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Case Report

Delayed Delivery After Expulsion of a Papyraceous Twin Product of Selective Reduction in a Dichorionic Diamniotic Pregnancy: Case Report and Review of the Literature

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Abstract

Objective: To present the case report of the delayed delivery of a twin after the expulsion of a papyraceous twin resulting from selective reduction due to a high suspicion of aneuploidy.

Materials and Methods: We present the case of a patient with a bi-chorionic bi-amniotic twin pregnancy, undergoing selective reduction at 18 4/7 week of one of the fetuses due to high suspicion of aneuploidy. Later, at 31 4/7 week she presented expulsion of the papyraceous fetus without complications. The delivery of the surviving twin was delayed, and the pregnancy ended at 34 5/7 weeks. A systematic search of information was conducted in the PUBMED database regarding indications, technique, and maternal and neonatal outcomes of twin pregnancies undergoing selective reduction of one of the fetuses. The narrative summary of the findings is presented.

Results: After filtering the information, ten (10) articles were reviewed and subsequently a clinical practice guideline and an ACOG bulletin were included as gray literature. It was found that the most frequent indication for selective reduction was structural and chromosomal anomalies, especially in procedures performed over 15 weeks. The aOR for the reduction of preterm delivery was 0.20 (95% CI 0.29 - 0.81) with rates of preterm delivery of 18%, likewise a higher average birth weight was found in patients undergoing fetal reduction before 15 weeks. The mean age at delivery was higher in this group of patients. The survival percentage was higher in late intervention, likewise delivery before 32 weeks had lower proportions in those patients taken to late intervention, favoring its performance at this gestational age.

Keywords: *Multiple Pregnancy; Fetal Reduction; Selective Reduction; Papyraceous Twin; Case Report*

Introduction

Multiple pregnancy has been associated with an increasing morbidity rate for the mother and the fetus; the risk is augmented in the measure of the number of fetuses and also depends on chorionicity. Some maternal complications as hypertensive disorders, gestational diabetes, anemia, postpartum hemorrhage and maternal death are 3 to 7 times more frequent in comparison with singleton pregnancies, especially in patients with risk factors as chronic maternal disease, uterine anomalies and preterm labor in a previous pregnancy [1-3]. On the other hand, the fetal complications have been associated specially with the own effects of prematurity below 32 weeks and the low birthweight below 1500 grams, where multiple pregnancies correspond to the 20.3% of the preterm labor [4-6].

The American College of Obstetricians and Gynecologists (ACOG) estimates the medical expenses are four (4) times bigger for the management of twins and ten (10) times bigger in pregnancies of higher order, making this a important phenomenon not only from the medical and economical point of view, but also social, with higher rates of maternal depression and child abuse [1]. Multiple pregnancy supposes an increase in the risk for chromosomic and structural anomalies [7]. Monochorionic twin pregnancy are 3 to 4 times more related with structural anomalies in comparison with singleton pregnancies, of which only 10 to 15% of cases present with compromise of both fetuses; meanwhile the risk of aneuploidy is similar among these two [7,8]. On the other hand, dichorionic twin pregnancies have a twofold risk for structural and chromosomic anomalies in comparison with singleton pregnancies, given the fact that each fetus has an independent risk. Out of these pregnancies, 80% of structural or chromosomic anomalies affect only one of the fetuses [7,9]. The presence of any anomaly constitutes a threat for the healthy twin with a significant increase in the risk of preterm labor and perinatal mortality. Additionally, the risk rises with the severity of the anomaly [9].

Multifetal reduction is the procedure for which one or various pregnancies are interrupted with the objective of reducing the number of evolving fetuses and with that reduce the maternal and perinatal risks, as well as reducing the socioeconomical and familiar challenges. It is recommended that the reduction is performed in pregnancies of superior order (3 or more fetuses) due to higher mortality rates that each additional fetus offers [1,2,7], as well as the cases in which coexist the risk for preterm birth, personal history or the patient expresses her desire for the procedure to be performed [4,7].

