



Selective fetal termination for preeclampsia treatment in a dichorionic gestation with a triploid fetus: A case report

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ABSTRACT

Background: Triploidy is commonly associated with the development of early-onset preeclampsia. While previable preeclampsia is often a contraindication to prolonging pregnancy, there may be rare circumstances in which an alternative approach may be offered.

Case: A nulliparous patient with a dichorionic twin gestation, recently diagnosed triploidy in one twin, and history of chronic hypertension presented at 18 weeks of gestation with signs and symptoms suggestive of preeclampsia. After symptomatic therapy and laboratory evaluations, selective fetal termination of the affected twin was elected and performed without complications. The patient subsequently delivered a healthy newborn at 37 weeks of gestation.

Conclusion: Selective fetal termination may be considered a management option for previable preeclampsia in a dichorionic gestation with triploid fetus and was associated with a favorable outcome in this case.

1. Introduction

Preeclampsia is a common complication in pregnancy and represents a leading cause of maternal morbidity and mortality worldwide [1]. Although often diagnosed in the third trimester, preeclampsia occurring in the previable period or prior to 20 weeks of gestation has been described [2]. Fetuses without expectation for survival at the time of maternal diagnosis (e.g. extreme prematurity, lethal anomaly) are conditions precluding expectant management and, as a result, termination of pregnancy is recommended [1]. However, there may be rare circumstances where an alternative approach may be offered. This report concerns a case of previable preeclampsia in a dichorionic twin gestation with a triploid fetus managed with selective fetal termination and expectant management until delivery at term.

2. Case Presentation

A 28-year-old woman, gravida 2 para 0010, presented to labor and delivery at 18 5/7 weeks of gestation with complaints of a new-onset severe headache, nausea, vomiting, and epigastric pain. This was a twin pregnancy complicated by the diagnosis of triploidy (69, XXY) for

twin B on amniocentesis performed for sonographic findings of severe fetal growth restriction, oligohydramnios, and placentalomegaly five days prior. The patient had previously declined prenatal diagnosis after a first-trimester ultrasound revealed a cystic hygroma for twin B; cell-free DNA screening was low-risk for fetal aneuploidy. Her medical history was significant for chronic hypertension diagnosed at the beginning of pregnancy, which had been adequately controlled with extended-release nifedipine 30 mg daily. Baseline laboratory and urine evaluation for end-organ involvement was within normal limits.

On presentation, the patient reported her headache was frontal and bilateral in location, which included pain behind her eyes, and that she had not taken medication for relief of her symptoms. She had no obstetric complaints. Physical examination was notable for severe hypertension (blood pressure 152/116 mmHg) and a nontender abdomen on palpation. Initial laboratory studies were significant for a mildly elevated aspartate aminotransferase (AST; 43 units/L), uric acid (7.6 mg/dL), lactate dehydrogenase (LDH; 406 units/L), and proteinuria (protein:creatinine ratio 0.7). Serum hemoglobin, platelet count, creatinine, and coagulation studies were within normal limits. Subsequent blood pressure evaluations were within the mild range and normal fetal heart rates for both fetuses were confirmed on ultrasound.

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Table 1

Case descriptions of selective fetal termination as a treatment for multiple gestations complicated by early-onset preeclampsia.

Case Report	Gestational age at presentation (weeks)	Indication	Ultrasound findings	Outcome
Audibert et al., 2003 ¹²	28	Preeclampsia (headache, severe hypertension, proteinuria)	Severe fetal growth discordance, reversed umbilical artery blood flow in one twin	Resolution of symptoms and controlled blood pressure, vaginal delivery at 38 weeks
Heyborne & Porreco, 2004 ¹³	26	Preeclampsia (mild hypertension, elevated liver function tests)	Severe fetal growth discordance, anhydramnios, absent end diastolic velocity in umbilical artery blood flow in one twin	Resolution of symptoms and laboratory abnormalities, vaginal delivery at term
	24	Preeclampsia (severe hypertension, elevated liver function tests, proteinuria)	Severe fetal growth discordance, oligohydramnios, absent end diastolic velocity in umbilical artery blood flow in one twin	Resolution of laboratory abnormalities, vaginal delivery at 34 weeks after preterm labor
	16	Preeclampsia (mild hypertension, generalized edema, thrombocytopenia, elevated liver function tests)	Severe hydrops in one twin, suspected Ballantyne syndrome	Resolution of hypertension, edema, and laboratory abnormalities, repeat cesarean birth at term
Guerby et al., 2019 ¹⁴	25	Preeclampsia (severe hypertension, headache, epigastric pain, pulmonary edema, HELLP syndrome)	Severe fetal growth discordance, reversed umbilical artery blood flow in one twin	Resolution of symptoms and laboratory abnormalities, cesarean birth at 29 weeks after preterm labor

Despite the early gestational age, and in the setting of a triploid fetus, the patient's clinical picture was concerning for superimposed preeclampsia. Intravenous magnesium was initiated for seizure prophylaxis and daily nifedipine was increased to 60 mg daily while symptomatic treatment and serial laboratory evaluations were underway. The patient was counseled on the risks, benefits, and alternatives of all available options, including expectant management despite the increased maternal risks, termination of both fetuses, which is most commonly recommended for management of previable preeclampsia, and selective fetal termination of twin B. Counseling regarding selective fetal termination included detail of a few anecdotal reports suggesting this as an option for treatment of preeclampsia in twin gestations and that subsequent termination of twin A may be indicated at a later gestational age if contraindications to continued expectant management developed after the procedure. Utilizing shared decision-making, the patient opted for selective fetal termination of twin B. An intracardiac potassium chloride injection (2.5 mL; 2 mEq/mL) was performed under continuous ultrasound guidance using sterile technique without complications. Cardioplegia of twin B was confirmed after the procedure, while the fetal heart rate of twin A was normal. The patient was discharged after four days of observation. During that time, her symptoms resolved, blood pressure remained controlled on nifedipine 60 mg daily, and her AST and LDH normalized while uric acid remained elevated at 7.2 mg/dL. A 24-h urine collection was significant for 632 mg of protein.

