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Complete Hydatidiform Molar Pregnancy with Coexisting Fetus: A Case Report with Perioperative Considerations

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Abstract

A Complete Hydatidiform Molar pregnancy with Coexisting Fetus (CHMCF) is becoming reported more frequently in the literature. We present a case of CHMCF, with emphasis on interdisciplinary care and management in the perinatal and perioperative period. A 37-year-old G6P1041 patient presented to our institution with concern for CHMCF at 30-weeks gestational age. A multidisciplinary team was established and she underwent a cesarean total hysterectomy with delivery of a live neonate at 32-weeks' gestation. For care of patients with CHMCH, multidisciplinary teams, extensive coordination of care, Level 3 or 4 maternal hospital, and emphasis on the perioperative concerns should be considered.

Introduction

A Complete Hydatidiform Molar pregnancy with Coexisting Fetus (CHMCF) is becoming reported more frequently in the literature. Prevalence estimations are varied ranging from 1 in 20,000 to 100,000 pregnancies [1,2]. Management of CHMCF may be associated with perioperative complications including hyperthyroidism, thyroid storm, anemia, hypertension, and acute cardiopulmonary failure [2-4]. We present a case of CHMCF, with emphasis on interdisciplinary care and management in the perinatal and perioperative period. The patient has provided written Health Insurance Portability and Accountability Act (HIPAA) authorization to write and publish this case report.

Case Presentation

A 37-year-old G6P1041 was transferred to our institution at 30-weeks Gestational Age (GA) for delivery planning due to concern for CHMCF with possible invasion of the complete molar pregnancy into the right parametrium and right colon.

Evidence of CHMCF was discovered on ultrasound at 15-weeks GA. Amniocentesis revealed 46XX, suggesting CHMCF as opposed to partial molar pregnancy. Transfer to a tertiary care center was initiated due to light vaginal bleeding, concern for invasion and potential for cardiovascular collapse necessitating a multidisciplinary team.

Repeat Magnetic Resonance Imaging (MRI) showed a gravid uterus containing a single intrauterine placenta and fetus, and a 20 cm multi-cystic intrauterine mass along the right lateral margin of the amniotic sac abutting the margin of the placenta. There was bulging of this mass with myometrial thinning and prominent parametrial vessels, raising concern for abnormal adherence/percreta spectrum. Ultrasound depicted a viable fetus measuring 1553 g 30% tile with a posterior placenta on maternal left; and on maternal right a complex cystic hyperechoic mass abutting the placenta, with blurring of the suspected molar pregnancy-myometrium interface with increased vascularity and bulging (Figure 1).

Admission labs were: Thyroid Stimulating Hormone (TSH) 1.913 uIU/mL, Hemoglobin 9.2 G/DL, Hematocrit 28.9%, quantitative Human Chorionic Gonadotropin (qHCG) 443,590 mIU/mL. The remainder of her labs and blood pressures were normal and she was admitted for inpatient management until delivery at 32-weeks GA due to suspected invading molar pregnancy and vaginal

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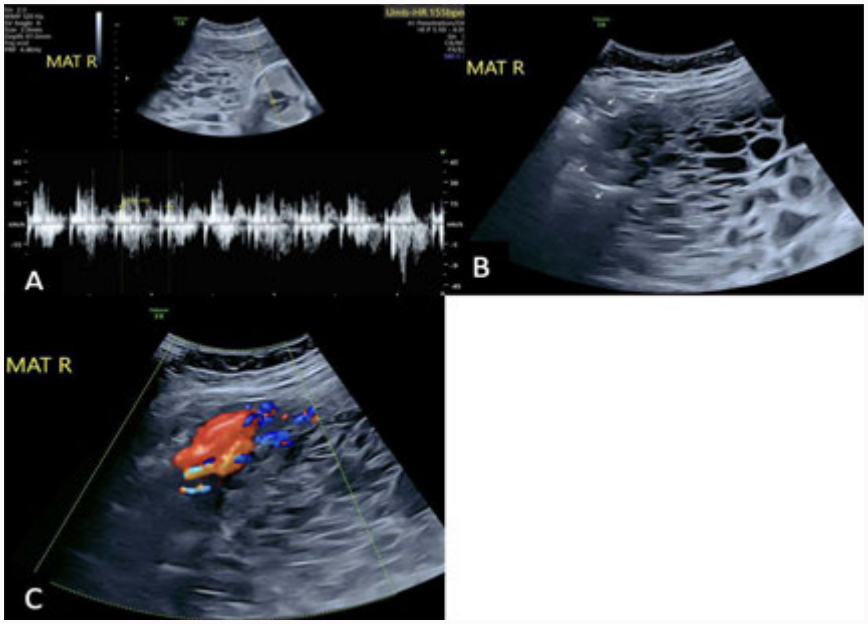

Figure 1: Fetal Ultrasound at 31 weeks gestational age depicting both complete molar pregnancy and coexisting viable fetus. A. Right side with complex cystic hyperechoic mass abutting the placenta and left side with viable fetus with fetal heart rate of 155 bpm. B. Depicted by arrows, blurring of molar pregnancy-myometrium interface on right aspect of complete molar pregnancy. C. Right aspect of complete molar pregnancy with increased vascularity.

bleeding.

