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Case Report

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

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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, neonatologists 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.

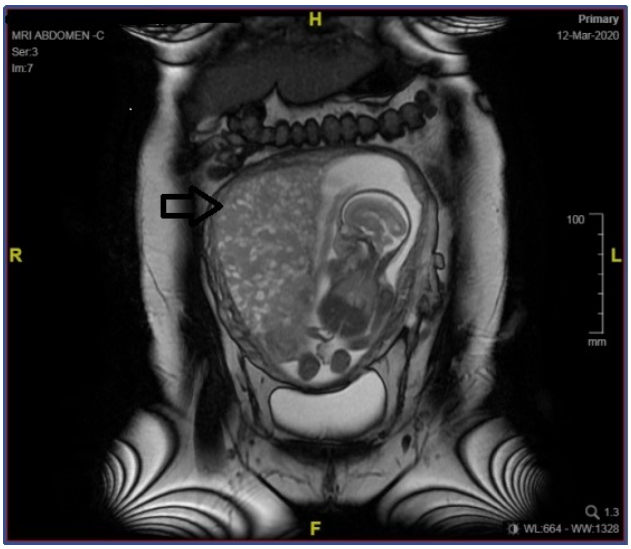

Fig. 1. Abdomen MRI at 20 weeks gestation. Twin pregnancy with a complete hydatidiform mole (arrow) can be seen along with a normal fetus and a normal placenta.

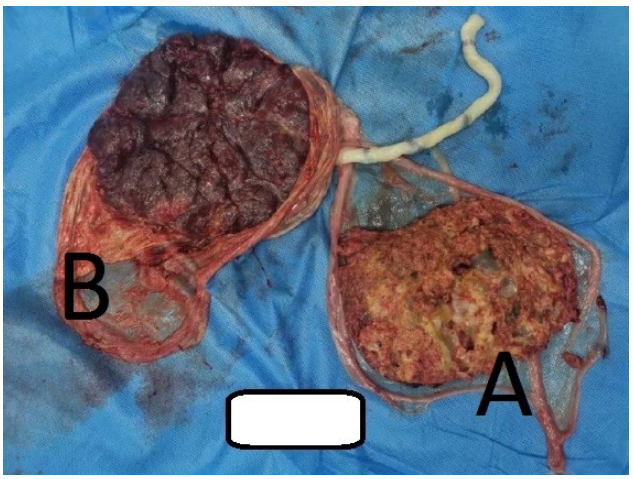

Fig. 2. Photograph of the molar (A) and normal (B) placentas obtained after the cesarean section.

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 antenatal 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
Altaras *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
Suksai *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	27,750/16	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, PIH	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	645,456/16	TOP	16	Yes
	32	-	30	22	VB	NA	383,000/22	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

