

# Complete hydatidiform mole with coexisting live fetus: case report

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## ABSTRACT

Hydatidiform mole (H mole) is a rare complication of pregnancy, characterized by an atypical trophoblastic proliferation and swelling of the chorionic villi, with or without a live fetus. A clinical diagnosis of H mole is confirmed by significantly high serum beta-hCG levels and/or the presence of characteristic sonographic and histopathologic findings. Differentiation between the two types—complete or partial—is important, since complete moles pose a higher risk for malignancy. An H mole can be further complicated by the presence of a coexisting live fetus. Complete molar pregnancy with a coexisting live fetus has an incidence of only 1 in 22,000 to 100,000 pregnancies. Termination of pregnancy is usually considered, especially when high risks of choriocarcinoma and maternal prenatal complications are present. We present the case of a 27-year-old female diagnosed as having a complete H mole with a coexisting live fetus. We terminated the pregnancy due to profuse vaginal bleeding that could cause maternal and fetal compromise.

**Keywords.** choriocarcinoma, placental mesenchymal dysplasia, methotrexate, gestational trophoblastic disease

## INTRODUCTION

Hydatidiform mole (H mole), a type of gestational trophoblastic disease, is characterized by an abnormal trophoblastic proliferation and the presence of hydropic chorionic villi, with either an absent or a deformed, nonviable fetus.<sup>1,2</sup> In North America, Europe, and Southeast Asia, the incidence of H mole is 1 to 2 in 1000 pregnancies. However, in the Philippines, the incidence is 1 in 250 pregnancies.<sup>3</sup> H moles are classified into one of two types—complete or partial—based on their cytogenetic, morphologic and histopathologic features.<sup>4</sup> Complete H mole is the more common of the two types.<sup>5</sup> It is important to differentiate one type from the other, since complete H moles are more invasive and have poorer prognosis.<sup>1,6</sup>

H mole coexistent with a twin fetus (HMTF) is an even rarer condition, with an incidence of only 1 in 20,000 to 100,000 pregnancies.<sup>7</sup> There are three different possible mechanisms of HMTF formation: a complete mole coexistent with a normal diploid fetus, a partial mole coexistent with a normal diploid fetus, and a partial mole with an abnormal triploid fetus.<sup>8,9</sup>

The risk of developing choriocarcinoma, a common subtype of gestational trophoblastic neoplasia (GTN), is 15% among patients with complete mole and 0.5% among those with partial mole.<sup>10</sup> The risk is significantly higher in patients with complete HMTF (50%) than in those with complete hydatidiform mole alone (12.5%).<sup>8</sup> There is also a higher risk of GTN among patients

with signs and symptoms indicative of marked trophoblastic proliferation (i.e., persistent vaginal bleeding, unexplained weight loss, abdominal swelling, etc.). Other risk factors for postmolar GTN include maternal age  $\geq 35$  years, gravidity  $\geq 4$ , inappropriately large uterine size after 6 weeks age of gestation (AOG, beta-hCG titer  $\geq 100,000$  mIU/mL, and presence of theca lutein cysts  $\geq 6$  cm.<sup>11</sup> Delayed detection and treatment of a complete mole usually lead to the development of choriocarcinoma.

We report the case of a 27-year-old woman who had a complete molar pregnancy with a coexisting live fetus. We performed delivery of the fetus and evacuation of the H mole by cesarean section. The mother had an uneventful course in the wards, but the baby died 3 days postpartum due to congenital diaphragmatic

