

Fetus Papyraceous in Uniovular Twin; Death of One Twin in Early Third Trimester and Successful Outcome of Other Twin at Term: A Rare Case Report

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Abstract

Fetus papyraceous is a rare obstetric complication in multiple gestations, the incidence of which in case of twin is 1 in 12,500. It may be associated with high maternal and fetal complications including disseminated intravascular coagulation induced death and organ damage. Fetus papyraceous can occur both in uniovular and binovular twin pregnancies. However intrauterine death is three times more common in case of uniovular twin because of frequency of vascular connections (85-98%) in monochorionic placenta. Here, we report a case of uniovular twin pregnancy with one twin fetus papyraceous in early third trimester and pregnancy continued till term with a successful outcome of surviving co-twin.

Keywords: Fetus papyraceous; Uniovular twin; Binovular twin; Twin-twin transfusion syndrome

Introduction

The term fetus papyraceous is used to describe a flattened, mummified fetus associated with a viable multiple gestations. In a multiple pregnancy a dead fetus becoming papyraceous and the surviving fetus growing normally is a rare event in obstetrics [1]. Incidence of fetus papyraceous in twin is 1 in 12,500. Fetus papyraceous occurs when a fetus dies in utero usually in second trimester and is not expelled out, resulting in its atrophy and mummification [2]. Here, we report a case of uniovular twin pregnancy with one twin fetus papyraceous in early third trimester and pregnancy continued till term with a successful outcome of surviving co-twin.

Case Report

A 23 year old pregnant women with 9 months of amenorrhoea attended emergency labour room of our hospital on 17th July, 2013 with complaints of labour pain. She was G₂P₁ with no living issue. Her previous pregnancy was a term intrauterine death which she delivered spontaneously vaginally. No cause of her previous pregnancy mishap was known.

Her current pregnancy was spontaneous conception and there was no family history of multiple pregnancy or child with congenital abnormality. She had only one ante-natal checkup in first trimester in a local primary health center and was irregularly taking folic acid tablets. In second trimester she took two doses of tetanus toxoid injection and had undergone her first ultrasonography done at 20 weeks of gestation. She was referred to our hospital with the sonography report. The ultrasonography showed twin live gestation in-utero with both breech with single placenta with normal parameters corresponding to 20 weeks 6 days in the first twin and 21 weeks 1 day in the 2nd twin (Figure 1). Appropriate counseling was done and she was advised for regular follow up in our hospital, but she wanted to see local primary health center in view of long travel distance to our hospital.

The patient again came for a follow up in our hospital at about 7 months of gestation. An ultrasonography was done in our hospital which showed twin live fetus in utero with single placenta in fundus and anterior wall without any evidence of inter-twin membrane suggestive of monochorionic monoamniotic twin pregnancy (Figure 2) with first twin at 27 weeks 2 days in cephalic presentation and 2nd twin at 27 weeks in breech presentation. No gross anomaly was noted in either of the twin. She was prescribed iron and calcium tablets in high doses and advised for follow up visit in 2 weeks.

Ten days later the patient had decreased fetal movement and she went to the local primary health center and where doctor performed

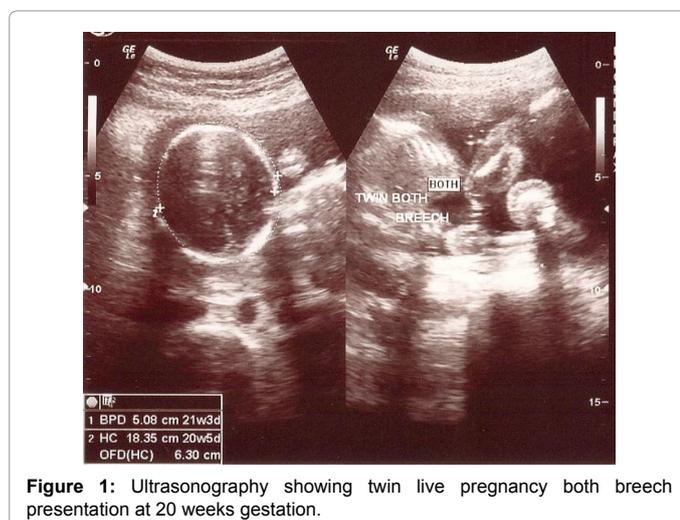


Figure 1: Ultrasonography showing twin live pregnancy both breech presentation at 20 weeks gestation.

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Figure 2: Ultrasonography showing single placenta in fundus and anterior wall without any evidence of intertwine membrane suggestive of monochorionic monoamniotic twin (Uniovular twin).



