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CASE REPORT

Viable triplet pregnancy coexisting with a complete molar pregnancy

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Key Clinical Message

This case is extraordinary because it was never before described in English literature. The case describes a long-standing debate about the safety of carrying this pregnancy to term. Some authors are for and some are against. The risks and benefits should be thoroughly reviewed before a decision is made.

Keywords

Artificial reproductive technology, molar pregnancy, triplet pregnancy.

Case Report

A 24-year-old in her third pregnancy, 11 weeks and 3 days gestation, turned for evaluation, following an episode of mild vaginal bleeding, diarrhea, palpitations, and dyspnoea. Her obstetric history includes two previous successful pregnancies stimulated by Pergonal (Serono®) injections. Her medical history was within normal limits. She started using ovarian stimulatory agents after one and a half year of idiopathic infertility and 2 years after her last delivery. She successfully conceived using a 5-day pretreatment of 50 mg clomiphene citrate (TEVA®) and timed intercourse. Upon arrival, her pulse was noted to be 160 beats per minute and blood pressure 117\58. Her temperature was 36.6°C. Her physical examination was within normal limits. Sinus tachycardia with no ischemic changes was diagnosed on the ECG in emergency room. Hemoglobin was 9.8 g\dL, Fibrinogen 307 mg\dL, PT-INR 1.22, TSH 0.09 UIU\mL, FFT3 4.3 ng\mL, FT4 3.77 ng\dL, and β hCG 1002010 international units (IU). Trichorionic triamniotic triplets and an additional dense hyperechoic tissue measuring 9.5 * 6 cm (Fig. 1A) separated from the three sacks which fitted the sonographic description of a complete molar pregnancy was visible on ER ultrasound. Further evaluation was conducted in the maternal fetal medicine (MFM) unit. US specialist visualized three separate sacks and three separate placentas. Crown-rump length was measured and was appropriate for gestational age. In addition, nuchal translucency of all three fetuses was within normal limits and three different pulses were observed. The additional tissue measured 9.5 * 8.2 cm (Fig. 1B), was separate from all three sacks and sonographically appeared as a complete molar pregnancy, which further strengthened the suspected diagnosis of a normal triplet pregnancy along with a complete molar pregnancy. Before further investigations were performed, a chest X-ray was performed and was reassuring. The patient was sent to a tertiary facility for a second opinion. The second opinion correlated exactly with ours. After considering all risks and benefits, she was recommended to terminate the pregnancy. The explanation was the increased risk for persistent trophoblastic disease, early onset preeclampsia, fetal death, and premature labor. After patient consent, she was sent to a tertiary facility for evacuation and β-hCG follow-up. The procedure was performed without any complications and the material obtained was sent for a pathological evaluation. The pathological report indicated a healthy placental material correlating with the live fetuses alongside a molar diseased tissue correlating with a complete mole

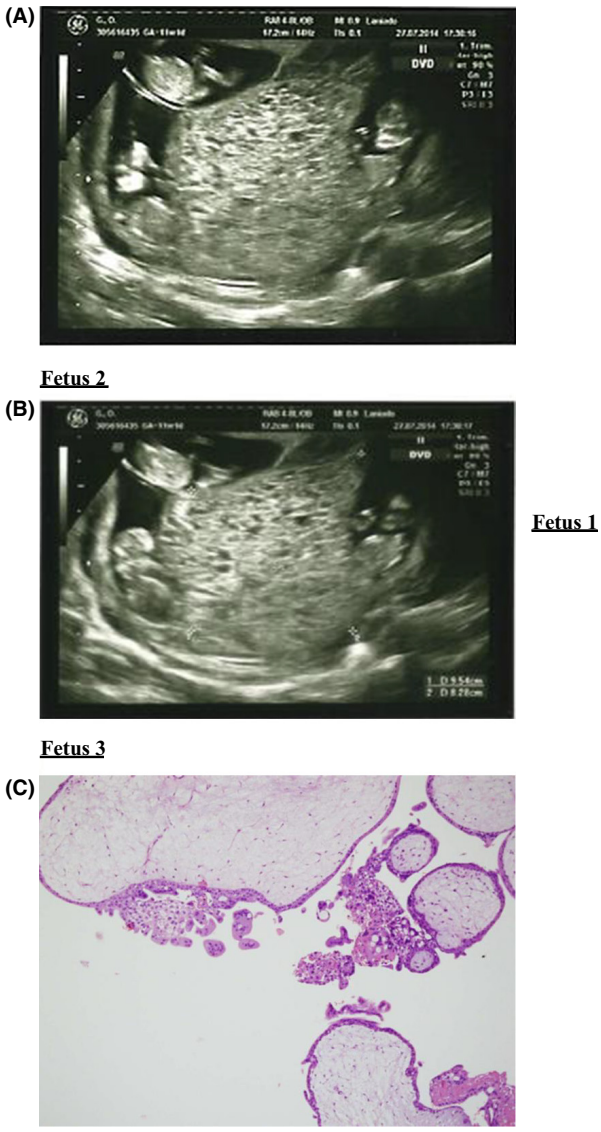

Figure 1. (A) Three fetuses together with a complete molar pregnancy. (B) Measurement of molar pregnancy size. (C) Histology of the intrauterine evacuated material showing cisterns and focal trophoblastic proliferation.

