Case report

FETUS PAPYRACEOUS: A RARE CLINICAL ENTITY IN DICHORIONIC-DIAMNIOTIC TWIN PREGNANCY

**ABSTRACT**

Fetus papyraceous, a high order pregnancy complication with an incidence of 1:12:0000 pregnancies and 1:200 in twin pregnancies, is a rare condition in which an intrauterine death of one fetus occurs causing mechanical compression over a period. The most concern for the pregnant woman remains the status of the life fetus and any unforeseeable complications. Genetically, the cause is unknown but may be related to poor antenatal screening, low socioeconomic status and certain maternal co-morbidities. Rarely, it could result in intrauterine death of the surviving twin, preterm labor, coagulopathy, obstructed labor and postpartum hemorrhage. Hereto, we present a case of a 37-year-old female Gravida 3 Para 2(1 dead), with dichorionic-diamniotic twins who developed fetal papyraceous of one twin

*Key Words: Fetus Papyraceous, Dichorionic-Diamniotic, Twin Gestation*

**1. INTRODUCTION**

Multiple pregnancies have become one of the high-risk situations that obstetricians encounter most often. Twins account for about 3% of every live birth (1). Multiple births represent 17% of all premature deliveries and 26% of all infants with very low birth weight, who face a seven-fold greater risk of dying before their first birthday (1). Fetus papyraceous, or fetus compressus, refers to the intrauterine demise of a fetus in early twin gestation, with the retention of that fetus for as long as 10 weeks; this leads to mechanical compression of the small fetus, making it resemble parchment paper due to the loss of fluids (2). Fetus papyraceous, also called mummified fetus, is a rare complication with incidence of 1:12,000 live births and 1:184-1:200 twin pregnancies (2). Fetus papyraceous is an occasional condition in dichorionic-diamniotic twin pregnancies, where one twin dies early in gestation and is compressed and flattened, resembling parchment paper, within the uterus. This occurs because the dead fetus is retained and its fluids are absorbed, leaving it flattened against the uterine wall and the membranes of the living twin (3). It typically occurs when there is intrauterine death of one twin in the late first or second trimester with retention of this twin, leading to compression and mummification with eventual appearance like a parchment (dry and paper like), hence the name papyraceous (2,3).

The etiology is unknown but it is thought to be associated with fetal genetic abnormalities, twin-to-twin transfusion Syndrome (TTTS) and cord abnormalities such as velamentous cord insertion, lethal nuchal cord or true knot (4). Occasionally, it could have both maternal and fetal complications of surviving twin. The death of one twin typically occurs in the first or early second trimester. The dead fetus is compressed, dehydrated, and mummified, taking on a parchment-like appearance (2,4). While the surviving twin may be unaffected, complications like antepartum stillbirth, intrauterine growth retardation, and maternal complications (postpartum hemorrhage, infection) are associated with fetus papyraceous. Fetus papyraceous can be diagnosed via ultrasound, allowing for early detection and monitoring of the pregnancy. Expectant management with close monitoring is often recommended when the diagnosis is made early. After delivery, careful examination of the placenta and fetus is crucial for confirmation and assessment (2).

**2. CASE PRESENTATION**

A 37-year-old Gravida 3 Para 1(dead) + 1(medical termination of pregnancy), presented for antenatal booking at the antenatal clinic of Tema General hospital, Tema, Ghana. Her dating ultrasound scan showed Dichorionic-Diamniotic twins, at 12 weeks 3 days with an EDD of 12/03/2024. Antenatal booking bloods were unremarkable. Past obstetric history revealed two years prior to this pregnancy, she had had an early neonatal death due to fetal distress occurring in labor, requiring an emergency delivery via caesarean section and a medical termination of pregnancy five (5) years earlier for an unwanted pregnancy. She had an uneventful antenatal appointment from booking to 16 weeks with no co-morbidity and was on routine antenatal medications consisting of ferrous sulphate 200mg twice daily, folic acid 5mg daily and calcium 500mg twice a day.