Figure 3: Healthy baby 2.9 kg, Fetus papyraceous, and single placenta

an ultrasonography and noted intrauterine demise of one twin (only report available, no formal image was available). She was counseled and advised to attend tertiary hospital; however she ignored the advice of the doctor. She attended our hospital at 37 completed weeks of gestation with labor pain.

General physical examination was unremarkable at the time of admission. Her routine laboratory examination and coagulation profile were within normal limits. Obstetrical examination revealed height of the fundus at 38 cm and abdominal girth 96 cm and fetal heart rate was 146/min regular and clear. An urgent ultrasonography showed first twin in breech presentation and other twin was fetus papyraceous compressed by healthy twin. Pelvic examination revealed is 2 cm dilated and cervix 50% effaced with intact membrane.

An emergency cesarean section was performed in view of breech presentation and her previous bad obstetric history. At cesarean section the first baby delivered was a healthy female 2.9 kg with Apgar score of 6/10 and 9/10 at one and 5 minutes respectively. Following the delivery of the healthy baby a grey white, mummified and compressed fetus papyraceous was delivered which weighed 700 grams (Figure 3). The sex was female. The head, trunk and extremities could be distinctly identified although there were marked distortions because of compression. There were gross bony

malformations of the upper and lower extremity (Figure 4). The placenta measured approximately 16 cm×17 cm×2 cm and weighed 650 grams. The placenta was monochorionic monoamniotic and no visible abnormality was noted in placenta or umbilical cord.

Postoperatively both mother and surviving twin baby had uneventful recovery and were discharged on 5th day in a healthy state. The patient was advised for postnatal check up at 6 weeks. The baby was thoroughly examined by pediatrician and has been perfectly normal. Immunization was done as per national immunization schedule.

Discussion

Death of a twin in the first trimester rarely complicates the pregnancy. However, death of a twin in second or third trimester is a rare occurrence and may be associated with high fetal and maternal complications [3]. Fetus papyraceous can occur in both uniovular and binovular twins which may be due to twin-twin transfusion syndrome, cord complications, placental insufficiency and congenital anomalies [4]. Main concern of fetus papyraceous is its effect on mother and surviving co-twin which depends on gestational age. Maternal complications include preterm labor, infection from a retained fetus, severe puerperal hemorrhage, consumptive coagulopathy and obstructed labor by a low-lying fetus papyraceous causing dystocia leading to cesarean delivery. The effects on surviving twin include risk of cerebral palsy, congenital abnormalities like neural tube defects (NTDs), optic nerve hypoplasia, hypoxic ischemic lesions of white matter, microcephaly, post hydrocephalus, bilateral renal cortical necrosis, unilateral absence of kidney, gastrointestinal tract atresia, gastroschisis, hemifacial microsomia and aplasia cutis [5]. The surviving twin may be complicated by twin embolization syndrome. The emboli



Figure 4: Fetus papyraceous with visible gross bony malformations of upper and lower extremity

results from placental and fetal thromboplastin or necrosed fragments of the dead placenta causing disseminated intravascular coagulation which can damage organs like brain, kidney and cause intrauterine death or premature birth [6]. However, the incidence of this complication is very low in case of twin pregnancies [7]. In our case the surviving twin was healthy with good birth weight of 2900 grams. There were obvious musculoskeletal abnormalities in the dead fetus, which may have resulted from prolonged maceration and compression in utero.

If fetus papyraceous is diagnosed antenately serial evaluation of the surviving fetus by sonography, biophysical profile, doppler and maternal coagulation factors should be done serially. Zygosity and chorionocity evaluation should be performed antenately. The timing and procedure for the termination of a pregnancy with a surviving twin are determined primarily by the maturity of the fetus and type of placenta. In case of dizygotic twin risk to the surviving twin is not increased and spontaneous onset of labor at term can be awaited under close monitoring of maternal and fetal parameters. In case of monochorionic twin risk to surviving twin is significantly increased through shunt communications which can lead to DIC induced death and organ damage [8,9]. Okamura et al. [10]. found out that survival difference due to chorionocity is because of frequency of vascular connections (85-98%) in monochorionic placenta. Close supervision of the condition of the mother and surviving child is essential and the mother should be admitted to hospital for this purpose if required.

In conclusion fetal death in a multiple pregnancy with one or more normally surviving fetus is unusual. It is very important

to make a diagnosis in time to prevent severe complications. It is important to reassure the patient that normal outcome is expected in most of the cases. If possible delivery should be scheduled at tertiary center.

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