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
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, PIH, 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
	41	-	27	18	VB	NA	NA	SA	18	No
Buke *et al.* (2014) [48]	42	-	21	17	VB, PP, PA, PL	NA	77,509/17	LB	CS/32	No
Chen *et al.* (2014) [49]	43	IUI	32	9	-	NA	551,600/10	TOP	14	No
Dalmia *et al.* (2013) [50]	44	-	20	10	VB	46, XX	NA	LB	VD/37	No
Loza *et al.* (2019) [51]	45	-	34	17	VB, HT, PL	NA	942,000/17	LB	CS/32	No
Dare *et al.* (1999) [52]	46	-	30	NA	PPROM, cord prolapse	NA	NA	LB	CS/NA	No
Devall *et al.* (2006) [53]	47	-	25	12	VB, PE, TLC	NA	NA	TOP	16	Yes
Ernst *et al.* (2009) [54]	48	-	28	NA	-	NA	NA	SB	27	NA
	49	-	29	NA	PIH, PL	NA	NA	LB	CS/30	NA
	50	-	32	NA	PL	NA	NA	LB	CS/35	NA
Ferraz *et al.* (2013) [55]	51	ICSI	39	12	HT	NA	1,402,565/14	TOP	14	Yes
Freis *et al.* (2016) [56]	52	-	33	14	VB, abruptio placenta	NA	NA	LB	CS/31	Yes
Nobuhara *et al.* (2018) [57]	53	IVF	42	9	VB	NA	647,000/8	TOP	10	Yes
Marcorelles *et al.* (2005) [19]	54	-	26	12	VB	46, XX	10,000/32	LB	CS/32	No
	55	-	25	18	-	46, XX	NA	LB	VD/38	No
	56	-	37	14	VB, TLC	46, XX	409,970/14	TOP	15	No
	57	-	41	15	VB, PE	46, XX	920,000/15	SA	21	Yes
Kashimura *et al.* (2001) [58]	58	IUI	30	13	VB	46, XY	10,260/13	TOP	14	Yes
Montes-de-Oca-Valero *et al.* (1999) [59]	59	IVF	41	16	VB, PE	46, XX	840,000/16	LB	CS/27	No
Moini *et al.* (2011) [3]	60	ICSI	39	18	VB	46, XX	NA	LB	CS/39	No
Giorgione *et al.* (2017) [60]	61	IVF	39	20	VB, HG	NA	NA	SA	20	Yes
	62	-	19	16	VB, HG, PP	NA	NA	SA	21	No
	63	ICSI	33	12	HT, TLC	NA	NA	TOP	13	Yes
	64	ICSI	31	18	VB, HT, HG	NA	NA	TOP	20	No
	65	-	39	20	PL	NA	NA	LB	CS/34	Yes
	66	-	37	19	PL	NA	NA	LB	CS/27	No
	67	OI	22	16	HT, PL	NA	NA	LB	CS/26	No
	68	-	NA	17	VB, HT	NA	NA	LB	VD/37	No
	69	-	42	15	VB	NA	NA	SA	15	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	VD/23	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	TOP	NA	NA
Winter *et al.* (1999) [20]	76	-	24	18	-	NA	287,000/17	LB	VD/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	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	NA	321,000/19	TOP	20	Yes
Makrydimas *et al.* (2002) [74]	88	-	28	15	VB	46, XX	NA	LB	CS/36	No
Peng *et al.* (2014) [21]	89	-	34	20	-	46, XX	310,277.7/20	LB	CS/37	Yes
Narlawar *et al.* (2000) [75]	90	-	29	22	VB, TLC, PL	46, XX	120,000	LB	VD/28	No
Rao *et al.* (2015) [76]	91	ICSI	29	16	VB, PL	NA	190,090/12	LB	CS/31	No
Garcia-Aguayo *et al.* (1992) [77]	92	-	25	14	VB	NA	149,333/14	TOP	14	Yes
Ozarpaci *et al.* (2005) [78]	93	-	28	16	VB	NA	530,000/16	TOP	16	No
Garbin *et al.* (1995) [79]	94	OI	30	23	HG, PE, PP	46, XX	134,600/25	TOP	27	No
Grenman *et al.* (1990) [80]	95	OI	20	19	VB, PE	NA	800,000/19	TOP	19	Yes
Harada *et al.* (1997) [81]	96	-	26	15	VB, PE	NA	1,207,600/15	TOP	15	Yes
He *et al.* (2014) [82]	97	-	20	18	-	46, XX	121,659.1/18	SA	22	No
Hirose *et al.* (1999) [83]	98	-	23	13	VB, HT	NA	1,024,000/13	TOP	13	No
Hsu *et al.* (1993) [84]	99	NA	29	15	VB, PE	46, XX	NA	TOP	15	NA
Ishii *et al.* (1998) [22]	100	-	30	24	VB	NA	NA	SB	25	Yes
	101	-	27	14	VB	NA	NA	SA	14	No
	102	OI	35	16	VB	NA	NA	ND	CS/22	Yes
	103	-	31	15	-	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
	105	-	37	22	-	NA	NA	LB	VD/40	Yes
Kaa *et al.* (1995) [85]	106	-	30	NA	VB	46, XX	NA	TOP	13	No
	107	-	25	NA	VB	46, XX	NA	TOP	8	Yes
	108	OI	36	NA	VB, PL	46, XX	NA	SA	18	No
	109*	OI	31	18	VB, PL	46, XX	327,150/18	SB/SB	25	No
	110	-	30	NA	VB	46, XX	NA	LB	NA/38	No
	111	-	32	11	VB, HG	46, XY	700,000/11	TOP	11	Yes
Koyama *et al.* (2010) [86]	112	-	20	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	310,351/17	SA	21	No
	119	-	24	11	-	46, XY	351,660/11	SA	11	No
Lee *et al.* (2010) [17]	120	-	26	13	VB, PE	46, XX	500,000/20	SA	20	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*	-	30	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

