Successful delivery of a twin pregnancy with complete hydatidiform mole and coexistent live fetus: a case report and review of literature

Neha Sethi1,2, Ann Gee Tan1, Maherah Kamarudin1, Sofiah Sulaiman1

1Department of Obstetrics and Gynaecology, Faculty of Medicine, University Malaya, 50603 Kuala Lumpur, Malaysia
2Correspondence: s_neha26@um.edu.my (Neha Sethi)

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Background: A twin pregnancy consisting of either a complete or partial hydatidiform mole and a fetus is rare. The reported incidence ranges from 1:22,000–100,000 pregnancies, and complete hydatidiform mole with a coexistent fetus (CHMCF) comprises the majority of these cases. The management of CHMCF is controversial, as maternal risk with continuation of the pregnancy should be weighed against fetal survival. Women with CHMCF are at risk of developing preeclampsia, gestational diabetes, hyperthyroidism, antepartum hemorrhage, and gestational trophoblastic neoplasia. Case: We report a case of a healthy 32-year-old woman in her third pregnancy. She presented at 18 weeks gestation with vaginal bleeding and a significantly large uterus relative to the gestational age. Ultrasound showed CHMCF with a beta-hCG value of 398,800 IU/L. After careful discussion with the patient and after considering her options, she elected to continue the pregnancy. She was closely monitored for complications and had no maternal or fetal concerns. An elective cesarean delivery was performed at 32 weeks. A live female infant was delivered together with a normal placenta and a complete mole. The mother and baby were discharged in good condition after 2 days. A histopathological examination of the molar tissue confirmed the CHMCF diagnosis. No finding of gestational trophoblastic neoplasia (GTN) was discovered throughout one-year follow-up. Conclusion: Successful pregnancy outcomes can be achieved in cases of CHMCF. Comprehensive counseling with the patient regarding possible complications is important. Closely monitoring the mother for any complications and performing ongoing fetal surveillance are essential. Delivery should be planned at a tertiary center with good facilities and neonatal support.

Keywords
Complete hydatidiform mole, Twin pregnancy, Coexistent live fetus, Gestational trophoblastic neoplasia

1. Introduction

Luker [1] first described a twin pregnancy consisting of a complete or partial hydatidiform mole in 1914. Over the previous decades, only a few cases were reported. The combination could be a live fetus with a complete hydatidiform mole or a live fetus with a partial hydatidiform mole [2–5]. Therefore, as the prognosis and management of each are different, distinguishing between both possibilities is crucial [6]. The fetus that is accompanied by a partial hydatidiform mole is malformed and usually does not survive past midpregnancy [7–9], pregnancy termination is recommended once the diagnosis is made [10]. Complete hydatidiform mole with a coexistent fetus (CHMCF) can result in a viable fetus that may survive until delivery [2, 3]. However, the management of patients with CHMCF is difficult due to its rarity and complexity [7, 10–12]. Although the fetus in CHMCF can be alive [2, 3], the pregnancy is usually terminated due to consequences that can threaten the lives of both the mother and fetus [2, 3, 13–16]. Prior reports note a high risk for haemorrhage requiring uterine evacuation [17]; however, several case reports have described safe continuation of the pregnancy [18–24]. In this case report, we present a case of CHMCF that resulted in a healthy newborn with no significant maternal complications throughout the pregnancy. In this report, we first describe a case of CHMCF and then provide a review and summary of the entire literature available regarding this rare condition in pregnancy.

2. Case presentation

A healthy 32-year-old woman, who was in her third pregnancy, presented to the University Malaya Medical Centre (UMMC) at 18 weeks gestation for vaginal bleeding. She did not experience abdominal pain, excessive nausea, or vomiting.

This was a planned pregnancy and a spontaneous conception. The patient was at risk for a miscarriage due to bleeding at 11 weeks, at which point she sought advice from a private practitioner. An ultrasound scan (USS) performed at that time revealed a viable fetus, and she was given a revised expected delivery date. No abnormalities were observed at that time.

She had a history of a complete miscarriage at 7 weeks gestation that required surgical intervention 4 years earlier and one uneventful full-term spontaneous vaginal delivery of a healthy baby girl 3 years earlier.
On examination, she did not appear pale and her vitals were stable. The abdomen was soft and non-tender. The fundal height was palpable at 28 weeks gestation.

USS at 18 weeks gestation showed an active fetus with parameters corresponding to the gestational age, and no obvious structural abnormalities were seen. The placenta was posterior and did not cover the cervical os. A large cystic mass measuring 16 × 8 cm with mixed echogenicity and a honeycomb appearance was observed within the uterus, which was separated by a membrane. This led to a diagnosis of a twin pregnancy with a coexistent molar pregnancy. No theca lutein cyst was detected on ultrasound. The patient declined chromosomal analysis to determine the karyotypes of the fetus and mole. No structural anomalies or soft markers were observed that suggested aneuploidy. The hydatidiform molar tissue was distinctly separated from the fetus and placenta.

Magnetic resonance imaging (MRI) was performed to support the diagnosis and delineate the distinct junction between the myometrium and molar tissue. Blood tests revealed normal thyroid function and a beta-human chorionic gonadotropin (hCG) level of 398,800 IU/L.

MRI revealed a well-formed fetus within the amniotic sac occupying the left posteroinferior aspect of the uterine cavity and a well-defined mass measuring 7.0 × 10.3 × 15.0 cm (AP × W × CC) outside the amniotic sac of the fetus occupying the right side of the uterine cavity. Multiple cystic areas were noted within the mass, as was evidence of subacute bleeding over the inferior pole of the lesion. However, no evidence of placental invasion by the mass was observed (Fig. 1).

The patient was informed of the possible complications of continuing the pregnancy, which included persistent vaginal bleeding, gestational trophoblastic neoplasia (GTN), preeclampsia, preterm delivery, and fetal growth restriction. At this time, pregnancy termination was discussed, but she elected to continue the pregnancy.

The pregnancy care plan was outlined, and she was compliant. She remained well, euthyroid, and normotensive throughout the pregnancy. No additional vaginal bleeding was seen after 21 weeks gestation.

The serial hemoglobin measurement, platelet count, thyroid function tests, liver function tests, and coagulation profiles were normal throughout the pregnancy. Oral glucose tolerance tests also ruled out gestational diabetes mellitus. The beta-hCG level at 22 weeks gestation measured 170,400 IU/L, which decreased to 80,385 IU/L after 4 weeks. The beta-hCG level exhibited a decreasing trend after 2 weeks with a value of 46,067 IU/L 1 week before delivery.

Serial fetal surveillance was satisfactory according to the Doppler findings. The molar aspect remained approximately the same size. No sonographic evidence that indicated invasion of the uterine myometrium by the mass was found.

A multidisciplinary team discussion that involved obstetricians, obstetrics anesthetists, neonatalogists and gynaecologic oncologists was arranged, and an elective cesarean section was performed at 32 weeks gestation. Antenatal corticosteroids for fetal lung maturity were administered prior to the cesarean section.

A lower-segment cesarean section was performed with no intraoperative complications. A normal baby girl weighing 1.98 kg was successfully delivered with an APGAR (Appearance, Pulse, Grimace, Activity, and Respiration) score of 6 at 1 minute and 10 at 5 and 10 minutes. The placenta, which was delivered via controlled cord traction, appeared grossly normal and weighed 285 grams (Fig. 2). The mass of the hydatidiform mole containing the vesicular lesion weighed 235 grams and was also delivered (Fig. 2). The operation was uneventful, and an intravenous infusion of 80 IU of oxytocin was immediately administered after delivery.
The baby girl was observed in the neonatal unit for 5 days, was then treated for congenital pneumonia and pathological jaundice, and subsequently discharged in good condition.

The histopathological examination of the molar tissues confirmed the clinical diagnosis of CHMCF. This patient had normal lochia, and a postpartum examination showed satisfactory involution of the uterus. The immediate postdelivery serum beta-hCG level was 5916 IU/L and was subsequently measured to be 123 IU/L, 12 IU/L, and less than 2 IU/L at 1, 2 and 6 weeks postpartum, respectively. Subsequent follow-up serum beta-hCG showed no sign of GTN and the patient remained clinically well up to one year after childbirth.

3. Discussion

The diagnosis for CHMCF is clinically challenging as it may be mistaken for partial hydatidiform mole. Since the management of these two conditions differs, distinguishing between them is important. This is a rare phenomenon [7, 10, 11], and as a result, not many of us are acquainted with this condition. In cases of a partial hydatidiform mole, pregnancy termination is advised, as the fetus is chromosomally abnormal. On the contrary, the coexistent fetus in CHMCF is viable and normal [2, 3].

A systematic search of published literature in English language from January 1990 to December 2020 from PubMed and MEDLINE using the terms “complete hydatidiform mole” or “twin hydatidiform mole”. The search was limited to case reports and case series which involved complete hydatidiform mole coexisting with at least one live fetus. 167 cases were discovered and their findings were summarized in Tables 1 (Ref. [3, 5, 6, 12, 14–102]) and Table 2.

The incidence of CHMCF is predicted to rise in the future, as the ultrasonographic technology used in detecting CHMCF advances and the extensive use of ovulation induction techniques increases [12, 59, 103]. 29.94% (n = 50) of the pregnancies in the present literature review were conceived with the aids of several assisted conception methods (Table 2). Extremes of maternal age (<20 and >40 years old) has been a well-known risk factor associated with complete hydatidiform mole [104–107]. 16 (9.58%) women with CHMCF pregnancies in our literature review were from these two age groups.

CHMCF should be a differential diagnosis when a pregnant woman presents with vaginal bleeding, hyperemesis gravidarum, features of hyperthyroidism, or a uterus larger than expected [2, 4, 14, 97, 103, 108, 109].

