COMPLETE HYDATIDIFORM MOLE IN A TRIPLET PREGNANCY WITH COEXISTING TWO LIVE FOETUSES- A CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

BACKGROUND

Hydatidiform mole with coexisting twin foetus is an infrequent condition that has been reported in a limited number of cases.⁽¹⁻⁴⁾ Only 12 cases of triplet pregnancy with complete mole and two foetuses following ovulation induction, Assisted Reproductive Techniques (ART) and spontaneous conceptions are reported.⁽¹⁾ We describe a case of complete hydatidiform mole with coexisting viable two foetuses following ovulation induction. A 35-year-old lady was pregnant following ovulation induction with clomiphene citrate. A triplet pregnancy with two live foetuses and a complete mole was delivered at 32 weeks of gestation. The antenatal period was uneventful. Continuation of pregnancy as long as maternal complications are absent or controllable can be a safe and rewarding effort in managing the patient.

KEYWORDS

Artificial Reproductive Techniques, Complete Hydatidiform Mole, Preterm, Triplet Pregnancy.

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BACKGROUND

The incidence of twin pregnancies with a live foetus and coexisting complete hydatidiform mole is 1 per 22,000 to 100,000 conceptions.⁽⁵⁾ Triplet pregnancies with two live foetuses and coexisting complete hydatidiform mole are even rarer. Only 13 cases of hydatidiform mole with two foetuses were reported following ovulation induction, ART and spontaneous conception.⁽¹⁾ And here we present a case of triplet pregnancy with two live foetuses and coexisting complete hydatidiform mole reported in our Institute.

CASE REPORT

A 35-year-old woman with history of late marriage and primary infertility underwent ovulation induction by clomiphene citrate 50 mg for 5 days in a cycle. She was diagnosed as a hypothyroid and was on T. Eltroxin 50 micrograms once daily. She conceived after 3rd cycle of ovulation induction. Investigations done at 3rd month of gestational age were found to be within the normal limits except for Beta-HCG, which was elevated up to 1,05,185. Antenatal scan was taken and revealed a twin gestation Fig. 1. She did not have any specific symptoms. She did not continue her regular antenatal checkup. Third trimester scan was taken, showed molar changes (Fig. 2). Then she had a pre-term pain at 32 weeks of gestational age. Inj. Betnesol IM was given. On examination uterus overdistended, mildly acting and multiple foetal parts were palpated.

As there was no progress in labour, scan was done and first baby was found to be in transverse lie. Emergency LSCS was done and delivered two female preterm babies. Twin I weighed 2100 grams and Apgar was 8/10. Twin II weighed 3213 grams and Apgar was 9/10. Also delivered monochorionic diamniotic placenta with a complete vesicular mole. Both the babies survived well. Postpartum period was uneventful.

The placenta with molar changes are subjected to histopathological examination. Macroscopically, the placenta was monochorionic diamniotic with a distinct groove in between and showed cord attachment. Attached to this was a complete vesicular mole composed of fluid filled vesicles covered by a membrane (Fig. 3).

Microscopically, molar placenta showed oedematous avascular chorionic villi with circumferential proliferation of trophoblast. There was a moderate degree of trophoblastic proliferation with atypia (Fig. 4). Other 2 normal appearing placentae and cords showed normal histology. As p57 will be positive in normal placenta and partial mole and absent in complete mole, p57 was done and found to be negative (Fig. 5).

Figure 1. Arrow shows Membrane between Two Gestational Sac indicating Multiple Gestations

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Figure 2. Arrow shows Vesicular Changes at Posterior Fundal Pole

Figure 3. Arrows show Grooves in between Placentae

Figure 4. Shows Oedematous Avascular Chorionic Villi with Circumferential Proliferation of Trophoblast

Figure 5. P57 Negative in Trophoblasts and in Stromal Cells

DISCUSSION

Triplet pregnancy with coexistence of complete hydatidiform mole is a rarer condition, which has been reported previously in 13 cases (as summarised in the Table).⁽¹⁾ For past 5 years, we have reported 4 cases of multiple pregnancies with hydatidiform change. Among them 3 cases were twin pregnancies with single live foetus and 1 hydatidiform mole. This was the first case we reported a triplet pregnancy with 2 live foetuses and coexisting complete hydatidiform mole.