At 20 weeks, an anomaly scan was carried out which showed Dichorionic- Diamniotic (DCDA) twin gestation with demise of leading Twin (Twin A). Twin A also exhibited Spalding sign consistent with intrauterine fetal demise and anhydramnios. The estimated gestational age was reported as15weeks + 3days, and estimated fetal weight was 160g. The placenta was anterio-fundal. Twin B was a live fetus with adequate liquor volume (AFI-10cm), cephalic presentation, estimated gestational age at 22weeks + 5days and estimated fetal weight of 373g. Preterm premature rupture of membranes was ruled out. Patient was counselled about the condition and the risk to the her and the life fetus. She had 2 weekly antenatal monitoring with ultrasound scan, hematological, biochemical and coagulation screen which were all normal. Extensive emotional and psychological support was offered throughout the pregnancy especially given her background of early neonatal death. At the 36weeks appointment, the ultrasound done showed a single intrauterine fetus with estimated fetal weight of 3100g, liquor volume extensively reduced with AFI-2.3cm (only one measurable pool) and normal umbilical artery doppler studies. Preterm premature rupture of membranes was ruled out. CTG done was normal.She was counselled for delivery and an emergency cesarean section was carried out with findings of a live female, with birthweight 2900g and APGARS 8/10 and 9/10 at 1 and 5 minutes. No gross abnormality detected. A fetus papyraceous of approximately 16 weeks gestation with weight 179g and height of 14cm was also delivered (FIGURE 1 and 2). Detailed newborn assessment of the surviving twin was done to rule out any congenital malformations and none was found.

**3. DISCUSSION**

If fetal demise of one twin occurs early in the first trimester, it results in intrauterine absorption of the dead fetus leading to vanishing twin but when it occurs in the late first or second trimester, then fetus papyraceous can ensue, where the dead fetus is compressed and mummified into a parchment state (hence its name).

Fetus papyraceous is a rare condition of multiple pregnancy occurring after the demise of one twin with retention of this twin for at least 10 weeks, to allow absorption of amniotic fluid, placenta and fluid content of the dead fetus (4). It can occur in both monochorionic and dichorionic twins but it is more common in monochorionic twins, due to shared vascular connections with resultant increase in fetal demise risk by three-fold (5). The resultant maternal and fetal complications may include bleeding and miscarriage in the first trimester, preterm premature rupture of membranes, preterm labor, cord complications, congenital disorders, sepsis and coagulopathy in the second and third trimester. The surviving fetus in Diachronic-Diamniotic twins (DCDA) rarely has complications compared to Monochorionic twins (6), where the risk of cerebral palsy, aplasia cutis and anomalies such as microcephaly, cerebral encephalomalacia or hydrocephalus are high (7,8,9). The pathophysiology is hypothesized to be due to reverse transfer of thromboplastin to the surviving twin leading to disseminated intravascular coagulation and severe intrauterine central nervous system damage (10). If fetus papyraceous is diagnosed antenatally, close monitoring is of utmost importance. Monitoring of the mother and fetus is done. Severe maternal complications rarely occur and most times, decision to end the pregnancy is due to fetal complications. Mode of delivery is according to local guidelines for management of twin pregnancy with associated fetal complications but majority will require earlier delivery.

In cases of uniovular twins, intrauterine death is three times more frequent due to the high incidence of vascular connections (85–98%). Maternal issues encompass preterm labor, consumptive coagulopathy, labor dystocia, and sepsis due to the retention of a deceased fetus (5). Fetal complications that can affect survival include congenital abnormalities, restricted growth within the uterus, early birth, and mortality. So far, the congenital anomalies reported in the live fetus include gastroschisis, aplasia cutis, intestinal atresia, central nervous system damage, absent ear, and heart anomalies. These abnormalities are primarily due to thrombus and other coagulation elements released from the deceased fetus, which embolizes the living twin, causing vascular obstruction (4). If the fetus is entirely absorbed during the first trimester, there are typically no additional complications in the pregnancy. Our patient was diagnosed with fetal papyraceous early in the second trimester thanks to excellent diagnostic resources. The complication may be more serious in a monochorionic placenta than in a dichorionic placenta. No link between parity and maternal age was identified. In general, both the mother and the surviving twin stay healthy without complications, as observed in our case (8). Monitoring the mother’s biochemical, hematological, and ultrasonography status during the antenatal period and post-delivery is crucial for evaluating consumptive coagulopathy and maternal infection (6). A thorough examination of the newborn is essential to exclude congenital malformations. Histopathological analysis of the placenta during the postpartum period can aid in understanding the reasons for fetus papyraceous

**4. CONCLUSION**

Fetus papyraceous, a rare phenomenon in twin gestation can have complications for both the mother and the surviving twin. These complications are vast and close follow up is needed to improve outcome.