With the recent advances in ultrasonography, CHMCF can even be incidentally detected starting late in the first trimester [103, 108]. In this condition, the complete mole typically presents with a classical snowstorm appearance together with the presence of a normal placenta and a viable fetus [4, 7, 12, 103, 110]. Occasionally, a theca lutein cyst is detected on ultrasound due to a significantly elevated level of serum beta-hCG, suggesting a higher probability of CHMCF [45, 103, 109, 111, 112]. Ultrasound is sufficient for a clinical diagnosis. When the physician is experienced, this condition can be diagnosed as soon as the end of the first trimester. In these patients, MRI supports the diagnosis, differentiates it from placental mesenchymal dysplasia, and also assesses invasion of the myometrium by the molar tissue [113–115].

Fetal karyotyping has been advocated to compare the normal chromosome number in CHMCF compared with the triploidy seen in a partial mole [2, 4, 6, 10, 39, 45, 109, 116, 117]. Complete mole is exclusively diploid and paternal in origin, occurring when an “empty” ovum is being fertilized by a single haploid sperm that duplicates (46, XX) or by two haploid sperms (46, XX or 46, XY) [2, 4, 5, 45]. Cytogenetic studies in the literature review have shown that majority (37.13% versus 5.99%) of CHMCF have a 46, XX karyotype (Table 2).

Histopathological examination of the trophoblastic tissue after delivery will confirm the final diagnosis of CHMCF [103, 118].

Immediate pregnancy termination upon diagnosis has typically been recommended in the past due to the potentially fatal complications that can occur if the pregnancy is continued [3, 14, 118]. GTN, which is one of the most serious maternal conditions that can develop in patients with CHMCF [3, 11, 13, 38, 45, 103, 118, 119], has a reported incidence ranging from 19% to 50% [2, 10, 90, 97, 120, 121]. Of the reported cases in the present literature, 32.93% (n = 55) of the CHMCF pregnancies progressed to GTN (Table 3) (Ref. [15–17, 19, 21, 22, 26, 27, 31–33, 38, 41, 42, 44, 53, 55–58, 60, 63, 65–67, 69, 72, 73, 77, 80, 81, 85, 87, 90, 94, 100, 102]). Among them, 15 (27.27%) of the GTN cases have progressively metastasize to distant organs, with lungs being the commonest site of metastasis. 6 (10.91%) women even required hysterectomy to cure from GTN but none of the patient in our literature review died from GTN or its complications. However, it has been demonstrated that the risk of GTN is independent of gestational age, meaning that the risk of GTN in patients who choose conservative management until delivery is the same as that in those who decide to terminate the pregnancy [2, 11, 12, 60, 118, 119]. Therefore, in the recent years, continuation of the pregnancy has become an option [2, 3, 12, 14, 116, 118], provided that the patient has access to a high standard of care under a multidisciplinary team at a tertiary hospital, does not develop any serious uncontrollable complications throughout the pregnancy, and can maintain compliance with regular follow-up during close surveillance [2, 3, 10, 12, 14, 108, 119, 122]. Comprehensive counseling involving obstetricians, gynaecologic oncologists, anesthetists and neonatologists with the couple must be performed, and they need to understand the risk of possible obstetric complications before this major decision is made [9, 10, 14, 38, 116, 118, 119]. The prenatal care included serial beta-hCG, haemoglobin and thyroid function measurements as well as monitoring of the progression of molar mass, theca lutein cysts and fetal growth [34, 41, 44, 56]. Blood pressure and urine protein should be closely evaluated to exclude pre-eclampsia [20, 56]. As with the development of any
## Table 1. CMCF cases from literature review [1990–2020].

| Author (year) | Case Assisted conception | Maternal age (year) | Ultrasound diagnosis (week) | Maternal complications | Molar karyotype | Peak hCG (IU/L) | Fetal outcome | Delivery mode/GA (week) | GTN |
|---------------|--------------------------|---------------------|----------------------------|------------------------|----------------|----------------|--------------|------------------------|------|
| Johnson et al. (2019) [25] | 1 | - | 27 | 16 | VB | 46, XX | 226,910/21 | LB | CS/34 | No |
| Lipi et al. (2020) [26] | 2 | - | 24 | 28 | HELLP syndrome with impending eclampsia | NA | 285,000/28 | LB | CS/33 | Yes |
| Alpay et al. (2020) [27] | 3 | ICSI | 33 | 12 | PE | NA | 425,000/12 | LB | CS/26 | Yes |
| Sheik et al. (2015) [14] | 4 | - | 32 | 13 | VB, TLC | NA | 1,386,570/13 | TOP | 17 | No |
| Raj et al. (2019) [28] | 5 | - | 24 | 13 | VB, HT, PE | NA | NA | LB | CS/24 | No |
| Piura et al. (2008) [29] | 6 | OI | 29 | 9 | VB, PL | NA | 697,930/12 | LB | CS/28 | No |
| Ray et al. (2020) [30] | 7 | OI | 27 | 13 | VB, HG, TLC | NA | 198,880/13 | TOP | 13 | No |
| Imafuku et al. (2018) [31] | 8 | OI | 24 | 12 | VB, HG | 46, XX | 239,100/12 | TOP | 21 | Yes |
| | 9 | - | 27 | 14 | - | 46, XX | 296,052/14 | TOP | 15 | No |
| Sharon et al. (2019) [32] | 10 | IVF | 41 | 11 | - | NA | 353,029/10 | TOP | 14 | No |
| | 11 | OI | 27 | 11 | VB | NA | 1,298,000/11 | TOP | 11 | Yes |
| | 12 | - | 26 | 12 | VB, PE | NA | 3,000,000/12 | TOP | 14 | No |
| Peng et al. (2014) [33] | 13 | OI | 30 | 8 | VB | 46, XY | 1,069,300/8 | TOP | 13 | Yes |
| | 14 | OI | 24 | 9 | VB | NA | 1,425,000/13 | TOP | 13 | Yes |
| | 15 | - | 37 | 10 | VB | 46, XX | 118,200/10 | TOP | 20 | No |
| | 16 | OI | 22 | 11 | VB | 46, XX | 108,200/11 | TOP | 24 | No |
| Rai et al. (2014) [34] | 17 | OI | 25 | 12 | VB, TLC | NA | 374,747/13 | LB | CS/36 | No |
| Altares et al. (1992) [35] | 18 | OI | 22 | 21 | VB, PL | NA | 10,000/6 | SA | 21 | No |
| Albers et al. (2001) [18] | 19 | - | 21 | 28 | - | 46, XX | 53,953/40 | LB | VD/40 | No |
| Bajaj et al. (2014) [12] | 20 | - | 25 | 16 | HT, TLC | NA | 811,780/16 | SA | 22 | No |
| Hyodo et al. (2005) [36] | 21 | - | 30 | 20 | - | 46, XY | 367,747.8/20 | LB | VD/28 | No |
| Gabra et al. (2020) [37] | 22 | - | 21 | 15 | VB, HG | 46, XX | 375,954/15 | TOP | 17 | No |
| Sukxsi et al. (2017) [16] | 23 | - | NA | 19 | VB, PE, HT | 46, XX | NA | TOP | 19 | No |
| | 24 | - | NA | 16 | HT, TLC | 46, XX | NA | TOP | 16 | Yes |
| Ogura et al. (2006) [38] | 25 | - | 27 | 15 | PE, HT | NA | 27,500/16 | TOP | 17 | Yes |
| | 26 | - | 30 | 20 | VB, PP | NA | 5,265/20 | TOP | 21 | No |
| Soysal et al. (1996) [39] | 27 | - | 27 | 14 | VB | 46, XX | 230,000/14 | TOP | 14 | No |
| Aggarwal et al. (2004) [40] | 28 | - | 28 | 20 | VB, HT, HG | 46, XX | 150,000/20 | TOP | 20 | No |
| Dolapcioglu et al. (2009) [5] | 29 | ICSI | 34 | 13 | VB, PH | NA | 198,000/13 | LB | CS/29 | No |
| | 30 | - | 18 | 15 | VB, TLC | NA | 512,000/15 | TOP | 17 | No |
| Miller et al. (1993) [41] | 31 | - | 27 | 16 | VB, HG | 46, XX | 649,456/16 | TOP | 16 | Yes |
| | 32 | - | 30 | 22 | VB | NA | 385,000/22 | LB | VD/38 | No |
| | 33 | - | 32 | 18 | VB, HG, PE | 46, XX | 1,620,000/18 | TOP | 18 | Yes |
| | 34 | - | 33 | 17 | VB, HG | 46, XY | 3,200,000/19 | TOP | 19 | Yes |
| Osada et al. (1995) [42] | 35 | - | 30 | 24 | VB, PP | NA | 478,000/24 | SB | 25 | Yes |
| Author (year)            | Case  | Assisted conception | Maternal age (year) | Ultrasound diagnosis (week) | Maternal complications | Molar karyotype | Peak hCG (IU/L) | Fetal outcome | Delivery mode/GA (week) | GTN |
|-------------------------|-------|---------------------|---------------------|-----------------------------|------------------------|----------------|----------------|--------------|--------------------------|-----|
| Abbi et al. (1999) [43] | 36    | -                   | 26                  | 36                          | VB                     | NA             | 95,000/36      | LB           | CS/37                     | No  |
| Aguilera et al. (2012) [44] | 37    | -                   | 48                  | 14                          | VB, PE, PP, PA         | NA             | 290,000/14     | LB           | CS/34                     | Yes |
| Albayrak et al. (2010) [45] | 38    | -                   | 30                  | 17                          | PL                     | NA             | 69,000/17      | LB           | CS/33                     | No  |
| Barrera et al. (2013) [46] | 39    | -                   | 37                  | 12                          | VB, PH, HT             | NA             | 1,000,000/12   | SA           | 13                       | No  |
| Bhutta et al. (1996) [47] | 40    | -                   | 25                  | 18                          | HG, VB, TLC            | NA             | 618,850/18     | LB           | CS/26                     | No  |
| Buke et al. (2014) [48]  | 41    | -                   | 27                  | 18                          | VB                     | NA             | NA             | SA           | 18                       | No  |
| Chen et al. (2014) [49]  | 42    | -                   | 21                  | 17                          | VB, PP, PA, PL         | NA             | 7,500/17       | LB           | CS/32                     | No  |
| Dalmia et al. (2013) [50] | 43    | -                   | 32                  | 9                           | -                      | NA             | 551,600/10     | TOP          | 14                       | No  |
| Loza et al. (2019) [51]  | 44    | -                   | 20                  | 10                          | VB                     | 46, XX         | NA             | LB           | VD/37                     | No  |
| Dare et al. (1999) [52]  | 45    | -                   | 34                  | 17                          | VB, HT, PL             | NA             | 942,000/17     | LB           | CS/32                     | No  |
| Devall et al. (2006) [53] | 46    | -                   | 30                  | 12                          | PPROM, cord prolapse   | NA             | NA             | LB           | CS/NA                     | No  |
| Ernst et al. (2009) [54] | 47    | -                   | 28                  | 12                          | NA                     | -              | NA             | NA           | SB                       | 27  |
| Ferraz et al. (2013) [55] | 48    | -                   | 29                  | 13                          | NA                     | PIH, PL        | NA             | NA           | CS/30                     | NA  |
| Freis et al. (2016) [56] | 49    | -                   | 32                  | 12                          | NA                     | PL             | NA             | LB           | CS/35                     | NA  |
| Nobuhara et al. (2018) [57] | 50    | -                   | 39                  | 12                          | HT                     | NA             | 1,402,565/14   | TOP          | 14                       | Yes |
| Marcorelles et al. (2005) [19] | 51    | ICSI                | 39                  | 13                          | NA                     | VB, abruptio placenta | NA          | NA           | LB                       | CS/31| No |
| Kashimura et al. (2001) [58] | 52    | -                   | 33                  | 14                          | VB                     | NA             | 647,000/8      | TOP          | 10                       | Yes |
| Montes-de-Oca-Valero et al. (1999) [59] | 53    | IVF                 | 42                  | 9                           | VB                     | NA             | 10,000/32      | LB           | CS/32                     | No  |
| Montes-de-Oca-Valero et al. (1999) [59] | 54    | -                   | 26                  | 12                          | VB                     | 46, XX         | 409,970/14     | TOP          | 15                       | No  |
| Montes-de-Oca-Valero et al. (1999) [59] | 55    | -                   | 25                  | 18                          | VB, TLC                | 46, XX         | 920,000/15     | SA           | 21                       | Yes |
| Montes-de-Oca-Valero et al. (1999) [59] | 56    | -                   | 37                  | 14                          | VB, PE                 | 46, XX         | 840,000/16     | LB           | CS/27                     | No  |
| Montes-de-Oca-Valero et al. (1999) [59] | 57    | -                   | 41                  | 15                          | VB, PE                 | 46, XX         | 1,026/10       | TOP          | 14                       | Yes |
| Moini et al. (2011) [3]  | 58    | IUI                 | 30                  | 13                          | VB                     | 46, XY         | 10,260/10      | TOP          | 14                       | Yes |
| Montes-de-Oca-Valero et al. (1999) [59] | 59    | IVF                 | 41                  | 16                          | VB, PE                 | 46, XX         | 10,260/10      | TOP          | 14                       | Yes |
| Moini et al. (2011) [3]  | 60    | ICSI                | 39                  | 20                          | VB, HG                 | NA             | NA             | SA           | 20                       | Yes |
| Montes-de-Oca-Valero et al. (1999) [59] | 61    | IVF                 | 39                  | 19                          | VB, HG, PP             | NA             | NA             | SA           | 21                       | No  |
| Giorgione et al. (2017) [60] | 62    | -                   | 33                  | 12                          | HT, TLC                | NA             | NA             | TOP          | 13                       | Yes |
| Giorgione et al. (2017) [60] | 63    | -                   | 31                  | 18                          | VB, HT, HG             | NA             | NA             | TOP          | 20                       | No  |
| Giorgione et al. (2017) [60] | 64    | -                   | 39                  | 20                          | PL                     | NA             | NA             | LB           | CS/34                     | Yes |
| Giorgione et al. (2017) [60] | 65    | -                   | 37                  | 19                          | PL                     | NA             | NA             | LB           | CS/37                     | No  |
| Giorgione et al. (2017) [60] | 66    | -                   | 22                  | 16                          | HT, PL                 | NA             | NA             | LB           | CS/26                     | No  |
Table 1. Continued.

