include a maximum of 10 references.

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Manuscripts should be designed in the following order: title page, abstract, main text, references, and tables, with each typeset on a separate page:

Page 1 - Title page
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Next Page - References
Next Page - Table heading and tables (each table should be placed in separate pages)
Next Page - Figure legends and figures (each figure should be placed in separate pages)
Last Page - Appendices (patient forms, surveys etc.)

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This page should only include the title of the manuscript, which should be carefully chosen to better reflect the contents of the study. No anusual abbreviations should be used in the title of the manuscript. A short title as running heading not exceeding 40 characters should be given which is desired to appear on top part of continuing pages when journal is published.

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— Abstracts of ***case reports*** should be max. 125 words and structured in three paragraphs using the following subtitles: Objective, Case, Conclusion. Following the abstract, each abstract page should include max. 3 key words separated with comma and written in lower cases.

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The Methods section should contain a detailed description of the material, the study design and clinical and laboratory tests, and statistical methods used. A statement regarding the ethical issues should also be given in this section.

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The Discussion section should mainly rely on the results derived from the study, with relevant citations from the most recent literature.

The Conclusion section should briefly and claearly present the conclusions derived from the results of the study. It should be in compliance with the aim of the work and and point out its application in clinical practice.

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References used in the text should be directly related to the topic, as recent as possible and in enough numbers. They should be numbered in square brackets in the order in which they are mentioned in the text including Tables and Figures. Citation order should be checked carefully.

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Direct use of references is strongly recommended and the authors may be asked to provide the first and last pages of certain references. Publication of the manuscript will be suspended until this request is fulfilled by the author(s).

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— ***Book:*** Jones KL. Practical perinatology. New York: Springer; 1990. p. 112-9.

— ***Chapter in a book:*** Sibai BM, Frangieh AY. Eclampsia. In: Gleicher N, editors. Principles and practice of medical therapy in pregnancy. 3rd ed. New York: Appleton&Lange; 1998. p. 1022-7.

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Each table should be prepared on a separate page with table heading on top of the table. Table heading should be added to the main text file on a separate page when a table is submitted as a supplementary file.

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Multicentric Multiple Pregnancy Study III: Triplets

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Abstract

Objective: The aim of the study is to evaluate demographic and clinical characteristics of triplet pregnancies and to compare with those of twins.

Methods: The prevalence of triplets was found as 0.97/1000 and the ratio of them within multiple pregnancies was determined as 4.9%. At least one fetus died in the antenatal period or at birth at ratio of 32.43% in triplets. Perinatal mortality ratio was 234.2 per thousand in triplets. Perinatal mortality was high in cases born before 29th gestational week, and less than 1000 g.

Results: Demographic data related to the triplets delivered between the period of 2003 and 2004, including mean maternal age, parity, fetal and perinatal mortality, gestational week at delivery, mode of delivery and maternal morbidity, fetal or newborn's weight and sex were determined from the questionnaire forms and data obtained from 15 obstetrics centers. They were compared with those of twins. Chi-square, Fischer's exact and Student's t tests are used for statistical analyses.

Conclusion: Assisted reproduction techniques increased high order multiple pregnancies causing high fetal morbidity and mortality and requiring perinatal center care.

Keywords: Triplet, perinatal mortality, multicentric study.

Çok merkezli çoğul gebelik çalışması III - üçüz gebelikler

Amaç: Bu çalışmada ülkemizdeki çoğul gebelikler içinde saptanan üçüz gebeliklerin demografik ve klinik özellikleri saptanarak, ikizler ile karşılaştırıldı.

Yöntem: 2003-2004 yıllarındaki çoğul gebelikler içindeki üçüzlerde anne yaşı, gebelik ve doğum sayıları, doğum haftası ve şekli, yenidoğan ağırlıkları, cinsiyetler ile fetus ve yenidoğan mortalitesi ile anne morbiditesi gibi demografik veriler araştırıldı, ikizler ile karşılaştırıldı. Veriler onbeş farklı Kadın Hastalıkları ve Doğum Kliniğinin anket ve veri formları yardımı ile elde edildi. İstatistiksel yöntem olarak Chi-square, Fisher's exact ve Student t testleri kullanıldı.

Bulgular: Üçüzlerin prevalansı 0.97/1000 ve çoğul gebelikler içindeki oranı %4.9 olarak bulundu. Üçüzlerin %32.43'ünde gebelikte en az bir fetusun antenatal dönemde veya doğumda kaybedilmiş olduğu, perinatal mortalite oranının binde 234.2 olduğu belirlendi. Gebeliğin 29. haftasından önce veya 1000 g altında doğanlarda, perinatal mortalite daha yüksek oranda bulundu.

Sonuç: Yardımla üreme tekniklerinin kullanımı sonucunda üçüz gebelikler artmaktadır. Bu durum yüksek perinatal mortalite ve morbiditeye sahip olduğundan perinatal merkezlerde takibi gerektirmektedir.

Anahtar Sözcükler: Üçüz, perinatal mortalite, çok merkezli çalışma.

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Introduction

With the introduction of artificial reproduction techniques (ART), the ratio of pregnant women has increased within the last 20 years in our country. The success of centers using induction of ovulation, in vitro fertilization (IVF) or intracytoplasmic sperm injection (ICSI) is measured by the ratio of pregnancy, meanwhile, the real measure, the healthy fetus at term (favorably singleton) is sometimes ignored.

Because of the reduced pregnancy rates by the conservative approaches in the favor of Singleton pregnancies and the psychological stress brought by failed pregnancy after a long and expensive treatment processes and to prevent the decrease in the success of the Assisted Reproductive Technique (ART) centers, implementation of aggressive therapies which may result multiple pregnancies are preferred physicians as well as by the patients, though well-known complications. This is accompanied by a higher number of twin and multiple pregnancy rates. Triplet pregnancy rates, once one per ten thousand,[1] has increased up to seven per ten thousand in recent years.[2]

As known, multiple pregnancies are responsible for 10% of perinatal mortality.[3] In triplet pregnancy perinatal mortality rate is 9 times as much as those of Singleton pregnancies and twice as much in twins.[3] The main factor related to this ratio is the presence of premature birth.[4] The aim of this cross-sectional multi-center study was to reveal the basic epidemiological parameters and mortality ratios in the triplet pregnancies seen in various Obstetrics and Gynecology clinics in our country.

Methods

This study, covering the years 2003-2004, has been performed with the help of survey and clinical information forms sent to the 15 Gynecology and Obstetrics Clinics of University Hospitals and the Training and Research Hospitals in various regions of Turkey. Maternal age, number of pregnancies and births, the number of fetuses, gestational age, birth type, birth weight, gender, mortality and maternal mortality-morbidity parameters were investigated in the survey. Some unreported data in the survey were re-questioned and the missing ones were completed. The data of multiple pregnancy ones with mortality in the perinatal period was compared to those without mortality using statistical tests of chi-square, Fisher's exact and Student's t-test.

The term “stillbirth” has been defined for the death of the fetus, who (or any of his twins) is at least 400 grams of weight or who completed 20 th gestational week, before birth or no respiration after birth or no heart beat. The term “early neonatal death” has been defined for neonatal deaths within the first 7 days after birth. Only ovulation induction, ICSI and IVF pregnancies have been accepted as assisted reproductive techniques.

Results

Nine of the fifteen centers included in the study were University Hospitals and the rest were Training and Research Hospitals. Though all centers completed the first survey data forms, only ten centers completed both parts.

The total number of births in 15 centers participating in the study in 2003-2004 was 70,091; 1310 of them were twins and 53 of them were triplets. Accordingly, the prevalence of twin birth was 18.6/1000 and the prevalence of triplet birth was 0.75/1000. According to 10 centers who provided detailed results, the total number of births was 43.258; 818 of them were twins (18.9/1000) and 42 of them were triplets (0.97/1000). The ratio of triplet pregnancies in the multiple pregnancies was found to be 4.9%. Comparative demographic data of total of 829 multiple pregnancies (792 twins and 37 triplets), assessed after removing those with

Table 1. Emographic and clinical data in multiple pregnancies.

	Twins (n:792)	Triplets (n:37)	p
Age (Mean ±SD)	27.91±5.36	28.76±4.23	0.346
Number of pregnancies (Mean± SD	2.40±1.90	2.20±2.41	0.555
Number of biths (Mean± SD))	1.10±1.66	0.97±2.19	0.670
Number of biths (Mean± SD)	34.33±3.52	30.65±3.35	**0.000**
Fetal neonatal weight (grams) (Mean± SD)	2167±638	1443±526	**0.000**

missing, are shown in Table 1. The birth week and the neonatal weight of triplets were found to be statistically different than the ones of twins (p <0.000).

Perinatal mortality rate in twin pregnancies was 106.9 per thousand (136/1272) and perinatal mortality rate in triplets was 234.2 per thousand (26/111) and the difference was more than 2-fold (Table 2). At least one fetus loss was present in 14% of twin pregnancies and 32% of triplet pregnancies (Table 2). The chance of pregnancy without fetal loss was 85% in twins and 67% in triplets. In triplet pregnancies, death rate in male fetuses was 31% and death rate in female fetuses was 33%. The loss of all fetuses in the multiple pregnancies was found to be 7.58%.

With the assessment of multiple pregnancies for mortality, it was found that fetus(es) with lower weight died in 78% (35/45) of twins and in 75% (6/8) of triplets. In these pregnancies in which at least one fetus was alive, the rate of cesarean section was 64% in twins and 100% in triplets. The comparison of demographic and clinical data of triplet pregnancies with and without mortality is shown in Table 3.

When evaluated for premature birth, available data has shown that 44% of twins and 87% of triplets was born at 34th week or earlier. The probability of giving birth of triplet pregnancies at 24th, 28th and 34th weeks were found to be twice, thrice and twice higher than the one in twin pregnancies at respective weeks.

When the perinatal mortality in triplet pregnancies is evaluated acording to the parity, the mortality ratio was 333 (7/21) per thousand in primigravida and 286 (4/14) per thousand in multigravida. The difference was not statistically significant (p >0.05). However, double and triple losses were more frequently observed in primigravida, single losses were more in multigravida. When the mortality in triplet pregnancies is evaluated according to the gestational week, it is seen that loss of all fetuses occured before 29 weeks of gestation. Especially under the 29 weeks of gestation this high mortality rate was statistically significant (p<0.001) and the mortality rate decreased with ongoing gestational weeks (p <0.05). However, there was no change

Table 2. Perinatal mortality in multiple pregnancies (673 cases with data).

	Twins (n:636)	Triplets (n:37)	Total (n:673)
All deceased	45 (%7.07)	6 (%16.21)	51 (% 7.58)
One alive	46 (%7.23)	2 (% 5.40)	48 (% 7.13)
Two of them alive	545 (%85.69)	4 (%10.81)	549 (%81.57)
Three of them alive	-	25 (%67.56)	25 (%3.71)
At least one deceased	91 (%14.30)	12 (%32.43)	103 (%15.30)

Table 3. The comparison of demographic and clinical data of triplet pregnancies with and without mortality (n: 37) (Values are shown as Mean ± Standard deviation).

	One deceased (n:4)	Two deceased (n:2)	Three deceased (n:6)	Any deceased (n:12)	All three alive (n:25)	P
Age	28.50±4.79	30.50±3.19	28.67±3.00	28.92±4.32	28.68±4.27	>0.05
Gestation	1.75±0.50	1.00±0.00	1.20±0.44	1.36±0.50	2.58±2.83	<0.05
Parity	0.80±0.50		0.20±0.40	0.40±0.50	1.26±2.61	>0.05
Birth Week	30.75±3.30	31.50±3.50	25.67±2.07	28.33±3.70	31.76±2.55	**<0.001**
Weight	1417±494 g	1640±167 g	711±205 g	1101±513 g	1606±482 g	**<0.001**
Weight of live fetus	1426±457 g	1655±219 g	-	1472±421 g	1606±482 g	>0.05
Weight of dead fetus	1400±637 g	1633±274 g	711±205 g	959±478 g	-	-
Primigravida	_	2/2	5/6	8/12	15/25	>0.05
Cesarian Section	4/4 (%100)	2/2 (%100)	1/6 (%17)	7/12 (%58)	23/25 (%92)	**<0.001**

Table 4. The comparison of triplet pregnancies with and without mortality according to the gestational week (n: 37).

