Accepted Manuscript

Submission Date: 2024-03-06
Accepted Date: 2024-03-27
Accepted Manuscript online: 2024-04-04

American Journal of Perinatology Reports

Expectant management of a triploid partial molar pregnancy at 26 weeks' gestation: a case report

Karen Wong, Mohannad Ali, Marc Stalder, Brigitte Bonin, Darine El-Chaar.

Affiliations below.

DOI: 10.1055/a-2299-4409

Please cite this article as: Wong K, Ali M, Stalder M et al. Expectant management of a triploid partial molar pregnancy at 26 weeks' gestation: a case report. American Journal of Perinatology Reports 2024. doi: 10.1055/a-2299-4409

Conflict of Interest: The authors declare that they have no conflict of interest.

Abstract:

Introduction

Triploid partial molar pregnancies are not viable, and confer maternal risks including preeclampsia, hemorrhage, gestational trophoblastic neoplasia, and trophoblastic embolization. We report a case managed expectantly until 26 weeks' gestation in a patient requesting continuation of pregnancy.

Case

This G2P1 presented with fetal anomalies indicative of triploid partial molar pregnancy. The pregnancy was complicated by anemia, hyperthyroidism, supraventricular tachycardia, and threatened preterm labour.
Her care involved Maternal Fetal Medicine collaborating with Internal Medicine, Palliative Care, Anesthesia and Critical Care. Labor was augmented at 26 weeks' gestation, resulting in vaginal delivery. Postpartum course was notably complicated by acute respiratory distress in the immediate post-partum period, which self-resolved. Postpartum hemorrhage and retained products of conception were additional complications.

Conclusion

This unique case highlights the role of multidisciplinary collaboration and shared decision making in challenging circumstances.

Corresponding Author:
Dr. Karen Wong, University of Ottawa Faculty of Medicine, Obstetrics & Gynecology, 451 Smyth Road, K1H 8M5 Ottawa, Canada, kwong005@uottawa.ca

Affiliations:
Karen Wong, University of Ottawa Faculty of Medicine, Obstetrics & Gynecology, Ottawa, Canada
Mohannad Ali, University of Ottawa Faculty of Medicine, Obstetrics & Gynecology, Ottawa, Canada
Marc Stalder, University of Ottawa, Department of Anesthesiology and Pain Medicine, Ottawa, Canada
Brigitte Bonin, Ottawa Hospital, Department of Obstetrics, Gynecology and Newborn Care, Ottawa, Canada
Darine El-Chaar, Ottawa Hospital, Department of Obstetrics, Gynecology and Newborn Care, Ottawa, Canada

Expectant management of a triploid partial molar pregnancy at 26 weeks' gestation: a case report

Karen Wong[1,2], MD MSc
Mohannad Ali, MD[1,2]
Marc Stalder[3], MD
Brigitte Bonin[1,2], MD
Darine El-Chaâr[1,2,4,5], MD MSc

[1]Department of Obstetrics and Gynecology, Faculty of Medicine, University of Ottawa, Ottawa, ON, Canada
[2] Department of Obstetrics, Gynecology and Newborn Care, The Ottawa Hospital, Ottawa, Canada
[3]Department of Anesthesiology and Pain Medicine, University of Ottawa, Ottawa, Ontario, Canada
[4]School of Epidemiology and Public Health, University of Ottawa, Ottawa, Ontario, Canada
[5]Clinical Epidemiology Program, Ottawa Hospital Research Institute, Ottawa, Ontario, Canada

Corresponding author:

Darine El-Chaâr, MD, MSc, FRCSC
Clinical Epidemiology Program, Ottawa Hospital Research Institute
Ottawa, Ontario, Canada
Email: delchaar@toh.ca

Abstract

Introduction

Triploid partial molar pregnancies are not viable, and confer maternal risks including preeclampsia, hemorrhage, gestational trophoblastic neoplasia, and trophoblastic embolization. We report a case managed expectantly until 26 weeks' gestation in a patient requesting continuation of pregnancy.

Case

This G2P1 presented with fetal anomalies indicative of triploid partial molar pregnancy. The pregnancy was complicated by anemia, hyperthyroidism, supraventricular tachycardia, and threatened preterm labour.

Her care involved Maternal Fetal Medicine collaborating with Internal Medicine, Palliative Care, Anesthesia and Critical Care. Labor was augmented at 26 weeks' gestation, resulting in vaginal delivery. Postpartum course was notably complicated by acute respiratory distress in the immediate post-partum period, which self-resolved. Postpartum hemorrhage and retained products of conception were additional complications.

