Case Report Fetus Papyraceous : Rarity Among Rarities

RASHEED F.¹, AMJAD N.²

Address or Correspondence: Dr Farhat Rasheed, Consultant Gynaecologist & Head of the Department, Ittefaq Hospital Trust, Lahore.

A 28 years old G3 P2 A0 with previous two cesarean sections presented for routine antenatal care at 10 weeks of gestation. She had family history of twin pregnancy in her real sister. Ultrasonography showed viable monochorionic, diamniotic twin gestation. She was advised regular antenatal visits. Ultrasound at 16 weeks showed intrauterine death of one of the fetuses with normal growth parameters of the second twin. Vigilant feto-maternal monitoring was done throughout the rest of her pregnancy and elective Cesarean section at 38 weeks yield a male baby of 3.4 kg. Examination of the placenta showed two cords one of which was attached to fetus papyraceous.

Key Words: Twin pregnancy, Fetus Papyraceous, Vanishing twin.

Introduction

Fetus papyraceous or compressus is the compressed, mummified, parchment-like remains of a dead twin which is retained in-utero after intrauterine death in the second trimester.¹⁻³ It is usually discovered among the placenta and membranes of its well-developed twin. This process is unusual, since fetal demise usually results in spontaneous abortion, stillbirth or complete resorption in early pregnancy. Fetus papyraceous is a relatively rare complication in twin pregnancy. The occurrence of fetus papyraceous is frequently associated with perinatal morbidity in the other twin as surviving fetus can have problems due to twin embolization syndrome (TES), making antenatal diagnosis of this condition desirable. Ultrasound detection is not always possible due to anatomical position and technical difficulties. It may demonstrates a small but recognizable nonviable fetus.

The incidence of fetus papyraceous has been reported at 1 in 17,000 to 20,000 pregnancies.³ This intrauterine accident occurs in 1 in 184 twin pregnancies $(0.54\%)^4$ and in about 1 in 8000 triplet pregnancies⁵. Incidence of fetus papyraceous in triplets around 1 in 32,800⁶ Two papyraceous fetuses in a triplet pregnancy is exceedingly rare. Fetus papyraceous can occur in both uniovular and binovular twins but is more common in uniovular twins. We present a case of fetus papyraceous diagnosed at 16 weeks of gestation in a twin pregnancy and surviving twin was delivered at 38 weeks of gestation in a healthy state.

Case Report

The patient was a 28 years old lady with history of previous two cesarean sections due to Cephalopelvic disproportion. Her last child born was 3 years old. She had regular menstrual cycle of 7/28 days. She was sure of her dates and presented at 10 weeks of gestation for routine antenatal checkup. She had family history of twin pregnancy in her real elder sister. Gynaecological, obstetrical, past medical and surgical history was unremarkable. General physical and systemic examination revealed no abnormality. She was advised Tab folic acid 5 mg daily, routine baseline investigations and obstetrical ultrasound. She was advised to come for follow up with the test reports.

Complete blood and urine examination was within normal limits. Blood group was 0 positive. Blood sugar level was within normal limits. Obstetrical ultrasound showed viable twin pregnancy of 10 weeks. Placenta was developing anteriorly. There was no other significant finding. She was counseled that twin pregnancy is a high risk pregnancy, associated feto-maternal risks were also explained to her and was advised regular frequent follow ups.

At twelve weeks of gestation, oral haematinics and calcium was started. Repeat ultrasound was done at 16 weeks of gestation for fetal well being. Unfortunately, it showed intrauterine death of one of the twins. However, the other twin was growing at a normal pace. The family was once again counseled in detail about the whole situation and risks to the surviving fetus were also explained to them. She was called for antenatal visits fortnightly, serial growth scans were done every 15 days for the surviving twin along with maternal coagulation studies.

After 28 weeks, along with obstetrical ultrasounds, her pregnancy was followed by weekly biophysical profile and Doppler ultrasound fortnightly. Aim was to prolong the pregnancy to as close to term as possible. Her Antenatal period remained uneventful and the surviving fetus showed normal growth on serial fetal biometries. Elective cesarean section was done at 38 completed weeks of gestation because of previous two cesarean sections due to Cephalo-pelvic disproportion. Outcome was an alive, male baby weighing 3.4 kg with good APGAR scores at 1 and 10 minutes. No obvious congenital abnormality was present. Close examination of the placenta showed a separate amniotic sac containing fetus papyraceous attached to the placenta by a separate umbilical cord (Fig. 1).



Fig. 1: Single Placenta showing a separate amniotic Sac.

The papyraceous fetus had a crown-heel length of 14 cm, head circumference of 7 cm, and weight of 40 g (Fig. 2). The mother had a normal puerperium. She was discharged on 4^{th} post-op day in a stable condition along with the baby.



Fig. 2: Fetus Papyraceous showing separate umbilical cord.

Discussion

In this case, the surviving fetus grew normally and no gross morbidity occurred at birth. Fetus Papyraceous is a macerated, tiny, fully, formed fetus which is usually dry and papery because the amniotic fluid and fluid content of the dead fetal tissues and of the placental tissue gets absorbed and the dead fetus gets flattened and compressed between the membranes of the living co-twin and the uterine wall. Most of the fluid will get absorbed from the fetus if it is retained for more than8 weeks in the uterus. Death of a binovular twin helps the surviving twin. The surviving twin often has the sequelae of twin embolization syndrome which is a complication following in-utero demise of the co-twin. It results from the embolization of placental and fetal thromboplastin or necrosed fragments of the dead placenta causing dissemedated intravascular coagulation or even end arteritis.^{7,8} The emboli predominantly damage highly vascularised organs such as brain and kidney but can affect almost all organ systems. In the central nervous system these emboli can result in ventriculomegaly, porencephaly, cysticencephalomalacia, microcephaly, and cerebral palsy. Prevalence of cerebral pasly in surviving twins was over 50 times higher then normal⁹. Extracranial abnormalities include small bowel atresia, gastroschisis, hydrothorax, renalcortical necrosis and aplasia cutis. Aplasiacutis congenita is a rare disorder characterized by localized absence of skin at birth.¹⁰ If fetus papyraceous is diagnosed antenatally serial evaluation of the surviving fetus by sonography, biophysical profile, doppler and maternal coagulation factors should be done serially. Zygosity and chorionicity evaluation should be performed antenatally. If possible delivery should be scheduled at tertiary center. These measures usually bring a good outcome for the surviving fetus.

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