**CONSENT**

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

**ETHICAL APPROVAL**

No ethical approval required.

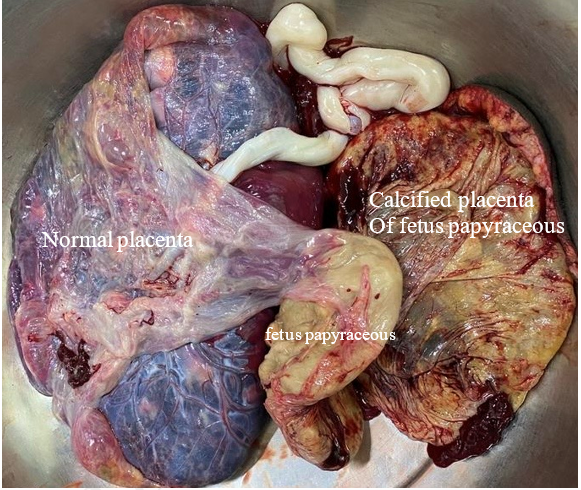
Disclaimer (Artificial intelligence)

Author(s) hereby declare that NO generative AI technologies such as Large Language Models (ChatGPT, COPILOT etc.) and text-to-image generators have been used during the writing or editing of this manuscript.

**REFERENCES**

1. Pharoah, P.O.D., Glinianaia, S.V., & Rankin, J. (2010). Congenital anomalies in multiple births after early loss of a conceptus. Human Reproduction, 24, 726-731
2. Kumar, R., Upreti, P., Malik, N., Kumari, O., & Dagar, N. (2023). Fetus Papyraceous in Diamniotic Dichorionic Twins: A Rare Entity. Journal of South Asian Federation of Obstetrics and Gynecology, 15(3), 347–348. <https://doi.org/10.5005/jp-journals-10006-2240Dahiya>
3. Dahiya, P., & Bains, R. (2014). Conservative Management of Fetus Papyraceus: A Report of Two Cases. Oman Medical Journal, 29(2), 132–134. https://doi.org/10.5001/omj.2014.32
4. S., L., & K., R. (2019). A rare case of fetus papyraceous presenting in monozygotic diamniotic twins. International Journal of Reproduction, Contraception, Obstetrics and Gynecology, 9(1), 448. <https://doi.org/10.18203/2320-1770.ijrcog20196066>
5. Vashistha, P., Kashyap, P., Kumar, A., Sethi, C. A., & Rana, S. (2020). Fetus papyraceous, a rare complication of twin pregnancy: case report. International Journal of Reproduction, Contraception, Obstetrics and Gynecology, 10(1), 361. <https://doi.org/10.18203/2320-1770.ijrcog20205799>
6. Akbar, M., Saeed, R., Ikram, M., Saeed, M., & Talib, W. (2005). Fetus Papyraceous. The Professional Medical Journal, 12(03), 351–353. <https://doi.org/10.29309/tpmj/2005.12.03.5136>
7. Srinidhi, C., & Jajoo, S. (2023). Fetus papyraceous: a rare clinical image. Pan African Medical Journal, 44. <https://doi.org/10.11604/pamj.2023.44.28.38689>
8. A Case Report: Aplasia Cutis Congenita Secondary to Fetus Papyraceus. (2021). Pediatrics and Neonatal Medicine, 1(1). <https://doi.org/10.33425/2768-0363.1005>
9. Anand, D., Platt, M. J., & Pharoah, P. O. D. (2007). Vanishing Twin: A Possible Cause of Cerebral Impairment. Twin Research and Human Genetics, 10(1), 202–209. <https://doi.org/10.1375/twin.10.1.202>
10. Pharoah, P. O. D. (2007). Prevalence and pathogenesis of congenital anomalies in cerebral palsy. Archives of Disease in Childhood - Fetal and Neonatal Edition, 92(6), F489–F493. <https://doi.org/10.1136/adc.2006.107375>
11. Benirschke, K. (1992). The contribution of placental anastomoses to prenatal twin damage. Human Pathology, 23(12), 1319–1320. <https://doi.org/10.1016/0046-8177(92)90048-8>

FIGURE 1: Normal Placenta on the left, Fetus Papyraceous with its placenta on the right



.

FIGURE 2: Fetus Papyraceous