| Author (year) | Case | Assisted conception | Maternal age (year) | Ultrasound diagnosis (week) | Maternal complications | Molar karyotype | Peak hCG (IU/L) | Fetal outcome | Delivery mode/GA (week) | GTN |
|---------------|------|---------------------|---------------------|-----------------------------|------------------------|----------------|----------------|----------------|-------------------------|-----|
| 70            | -    | 42                  | 13                  | VB                          | NA                     | NA             | SA             | 13             | Yes                     |     |
| 71            | -    | 24                  | 15                  | VB                          | NA                     | NA             | SA             | 17             | No                      |     |
| 72            | -    | 30                  | 16                  | -                           | NA                     | NA             | ND             | 13             | No                      |     |
| 73            | -    | 29                  | 20                  | VB, PL                      | NA                     | NA             | LB             | CS/30          | No                      |     |
| Singh et al. (2011) [61] | 74 | -                    | 29                  | 12                         | VB, PE                 | NA             | NA             | LB             | CS/36                   | No  |
| Wang et al. (2013) [62] | 75 | NA                   | 25                  | 16                         | VB                      | NA             | >1,000,000/16/16 | TOP           | NA                     |     |
| Winter et al. (1999) [20] | 76 | -                    | 24                  | 18                         | -                      | NA             | 287,000/17     | VB             | CS/36                   | No  |
| Vandenhove et al. (2008) [63] | 77 | IVF                  | 31                  | 15                         | VB, HT                 | 46, XX          | 1,638,200/15   | TOP            | 18                     | Yes |
| Sumigama et al. (2007) [64] | 78 | OI                   | 37                  | 10                         | -                      | 46, XX          | 218,000/10     | TOP            | 10                     | No  |
| Sanchez-Ferrer et al. (2014) [65] | 79 | -                    | 35                  | NA                         | VB, HT, uterine rupture | NA             | 963,971/NA     | TOP            | 15                     | Yes |
| Suri et al. (2009) [66] | 80 | -                    | 32                  | 19                         | VB, PP                 | NA             | 113,324/19     | LB             | CS/28                   | Yes |
| Sanchez-Ferrer et al. (2013) [67] | 81 | -                    | 28                  | 11                         | VB, HT, PIH            | NA             | 939,390/13     | TOP            | 13                     | Yes |
| Slevin et al. (2000) [68] | 82 | -                    | 20                  | 12                         | VB, HG, PE, TLC, HT    | 46, XX          | 1,298,000/17   | TOP            | 17                     | No  |
| Jinno et al. (1994) [69] | 83 | IVF                  | 35                  | 12                         | VB, HT, PIH            | 46, XX          | 1,024,000/14   | ND             | CS/31                   | Yes |
| True et al. (2007) [70] | 84 | -                    | 35                  | 23                         | VB, HT, PL             | NA             | >1,058,000/25/25 | LB             | NA/26                   | No  |
| Hamanoue et al. (2006) [71] | 85 | ICSI                 | 40                  | 7                          | VB, PL                 | 46, XX          | NA             | LB             | CS/33                   | No  |
| Hurteau et al. (1997) [72] | 86 | -                    | 33                  | 9                          | -                      | 46, XX          | 600,000/9      | TOP            | 10                     | Yes |
| Kwon et al. (2002) [73] | 87 | IVF                  | 35                  | 19                         | VB                      | 46, XX          | NA             | LB             | CS/36                   | No  |
| Makrydimas et al. (2002) [74] | 88 | -                    | 28                  | 15                         | VB                      | 46, XX          | NA             | LB             | CS/36                   | No  |
| Peng et al. (2014) [75] | 89 | -                    | 34                  | 20                         | -                      | 46, XX          | 310,277/7/20   | LB             | CS/37                   | Yes |
| Narlawar et al. (2000) [76] | 90 | -                    | 29                  | 22                         | VB, TLC, PL            | 46, XX          | 120,000        | LB             | VD/28                   | No  |
| Rao et al. (2015) [77] | 91 | ICSI                 | 29                  | 16                         | VB, PL                 | NA             | 190,090/12     | LB             | CS/31                   | No  |
| Garcia-Aguayo et al. (1992) [78] | 92 | -                    | 25                  | 14                         | VB                      | NA             | 149,333/14     | TOP            | 14                     | Yes |
| Ozarpaci et al. (2005) [79] | 93 | -                    | 28                  | 16                         | VB                      | NA             | 530,000/16     | TOP            | 16                     | No  |
| Garbin et al. (1995) [80] | 94 | OI                   | 30                  | 23                         | HG, PE, PP             | 46, XX          | 134,600/25     | TOP            | 27                     | No  |
| grenman et al. (1990) [81] | 95 | OI                   | 20                  | 19                         | VB, PE                 | NA             | 800,000/19     | TOP            | 19                     | Yes |
| Harada et al. (1997) [82] | 96 | -                    | 26                  | 15                         | VB, PE                 | NA             | 1,207,600/15   | TOP            | 15                     | Yes |
| He et al. (2014) [83] | 97 | -                    | 20                  | 18                         | -                      | 46, XX          | 121,659/18     | SA             | 22                     | No  |
| Hirose et al. (1999) [84] | 98 | -                    | 23                  | 13                         | VB, HT                 | NA             | 1,024,000/13   | TOP            | 13                     | No  |
| Hsu et al. (1993) [85] | 99 | NA                   | 29                  | 15                         | VB, PE                 | 46, XX          | NA             | TOP            | 15                     | NA  |
| Ishii et al. (1998) [86] | 100| -                    | 30                  | 24                         | VB                      | NA             | NA             | SB             | 25                     | Yes |
| 101 | - | 27                  | 14                  | VB                          | NA                     | NA             | NA             | SA             | 14                     | No  |
| 102 | OI | 35                  | 16                  | VB                          | NA                     | NA             | ND             | CS/22          | Yes                     |     |
| 103 | - | 31                  | 15                  | -                           | NA                     | NA             | NA             | SA             | 15                     | No  |
| 104 | - | 22                  | 11                  | VB                          | NA                     | NA             | LB             | CS/39          | No                      |     |
# Table 1. Continued.