	One deceased (n:4)	Two deceased (n:2)	Three deceased (n:6)	Any deceased (n:12)	All three alive (n:25)	P
< 29 weeks	-	-	6	6	3	**<0.001**
29-32 week	2	1	-	3	9	>0.05
> 32 week	2	1	-	3	13	>0.05

in the probability of pregnancy without any loss (Table 4). Survival rate in triplet pregnancies is shown in the Table 5 and survival graph (Figure 1). When the perinatal mortality in triplet pregnancies is evaluated according to the birth weight, the mortality ratio was 76% (19/21) in babies born with a birth weight of 1000 grams or lesser and the ratio was 7.7% (7/90) in babies

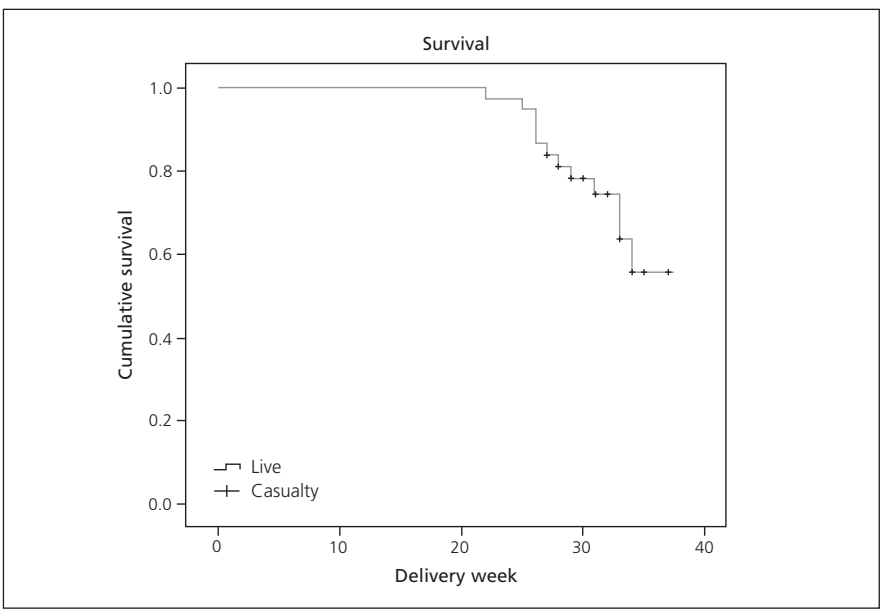

Figure 1. Survival graphic in triplets.

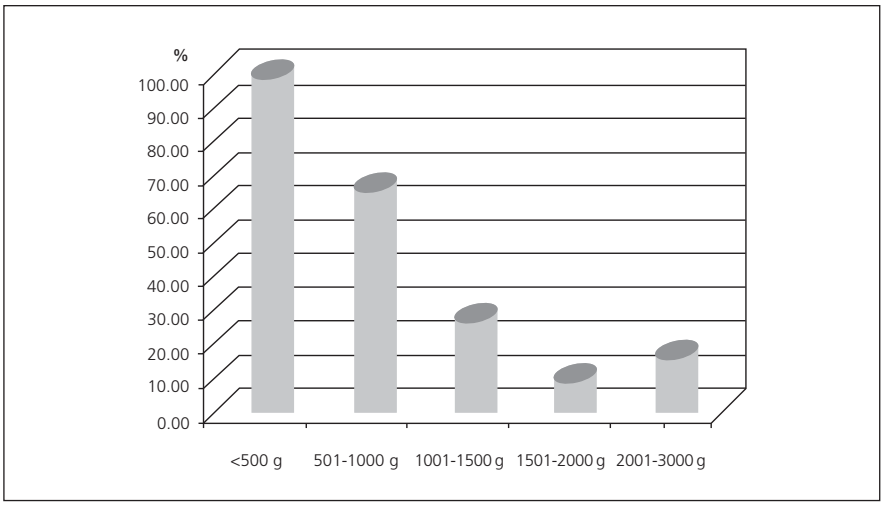

Figure 2. Mortality ratios in triplet pregnancies according to birth weight

Table 5. The ratio of living fetuses in triplet pregnancies according to the gestational week.

Gestational Week	Living Fetus (%)
20-24 Weeks	97
25-27 Weeks	84
28-30 Weeks	78
31-33 Weeks	70
34-36 Weeks	68
> 36 Weeks	68

born with a birth weight more than 1000 grams. The difference was statistically significant (p <0.001). However, there was no statistical difference between rates of pregnancy without loss (Table 6). Table 7 and Figure 2 shows mortality rates according to the weight of fetuses.

When the perinatal mortality in triplet pregnancies is evaluated according to the gender, it was seen that 24.19% of male fetuses (15/62)

Table 6. The comparison of triplet pregnancies with and without mortality according to the birth weight (n: 37).

	One deceased (n:4)	Two deceased (n:2)	Three deceased (n:6)	Any deceased (n:12)	All three alive (n:25)	P
≤ 1000 g	1/3	-	18/18	19/21	6/75	<0.001
1001-2000	2/7	4/6	-	6/13	53/75	>0.05
>2000	_	-	-	_	16/75	>0.05

Table 7. The relation of fetal-neonatal mortality with birth weight.

	Fetal-neonatal mortality (+)	Fetal-neonatal mortality (-)	Total	Mortality %
< 500 g	1	-	1	100
501-1000 g	6	3	9	66,7
1001-1500 g	3	8	11	27,3
1501-2000 g	1	9	10	10,0
2001-3000 g	1	5	6	16,7
> 3000	-	-	-	-
Total	12	25	37	

and 22.44% (11/49) of female fetuses died. But, the difference was not statistically significant (p >0.05). When the maternal morbidity in triplet pregnancies was assessed, though data is inadequate, available data showed that some treatment was managed for premature birth and the caesarian section ratio was 93% in case of living fetuses.

Discussion

The main reasons of high perinatal mortality rate in multiple pregnancies are the premature births and complications related to it such as respiratory distress syndrome, necrotizing enterocolitis, and intraventricular hemorrhage. Fetal malformations, intrauterine inappropriate and complications related to the placenta are the other reasons.[4]

In our country, antenatal and neonatal care services could not keep pace with the advances in the reproductive technologies.[5] Therefore, specialist and minor associations and the Ministry of Health, as well, involved in a quest. Indeed, their results were seen in recent years, and the number of embryos transferred in IVF programs was limited with a regulation.[6]

Concerning the triplet pregnancy outcomes in some centers in our country, our study found the prevalence of triplets as 9.7 per ten thousand. Probability of premature birth was 87%, perinatal mortality was 234.2 per thousand and the ratio of pregnant women losing any fetus in the antenatal period or birth was 32% and the ratio of pregnant women losing all fetuses was 16.21%. Clinical studies reported from Turkey, the ratio triplets in multiple pregnancies were reported within the range of 3-26% and their mortality rate was reported % in the range of 3.5-37%.[7-11] Our ratio of triplets approximating 4% is in accordance with the ratios of other studies. In our series, 32% loss rate is suggested to be related to the inclusion of ones with small gestational ages.

Ulug et al reported the mean birth week as 33.4 weeks, mean birth weight as 1824 grams and the mortality rate as 5% in the series of 55 viable triplet pregnancies.[12] The results of our study are different as our study also covered the results of those previable. The neonatal mortality in the triplets in our study is highest in the births before 29 weeks of gestation at birth is highest. The mean birth week in the triplet pregnancies with mortality (28.33±3.70), was approximately 3.4 weeks earlier than the ones without mortality (31.76±2.55). When the losses in the triplets were examined, the survival chance of births before 29 weeks was the lowest. The ratio of one loss was 10.81%, the ratio of two losses was 5:40% and the ratio of three losses was 16.21%. Overall perinatal mortality was 324 per thousand in triplets. This ratio was found as 370 per thousand in the study of Yayla et al.[11] When compared with the singleton pregnancies, birth weights of twins are 25% lesser, birth weights of triplets are 50% lesser and the birth weights of quadruplets or more multiple pregnancies are 70% lesser.[1] In multiple pregnancies, the fetus died, was the one with lower weight in about three quarters. The living fetus with a lost twin was 770 grams heavier than the lost one but was 325 grams lighter than those both born alive. This may be due to born of normal fetus immediately after death of twin or continuing growth of the other fetus after the death of twin. Similar to twins, in triplets the loss of fetuses was more in fetuses with a weight lower than 1000 grams.

The discordant development between twins has been suggested (discordance) as an important factor affecting mortality in twins and poorer prognosis was reported in the discordant twins even in the absence of twin-twin transfusion syndrome or congenital anomaly.[13] A similar situation may also apply to the triplets.

In our study, mortality in those born before 25 weeks or 500 grams of birth weight was 100%, while the pregnancy progresses, mortali-

ty has dropped to 10%. 29 weeks gestational week and weight limit of 1000 grams are comforting parameters in the follow up.

Although the limited data to make comparison in terms of maternal morbidity, there was a need of treatment for premature birth in one of two triplet pregnancies and cesarean section rate was over 90%.

Conclusion

In our study, the rate of multiple pregnancies with more than two fetuses was approximating one per thousand; this is 6-10 times higher than expected. Though perinatal mortality in triplet pregnancies was associated with gestational week and birth weight, chance for all fetuses to live showed no difference according to the gestational week and birth weight. This suggests that triplet pregnancies are prone to increase perinatal mortality. Therefore, prevention of multiple pregnancies with more than two fetuses may help reducing perinatal mortality.

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Assessment of Maternal and Perinatal Outcomes in Pregnancies Complicated by Epilepsy

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Abstract

Objective: The study was undertaken to assess the maternal outcomes of pregnancies complicated by epilepsy that is a common neurologic disorder of pregnancy, and to compare the perinatal outcomes of these pregnancies with healty controls.

Methods: Sixty-five pregnant women with epilepsy disorder and 69 healty controls who delivered at our clinic between April 2005 and March 2009 were included in this study. Age, number of previous pregnancies, administered anti epileptic treatments, disease duration and frequency of the seizures were examined in epileptic women. Birth weight and week, Apgar score at 5 minute, intensive care unit admission, perinatal complications, congenital anomalies and perinatal mortality were compared between two groups.

Results: Sixty-five pregnant women with epilepsy disorder and 69 healty controls were included in this study. Age, number of previous pregnancies, administered anti epileptic treatments, disease duration and frequency of the seizures were examined in epileptic women. Birth weight and week, Apgar score at 5 minute, intensive care unit admission, perinatal complications, congenital anomalies and perinatal mortality were compared between two groups.

Conclusion: Possible complications of pregnancies should be explained to epileptic women before conception, and perinatal outcomes should be improved with appropriate approach and follow-up strategies during pregnancy.

Keywords: Epilepsy, pregnancy, maternal and perinatal outcomes.

Epileptik gebeliklerde maternal ve perinatal sonuçların değerlendirilmesi

Amaç: Bu çalışmada, gebelikte sık rastlanan nörolojik bozukluklardan biri olan epilepsili gebelik olgularının maternal sonuçlarını değerlendirmek ve bu gebeliklerin perinatal sonuçlarını sağlıklı gebelerle karşılaştırmak amaçlandı.