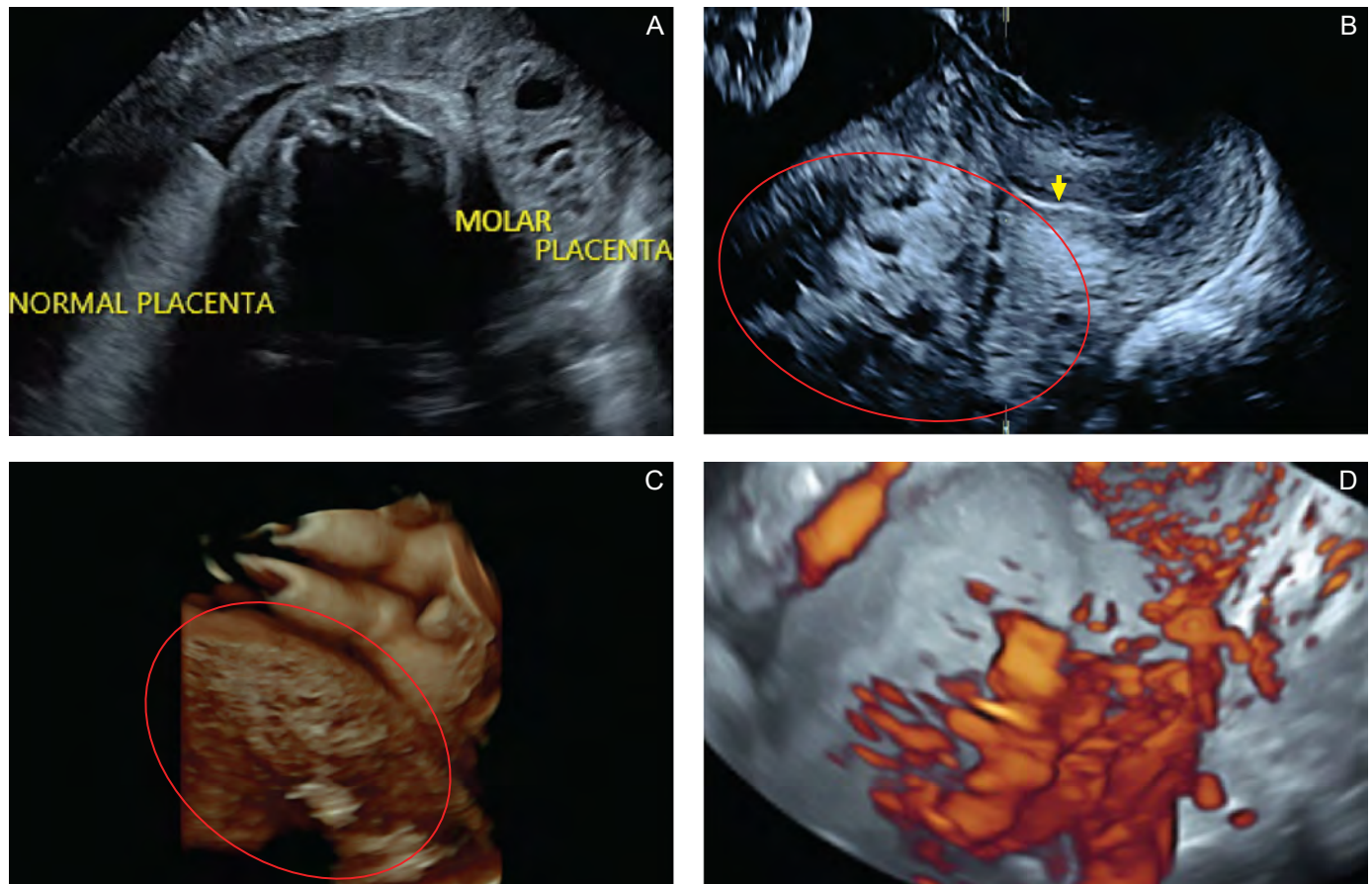
## IN ESSENCE

Complete hydatidiform mole with coexisting fetus is a very rare obstetric problem that poses a challenge in clinical decision-making and management.

In this case report, we describe a 27-year-old woman with a complete molar pregnancy with coexisting live fetus who presented with preterm labor at 31 1/7 weeks of gestation.

Due to profuse vaginal bleeding and uncontrolled labor, we terminated the pregnancy. We also administered a course of methotrexate as prophylaxis of postmolar gestational trophoblastic neoplasia.





**Figure 1** Abdominal 2D (A), transvaginal 2D (B), 3D rendering (C), and power Doppler (D) ultrasonographic images of complete hydatidiform mole with coexisting fetus at 31 weeks age of gestation, showing normal placenta (A) and the hypervascular molar tissue (B and C: red ring, and D)—with “snowstorm” or “honeycomb” pattern—completely covering the internal os of the cervix (B: yellow arrow).

hernia.

