

## DEMISE OF CO-TWIN IN SECOND TRIMESTER LEADING TO FETUS POPYRACEOUS – SUCCESSFUL OUTCOME IN SURVIVING TWIN

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### ABSTRACT

Foetus 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. It is an uncommon finding. The incidence of foetus papyraceous is reported as 1 in 17,000 to 1 in 20,000 pregnancies and in twin pregnancy it is 1 in 184 to 1 in 200 pregnancies. A case series of foetus papyraceous found in three different twin pregnancies over one year are reported here.

**Keywords:** Foetus papyraceous, Intrauterine death, Twin pregnancy

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### 1. Introduction

Foetus 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<sup>1-3</sup>. It is usually discovered among the placenta and membranes of its well-developed twin<sup>2</sup>. The cause is thought to be death of one twin, amniotic fluid loss, or reabsorption and compression of the dead fetus<sup>3</sup>. The incidence of fetus papyraceous has been reported to be 1 in 17,000 to 20,000 pregnancies<sup>3</sup> and in twin pregnancy it is 1 in 184 to 1 in 200 pregnancies<sup>4</sup>. Death of one twin in first trimester with vanishing twin syndrome is relatively common (up to 29%) and the pregnancy usually continues with little adverse effect on the mother and twin. But death of one twin in second or third trimester is more serious with an increased risk for surviving twin and possibility of maternal disseminated intravascular coagulation (DIC). It is emphasized that a close high-risk obstetric management must be used and a careful follow up of the neonate must be done. Three cases of twin pregnancies with death of co twin leading to foetus papyraceous are described and reviewed here.

### 2. Case Series

**2.1. Case Report 1:** A 25 years old woman, second gravid with one abortion with twin gestation was admitted in a rural, tertiary care centre at 35+4 weeks gestation with complaints of lower abdominal pain. The patient had conceived spontaneously. On examination, general condition was fair and systemic examination revealed no abnormality. Fundal

height was more than the period of gestation and hence obstetric scan was advised which showed a viable twin (“A”) with 35 weeks gestation with cephalic presentation and fundal posterior placenta and a non viable twin (“B”) of 17 weeks gestation and anterior fundal placenta. Her routine investigations and coagulation profile were done and were found to be within normal limits. Patient was managed conservatively for few days and discharged with advice for regular antenatal check-up. Follow up ultrasound examination was done at 37 weeks, all reports were corresponding with her gestational period and presentation was cephalic. Patient was admitted again at 38 weeks of pregnancy with spontaneous onset of labour. The patient delivered normally a live twin “A” male baby with an Apgar score of 8/10 and birth weight of 2.640 kg and weight of placenta 400 gm. Patient delivered a twin “B”, dead foetus papyraceous weighing about 100 gm with placenta weighing 50gm. Inspection of placenta showed diamniotic dichorionic variety. Amount of blood loss during normal delivery was average. Postpartum period was uneventful and mother- baby pair was discharged after 3 days.

**2.2. Case Report 2:** A 27 years old, un booked woman second gravida , para one with one live issue with previous one cesarean delivery came to the obstetric out patient at 19+5 weeks gestation for routine antenatal check-up. Her ultrasound report revealed twin gestation with first twin live with breech presentation at 19 weeks and placenta upper posterior and a nonviable second twin of 14 weeks gestation with placenta upper anterior. She was admitted for observation and investigations. Coagulation

profile was done which came out to be within normal limits. Patient was treated conservatively and discharged with advice for regular antenatal check-up. Follow up ultrasound examination was done at 26 weeks, 32 weeks and 37 weeks and all reports were corresponding with her gestational periods and presentation was breech at term. Patient was admitted at 38 weeks and elective cesarean section was performed for breech presentation of first twin and previous history of LSCS for cephalopelvic disproportion. A live female with an Apgar score of 8/10 was delivered with birth weight of 2.539 kg and weight of placenta 410 gm. Second sac contained a dead foetus papyraceous, parchment like stuck to the amniotic membrane. Placenta was diamniotic dichorionic. Post operative period was uneventful and mother- baby pair was discharged after 8 days.

**2.3. Case Report 3:** A 20 years old unbooked, uninvestigated, primigravida woman presented in emergency hours at 38+6 weeks gestation to labour delivery unit with chief complaints of spontaneous onset of labor pains since one day. On general examination her vitals were normal and on per abdominal examination uterine height corresponded to the period of gestation. Single foetal heart sound was heard. On per vaginal examination cervix was 7-8 cms dilated and fully effaced with the presenting part (vertex) at +2 stations. Patient delivered normally a live male baby weighing 2.714 Kgs with 9/10 Apgar score. While attempting to deliver the placenta it was diagnosed that it is a twin pregnancy with second sac bulging out. Membranes of second sac were ruptured and within 10 minutes a dead, macerated, parchment like baby weighing 500 gms was delivered. Inspection of the placenta showed monochorionic diamniotic placenta.

Postpartum period was uneventful and mother-baby pair was discharged in good condition after 3 days.

