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The Co-Existence of a Live Fetus in a Twin Pregnancy with Hydatidiform Mole: Cross-Sectional Study

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ABSTRACT

Background & Objective: An unusual manifestation of prenatal trophoblastic illness is a twin molar pregnancy with a live fetus and a hydatidiform mole. Due to increased ovulation stimulation, this unusual scenario is rising. Most hydatidiform moles are complete with a fetus, while 0.005 to 0.01% of gestations have a partial mole. This report examined the clinical appearance and outcome of live fetuses with molar pregnancies.

Materials & Methods: This study was done at Adiwaniyah, where the Maternity and Pediatrics Teaching Hospital has one obstetrics center. The researcher reviewed 20 years of obstetric reports and found 12 cases of hydatidiform mole pregnancies, with ultrasound reports and/or postpartum examination of the product of gestation indicating twin pregnancy with a single viable fetus.

Results: The average age was 27.00 ±6.87 years, with a range of 18–37 years. The parity breakdown was eight (66.7%) primiparous and four (33.3%) multiparous. The average gestational age upon diagnosis was 19.50 ±3.09 weeks, ranging from 15 to 25 weeks. The average gestational age at delivery commencement was 26.67 ±7.09 weeks, with a range of 17–35 weeks. Seven (58.3%) amniocenteses were normal. Maternal complications included bleeding, anemia, blood transfusion, threatening miscarriage, and pre-eclampsia. End-of-gestation outcomes comprised five (41.7%) living and healthy newborns and three (25.0%) dead babies. There were five (41.7%) men and two (16.7%) women. Moreover, cesarean sections, early labor, and miscarriages were documented.

Conclusion: Twin pregnancies with a live viable fetus and a molar pregnancy is rare, hence early detection and care are crucial to prevent difficulties for the mother and fetus.

Keywords: Molar, Pregnancy, Twin, Living Fetus

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Introduction

Hydatidiform mole in pregnancy is one of a spectrum of related obstetric conditions known collectively as “trophoblastic disease of pregnancy” **(1, 2)**. These disorders arise out of the placenta and include a number of well-defined entities such as the benign placental tumor (Hydatidiform mole), and the malignant trophoblastic disorders that are associated with pregnancy (invasive mole, postmolar gestational trophoblastic neoplasia, epithelioid trophoblastic tumor, gestational choriocarcinoma and placental site trophoblastic tumor) **(3)**. The incidence rates of various kinds of these disorders vary globally. “In the United States”, the incidence rate is approximately one in every 1000 gestations **(4, 5)**. The estimated incidence rate of choriocarcinoma is in the range of one in every 20,000 pregnancies to one in every 40,000 pregnancies **(4, 5)**. Epithelioid trophoblastic tumors and “placental site trophoblastic tumors” may originate following any form of gestation; however, they are extremely rare forms **(6)**. Hydatidiform mole is sub-grouped into a partial and complete mole and is often regarded as the non-invasive form of trophoblastic disease of pregnancy. Hydatidiform moles possess the ability to develop into invasive and maligmole andons, even though they are typically thought of as benign **(7-9)**. The complete mole is different from the partial mole kind by having no fetal parts. It arises when an enucleated egg is fertilized by a sperm (haploid) that then duplicates or by 2 sperms, thus it is diploid and contains 46, XY or 46, XX chromosomes in 10% and 90% of cases respectively. Therefore, a complete mole contains paternal DNA only. On the other hand, partial more contains some fetal parts and harbors paternal and maternal DNA as it arises out of fertilization of an egg that is haploid by two sperms or a single sperm that then duplicates. Partial more karyotype is either 69, XXX or 69, XXY **(10-12)**. A special kind of presentation of gestational trophoblastic disease is a

twin molar gestation with a coexisting live fetus and a hydatidiform mole. This situation is rare, but it is demonstrating an increasing trend because of the increase in the ovulation stimulation procedures. The estimated incidence is between one in 20 000 to one in 100 000 pregnancies **(13, 14)**, with the majority of "complete hydatidiform moles with a fetus"; nevertheless, the estimated prevalence rate for a coexisting fetus with a partial mole is 0.005 to 0.01% of gestations **(15)**. The data about the co-existence of a live fetus with molar pregnancy in Iraq are scanty, thus, the purpose of the present research was to shed light on the medical presentation and net results of cases of co-existence of a live fetus with molar pregnancy in Aiwaniayh Province, Iraq.