Selective reduction looks for the interruption of the pregnancy of fetuses with structural anomalies as discordant anomalies, intrauterine growth restriction and chromosomic anomalies in a multiple pregnancy. The goal of the procedure is to improve the overall prognosis of the pregnancy when the abnormal fetus is presented [2,10]. This is the case that we present up next, in which by the ultrasound findings, the discordance among the fetuses made the medical team offer a patient a selective reduction of the abnormal fetus with the goal of improving the overall prognosis of the healthy fetus. The term papyraceous twin is conferred to the reduced twin or the one that presents with fetal demise, ceases to grow, and has an appearance of mummification [11].

The procedure technique for the fetal reduction or selective reduction according to the case, will depend on the chorionicity of the multiple pregnancy. In the bichorial twin pregnancy the procedure will be performed by the intracardiac, or intra-aortic injection of potassium chloride concentrate 15% with a spinocan needle under sonographic visualization, as in the monochorionic twin pregnancies the procedure will be the ablation of the umbilical cord flow, as the potassium chloride can affect the healthy fetus by crossing the placental vessels that communicate both fetuses [2-4,6,8].

Case Presentation

A 37-year-old patient with her first pregnancy, presents with a bichorial-biamniotic pregnancy of 18 weeks and 2 days calculated by her Last Menstrual Period (LMP) at the Maternal Fetal Medicine Department at Clinica Colsanitas in Bogota, Colombia for evaluation in the Congenital Anomalies Board. The patient had a first trimester ultrasound in which was found a reverse ductus venosus and a discor-

dance in one of the fetuses growth of 33% by Crown-Rump length (Figure 1). As relevant medical history, the patient had had am endometrial polyp resection by hysteroscopy and uterine leiomyomatosis.

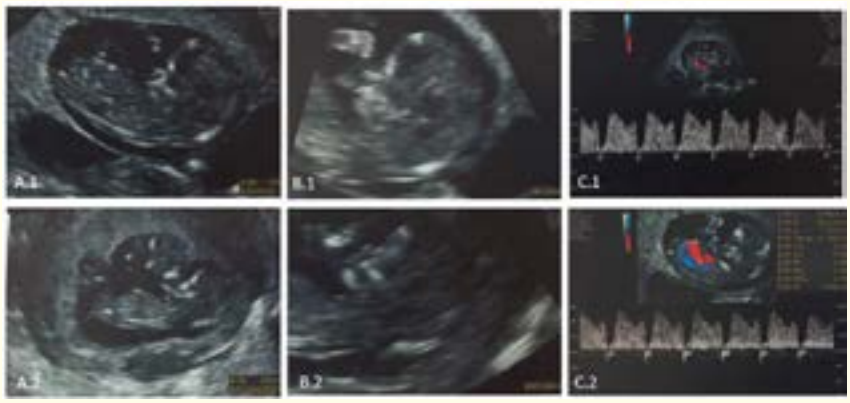

Figure 1: *First trimester US. A. Discordance in CRL of both fetuses: Twin A with CRL for 11 weeks and 3 days (A.1) and Twin B with CRL for 12 weeks and 5 days (A.2). B. Measure of nucal translucency: Twin A (B.1) Twin B (B.2). C. Ductus venosus: Reverse ductus for Twin A (C.1) and normal ductus for Twin B (C.2). Pictures taken with patients' authorization.*

An ultrasound evaluation was performed finding: Fetus A with anhydramnios, large echogenic kidneys with absent bladder, with growth percentile in below 1%; Fetus B with normal anatomic evaluation, normal amniotic fluid with growth percentile at 60%. Weight discordance was about 49%. It was considered fetus A had severe Intrauterine Growth Restriction with poor perinatal prognosis (Figure 2).