### CLINICAL FEATURES

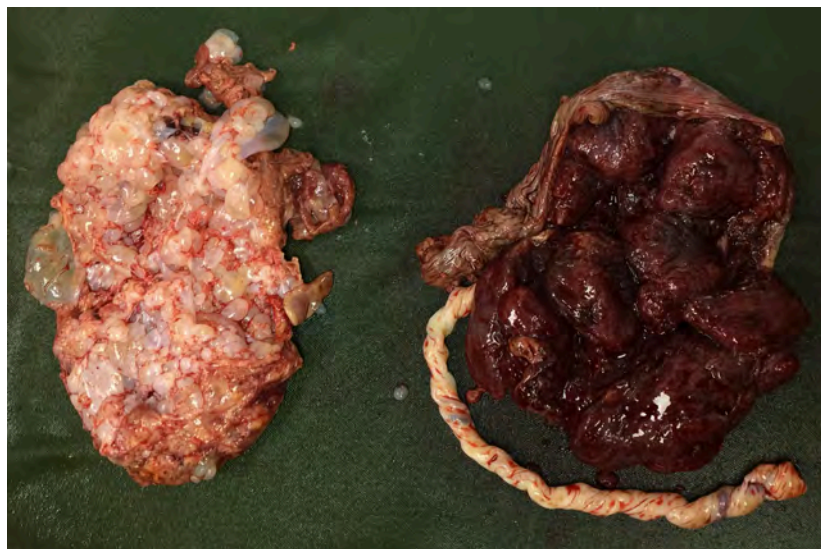
A 27-year-old woman, gravida 2 para 1, AOG 31 weeks and 1 day, came to our emergency room with a chief complaint of heavy vaginal bleeding. She reported that she had vaginal spotting at the 12th week of her gestation. She then consulted a general practitioner. An abdominal ultrasonography done revealed a live intrauterine fetus with normal placenta, coexisting with another placental mass that contains multiple cystic structures indicative of an H mole. The general practitioner prescribed a week's course of oral dydrogesterone and nifedipine for tocolysis. After taking the prescribed medications, the vaginal spotting would still occur infrequently, once or twice a month, for which the patient would self-medicate with dydrogesterone and nifedipine. Ten hours prior to her admission, she started to have profuse vaginal bleeding. A repeat ultrasonography done showed a complete molar pregnancy in the lower

uterine portion, coexistent with a 31-week-old fetus in transverse lie position in the upper uterine segment. She was referred to our care for further evaluation and management.

Two years before the present pregnancy, the patient delivered a live, full-term baby girl who eventually died at two weeks of life due to a congenital heart disease. The rest of the patient's history was unremarkable.

On physical examination, the uterus had a fundic height of 27 cm. The fetal heart rate at that time was 130 beats per minute. By Leopold's maneuver, it was determined that the fetus was in transverse lie position. Within the first day of admission, her blood pressure ranged from 120/80 to 150/80 mmHg. Our admitting diagnosis was: twin pregnancy with H mole and coexisting live fetus. We counselled the patient regarding the risks involved with this type of pregnancy. Since the patient lost her first child, she made a conscious decision to continue the pregnancy at all costs.





**Figure 2** Comparative size of the molar tissue (left) and the normal placenta (right). The molar tissue measures 15 x 13 x 5 cm and weighs 300 grams. The normal placenta measures 17 x 14 x 4 cm and weighs 340 grams.

### DIAGNOSTIC APPROACHES

On admission, we did several ultrasonographic imaging studies (Figure 1), which confirmed a twin pregnancy, consisting of a complete mole with a coexisting live fetus. The posterior portion of the uterus was occupied by the complete mole measuring 12.8 x 8.3 x 5.4 cm at the left posterolateral area and containing multiple cystic structures. The fetus was breech in presentation with good fetal movements. The normal placenta had occasional basal calcification consistent with 30-38 weeks AOG (Grade II), located anteriorly in the fundal area. The estimated fetal weight was 1,500 grams, which was appropriate for gestational age. We noted several episodes of fetal bradycardia. Doppler flow indices showed normal values for umbilical and uterine arteries. The cervical length was 3.2 cm. On admission, the patient's serum beta-hCG level was 97,879 mIU/mL. Her complete blood count showed leukocytosis ( $27.69 \times 10^3/\mu\text{L}$ ) with neutrophilic predominance (88%). Her serum electrolyte levels were within normal limits.

