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Two fetal papyraceous in triplet pregnancy in a resource limited setting: A case report

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Abstract
Introduction: Fetal papyraceous results when an early fetal death occurs in utero (usually in early 2nd trimester between 15 to 20 weeks) and the fetus is not expelled out. A single fetal death in multiple pregnancy is not uncommon. However the death of one or more fetuses in multiple pregnancy leads to increased morbidity and mortality of the surviving fetus/ fetuses.
Case Discussion: A case of a 43 year old primigravida with a triplet pregnancy who had fetal demise of two of the fetuses. She was monitored closely and eventually had an Emergency caesarean section at 30 weeks gestation, with delivery of a live fetus and two fetal papyraceous.
Conclusion: Fetal papyraceous diagnosed in the second or third trimester is rare. Careful fetal and maternal monitoring is required to improve fetal and maternal outcomes.

Keywords: Fetal demise, fetal papyraceous, triplet, multiple gestation

Introduction
The last three decades has seen an enormous rise in the incidence of multiple pregnancies and this is due to increases in ovulation induction, *in vitro* fertilization (IVF) as well as childbearing at older ages [1].
A single fetal death in multiple pregnancy is not uncommon [2]. However the death of one or more fetuses in multiple pregnancy leads to increased morbidity and mortality of the surviving fetus/fetuses [2]. The rate of intrauterine accident is 1 in 184 twin pregnancies (0.54%) and about 1 in 8000 triplet pregnancies [3]. Fetal papyraceous occurs in about 1 in 12, 500 twin pregnancies and 1 in 32,800 triplet pregnancies [3]. Two papyraceous fetuses in a triplet pregnancy is exceedingly rare [3].
Fetal papyraceous results when an early fetal death occurs in utero (usually in early 2nd trimester between 15 to 20 weeks) and the fetus is not expelled out [3]. This results in atrophy and mummification of the fetus. A fetal papyraceous is a macerated, small, completely formed fetus, it is commonly dry and papery as a result of the amniotic fluid and fluid content of the fetus being absorbed and the fetus compressed between the living fetus and the uterine wall [3].
The true incidence of fetal papyraceous is difficult to ascertain in Nigeria, as majority of women do not receive antenatal care and deliver at home [4].
There are no established guidelines in management of these cases due to its rarity, however if the surviving fetus is near term, delivery of the baby appears rational. If the fetal deaths occur before term, conditions such as a consumptive coagulopathy, risk of infection, or impending death of the surviving fetus would inform delivery [5].

Case report
A 43 year old Primigravida presented to the Obstetric emergency at a gestational age of 30 weeks and one day. Pregnancy was conceived through In-vitro fertilization with 2 embryos transferred, however one of the embryo split resulting in a triplet pregnancy, with ultrasound scan done a 7 weeks showing a triplet pregnancy. She had been previously managed as a case of chronic hypertension in pregnancy with Tabs alpha-methyl dopa and nifedipine. At her 24 weeks routine Ultrasound scan she was found to have fetal demise of two of the triplet fetuses. She was subsequently closely monitored with serial fetal surveillance using twice weekly fetal biophysical profile and maternal coagulation profile.

She presented at 30 weeks and one day with a history of reduced fetal movement, there was no bleeding per vagina and no drainage of liquor. Fetal biophysical profile done at presentation was abnormal with a score of 4. Her blood pressure at presentation was 130/90 mmHg. She was counseled for an emergency lower segment caesarean section which she consented to. Intraoperative findings were a live female neonate with birth weight of 1.3 kg and Apgar scores of 8 and 9 in the 1st and 5th minutes respectively and two mummified fetuses, each fetus attached to its individual placenta and in different amniotic sacs.

The baby was admitted to the special care baby unit. Post-operatively mother and baby remained stable and mother was subsequently discharged on the 5th post-operative day. Baby was discharged at a weight of 1.5kg.

Discussion

There is a paucity of reports on intrauterine fetal demise of two fetuses in a triplet pregnancy [2]. Teliga- Czajkowska *et al*. in 2003 reported a fetal demise of two fetuses in a triplet pregnancy with eventual delivery of the surviving fetus by emergency caesarean section at 31 weeks as a result of suspected maternal compromise. This was similar to our index case [6].

It is often difficult to diagnose fetal papyraceous if obstetric ultrasounds are not done during routine antenatal visits, as a result many are diagnosed only after delivery [3]. High order multiple gestations can be diagnosed by transvaginal ultrasonography as early as the 4th week. In the late second and third trimesters, it is often difficult to diagnose fetal papyraceous by ultrasound [3].

A study performed by Dickey, et al., examined the relationship between the number of fetuses and fetal demise [7]. They found that spontaneous reduction of one or more gestational sacs and or embryos occurred before the 12th week of gestation in 53% of triplet, and 65% of quadruplet pregnancies [7]. Fetal death occurring after 20 weeks gestation is rare, with an incidence of 2.6% of twin and 4.3% of triplet gestations [8]. When fetal death occurs, the survival of the remaining fetuses is inversely related to the time of the first fetal demise, with an increased risk of fetal death seen with monochromic placentation and monozygosity [8]. Our index patient had fetal demise of two fetuses at 24 weeks with each fetus having a separate placenta.

If fetal papyraceous is diagnosed antenatally serial surveillance of the surviving fetus should be done by sonography, biophysical profile, doppler and maternal coagulation factors should be done serially [4]. Complications of a fetal papyraceous on the surviving fetus include intrauterine growth restriction, congenital disorders, cord complications, pre-term labour, and prematurity. Risk of cerebral impairment of the surviving fetus is about 20% with corresponding increase in the incidence of cerebral palsy [4].

Conclusion

Fetal papyraceous diagnosed in the second or third trimester is rare. Careful fetal and maternal monitoring is required to improve fetal and maternal outcomes.

Fig 1: Live neonate with two fetal papyraceous

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