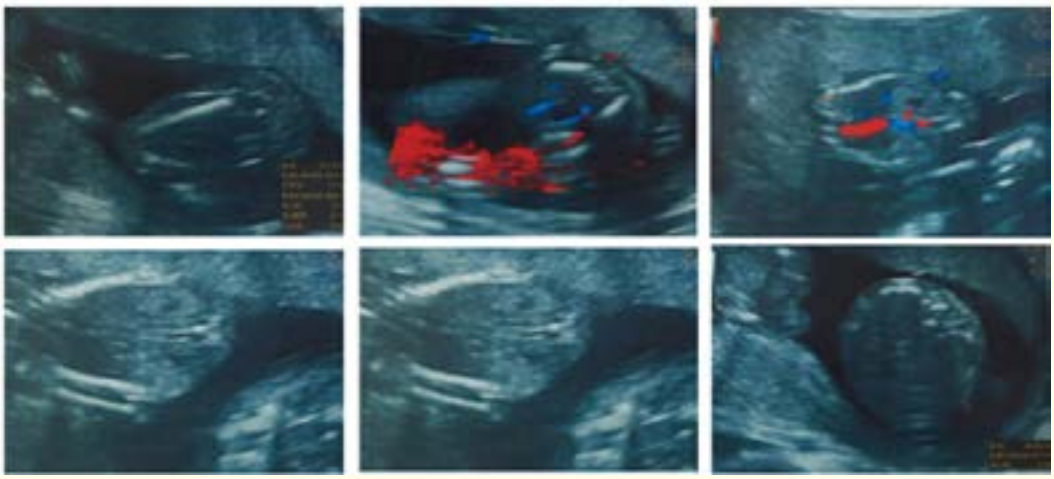

Figure 2: *Pictures of the ultrasound taken at the anomalies board. Pictures taken with the patient´s authorization.*

A prenatal counselling was offered regarding pregnancy prognosis. The selective reduction of the affected fetus was offered. At 18 weeks, under sonographic visualization, intracardiac injection of 1 cc of potassium chloride was performed until asystole was achieved. After an uncomplicated procedure and confirmation of asystole of fetus A, the wellbeing of fetus B was confirmed. The patient was discharged from the hospital 24 hours after the procedure.

At the outpatient facilities the patient was assessed 15 days later by the Board of Anomalies finding a healthy patient. With the ultrasound evaluation a healthy fetus was found, with an estimate weight of 417 grams, adequate volume of amniotic fluid, with an anterior corporal placenta. Adjacent to the healthy fetus, the reduced fetus was observed with no relevant findings. During the consecutive prenatal assessment no other abnormality was found.

At 31 weeks and 4 days patient consulted the emergency department for mass sensation in the vaginal canal. On admission the patient was found with no amniorrhea or genital bleeding. During physical examination, the presence of mass in the cervical canal was evident and the cervicometry showed papyraceous fetus in the cervical canal (Figure 3A). It was proceeded to manual extraction of papyraceous fetus which was covered by membranes. The manual removal was uncomplicated (Figure 3B). It drew attention the modifications of the cervix (dilation of 3 cm with short cervix) (Figure 3C) and it was decided to hospitalize the patient for surveillance and follow-up. During hospitalization, the patient received fetal maturation regimen and prophylactic antibiotic with clindamycin/gentamicin.

As part of the approach to preterm labor, an infectious profile was requested with finding of leukocytosis and neutrophilia, negative C-reactive protein and negative procalcitonin. Fetal well-being was verified by biophysical profile 10/10, estimated fetal weight was 1957 grams. Follow-up cervicometry was performed finding residual cervix of 1 cm (Figure 3D). Surveillance was indicated for 48 hours and because of satisfactory evolution it was decided to discharge her with recommendations.

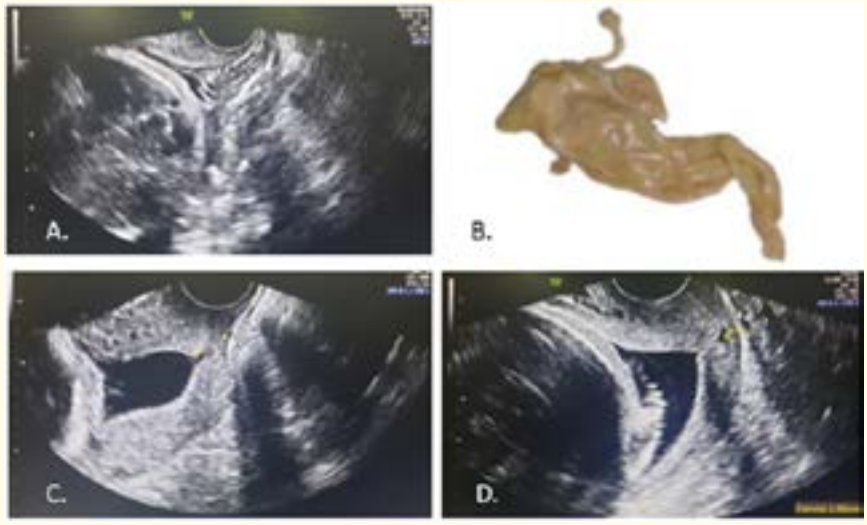

Figure 3: *(A) Ultrasound performed of papyraceous twin in endocervical canal, the fetal spine of the reduced twin is observed. (B) Photograph of papyraceous twin extracted from vaginal canal (C) Ultrasound of cervical canal after expulsion of papyraceous twin. (D) Cervicometry performed 24 hours after expulsion of papyraceous twin). Photo taken with patient's permission.*