Given the surgical complexity and potential for significant danger surrounding delivery, an interdisciplinary team was established: Maternal Fetal Medicine, Obstetric Anesthesiology, Cardiac Anesthesiology, Interventional Cardiology, Interventional Radiology, Blood Bank, Gynecologic Oncology, General Surgery and Neonatal Intensive Care Unit (NICU). Ultimately, the plan was made for delivery in a cardiac operating room with a cesarean hysterectomy.

Vascular access included a right internal jugular Multi-lumen Access Catheter (MAC) introducer, pre-induction arterial line and 16 g and 18 g peripheral lines. IR placed Fogarty catheters into bilateral internal iliac arteries for perioperative balloon occlusion as well as 5 French sheaths in both common femoral veins to expedite potential extracorporeal membrane oxygenation cannulation. In preparation for Massive Transfusion Protocol (MTP), 10 RBCs, 10 FFP, 1 platelet, and 2 cryoprecipitate were prepared by the blood bank.

After the placement of American Society of Anesthesiologists standard monitors and pre-oxygenation, general endotracheal anesthesia was initiated *via* rapid sequence induction. Maintenance of anesthesia was achieved with remifentanil 0.2 mcg/kg/min and propofol 125 mcg/kg/min infusions. Intraoperative transesophageal echocardiography was utilized by a cardiothoracic anesthesiologist with the goal of rapidly identifying the etiology of the potential shock. The patient's abdomen and chest were prepped with chlorhexidine in event of cardiac decompensation necessitating intervention.

A midline vertical skin incision and a left lateral classical uterine incision were employed to avoid disrupting the molar pregnancy. At time of delivery, bilateral internal iliac artery balloons were inflated. A viable neonate was delivered at 32-weeks GA with APGARs of 3 and 7. The amniotic fluid was brown; gray hydropic appearing tissue consistent with hydropic villi was protruding through the hysterotomy; and a large gravid appearing uterus was identified with serosal thinning on the right cornual aspect of the uterus. A total

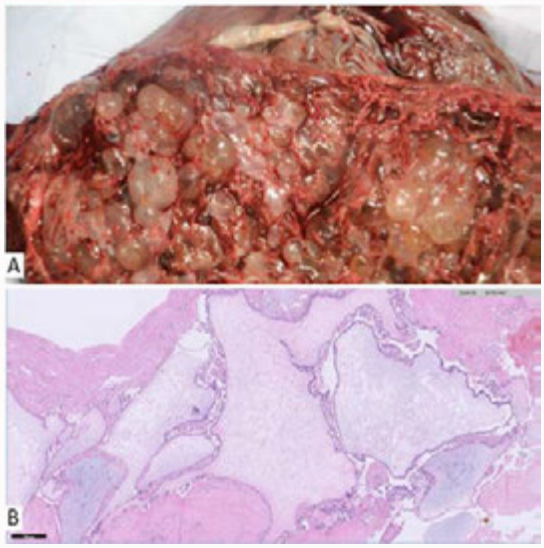

Figure 2: Pathological diagnosis. A. Gross examination revealed a 30 cm intrauterine mass composed of numerous large fluid-filled chorionic villi; B. Microscopic examination showed large hydropic chorionic villi with circumferential proliferation of trophoblasts. The villi had invaded into the inner half of uterine wall, consistent with invasive complete hydatidiform mole.

hysterectomy was performed. The iliac balloons were deflated after the specimen was removed. Total inflation time was 37 min. Total blood loss during the case was 800 cc. Nitroglycerine and clevidipine were required to maintain blood pressure <160/110.

The patient was transferred to the Surgical Intensive Care Unit (SICU) for increased monitoring. On postoperative day 0, preeclampsia with severe features was diagnosed by blood pressure criteria, requiring both infusion and oral antihypertensive agents. On postoperative day 1, repeat labs were: TSH 2.160 uIU/mL, qHCG 127,368.2 mIU/mL. The patient was discharged home on

postoperative day 3 with plans to follow qHCG weekly then monthly until complete resolution. Gross examination of the hysterectomy revealed a postpartum uterus (4,076 g) containing a 19 cm mature placenta and an adjacent 30 cm mass of numerous large fluid-filled chorionic villi (Figure 2a). Microscopic examination of the mass revealed hydropic chorionic villi with circumferential proliferation of trophoblasts. The villi had invaded into the inner half of the uterine wall, consistent with invasive complete hydatidiform mole (Figure 2b).