Conclusion

This unique case highlights the role of multidisciplinary collaboration and shared decision making in challenging circumstances.

Introduction

Partial molar pregnancies are abnormal gestations, characterized by a triploid genotype. Triploidy is not compatible with long-term survival; most pregnancies spontaneously abort in the first trimester. Additionally, partial molar pregnancy itself confers maternal risks such as preeclampsia, hemorrhage, and gestational trophoblastic neoplasia. There may also be a risk of trophoblastic embolization, particularly when the uterus is greater than 16 weeks in size [1]. Most existing case reports of partial molar pregnancy in the second trimester involve induced abortion upon diagnosis [2–4].

Induced abortion is not an option for some individuals, and expectant management of even life-limiting fetal diagnoses is required. However, there is limited evidence in the literature regarding the management of ongoing partial molar pregnancies. To our knowledge, there is one case report describing expectant management until preeclampsia prompted delivery by Cesarean section at 31 weeks' gestation [5]. There is no information regarding vaginal delivery in this context.

We present a case of suspected partial molar triploid pregnancy managed expectantly into the late second trimester, with details on the precautions undertaken for planned vaginal delivery.

Case Presentation

Events in the case are summarized in Figure 1.

Patient information

A 25-year-old G2P1 was referred to the Maternal Fetal Medicine (MFM) service at 18 weeks 3 days gestation with the findings of thick nuchal translucency (NT), highly abnormal maternal serum screening analytes, and multiple anomalies on ultrasound. She had intermittent supraventricular tachycardia (SVT) in her prior pregnancy, though delivery was uncomplicated, with spontaneous vaginal delivery of a healthy female infant. SVT had resolved in the inter-pregnancy period.

Clinical presentation

A detailed ultrasound demonstrated a fetus with posterior fossa cysts, absent cerebellum, cystic hygroma, ventricular septal defect, and omphalocele containing both liver and gallbladder. Further, the placenta was noted to be thick and cystic (See Figure 2). Maternal ovaries were bilaterally enlarged, suggesting theca lutein cysts. Based on these clinical features, a diagnosis of triploid partial molar pregnancy was strongly suspected.

The patient was counselled regarding the poor prognosis of a triploid pregnancy, and the maternal risks of a partial molar pregnancy, including preeclampsia, obstetrical hemorrhage, thyrotoxicosis, respiratory compromise, and gestational trophoblastic neoplasia (GTN).

Assessment

Understanding both the poor prognosis and risks, the patient declined immediate induced abortion. She valued the time spent with the fetus in utero, and wished to continue

the pregnancy. However, she would accept an early induction of labour if her health deteriorated. The intent of an early induction of labour, in contrast to induced abortion, would be livebirth followed by palliative care. She wished to avoid cesarian delivery, except for maternal indications.

She declined amniocentesis, however accepted non-invasive prenatal testing (NIPT) using single nucleotide polymorphism (SNP)-based technology. This test found excess genetic material consistent with either triploidy or a vanishing twin. In the context of the ultrasound findings, this was interpreted as triploidy and supportive of the working diagnosis.

Management

Her prenatal care continued with weekly appointments. Perinatal Palliative Care was consulted to prepare the family for the delivery and develop a palliation plan. She was advised to monitor her blood pressure at home.

At 20 weeks' gestation, she was anemic (hemoglobin 82 g/L), and sub-clinically hyperthyroid (TSH 0.01 mIU/mL and thyroxine 10 pmol/L). She became persistently tachycardic and developed a non-pruritic petechial rash. Cardiology and Obstetric Medicine services were consulted. Investigations for underlying hematologic, cardiac, or thyroid abnormalities were negative. She was treated with intravenous iron and oral metoprolol.

By 24 weeks, the patient developed significant pedal edema limiting her mobility; shortness of breath on mild exertion; and threatened preterm labour. She was admitted to hospital with regular painful contractions. The cervix was closed, however transvaginal ultrasound demonstrated a short cervix, measuring 7 mm. She had an episode of SVT requiring chemical cardioversion with diltiazem.

With her worsening medical condition and risk of premature precipitous delivery with ensuing postpartum hemorrhage, she was offered and accepted induction of labor.

The peripartum plan was developed in collaboration with the Anesthesia service to mitigate risks of massive hemorrhage and/or trophoblastic embolization. The Critical Care service was notified of her expected delivery.

