

Twin Fetuses Papyraeci in a Spontaneous Triplet Pregnancy Presenting with Unexplained Preterm Contractions

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Abstract

Fetus papyraecia in a triplet pregnancy is indeed rare and can pose serious management challenges. These challenges are more pronounced where facilities for monitoring are either inadequate or nonexistent. A 39-year-old, grand multipara multipara was referred to the University of Maiduguri Teaching Hospital at 27 weeks gestation with preterm contractions. Materno fetal monitoring did not reveal the cause of the preterm contractions. She was delivered via caesarean section, at 36 weeks of gestation, on account of decreased fetal movement and the products were a live female fetus weighing 2.3 kg and two male papyraceous fetuses weighing 150 g and 130 g, respectively.

Keywords: Caesarean section, Fetus papyraceous, Preterm contractions, Triplet pregnancy

Introduction

The retention of a mummified parchment – like remains of a dead fetus (es) in multiple pregnancy in the second trimester of pregnancy in association with a viable twin is known as fetus papyraceous.^[1,2] Its incidence is 1 in 17,000-20,000 pregnancies or 2.3% of all twin pregnancies.^[2,3] The incidence of two fetuses papyraceous in a triplet pregnancy is 1 in 32,800.^[4] The incidence quoted is likely to be higher in Nigeria, a country with the highest twinning rate in the world.^[5] In Nigeria were 36% of mothers do not receive antenatal care and with 62% of births taking place at home^[6] the true incidence of fetus papyraceous or any complication of multiple pregnancy is likely to be underreported. Disseminated intravascular coagulation (DIC) which complicates demise of singleton pregnancies especially if prolonged is uncommon with fetus papyraceous.^[7]

Case Report

A 39-year-old Nigerian grand multipara, was referred to University of Maiduguri Teaching Hospital at 27 weeks of gestation on account of persistent uterine contraction of

3 days prior to presentation. An accompanying ultrasound scan report revealed a live singleton fetus at 27 weeks 2 days gestation, presenting breech, with a fundally placed placenta. The history and general examination were unremarkable. The pulse rate was 86 beat per min and her blood pressure was 130/80 mmHg. The fundal height was 29 cm which was compatible with her date of 27 weeks. The fetal heart rate was 142 beats per min. There was one palpable uterine contraction, of moderate intensity and lasted 25 s. Vaginal examination was unremarkable. Results of initial investigations were: Packed cell volume (PCV) 28%, white blood cells $8 \times 10^9/L$, platelet normal, high vaginal swab (HVS) for microscopy, culture and sensitivity (MCS) – normal, urine MCS – normal, fetal kick count – normal, up until 31st March, 2010. A repeat ultrasound scan confirmed earlier report from the referring centre. She was placed on intravenous fluid, cefuroxime axetil 500 mg twice daily for 10 days, pentazocin 60 mg once in 8 h for 24 h and continued on tablets of ibuprofen. During the 9 weeks period of admission she had received steroids for fetal lung maturity, nifedipine, phenobarbitone, and hematinics. The contractions never abated completely but it waxed and waned despite the tocolytics. An ultrasound scan at 34 weeks put the estimated fetal weight at 2320 g. She complained of reduced fetal movement at 36 weeks, 2 days and a review of the fetal kick count showed 5 kicks in the preceding 14 h. Her PCV then was 32% and urinalysis was negative for protein and glucose. She was offered emergency caesarean section and the products were two macerated male fetuses with monochorionic placenta, weighing 150 g and 130 g. The live female fetus weighed 2.3 kg with Apgar score of 7 and 10 in 1st and 5th min,

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respectively. The placenta of the surviving twin and that of the twin papyraceous weighed 500 g and 200 g, respectively. She was discharged on the third post-operative day with her baby. Both had no complaints at the 6th week post-natal visit.

Discussion

The intrauterine death of a fetus (es) with a surviving twin is a challenging case to manage. The chorionicity, gestational age at diagnosis and other complications specific to the pregnancy may influence the management option.^[8] Ultrasound scan is invaluable in the diagnosis of twin pregnancy and its complication.^[9] The failure of serial ultrasound scan to detect fetuses papyraceous in the index case has also been reported elsewhere.^[10,11] This is thought to be due to anatomical position and technical difficulties.^[10] Maternal complications associated with fetus papyraceous include DIC, preeclampsia, hydramnious, antepartum hemorrhage, preterm labor, and anemia.^[12,13] The index case presented with premature contractions before the age of viability (28 weeks in Nigeria). Preterm delivery commonly by caesarean section is the usual course in fetal death in triplet pregnancies.^[12] Our case had caesarean section at 36 weeks on account of decreased fetal movement. The estimated fetal weight on ultrasound 2 weeks prior to the caesarean delivery was 2320 g and the weight at delivery was 2.3 kg. This may be a reflection of growth retardation which is a frequent complication before fetal death.^[12] Where cardiotocograph (CTG) is not available as in our case the fetal kick count was valuable in the decision to terminate the pregnancy. Although, the coagulation status was not monitored, as the cause of the preterm contraction became obvious only after the caesarean section, no obvious coagulopathy was noticed during the 9 weeks period on admission. Similar observations on intact coagulation system after a fetus papyraceous was diagnosed have been reported.^[7] The patient, on admission at 27 weeks had mild anemia but it was difficult to ascribe the anemia to fetus papyraceous as the commonest cause of anemia in Nigeria is nutritional deficiency.^[13] The surviving twin of a papyraceous is prone to neurological complications, cerebral palsy, renal failure, intrauterine or neonatal deaths.^[14,15] Because the twin papyraeci shared the same placenta,[Figure 1] one seem to have died shortly after the other as may be inferred from the differential fetal weights of 150 g and 130 g. The postulated cause of death in a monochorionic placentation as in this case of the twin papyraeci may be that thromboplastic proteins from the first twin to die (130 g) were transferred to the then surviving (150 g) circulation, resulting in DIC. The other explanation may be that massive blood loss may have taken place from the second twin to die into the first to die who had a more relaxed circulation through vascular anastomosis.^[16] A more plausible explanation is that fetofetal transfusion imbalance may lead to fetal or infant death or cerebral palsy and/or congenital anomaly in surviving twins.^[17-19] Fetal death could also be a result of congenital malformations, umbilical cord prolapsed or hydrops.^[12] The survivor in



Figure 1: Picture of twin fetus papyraeci with the placenta of the surviving twin

this triplet pregnancy had no obvious complications at birth probably because of its separate placenta without vascular anastomosis.^[14] Cerebral palsy is unlikely in this case as the survivor has a separate placenta from the twin papyraeci.

This case report suggests that the cause of “unexplained preterm contractions” could be complications associated with multiple pregnancies. When such cases are encountered, detailed sonographic assessment by an experienced sonologist may be all that is required to unravel the cause for the “unexplained contractions” especially in regions with high twinning rate like Nigeria.

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