Yöntem: Çalışmaya Nisan 2005-Mart 2009 yılları arasında hastanemizde doğum yapmış ve epilepsi hastalığı bulunan 65 gebe ile herhangi bir hastalığı olmayan 69 sağlıklı gebe alındı. Epileptik gebelerin yaşları, gebeliklerinin sayısı, alınan medikal tedaviler, hastalığın süresi ve gebelik boyunca geçirilen atakların sıklığı belirlendi. Perinatal sonuçlar olarak doğum ağırlıkları, doğum haftası, 5. dakikadaki APGAR skorları, yoğun bakımda ihtiyacı, perinatal komplikasyonlar, konjenital anomaliler ve perinatal mortalite oranları belirlendi ve bu sonuçlar iki grup arasında karşılaştırıldı.

Bulgular: Epileptik gebelerin yaş ortalaması 26.6+4.7/yıl, hastalık süresi 8.11+4.5/yıl olarak saptandı. Hastalardan 38'inin gebelikte atak geçirirken, en sık atak 1. trimesterde (%47.3) kaydedildi. Hastaların 53'ü (%81.5) gebelikte ilaç kullanırken, en sık kullanılan ilaç karbamazepin olarak tespit edildi. Doğum haftası ve ağırlığı, operatif doğumların oranı, yoğun bakım ihtiyacı, perinatal komplikasyon ve konjenital anomali oranı anlamlı olarak epileptik hastalarda yüksek bulundu ($p<0.05$).

Sonuç: Epileptik hastalar doğum öncesi dönemde gebelikte gözlenebilecek olası komplikasyonlar hakkında bilgilendirilmeli ve gebelik boyunca sağlanacak uygun yaklaşım ve takiplerle perinatal sonuçlar iyileştirilmelidir.

Anahtar Sözcükler: Epilepsi, gebelik, maternal ve perinatal sonuç.

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Introduction

Epileptic disorders are one of the major neurological complications affecting nearly 1% of society and observed most frequently in pregnancy after migraine. It was shown that approximately 0.3-0.5% of pregnancies have epilepsy.[1] Today, by means of developments in the diagnosis and treatment of epilepsy, many women with epilepsy have been able to maintain a normal life and get pregnant. Possible negative effects of antiepileptics on fetus used for epileptic pregnancies constitute a significant risk group associated with the increase of convulsion frequencies and malformations that may appear in their children. There are publications showing the increase in the frequency of epileptic attacks in pregnancy and teratogenic effects of anticonvulsants.[2] Also it is known that convulsions appeared during pregnancy pose threat in terms of mother and fetus.[3]

In many studies, it was shown that there was an increase in the risks of miscarriage, stillbirth, preterm delivery, lower birth weight, intrauterine growth retardation and low mental and motor retardation in newborns during epileptic pregnancies in the long period.[4-6] Besides, rates of maternal complications such as hypertensive diseases, antepartum hemorrhage, delivery by cesarean, intervened delivery were found as increased among epileptic pregnancies.[7] Therefore, epileptic pregnancies are the cases where convenient approaches and teamwork should be provided to obtain positive outcomes in terms of mother and fetus. The first condition to obtain positive outcomes in terms of pregnancy in epileptic women is to do pregnancy by planning. This enables to arrange antiepileptic drugs (AED), and to take required precautions by informing family in terms of fetal malformations and especially neural tube defects.

Our aim in this study is to evaluate maternal and fetal outcomes of epileptic pregnancies observed in our clinic which is a reference center and to compare their perinatal outcomes with healthy pregnants.

Method

65 pregnants who were diagnosed as epilepsy before or during their pregnancies as being followed-up in the Department of Obstetrics and Gynecology Department of Meram Medicine Faculty in between April 2005-March 2009 were included into the study. Also 69 healthy pregnants who were similar for age, gravida and parity but do not have epilepsy or any systemic disease were taken as control group to evaluate perinatal outcomes. Control group members were determined randomly among pregnants who delivered in our clinic during the period mentioned above. Those whose pregnancies were ended due to severe epileptic attacks or problems at early gestational weeks and those with suspected epileptic attack history were excluded from the study. Demographic characteristics of epileptic pregnants such as age, gravida, parity, abortus count as well as the duration of the disease, drugs used, whether they had epileptic attack or not during pregnancy and the rate of attacks as to trimesters were evaluated.

Neurological examination and obstetric ultrasonography were performed on each pregnant who had cardiac disease before delivery. Neurology Clinic decided the delivery type to be preferred according to disease rate of epileptic pregnants. Total hospitalization duration of these pregnants, delivery type and the existence of any accompanying medical disease were determined. Delivery type as perinatal outcomes, gestational weeks at delivery, birth weights, existence of any perinatal complication accompanying pregnancy such as oligohydramnios, intrauterine growth retardation and preeclampsia, antepartum neonatal death, fetal malformations, intense care requirement of babies and 5th minute Apgar score being below 5 were evaluated. Preterm delivery was defined as deliveries below 36th week and growth retardation was defined as deliveries below 10% according to delivery week. All these parameters were compared between epileptic pregnants and healthy pregnants.

Statistical analyses were done by evaluating in SPSS database. In the comparison of perinatal outcomes, chi-square test and Student T-test were used. P value being <0.05 was accepted as statistically significant.

Results

Maternal Results

Maternal results of the epileptic patients and the control group were shown in Table 1. No difference was observed between two groups in terms of mean age, gravida, parity and abortus (Table 1). On the other hand, hospitalization duration was found significantly high in epileptic group. Mean age of patients in epileptic group was found 26.6 + 4.77/year while mean disease period was found as 8.11 + 4.5/year. It was observed that 38 patients (58.4%) had epileptic attacks during pregnancy and that 47.3% of them were at first trimester. The gestational period with the least attack was third trimester. 53 of these patients (81.5%) used any AED during pregnancy and the most frequently used drugs were carbamazepine (41.4%) and sodium valproate (34%). The distribution of AEDs used during pregnancy was shown in Table 2. It was observed that 43 (66.2%) of epileptic pregnants were delivered by cesarean and 22 (33.8%) of them were delivered vaginally. 32.3% of patients had other medical diseases accompanying to epilepsy and the most frequently seen disease was diabetes mellitus. No maternal mortality was seen in any pregnant.

Perinatal Results

Results of epileptic and healthy pregnants were shown in Table 3. Delivery week and birth weights in epileptic pregnants (36.8 ± 2.7/week and 2758 ± 664/g) were lower than those in healthy pregnants (37.7 ± 1.4 week and 3122 ± 461 g) and the difference was statistically significant (p<0.036 and 0.01). While the rate of delivery by cesarean was 66.2% in epileptic pregnants, it was 40.6% in health pregnants and the difference was statistically significant (p<0.003).

Table 1. Maternal characteristics of pregnancies with epilepsy and control group.

Maternal characteristics	Epileptic group (n=65)	Control group (n=69)	P value
Age* (year)	26.6±4.7	28.2±5.2	>0.05
Gravida*	2.4±1.31	2.7±1.23	>0.05
Parity*	1.2±1.09	1.4±1.17	>0.05
Abortus*	0.32±0.77	0.43±0.82	>0.05
Disease period (year)*	8.11±4.5	-	-
Week of attacks at pregnancy (n=38)		-	-
1st Trimester	18/38 (47.3%)	-	-
2nd Trimester	8/38 (21%)	-	-
3rd Trimester	12/38 (31.7%)	-	-
Medical treatment at pregnancy		-	-
Received	53 (81.5%)	-	-
Not received	12 (18.5%)	-	-
Delivery type		-	-
Cesarean	43 (66.2%)	28 (40.6%)	<0.05**
Vaginal	22 (33.8%)	41 (59.4%)	<0.05**
Hospitalization period (day)*	3.2±2.3	1.6±1.8	<0.05**
Accompanying medical disease	3.2±2.3	1.6±1.8	<0.05**
Exists	21 (32.3%)	-	
Does not exist	44 (67.7%)	-	
Maternal mortality	0	0	

:** *Mean ± SD (standard deviation);* *:** *Statistically significant*

Table 2. The distribution of AED used during pregnancy.

Drug	N=53	%
Carbamazepine	22	41.4
Sodium valproate	18	34
Oxycarbazepine	4	7.5
Lamotrigine	2	3.8
Levetiracetam	1	1.8
Carbamazepine + Sod. valproate	2	3.8
Carbamazepine + Levetiracetam	3	5.7
Oxycarbazepine + Sod. valproate	1	1.8

Table 3. Perinatal outcomes of epileptic and healthy pregnants.

Perinatal outcomes	Epileptic (n=65)	Healthy (n=69)	P value
Delivery week*	36.8±2.7	37.7±1.4	0.036**
Birth weight	2758±664	3122±461	0.01**
Operative birth	43 (66.2%)	28 (40.6%)	0.003**
Perinatal mortality	4 (6.1%)	2 (2.8%)	0.062
Apgar 5th minute <5	13 (20%)	8 (11.6%)	0.18
Being taken to intense care	18 (27.7%)	9 (13%)	0.035**
Perinatal complication	22 (33.8%)	11 (15.4%)	0.043**
Congenital malformation	9 (13.8%)	3 (4.6%)	0.021**

:** *Mean ± SD (standard deviation);* *:** *Statistically significant*

Perinatal mortality was found in 4 pregnants within epileptic group and in 2 pregnants within healthy group. Perinatal mortality in epileptic group was caused by hydrocephaly in 1 case and by respiratory distress syndrome and accompanying anomalies due to prematurity in 3 cases. While the rate of those with 5th minute Apgar score <5 was 20% in epileptic group, it was found as 11.6% in healthy group. On the other hand, the difference was not statistically significant in terms of perinatal mortality and 5th minute Apgar score ($p>0.05$). The rate of pregnants who need intense care and found perinatal complication was again significantly higher than the epileptic group (27.7% vs 13%). It can be considered that the requirement of intense care by babies of healthy pregnants can be higher than normal society. However, this may be caused that pregnancies are chosen randomly though pregnants themselves are healthy. These complications in epileptic group were recorded as oligohydramnios in 7 patients, polyhydramnios in 5 patients and preeclampsia in 2 patients. Congenital malformation was found in 9 (13.8%) newborns within epileptic group and in 3 newborns (4%) within control group ($p<0.021$). These anomalies were detected as encephalocoele in 2 cases, hydrocephaly in 3 cases, ventriculomegaly in 2 cases, spina bifida in 1 case and phocomelia in 1 case within the first group. One cardiac anomaly, 2 hydrocephaly and meningocele were observed in other cases.