Discussion

There are several concerns regarding CHMCF, including hemorrhage, preeclampsia, hyperthyroidism, molar tissue embolization, cardiovascular compromise, respiratory distress, gestational trophoblastic neoplasia, fetal growth restriction, prematurity, and intrauterine fetal demise [4-6]. This case report represents a unique case in which a patient with CHMCF did not have severe complications antenatally and instead developed systemic manifestations in the postpartum setting. Furthermore, this case report adds to the existing literature by an in-depth review and considerations regarding the perioperative and postoperative considerations.

This prenatal course demonstrates many considerations, the first being differentiation between a CHMCF and a partial molar pregnancy. In this case, the patient underwent an amniocentesis and the karyotype was not suggestive of a partial mole, as a triploid karyotype would be expected. This is an important distinction as partial molar pregnancies are non-viable, unlike CHMCF [7]. While there was no immediate postpartum hemorrhage, the reported patient did have daily vaginal spotting since approximately 20-weeks gestational age. Prior studies have reported vaginal hemorrhage ranges from 0% to 34% in cases with CHMCF [6]. This stresses the importance of receiving care at institutions with large blood banks and maintaining IV access [6].

While the majority of case reports describe patients with CHMCF who develop preeclampsia antenatally, this patient did not develop preeclampsia until after delivery. It is important to monitor blood pressure values, possibly laboratory assessment and symptoms for signs of preeclampsia. In this patient, we assessed blood pressure daily, symptoms daily, and laboratory assessment every 72-h while inpatient. Regarding concern for hyperthyroidism, consider baseline laboratory assessment, possibly every trimester, with further evaluation postpartum and/or if symptoms were to present. Regarding the fetus, there have been reports of growth abnormalities and growth restriction. Throughout this patient's prenatal course, the fetus remained appropriate for gestational age, however, consider close fetal growth surveillance.

Communication between anesthesiologists and obstetricians should begin promptly after diagnosis of CHMCF to provide adequate time for a comprehensive workup. Particular emphasis is placed on cardiopulmonary status, electrolyte abnormalities, coagulation status and oxygen-carrying capacity. To safely care for these patients, these cases should be performed at a level 3 or 4 maternal hospital where there is a large blood bank, surgical resources and access to a multi-disciplinary team including Maternal Fetal Medicine, Obstetric Anesthesiology, Cardiac Anesthesiology, Interventional Cardiology, Interventional Radiology, Blood Bank, Gynecologic Oncology, General Surgery and NICU.

Symptoms of cardiopulmonary failure usually develop within 4 h to 12 h after evacuation of the uterus and vary in severity, with some requiring mechanical ventilation, vasopressors, and invasive monitoring. Acute cardiopulmonary distress has been observed after evacuation of molar pregnancy in 27% of the cases, especially in patients with a uterine size of 16-weeks or greater [8]. It usually develops within 12 h of evacuation making the immediate post-partum period high risk for complications. Trophoblastic embolization is the etiology in more than half of these cases [8]. In the most severe cases, massive trophoblastic embolism can lead to death. For this reason, consider recovery in SICU level care for the first 24 h to 48-h postoperatively.

Hyperthyroidism may occur due to excessive levels of circulating human chorionic gonadotropin released from the mole. Prevalence of hyperthyroidism during complete molar pregnancy is as high as 7%. Pulmonary hypertension, which is correlated with inadequately controlled hyperthyroidism, can result in heart failure and pulmonary edema. Thyroid storm has been reported in patients with molar pregnancy during the induction of anesthesia.

Anemia may be secondary to chronic bleeding from the placenta and from perioperative blood loss. Chronic bleeding from a molar placenta can stress the clotting mechanisms leading to disseminated intravascular coagulation and severe hemorrhage following evacuation. Because of the potential for substantial blood loss, adequate intravenous access, need for invasive monitoring, need for IR or ECMO, and the immediate availability of blood products should be established before induction.

General anesthesia with endotracheal intubation is often the technique of choice because of the potential for intraoperative hemodynamic instability. However, inhalational anesthetics may contribute to bleeding from excessive uterine relaxation.

In summary, CHMCF represents a serious obstetric condition with the possibility for rapid decompensation. This case emphasizes the importance of a multidisciplinary care team, a level 3 or 4 maternal hospital, and consideration of the possible maternal comorbidities. While more research is needed, this case greatly contributes to the literature and expands upon the complex care coordination required for these patients.

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