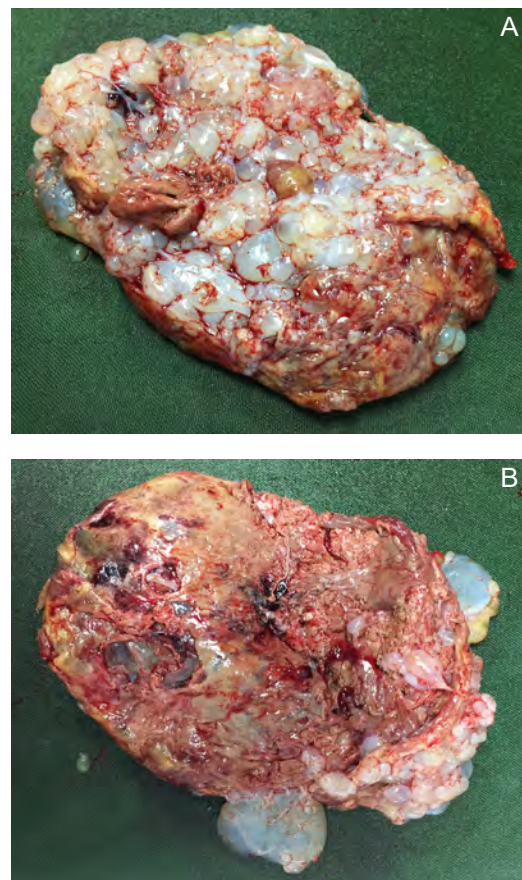
### THERAPEUTIC APPROACHES

We tried to manage the preterm labor with oral isoxsuprine on admission. On the second hospital day, the patient had several episodes of elevated blood pressure and persistent uterine contractions. Since uterine contractions were not controlled, we shifted the patient from isoxsuprine to magnesium sulfate for both tocolysis and neuroprotection of the fetus. We also started the patient on a course

of intramuscular dexamethasone for fetal lung maturation. By the third hospital day, the patient had already completed the 24-hour course of magnesium sulfate and four doses of dexamethasone. The patient had an episode of vaginal bleeding amounting to 300 mL, associated with persistent uterine contractions, hence we decided to do an emergency caesarean section.

The patient delivered a male baby weighing 1.5 kg with moderate asphyxia, with Apgar scores of 4, 6, and 6 at 1, 3, and 5 minutes, respectively. Both the normal placenta and the molar tissue were extracted manually. The placenta was roughly disc-shaped with a three-vessel umbilical cord attached centrally. The surgery was uncomplicated, with no significant uterine bleeding or other complications. The baby was admitted to the newborn intensive care unit due to prematurity.

Postoperatively, the patient had febrile episodes, hence we gave her intravenous clindamycin. We also started the patient on methotrexate chemoprophylaxis, which we administered intramuscularly once a day for five days right after molar evacuation. On



**Figure 3** Fetal (A) and endometrial (B) sides of the molar tissue, showing diffuse variable-sized vesicles.

discharge, we advised the patient to take oral contraceptives to avoid getting pregnant until the beta-hCG levels have normalized.

The pathological report confirmed our initial diagnosis of complete mole with co-existing live fetus. The specimen we sent consisted of normal placental tissue, which measured 17 x 14 x 4 cm and weighed 340 grams, and molar tissue, which measured 15 x 13 x 5 cm and weighed 300 grams (Figure 2). The umbilical cord was 40 cm in length with a maximum diameter of 2 cm. Grossly, the molar tissue had a diffuse vesicle formation. Vesicles within the molar tissue varied in size, with a maximum diameter of 4 cm (Figure 3). Histological examination of the molar tissue showed diffuse trophoblastic hyperplasia with mild-to-moderate atypia. The cores of the villi had fluid-filled cavities (cisterns). Taken together, the findings are consistent with complete hydatidiform mole (Figure 4).