Discussion

Gestational periods of women with epileptic disorder are mostly non-problematic. Yet, some complications may be seen higher in epileptic pregnants than society.[8] It is known that the rates of fetal loss, congenital malformation and psychomotor growth disorder in epileptic pregnancies are higher compared to general population.[4,5,9] There are publications showing that extremity anomalies (distal phalanx and nail hypoplasia), craniofascial anomalies (lip-palate clefts), congenital cardiac diseases and NTD prevalence associated with valproic acid and carbamazepine increase.[10] It was shown in the studies of Richmond et al. that major congenital malformations (heart, orofacial defects, neural tube defects, intestinal atresia and urogenital anomalies) were approximately two times higher and minor anomalies were three times higher.[1]

AEDs, epileptic attacks and maternal genes that can cause epilepsy are considered as the factors which may cause congenital malformations. It is antiepileptic drug use among the factors mentioned which is clearly showed as having the most close relation with malformations.4 In a multi-centric study, 9.9% malformation rate was reported in living newborns. This rate was 11.5% in the group who did not have AED and 2.3% in the group who had the drug and the malfunction rate was reported as 5 times higher.[11] Again in the study of Katz et al. malformation rates with 2 times increased AED use were reported.[12] The classic AEDs used (carbamazepine, pheno-barbital, phenytoin, primadon, valproate) are in the D category in terms of their effects on fetus and they are the drugs which can be used during pregnancy since their benefits accepted as teratogenetically effective were more than their harms.[13] Nakane et al. showed that there was an increase in malformation rates as the number of used antiepileptic drugs increased. While the rate was less than 5% by monotherapy, malformation rate was shown as higher than 20% when drug number reaches four.11 In a study performed in our country found that anomaly rate in epileptic pregnancies were 5.09 times higher than the healthy group.[14] Congenital malformation risk proportionally increases by the number of antiepileptic drug used (polytherapy and monotherapy) and the increase in the dose.[15] In our epileptic patient group, we found 13.8% fetal malformation (2 encephalocele, 3 hydrocephaly, 2 ventriculomegaly, 1 spina bifida, 1 phocomely). A similar result was presented in the studies of Koch et al. and they reported that there was no difference in the malformation rate of mothers using single or multiple drugs.[16] There are publi-

cations reporting 3-5 times higher fetal loss rates associated with miscarriage and preterm delivery. In our case group, fetal loss rate was 6.1% (4/65) and it was observed that they were due to fetal malformation and prematurity. However, Hiilesmaa et al. showed in their studies that there was no difference between groups in terms of perinatal death.[17] Yerby et al. reported that there was increase in preeclampsia prevalence together with 2.79 times of increase in the rate of babies with low birth weight in epileptic women.[8] Similarly, Hvas et al. found in the study that birth weights of babies delivered by epileptic mothers were 208 gram lesser than those in the control group.[18] We found mean birth weight as 2758 ± 664 gram in epileptic pregnants. Preeclampsia developed in 2 epileptic patients and one of them also had hellp syndrome. None of the patients in control group had preeclampsia or hellp syndrome. While Hiilesmaa and Viinkainen found no evident difference in the gestational complications in epileptic women, preeclampsia and preterm delivery and perinatal death prevalence, Viinkainen et al. found SGA baby rate as considerably increased.[17,19] In our study, preeclampsia was found in the epileptic group while it was not found in the control group. These results support that preeclampsia risk is observed more in epileptic pregnants.

Although vaginal delivery is suggested to pregnants who have epileptic disorder, the uncertainty of complications which may develop in the management of delivery and attacks which may arise due to stress and sleeplessness associated with delivery increase cesarean rates in this group. The delivery is especially a risky period in terms of the formation of epileptic attack and there are publications showing that the possibility of having an attack is increased approximately 9 times.[20] In our follow-ups, we found that the rate of delivery by cesarean in epileptic group was high as 66.2% (43/65). This rate was significantly higher than cesarean rate (40.6%) in control group. Similarly, Katz et al. showed in their study that epilepsy is an independent risk factor for delivery by cesarean.12 However, Hiilesma et al. did not found increase in the operative delivery rates in this group.[17]

Today, by founding new anticonvulsants and the traceability of anticonvulsant levels, attack prevalence may stay same or decrease by patient compliance and close follow-up in the delivery period of many epileptic patients. Schmidt et al. indicated in their study that attack prevalence reduced or did not change in 63% of patients, and that attacks increased only in 37% of them.[21] It was observed that 58.3% of 65 epileptic pregnants consisting of our study group had epileptic attacks during their follow-up and these attacks were frequently at first trimester. One of the factors effective on attack prevalence is hormonal change. Increases and changes in estrogen and progesterone hormones affect the formation and prevalence of epileptic attacks.[22] One of the most important factors increasing epileptic attacks is the discontinuance of treatment by pregnants especially within first three months to minimize the risk of AED exposure by fetus. Similarly, in a study reported in our country indicated that epileptic attacks mostly observed in the first trimester.[23] The pregnancy of an epileptic woman also should certainly be planned. Another benefit of a preconceptional advisor is the enablement of folic acid prophylaxis. It is reported that the use of 4 mg preconceptional folic acid decreases the formation of neural tube defect at a rate of 50%.24

Conclusion

The aim in follow-up of epileptic pregnants is to give folic acid prophylaxis by pre-gestational consultation, and the good control of epileptic attacks by monotherapy and the possible lowest drug dose. Also these patients should be informed before pregnancy about possible perinatal complications which may occur during these pregnancies. In order to improve perinatal results of these pregnancies, proper approaches and gestational follow-ups should be provided.

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A Case of Parapagus Dicephalus Conjoined Twins Diagnosed at 17th Weeks of Gestation

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Abstract

Objective: Conjoined twins are a rarely seen anomaly with an incidence of 150.000 1100.000 gestations and together with severe mortality and morbidity. Early diagnosis and treatment seems to be very important in the followup.

Case: An 32 yearsold, nulliparous woman whom personal and family background were uneventful was referred to our clinic with a diagnosis of conjoined twins at 17th weeks of gestation. Ultrasonographic examination revealed a singleton fetus with mesurements consistent with a gestational age of 17 weeks and the fetus with 2 heads with seperate necks, 2 arms, 2 legs, only one medulla spinalis and vertebral column.

Conclusion: Conjoined twins are a rare form of twinning. Early diagnosis and management of the pregnancy regarding to social, ethical and economic problems seems to be very important.

Keywords: Conjoined twins, prenatal ultrasound.

Prenatal tanısı 17. Gebelik haftasında konan parapagus disefalus yapışık ikiz: olgu sunumu

Amaç: Yapışık ikiz, insidansı 150.000–1100.000 olan, ağır mortalite ve morbidite ile seyreden nadir bir anomalidir. Erken tanı ve tedavi gebelik takibinde önemlidir.

Olgu: 32 yaşında, nullipar, özgeçmişi ve soy geçmişinde özellik bulunmayan hasta yapışık ikiz ön tanısı ile, 17. gebelik haftasında başka bir merkezden hastanemize refere edildi. Yapılan USG incelemesinde intrauterin 17 hafta ile uyumlu, iki başı, tek gövdesi, iki kol ve iki bacağı, tek medulla spinalis ve vertebral kolonu bulunan fetüs izlendi.

Sonuç: Yapışık ikiz, ikiz gebeliğin nadir görülen bir formudur. Erken gebelik döneminde tanı konulması ve gebeliğin mevcut duruma göre yönetilmesi, sosyal, etik ve ekonomik sorunlar düşünüldüğünde oldukça önemli görünmektedir.

Anahtar Sözcükler: Yapışık ikiz, prenatal ultrasonografi.

Introduction

Conjoined twin is a quite rare congenital anomaly which can be seen in 1/50.000 – 1/100.000 pregnancies.[1] Considering that 60% of these fetuses die in a short time after delivery or born dead, real incidence of live birth is calculated as 1/200.000.[2] Though the exact etiology of conjoined twins are not know, two theories are suggested: According to the first theory, monovular embryo is divided incompletely at 13th-15th days of conception. In fusion theory, a secondary fusion occurs between two monovular embryonic dics.[3] It has been argued in the literature that fusion theory can explain all conjoined twin cases; by spherical theory, it has been explained that conjoined twins are

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attached to each other asymmetrically or through different body parts.[4,5]

Classification of conjoined twins was widely accepted by the classification suggested by Spencer; according to this, ventral or dorsal fusion may occur as to fusion point of embryonic disc.4 Body part numbers are expressed as di- (two), tri- (three), tetra- (four) and body parts expressed by Latin words [for instance, brachius (arm), -pus (lower extremity), prospus (face) etc.]. When different fusions are compared, conjoined twins with central fusion are seen more frequently since somite development begins from the central and proceeds towards caudal and cranial.[6] Conjoined twins with ventral fusion cover 87% of all cases; 11% of them are cephalopagus cases, 19% of them are thoracopagus cases, 18% of them are omphalopagus cases, 11% of them are ischiopagus cases and 28% of them are parapagus cases.[7]

In the article that we present, the significance of early diagnosis and treatment for dicephalic parapagus case diagnosed at 17th gestational week will be emphasized.

Case

Thirty-two years old woman with gravida 1 who had eventless personal and family backgrounds without kin marriage was referred to our clinic by a pre-diagnosis of conjoined twins through external center at her 17th gestational week. Obstetric ultrasonographic examination (USG) performed by Voluson 730 PRO (General Electric, Healthcare, Milwaukee, WI) revealed a singleton fetus with measurements consistent with intrauterine gestational age of 17 weeks having 2 heads and 2 necks on one body, 2 arms and 2 legs, single or conjoined two medulla spinalis and vertebral column (Figure 1). It was observed in the transverse and longitudinal cross-sections that fetus had one heart, one stomach, one bladder, one placenta and umbilical cord.

The patient and her husband were informed in detail about the current status of pregnancy and that their babies would have a low chance to live when surgical separation process is performed after delivery. The case was discussed in the council of Perinatology Clinic of our hospital; and gestational termination was conclud-

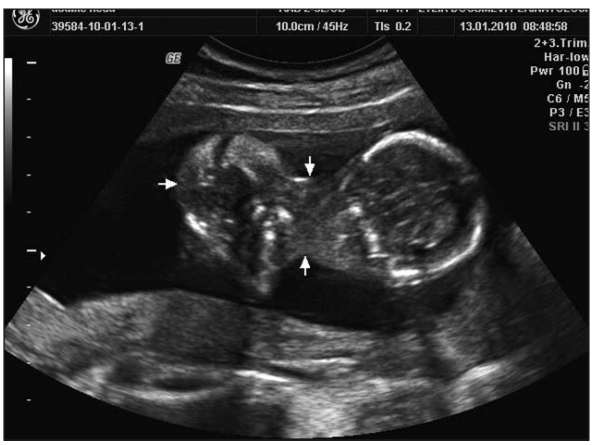

Figure 1. Ultrasonographic view of conjoined twin case: the view of two heads and two necks of a case consistent with intrauterine gestational age of 17 weeks having 2 heads and 2 necks on one body, 2 arms and 2 legs, single or conjoined two medulla spinalis and vertebral column.

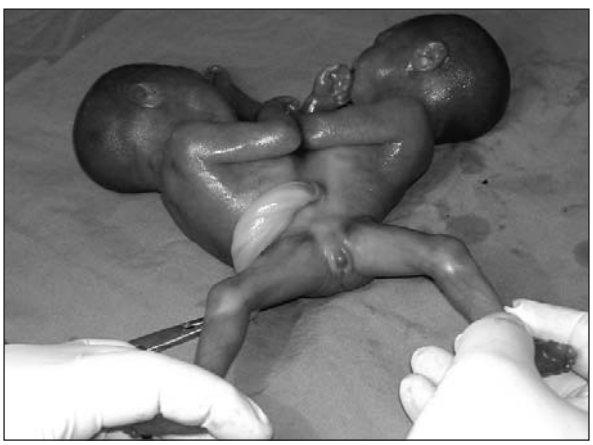

Figure 2. The postnatal view of terminated conjoined twin; two heads, two necks, two arms and two legs of the fetus.

ed after written consent of the patient and her husband was taken. Karyotype analysis was not planned as it is known that heredity is not seen in conjoined twins together with karyotype anomaly. In the macroscopic examination of fetus after termination, it was seen that fetus had two heads, two necks, two arms and two legs (Figure 2). One anus and one external genitalia existed on one body. Single or conjoined two vertebral colon(s) and one sacrum were observed in the x-ray film (Figure 3).

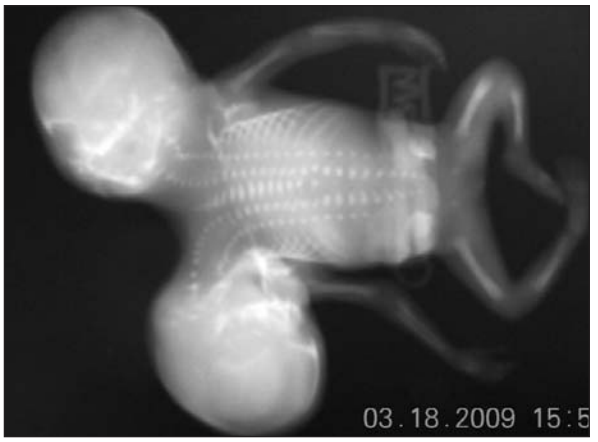

Figure 3. X-ray view of conjoined twin case: Single or conjoined two vertebral colon(s) and one sacrum.