The patient returned to labor and delivery for evaluation two days later after presenting with severe hypertension (blood pressure range 140–170/90–110 mmHg) at an outpatient ultrasound center. She denied any symptoms consistent with severe features of preeclampsia, her blood pressures remained in mild-range and her laboratory evaluation was unremarkable. After extensive counseling again regarding all available options, the patient desired continued expectant management and was discharged after 24 h of observation.

The remainder of the patient's pregnancy was managed under intensive maternal and fetal surveillance as an outpatient. The patient's antihypertensive requirements decreased to a point where nifedipine was transiently discontinued between 28 weeks and 34 weeks, there was no recurrence of symptoms or laboratory abnormalities consistent with preeclampsia, and her proteinuria resolved. An uncomplicated primary cesarean birth was performed under regional anesthesia at 37 3/7 weeks of gestation for a non-reassuring fetal heart tracing following induction of labor. A 2520 g female liveborn was delivered with Apgar scores of 9 and 9 at 1 and 5 min, respectively. The patient's postpartum course was uneventful and she was discharged on postoperative day 2 in stable condition with nifedipine 60 mg daily.

Written informed consent was obtained from the patient for publication of this case report.

3. Discussion

Triploidy is a rare fetal disorder in pregnancy defined as the presence of an additional set of 23 chromosomes (for a total of 69), which are either maternal (i.e. digynic) or paternal (i.e. diandric) in origin [3,4]. Given that triploidy is not compatible with life, most affected pregnancies will result in an early first-trimester loss [5,6]. However, diagnosis in the second trimester is possible and commonly associated with the development of pregnancy-related complications such as preeclampsia [3,4,7,8]. The association of triploidy with early-onset preeclampsia has been known for decades, with early reports suggesting a risk of second-trimester preeclampsia in up to 35% of cases [8]. An overlapping etiology with molar pregnancies has been suggested as placental studies in pregnancies complicated by triploidy and preeclampsia have revealed hydropic and hydatidiform molar degeneration [8,9]. This is further evident by the commonly accompanying sonographic appearance of placentomegaly and markedly elevated maternal serum free β -human chorionic gonadotropin (β -hCG) in affected pregnancies [3,8], both of which were present in this case.

Multiple gestations are also at increased risk for a number of perinatal complications [10]. Selective fetal termination is an intervention available in which a fetal reduction technique is applied to the abnormal fetus in a dichorionic gestation, which may be secondary to significant growth discordance or fetal anomalies detected at early gestational ages [10]. Despite the risk of unintended pregnancy loss of the co-twin, prolongation of pregnancy resulting in the birth of a healthy infant at or near term occurs in the majority of cases [11]. Although often performed for fetal indications, this technique has been utilized for maternal benefit in pregnancies complicated by early-onset preeclampsia [12–14].

Several cases of selective fetal termination as a treatment for multiple gestations complicated by early-onset preeclampsia resulting in prolongation of pregnancy have been reported in the literature (Table 1) [12–14]. Of the 5 reported cases, 3 (60%) delivered at term. In most of these cases, preeclampsia presented at a later gestational age with severe fetal growth discordance and the absence of a known chromosomally abnormality [12–14]. Based on these previous reports, the managing team believed selective fetal termination was a reasonable option for this patient. After the procedure, the patient's symptoms and laboratory abnormalities resolved and her antihypertensive requirements decreased. Hypothesized mechanisms for the resolution of preeclampsia after selective fetal termination include the disruption of placental blood flow, placental site involution, and cessation of the production of angiogenic factors that have been implicated in the pathophysiology of the disease [12–14].

The development of preeclampsia at an early gestational age in this

patient was in all likelihood related the presence of a triploid fetus. This patient's presentation highlights the importance of surveillance for signs and symptoms of preeclampsia in pregnancies affected by triploidy and considering selective fetal termination as an option for those encountering this clinical scenario when there are no significant contraindications precluding expectant management (e.g. uncontrolled persistent hypertension, toxic symptoms unrelieved by medication, persistent laboratory abnormalities). Patients with a triploid fetus in a multiple gestation should be counseled on the increased risk of developing previable preeclampsia, with all management options discussed even prior to the diagnosis in a timely manner. Shared decision making should be utilized and counseling regarding the potential that complete resolution of preeclampsia might not occur is of importance, as it may significantly increase maternal risk. Furthermore, in previable patients undergoing this intervention, it is possible that preeclampsia may return and worsen in the periviable period where the risk of neonatal morbidity and mortality is at its highest for the normal twin should delivery be indicated [15]. Thus, intensive surveillance as an outpatient with frequent prenatal visits, home blood pressure monitoring, and serial laboratory evaluations should be considered throughout gestation. Whether this option should be routinely recommended in all dichorionic twin gestations affected by previable preeclampsia is unclear; however, this patient's outcome in addition to those from previously reported cases are encouraging.

Contributors

All named authors were involved in this patient's care and have made substantial contributions to the concept and design, or analysis and interpretation of data, clinical assessment, or drafting the manuscript, and revising it for important intellectual content.

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The authors declare that they have no conflict of interest regarding the publication of this case report.

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