Spontaneous triplet pregnancy with twin fetuses papyraeci: a rare case report and review of the literature

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Abstract

A fetal death in a multiple pregnancy with one or more normally surviving fetus is unusual. Fetus papyraceous (FP) is a rare obstetric complication in multiple gestations. It is defined as retention of a mummified parchment like remains of a dead fetus in multiple pregnancy associated with a viable twin. It is important to reassure the patient of the normal outcome expected in most of the cases. Herein, we report a rare case of twin FP in a spontaneous triplet pregnancy with a literature review of maternal and neonatal outcomes and management of similar cases.

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Introduction

Multiple pregnancies have become one of the most common high-risk conditions encountered during pregnancy especially after the era of assisted reproductive techniques. Triplet pregnancy and higher-order births have increased 470% and the incidence of triplets reaches a frequency of 1 in 500 deliveries.[1]

Fetus papyraceous (FP) is a medical condition defined as retention of a mummified parchment like remains of a dead fetus(es) in multiple pregnancy associated with a viable twin.[2] Its incidence is 1 in 17,000-20,000 pregnancies.[3] However, the presence of twin fetuses papyraeci in a triplet pregnancy is much rarer with an incidence 1 in 32,800.[4]

The condition is difficult to diagnose if antenatal visits with obstetric ultrasound are not done, thus many times diagnosis is only made after delivery. It is usually discovered accidentally among the placenta and membranes of its well-developed co-twin due to amniotic fluid loss, or resorption and compression of the dead fetus.[3]

Maternal complications associated with

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fetus papyraceous include disseminated intravascular coagulation (DIC), preeclampsia, antepartum hemorrhage, polyhydramnios, and preterm labor.[5] However, expectant management with close maternal and fetal monitoring is advised if diagnosed antenatally.

Here, we report a case of spontaneous triplet pregnancy with twin FP diagnosed in the second trimester that was managed conservatively and delivered at full term with no maternal or fetal complications.

Case presentation:

A 27 years old woman, gravida 2, para 1, pregnant 9 weeks (based on her last menstrual period) presented to the Outpatient Antenatal Clinic of Assiut Woman's Health Hospital for routine antenatal care. Her ultrasound scan reported triplet gestation with three living embryos; their crown rump lengths (CRL) average between 8-9 weeks gestation.

She had one living child delivered by an elective cesarean section (CS) at term of 2 years duration. She got pregnant spontaneously without using any ovulation induction drugs. Past history was unremarkable of any obstetric or gynecological events. There was no family history of multiple pregnancy. We recommended folic acid intake 5 mg daily with regular antenatal follow-up.