| Author (year) | Case | Assisted conception | Maternal age (year) | Ultrasound diagnosis (week) | Maternal complications | Molar karyotype | Peak hCG (IU/L) | Fetal outcome | Delivery mode/GA (week) | GTN |
|---------------|------|---------------------|---------------------|-----------------------------|------------------------|----------------|-----------------|----------------|------------------------|-----|
| Kaa et al. (1995) [85] | 105 | - | 37 | 22 | - | NA | NA | LB | VD/40 | Yes |
| Koyama et al. (2010) [86] | 112 | - | 15 | - | - | 46, XX | 272,397/15 | TOP | 16 | No |
| Kutuk et al. (2014) [87] | 113 | IUI | 25 | 23 | PE | 46, XX | 100,048/23 | LB | NA/34 | No |
| 114 | - | 23 | 20 | - | 46, XX | 15,774/20 | TOP | 23 | No |
| 115 | - | 24 | 18 | PPROM | 46, XY | 141,720/18 | SA | 21 | No |
| 116 | - | 29 | 12 | HG, HT | 46, XX | 310,270/12 | TOP | 14 | No |
| 117* | OI | 26 | 12 | VB, HG | 46, XY | 125,220/12 | SA | 14 | Yes |
| 118 | - | 26 | 17 | VB, HT, TLC | 46, XX | 310,315/17 | SA | 11 | No |
| 119 | - | 24 | 11 | - | 46, XY | 351,660/11 | SA | 10 | No |
| Lee et al. (2010) [17] | 120 | - | 26 | 13 | VB, PE | 46, XX | 500,000/20 | SA | 10 | No |
| 121 | - | 28 | 14 | VB, HT | 46, XX | 245,000/14 | LB | VD/38 | Yes |
| 122 | IVF | 27 | 14 | VB, HT, PE | NA | >500,000/14 | TOP | 21 | No |
| 123* | IVF | 27 | 13 | VB, HT | NA | 665,105/14 | TOP | 14 | Yes |
| 124 | IVF | 35 | 12 | VB, HG, TLC | NA | 371,000/12 | SA | 18 | No |
| 125 | IVF | 39 | 12 | HT | NA | 1,307,693/13 | TOP | 13 | Yes |
| Wu et al. (2005) [88] | 126 | IVF | 36 | 8 | VB, PL | NA | 685,000/19 | SB | 24 | No |
| Gejin et al. (1992) [89] | 127* | GIFT | 31 | 17 | VB, PL | 46, XX | 327,150/19 | ND/ND | VD/24 | No |
| Niemann et al. (2007) [90] | 128 | NA | 19 | 21 | VB | 46, XX | 182,480/21 | TOP | 23 | No |
| 129 | NA | 22 | 18 | - | NA | NA | TOP | 18 | No |
| 130 | NA | 33 | 18 | VB | 46, XX | 1,142,260/18 | LB | CS/27 | No |
| 131 | NA | 26 | 20 | VB | 46, XY | NA | SA | 20 | Yes |
| 132 | NA | 32 | 9 | VB | 46, XX | 254,880/9 | TOP | 11 | No |
| 133 | NA | 24 | 10 | VB | 46, XX | 180,000/10 | TOP | 14 | No |
| 134 | NA | 26 | 6 | VB | 46, XX | 492,500/6 | TOP | 11 | Yes |
| 135* | NA | 27 | 10 | - | 46, XX | 1,216,888/10 | TOP | 14 | No |
| Azuma et al. (1992) [91] | 136* | OI | 24 | NA | VB | NA | 110,000/18 | SA | 18 | No |
| Author (year) | Case | Maternal age (year) | Ultrasound diagnosis (week) | Maternal complications | Molar karyotype | Peak hCG (IU/L) | Fetal outcome | Delivery mode/GA (week) | GTN |
|--------------|------|---------------------|-----------------------------|------------------------|----------------|----------------|---------------|-----------------------|------|
| Malhotra et al. (2001) [6] | 137* | - | 29 | 16 | VB | NA | 250,000/17 | SA | 21 | No |
| Okumura et al. (2014) [92] | 138 | - | 27 | 15 | PE | NA | >200,000/15 | LB | CS/32 | No |
| Ozumba et al. (1994) [93] | 139 | - | 56 | 20 | VB, PL | NA | NA | SB | 26 | No |
| Shoa et al. (1998) [94] | 140 | GIFT | 31 | 15 | VB | 46, XX | 2,000,000/15 | TOP | 16 | No |
| 141* | IVF | 31 | 12 | VB | 46, XX | 6,400,000/15 | TOP | 15 | Yes |
| Wax et al. (2003) [23] | 142 | IVF | 41 | 10 | - | 46, XX | 179,933/10 | LB | CS/36 | No |
| Kashani et al. (2009) [95] | 143 | ICSI | 29 | 19 | PE | 46, XX | 73,000/19 | SA | 19 | No |
| 144 | - | 19 | NA | VB, PL | NA | NA | LB | NA/35 | No |
| Lambert-Messerlian et al. (2005) [96] | 145 | - | 18 | NA | PL | 46, XY | 176,000 | LB | NA/23 | NA |
| 146 | IVF | 30 | NA | - | 46, XX | 279,000 | LB | CS/28 | No |
| Bovicelli et al. (2004) [97] | 147* | IVF | 32 | 9 | VB | 46, XX | 300,000/24 | LB/SB | CS/31 | No |
| Klatt et al. (2006) [98] | 148 | - | 35 | 19 | VB, PP | 46, XX | 195,575/18 | LB | CS/31 | No |
| Miskovic et al. (2006) [24] | 149 | - | 32 | 18 | - | 46, XX | 199,000/28 | LB | VD/37 | No |
| Cheng et al. (1995) [99] | 150 | IVF | 29 | 15 | VB, PL, PP | 46, XX | 501,808/15 | LB | CS/29 | No |
| Massardier et al. (2009) [15] | 151 | NA | 27 | NA | - | NA | NA | TOP | 17 | No |
| 152 | NA | 28 | NA | - | NA | NA | TOP | 12 | Yes |
| 153 | NA | 37 | NA | HT | NA | NA | LB | CS/27 | No |
| 154 | NA | 43 | NA | - | NA | NA | SA | 12 | Yes |
| 155 | NA | 27 | NA | PE | NA | NA | TOP | 16 | Yes |
| 156 | NA | 31 | NA | VB | NA | NA | TOP | 15 | Yes |
| 157 | NA | 26 | NA | HT | NA | NA | SA | 24 | Yes |
| 158 | NA | 30 | NA | HT | NA | NA | TOP | 14 | No |
| 159 | NA | 21 | NA | PE | NA | NA | SA | 17 | No |
| 160 | NA | 32 | NA | PE | NA | NA | TOP | 16 | Yes |
| 161 | NA | 27 | NA | PPROM | NA | NA | TOP | 22 | No |
| 162 | NA | 37 | NA | - | NA | NA | LB | VD/25 | No |
| 163 | NA | 27 | NA | PE | NA | NA | LB | VD/38 | No |
| 164 | NA | 30 | NA | HT | NA | NA | TOP | 11 | Yes |
| Makary et al. (2010) [100] | 165 | - | 19 | 25 | PE | NA | 228,000/25 | LB | CS/25 | Yes |
| Huang et al. (2014) [101] | 166 | OI | 29 | 12 | VB | 46, XX | NA | TOP | 15 | No |
| Yamada et al. (2008) [102] | 167 | ICSI | 33 | 10 | VB, HG, PE | 46, XX | 774,840/10 | TOP | 16 | Yes |

*, triplet pregnancy involving 2 fetuses; **, quadruplet pregnancy involving 3 fetuses; hCG, human chorionic gonadotropin; GTN, gestational trophoblastic neoplasia; OI, ovulation induction; ICSI, intra-cytoplasmic sperm injection; IVF, in vitro fertilization; GIFT, gamete intra-fallopian transfer; IUI, intra-uterine insemination; VB, vaginal bleeding; HELLP syndrome, Haemolysis, Elevated Liver enzyme and Low Platelet count syndrome; PE, pre- eclampsia; PIH, pregnancy induced hypertension; HT, hyperthyroidism; HG, hyperemesis gravidarum; TLC, theca lutein cysts; PL, preterm labour; PP, placenta praevia; PA, placenta accreta; PPROM, preterm premature rupture of membranes; LB, live birth; TOP, termination of pregnancy; SA, spontaneous abortion before 24 weeks gestation; SB, stillbirth equal or more than 24 weeks gestation; ND, neonatal death; CS, caesarean section; VD, vaginal delivery; NA, not available.
Table 2. Characteristics of 167 reviewed cases of CMCF.