Discussion

Parapagus conjoined twin cases developed due to parallel duplication of two notochords at proximal level are a kind of ventrolateral conjoin twins; these fetuses share umbilicus, abdomen and pelvis. The fusion includes pelvis which has single or double sacrum(s) and single symphysis pubis; parapagus twin case with sacral agenesis was also reported in the literature.[8] When thoraxes of twins are separate, they are called as dithoracic conjoined twins and when they have two heads on single body as the case that we presented, they are called as dicephalic conjoined twins. Their arm and leg numbers may vary between 2 and 4. Although all visceral organs are rarely double in parapagus cases, there are cases reported in the literature.[3] In the case that we presented, there were two arms, two legs on a single body, the case was called as parapagus dicephalous dibrachius dipus; also visceral organs were displayed separately. The prognosis in conjoined twin cases depends on the scale of fusion and the decision for maintaining pregnancy should be taken into the cases which do not have other organ anomalies and on which surgical separation operations can be performed.[9] In parapagus cases with single heart as in the presented case, this situation is a factor negatively affecting prognosis. Although each fusion type possibly may have cardiac anomaly, thoracopagus cases are at the top in terms of cardiac anomaly rate. Dibrachius dipus cases generally have one heart; however, the heart is duplicated or conjoined in most of the cases.[10] Intracardiac pathology existence and cardiac fusion rates in conjoined twins are important due to the success for surgical procedures and their effects on long-term survival. Due to the existence of distorted heart in one of two thoracopagus cases presented in the study of Şen et al. in 2003, the case was terminated at 19th gestational week; the second thoracopagus case was diagnosed at 25th week and the pregnancy was followed up until 38th gestational week since conjoined liver with separate vascular structure and conjoined non-pericardium heart on the sides of ventricles facing each other were found and there was no accompanying anomaly, then the surgical separation operation was successfully performed at 10th month after delivery by cesarean.[9] In another study, stillborn parapagus dicephalous conjoined twin case was presented which admitted to clinic at 38th gestational week without any follow-up and was delivered vaginally with difficulty.[11] This case is significant in terms of showing the significance of prenatal diagnosis and antenatal follow-up in conjoined twin cases. In the case that we present, visceral pathologies (cardiac fusion) were not confirmed by autopsy since the family did not

approve the autopsy though they were informed; however, no intracardiac pathology was seen in the prenatal echocardiography.

Prenatal diagnosis of conjoined twins is important when considered together with high mortality rate and postnatal ethic, social and economical problems. Conjoined twins can be diagnosed by transvaginal or transabdominal USG as from first trimester. If single yolk sac is followed up together with two fetuses at first trimester, or if there is the case of monaoamniotic twin, conjoined twins should be considered.[3] After diagnosis, fusion existence should be checked in brain, liver, heart, extremities and spinal cord. Also revealing vascularization in conjoined vital organs is determinant in terms of the prognosis of surgical separation operations as well as revealing additional accompanying fetal anomalies. Especially three-dimensional USG can present complex fetal anatomy and case type in conjoined twin cases and may provide selective termination in this way.[7]

Conclusion

Fetal anomalies can be diagnosed at early period by ultrasonography and prenatal invasive initiatives and pregnancies with fetal anomaly can be terminated. Conjoined twin is a rare anomaly and may progress with high mortality and morbidity according to the level of organ share. Diagnosing this situation at early prenatal period would have a significant role in advanced periods of pregnancy, informing family and terminating pregnancy when required.

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Prenatal Diagnosis and Management of a Case With Type-1 Congenital Cystic Adenomatoid Malformation

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Abstract

Objective: To present a prenatally diagnosed case of type I congenital cystic adenomatoid malformation.

Case: A 13.9 X 10.8 mm unilocular and a 16.7 X 14.9 mm multilocular anechoic cystic structures at the left lung field with a marked mediastinal shift to the right were observed on ultrasonography of a fetus at 21 weeks and 3 days of gestation. With a tentative diagnosis of type I congenital cystic adenomatoid malformation, the pregnancy was followed with regular serial antenatal sonograms. After 35th weeks of gestation, the lesion started to reduce in size and the baby had a good prognosis after birth.

Conclusion: Unless hydrops fetalis or lifethreatening malformations coexist, a prenatally diagnosed isolated, type I congenital cystic adenomatoid malformation can be managed conservatively with a favorable prognosis.

Keywords: Congenital cystic adenomatoid malformation, prenatal diagnosis, management.

Tip-1 konjenital kistik adenomatoid malformasyonlu bir olgunun prenatal tanısı ve yönetimi

Amaç: Prenatal tanı almış bir tip I konjenital kistik adenomatoid malformasyon olgusunu sunmak.

Olgu: Bir fetüsün 21 hafta 3 günlük iken yapılan ultrasonografisinde, sol akciğer alanında sağa belirgin mediastinal yer değiştirmeye neden olan, 13.9 X 10.8 mm uniloküler ve 16.7 X 14.9 mm multiloküler anekoik kistik yapılar gözlendi. Tip I konjenital kistik adenomatoid malformasyon ön tanısıyla, düzenli seri antenatal sonogramlarla gebelik takibine devam edildi. Otuzbeşinci gebelik haftasından sonra lezyon küçülmeye başladı ve doğumdan sonra bebeğin prognozu iyiydi.

Sonuç: Hidrops fetalis veya hayatı tehdit edici malformasyonlar eşlik etmediği sürece, prenatal tanı alan izole tip I konjenital kistik adenomatoid malformasyon olguları iyi bir prognoz beklentisiyle konservatif takip edilebilir.

Anahtar Sözcükler: Konjenital kistik adenomatoid malformasyon, prenatal tanı, yönetim.

Introduction

Congenital cystic adenomatoid malformation (CCAM) is a developmental hamartomatous abnormality of the lung and represents approximately 25% of all congenital lung lesions. As a distinct entity, it was firstly described by Ch'in and Tang in 1949 and is characterized by overgrowth of the terminal bronchioles[1]

The pathologic classification of CCAM is based on cyst size and includes three types.[2] Type I has 1 or more large (>2 cm) multiloculat-

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ed cysts while type II has smaller uniform cysts (<1 cm). Type III is not grossly cystic and referred to as the "adenomatoid" type. During routine prenatal ultrasonography, CCAM is usually identified as a mass in the fetal chest which may be solid, cystic (solitary or multiple) or both. With large CCAMs, mediastinal shift, polyhydramnios, cardiac compression and fetal hydrops may also be found. The prognosis depends on the histologic type and is also influenced by the associated findings.

In this report, we aimed to present a case of large type I CCAM with mediastinal shift which was diagnosed prenatally at 21 weeks and 3 days of gestation and had a good prognosis after birth.

Case

A 29-year-old, gravida 1, para 0 woman was referred to our hospital for fetal evaluation following the identification of a large fetal thoracic cyst at 21 weeks and 3 days of gestation with a presumptive diagnosis of diaphragmatic hernia. Before referral, the course of the pregnancy had been uneventful. We observed a 13.9 X 10.8 mm unilocular and a 16.7 X 14.9 mm multilocular anechoic cystic structures at the left lung field with a marked mediastinal shift to the right on B-mode imaging which was performed with a Voluson E8 Expert (GE Healthcare, Wauwatosa, WI, USA) ultrasound machine (Figure 1). The diaphragm was intact (Figure 2) and there were no hydrops fetalis or associated congenital anomalies. Amniocentesis was performed, which revealed a normal karyotype of 46, XY. The tentative diagnosis was type I congenital cystic adenomatoid malformation. After detailed counseling, the parents opted for continuation of the pregnancy. Regular serial antenatal sonograms revealed that the fetus had a persistent left lower lung cystic lesion until 35th weeks of gestation. Thereafter, it started to reduce in size and nearly 90% of it disappeared until delivery. A live male baby weighing 3170 g was delivered by cesarean section at 39 weeks

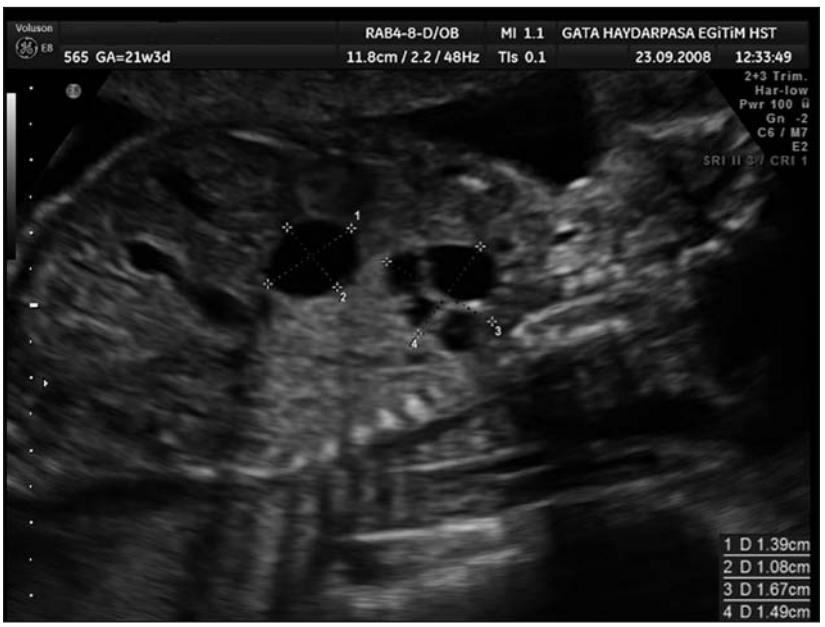

Figure 1. Shape 1. Sagittal ultrasonographic section of thorax in the fetus with type I congenital cystic adenomatoid malformation, showing cystic lesions in the left lung region.

and 5 days' gestation. Apgar scores were 9 at one minute and 10 at 5 minutes. The newborn breathed well and the signs of respiratory distress such as tachypnea, cyanosis, or dyspnea were not observed at birth. A magnetic resonance imaging (MRI) scan of the chest after birth revealed only a small remnant lesion of CCAM. Postnatal follow-up of the baby was uneventful until 7th month and the baby still receives regular follow-up.

Discussion

CCAM is believed to result from an arrest of normal fetal pulmonary maturation before the seventh week of gestation, leading to the development of dysplastic bronchopulmonary tissue.[3] Approximately 4-26% of cases and mostly the ones with type II can be associated with other congenital abnormalities such as diaphragmatic hernia, skeletal malformations, extra lobar pulmonary sequestration, bilateral renal agenesis/dysgenesis, jejunal atresia, cardiovascular malformation, pulmonary hypoplasia, depending on the time and type of insult. Syndactyly, lomber bifid spine and imperforation of the anus have also been shown in a case with type III.[4] However, our case, CCAM type I, was not associated with any congenital anomalies.

Although ultrasonography is quite helpful in the prenatal diagnosis of CCAM, diagnostic errors are also possible. Mediastinal lesions, such as cystic teratoma or neurenteric cyst; diaphragmatic hernia, extralobar and intralobar bronchopulmonary sequestration and bronchogenic cyst should be considered in the differential diagnosis and excluded by careful ultrasonographic assessment with the help of color-flow Doppler.[5-6] In our case, the patient was also referred to our clinic with a diagnosis of fetal diaphragmatic hernia and it was excluded by observing an intact diaphragma, normal stomach and abdominal anatomy.