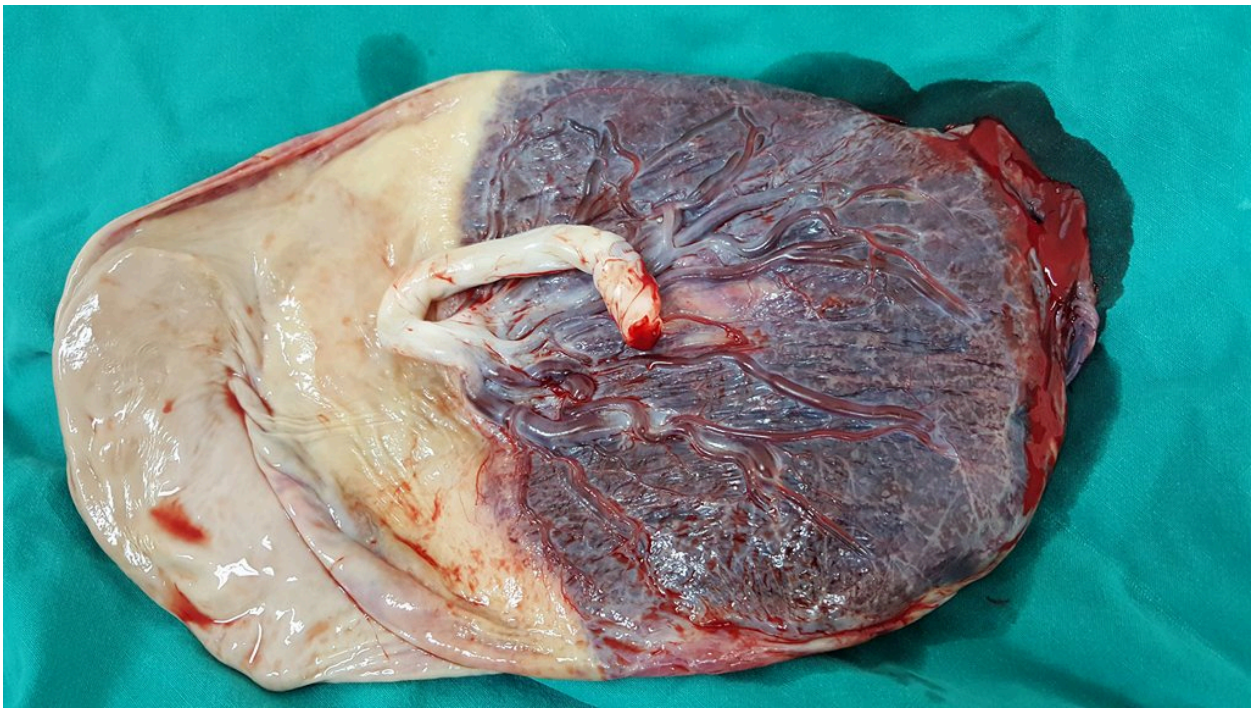

Figure 1: The fetal surface of the placenta showing eccentrically attached umbilical cord of the surviving baby.