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
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
Shozu *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)

Table 3. CMCF cases that resulted in gestational trophoblastic neoplasia from literature review (n = 55).

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
2 [26]	285,000/28	2,900/6	3,500/8	-	Methotrexate	9	No
3 [27]	425,000/12	6,000/8	Plateau	Lungs	Methotrexate	6	No
8 [31]	239,100/12	NA	NA	Lungs	Methotrexate	6	No
11 [32]	1,298,000/11	High	High	-	Methotrexate	NA	No
13 [33]	1,069,300/8	High	High/10	-	5-FU + KSM	5 courses	No
14 [33]	1,425,000/13	NA	NA	Invasive mole	5-FU + KSM	6 courses	No
24 [16]	NA	Normal	High/18	-	Methotrexate	3	No
25 [38]	27,750/16	NA	NA	Lungs	Methotrexate + citrovorum factor	10 courses	No
31 [41]	645,456/16	NA	3900/6	Right lung	Methotrexate >actinomycin D >EMA + cisplatin	NA	No
33 [41]	1,620,000/18	56	Plateau		Methotrexate + actinomycin D	3	Yes
34 [41]	3,200,000/19	77,000	Plateau		Methotrexate + actinomycin D	NA	No
35 [42]	478,000/24	Increase	41,600/7	Lungs	Etoposide	6 courses	No
37 [44]	290,000/14	>1000	Plateau	-	Methotrexate	NA	Yes
47 [53]	NA	37,946/4	-	Invasive mole	Methotrexate >EMA-CO	NA	No
51 [55]	1,402,565/14	28.74/8	Plateau	-	NA	NA	No
52 [56]	NA	Increase/8	Increase/32	-	Methotrexate	2	No
53 [57]	647,000/8	310,000/5	-	Lungs, diagnosed as chorio-carcinoma	EMO-CO	11	Yes
57 [19]	920,000/15	Decrease/1	Increase/6		Methotrexate (chemoprophylaxis) >Actinomycin D + etoposide	NA	No
58 [58]	10,260/13	1,680/1		Left lung	Methotrexate	3 courses	No
61 [60]	NA	NA	NA	NA	NA	NA	No
63 [60]	NA	NA	NA	NA	NA	NA	No
65 [60]	NA	NA	NA	NA	NA	NA	No
70 [60]	NA	NA	NA	NA	NA	NA	No
77 [63]	1,638,200/15	Increase/3	-	-	Methotrexate	Modified bagshawe regime	No
79 [65]	963,971/NA	2,832/2	Plateau	Lungs, invasive mole	EMA-CO	5 courses	Yes
80 [66]	113,324/19	High	Plateau	-	Methotrexate >actinomycin D	NA	No
81 [67]	939,390/13	High	Plateau	Lungs	Methotrexate	14	No
83 [69]	1,024,000/14	High	-	Lungs	Methotrexate + actinomycin D	6 courses	No
86 [72]	600,000/9	Increase/4	-	-	Methotrexate >actinomycin D	NA	No

Table 3. Continued.

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]	310,277.7/20	995.3/1	268.1/16	Left lung	Methotrexate + folinate >EMA-CO	3 > 4	No
92 [77]	149,333/14	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 metasta-size 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 + Kengshengmycin; 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, Intra-Cytoplasmic 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, Preterm Labour; PP, Placenta Praevia; PPROM, 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.

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