Methods

This research work was carried out in Adiwaniyah province, Iraq, in which there is a single center for obstetrics that belongs to the Maternity and Pediatrics Teaching Hospital. The researcher reviewed obstetric reports for the last 20 years and obtained 12 cases of hydatidiform mole pregnancies in which there was an evidence of twin pregnancy comprising a single viable fetus and a mole pregnancy based on ultrasound reports and or postpartum examination of the product of gestation. The following variables were obtained and included in the study: age of mother, parity, gestational age at time of diagnosis, "gestational age at end of pregnancy", maternal complications and outcome of gestation. Ethical approval was issued by "the ethical approval committee of the college of Medicine / University of Al-Qadisiyah". There was no need for written consent of patients because the study was retrospective and included collection of data already present in patients' reports.

Results

The data obtained from the reports of 12 women is shown in Table 1. The general features of women involved in this research are shown in Table 2. The average age was 27.00 ±6.87 years and the range of age was 18-37 years. According to parity, there were 8 (66.7 %) primiparous women and 4 (33.3 %) multiparous women. The average gestational age at onset of diagnosis was 19.50 ±3.09 weeks, and it ranged from 15 to 25 weeks. The average gestational age at onset of delivery was 26.67 ±7.09 weeks, and the range was 17-35 weeks. The Outcome of gestation is shown in Table 3. Amniocentesis was normal in 7 (58.3 %) cases. Maternal complications included bleeding, anemia, blood transfusion, threatened miscarriage and pre-eclampsia. The outcome at end of gestation included alive and healthy babies and dead babies, 5 (41.7 %) and 3 (25.0 %), respectively. There were 5 (41.7 %) males and 2 (16.7 %) females. Miscarriage, preterm labor and cesarean sections were also reported.

Table 1. The data obtained from the reports of 12 women

No.	History	Gestational age at diagnosis (weeks)	Amniocentesis (Kt)	Maternal complication	Gestational age at delivery (weeks)	Outcome
1	Primi, 23 years	22	Normal	Anemia, blood transfusion	32	Preterm labor (PTL), female, alive and healthy
2	Multi, 28 years	15	Normal	Threatened miscarriage	18	Spontaneous miscarriage
3	Primi, 30 years	23	Normal	Pre-eclampsia (PE), blood transfusion	30	Emergency LSCS for fetal distress, male baby, alive and healthy
4	Primi, 32 years	20	Not done	Threatened miscarriage, vaginal bleeding	20	Inevitable miscarriage
5	Primi, 20 years	25	Normal	Antepartum hemorrhage, Postpartum hemorrhage	35	Elective LSCS for FGR, male alive and healthy
6	Primi, 20 years	18	Normal	Threatened miscarriage, anemia, blood transfusion	17	Spontaneous miscarriage
7	Primi, 18 years	16	Not done	Anemia, blood transfusion	25	PTL, male, dead
8	Multi, 37 years	20	Not done	APH	30	PTL, female, dead
9	Multi, 33 years	18	Normal	None	27	PTL, male, dead

No.	History	Gestational age at diagnosis (weeks)	Amniocentesis (Kt)	Maternal complication	Gestational age at delivery (weeks)	Outcome
10	Primi, 35 years	22	Normal	None	34	PTL, alive
11	Multi, 30 years	19	Not done	PPH	35	Male, alive
12	Primi, 18 years	16	Not done	Vaginal bleeding	17	Miscarriage

Table 2. Characteristics of women enrolled in this study

Characteristic	Result
Age (years)	
Mean ±SD	27.00 ±6.87
Range	18-37
Parity	
Primiparous	8 (66.7 %)
Multiparous	4 (33.3 %)
Gestational age at diagnosis (weeks)	
Mean ±SD	19.50 ±3.09
Range	15-25
Gestational age at delivery (weeks)	
Mean ±SD	26.67 ±7.09
Range	17-35

SD: standard deviation

Table 3. Outcome of gestation

Characteristic	Result
Amniocentesis	
Normal	7 (58.3 %)
Not done	5 (41.7 %)
Maternal complication	
Anemia	3 (25.0 %)
Blood transfusion	4 (33.3 %)
Threatened miscarriage	3 (25.0 %)
Pre-eclampsia	1 (8.3 %)
Vaginal bleeding	6 (50.0 %)
Outcome of gestation	
Alive and healthy	5 (41.7 %)
Dead	3 (25.0 %)
Male	5 (41.7 %)
Female	2 (16.7 %)
Spontaneous miscarriage	3 (25.0 %)

Characteristic	Result
Inevitable miscarriage	1 (8.3 %)
Preterm labor	5 (41.7 %)
Emergency cesarean section for fetal distress	1 (8.3 %)
Elective cesarean section for fetal growth restriction	1 (8.3 %)