| Characteristics                                    | Frequency, n (%) |
|----------------------------------------------------|------------------|
| **Age group (n = 167)**                            |                  |
| - Less than 20 years old                           | 6 (3.59)         |
| - 20 to 40 years old                               | 148 (88.62)      |
| - 40 years old and above                           | 10 (5.99)        |
| - Not stated                                       | 3 (1.80)         |
| Mean diagnostic gestational weeks on ultrasound ± standard deviation (n = 140) | 15.42 ± 4.60 weeks |
| **Method of conception (n = 167)**                  |                  |
| - Spontaneous                                      | 93 (55.69)       |
| - Ovulation induction                              | 19 (11.38)       |
| - In vitro fertilization                           | 16 (9.58)        |
| - Intra-cytoplasmic sperm injection                | 10 (5.99)        |
| - Intra-uterine insemination                       | 3 (1.80)         |
| - Gamete intra-fallopian transfer                  | 2 (1.20)         |
| - Not stated                                       | 24 (14.37)       |
| **Common maternal complications (n = 167)**         |                  |
| - Vaginal bleeding                                 | 108 (64.67)      |
| - Pre-eclampsia                                    | 28 (16.77)       |
| - Hyperthyroidism                                  | 30 (17.96)       |
| - Hyperemesis gravidarum                           | 18 (10.78)       |
| - Theca lutein cysts                               | 14 (8.38)        |
| - Preterm labour                                   | 23 (13.77)       |
| - Placenta praevia                                 | 9 (5.39)         |
| **Molar karyotype (n = 167)**                       |                  |
| - 46, XX                                           | 62 (37.13)       |
| - 46, XY                                           | 10 (5.99)        |
| - Not stated                                       | 95 (56.89)       |
| **Fetal outcome (n = 177 fetuses)**                 |                  |
| - Termination of pregnancy                         | 75 (42.37)       |
| - Spontaneous abortion                             | 31 (17.51)       |
| - Stillbirth                                        | 8 (4.52)         |
| - Live birth                                       | 58 (32.77)       |
| - Neonatal death                                   | 5 (2.82)         |
| **Mode of delivery for live births and neonatal death (n = 62 pregnancies that resulted in live births + neonatal deaths)** |                  |
| - Caesarean section                                | 42 (67.74)       |
| - Vaginal delivery                                 | 15 (24.19)       |
| - Not stated                                       | 5 (8.06)         |
| **Progression to gestational trophoblastic neoplasia (n = 167)** |                  |
| - Yes                                               | 55 (32.93)       |
| - No                                                | 106 (63.47)      |
| - Not stated                                       | 6 (3.59)         |
| Case | Peak hCG (IU/L) | Initial hCG (IU/L) | Follow-up hCG (IU/L) | Metastasis/invasion | Chemotherapy | Cycles of chemotherapy | Hysterectomy |
|------|----------------|--------------------|----------------------|---------------------|--------------|------------------------|--------------|
| 2    | 285,000/28     | 2,900/6            | 3,500/8              | -                   | Methotrexate  | 9                      | No           |
| 3    | 425,000/12     | 6,000/8            | Plateau              | Lungs               | Methotrexate | 6                      | No           |
| 8    | 239,100/12     | NA                 | NA                   | Lungs               | Methotrexate | 6                      | No           |
| 11   | 1,298,000/11   | High               | High                 | -                   | Methotrexate | NA                     | No           |
| 13   | 1,069,300/8    | High               | High/10              | -                   | 5-FU + KSM   | 5 courses              | No           |
| 14   | 1,425,000/13   | NA                 | NA                   | Invasive mole       | 5-FU + KSM   | 6 courses              | No           |
| 24   | NA             | Normal             | High/18              | -                   | Methotrexate | 3                      | No           |
| 25   | 27,750/16      | NA                 | NA                   | Lungs               | Methotrexate + citrovorum factor | 10 courses | No |
| 31   | 645,456/16     | NA                 | 3900/6               | Right lung          | Methotrexate >actinomycin D >EMA + cisplatin | NA | No |
| 33   | 1,620,000/18   | 56                 | Plateau              | Methotrexate + actinomycin D | 3 | Yes |
| 34   | 3,200,000/19   | 77,000             | Plateau              | Methotrexate + actinomycin D | NA | No |
| 35   | 478,000/24     | Increase           | 41,600/7             | Lungs               | Etoposide    | 6 courses              | No           |
| 37   | 290,000/14     | >1000              | Plateau              | -                   | Methotrexate | NA                     | Yes          |
| 47   | NA             | 37,946/4           | -                    | Invasive mole       | Methotrexate >EMA-CO | NA | No |
| 51   | 1,402,565/14   | 28.74/8            | Plateau              | -                   | NA           | NA                     | No           |
| 52   | NA             | Increase/8         | Increase/32          | -                   | Methotrexate | 2                      | No           |
| 53   | 647,000/8      | 310,000/5          | -                    | Lungs, diagnosed as choriocarcinoma | EMO-CO | 11 | Yes |
| 57   | 920,000/15     | Decrease/1         | Increase/6           | Methotrexate (chemoprophylaxis) >Actinomycin D + etoposide | NA | No |
| 58   | 10,260/13      | 1,680/1            | Left lung            | Methotrexate        | 3 courses    | No                     |
| 61   | NA             | NA                 | NA                   | NA                  | NA           | NA                     | No           |
| 63   | NA             | NA                 | NA                   | NA                  | NA           | NA                     | No           |
| 70   | NA             | NA                 | NA                   | NA                  | NA           | NA                     | No           |
| 77   | 1,638,200/15   | Increase/3         | -                    | -                   | Methotrexate | Modified bagshawe regime | No |
| 79   | 963,971/NA     | 2,832/2            | Plateau              | Lungs, invasive mole | EMA-CO | 5 courses | Yes |
| 80   | 113,324/19     | High               | Plateau              | -                   | Methotrexate >actinomycin D | NA | No |
| 81   | 939,390/13     | High               | Plateau              | Lungs               | Methotrexate | 14 | No |
| 83   | 1,024,000/14   | High               | -                    | Lungs               | Methotrexate + actinomycin D | 6 courses | No |
| 86   | 600,000/9      | Increase/4         | -                    | -                   | Methotrexate >actinomycin D | NA | No |
| Case | Peak hCG (IU/L) /GA (week) | Initial hCG (IU/L) /postpartum (week) | Follow-up hCG (IU/L) /postpartum (week) | Metastasis/invasion | Chemotherapy | Cycles of chemotherapy | Hysterectomy |
|------|-----------------------------|----------------------------------------|------------------------------------------|--------------------|--------------|------------------------|--------------|
| 87 [73] | 321,000/19 | 449/16 | 1,449/20 | - | Methotrexate + citrovorum factor | 1 | No |
| 89 [21] | 510,277.7/20 | 995.3/1 | 268.1/16 | Left lung | Methotrexate + folinate > EMA-CO | 3 > 4 | No |
| 92 [77] | 149,335/14 | 1000/1 | Plateau | Invasive mole | Methotrexate | 2 courses | No |
| 95 [80] | 800,000/19 | Decrease/1 | Increase/4 | Myometrial invasive mole | Methotrexate | 3 courses | No |
| 96 [81] | 1,207,600/15 | Increase/2 | - | Invasive mole | Methotrexate | 7 courses | No |
| 100 [22] | NA | Increase | - | Lungs | Etoposide | 6 | No |
| 102 [22] | NA | Increase | - | - | Methotrexate > MEA | 3 > 5 | No |
| 105 [22] | NA | Decrease | - | Lungs | Methotrexate | NA | No |
| 107 [85] | NA | 9,600/1 | 14,400/2 | - | Curettage | - | No |
| 111 [85] | 700,000/11 | 2000/1 | 12,000/3 | - | Methotrexate | 4 courses | No |
| 117 [87] | 125,220/12 | NA | NA | - | - | - | Yes |
| 121 [17] | 245,000/14 | 510/4 | 760/6 | - | Methotrexate | 2 courses | No |
| 123 [17] | 665,105/14 | 289/5 | 469/7 | - | Methotrexate- citrovorum factor | 1 course | No |
| 125 [17] | 1,307,693/13 | 1004/3 | 1719/4 | Lungs | Methotrexate- citrovorum factor | 7 | No |
| 131 [90] | NA | NA | NA | NA | Methotrexate | 1 | No |
| 134 [90] | 492,500/6 | NA | NA | NA | Methotrexate | 3 | No |
| 141 [94] | 6,400,000/15 | 400/3 | 8,000/4 | Invasive mole | Methotrexate + actinomycin D | 6 courses | No |
| 152 [15] | NA | NA | NA | NA | Methotrexate | NA | No |
| 154 [15] | NA | NA | NA | NA | Methotrexate | NA | No |
| 155 [15] | NA | NA | NA | NA | Methotrexate | NA | No |
| 156 [15] | NA | NA | NA | NA | Methotrexate > multi-agent chemotherapy | 13 > NA | No |
| 157 [15] | NA | NA | NA | NA | Methotrexate | NA | No |
| 160 [15] | NA | NA | NA | NA | Methotrexate | NA | No |
| 164 [15] | NA | NA | NA | NA | Methotrexate | NA | No |
| 165 [100] | 228,000/25 | 301,500/8 | - | Choriocarcinoma metastasis to left kidney and lungs | EMA | 8 | No |
| 167 [102] | 774,840/10 | 22,865/6 | - | Invasive mole | Methotrexate > EMA-CO | 2 courses > 5 courses | Yes |

GA, gestational age; hCG, human chorionic gonadotropin; NA, not available; 5-FU + KSM, 5-fluorouracil + kengshengmycin; EMA, etoposide + methotrexate + actinomycin D; EMA-CO, etoposide + methotrexate + actinomycin D + cyclophosphamide + vincristine.
life-threatening complication, such as heavy vaginal bleeding, severe preeclampsia, GTN, or intrauterine fetal death, [2–4, 11, 14, 19, 45, 97, 109, 118, 119, 123, 124] immediate evacuation is required regardless of the gestational age [2, 14, 19, 108].

Several authors have suggested that, when a less aggressive trophoblast is noted, i.e., a smaller molar component, declining serum hCG levels in the second trimester, and a uterus that is not abnormally large for the gestational age, chances for a successful pregnancy outcome are increased, as in our patient [125]. In addition, she did not develop any serious obstetrical complications.