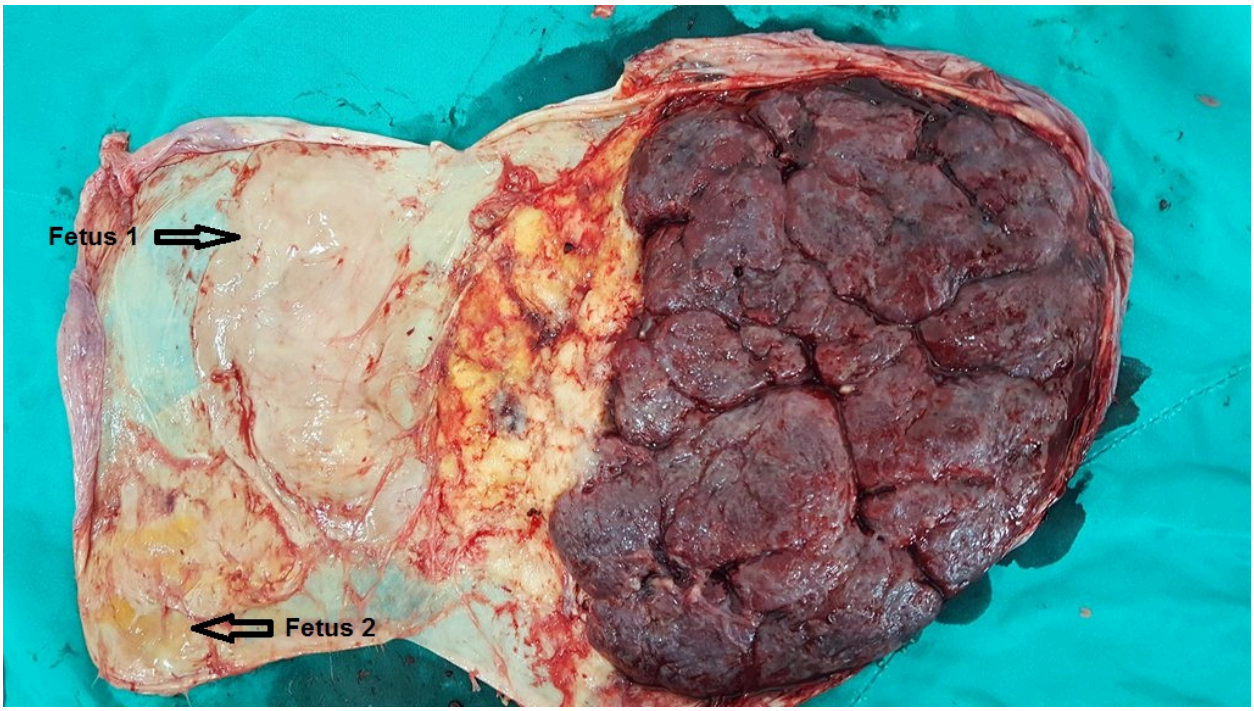

Figure 2: The maternal surface of the placenta of the surviving twin with twin fetuses papyraeci.

At 15th week, a trans-abdominal ultrasound scan revealed only a single living fetus, which measured 14.5 weeks on average biometry. The other two fetuses were non-viable with CRL of 12 and 10 weeks. There was no history of vaginal bleeding. She was admitted for observation of her condition and to conduct initial investigations to exclude any complications. A complete blood picture, with prothrombin time, concentration and activated partial thromboplastin time was done with normal values. The patient was counseled with regard to fetomaternal risks for expectant management and discharged with advice for more regular antenatal check-up. Low molecular weight heparin (LMWH) prophylaxis was introduced at a dose of 5000 IU/ day till delivery as a prophylaxis against the occurrence of DIC.

An ultrasound scan at 20 weeks for anatomy revealed no major abnormality in the living fetus with a normal rate of growth which was had follow-up ultrasounds every 4 weeks. The placenta was anterior high with average amniotic fluid. Serial antepartum fetal surveillance by cardiotocograph every 4 weeks was carried out and expressed adequate fetal movements and normal heart rate patterns.

The fetus presentation at term was breech, so the patient was counseled for repeat CS and informed consent was obtained. She was admitted at 38 weeks and elective CS was performed under spinal anesthesia by a senior obstetrician. A healthy female neonate with an Apgar score of 10/10 at 1 minute and birth weight of 3100 gm was delivered through a low transverse uterine segment incision.

The placenta was carefully delivered completely along with the membranes. The umbilical cord of the healthy baby was eccentrically attached to the placenta (Figure 1). There were twin fetus papyraeci flattened parchment-like and compressed beneath the membranes (Figure 2). They measured 13.5 and 8 centimeters in length (Figure 3), and the whole placenta weighed 550 grams. No complications occurred in the third stage. The uterus was closed in double layers followed by routine closure of other layers.

