CASE REPORT

Complete Hydatidiform Mole and Co-Existing Foetus with Severe Maternal Thyrotoxicosis: A Case Report

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ABSTRACT

Twin molar pregnancy refers to the coexistence of a complete or partial hydatidiform mole alongside a foetus. A complete hydatidiform mole with co-existing foetus (CHMCF) is an exceptionally rare obstetric phenomenon, with an incidence of approximately 1 in 20,000 to 100,000 pregnancies. Owing to its rarity, no standardized management guidelines currently exist. We report a case of a 33-year-old woman, G5P4, diagnosed with CHMCF at 15 weeks gestation, who developed severe maternal thyrotoxicosis and rapid uterine enlargement necessitating termination of pregnancy for maternal safety. Following termination of pregnancy, she was monitored closely with β-hCG surveillance due to the potential risk of gestational trophoblastic neoplasia. This case highlights the diagnostic dilemma, as well as challenges of management and post-treatment surveillance in CHMCF. Early diagnosis and multidisciplinary care are essential for optimal maternal outcomes.

INTRODUCTION

Molar pregnancy (hydatidiform mole) occurs approximately 1-3 per 1,000 pregnancies and is classified into complete or partial hydatidiform mole. Complete hydatidiform mole occurs when an empty ovum fertilized by one sperm that duplicates its paternal genome or results from when one ovum is fertilized by two sperms. Consequently, no embryonic or foetal tissue develop. Suction curettage remains the mainstay of treatment.

In contrast, CHMCF refers to the coexistence of a complete hydatidiform mole alongside a foetus. CHMCF was first described in 1914[2] and is more common than partial hydatidiform mole with a co-existing foetus (PHMCF). In comparison, CHMCF has a higher risk of gestational trophoblastic neoplasia (GTN) with a possibility of live birth of 40-60%[1,3,4,10], while PHMCF has a high likelihood of miscarriage or intrauterine foetal death due to severe chromosomal abnormalities.[4] Owing to its rarity and lack of standardized guidelines, CHMCH presents considerable diagnostic and management challenges.

CASE REPORT

We report a very rare case of twin molar pregnancy of the CHMCF type in Malaysia. A 33-year-old woman (G5P4) presented at 10 weeks' gestation with vaginal spotting. An ultrasound showed a single viable foetus, which corresponded to her gestational age. She was discharged and treated as threatened miscarriage.

At 12 weeks gestation, she experienced hyperthyroid symptoms, primarily palpitations. Clinical assessment showed that the uterus was larger than dates, measuring approximately18 weeks, with a doughy consistency. Ultrasonography showed a single viable foetus corresponding to 12 weeks' gestation and the presence of multiple hypoechoic cystic lesions within the placenta. The differential diagnoses included a partial mole, a molar pregnancy with co-existing normal twin, a singleton pregnancy with placental mesenchymal dysplasia, or an exaggerated placental site tumour. Her β-hCG level was 453,800 IU/L, exceeding the normal range for a singleton

pregnancy between 9 and 12 weeks (25,700-288,000 IU/L).[5] Her thyroid function was also raised with free T4 elevated at 44.3 pmol/L. Therefore, propylthiouracil at a dose of 50 mg twice daily was initiated.

At 15 weeks gestation, the ultrasound revealed a viable foetus with two distinctive placental morphologies: anteriorly to the foetus, there was a multicystic appearance of the placenta and the presence of another normal, homogenous placenta adjacent to it extending posteriorly (Figure 1). Bilateral theca lutein cysts were observed. These findings were aligned with the ultrasound findings of CHMCF, as reported in the case series by Giorgione et al. Because the molar placenta was located anteriorly, amniocentesis was not performed due to the risk of massive bleeding.

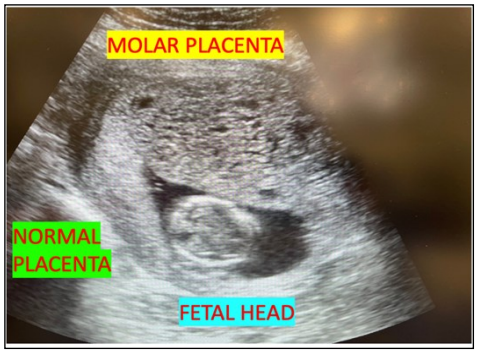

Figure 1: Transabdominal ultrasound at 15 weeks gestation demonstrating the viable fetus (F), the adjacent multicystic 'molar' placenta (MP), and the separate normal placenta (NP). Bilateral theca lutein cysts are also noted (not shown)."