### OUTCOMES

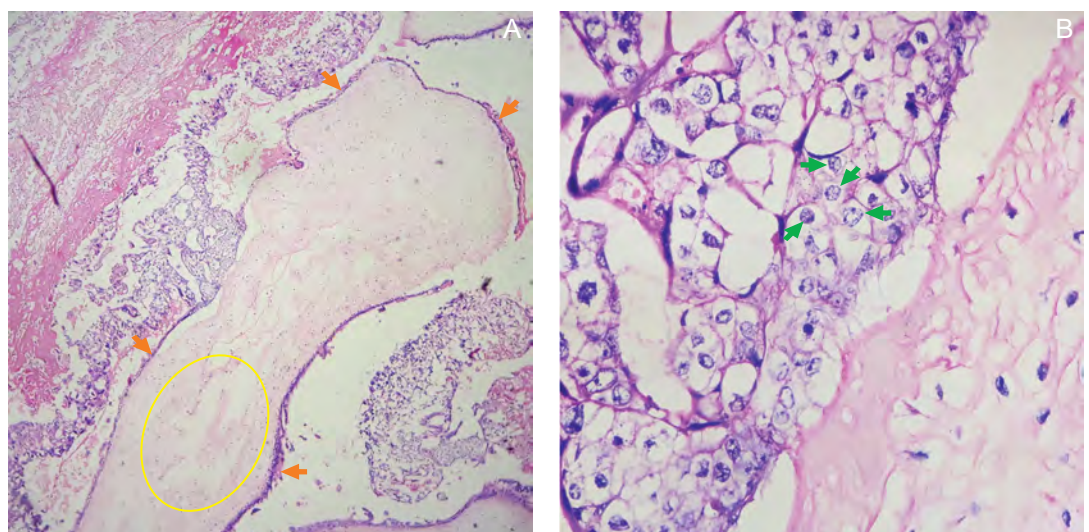
The patient's postoperative course in the ward was uneventful. During the same admission, the patient's serum beta-hCG levels went down to 7480 mIU/mL and 1131 mIU/mL on the 3rd and 7th days postpartum, respectively. She was discharged 11 days postpartum. We instructed her to return for follow up every two weeks until a beta-hCG level of  $\leq 5$  mIU/mL is achieved. However, the patient only followed up at the Outpatient Department (OPD) on the 16th and 30th days postpartum, with beta-hCG levels of 139.25 mIU/mL and 20.71 mIU/mL, respectively.

The baby was born with moderate asphyxia. He was then intubated and placed on mechanical ventilation. Arterial blood gas monitoring revealed uncompensated metabolic acidosis with hypoxemia five hours after birth, and respiratory acidosis eight hours after birth. A chest x-ray taken around this time revealed the presence of irregular lucencies representing bowel segments that were seen occupying the left hemithorax, and were insinuating from the left upper hemiabdomen. The left hemidiaphragm was not appreciated, and there was shifting of the midline structures to the right. These findings suggest the presence of a left diaphragmatic hernia, probably Bochdalek type. Chest ultrasound also revealed the same findings. He was immediately referred to the Pediatric Surgery Service. Two-dimensional echocardiography and congenital work-up were requested, and subsequent repair of the diaphragmatic hernia was scheduled. However, before the work-up could be done, on the third day of life, the baby was noted to be cyanotic and bradycardic, with skin mottling. Despite aggressive treatment, he went into respiratory arrest, which led to his eventual demise.

### DISCUSSION

#### Hydatidiform mole

The diagnosis of H mole is based on the clinical presentation of patients, combined with distinctive ultrasound findings, beta-hCG serum levels, and histopathological characteristics.<sup>11</sup> Patients with complete moles usually present with a pre-evacuation beta-



**Figure 4** Histopathology of the hydatidiform mole, showing a hydropic villus containing fluid-filled cavities (cisterns) (A: yellow ring) and lined with hyperplastic trophoblastic sheet (A: orange arrows). Magnified image of trophoblastic sheet revealing atypical cells (B: green arrows) (hematoxylin-eosin stain, A: x4, B: x10).



hCG titer greater than 100,000 mIU/mL.<sup>12</sup>

Several factors have been implicated in the development of H mole, but the exact etiology is still unknown. These factors include maternal age <15 or >40 years old, paternal age >45 years old, previous molar pregnancy, nulliparity with a history of miscarriage, and history of a twin pregnancy conceived by artificial insemination.<sup>11</sup> However, we could not elicit the presence of any of these risk factors from our patient.

### Complete hydatidiform mole with coexisting fetus

Complete hydatidiform mole with coexisting fetus (CHMCF) is seen mainly in patients between 21 and 41 years of age.<sup>13</sup> The coexistence of a complete mole with a live fetus is rare, with an incidence of only 1 in 22,000 to 100,000 pregnancies.<sup>14-15</sup> The condition is more common in Southeast Asia and Nigeria, compared to elsewhere in the world, with incidences of 2 in 1000 and 45 in 10,000 pregnancies, respectively.<sup>16</sup>

A complete hydatidiform mole with coexisting fetus (CHMCF) commonly presents with heavy vaginal bleeding, larger-than-gestational-age uterus, hyperemesis, hypertension, and preeclampsia.<sup>14-17-19</sup> Our patient presented with heavy vaginal bleeding and several episodes of elevated BP prior to delivery.

Some cases (22% to 30%) of CHMCF have been reported following ovulation induction,<sup>20-22</sup> however, no causal relationship between ovulation-inducing drugs and CHMCF has been established.<sup>22-23</sup>

The diagnosis of complete molar pregnancy with a coexisting fetus can be clinically challenging. This condition needs to be differentiated from a partial hydatidiform mole, placental mesenchymal dysplasia and chorioangioma.