(Fig. 1C). Karyotyping was not performed. Before the procedure, β-hCG level was 1,500,000 IU. After the procedure, β-hCG follow-up was initiated. Due to lack of compliance of the patient, two β-hCG tests were done for 2 months. After 1 month, her β-hCG level was 10,000 IU and after 2 months, it was 150 IU.

Discussion

A complete Hydatiform mole with a coexisting fetus is a rare entity seldom described in English literature. The incidence varies from 1:22,000 to 1:100,000 pregnancies [1]. The incidence of a complete Hydatiform mole with two coexisting fetuses is unknown due to the rarity of case reports. A complete Hydatiform mole along with three coexisting fetuses was never described before in English literature. Diagnosing this kind of case is difficult and requires a high level of suspicion. It might be misdiagnosed as a growing subchorionic hematoma 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 when diagnosing an inconclusive case [2]. The infertility factor and the importance of pregnancy make treatment decision difficult for the family and medical staff. GTD is divided into several groups including: (1) Complete\partial Hydatiform mole; (2) Persistent\invasive gestational trophoblastic neoplasia; (3) Choriocarcinoma; (4) Placental site trophoblastic tumor [3,4]. The incidence of PTD after a singleton complete molar pregnancy is between 15–25% and 3–5% after a singleton partial molar pregnancy. Localized disease usually occurs in 15%, while distant metastasis in 4% of cases [5]. When discussing our case, the risks are much higher than that. It is known that the risk of PTD, distant metastasis, and chemotherapeutic treatment increase in a complete molar pregnancy with a coexisting live fetus. PTD incidence was shown to be as high as 55%, if pregnancy was chosen to be continued in one study [6]. Considering the increased linear risk curve described in literature, the risk in our case is even higher [7]. The exact incidence of a quadruplet pregnancy with three coexisting fetuses isn't known due to rarity of described cases. In contrast, according to Wee et al., gestational age at diagnosis does not change postevacuation outcomes or PTD prognosis. Adding this to our management consideration makes the decision a difficult one. Severe clinical symptoms such as: vaginal bleeding, pelvic pain and pressure, hyperthyroidism, hyperemesis gravidarum, anemia, 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. Full-blown hyperthyroidism almost never occurs in a singleton molar pregnancy even though the subclinical form occurs in 5% of cases. In a molar pregnancy with a single coexisting fetus, full-blown hyperthyroidism occurs in 0.05% and the incidence increases with increased coexisting fetal number. Full-blown disease should be addressed immediately in order to prevent thyroid storm with its complications. The best treatment is termination of pregnancy (TOP), which will treat the source and eliminate the disease. Our patient exhibited hyperthyroid conditions in the form of severe diarrhea and tachycardia of 160 beats per minute. Furthermore, thyroid function tests indicated an acutely active disease. Severe pregnancy complications should also be thoroughly considered and

explained to the patient before making the decision to continue or terminate the pregnancy [8]. Pregnancy complications include early onset preeclampsia, which is estimated to occur in 27% of complete molar pregnancies with a coexisting fetus. No data exists about more than one coexisting fetus, but the incidence is estimated to be higher. Preterm birth before 28 weeks gestation is estimated to occur in 62% of cases. As previously emphasized, the rate in more than one coexisting fetus is not known, but estimated to be higher. Spontaneous fetal loss is estimated to occur in 62% of cases. The incidence seems to be higher in cases with more than one coexisting fetus [9]. As a result of these pregnancy complications along with the estimated high risk for PTD, we recommend TOP with β-hCG follow-up. In contrast to our professional opinion, there is an opposite opinion in the literature advising conservative treatment. This advice is built on the hypothesis that after considering all pregnancy complications, 40% of patients give birth to a live fetus.

There is no data on fetal morbidity. In addition, the reported incidence of PTD and chemotherapy treatment was not significantly higher than in a singleton complete molar pregnancy according to some reports. Added to these factors, gestational age at diagnosis was not different in PTD incidence or disease prognosis [10]. All these risk calculations were considered when the discussion was of a complete molar pregnancy with one or two coexisting fetuses, but in our case, there were three coexisting fetuses. The recommendation of TOP was made due to estimated increased maternal and fetal risks. These risks outweigh the benefits. Pregnancy complications, PTD, fetal and maternal morbidity, and mortality, which were never explored because of the case rarity considered in this case. The subject of complete molar pregnancy coexisting with more than one live fetus should be further investigated and shed some light on the best way of management.

Conflict of Interest

None declared.

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