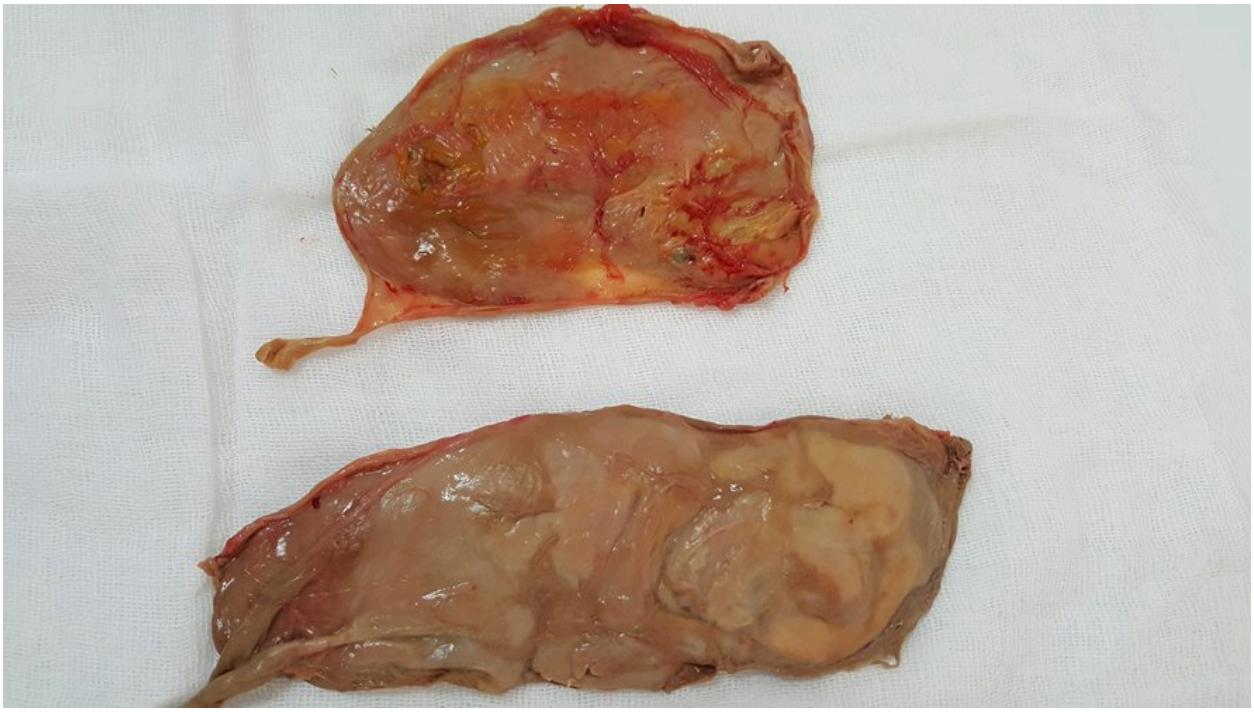

Figure 3: Gross specimen of the two mummified fetuses.

The patient and her normal healthy baby had a smooth postoperative course. They were discharged one day later with uneventful postpartum period.

Discussion

The current case report is one of the few cases published in the literature about twin fetuses papyraeci in triplet pregnancy. Most of the data concerning this rare condition comes from cases reported in the literature. To the best of our knowledge, there are only five reported cases, in the current century, of twin FP in triplet pregnancies [6-10] in addition to our case (Table 1).

Death of one or more fetuses intrauterine with a surviving twin is a challenging issue in the management of multiple pregnancy. The surviving twin is more prone to neurological complications, cerebral palsy, renal failure or intrauterine death especially if sharing the same placenta with the FP.11 This may be attributed to the feto-fetal transfusion imbalance leading to congenital anomaly or death of the surviving twin.[12,13]

In our case, the surviving fetus in the

triplet pregnancy had no evident complications at delivery mostly due to its separate placenta with no vascular anastomosis with the twin papyraeci. Cerebral palsy is uncommon if the survivor has a separate placenta from the twin papyraeci.[14] However, a previous study reported that the surviving twin had lower scores on the Mental and Development Scales if compared to singleton pregnancies.[15]

Management of FP with a surviving twin mainly depends on the chronicity; gestational age at diagnosis and occurrence of complications during pregnancy.[16] If the condition occurs in the first trimester, the fetus usually completely absorbed with no more pregnancy complications.[17] However, if it occurs after the first trimester, the risks of the aforementioned maternal and fetal complications will be high. Occasionally at term, a low-lying FP may obstruct the vaginal delivery pathway that necessitates a CS to deliver the surviving twin[18]. In the current case, FP had occurred at the end of first trimester, so expectant management was offered to the patient and the course of pregnancy was completed successfully without any complications till 38 weeks gestation.

The condition of twin FP is difficult to diagnoses without sonographic scans during antenatal visits. In the previously reported cases, two cases were diagnosed only after delivery[7,10] due to poor antenatal care. In our case, the condition was diagnosed at the 15th week of pregnancy. This allowed us to add LMWH therapy to the patient's treatment as a prophylactic line against development of thrombosis and its harmful consequences.

