Management Dilemma Posed By Intrauterine Fetal Death of A Co-twin Remote from Term: A Case Report

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Primigravida, aged 20 years, presented at 26 weeks of gestation for antenatal check-up with no associated problems. The uterus was large for date, ultrasonography revealed twin pregnancy with one fetal death. The couple decided to continue the pregnancy and was followed-up closely clinically, with serial sonography and coagulation profile. A live female baby was delivered by emergency cesarean section at 36 weeks with ruptured membranes in early labor with the fetus papayraceous lying transversely in the lower segment covering the internal os causing obstruction to the delivery of the viable twin. Both mother and surviving neonate did not have any complications at and after birth. The infant attained all developmental milestones till one year of follow-up.

Keywords: fetal death, fetus papyraceous, twin pregnancy.

The incidence of multifetal pregnancies has been increasing worldwide due to increasing use of assisted reproductive techniques. In one hospital based study from eastern part of Nepal the incidence of twin pregnancy was reported as 9.2 /1000 births[1] while from another hospital located in western part it was

reported the as 1.9/1000 births.[2] Death of one twin remote from term with continuation of other pregnancy can occur at times posing management dilemmas. Loss of one twin in the first trimester does not appear to impair the development of the surviving twin.[3] Fetal death occurring in second or third trimester may increase the risk of intrauterine growth retardation, preterm labor, preeclampsia, and perinatal mortality as well as risk of maternal coagulation complications.[4] The exact cause of fetal death is usually unknown and can vary including twin–twin transfusion, placental insufficiency, intrauterine growth retardation related to preeclampsia, cord accidents, velamentous insertion of the cord, cord stricture, cord around the neck, fetal chromosomal abnormalities and congenital abnormalities.[4,5] Though the maternal and fetal complications in affected cases can be severe, we report one case of fetus papyraceus managed conservatively without any complications. Successful outcome is related to careful monitoring during pregnancy and delivery.

Case Report

A primigravida, 20-year-old, presented at 26 weeks of amenorrhea for routine antenatal check up. She had attended antenatal check-up once at five months of pregnancy in a health post for the first time and was apparently all right. She had been married for two years and had no significant past medical or surgical history and no associated co-morbidity. She was perceiving fetal movements and did not have any complications till date. Fundal height was more than the period of gestation corresponding to 32 weeks of gestation. Routine antenatal blood investigations and ultrasonography was advised. Ultrasonography revealed twin pregnancy with one fetus dead. The alive fetus was estimated to be around 26 weeks of gestation while the dead fetus appeared lying in lower part of the uterus and was smaller, about 20 weeks on sonographic measurements. One placenta was identified which appeared to be fundal and dividing amniotic membranes were not seen separately. Hemogram parameters and serology were within normal limits. A diagnosis of twin pregnancy with intrauterine fetal death of a co-twin was made. She did not have any history of infertility treatment, use of ovulation induction drugs or family history of multiple pregnancy. Since sonography had not been done earlier definite diagnosis of chorionicity could not be made at that time. The woman and her family members were informed about the death of a co-twin and counselling was done regarding the possible complications that could arise. The couple decided to continue the pregnancy. Coagulation profile was within normal limits and the need for close fetal surveillance was stressed. She was followed up closely with check-ups every two weeks including sonographic assessment at 2-3 weekly interval. Serial sonography and maternal blood and urine parameters that were done till 35 weeks were within normal

limits. Sonography done at 35 weeks revealed alive fetus growing normally with the dead fetus lying in the lower part of the uterus transversely.

She presented a week later with ruptured membranes in early labor at 36 weeks of gestation. Fetal heart rate was within normal range, liquor was clear, she was four centimeters dilated with fully effaced cervix. Only irregular bony hard fetal parts were palpable on vaginal examination. Fetal head of the alive twin was palpable on per abdominal examination as freely ballotable. Emergency cesarean section was done and alive female baby weighing 2.8 kg with good Apgar score was delivered as vertex. The fetus papyraceous was lying transversely in lower uterine segment. One half of the placenta looked infarcted and calcified irregularly while one half appeared normal grossly. Dividing amniotic membranes could not be separated into layers grossly. Fetus papayraceous was of female sex and weighed 650 grams while the whole placenta weighed 380 grams. The woman and family did not consent to pathological examination of placenta citing religious reasons. The surviving baby did not have any gross congenital anomalies and was followed up till one year of age who attained all developmental milestones during that period.