Although many patients choose a conservative approach, only 35.59% (n = 63) of the pregnancies in the present literature review resulted in successful delivery of a viable live born fetus (Table 2). Many of the pregnancies still ultimately resulted in elective (42.37%, n = 75) and spontaneous (17.51%, n = 31) termination due to obstetric complications, as discussed in many reports (Table 1). According to our literature review, vaginal bleeding (64.67%, n = 108) is the most common maternal complication in CHMCF pregnancy, followed by hyperthyroidism (17.96%, n = 30) and pre-eclampsia (16.77%, n = 28) (Table 2). Lower hCG levels at presentation, later gestational age upon detection, and absence of maternal complications are favorable prognostic factors for better pregnancy outcomes [16, 125].

Cesarean section (67.74%, n = 42) is the recommended mode of delivery in patients with CHMCF, and delivery should be performed by a dedicated team of experts [9, 119]. Intensive neonatal care must be accessible since the newborns in these cases are usually very premature [119]. No consensus on the optimal gestation time for delivery has been established, but in our patient, delivery was scheduled for between 32 and 34 weeks gestation. However, a cesarean section was performed at 32 weeks gestation due to a plateau of fetal growth and because the patient had also complained of uterine contractions. Regular monitoring of the hCG levels throughout the pregnancy and postpartum period is necessary to detect GTN [116, 119, 122]. Furthermore, the patient should be treated with chemotherapy [11, 13, 38] when GTN is suspected.

In conclusion, obstetricians should bear in mind that CHMCF can be one of the possible diagnosis when performing an ultrasound in the early gestational period in women with persistent vaginal bleeding and those with excessive symptoms during early pregnancy. However, the management of this condition remains controversial. Here we report a successful pregnancy outcome despite the late presentation to a tertiary center. However, further studies are warranted to evaluate the most appropriate management strategy for these patients.

Abbreviations

CHMCF, Complete Hydatidiform Mole with Coexistent Fetus; CS, Caesarean Section; EMA, Etoposide + Methotrexate + Actinomycin D; EMA-CO, Etoposide + Methotrexate + Actinomycin D + Cyclophosphamide + Vincristine; GA, Gestational Age; 5-FU + KSM, 5-Fluorouracil + Kensorhengycin; GIFT, Gamete Intra-Fallopian Transfer; GTN, Gestational Trophoblastic Neoplasia; hCG, Human Chorionic Gonadotropin; HELLP Syndrome, Haemolysis, Elevated Liver Enzyme And Low Platelet Count Syndrome; HG, Hyperemesis Gravidarum; HT, Hyperthyroidism; ICSI, Intracytoplasmic Sperm Injection; IUI, Intra-Uterine Insemination; IVF, In Vitro Fertilization; LB, Live Birth; MRI, Magnetic Resonance Imaging; ND, Neonatal Death; OI, Ovulation Induction; PA, Placenta Accreta; PE, Pre-Eclampsia; PIH, Pregnancy Induced Hypertension; PL, Preternum Labour; PP, Placenta Praevia; PROM, Preterm Premature Rupture of Membranes; SA, Spontaneous Abortion Before 24 Weeks Gestation; SB, Stillbirth Equal Or More Than 24 Weeks Gestation; TLC, Theca Lutein Cysts; TOP, Termination Of Pregnancy; UMMC, University of Malaya Medical Centre; USS, Ultrasound Scan; VB, Vaginal Bleeding; VD, Vaginal Delivery.

Author contributions

NS prepared the manuscript and is responsible for the overall content as guarantor. SS, MK, AGT reviewed the manuscript. All authors read and approved the final manuscript.

Ethics approval and consent to participate

Written informed consent was obtained from the patient for the publication of this case report. Ethical approval is waived for the case report.

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Conflict of interest

The authors declare no conflict of interest.

References

[1] Luker G. Twin pregnancy; hydatidiform mole associated with normal ovum; abortion at four months. Proceedings of the Royal Society of Medicine. 1914; 7: 113.
[2] Matsui H, Sekiya S, Hando T, Wake N, Tomoda Y. Hydatidiform mole coexistent with a twin live fetus: a national collaborative study in Japan. Human Reproduction. 2000; 15: 608–611.
[3] Moini A, Ahmadi F, Eslami B, Zafarani F. Dizygotic twin pregnancy with a complete hydatidiform mole and a coexisting viable fetus. Iranian Journal of Radiology. 2011; 8: 249–252.
Johnston KM, Steele EK, Magee SE. Twin pregnancy with a living fetus and coexisting complete hydatidiform mole. The Ulster Medical Journal. 2000; 69: 168–170.

Dolapcioglu K, Gungoren A, Hakverdi S, Hakverdi AU, Egilmez E. Twin pregnancy with a complete hydatidiform mole and coexisting live fetus: two case reports and review of the literature. Archives of Gynecology and Obstetrics. 2009; 279: 431–436.

Malhotra N, Deka D, Takkar D, Kochar S, Goel S, Sharma MC. Hydatidiform mole with coexisting live fetus in dichorionic twin gestation. European Journal of Obstetrics, Gynecology, and Reproductive Biology. 2001; 94: 301–303.

Odedra D, MacEachern K, Elit L, Mohamed S, McCreaddy E, DeFrance B, et al. Twin pregnancy with metastatic complete molar pregnancy and coexisting live fetus. Radiology Case Reports. 2020; 15: 195–200.

Jauniaux E, Brown R, Snijders RJ, Noble P, Nicolaides KH. Early prenatal diagnosis of triploidy. American Journal of Obstetrics and Gynecology. 1997; 176: 550–554.

Zeng C, Chen Y, Zhao L, Wan B. Partial hydatidiform mole and coexistent live fetus: a case report and review of the literature. Open Med. 2019; 14: 843–846.

Vaisbuch E, Ben-Arie A, Dgani R, Perlman S, Sokolovsky N, Haggay Z. Twin pregnancy consisting of a complete hydatidiform mole and coexistent fetus: report of two cases and review of literature. Gynecologic Oncology. 2005; 98: 19–23.

Sebire NJ, Foskett M, Paradinas FJ, Fisher RA, Francis RJ, Short D, et al. Outcome of twin pregnancies with complete hydatidiform mole and healthy co-twin. Lancet. 2002; 359: 2165–2166.

Bajaj SK, Misra R, Gupta R, Nisha B, Thukral BB. Complete hydatidiform mole with coexisting twin fetus: usefulness of MRI in management planning. Journal of Obstetrics and Gynaecology of India. 2014; 64: 9–13.

Fishman DA, Padilla LA, Keh P, Cohen L, Frederiksen M, Lurain JR. Management of twin pregnancies consisting of a complete hydatidiform mole and normal fetus. Obstetrics and Gynecology. 1998; 91: 546–550.

Sheik S, Al-Riyami N, Mathew NR, Al-Sukaiti R, Qureshi A, Mathew M. Twin pregnancy with a complete hydatidiform mole and a coexisting live fetus: rare entity. Sultan Qaboos University Medical Journal. 2015; 15: e550–e553.

Massardier J, Gollier F, Journet D, Frappart L, Zalaquett M, Schott MG, Gonzalez MG, Bullock HN, Hill MG. Cell-Free DNA testing for prenatal diagnosis of triploidy. Gynecologic Oncology Reports. 2020; 31: 100519.

Zilberman Sharon N, Melzer Y, Maymon R. Is a complete hydatidiform mole and a co-existing normal fetus an iatrogenic effect? The Israel Medical Association Journal. 2019; 21: 653–657.

Peng M, Li L, Zheng J, Ding Y, Yu L, Huang J. Termination of twin pregnancies with hydatidiform moles: a case series of four patients. Iranian Journal of Public Health. 2014; 43: 1000–1006.

Rai L, Shirrip H, Guruvare S, Prashanth A, Mundkur A. Twin pregnancy with hydatidiform mole and co-existent live fetus: lessons learnt. The Malaysian Journal of Medical Sciences. 2014; 21: 61–64.

Hyodo M, Samura O, Miharu N, Ohama K, Kudo Y. Molar DNA in maternal serum in a case of 46,XY heterozygous complete hydatidiform mole coexisting with a 46,XX live twin fetus. Clinical Chemistry. 2005; 51: 676–677.

Ogura T, Katoh H, Satoh S, Tsukimori K, Hirakawa T, Wake N, et al. Complete mole coexistent with a twin fetus. Journal of Obstetrics and Gynaecology Research. 2006; 32: 593–601.

Soysal M, Kara S, Ekici E. Twin pregnancy with a fetus and a complete hydatidiform mole. Archives of Gynecology and Obstetrics. 1996; 259: 41–44.

Aggarwal N, Suri V, Deo ND, Malhotra S, Vasishtha K. Twin pregnancy with a complete hydatidiform mole and co-existing fetus. Acta Obstetricia Et Gynecologica Scandinavica. 2004; 83: 604.
Miller D, Jackson R, Ehlen T, McMurtrie E. Complete hydatidiform mole coexisting with a twin live fetus: clinical course of four cases with complete cytogenetic analysis. Gynecologic Oncology. 1993; 50: 119–123.

Osada H, Itsuka Y, Matsui H, Sekiya S. A complete hydatidiform mole coexisting with a normal fetus was confirmed by variable number tandem repeat (VNTR) polymorphism analysis using polymerase chain reaction. Gynecologic Oncology. 1995; 56: 90–93.

Abbi M, Kripiani A, Uppal R, Takkar D. Term twin pregnancy with complete hydatidiform mole and a normal fetus. Archives of Gynecology and Obstetrics. 1999; 262: 189–191.

Aguilera M, Raus F, Ghebre R, Ramiz K. Complete hydatidiform mole presenting as a placenta accreta in a twin pregnancy with a coexisting normal fetus: case report. Case Reports in Obstetrics and Gynecology. 2012; 2012: 405085.

Albayrak M, Ozer A, Demir OF, Ozer S, Erkaya S. Complete mole coexisting with a twin fetus. Archives of Gynecology and Obstetrics. 2010; 281: 119–122.

Barrera JR, Sandoval MAS, Quiva LQ, Paz-Pacheco E. The interplay of Graves' disease and twin molar pregnancy. BMJ Case Reports. 2013; 2013: bcr20130300604.

