Case Report

Fetus papyraceous in a monozygotic twin with a normal co-twin

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A fetus papyraceous was delivered by a 26 year primigravida (G1P1A0) in a normal vaginal delivery with a healthy female baby weighing 2.96 kg. The two fetuses were attached to a single placenta by two umbilical cords one longer and the other short. Ultrasonography showed monochorionic diamniotic pregnancy. On examination, placenta was bilobed, and adherent with velamentous insertion. Detailed study on the prenatal history of the mother, type of placentation, and prenatal diagnosis of fetuses were done. The causes of fetus papyraceous and the effects of fetus papyraceous on the mother and surviving fetus were studied in detail. It was observed that there were no hereditary and maternal reasons for this unusual event. The single fetal death might be due to circulatory problems followed by monochorionicity and velamentous insertion of placenta. It was also noted that the surviving twin was healthy and no congenital anomalies were found.

Key words: Intrauterine death, papyraceous, monochorionic, velamentous, bilobed placenta.

INTRODUCTION

Monozygotic twins are derived from a single fertilized ovum and their placentation depends upon the stage at which duplication occurs. If splitting occurs between 3rd and 8th day after fertilization, the blastocyst separates into two embryos and the trophoblast will develop as a single placenta. The two embryos implant in close proximity in two amniotic sacs, this placentation is categorized as monochorionic diamniotic [Djafar et al., 2006]. Single fetal death in twin pregnancy is not rare but it causes serious maternal and fetal risks. Fetus Papyraceous is the compressed and mumified parchment-like remains of a dead twin which is retained in utero in early second trimester. It is usually discovered among the placenta and membranes of its well developed co-twin [Airede and Ahmed, 2005]. It is a macerated, tiny, full formed fetus which is usually dry and papery because the amniotic fluid and fluid content of the dead fetus and of placental tissue gets absorbed and the fetus gets flattened and compressed between membranes of the living co-twin and the uterine wall [Mittal and Khanna, 2007].

This process is unusual since fetal demise usually results in spontaneous abortion, still birth or complete resorption in early pregnancy. The incidence of fetus papyraceous occur 1 in 17000-20000 pregnancies and 1 in 184 to 200 twin pregnancies. Fetus papyraceous can occur both in uniovular and biovular twins but is more common in uniovular twins. The occurrence of fetus papyraceous frequently associated with perinatal morbidity in the other twin as the surviving fetus can have problems due to Twin Embolisation Syndrome (TES) making antenatal diagnosis of this condition desirable [Rasheed and Amjad, 2008]. The present case report is of a fetus papyraceous diagnosed at 26th weeks of gestation in a monochorionic twin pregnancy while the surviving twin was delivered at 39th week of gestation in a healthy state. The fetus papyraceus and the live fetus were attached to a single placenta by a velamentous insertion of umbilical cord.

CASE REPORT

A 26 year old primigravida with a normal menstrual history was diagnosed at six weeks gestation with monochorionic diamniotic twin pregnancy by sonography. Past history was unremarkable of any surgical,
gynecological or obstetrical events. There was no history of twinning in family. Biochemical investigations were normal. Sonography at 17th week showed two intrauterine diamniotic fetuses. Only a single placenta was detected. The fetuses measured, 216 grams and 179 grams. Repeat sonography at 26th weeks showed only one live fetus. The two fetuses appeared to be separated by an amniotic membrane. The family was counseled with regard to fetomaternal risks. Sonography revealed a normal growing live fetus. Placenta, amniotic fluid and membranes were normal. The dead fetus measured BPD-36.8 mm, HC-154 mm and FL-32.3 mm. At 31st week the patient was admitted for reduced fetal movements. On examination leaking and bleeding were not found. The patient felt fetal movements after 24 h and was discharged. Results of the sonography at 36th week revealed normal vitals and the fetal weight of 2621 ± 218 gram. The amniotic fluid was adequate. At 39th week the patient was admitted and oxytocin given. The fetus papyraceous was delivered early followed by the second twin weighing about 2.96 kg. After five minutes, the placenta (about 950 grams) and the membranes were expelled. The placenta was monochorionic diamniotic, bilobed, adherent with velamentous insertion of umbilical cord (Figures 1 and 2). Fetus papyraceous was attached to the same placenta by a separate umbilical cord. The patient was discharged after three days in a healthy state.

DISCUSSION

Twin gestation or multiple pregnancy is a common finding in today’s era due to Assisted Reproductive techniques, in vitro fertilisation and highly advanced prenatal methods [Sinha R et al. 2010]. A fetal death in multiple pregnancies with one or more surviving fetus is unusual [Upadhyaya I et al. 2009]. Incidence of fetus papyraceous in twins is one in 12,500 [Mittal PS, Khanna M, 2007]. Intrauterine death of a single fetus in the 2nd or 3rd trimester is uncommon and has been associated with increased risk of mortality and morbidity to the surviving twin. Prenatal mortality is high in monochorionic twin pregnancies than dichorionic twin pregnancies and the prevalence is about 50 to 70% [Woo et al., 2000]. In the present case the IUD occurred approximately in 26th week according to ultra sonography and was retained in utero till 39th week without any complications to the surviving fetus and mother.