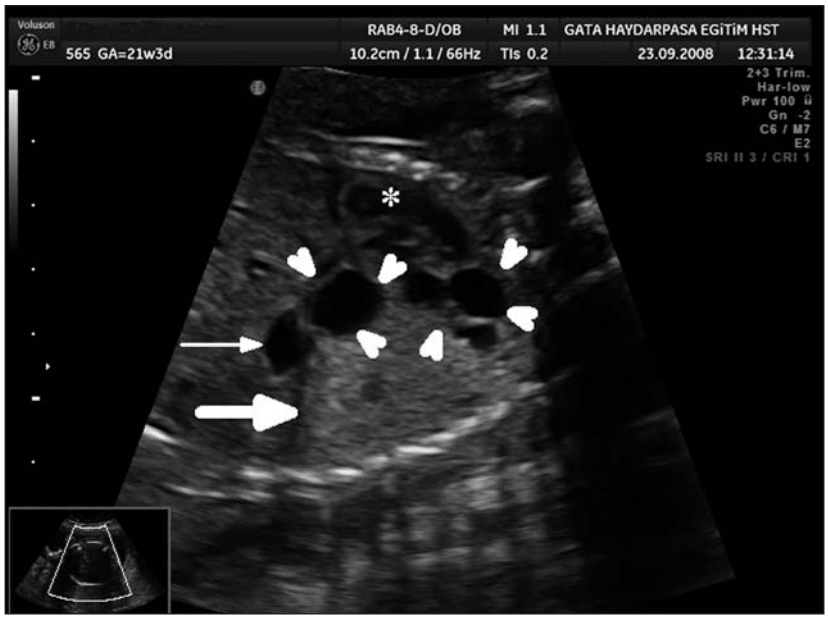

Figure 2. Shape 2. Sagittal ultrasonographic section of thorax and upper abdomen in the fetus with type I congenital cystic adenomatoid malformation, showing intact diaphragma (thick arrow), stomach (thin arrow), cystic lesions (arrowheads) in the left lung and heart (asterix).

There are conflicting opinions in the literature about the association of CCAM and chromosomal abnormalities. When a CCAM has been antenatally identified, although some authors advise to obtain a fetal karyotype only if there is an associated finding requiring chromosomal analysis,[7] the others advise fetal and karyotyping in all cases of CCAM, because fetal prognosis has been improved in recent years with intrauterine and/or postpartum therapy in these pregnancies.[8] More specifically, beside the reports indicating that CCAM Type II appears to be more frequently associated with other fetal abnormalities and aneuploidy,[9] there is a case series not revealing any abnormal karyotype in a review of 18 CCAM patients (9 of them are CAM Type II).[10] As proposed by International Society of Ultrasound in Obstetrics and Gynecology & Fetal Medicine Foundation,[11] our clinical approach is to offer fetal karyotyping when major defects are demonstrated on 18–23-week scan even if these defects are apparently isolated.

Most of the time, the lesions seen in CCAM are unilateral. However, very rarely, bilateral lesions may be encountered.[4]

Although it has not been encountered in other series, Sapin et al. observed the great predominance of left-sided lesions in their patients.[12] In our case, the lesion was unilateral and left-sided. According to the classification of CCAM using ultrasonographic findings, our case was CCAM type I and had a good prognosis. In general, this type is the most common form of CCAM and has the best prognosis of the three types, with a survival rate of 69%. Both of type II and type III have a poor prognosis (pulmonary and cardiac insufficiency in the newborn) with a mortality rate of 100%.[2,13] Baytur et al. also reported a case with type I CCAM in which the prognosis was also good.[14] Therefore, pregnancy termination may be advised when an extensive type II or type III malformation is detected before viability is reached while referring the patient to a perinatal center for scheduling the delivery and providing optimum neonatal care should be considered if the anomaly is detected later and if the findings are type I.[13] The prognosis is also considered to be worse if CCAM is microcystic (5 mm or less) and associated with a mediastinal shift, polyhydramnios or hydrops.[15] In our case with type I CCAM, the lesion was not microcystic and the only associated finding was a mediastinal shift. The pregnancy was decided to go ahead and the prognosis of the baby was not poor. Therefore, in our opinion, the associated findings other than nonimmune hydrops fetalis or accompanying life-threatening malformations may not affect the clinical outcome of the patients significantly. In our case, gradual reduction in the size of the mass and shift of the mediastinum back to the midline was observed during the last trimester of pregnancy. Postnatal magnetic resonance imaging also confirmed the spontaneous regression of the lesion. The exact mechanism by which these lesions shrink is unclear. Although the natural history of prenatally diagnosed pulmonary lesions is variable, Adzick reported that approximately 15% of CCAM lesions may decrease in size during gestation.[16] Therefore it is advised to follow-up these cases with serial prenatal sonograms and postnatal imaging studies.[6]

Conclusion

As a conclusion, after extensive counseling, a prenatally diagnosed isolated, CCAM can be managed conservatively with a favorable prognosis unless hydrops fetalis or life-threatening malformations coexist. Since the lesion might regress spontaneously, a serial scan should be arranged to monitor the size of the lesion in all affected fetuses.

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Pandemic Influenza A (H1N1) and Pregnancy: Case Reports

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Abstract

Objective: To discuss two cases who had Pandemic Influenza A (H1N1) virus infection in their pregnancies.

Case: Two pregnant cases in the third trimester were submitted to the hospital with high fever and dyspnea. These patients who were diagnosed as Pandemic Influenza A (H1N1) virus infection were delivered . The patients and newborns were followed up in the intensive care unit and were discharged uneventfully.

Conclusion: Pandemic Influenza A (H1N1) virus infection may proceed severe in the pregnancy. Early diagnosis and treatment are important.

Keywords: Pandemic influenza A (H1N1) infection, pregnancy.

Pandemik influenza A (H1N1) ve gebelik: olgu sunumu

Amaç: Gebeliklerinde Pandemik İnfluenza A (H1N1) enfeksiyonu geçiren iki olgunun klinik seyrini tartışmak.

Olgu: Üçüncü trimesterde gebeliği olan iki olgu, yüksek ateş ve nefes darlığı şikayeti ile hastaneye başvurdu. Pandemik İnfluenza A (H1N1) enfeksiyonu tanısı konulan hastaların doğumları gerçekleştirildi. Yoğun bakım şartlarında takip edilen anne ve bebekleri sorunsuz olarak taburcu edildiler.

Sonuç: Pandemik İnfluenza A (H1N1) enfeksiyonu gebelikte ağır seyredebilmektedir. Erken tanı ve tedavi önemlidir.

Anahtar Sözcükler: Pandemik influenza A (H1N1) enfeksiyonu, gebelik.

Introduction

Influenza viruses are RNA viruses from orthomyxovirus family. Influenza has three types as A, B, and C. Influenza A is separated subtypes hemagglutinin and neuroaminidase antigens. H1N1 virus is also the subtype of Type A influenza. Hemagglutinin virus is responsible for adsorption and neuroaminidase is responsible for the spread of virus.[1] Clinical profile has a course of fever, cough, sore throat, nasal flow, myalgia and diarrhea. Disease profile associated with secondary pneumonia may develop. Symptoms may arise within 7 days after being exposed to the virus. Although the disease has no complication in many pregnants, influenza morbidity risk is higher in pregnancy.[2] Taking the previous influenza pandemics into consideration, mortality rates are high especially at third trimester. In 1918 pandemics, 50% of 1350 pregnants got pneumonia and fatality rate was found as 27%.[3,4] Final diagnosis was found by reverse transcription polymerase chain reaction (TR–PCR) and virus culture.[3] Centers for Disease Control (CDC) suggests to use

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oseltamivir 75 mg for 5 days as 2 tablets per day. Though the gestational category of oseltamivir is C, no negative effect was reported on fetus and pregnants.[5]

Patients should be interfered in an isolated room by mask, gloves and protective clothing.

In this study, two pregnants admitted to our clinic and diagnosed with Pandemic Influenza A (H1N1) virus infection were presented and discussed in the light of the literature.

Cases

Case 1

Twenty-five years old pregnant with gravida 2, parity 1 and 28 weeks and 4 days of twin admitted to the hospital with complaints of high fever, sore throat and respiratory distress. Vital findings of the patient were normal except 38.0 ºC fever. In her obstetric ultrasonography, she had twin pregnancy conforming to the gestational week. Collum was found as closed in the obstetric examination. Her NSTs were observed as reactive. Rales were found in the respiratory system examination evaluating pulmonary diseases. The patient did not want to have a pulmonary radiography though she was informed that her belly would be protected by lead vest. Due to the suspicion of Pandemic Influenza A (H1N1) virus infection, oseltamivir 75 mg as 2 tablets per day and sulbactam – ampicilline 1.5 gr, 4x1 doses were initiated. Nasopharynx wipe sample was taken by provincial health directorate and our department was informed that the patient was positive for Pandemic Influenza A (H1N1) virus 48 hours later.

It was seen in the laboratory examination that AST value was 65 IU/L and ALT value was 87 IU/L while other laboratory values were found as normal. Betamethazone (6 mg and at the dose of 2 x 2 ampoule) was applied intramuscularly for fetal lung maturation. The patient was urgently taken to the cesarean at the 7th day of her hospitalization when respiratory distress increased in her clinical follow-up, oxygen saturations were below 90% despite the mask and nasal valve and oxygen treatment, AST and ALT values increased more, and late decelerations occurred at NST. Two living girls (1500 g and 1330 g) were delivered. The patient was taken to normal service when recovery was observed in her general condition after 48th hour of her postoperative follow-up. Her laboratory values regressed. The patient was discharged healthily 14 days later.

The first baby (1500 g) was intubated due the diagnosis of respiratory distress. Surfactant treatment was applied. Only oxygen treatment was applied to the second baby. Ampiric ampicilline and amicasin treatment was applied in order to prevent secondary bacterial infection. No reproduction occurred in blood cultures. Respiratory distresses of babies regressed and they were discharged healthily on 18th and 19th postpartum days.

Case 2

Twenty-six years old pregnant with gravida 7, no parity, and pregnancy evacuation 6 on her 35th gestational week admitted to the emergency service for respiratory distress and sore throat.

In the evaluation of the patient during the application, general status of the patient was moderate and the consciousness was open. Her body temperature was 37,6°C, respiration rate was 28/min, pulse was 130/min and oxygen saturation was 99%. There was asthma bronchial and smoking habit (20 package years) in the history of the patient. Single pregnancy was observed as compliant with gestational week in the obstetric ultrasonography. Collum was closed in the obstetric examination and NSTs were reactive. The patient was hospitalized in the service by the diagnosis of Pandemic Influenza A (H1N1). The patient was taken into follow-up and treatment in an isolated room in

service conditions by taking protection precautions for health personnel. Oseltamivir treatment was initiated, and nasopharynx wipe sample was taken. Her lung radiography was taken by protecting her belly with a lead vest. There were pneumonic infiltration diagnoses in the lung radiography. Sulbactam – ampicilline, steroid, and bronchodilator treatments were initiated for thoracic diseases. Since oxygen saturation values continued to reduce despite the oxygen treatment under service and her general condition deteriorated, the patient was taken to urgent cesarean and a 2440 g living boy was delivered. Oxygen treatment was applied and the patient was discharged healthily on postpartum 3rd day. Postoperative follow-up of the patient was performed in the intense care unit and respiratory support was provided by mechanical ventilator.

Wipe sample taken by provincial health directorate was reported to our clinic as Pandemic Influenza A (H1N1) virus infection.

The patient was connected to mechanical ventilator for 8 days. No reproduction was observed in the blood, urine and tracheal aspirate samples of the patient. The patient with regression in the infiltration of her lungs according to the thorax computed tomography was extubated. When the spontaneous respiration of the patient became normal, she was discharged on the 11th day of her hospitalization.

Discussion

Maternal and fetal risks associated with Influenza virus infection at pregnancy are at high rates due to hormonal, immunological and mechanical changes occurred.[6,7] Like in our cases, complication and hospitalization rates increases as gestational week increases.[8]

CDC reported mean hospitalization period as 2-15 days for Pandemic Influenza A (H1N1).[9] This period was 11 and 14 days in our cases. The existence of asthma bronchial and smoking habit in the history of our second case caused infection clinic to proceed rapidly and become more severe.