Bhutta SZ. Twin pregnancy with complete hydatidiform mole and coexistent foetus. Journal of Pakistan Medical Association. 1996; 46: 180–181.

Büke B, Topçu HO, Bulgu E, Eminov E, Kazandi M. Complete hydatidiform mole presenting as placenta previa in a twin pregnancy with a coexisting normal foetus: case report. Journal of the Turkish German Gynecological Association. 2014; 15: 256–258.

Chen C, Ko T, Chen C, Wang T, Chen S, Kuo Y, et al. First-trimester molecular diagnosis of complete hydatidiform mole associated with dizygotic twin pregnancy conceived by in vitro fertilization. Taiwanese Journal of Obstetrics & Gynecology. 2014; 53: 572–578.

Dalmia S, Sivakumar K, Kathirvel R, Phillips K, Coady AM. Twin pregnancy with a complete hydatidiform mole and co-existing healthy fetus: unusual case of complete resorption of molar pregnancy. Journal of Obstetrics and Gynaecology. 2013; 33: 312–313.

Loza AJ, Fang YMV. Complete molar pregnancy coexisting with a normal fetus in the third trimester. American Journal of Obstetrics and Gynecology. 2019; 220: 600–601.

Dare FO, Adelusola A, Adetiloye V, Makinde ON, Orji E. Twin pregnancy involving complete hydatidiform mole. Journal of Obstetrics and Gynaecology. 1999; 19: 318–319.

Devall A, Thompson F, Taylor AA, Pakarian F. Early diagnosis of a complete hydatidiform mole in a twin pregnancy with a viable fetus. Journal of Obstetrics and Gynaecology. 2006; 26: 169–171.

Ernst LM, Chou D, Parry S. Fetal thrombotic vasculopathy in twin placentas with complete hydatidiform mole. Pediatric and Developmental Pathology. 2009; 12: 63–67.

Ferraz TJSDM, Bartosch CMM, Ramalho CMA, Carvalho FAGD, Carvalho BCCLD, Brando OGBC, et al. Complete mole in a dichorionic twin pregnancy after intracytoplasmic sperm injection. Revista Brasileira De Ginecologia E Obstetricia. 2013; 35: 39–43.

Freis A, Elässer M, Sohn C, Fluhir H. Twin pregnancy with one fetus and one complete mole—a case report. Geburtshilfe Frauenheilk. 2016; 76: 819–822.

Nobuhara I, Harada N, Haruta N, Higashiyura Y, Watanabe H, Watanabe S, et al. Multiple metastatic gestational trophoblastic disease after a twin pregnancy with complete hydatidiform mole and coexisting fetus, following assisted reproductive technology: case report and literature review. Taiwanese Journal of Obstetrics & Gynecology. 2018; 57: 588–593.

Kashimura Y, Tanaka M, Harada N, Shinmoto M, Morishita T, Morishita H, et al. Twin pregnancy consisting of 46, XY heterozygous complete mole coexisting with a live fetus. Placenta. 2001; 22: 323–327.

Montes-de-Oca-Valero F, Macara L, Shaker A. Twin pregnancy with a complete hydatidiform mole and co-existing fetus following in-vitro fertilization: case report. Human Reproduction. 1999; 14: 2905–2907.

Giorgione V, Cavorotto G, Cormio G, Valsecchi L, Vimercatti A, De Gennaro A, et al. Prenatal diagnosis of twin pregnancies with complete hydatidiform mole and coexistent normal fetus: a series of 13 cases. Gynecologic and Obstetric Investigation. 2017; 82: 404–409.

Singh M, Shahzad N, Emovon E. Twin pregnancy with complete hydatidiform mole and co-existent viable fetus. Journal of Obstetrics and Gynaecology. 2011; 31: 767–768.

Wang PS, Horro MM. Twin pregnancy with complete hydatidiform mole and normal coexisting fetus. Ultrasound Quarterly. 2013; 29: 219–220.

Vandenhove M, Amant F, van Schoubroek D, Cannie M, Dy markowski S, Hansens M. Complete hydatidiform mole with co-existing healthy fetus: a case report. The Journal of Maternal-Fetal & Neonatal Medicine. 2008; 21: 341–344.

Sugimama S, Itakura A, Yamamoto T, Nagasaka T, Yamamoto E, Ino K, et al. Genetically identified complete hydatidiform mole coexisting with a live twin fetus: comparison with conventional diagnosis. Gynecologic and Obstetric Investigation. 2007; 64: 228–231.

Sánchez-Ferrer ML, Hernández-Martínez F, Machado-Linde F, Ferri B, Carbonel P, Nieto-Díaz A. Uterine rupture in twin pregnancy with normal fetus and complete hydatidiform mole. Gynecologic and Obstetric Investigation. 2014; 77: 127–133.

Suri S, Davies M, Jauinias E. Twin pregnancy presenting as a praevia complete hydatidiform mole and coexisting fetus complicated by a placental abcess. Fetal Diagnosis and Therapy. 2009; 26: 181–184.

Sánchez-Ferrer ML, Machado-Linde F, Martínez-Espejo Cerezo A, Peñalver Parres C, Ferri B, López-Exposito I, et al. Management of a dichorionic twin pregnancy with a normal fetus and an androgenetic diploid complete hydatidiform mole. Fetal Diagnosis and Therapy. 2013; 33: 194–200.

Slevin J, Gleeson R, McKenna P. A rare twin pregnancy: a normal fetus and a complete hydatidiform mole. Journal of Obstetrics and Gynaecology. 2000; 20: 319–320.

Jinno M, Ubukata Y, Hanui M, Satou M, Yoshimura Y, Nakamura Y. Hydatidiform mole with a surviving coexistent fetus following in vitro fertilization. Human Reproduction. 1994; 9: 1770–1772.

True DK, Thomsett M, Lilley H, Chitturi S, Cincotta R, Morton A, et al. Twin pregnancy with a coexisting hydatidiform mole and liveborn infant: complicated by maternal hyperthyroidism and neonatal hypothyroidism. Journal of Paediatrics and Child Health. 2007; 43: 646–648.

Hamanoue H, Umezlu N, Okuda M, Harada N, Ohata T, Sakai H, et al. Complete hydatidiform mole and normal live birth following intra-uterine sperm injection. Journal of Human Genetics. 2006; 51: 477–479.

Hurteau JA, Roth LM, Schilder JM, Sumners J. Complete hydatidiform mole coexisting with a live twin fetus: clinical course. Gynecologic Oncology. 1997; 66: 156–159.

Kwon H, Park E, Kim S, Chae H, Won H, Kim C, et al. A case of twin pregnancy with complete hydatidiform mole and coexisting fetus following IVF-ET. Journal of Assisted Reproduction and Genetics. 2002; 19: 144–148.

Makrydimas G, Sebire NJ, Thornton SE, Zagorianakou N, Lolis D, Fisher RA. Complete hydatidiform mole and normal live birth: a novel case of confined placental mosaicism. Human Reproduction. 2002; 17: 2459–2463.

Narlawr RS, Shah J, Patkar D. Images in radiology: complete hydatidiform mole with live pregnancy in a twin gestation. Journal of Postgraduate Medicine. 2000; 46: 291–292.

Rao A, Dafe K, Padmashri G, Rao D, Sivakumar N. Pregnancy outcome with coexisting mole after intrauterine sperm injection: a case series. Journal of Human Reproductive Sciences. 2015; 8: 178.
García-Aguayo FJ, Menargues Irles MA. Evolution of diamniotic-dichorionic pregnancy into complete hydatidiform mole and normal fetus. Journal of Clinical Ultrasound. 1992; 20: 604–607.

Ozarpaci C, Yalcin S, Gürgöz B, Ceylan S, Cakar Y. Complete hydatidiform mole with coexistent live fetus in dichorionic twin gestation. Archives of Gynecology and Obstetrics. 2005; 271: 270–273.

Garbin O, Favre R, Weber P, Arbogast E, Gasser B. How to deal with a rare entity: the coexistence of a complete mole and a healthy egg in a twin pregnancy? Fetal Diagnosis and Therapy. 1995; 10: 337–342.

Grenman SE, Salmi T, Meurman L. Post-molar trophoblastic disease following coexisting molar pregnancy and living fetus subsequent to clomiphene citrate therapy. International Journal of Gynaecology and Obstetrics. 1990; 32: 381–385.

Harada I, Tsutsui O, Takai Y, Iida T, Sakai M, Yoshikawa H, et al. DNA polymorphism analysis of a case of complete hydatidiform mole coexisting with a fetus. Human Reproduction. 1997; 12: 2563–2566.

He Y, Li F, He X, Qi Z, Zhang Y, Li D. Rapid prenatal diagnosis of complete mole with co-existing twin by QF-PCR analysis. Journal of Obstetrics and Gynaecology. 2015; 35: 526–544.

Hirose M, Kimura T, Mitsuuno N, Wakah S, Takakura K, Fujita J, et al. DNA flow cytometric quantification and DNA polymorphism analysis in the case of a complete mole with a coexisting fetus. Journal of Assisted Reproduction and Genetics. 1999; 16: 263–267.

Hsu CC, McConnell J, Ko TM, Braude PR. Twin pregnancy consisting of a complete hydatidiform mole and a fetus: genetic origin determined by DNA typing. British Journal of Obstetrics and Gynaecology. 1993; 100: 867–869.

Kaa CAVD, Robben JCM, Hopman AHN, Hanselaer AGJM, Voois GP. Complete hydatidiform mole in twin pregnancy: differentiation of complete mole with interphase cytogenetic and DNA cytometric analyses on paraffin embedded tissues. Histopathology. 1995; 26: 123–129.

Koyama S, Tomimatsu T, Sawada K, Kanagawa T, Isobe A, Kinugasa Y, et al. A case of complete hydatidiform mole with coexistent fetus: conclusive diagnosis of androgenesis of the molar placenta by variation of paternal acrocentric short arm. American Journal of Perinatology. 2010; 27: 143–149.