The complete, classical mole has a diploid karyotype, hydropic placental villi, trophoblastic hyperplasia with no embryo or amnion.<sup>12</sup> A partial mole usually has a triploid karyotype, identifiable embryo, umbilical cord and amniotic membrane with only focal changes of placental villi and trophoblasts.<sup>17-24</sup>

An extremely rare condition to be considered is a partial mole in one amniotic sac with one normal twin in another.<sup>24</sup> Similar to CHMCF, the partial mole is characterized by a thickened placenta with hypoechoic, multicystic areas on ultrasonography and a large-for-gestational age uterus with several cystically dilated vesicles upon gross examination. In contrast to a complete mole, however, a partial mole has less prominent dilated stem vessels with trophoblastic proliferation.<sup>25</sup> Our patient's molar placenta had diffuse vesicle formation

**Table 1** Comparison of characteristics of complete and partial hydatidiform moles (H moles)

| Parameters         | Complete H mole  | Partial H mole   |
|--------------------|--|--|
| Signs and symptoms | <ul style="list-style-type: none"> <li>vaginal bleeding<sup>1 11 26 29</sup></li> <li>hyperemesis<sup>1 11</sup></li> <li>signs and symptoms of hyperthyroidism, including tachycardia and tremors<sup>1 11 26</sup></li> <li>pre-eclampsia<sup>1 11 29</sup></li> <li>larger-than-gestational-age uterus<sup>1 11 26 29</sup></li> </ul>                  | <ul style="list-style-type: none"> <li>less dramatic presentation<sup>1 11</sup></li> <li>vaginal bleeding<sup>1 11 27</sup></li> <li>occasionally, smaller-than-gestational age uterus<sup>1</sup></li> </ul>   |
| Beta-hCG levels    | <ul style="list-style-type: none"> <li>markedly elevated, with majority of patients presenting with a titer of &gt;100,000 mIU/mL<sup>1 11 26 29</sup></li> </ul>  | <ul style="list-style-type: none"> <li>normal to slightly elevated<sup>1 11</sup></li> </ul>   |
| Cytogenetics       | <ul style="list-style-type: none"> <li>diploid karyotype: 90% are 46,XX and 10% are 46,XY<sup>1 26 28 29</sup></li> </ul>  | <ul style="list-style-type: none"> <li>triploid karyotype: 69,XXX 69,XXY or 69,XYY (diandric triploidy)<sup>1 27-29</sup></li> <li>occasionally, tetraploid<sup>29</sup></li> <li>rarely, a diploid karyotype<sup>29</sup></li> </ul>  |
| Ultrasound         | <ul style="list-style-type: none"> <li>heterogeneous mass with multiple anechoic spaces, which resemble a classical "snowstorm" appearance, usually seen in the second trimester of pregnancy<sup>1 11 26 28 29</sup></li> <li>occasionally, presence of theca lutein cysts<sup>26 28</sup></li> </ul>   | <ul style="list-style-type: none"> <li>focal cystic changes in the placenta and/or increased echogenicity of chorionic villi ("Swiss cheese pattern")<sup>11 28</sup></li> <li>presence of a growth-retarded fetus with multiple congenital anomalies<sup>11</sup></li> <li>ratio of the transverse to anteroposterior dimension of the gestational sac is &gt;1.51</li> </ul> |
| Gross morphology   | <ul style="list-style-type: none"> <li>bulky, bloody tissue with volume sometimes over 500cc, and a classical "bunch of grapes" appearance<sup>26 29</sup></li> <li>normal placental structures are absent, and all villi are enlarged with variable-sized vesicles (1 to 30mm)<sup>26 29</sup></li> <li>absent embryo or fetus<sup>26 29</sup></li> </ul> | <ul style="list-style-type: none"> <li>tissue volume is less than 300cc<sup>29</sup></li> <li>characteristic changes affect only a part of the placenta and only a proportion of the villi are vesicular<sup>11</sup></li> <li>a fetus with congenital malformations is usually present<sup>11 29</sup></li> </ul>   |
| Histopathology     | <ul style="list-style-type: none"> <li>swollen villi with marked circumferential villus trophoblasts<sup>11 29</sup></li> <li>generalized hydropic changes with excessive trophoblastic proliferation, often marked with cytologic atypia<sup>11 29</sup></li> </ul>   | <ul style="list-style-type: none"> <li>mixture of swollen and normal-sized villi<sup>11 27</sup></li> <li>trophoblastic proliferation is either absent or very mild<sup>11 27 29</sup></li> </ul>  |

on gross morphology, with marked trophoblastic proliferation seen on histopathology, which is characteristic of a complete mole.