Discussion

Twin pregnancy with death of one fetus remote from term and continuation of co-twin with normal development is not a common event. The term fetus papyraceus is used when intrauterine fetal demise of a twin occurs in second or third trimester, with retention of the fetus for a minimum of 10 weeks resulting in mechanical compression of the small fetus such that it resembles parchment paper.[6] The incidence of fetus papyraceous has been reported at 1 in 12,000 pregnancy, and ranges between 1:184 and 1:200 twin pregnancies.[7] Though the maternal and fetal complications in affected cases can be severe, we report a fetus papyraceus **(Figure 1)** with surviving co-twin managed conservatively without any complications with successful outcome of the surviving twin with close feto-maternal surveillance.

Figure 1: Fetus papyraceous after delivery

Upadhay et al have reported one case of antepartum fetal death diagnosed at 32 weeks with continuation of pregnancy to term with normal fetomaternal outcome of the surviving twin.[8] In our case report the approximate time of death of the co-twin

could not be ascertained as the woman presented for first time at 26 weeks with demise of a co-twin diagnosed on routine ultrasound. Dahiya and Bains have reported two cases of intrauterine death of co-twin remote from term managed conservatively with successful outcome of the surviving twin.[9] The main dilemma in continuing the pregnancy is the sequel to the surviving twin and risk of maternal coagulation disorders. Maternal psychological fear and emotional issues also need to be addressed. Jain and Purohit have reviewed 64 twin pregnancies with 5 single fetal deaths.[4] The authors report 3 out of 5 twin pregnancies with single intrauterine fetal death being successfully managed to term and 3 live fetus salvaged by close monitoring.[4]

Figure 2: Single placenta with infarcted and calicified half

Management guidelines are not uniform in this uncommon clinical entity and needs to be tailored. The RCOG Study Group on Multiple Pregnancy (published 22/2/2011) states that the death of a co-twin management depends on chorionicity, gestation and time since death. A conservative policy of increased surveillance with delivery at 37 weeks is recommended in dichorionic pregnancies. ACOG guidelines disagree about the preferred antepartum surveillance method and management once demise of a co-twin has occurred. Some investigators have advocated immediate delivery of the remaining fetuses. However, if the death is the result of an abnormality of the fetus itself rather than maternal or uteroplacental pathology, and the pregnancy is remote from term, expectant management may be appropriate.[7] The sequel to the surviving twin depends on zygosity and chorionicity. Monoamniotic twins are obviously at highest risk of fetal complications. In our case two separate amniotic sacs could not be identified separately during antenatal ultrasonography. Placenta was single and both fetuses were of same sex as noted after delivery. About half portion of the placenta corresponding to the dead co-twin appeared infarcted and dividing membranes could not be identified on gross examination **(Figure 2)**. Detailed pathological examination of the placenta could not be done as the family did not consent citing religious beliefs of cremating the placenta together with the dead fetus. Bajoria et al have reported outcome of twin pregnancies complicated by single intrauterine death in relation to vascular

anatomy of the monochorionic placenta. Determination of chorionicity with evidence of twin to twin transfusion syndrome and the type of anastomosis arterio-arterial, venovenous or arteriovenous has a bearing on the outcome of the twins.[5] Ascertaining the timing of fetal death is important because death of recipient twin first may lead to significant sequel in the surviving twin.[5] Embolization of placental and fetal thromboplastin or necrosed fragments of dead placenta can damage vital organs like kidney and brain of the surviving twin. Similar to singleton intrauterine fetal death with retention of the dead fetus for prolonged period has a possibility of triggering DIC in the mother, neurological and nephrological damage and preterm delivery.[8]

The management dilemmas in our case included the decision to continue pregnancy as the approximate time of fetal death and chorionicity could not be ascertained. The other dilemma was to decide the mode of delivery as the fetus papyraceous was lying in the lower uterine segment across the dilated internal os in early labor. In view of the benefit of the surviving twin emergency cesarean section was performed to avoid the risks of obstructed labor. Motevelo and Ndaboine have reported a case of fetus papyraceous causing labor dystocia in a rural setting needing emergency cesarean section.[6,9]

Conclusion

The primary concern of death of a co- twin remote from term is its effect on the surviving fetus and on the mother. To avoid possible complications, close feto-maternal surveillance by clinical, sonographic and coagulation profiles are necessary for successful outcome. The fact that fetus papraceous can sometimes cause labor dystocia due to its malpresentataion has to be considered during labor management.

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