Kutuk MS, Ozgun MT, Dolanbey M, Batukan C, Uludag S, Basbug M. Sonographic findings and perinatal outcome of multiple pregnancies associating a complete hydatidiform mole and a live fetus: a case series. Journal of Clinical Ultrasound. 2014; 42: 465–471.

Wu T, Shen S, Chang S, Chang C, Guo W. Magnetic resonance experience of a twin pregnancy with a normal fetus and hydatidiform mole: a case report. Journal of Computer Assisted Tomography. 2005; 29: 415–417.

van de Geijn EJ, Wedema CA, Hemioka DJ, Schutte MF, ten Velden JJ. Hydatidiform mole with coexisting twin pregnancy after gamete intra-fallopian transfer. Human Reproduction. 1992; 7: 568–572.

Niemann I, Sunde L, Petersen IK. Evaluation of the risk of persistent trophoblastic disease after twin pregnancy with diploid hydatidiform mole and coexisting normal fetus. American Journal of Obstetrics and Gynecology. 2007; 197: 45.e1–45.e5.

Azuma C, Saji F, Takemura M, Ohashi K, Kimura T, Miyake A, et al. Triplet pregnancy involving complete hydatidiform mole and two fetuses: genetic analysis by deoxyribonucleic acid fingerprint. American Journal of Obstetrics and Gynecology. 1992; 166: 664–667.

Okumura M, Fushida K, Francisco RPV, Schultz R, Zugmaier M. Massive necrosis of a complete hydatidiform mole in a twin pregnancy with a surviving coexistent fetus. Journal of Ultrasound in Medicine. 2014; 33: 177–179.

Ozumba BC, Ofodile A. Twin pregnancy involving complete hydatidiform mole and partial mole after five years of amenorrhea. European Journal of Obstetrics & Gynecology and Reproductive Biology. 1994; 53: 217–218.

[77] Shoumu Z, Akimoto K, Kasai T, Inoue M, Michikura Y. Hydatidiform moles associated with multiple gestations after assisted reproduction: diagnosis by analysis of DNA fingerprint. Molecular Human Reproduction. 1998; 4: 877–880.

[78] Kashani, E. and Boustan, P. and Roshandel, G. and Roshandel D. Case report molar pregnancy and co-existent foetuses: a report of two cases. Journal of Clinical and Diagnostic Research. 2009; 1: 1334–1337.

[79] Lambert-Messerlian G, Pinar H, Rubin LP, De Paepe ME, Tantravahi U, Steinhoff MM, et al. Second-trimester maternal serum markers in twin pregnancy with complete mole: report of 2 cases. Pediatric and Developmental Pathology. 2005; 8: 230–234.

[80] Boivicelli L, Ghiti T, Pilu G, Farina A, Savelli L, Simonazzi G, et al. Pre-natal diagnosis of a complete mole coexisting with a dichorionic twin pregnancy: case report. Human Reproduction. 2004; 19: 1231–1234.

[81] Klett TE, Franciosi RA, Cruikshank DP. Normal fetus with a twin presenting as both a complete hydatidiform mole and placenta previa. Obstetrics and Gynecology. 2006; 107: 527–530.

[82] Cheng PJ, Chang FH, Liang CC, Chang TC, Soong YK, Hsueh C. A twin pregnancy with a hydatidiform mole and an alive, coexistent baby after in vitro fertilization and embryo transfer. Journal of Assisted Reproduction and Genetics. 1995; 12: 389–392.

[83] Makary R, Mohammedi A, Rosa M, Shuja S. Twin gestation with complete hydatidiform mole and a coexisting live fetus: case report and brief review of literature. Obstetric Medicine. 2010; 3: 30–32.

[84] Huang X, Liang J, Huang Y, Huang J. Complete hydatidiform mole and a coexistent fetus following ovulation induction in a patient with Sheehan’s syndrome: a first case report and review of literature. Archives of Gynecology and Obstetrics. 2014; 289: 1145–1150.

[85] Yamada T, Matsuda T, Kudo M, Yamada T, Moriwaki M, Nishi S, et al. Complete hydatidiform mole with coexisting dichorionic diamniotic twins following testicular sperm extraction and intracytoplasmic sperm injection. Journal of Obstetrics and Gynaecology Research. 2008; 34: 121–124.

[86] Juainias E. Ultrasound diagnosis and follow-up of gestational trophoblastic disease. Ultrasound in Obstetrics and Gynaecology. 1998; 11: 367–377.

[87] Simms-Stewart D, McDonald G, Fletcher H, Bromfield M, Williams N, Bambury I, et al. A review of molar pregnancy at the university hospital of the West Indies over a 16-year period. Journal of Obstetrics and Gynaecology. 2013; 33: 298–300.

[88] Muliosa O, Roberts DJ, Sengupta ES, Agaba E, Laffita D, Tobias T, et al. Prevalence and Risk Factors Associated with Hydatidiform Mole among Patients Undergoing Uterine Evacuation at Mbarara Regional Referral Hospital. Obstetrics and Gynecology International. 2018; 2018: 9561413.

[89] Sebire NJ, Foskett M, Fisher RA, Rees H, Seckl M, Newlands E. Risk of partial and complete hydatidiform mole pregnancy in relation to maternal age. BJOG: An International Journal of Obstetrics and Gynaecology. 2002; 109: 99–102.

[90] Oikonomoumis P, Pergialiotis B, Pitsouni E, Natsis S, Lagkadis A, Giannakopoulou K. Repetitive complete molar pregnancy in a 54-year-old patient in a time distance of eighteen years from the first incident: case report and mini review. Case Reports in Medicine. 2011; 2011: 1–4.

[91] Sindiani A, Obeidat B, Alshdaifat E. Successful management of the first case of a metastasized complete mole in form of twin pregnancy in Jordan. American Journal of Case Reports. 2020; 21: 1–5.

[92] Woo L, Juainias E. Prenatal diagnosis and management of twin pregnancies complicated by a co-existing molar pregnancy. Prenatal Diagnosis. 2005; 25: 772–776.

[93] Sauerbrei EE, Salem S, Fayle B. Coexistent hydatidiform mole and live fetus in the second trimester: an ultrasound study. Radiology. 1980; 135: 415–417.

[94] Steller MA, Genest DR, Bernstein MR, Lage JM, Goldstein DP, Berkowitz RS. Natural history of twin pregnancy with complete...
hydatidiform mole and coexisting fetus. Obstetrics and Gynecology. 1994; 83: 35–42.

[112] Lurain JR. Gestational trophoblastic disease I: epidemiology, pathology, clinical presentation and diagnosis of gestational trophoblastic disease, and management of hydatidiform mole. American Journal of Obstetrics and Gynecology. 2010; 203: 531–539.

[113] Linn RL, Minturn L, Yee LM, Maniar K, Zhang Y, Fritsch MK, et al. Placental mesenchymal dysplasia without fetal development in a twin gestation: a case report and review of the spectrum of androgenetic biparental mosaicism. Pediatric and Developmental Pathology. 2015; 18: 146–154.

[114] Surti U, Hill LM, Dunn J, Prosen T, Hoffner L. Twin pregnancy with a chimeric androgenetic and biparental placenta in one twin displaying placental mesenchymal dysplasia phenotype. Prenatal Diagnosis. 2005; 25: 1048–1056.

[115] Starikov R, Goldman R, Dizon DS, Kostadinov S, Carr S. Placental mesenchymal dysplasia presenting as a twin gestation with complete molar pregnancy. Obstetrics & Gynecology. 2011; 118: 445–449.

[116] Deopty J. Complete hydatidiform mole with a twin pregnancy at 26 weeks: a rare obstetric complication. Autopsy & Case Reports. 2019; 9: e2019108.

[117] Renard N. Aggressive complete hydatidiform mole coexistent with a normal fetus during pregnancy: is there a correlation between outcome, and serum HCG levels? A report on 2 cases and review of the literature. Obstetrics and Gynecology Cases—Reviews. 2016; 3: 1–7.

[118] Vimercati A, de Gennaro AC, Cobuzzi I, Grassi S, Abruzzese M, Fascilla FD, et al. Two cases of complete hydatidiform mole and coexistent live fetus. Journal of Prenatal Medicine. 2013; 7: 1–4.

[119] Zilberman Sharon N, Maymon R, Melcer Y, Jauniaux E. Obstetric outcomes of twin pregnancies presenting with a complete hydatidiform mole and coexistent normal fetus: a systematic review and meta-analysis. BJOG: An International Journal of Obstetrics & Gynaecology. 2020; 127: 1450–1457.

[120] Sebire NJ. Prenatal diagnosis and management of twin pregnancies complicated by a co-existing molar pregnancy. Prenatal Diagnosis. 2006; 26: 373.

[121] Rohilla M, Singh P, Kaur J, Jain V, Gupta N, Prasad GRV. Individualistic approach to the management of complete hydatidiform mole with coexisting live fetus. European Journal of Obstetrics, Gynecology, and Reproductive Biology. 2015; 191: 39–42.

[122] Atuk FA, Basuni JBM. Molar pregnancy with normal viable fetus presenting with severe pre-eclampsia: a case report. Journal of Medical Case Reports. 2018; 12: 140.

[123] Yarandi F, Eftekhari Z, Izadi-Mood N, Elahi-Panah Z, Shojaei H. Twin pregnancy with hydatidiform mole and coexisting fetus: report of three cases and review of literature. Journal of Family and Reproductive Health. 2009; 3: 31–34.

[124] Lin LH, Maesta I, Braga A, Sun SY, Fushida K, Francisco RPV, et al. Multiple pregnancies with complete mole and coexisting normal fetus in North and South America: a retrospective multicenter cohort and literature review. Gynecologic Oncology. 2017; 145: 88–95.

[125] Bristow RE, Shumway JB, Khouzami AN, Witter FR. Complete hydatidiform mole and surviving coexistent twin. Obstetrical & Gynecological Survey. 1996; 51: 705–709.