The clinical, laboratory, radiologic, and histopathologic features of complete and partial moles are described in Table 1.

Placental mesenchymal dysplasia (PMD) is a rare placental vascular abnormality often mistaken for molar pregnancy based on similar sonographic findings of enlarged, abnormally thick placenta with cystic areas.<sup>30</sup> PMD is associated with elevated maternal serum AFP (msAFP) levels and normal to elevated serum beta-hCG.<sup>31</sup> The primary complications in PMD are mostly fetal, and 23% of such complications are Beckwith-Wiedemann syndrome and fetal growth restriction.<sup>32</sup> Grossly, the placenta in PMD contains markedly dilated and tortuous chorionic vessels on the fetal surface.<sup>25 32</sup> Excessive trophoblastic proliferation—a hallmark sign of gestational trophoblastic disease—is absent in PMD.<sup>25 30 32</sup> In contrast, the molar placenta of our patient on histological examination showed diffuse hydropic chorionic villi with marked trophoblastic proliferation.

Another diagnostic consideration in the context of placental abnormalities with a normal diploid fetus is chorioangioma.<sup>24</sup> This can be differentiated from CHMCF by ultrasonography, which would show a well-circumscribed lesion located on the fetal placental surface or protruding into the amniotic cavity, with echogenicity that is different from the rest of the placenta.<sup>25</sup> Chorioangioma is characterized by the presence of dilated small vessels and capillaries inside the tumor with the same pulsation rate as in the umbilical cord.<sup>24</sup> In pregnancies with chorioangioma, the fetus may appear normal.<sup>33</sup> In contrast, our patient presented with a diffuse multicystic placenta attached to one side of the uterus, concomitant with a normal placenta attached to the fetus.

As early as the first trimester of pregnancy, CHMCF can be detected through transvaginal ultrasonography as a “snowstorm” or “honey-comb” pattern in the pelvic area, which is indicative of a distinct, heterogenous placental mass with multiple cystic structures adjacent to a normal-appearing placenta in one sac.<sup>12 22 34</sup> On ultrasound, our patient’s uterus contained a live normal fetus attached to one placenta that is located anteriorly, and another placenta with multiple cystic structures located posterolaterally. A variety of ancillary techniques, including p57 immunohistochemical staining

and molecular genotyping, have been developed to improve the diagnosis CHMCF.<sup>35 36</sup> High levels of human placental lactogen may also indicate the presence of CHMCF.<sup>13</sup>

In CHMCF, it is extremely rare for the fetus to progress into a viable, healthy infant.<sup>37</sup> Current literature suggests a 90% fetal loss rate for CHMCF.<sup>38</sup> The overall incidence of live birth, if the pregnancy is not terminated, is around 16% to 56%. Fetuses are usually delivered in the 30th week of gestation.<sup>15</sup> In some cases (41%), the pregnancy is terminated due to complications such as vaginal bleeding or pre-eclampsia.<sup>38</sup>

The management of complete mole with a coexisting live fetus remains controversial. The dilemma between continuation and termination of pregnancy prior to term presented itself in our patient’s case, especially in the setting of preterm labor with a normal fetus present. We made the decision to terminate the pregnancy due to profuse vaginal bleeding, which would have compromised both the mother and the baby. Regarding the risks of developing gestational trophoblastic neoplasia and maternal medical complications, several studies have produced contrasting recommendations.<sup>39 40</sup> A few studies have suggested continuation of pregnancy, irrespective of the development of GTN, in the absence of fetal anomaly or pre-eclampsia.<sup>40-42</sup> The incidence of developing GTN is independent of gestational age, but maternal prenatal complications were noted to be associated with GTN.<sup>43</sup>