No other reason except Pandemic Influenza A (H1N1) virus infection was found to explain the increase of liver transaminases in the first case. Serum bile acids and normal thrombocyte and LDH levels supported transaminase increase associated with infection. Transaminase increases associated with influenza infection were reported by Monto et al. The increase in transaminase levels shows that clinical profile may follow an atypical route.[10]

The full effect of maternal influenza infection on fetus is not known completely today. It was shown that the risks of cerebral palsy, encephalopathy and neonatal death increased in the babies of mothers who had high fever.[11,12] This case affected the delivery decisions of current cases. No clinical and laboratorial findings of infection were found in the postpartum follow-up of the babies. Support treatment was applied in terms of prematurity. Breast milk was given when enteral feeding was initiated. No additional fetal antiviral treatment was applied due to maternal antiviral treatment. More cases are needed for the observation of Pandemic Influenza A (H1N1) virus infection at pregnancy. We wanted to present these two cases as they might be helpful in diagnosis and treatment planning of such infections at pregnancy.

Conclusion

Chemoprophylaxis and early hospitalization if needed should be considered within first 48 hours in the existence of clinical suspicion since Pandemic Influenza A (H1N1) virus infection may progress slowly at pregnancy. It can be suggested that this is the reason for non-existence of mortality in our current cases.

Antiviral treatment and vaccination at pregnancy should be suggested by taking benefit-harm balance into consideration.

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Subchorionic Hematoma Associated with Preeclampsia and Fetal Distress: Case Report

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Abstract

Objective: In this case report; we present a patient of 26 weeks pregnancy with a prominent subchorionic hematoma and discuss the differential diagnosis of placental masses with respect to the related literature.

Case: Twenty years old, 26 weeks pregnant patient, referred to our clinic with the presumptive diagnosis of PPROM (preterm prematrue rupture of the membranes), ablatio placentae and IUGR (intrauterine growth restriction) was evaluated. Doppler ultrasonography revealed; a hypoechoic mass lesion with a diameter of 5 cm in the placenta compatible with hematoma. The diagnosis subchorionic hematoma, was established by means of 3D sonography and clinical findings. Worsening of fetal and maternal wellbeing led to the early delivery. Histopathological examination of the placenta verified the subchorionic hematoma.

Conclusion: Differential diagnosis of subchorionic hematoma from ablatio placenta and chorioangioma should be made, because of the differences in the clinical followup and management. Doppler, 3D sonography and MRI are the main diagnostic tools. Worsening of the fetal or maternal wellbeing should prompt immediate delivery.

Keywords: Subchorionic hematoma, ablatio placentae, chorioangioma.

Preeklampsi ve fetal distres ile ilişkili subkoryonik hematom: olgu sunumu

Amaç: 26. gebelik haftasında geniş subkoryonik hematom olgusunu ve plasental kitlelere tanısal ve klinik yaklaşımla ilgili literatür bilgisi sunmak.

Olgu: Ablasyo plasenta, erken membran rüptürü ve gelişme kısıtlanması ön tanılarıyla refere edilen 20 yaşında SAT 'e göre 26 hafta G1P0 olgu değerlendirildi. Yapılan Doppler ultrasonografide plasenta içinde 5 cm çapında hipoekoik hematom ile uyumlu kitlede kan akımı tespit edilmedi. Üç boyutlu ultrasonografi ve klinik bulgularla Subkoryonik Hematom tanısı kondu. Fetal ve maternal durumda kötüleşme nedeniyle gebelik 27. haftada sonlandırıldı. Doğum sonrası tanı plasentanın patolojik incelemesiyle doğrulandı.

Sonuç: Subkoryonik hematomanın klinik izlem ve tedavide farklılıklar göstermesi nedeniyle Ablasyo plasenta ve koryoanjiomadan ayırıcı tanısı mutlaka yapılmalıdır. Doppler ve üç boyutlu ultrasonografi ve MRG tanıya ulaşmada önemlidir. Subkoryonik hematom olgularında fatal distres ve maternal durumda kötüleşme söz konusu olduğunda doğum gerçekleştirilmelidir.

Anahtar Sözcükler: Subkoryonik hematom, ablasyo plasenta, koryoanjioma.

Introduction

Our aim is to present wide subchorionic hematoma case at 26th gestational week and its literature information associated with diagnostic and clinical approach towards placental masses.

Cases

Twenty-years-old case with 26th week G1P0 according to SAT was referred with the prediagnoses of ablatio placenta, early membrane rupture and growth retardation. Her blood

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pleasure was measured as 180/110 mmHg at our polyclinic. It was seen in vaginal speculum examination that there was no amnion fluid broke. In the routine urine examination, protein 4 was found positive, protein within urine of 24 hours was 12 gr, ALT was 46 U/L and LDH was 289. In the abdominal ultrasonography performed by Voluson 730 Expert, C was found below 5% in the biometric measurements of fetus while it was between 5% and 25% in other measurements. Three hypoechoic solid masses were observed within placenta near the umbilical artery entrance with 5 cm diameter maximum (Figure 1). No flow was detected in the mass by Doppler ultrasonography (Figure 2). Three-dimensional ultrasonographic examinations were done on masses (Figure 3). The patient was hospitalized by pre-diagnoses of subchorionic thrombohematoma, chorioangioma and preeclampsia. Blurred vision developed in the clinical follow-ups of the patient. The delivery was performed at 48th hour after applying corticosteroid upon the non-reactive progress of NST, decrease of thrombocyte counts and increase of ALT. A living 590g male baby was delivered by cesarean and the baby

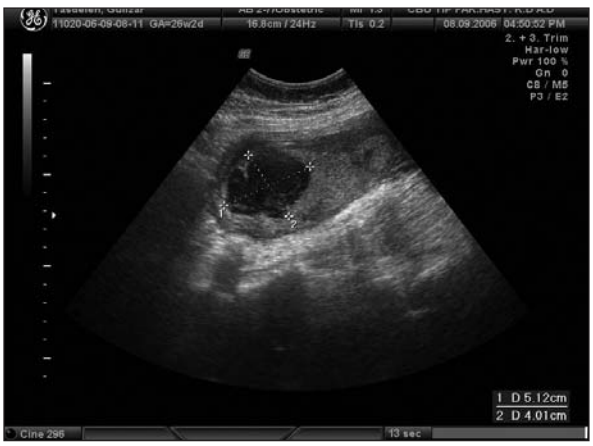

Figure 1. Hypoechoic mass within placenta.

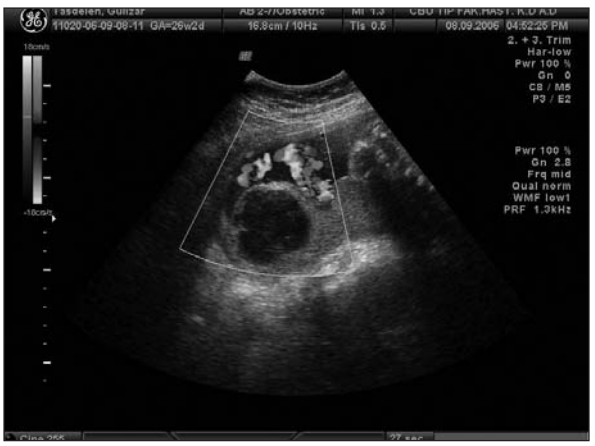

Figure 2. No flow was detected in the Doppler examination performed on the mass.

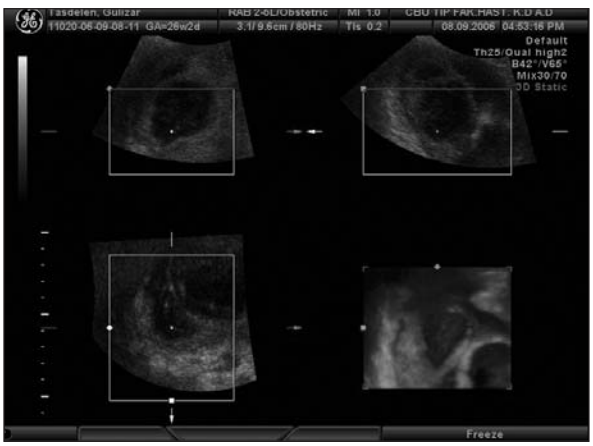

Figure 3. Three dimensional views of masses.

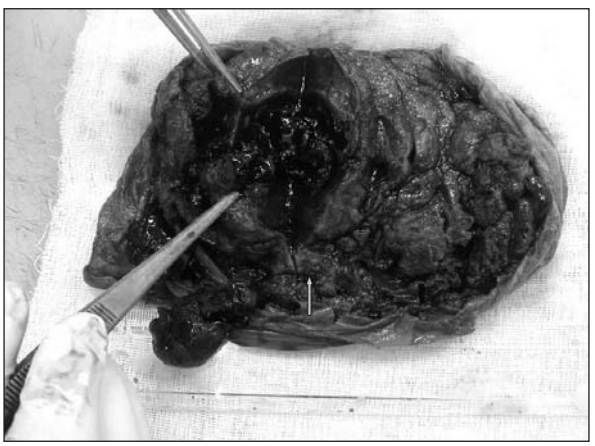

Figure 4. Intraplacental hematoma (thick arrow) and adjacent infarction areas (thin arrows).

was taken into newborn care unit. After delivery, a 7x5x3 cm intraplacental located hematoma with lobule contour was seen at macroscopic examination of placenta as well as numerous pale yellow – light brown colored infarction areas (Figure 4). Hematoma blood elements and mixed fibrin mass were used in microscopic examination of placenta. Ghost villi structures where their nuclei cannot be seen within infarction areas and characterized necrosis areas caught attention. Calcification focuses were seen both in necrosis areas and around hematoma.

Discussion

Subchorionic hematoma was first named as Breus's Mole by Breus in 1982 and it can be defined as massive maternal bleeding which separates chorionic villi from chorionic plate.[1] It is associated with serious gestational complications such as intrauterine growth retardation, fetal distress and fetal death.[2] Thrombolytic treatment in the development of subchorionic hematoma[3,4] was reported as the risk factor for advanced maternal age and chronic hypertension.[1] Also other placental masses and especially chorioangiomas should certainly be considered. Our most important tool for definitive diagnosis of masses within placenta is Doppler ultrasonography.

Sepulveda et al. evaluated seven pregnants by Doppler ultrasonography who had placental masses and pre-diagnosis of chorioangioma was confirmed in the pathological examination of four cases on whom flow was detected. 2 out of 3 cases on whom no flow was detected were found as having subchorionic thrombohematoma and one of them having subamniotic hematoma.[5] While hydrops and polyhdramnios development in fetus and blood flow within the mass makes us to think chorioangioma, like in our case, non-existence of blood flow within the mass, growth retardation, abnormal Doppler findings and accompanying oligohydramnios are the signs of subchorionic hematoma. However, growth retardation may also exist in chorioangiomas. In the 9 years of series of Prapas, seven cases were diagnosed as choriangioma by histopathological examination and polyhidramnios was seen in six of these cases while intrauterine growth retardation was seen in two of them.[6] Although these masses can be easily detected by two-dimensional Doppler ultrasonography, three-dimensional ultrasonography can contribute to accurate diagnosis.[7] Moreover, it was reported that MR may contribute to the diagnoses in cases which cannot be certainly diagnosed and that MR may recognize placental bleedings (retroplacental hematoma, intervillous thrombus, cubchorionic hematoma) and ischemic lesions.[8,9] The diagnosis was performed by two-dimensional ultrasonography and Doppler in our case and also three-dimensional ultrasonography was used. MR utilization was not considered as necessary for diagnosis.