Despite treatment, her clinical condition progressively deteriorated including palpitations, persistent vomiting, significant weight loss, reduced effort tolerance, and lower limb swelling. Clinical examination revealed fine tremors without overt signs of heart failure, and the gravid uterus had enlarged to a 24 weeks' size. Her β-hCG level exceeded 1,000,000 IU/L, and thyroid function tests confirmed severe thyrotoxicosis (TSH< 0.0005 mIU/L and free T4 at 42 pmol/L). Given the high risk of maternal deterioration, termination of pregnancy was advised after in-depth counselling and she was managed by a multidisciplinary team.

She underwent suction and curettage (S&C) under ultrasound guidance with cervical ripening facilitated by per vaginal misoprostol 400µg three hours prior to the procedure and was closely monitored for bleeding.

Intraoperatively, the foetus required piecemeal extraction because of the advanced gestational age using sponge forceps, followed by suction and curettage using a 10-mm Karman cannula. Oxytocin infusion was initiated at the beginning of the procedure to minimise the risk of excessive haemorrhage. The estimated blood loss was 1 litre, and she received one unit of packed red blood cells.

Post-procedural inspection revealed morphologically normal foetus with a normal placenta (Figure 2a) and vesicular tissue consistent with a molar pregnancy (Figure 2b), which was later confirmed by histopathological examination.

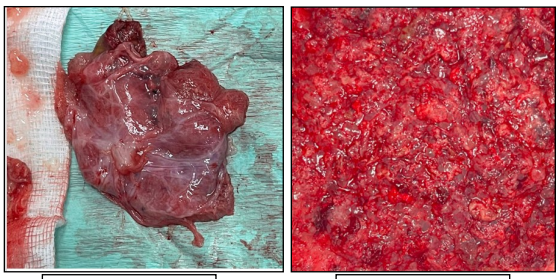

Figure 2a | Figure 2b

Figure 2: Gross specimen of placental tissues removed during suction and curettage.
(a) Normal-appearing placental tissue with smooth chorionic surface and membranous remnants.
(b) Molar tissue showing multiple translucent vesicles of varying sizes, consistent with complete hydatidiform mole.

Initially, serial weekly β-hCG levels declined, but by the fourth week post-S&C, levels plateaued at 9,787 IU/L (Figure 3). She declined chemotherapy recommended by oncology team.

At the tenth week post-S&C, a new intrauterine lesion was detected on imaging, with a β-hCG level of 3,585 IU/L. She underwent a repeat S&C. Histopathologic examination (HPE) did not reveal any evidence of gestational trophoblastic neoplasia.

Following the procedure, her β-hCG levels declined and eventually normalised, and her thyrotoxic symptoms resolved, allowing for the gradual discontinuation of antithyroid medication.

DISCUSSION

CHMCF is a very rare and complex condition. Here, we explore the diagnostic dilemmas, prenatal testing options,

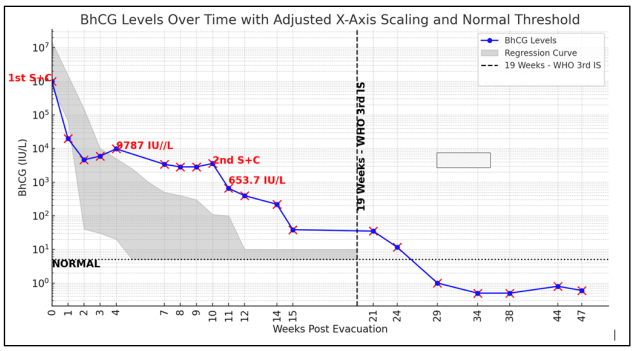

Figure 3: Post-evacuation **β-hCG trend** over time
(**X-axis:** weeks post-evacuation; **Y-axis:** β-hCG, IU/L; log scale).

DIAGNOSTIC DILEMMA

Ultrasound serves as a valuable tool for differentiating twin molar pregnancy from other placental abnormalities, such as placental mesenchymal dysplasia and exaggerated placental site reactions. Typically, the diagnosis of CHMCF is made between 12 and 14 weeks of gestation[4,7], through the concurrent visualisation of both a normal placenta and a molar placenta. Bilateral thecal lutein cysts are present in approximately one-quarter of CHMCF cases.[4,7]

β-hCG levels can aid in differentiation, as they have significantly elevated in twin molar pregnancies but only mildly increased in placental mesenchymal dysplasia.[8]

In cases where the diagnosis remains inconclusive, magnetic resonance imaging (MRI) can provide a further distinction between CHMCF and placental mesenchymal dysplasia.[9]

ROLE OF PRENATAL TESTING

Prenatal diagnosis using chorionic villous sampling, amniocentesis, or foetal cord sampling can help to differentiate between CHMCF and PHMCF. Cytogenetic analysis of CHMCF typically reveals a diploid karyotype of paternal origin in the molar placenta, while the coexisting foetus has a normal euploid karyotype. In contrast, cases of PHMCF often exhibit triploid or tetraploid karyotypes, with both the placenta and foetus being structurally abnormal and frequently resulting in miscarriage during the first trimester.