Although they are non-specific, there are some reported biochemical markers implicated in diagnosis of FP. An increase in pregnancy associated plasma protein-A, beta human chorionic gonadotropin and alpha fetoprotein levels were reported with FP.[19,20] If FP is diagnosed antenatally, serial evaluation of the surviving twin by biophysical profile and Doppler should be done in addition to serial evaluation of maternal coagulation factors.[21]

This case discussed three important clinical objects. Firstly, FP should be diagnosed by ultrasound scan as early as possible to provide more close and regular checkup of the woman during her antenatal course. Secondly, LMWH should be considered in the treatment of cases with early FP to avoid the thrombotic complications.[22] Thirdly, routine postnatal placental examination should be instituted searching for a FP in cases with poor antenatal care.

Conclusions

In conclusion, we report a case of twin fetuses papyraeci in a spontaneous triplet pregnancy with no maternal or fetal complications during pregnancy and delivery. Routine ultrasound scan during antenatal visits of pregnant women with multiple pregnancy is important for detection of such condition. This will allow early diagnosis of FP and prevent future obstetrical complications with reduction in the risk of mortality and morbidity for the surviving fetus.

Table 1: Maternal and neonatal outcomes and management of cases of twin fetuses papyraeci in triplet pregnancy

Case	Year	Maternal age (years)	Parity	Gestational age at diagnosis	Maternal Complications	Mode of delivery	Gestational age at delivery	Birth weight (grams)	Neonatal complications
Teliga-Czajkowska et al.[6]	2003	33	1 + 0	20 weeks	PROM	Emergency CS	31 weeks	1380	Respiratory distress
Mittal and Khanna[7]	2007	25	3 + 0	Not diagnosed	Nil	Vaginal delivery	Term	2600	Nil
Bukar et al.[8]	2013	39	Multi	27 weeks	Preterm labor	Emergency CS	36 weeks	2300	Nil
Gulati et al.[9]	2015	25	1 + 0	24 weeks	Nil	Vaginal delivery	Term	2250	Nil
Mynso et al.[10]	2015	------	2 + 0	Not diagnosed	Nil	Vaginal delivery	Term	3100	Nil
Current case	2016	27	1 + 0	15 weeks	Nil	Elective CS	38 weeks	3100	Nil

PROM; premature rupture of membranes, CS; cesarean section

References

1. American College of Obstetricians and Gynecologists Committee on Practice Bulletins-Obstetrics.; Society for Maternal-Fetal Medicine.; ACOG Joint Editorial Committee.. ACOG Practice Bulletin #56: Multiple gestation: complicated twin, triplet, and high-order multifetal pregnancy. Obstet Gynecol. 2004 Oct;104(4):869-83. https://doi.org/10.1097/00006250-200410000-00046 PubMed PMID: 15458915.

2. Fisk NM. Multiple pregnancy. In: Edmonds DK, editor. Dewhurst's textbook of obstetrics and gynaecology. 7th ed. Oxford: Blackwell; 2007. p. 166–176.

3. Bush M, Pernoll ML. Multiple pregnancy. In: DeCherney AH, Nathan L, editors. Current obstetric and gynecologic diagnosis and treatment. 9th ed. New York: McGraw-Hill; 2003. p. 315–25.

4. Hommel H, Festge B. [Triplet pregnancy with papyraceous foetuses (author's transl)]. Zentralbl Gynakol. 1979;101(13):845-7. German. PubMed PMID: 506533.

5. Børlum KG. Third-trimester fetal death in triplet pregnancies. Obstet Gynecol. 1991 Jan;77(1):6-9. PubMed PMID: 1984228.

6. Teliga-Czajkowska J, Dadalska E, Sopliński A. [Triplet pregnancy complicated by intrauterine death of two fetuses--case report]. Ginekol Pol. 2003 Oct;74(10):1302-5. Polish. PubMed PMID: 14669434.

7. Mittal PS, Khanna M. Two papyraceous fetuses in a triplet pregnancy. J Obstet Gynecol India. 2007Jan-Feb;57(1):77-8.

8. Bukar M, Chama C, Bako B, Jonathan B. Twin fetuses papyraeci in a spontaneous triplet pregnancy presenting with unexplained preterm contractions. Ann Med Health Sci Res. 2013 Nov;3(Suppl 1):S13-5. https://doi.org/10.4103/2141-9248.121210 PubMed PMID: 24349837; PubMed Central PMCID: PMC3853596.