Case	Hormone Therapy	Gestational Age at Delivery (In Weeks)	No. of Surviving Foetuses	Maternal Complications
1. Sauerbrey et al ⁽⁶⁾	Clomiphene	22	0	Vaginal bleeding, preeclampsia at 22 weeks
2. Ohmichi et al ⁽⁷⁾	hMG + hCG	17	0	Vaginal bleeding, persistent trophoblastic tumour
3. Azuma et al ⁽⁸⁾	hMG + hCG	19	0	Vaginal bleeding
4. Van de Geijn et al ⁽⁹⁾	GIFT	24	0	Vaginal bleeding
5. Shahabi et al ⁽¹⁰⁾	Clomiphene	17	0	Persistent trophoblastic tumour, choriocarcinoma, lung metastasis, hyperthyroidism
6. Shozu et al ⁽¹¹⁾	IVF - ET	15	0	Vaginal bleeding, persistent trophoblastic tumour
7. Higashino et al ⁽¹²⁾	Clomiphene + FSH + hCG	15	0	Preeclampsia, secondary hyperthyroidism, persistent trophoblastic tumour
8. Amr et al ⁽¹³⁾	Clomiphene + hCG	30	1	None
9. Rajesh et al ⁽¹⁴⁾	None	24	0	Vaginal bleeding
10. Malhotra et al ⁽¹⁵⁾	None	21	0	Vaginal bleeding
11. Takagi et al ⁽⁵⁾	hMG + hCG	28	2	Persistent trophoblastic tumour, lung metastasis
12. Bovicelli et al ⁽¹⁶⁾	IVF - ET	31	1	None
13. P. C. Ko et al ⁽¹⁾	IVF - ET	33	2	Preeclampsia
14. Present case	Clomiphene	33	2	None

Table 1. Cases of Triplet Pregnancies consisting of a Complete Hydatidiform Mole and Two Foetuses: Maternal and Foetal Outcomes (1)

hMG = human menopausal gonadotropin; hCG = human chorionic gonadotropin; GIFT = gamete intrafallopian transfer; IVF-ET = in vitro fertilisation and embryo transfer.

The incidence of a complete Hydatidiform mole with two coexisting fetuses was unknown due to the rarity of case reports. It might be misdiagnosed as a growing subchorionic haematoma or a twin gestation along with a partial molar pregnancy. Ovarian medical stimulation due to infertility is one of the major risk factors, which need to be considered. The infertility factor and the importance of pregnancy make treatment decision difficult for the family and medical staff. Gestational Trophoblastic Disease (GTD) is divided into several groups including: (1) Complete/partial Hydatidiform mole; (2) Persistent/invasive gestational trophoblastic neoplasia; (3) Choriocarcinoma; (4) Placental site trophoblastic tumour. It is known that the risk of GTD, distant metastasis and chemotherapeutic treatment increase in a complete molar pregnancy with a coexisting live fetus. GTD incidence was shown to be as high as 55%, if pregnancy was chosen to be continued. Severe clinical symptoms such as vaginal bleeding, pelvic pain and pressure, hyperthyroidism, hyperemesis gravidarum, anaemia and a rapidly growing uterus are what make the patient turn up for a medical evaluation. Symptoms worsen as the number of coexisting fetuses increase. In a molar pregnancy with a single coexisting fetus, full-blown hyperthyroidism occurs in 0.05% and the incidence increases with increased coexisting foetal number. Preterm birth before 28 weeks gestation is estimated to occur in 62% of cases. The rate in more than one coexisting fetus is not known, but estimated to be higher. Spontaneous foetal loss was estimated to occur in 62% of cases. The incidence seems to be higher in cases with more than one coexisting fetus.⁽¹⁷⁾

CONCLUSION

Complete hydatidiform mole with two live fetuses was a rare condition and mostly ends up in poor outcome. This patient had a history of primary infertility and ovulation induction delivered two live babies and a complete mole at 32 weeks of gestation. This case is presented for its rarity and favourable outcome.

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