The potential role of cell-free DNA (cfDNA) in diagnosing CHMCF was described by Simon et al. in 2015. This non-invasive method can detect anomalous foetal haplotypes and paternal uniparental disomy, a key feature of a complete hydatidiform mole. As a result, cfDNA testing may reduce the need for invasive prenatal procedures, particularly in cases where the risk of bleeding is high.

EXPECTED PERINATAL OUTCOMES

CHMCF has a high risk of complications, including vaginal bleeding, pre-eclampsia, hyperthyroidism, GTN, miscarriage, preterm birth, and intrauterine foetal demise. The likelihood of achieving a live birth ranges from 40% to 60%.[1,3,4,10]

Historically, 50% to 70% of women underwent elective termination due to maternal complications.[4] However, recent retrospective cohort studies (2017 & 2024) have shown that only 8% to 23% of women opted for elective termination, while 8% to 16% underwent termination due to maternal complications.[1,10]

Maternal complication rates have remained comparable between old and recent studies. A systematic review and meta-analysis by Zilberman Sharon et al. reported an overall antenatal maternal complication rate of 80.8%, with 70% of women experiencing vaginal bleeding, 23.3% developing hyperthyroidism, and 14.3% being diagnosed with pre-eclampsia. Intrauterine foetal demise was reported in 40.1% of cases, and preterm birth accounted for 78% of live births. A French retrospective cohort study (2024) found that the mean gestational age at delivery was 32 ± 5 weeks.[10] The risk of developing GTN following a CHMCF pregnancy has remained consistently high, ranging from 27% to 46% across various studies.[1,4,10]

More favourable obstetric outcomes have been observed in pregnancies without maternal complications and with β-hCG levels below 400,000 IU/L, with a higher likelihood of achieving a live birth.[13] However, in this case, the patient experienced multiple severe complications, including vaginal bleeding, progressively worsening hyperthyroid symptoms, and extremely

elevated β-hCG levels exceeding 1,000,000 IU/L. Hence, after in-depth counselling with the couple, termination of pregnancy was carried out.

CHALLENGES IN TERMINATION OF PREGNANCY

Maternal stabilisation is a priority and requires a multidisciplinary approach. Surgical challenges include a tightly closed cervix and an enlarged uterus with the coexistence of a formed foetus and molar tissue, which increase the risk of torrential bleeding. This case also highlights the technical challenges of second-trimester uterine evacuation in CHMCF, where evacuation of a formed foetus followed by complete removal of extensive molar tissue requires meticulous surgical planning to minimise haemorrhage and reduce the risk of retained trophoblastic tissue. Cervical ripening immediately before uterine evacuation is considered a safe approach. Ultrasound guidance during the procedure is recommended to reduce the risk of uterine perforation and ensure complete evacuation. Suction and curettage (S&C) remains the preferred method of uterine evacuation unless foetal parts obstruct suction. The International Federation of Gynaecology and Obstetrics (FIGO) recommends initiating oxytocin at the start of the procedure to minimise bleeding risk. However, physicians need to balance the benefits of bleeding control with the potential risk of embolisation of molar tissue.

MONITORING POST-TERMINATION OF PREGNANCY

CHMCF has a higher risk of GTN (27-46%) compared with single molar pregnancies (15%).[1,4,10] The median time for β-hCG normalisation in CHMCF is 12-16 weeks[1], and 8 weeks for single molar pregnancies. Therefore, pre-existing β-hCG regression nomograms for single molar pregnancies are not representative of the β-hCG reduction trends in CHMCF, and the optimal timeframe for β-hCG surveillance in CHMCF remains uncertain. Rising or plateauing β-hCG levels may indicate the development of GTN. Most cases are FIGO low-risk (FIGO score 0 to 6) and respond well to first-line single-agent chemotherapy (methotrexate), with an overall cure rate of 91%.[10] For less chemo-sensitive GTN, combination chemotherapy with surgery achieves a 100% cure rate.[10]

CONCLUSION

CHMCF presents significant diagnostic challenges, often becoming more readily recognisable on ultrasound after 12 weeks' gestation, with maternal complications being more frequent and severe than in single molar pregnancies. S&C is challenging, requiring the piecemeal extraction of the normal foetus into smaller pieces to achieve complete evacuation of molar tissue.

β-hCG levels in CHMCF are markedly elevated and regress more slowly than in singleton molar pregnancies. The lack of a specific β-hCG regression nomogram further complicates post-termination monitoring and management. A multidisciplinary team approach is essential to optimise patient care.

CONFLICT OF INTEREST

The authors declare no conflicts of interest related to the publication of this manuscript.

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