### 3. Discussion

Twin pregnancy or multiple gestations is a common finding in today's era especially due to artificial reproductive technology and in vitro fertilisation. Conception rate of multiple gestations are greater than birth rates<sup>5</sup>, thus the death of fetuses in multiple pregnancy is not uncommon. Among pregnancies with twin sacs or embryos, 30% will ultimately result in singletons and less than 10% will result in no foetuses<sup>5</sup>. Frequently, it takes the form of vanishing twin syndrome, in which a foetus

simply dies for no apparent or known reason. In most cases, the body of the mother reabsorbs the foetus, leaving few traces that it ever existed. However, in rare circumstances, the foetus may instead be flattened against the side of the uterus by the sibling creating a foetus papyraceous<sup>3</sup>. Frequently the only sign left of vanishing twin is the development of subtle abnormality of placenta such as well-defined cysts or sacs, areas of degenerated chorionic villi, fibrin deposition or fibrinoid degeneration, placental nodules or plaques and embryonic remnants. Causative factors for foetus papyraceous have been debated in literature. The role of velamentous and marginal insertion of placenta has been postulated<sup>6</sup>. The condition occurs more often in fetuses with genetic or chromosomal abnormalities<sup>5</sup>. Placental or foetal analysis frequently reveals diploidy, triploidy, and alternate sex chromosome on placental biopsy, foetal skin biopsies, and chorionic villous sampling<sup>5</sup>. Rare case of a foetus papyraceous due to maternal trauma has also been reported in the literature.

Earlier, foetus papyraceous was thought to be more common in monozygotic twins. But recently it was found that they occur with similar frequencies in both dichorionic (12%) and monochorionic twin (11%) pregnancies<sup>7</sup>. It is more difficult to predict viability of twins early in pregnancy, more so when the placenta is monochorionic. There is no prognostic effect of maternal age and spontaneous versus induced conception. The primary concern of foetus papyraceous is its effect on mother and surviving co-twin. In most cases no complications to the mother or to the surviving twin have been reported<sup>8</sup>. Morbidity and mortality are mainly related to the gestational age of foetus papyraceous. When vanishing twin syndrome occurs during the first trimester, morbidity is limited and the mother is most likely to develop mild vaginal bleeding and cramping. If the event occurs later in the pregnancy, the morbidity is high. Maternal complications include preterm labour, infection from a retained foetus, severe puerperal haemorrhage, consumptive coagulopathy, and obstruction of labour by a low-lying foetus papyraceous causing dystocia leading to caesarean delivery<sup>8</sup>. Vanishing twin in a pregnancy significantly increases both preterm (<37 gestational weeks) and very preterm (<32 gestational weeks) births<sup>8</sup>. The effects on surviving twin include risk of cerebral palsy, congenital abnormalities like neural tube defects

(NTDs), optic nerve hypoplasia, hypoxic ischemic lesions of white matter, microcephaly, post haemorrhagic hydrocephalus, bilateral renal cortical necrosis, unilateral absence of kidney, gastrointestinal tract atresia, gastroschisis, hemifacial microsomia and aplasia cutis<sup>6</sup>. It was reported that surviving co-twins had poorer scores on the Griffiths Mental and Development Scales when compared to singleton pregnancies<sup>9</sup>. Prior to the use of ultrasonography, the diagnosis of foetus papyraceous could only be made after delivery of the surviving twin. The advent of real time ultrasound permits the diagnosis of multiple gestations as early as four weeks after conception using the transvaginal probe<sup>6</sup>. The subsequent demise of one foetus could then be diagnosed before delivery. Recently various biochemical markers have been implicated in diagnosing foetus papyraceous. Foetus papyraceous or vanishing twin has been shown to increase pregnancy associated plasma protein-A (PAPP-A) and free beta human chorionic gonadotropin (hCG)<sup>10</sup>. Alpha-fetoprotein levels are elevated compared with values at similar junctures in both singleton pregnancy and normal twin pregnancy<sup>11</sup>. The rate of rise of beta-hCG is slower than that in a normal twin pregnancy<sup>12</sup>. If foetus papyraceous is diagnosed antenatally, serial evaluation of the surviving fetus by sonography, biophysical profile and doppler should be done. Maternal coagulation factors should also be done serially and zygosity/chorionocity evaluated antenatally. Death of a binovular twin may help the surviving twin. On the other hand the surviving twin often has the sequelae of twin embolization syndrome 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 disseminated intravascular coagulation or even end arteritis. As technology advances, the routine surveillance of early pregnancy will be helpful in early detection of foetus papyraceous before the development of complications.

In the test series we have reported, the death of co twin did not have any significant adverse effect on the surviving twin or the mother and successful outcome could be obtained in all mother –baby pairs.

#### 4. Conclusion

The sequelae of a single foetal death in twin pregnancy depend on the gestation and in the second and in the late third trimester there is

increase in mortality or morbidity in the surviving twin. Antenatal evaluation periodically by ultrasonography is important to reduce the potential risk. Conservative management remains the main stay but the risk of keeping the live foetus in the hostile intrauterine environment has to be weighed against the risk of preterm delivery.

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