In small and asymptomatic subchorionic hematoma cases, pregnancy complications and bad perinatal results are not expected and follow-up can be performed.[5] However, if a large mass or bleeding is in question which may significantly decrease the nourishment and oxygenation of fetus, growth retardation and fetal death may occur. As growth rate of placental masses are not known clearly, a close follow-up is significant. In a publication, it was reported that preterm labor occurred in 6 cases with chorioangioma and neonatal death in one case associated with rapid growth of tumor.[5] When reported cases were evaluated, it should be remembered that premature early membranous rupture, antenatal bleeding and intrauterine fetal death may occur in subchorionic hematoma cases.[10] Therefore, these cases should be followed up closely by Doppler ultrasonography and other fetal wellness tests. As in our case, delivery should be performed immediately when fetal distress in question. Asymptomatic cases far from term can be fol-

lowed conservatively as long as close follow-up is performed.

Conclusion

Since clinical approach and treatment options differ in the differential diagnosis of subchorionic hematoma, chorioangiomas and ablatio placenta should certainly be considered. When reported cases are evaluated, it should be remembered that premature early membranous rupture, antenatal bleeding and intrauterine fetal death may occur in subchorionic hematoma cases. As in our case, delivery should be performed immediately when fetal distress in question. Asymptomatic cases far from term can be followed conservatively as long as close follow-up is performed.

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Truncus Arteriosus Type1 With Prenatal Diagnosis: Case Report

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Abstract

Objective: Truncus Arteriosus complicates approximately 0,01 1000 live births. In this case report we discussed Truncus Arteriosus Type 1 case detected in utero with echocardiographic findings.

Case: A 31 year old pregnant woman at 26 weeks of gestation referred to our clinic. She hasn't got a consenginous history.According to the fetal echocardiographic findings we diagnosed Truncus Arteriosus Type 1.

Conclusion: The big part of the congenital heart diseaseses occurs in pregnancies with no risc factors. If we visualized the subaortic ventricular septal defect in the fetal heart, we can detected the associated abnormalities; such as Truncus Arteriosus.

Keywords: Truncus arteriosus, congenital heart anomalies, fetal echocardiography.

Prenatal tanı alan trunkus arteriozus tip 1: olgu sunumu

Amaç: İkinci trimesterdeki taramada tanısı konan, nadir görülen konjenital kalp hastalıklarından biri olan Trunkus Arteriyozus olgusu sunulmuştur.

Olgu: 31 yaşında, gebeliğin 26. haftasında kliniğimize başvuran, akraba evliliği hikayesi olmayan hastanın yapılan fetal ekokardiografisinde Trunkus Arteriozus Tip 1 tespit edildi.

Sonuç: Konjenital kalp hastalıklarının çok büyük bir kısmı risk faktörü içermeyen gebeliklerde meydana gelmektedir. Detaylı bir muayene ile subaortik ventriküler defektin izlenmesi altta yatan Trunkus Arteriozus'un yakalanmasına yardımcı olacaktır.

Anahtar Sözcükler: Trunkus arteriozus, konjenital kalp anomalileri, fetal ekokardiyografi.

Introduction

The incidence rate of congenital cardiac disease is 4-11/1000 live birth and this rate constitutes the most frequent congenital anomaly group among cardiac diseases. The most frequent cause of early neonatal deaths associated with congenital anomaly is cardiac diseases.[1,2] Truncus Arteriosus is characterized as single great artery coming out of heart and its incidence rate is 0.01 among 1000 live birth.[3] Truncus feeds systemic, coronary and pulmonary circulations. Echocardiographic finding is diagnosed by detecting also pulmonary arteries coming out from a single great artery wider than aorta and overlapping on ventricles.[3,4]

Like in other conotruncal anomalies, Truncus Arteriosus cannot perform in utero cardiac decompensation; however, decompensation may occur in first days of life. The surgical fixation is quite complex since it is needed to turn dysplastic truncal valve into aorta and to

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make a connection from right ventricle to pulmonary arteries. Even after a successful surgery, 10 years of survival rate is less than 80%.[3]

A major part of congenital cardiac anomalies occurs in pregnancies which do not include risk factors.[5,6] In our article, the case of Truncus Arteriosus Type 1 with prenatal diagnosis was discussed in accordance with the literature.

Cases

31 years-old G2 P1 pregnant without any consanguineous history admitted to our clinic at her 26th gestational week. No systemic disease was found in her anamnesis. Nothing was found in her family history except Type 2 Diabetes Mellitus of her mother. In the examination performed on her 12th gestational week, her nuchal transparency was measured as 2.5 mm. However, biochemical marker data of 1st and 2nd trimesters could not be found. In the detailed systemic examination performed by ultrasonography, no anomaly was observed except subaortic ventricular septal defect. Fetal growth was normal. Amniotic fluid volume was found normal. No notching was detected on bilateral uterine arteries. In the fetal echocardiography of the patient, heart was observed within left hemithorax and cardiac apex was observed as staying leftward. Atrial situs solitus was in existence. Heart chambers were in normal width and global heart contractions were detected as good. Atrioventricular concordance was full. Mitral tricuspid valve was normal. Intraventricualr septum developed; however, there was wide ventricular septal defect at outlet localization. A single great vessel (truncal vessel) output was observed from ventricles.

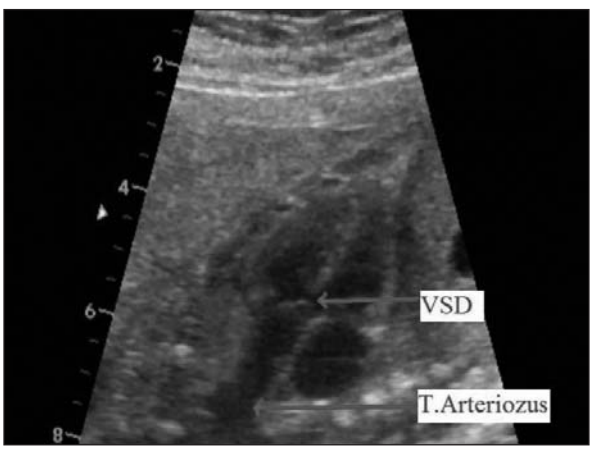

Figure 1. Truncus arteriosus and ventricular septal defect.

Though truncal vessel was slightly "overriding" the ventricular septal defect, it was coming out of right ventricular mainly and 80% of it was dextropositioned. Truncal vessel flow rate was found as 175 cm/sec. The pulmonary artery was just coming out of the distal of truncal valve as a single root. The pulmonary was separated into right and left branches and no stenosis was found on artery output. Truncal arcus was on rightward position and major vessels were observed, aorta coarctation or interruption. The finding was diagnosed as Truncus Arteriosus Type 1 according to the diagnoses mentioned above.

Genetic consultation was given to the family and karyotype analysis was suggested. Fetal blood sampling was performed to the patient in the same session. In the karyotype analysis performed on fetal blood, no numerical and structural anomalies were observed and chromosome formation was found as normal. In the examination performed by fluorescence in situ hybridization (FISH) on fetal blood, 22q11 micro-deletion was not found. Genetic consultation was given to the family about prognosis. The family stated that they wanted to maintain the pregnancy. The patient was called for control one week later. When the patient admitted to our clinic due to the reducing baby movements five days later, no fetal cardiac motions was detected. It was accepted as stillbirth and the patient was hospitalized in our department for delivery. Following the delivery induction, 1100 gr girl fetus was labored. In the postmortem examination, it was observed that pulmonary artery and aorta came out of right ventricle as a single root and thoracic aorta was descending rightwards on the high ventricular

septal defect (VSD) surface. No ductus arteriosus (Botalli) was observed. Prenatal diagnosis was confirmed.

Discussion

Prenatal diagnosis of congenital cardiac diseases is very significant since they create the most frequent anomaly at live births.[7,8] While prenatal evaluation of all other organ systems of fetus was improved rapidly through high standards and prevalent practices, it is hard to say the same for prenatal ultrasonographic evaluation of fetal heart despite the efforts which had began much earlier. In the study of Jaeggi et al., only 15% of all heart anomalies could be recognized at prenatal period.[9] While this rate was 30% in anomalies which could be recognized by cross-section of four chambers, prenatal diagnosis rate was only 6.7% in conotruncal anomalies associated with major vessel outputs.[9] The detection rate of heart anomalies was found as 26% in the study of Tegnander et al. by imaging four chamber in fetal heart anomaly scanning.[10] Bromler et al. showed that this rate could increase to 83% by including major vessels into the evaluation in addition to four chamber imaging.[11] Including major vessels into evaluation in fetal echocardiography does not only increase to intrauterine detection rate of congenital cardiac diseases but also provides to diagnose properly and effective intervention at postnatal period by consecutive examination technique.[5,10,11]

According to the classical classification, Truncus Arteriosus cases are gathered under three major groups. At Type-1 Truncus Arteriosus, the major pulmonary comes out of the location superior to artery truncal valve, the left posterolateral truncus and branches off to right and left pulmonary artery branches. In Type 2, right and left pulmonary artery branches (without the major pulmonary artery) come out of the same location as in Type 1 but separately. In Type-3, the outlet location is upper and separate from lateral. In this type, coronary artery anomalies are often.[12]

In our case, major pulmonary artery came out of the location superior to artery truncal valve and then branched off into right and left pulmonary artery branches. According to this definition, our case was found compatible with Truncus Arteriosus Type 1.

There are aortic arches located in the right side in 15-30% of Truncus Arteriosus cases.[13] Aortic arch was also located in the right side in our case.

It is known that there is ductus arteriosus agenesis in 50-75% of cases with Truncus Arteriosus.14,15 In our case, ductus arteriosus which could not be detected in fetal echocardiography was also not found in post-mortem examination.

It may be difficult to distinguish Truncus Arteriosus from Fallot tetralogy with pulmonary atresia in utero.[3] Final diagnosis is established by observing pulmonary artery leaving truncus. In pulmonary atresia cases with VSD, only aorta is followed instead of truncus and pulmonary artery is not followed.

There is also a relationship between congenital cardiac anomalies and structural chromosome anomalies. It is known that 22% of cardiac anomalies in fetus were seen together with aneuploid.16 The most frequent karyotype anomalies are Trisomy 18, 13, 21 and Monosomy X.[16] On the other hand, it was found in a study performed on 11-14 weeks old fetuses that 72.9% of fetuses with fetal achocardiography abnormality had karyotype anomalies.[17]

Beside major structural chromosome anomalies, it was found that conotruncal anomalies progressed together with 22q11 micro-deletion (Di George Syndrome; it is together with thymic aplasia, hypocalcemia, abnormal face shape and mental retardation). 22q11 micro-deletion was found in 35% of cases with Truncus Arteriosus.[12] In the examination performed on our case by FISH, no deletion was found and the karyotype analysis was reported as normal.

An opportunity will be obtained by fetal echocardiography for giving a full briefing to parents about prognosis and risks at 18th-22nd

gestational week and parent will be able to benefit the opportunity of termination on time in cases with severely poor prognosis. An overwhelming majority of congenital cardiac anomalies, as seen in our case, is formed of pregnancies not including risk factor. In Truncus Artoriosus cases, generally the view of normal four chambers is observed. In case of observing sub-aortic VSD in detailed examination during examining major vessels, truncus arteriosus diagnosis can be established. In fetuses with congenital cardiac diseases, performing fetal karyotype analysis and FISH will allow prenatal diagnosis of syndromes displaying association. In cases which are desired to continue pregnancy, genetic consultancy should be given for chromosome anomaly that can be followed with 14% frequency and for 22q11 deletion and Di George Syndrome that can be followed with 35% frequency. As seen in our case, congenital cardiac disease may also be responsible for fetal death cases. Performing postmortem examination will provide to establish the most proper diagnosis in stillborn fetuses that are not diagnosed as antenatal congenital cardiac disease.

Conclusion

An overwhelming majority of congenital cardiac diseases occur in pregnancies which do not have risk factors. By a detailed examination, observing sub-aortic ventricular defect would help to detect Truncus Arteriosus. Performing postmortem examination will provide to establish the most proper diagnosis in fetal death cases that are not diagnosed as antenatal congenital cardiac disease.

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