The most common complications of CHMCF is the formation of malignant neoplasia, such as choriocarcinoma.<sup>15</sup> Symptoms of choriocarcinoma are usually related to metastases to the lungs (85%) and other organs. Choriocarcinoma with lung involvement may present with pulmonary symptoms. On the other hand, headaches, dizziness and seizures are related to brain metastasis.<sup>44</sup> Complete work-up to detect choriocarcinoma is done in one or more of the following conditions: beta-hCG  $\geq 20,000$  mIU/mL after 4 weeks post-evacuation; rise in beta-hCG  $\geq 10\%$  during the 2 consecutive biweekly determinations; plateauing beta-hCG values ( $<10\%$  decline or rise) at any time after evacuation; clinical or histologic evidence of metastasis at any site; persistently elevated beta-hCG titer at 14 weeks post-evacuation; and elevation of a previously normal beta-hCG titer after evacuation.<sup>11</sup> Our patient’s beta-HCG levels consistently decreased up to

the 30th day post-evacuation. Hence, a complete work-up for choriocarcinoma was not indicated. Ideally, the recommended serial beta-hCG monitoring to detect malignant degeneration suggests that serum beta-hCG should be taken one week postmolar evacuation, then every two weeks until normal levels ( $\leq 5$  mIU/mL) are reached. After three consecutive normal determinations, monitoring is decreased to monthly for 6 months, then at two monthly intervals for the next six months.<sup>11</sup>

To reduce the risk of choriocarcinoma, preventive methotrexate chemoprophylaxis is usually given to the patient.<sup>45</sup> Chemoprophylaxis is useful in situations where patients are at high risk for postmolar GTN, and when postpartum beta-hCG monitoring is doubtful.<sup>11</sup> Patients with any medical complication—such as preeclampsia, thyrotoxicosis, pulmonary insufficiency, and disseminated intravascular coagulation—as well as those with previous history of molar pregnancy, may benefit from preventive chemoprophylaxis.<sup>11</sup> Frequent follow-up by the patient was uncertain because her residence was too distant from the hospital. For these reasons, we decided to start the patient on methotrexate chemoprophylaxis.

Other common outcomes of CHMCF include intrauterine growth retardation, uterine rupture, fetal distress, premature delivery, and intrauterine fetal demise.<sup>46</sup>

In summary, we were presented with a patient with a complete mole and a coexisting fetus. The patient's pregnancy was terminated at preterm gestation due to profuse vaginal bleeding. The baby who had congenital diaphragmatic hernia, developed respiratory failure, which eventually led to his demise. A twin pregnancy consisting of a hydatidiform mole and a coexisting live fetus requires thorough evaluation. Differentiating between a complete and partial mole is of utmost importance, since a complete mole poses a high risk for GTN, especially when a live fetus is present. Although maternal complications may arise, the decision to continue the pregnancy until term should be entertained, especially if considerations regarding fetal survival outweigh maternal risk.

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#### Patient consent

Obtained

#### Reporting guideline used

CARE Checklist

(<http://www.care-statement.org/downloads/CAREchecklist-English.pdf>)

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#### Competing interests

None declared

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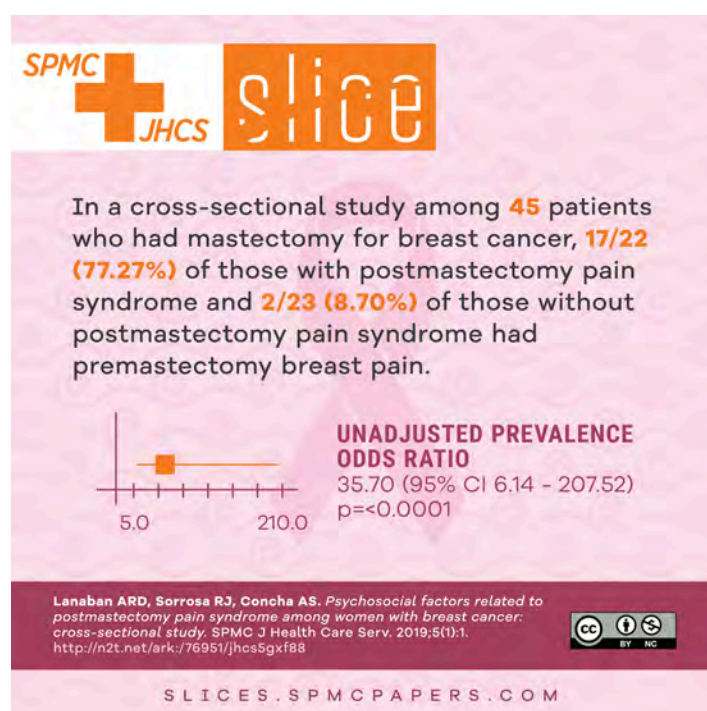


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