Mifepristone was administered for cervical ripening[6] at 26 weeks 0 days gestation as an inpatient, with the intention of subsequent doses of misoprostol. Two large bore IVs were established, and 6 units of packed red blood cells prepared. Spontaneous rupture of membranes and active labor occurred 17 hours later. An epidural and arterial line were placed, and she was brought to the operating room for delivery. Continuous electronic fetal monitoring was not utilized as she did not desire a cesarian delivery for fetal indications.

Outcome and follow-up

The second stage of labor lasted 15 minutes, with vaginal delivery of a stillborn female. The placenta was easily delivered intact (see Figure 3), followed by a large gush of blood. Uterine atony was treated with uterine massage, misoprostol, and oxytocin. Total blood loss for delivery was estimated at 1000mL. She received one unit of packed red blood cells.

About one hour after delivery, her vital signs deteriorated, with tachycardia (148 bpm), tachypnea (44 breaths per minute), fever (38.7˚ C), and hypertension (169/91 mmHg). She was drowsy, with shortness of breath and a cough productive of clear sputum. The lungs were clear on auscultation. She was transferred to the Intensive Care Unit by the Critical Care team. She received a single dose of propranolol 40mg orally for presumed thyroid storm and her vitals stabilized without further intervention. Of relevant investigations, TSH had normalized, and blood cultures were negative. Chest X-ray demonstrated interstitial edema and trace right pleural effusion. Hemoglobin was 79 g/L. Platelets, AST, ALT, and creatinine remained within normal range.

The differential diagnosis included sepsis, preeclampsia, gestational trophoblastic embolism, amniotic fluid embolism, thyroid storm, pulmonary embolism, transfusion reaction, and iatrogenic fluid overload. No clear diagnosis could be made for this presentation given her rapid recovery with limited intervention. She was discharged home on postpartum day 1 with stable vitals, and without additional medication.

Prior to discharge, the Palliative Care Team visited her following delivery to provide emotional support. The patient spent time with her stillborn daughter, and had mementoes created.

Placental pathology confirmed the diagnosis of partial molar pregnancy. All symptoms, including SVT, edema, and rash resolved by 6 weeks postpartum. However, lochia became heavy at about 8 weeks postpartum. Pelvic ultrasound demonstrated retained products of conception, which were evacuated by suction dilation and curettage. Theca lutein cysts had fully resolved. Serum β-hCG was done weekly until undetectable at 9 weeks postpartum.

The patient was counselled that the risk of recurrence of triploidy was low. She did conceive spontaneously following the events of this report, and had an uncomplicated pregnancy and vaginal delivery.

Discussion

Partial molar pregnancies are abnormal gestations, characterized by a triploid genotype. They are associated with considerable risk, for both the fetus and pregnant individual. This case report is the first to describe the expectant management and vaginal delivery of a singleton partial molar pregnancy into the second trimester.

Previous case series describe expectant management of *twin* pregnancies complicated by one complete molar twin, and one unaffected fetus.[7] Due to the presence of an unaffected

fetus, expectant management and delivery by cesarian delivery are often described in these series.

We highlight in this report the clinical measures and decision making involved in expectant management and vaginal delivery of a partial molar triploid pregnancy in the late second trimester.

In the antepartum period, we have demonstrated that where invasive diagnosis is declined, SNP-based NIPT may support the sonographic diagnosis. SNP-based NIPT deduces fetal genotype by comparison to maternal genetic material, in contrast to other NIPT technology which deduces triploidy by comparison to a reference chromosome. This allows SNP-based testing to identify the presence of a vanishing twin or triploid pregnancy, which is not possible with other NIPT technology.

The patient who does not undergo immediate induced abortion should be managed by Maternal Fetal Medicine and counselled regarding the risks of ongoing pregnancy. Unlike this case, hypertension and preeclampsia are described in most reports of partial molar pregnancy in the second trimester [2–5]. Blood pressure should be monitored closely. It is possible that the metoprolol taken by our patient for SVT masked rising blood pressure before gestational hypertension could be diagnosed.

Anemia and hyperthyroidism are less dramatic sequelae, but important to identify and manage in collaboration with Obstetric/Internal Medicine consultants. Normocytic anemia with normal ferritin, B12, and folate may be secondary to a significantly increased plasma volume without concurrent increase in red blood cell volume. Beta-blockade and/or thionamides may be required for symptomatic hyperthyroidism.