At 34 5/7 weeks the patient enters the emergency room with uterine activity and fluid outflow through the vagina. On physical examination it was found cervical dilation of 5 cm and 90% erasure with clear amniorrhea. Premature rupture of membranes and preterm labor in advanced stage were diagnosed and the patient was admitted in the delivery room for conduction and monitoring of labor. An unsatisfactory fetal status was evidenced by ACOG III fetal monitory staging and segmental cesarean section (Robson category I) was indicated, obtaining a live male newborn with a weight of 2460 grams and a height of 46 cm. The APGAR score at 5 minutes was 8 points.

During the adaptation process, the newborn presented respiratory difficulty with a 5-point Silverman, so the pediatric service decided to start CPAP ventilation and transfer to the neonatal unit for comprehensive management. Upon admission, a chest x-ray was performed with findings of interstitial opacities without consolidation zones. The newborn presented episodes of desaturation that required an increase in FiO_2, so diagnosis of respiratory distress due to pulmonary surfactant deficiency was made. After orotracheal intubation, pulmonary surfactant was passed through a smooth technique and first-line prophylactic antibiotic coverage (ampicillin/gentamicin) was initiated. The baby presented adequate clinical evolution with improvement of respiratory parameters so he was passed to non-invasive mechanical ventilation, chest x-ray control with evidence of anterior pneumothorax of < 20% without requiring invasive interventions. During his stay in the neonatal unit, he adequately tolerated oxygen by nasal cannula. Follow-up of chest x-ray showed resolution of pneumothorax and blood cultures taken at admission were negative, so after 10 days he was discharged with follow-up by kangaroo care program. The patient leaves after 24 hours due to satisfactory clinical evolution during the immediate puerperium.

Methods

We systematically searched on PUBMED database for information with the descriptors ((“Pregnancy, Twin”[Mesh]) AND (“Pregnancy Reduction, Multifetal”[Mesh])) AND (“Pregnancy Outcome”[Mesh])), limiting the research from 2017 to 2022. The studies searched for information regarding indications, technique and maternal and neonatal outcomes of twin pregnancies subjected to selective reduction of one of the fetuses.

Results

We found a total of 35 results: 18 articles were discarded by title and 7 by abstract. A total of 10 articles were reviewed: 1 literature review [2], 1 systematic literature review [5] and 8 retrospective cohort studies [3,4,6,8-10,12,13] were selected. A clinical practice guideline [7] and an ACOG bulletin were added as grey literature [1].

The 8 retrospective cohort studies included a total of 1115 patients leading to selective reduction and 2´018.015 patients who completed multiple pregnancy until the time of delivery. Within these studies, the analysis of the outcomes with respect to reduced pregnancies was carried out compared to those not led to reduction, the gestational age at which the intervention is performed, the indication of the selective reduction and the multiplicity to which the pregnancies were reduced.

Within the population of patients taken to selective reduction, it was identified that 67.3% of patients opted for selective reduction under the indication of structural and chromosomal abnormalities, 10.2% due to risk factors for preterm delivery and 20.4% due to the patient’s request [4]. Likewise, genetic and congenital alterations were found in greater proportion in those patients taken to reduction above 15 weeks due to the moment of the alterations screening and the decision making of the reduction [9].

It was found higher survival rates in those pregnancies led to late fetal reduction (> 24 weeks) and lower in those led to early (< 18 weeks) and intermediate (18 - 23.6 weeks) reduction (100%, 86% and 96.9% respectively) being the last option the intervention related to lower survival rates [8,13]. Lower rates of abortion were also identified in pregnancies leading to reduction before 15 weeks compared to those carried to reduction above this gestational age (11.4% vs 33.3% respectively) [9]. Pregnancy global loss rates of 9.6% were reported in patients operated [13] while the overall live birth rate depending on the gestational age at which the intervention was

performed was 66.6%, 100% and 100% for early, intermediate, and late interventions respectively ($p < 0.001$) [13]. The aOR for severe neonatal morbidity and perinatal death for unreduced vs. reduced pregnancies was 1.57 (95% CI 0.74 - 3.33) [10].