9. Gulati G, Das B, Deepika. Twin foetus papyraceous in triplet pregnancy. Int J Reprod Contracept Obstet Gynecol. 2015 Aug;4(4):1214-6. http://dx.doi.org/10.18203/2320-1770.ijrcog20150461

10. Mynso KS, Singh LB, Singh NN, Meetei LT, Devi KP, Sharma S. Fetus papyraceous – a case report with successful maternal and fetal outcome of the triplet. IOSR J Dent Med Sci 2015;14(2):9-11. http://DOI:10.9790/0853-14210911

11. Taylor CL, de Groot J, Blair EM, Stanley FJ. The risk of cerebral palsy in survivors of multiple pregnancies with cofetal loss or death. Am J Obstet Gynecol. 2009 Jul;201(1):41.e1-6. https://doi.org/10.1016/j.ajog.2009.02.003 PubMed PMID:19394587.

12. Gonen R. The origin of brain lesions in survivors of twin gestations complicated by fetal death. Am J Obstet Gynecol. 1991 Dec;165(6 Pt 1):1897-8. https://doi.org/10.1016/0002-9378(91)90059-Z PubMed PMID: 1750494.

13. Grafe MR. Antenatal cerebral necrosis in monochorionic twins. Pediatr Pathol. 1993 Jan-Feb;13(1):15-9. https://doi.org/10.3109/15513819309048188 PubMed PMID: 8474948.

14. Ikpeze OC, Nwosu OB. Management of multiple gestation in which fetus papyraceous is a co-twin. Trop J Med Res. 1998;2:84–6.

15. Anand D, Platt MJ, Pharoah PO. Comparative development of surviving co-twins of vanishing twin conceptions, twins and singletons. Twin Res Hum Genet. 2007 Feb;10(1):210-5. https://doi.org/10.1375/twin.10.1.210 PubMed PMID: 17539382.

16. Cleary-Goldman J, D'Alton M. Management of single fetal demise in a multiple gestation. Obstet Gynecol Surv. 2004 Apr;59(4):285-98. https://doi.org/10.1097/01.OGX.0000120165.52159.04 PubMed PMID:15024229.

17. Saidi MH. First-trimester bleeding and the vanishing twin. A report of three cases. J Reprod Med. 1988 Oct;33(10):831-4. PubMed PMID: 3057206.

18. Lau WC, Rogers MS. Fetus papyraceous: an unusual cause of obstructed labour. Eur J Obstet Gynecol Reprod Biol. 1999 Sep;86(1):109-11. https://doi.org/10.1016/S0301-2115(99)00053-6 PubMed PMID: 10471152.

19. Chasen ST, Perni SC, Predanic M, Kalish RB, Chervenak FA. Does a "vanishing twin" affect first-trimester biochemistry in Down syndrome risk assessment? Am J Obstet Gynecol. 2006 Jul;195(1):236-9. https://doi.org/10.1016/j.ajog.2006.01.044 PubMed PMID: 16626615.

20. Abbas A, Johnson M, Bersinger N, Nicolaides K. Maternal alpha-fetoprotein levels in multiple pregnancies. Br J Obstet Gynaecol. 1994 Feb;101(2):156-8. https://doi.org/10.1111/j.1471-0528.1994.tb13084.x PubMed PMID: 7508260.

21. Rasheed F, Amjad N. Case report, fetus papyraceus: rarity among rarities. Annals. 2008Oct-Dec; 14(4):154-5.

22. Shaaban OM, Abbas AM, Zahran KM, Fathalla MM, Anan MA, Salman SA. Low-Molecular-Weight Heparin for the Treatment of Unexplained Recurrent Miscarriage With Negative Antiphospholipid Antibodies: A Randomized Controlled Trial. Clin Appl Thromb Hemost. 2016 Aug 29. [Epub ahead of print] https://doi.org/10.1177/1076029616665167 PubMed PMID: 27572887.