The development of theca lutein cysts suggests high levels of β-hCG. Complications of these enlarged ovaries can include adnexal torsion and cyst hemorrhage. Patients should be

advised to monitor and report severe pelvic pain. Adnexectomy is not required, as these cysts resolve postpartum.

The peripartum risk of trophoblastic embolization has been suggested in case reports [8]. The incidence of trophoblastic emboli at molar evacuation has been reported to be about 2%, with greater risk associated with uteri over 16 weeks' size or 4 weeks greater than expected [1]. Some speculate that the diagnosis is rarer, and that respiratory complications peri- or post-operatively are more likely attributable to pulmonary edema, anemia, hyperthyroidism, and/or preeclampsia [9,10]. There is a risk of peripartum or postpartum hemorrhage, as the markedly enlarged placenta distends the uterus.

Preparation for these peripartum risks is imperative and includes collaboration with Anesthesia. It is prudent to establish two large bore IVs, an arterial line, and to have blood products available. A central venous line could be considered when the risk of trophoblastic embolization is high. Providers may additionally choose to deliver in the operating room for proximity to resuscitative equipment. Fluid balance should be monitored, as vigorous fluid or blood administration may contribute to respiratory complications postpartum.

Postpartum, these patients require serial β-hCG monitoring, as for all hydatidiform molar pregnancies given the risk of gestational trophoblastic neoplasia.

These pregnancies should be additionally supported by consultation with a Perinatal Palliative Care team.

This case report explores the management of partial molar pregnancy into the second trimester. Interdisciplinary care and close surveillance are recommended. Patients should be thoroughly counselled on, and clinicians prepared for the antepartum, peripartum, and postpartum risks if choosing expectant management.

Disclosures: No financial support or funding was received in the preparation of this work.
Conflict of interest: none declared

Acknowledgements: The authors would like to thank the patient, and staff at The Ottawa Hospital department of OBGYN for their contribution to this work and care of this case.

References

1. Kohorn EI. Clinical management and the neoplastic sequelae of trophoblastic embolization associated with hydatidiform mole. Obstet Gynecol Surv. 1987;42(8):484–8.
2. Prasannan-Nair C, Reynolds SF, Budden G. Partial molar pregnancy with severe pre-eclampsia at 19 weeks' gestation. J Obstet Gynaecol (Lahore). 2006 Nov 1;26(8):817.
3. Rahimpanah F, Smoleniec J. Partial mole, triploidy and proteinuric hypertension: Two case reports. Australian and New Zealand Journal of Obstetrics and Gynaecology. 2000;40(2):215–8.
4. Stefos T, Plachouras N, Mari G, Cosmi E, Lolis D. A case of partial mole and atypical type I triploidy associated with severe HELLP syndrome at 18 weeks' gestation. Ultrasound in Obstetrics and Gynecology. 2002;20(4):403–4.
5. Drummond S, Fritz E. Management of a Partial Molar Pregnancy A Case Study Report. Vol. 23, J Perinat Neonat Nurs.
6. Wing DA, Fassett MJ, Mishell DR. Mifepristone for Preinduction Cervical Ripening Beyond 41 Weeks' Gestation: A Randomized Controlled Trial. Obstetrics & Gynecology. 2000;96(4).
7. Wang G, Cao J, Xu X, Han X, Cui H. Delivery management of a complete hydatidiform mole and co-existing viable fetus: A meta-analysis and systematic review. J Gynecol Obstet Hum Reprod. 2022 Jan 1;51(1).
8. Twiggs LB, Phillips GL. Documentation of subclinical trophoblastic embolization with invasive cardiac monitoring in a woman with a molar pregnancy: a case report. Journal of reproductive medicine. 1986;31(4):277–9.
9. Hankins GDV, Wendel GD, Snyder RR, Cunningham FG. Trophoblastic embolization during molar evacuation: central hemodynamic observations. Obstetrics & gynecology. 1987;69(3):368–72.
10. Rosen T, Sutin K, Carreno CA, Hibbett E, Funai EF. Central hemodynamic monitoring in a woman with acute respiratory insufficiency after evacuation of a complete molar pregnancy: a case report. Journal of Reproductive Medicine. 2001;46(10):916–21.

Figure 1: Timeline of events

Figure 2: Transabdominal ultrasound image of an enlarged, cystic placenta consistent with partial molar pregnancy.

Figure 3: Photograph of suspected partial molar triploid placenta after spontaneous vaginal delivery.