Reported causes of IUD as Twin Transfusion Syndrome, cord knot, cord stricture, placental insufficiency, intrauterine growth retardation and congenital anomalies [Woo et al., 2000]. The routine placental determination by sonography is necessary because the mortality and morbidity depended more on chorionicity than zygosity. In this case the first ultrasound at 6th week of gestation itself detected the monochorionic nature. Ultrasound reported that the IUD occurred in the
late 2nd trimester (24th week) and the dead fetus showed all normal fetal measurements.

Normally blood vessels run from the placenta via the umbilical cord to the baby. In velamentous insertion, umbilical cord is attached to the membranes outside the placenta and the umbilical vessels ramify and anastomose there. The possibility of velamentous insertion is 1 to 2% in all pregnancies [Djafar et al., 2006]. The detailed study of umbilical cord is important because abnormal cord insertion is correlated with the low birth weight of live twin. In the present case examination of placenta showed velamentous insertion of the placenta but the birth weight of surviving twin was normal. The presence of vascular anastomosis in monochorionic placentas might lead to vascular disruption injury and result in placental infarct due to interruption of maternal blood supply to fetus [Woo et al., 2000]. Fetal development and functions need sufficient blood supply to placenta, so placental infarct result in prenatal fetal death [Djafar et al., 2006].

The primary concern of fetus papyraceous is its effect on surviving twin and mother [Sinha et al., 2010]. The reported effects of fetus papyraceous on surviving twin include risk of cerebral palsy, congenital abnormalities like neural tube defects, microcephaly, post hemorrhagic hydrocephalus, bilateral renal cortical necrosis, unilateral absence of kidney, GIT Atresia, hemifacial microsomia and aplasia cutis [Misbah et al., 2005]. The authors [Glinianaia et al., 2002] reported the increased risk of cerebral palsy in the surviving twin after the fetal death of a co-twin. The probable causes explained are consequences of prematurity, twin transfusion syndrome associated with monochorionicity. In the present study no congenital abnormalities and complications were detected in surviving twin during prenatal period and after birth.

Mortality and morbidity were mainly related to the gestational age of fetus papyraceous. If IUD occurred during 1st trimester, the morbidity was limited and it could be diagnosed by maternal complications like mild vaginal bleeding and cramping. If IUD occurred later, morbidity will be high and possible maternal complications include preterm labor, infection from retained fetus, severe purpural hemorrhage, consumptive coagulopathy and dystocia leading to caesarian delivery [Sinha et al., 2010]. In this case ultrasound showed that IUD had happened approximately in late 2nd trimester and the dead fetus was retained in utero till 39th week without any severe complications. If a dead fetus is retained in utero for a very long period that is more than 5 weeks, tissue thromboplastin from dead fetus and placenta might enter into maternal circulation due to vascular connections and gradually reduce fibrinogen leading to haemostatic impairment [Misbah A et al. 2005]. In the present case no
hematological abnormalities were reported both in mother and surviving twin.

Twin embolisation syndrome is a complication followed by fetal demise as a result of vascular coagulation and emboli formation. The emboli could damage highly vascularised organs like brain and kidney [Upadhyaya I et al. 2009]. Twin transfusion syndrome is diagnosed by acute hydramnios developed at the midtrimester of gestation and visible difference in the size of the twins in sonography [Djafar et al., 2006]. In this study, hydramnios was never reported in any of the ultrasound results and the size of the fetus showed a slight difference till the death of one fetus.

Antenatal death of one fetus in the late 2nd or 3rd trimester of a pregnancy posed an important management dilemma in obstetrics [Woo et al., 2000]. Prior to the use of ultrasound the diagnosis of fetus papyraceous could only be made out after delivery. The advent of real time ultrasound permitted the detection of multiple gestation as early as four weeks after conception using intravaginal probe [Anand et al., 2007]. In the presence of fetus papyraceous, maternal serum α-fetoprotein level and amniotic fetoprotein level might be elevated prenatally and amniotic acetyl cholinesterase might be increased [Luckoffc et al., 2002]. If fetus papyraceous is diagnosed antenataly, serial evaluation of surviving fetus by sonography, biophysical profile, doppler and maternal coagulation factors should be done. Zygosity and chorionicity should be detected antenataly [Rasheed and Amjad, 2008]. Long term follow-up of the surviving twin is necessary and it should start right from neonatal evaluation to detect neural, renal, circulatory and cutaneous defects by new techniques like high resolution ultra sonography of the brain, computed tomography, renal function studies and magnetic resonance imaging [Woo et al., 2000].

Conclusion

A case of monochorionic twin gestation with the outcome of a single live fetus with a fetus papyraceous, and its probable causes are described. If fetus papyraceous is detected earlier, antenatal determination of chorionicity and zygosity is important to assess the potential risks. Along with routine surveillance, detailed evaluation of fetal anomalies, a close fetal surveillance by ultrasonography, biochemical tests, psychological counseling is essential. After delivery, histopathological examination of placenta and membranes and postmortem examination of dead fetus should be done to detect the causes of IUD. Moreover a long term follow up of surviving fetus in physical, biological and psychological aspects is also important.

REFERENCES