For the decrease in the risk of prematurity, an aOR of 0.206 (95% CI 0.06 - 0.65) was found, with preterm birth rates of 18%. In patients with expectant management, higher OR were found for delivery before 37 weeks compared with deliveries before 34 weeks (5.62, 95% CI 3.67 - 8.61, $P < 0.001$ Vs 2.2, 95% CI 1.2 - 4.11, $p < 0.001$) [4]. Prematurity before 32 weeks presented higher proportions in those patients taken to early intervention compared to those taken to intermediate or late intervention (27%, 18.2% and 9.5% respectively) [8], likewise the average age at the time of delivery was higher in patients taken to fetal reduction with respect to that found in those non-reduced pregnancies (36.8 ± 3 - 35.4 weeks Vs 35.6 ± 2.4 Vs 34.1 sem) [4,6]. Similarly, the average gestational age at the time of delivery in patients with interventions performed early was 37 3/7 weeks and 34 6/7 weeks for late [4].

The average birth weight identified in studies in patients with reduced pregnancies was 2705.3 ± 708.7 and 2276.7 ± 543.9g in those not reduced [4]. Higher mean fetal weight ranges were identified in patients reduced before 15 weeks with respect to those reduced above that gestational age (2803 - 3058g vs 2449 - 2878g) [9]. For the very low birth weight outcome, expectant management had an OR of 2.7 (95% CI 1.53 - 4.76, $P < 0.001$) and the OR for low birth weight in the expectant management group found was 0.3 (95% CI 1.89 - 4.76, $p < 0.01$) [12].

Several studies reviewed favored late reductions (30 - 32 weeks) when identification of abnormalities or gestational age of decision-making required the procedure to be performed above 15 - 18 weeks. In this group, no data on pregnancy loss and preterm birth rates were found, although they were higher in the late intervention group had a lower proportion for extremely preterm birth outcomes compared to very preterm (0% for late intervention, 36.3% for intermediate interventions and 11.1% for early interventions) [13]. Comparing early interventions with late interventions we found an OR of 0.44 for delivery before 37 weeks (95% CI 0.21 - 0.94, p 0.033) and 0.0092 for delivery before 34 weeks (95% CI 0.016 - 0.54, p 0.008) [3].

Additional findings found favored fetal reduction vs conservative management in cesarean delivery outcomes (OR 5.53, 95% CI 3.60 - 8.49, $p < 0.001$) and in terms of increase of incidence of preeclampsia (OR 0.33, 95% CI 1.60 - 6.96, $p < 0.001$) [5,12].

Discussion

In the clinical case presented, the clinical course of a patient taken to selective reduction under suspicion of chromosomal anomaly and with evident structural anomalies is exposed. This intervention was performed in the gestational age range for intermediate intervention for some of the studies found and for other studies in late intervention ranges. The procedure could be performed without complications and the recovery and follow-up after it was adequate which differs in results found in some of the studies with respect to the interventions carried out at that gestational age. The spontaneous expulsion of a papyraceous twin and subsequent delayed delivery was not reported in any of the studies reviewed making the presentation of this case unusual. Similarly, the gestational age at the time of delivery in the patient (34 5/7 weeks) was consistent with data found in the outcomes presented in different articles.

The data found in the reviewed studies that favor the performance of the intervention of late fetal reduction at 30 - 32 weeks guide the performance of procedure in that gestational age in patients with decision of the procedure taken above 15 - 18 weeks. However, in many of the studies reviewed that were conducted in different countries, legislation regarding the maximum gestational age at which the intervention can be performed limits their external validity.

Conclusion

Selective reduction in multiple pregnancies becomes an important intervention to improve different outcomes among which are mainly gestational at the time of delivery, the rate of preterm births and birth weight. Likewise, the outcomes found regarding to gestational

age at the time of the reduction were better in patients with early reductions compared to those performed later in pregnancy. To make a recommendation to perform the intervention at gestational ages above 30 weeks, it is necessary to conduct a greater number of studies with a larger number of cases.

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