Discussion

Most of previous studies about the co-incidence of live viable fetus and a molar pregnancy were in the form of case reports **(16-20)**. In a case of twin molar pregnancy with a hydatidiform mole and a coexisting live fetus identified at gestational week 14, Braga, Obeica **(16)** presented the results of two-dimensional (2D) and three-dimensional (3D) ultrasonography and magnetic resonance imaging. They also discussed post molar follow-up and obstetric management. A case report of a twin pregnancy with one fetus and a coexisting mole identified at 13 weeks was presented by Rai, Shripad **(17)**. Serial S-hCG, ultrasound for fetal growth, measurement of the molar sac, and theca lutein cysts, which steadily shrunk in size throughout the second trimester of pregnancy, were all used to closely monitor the pregnancy. Due to the problems of the pregnancy, an urgent cesarean delivery was done at 36 weeks **(17)**. The case of a 32-year-old lady who visited "the Sultan Qaboos University Hospital in Muscat, Oman, in March 2012" at 13 gestational weeks with vaginal bleeding and abdominal pain was described by Sheik, Al-Riyami **(18)**. A viable fetus and a hydatidiform mole pregnancy were discovered during an ultrasound. The patient and her husband choose a conservative management strategy after receiving considerable counseling. Unfortunately, a hysterotomy had to be done at 17 weeks of pregnancy since there had been a serious hemorrhage. Histopathology results showed one whole mole with a coexisting fetus and a healthy placenta, and the postoperative phase was uncomplicated. Human chorionic gonadotropin levels in the patient's serum remained normal for 18 months after her operation. Makary, Mohammadi **(19)** described a case of a 19-year-old primigravida who was carrying a live fetus and had a full hydatidiform mole at 25 weeks of gestation **(19)**. After a brief dexamethasone course to hasten the fetal lung maturation and preeclampsia accompanied by hypertension that is uncontrolled, the termination of pregnancy was done by cesarean surgery. Without incident, the placenta and a live female fetus with normal morphology were delivered alongside a distinct mass of a whole mole. Choriocarcinoma of the uterus, with spread to the lung and left kidney that showed response to treatment with chemotherapy, complicated the postpartum course. Their case demonstrates the range of maternal problems that call for close pre- and post-natal observation and is a rare instance of a twin gestation involving a fully developed hydatidiform mole and a coexisting live fetus. Twenty examples were presented by Vimercati et al. **(20)**. In both cases, ultrasound scanning revealed a live, developing fetus that was developing healthily with a healthy placenta and another intrauterine echogenic lump that resembled a hydatidiform mole. Fetal karyotypes were normal and hCG levels were noticeably elevated. In the first example, a cesarean surgery performed at 28 weeks' gestation, and in the second case, one conducted at 26 weeks' gestation, both resulted in the birth of a live, healthy newborn with two adjacent placentas. Complete hydatidiform mole was determined by microscopic investigation of the malformed placentas. Serial maternal serum hCG levels exhibited a trend toward decline and eventually became undetectable a few months after delivery, and the neonates performed well. To examine the clinical course and the aftereffects of patients with hydatidiform mole coexistent with twin live fetus (HMTF), Matsui, Sekiya **(21)** reported 72 cases in a national collaborative study carried out in Japan. Based on histological criteria and gross appearance, 72 instances of HMTF were identified. Cytogenetically confirming androgenetic origin of the molar components in 18 cases (whole mole). The prevalence of persistent trophoblastic tumors (PTT) was 30.6% overall in HMTF patients, and it was 50.0% in the 18 patients with CHMTF (full mole with verified androgenetic coexistence with twin live fetus). The average gestational age at delivery or termination of pregnancy among these patients was 20.6 weeks for those who developed PTT (n = 9) and 19.4 weeks for those who did not (n = 9). Patients who later had PTT had a significantly greater incidence of severe maternal problems (P 0.05). In comparison to patients with a single complete mole in prior research, patients with CHMTF had a rate of PTT development that was significantly greater (50 vs. 12.5%). However, given that the risk of malignancy does not rise with increasing gestational age, patients with HMTF may be able to carry their pregnancies to term as long as severe mother problems are managed and the fetal development and karyotype are natural. "To the best of our knowledge", this is an initial Iraqi article that described 12 cases of co-existence of normal fetus with molar gestation in which we presented detailed account about the outcome of gestation.

Conclusion

The incidence of twin pregnancy with a live viable fetus and a molar pregnancy is extremely low, and early diagnosis and intervention is essential to avoid

unnecessary complications to the mother and to the fetus.

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Conflict of Interest

None.

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